114 年年會 海報論文展示：病例報告

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病例報告 114_C001

以胃鏡診斷的上腸系膜動脈症候群-病例報告

Superior Mesenteric Artery Syndrome Diagnosed Via Panendoscopy: A case report

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Introduction

Superior mesenteric artery syndrome (SMAS), also known as Wilkie's syndrome or cast syndrome, is a rare condition characterized by compression of the third portion of the duodenum between the aorta posteriorly and the superior mesenteric artery (SMA) anteriorly. The syndrome typically occurs following significant weight loss, which reduces the fat pad between the SMA and aorta, thereby decreasing the aortomesenteric angle and distance

Case Report

A 16-year-old female with no significant medical history presented with nausea and epigastric pain persisting for 10 days. The patient was born prematurely at 30 weeks gestational age but had no history of necrotizing enterocolitis or abdominal surgery. She reported anorexia, abdominal distension, and weight loss of 4 kilograms from 44 kilograms within two weeks. Her symptoms, particularly epigastric pain, vomiting, and abdominal distension, were exacerbated postprandially. She denied fever, dysphagia/odynophagia, chest pain, dyspnea, radiation pain, or gastrointestinal bleeding. Previous treatment at a local pediatric clinic had been ineffective.

Physical examination revealed a soft abdomen with mild tenderness over the epigastric region, without rebound pain or muscle guarding. Abdominal ultrasonography showed no significant abnormalities. An abdominal X-ray revealed fecal retention without evidence of obstruction or free air.

Panendoscopy demonstrated luminal compression of the third portion of the duodenum with obvious pulsation of the medial duodenal wall, raising suspicion for SMAS. Subsequent contrast-enhanced abdominal CT confirmed the diagnosis, showing a reduced aortomesenteric angle of 16.6° and diminished aortomesenteric distance, although complete duodenal obstruction was not present.

Discussion

This case highlights the diagnostic challenge of SMAS and the utility of panendoscopy in establishing the diagnosis. The classic clinical presentation of SMAS includes postprandial epigastric pain, early satiety, nausea, and vomiting, which are relieved by positional changes that increase the aortomesenteric angle, such as left lateral decubitus, prone, or knee-chest positions. Our patient exhibited these typical symptoms with postprandial exacerbation and significant weight loss, creating a self-perpetuating cycle characteristic of SMAS.

Conclusion

This case illustrates the importance of considering SMAS in the differential diagnosis for adolescents presenting with postprandial abdominal pain and weight loss. The direct visualization

of duodenal compression via panendoscopy served as a crucial diagnostic clue, prompting confirmation with CT imaging. This underscores the utility of panendoscopy in the diagnostic approach to SMAS, potentially expediting diagnosis in cases where clinical suspicion is high.

病例報告 114_C002

對稱性周邊壞疽：病例報告

Symmetric peripheral gangrene: A Case Report

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Introduction

Symmetrical peripheral gangrene (SPG) is a rare but severe condition characterized by distal ischemic necrosis of two or more extremities without large vessel obstruction, most commonly caused by sepsis, disseminated intravascular coagulation (DIC), or low-flow states.

Case Report

An 83-year-old woman with diabetes and hypertension was referred from a local hospital due to an infected right perirenal hematoma with septic shock and acute renal and respiratory failure following double-J stent insertion for a ureteral stone with obstructive uropathy and *E coli* bacteremic urosepsis. On admission to our intensive care unit, physical examination showed symmetric gangrene involving both hands and lower legs, with palpable peripheral pulses. She received antibiotic therapy and pigtail drainage for the infected perirenal hematoma, which cultured the same pathogen. Her clinical course was complicated, but she ultimately recovered from the systemic infection, with residual dry gangrene of multiple fingers and scarring of the forearms and lower legs. She declined amputation and chose conservative management.

Discussion

SPG results from microcirculatory failure in the setting of sepsis, DIC, and hypoperfusion, despite preserved large-vessel flow. Treatment is mainly supportive, aiming at infection control, hemodynamic optimization, and wound care. Amputation is usually postponed until tissue necrosis becomes well demarcated, though eventual multi-limb loss remains a common outcome.

Conclusion

This case demonstrates a typical pattern of SPG that emerged in the setting of gram-negative sepsis. The development of distal limb ischemia was likely driven by a combination of septic shock and DIC. Prompt recognition and early initiation of supportive therapies are vital.

病例報告 114_C003

假性副甲狀腺低下症 Ib: GNAS 甲基化異常造成腎臟對有 PTH 阻抗性及低血鈣

Pseudohypoparathyroidism type Ib with abnormal GNAS gene methylation presented with renal limited PTH resistance and hypocalcemia

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Introduction

The well-known clinical manifestations of pseudohypoparathyroidism may include neurocognitive dysfunction, brachydactyly, ectopic ossification, and parathyroid hormone resistance. We reported a case who was diagnosed with Pseudohypoparathyroidism type Ib due to abnormal GNAS gene methylation and he presented with renal-limited PTH resistance and hypocalcemia.

Case Report

Mr. Lin is a 29-year-old male with normal developmental history, intelligence, and height. He presented with a two-year history of limb numbness. During a physical examination, hypocalcemia was incidentally discovered (serum calcium 5.6 mg/dL), and he was referred to the nephrology outpatient clinic.

Mr. Lin's renal function was normal, but he had elevated parathyroid hormone levels (iPTH 333.5 pg/mL) and slightly reduced 25-hydroxyvitamin D (25-(OH)D). Common causes of vitamin D deficiency, including insufficient intake, limited sunlight exposure, malabsorption, hepatobiliary disease, and medications were sequentially excluded. Moreover, serum 25-hydroxyvitamin D would typically need to fall below 10 ng/mL to cause hypocalcemia. In addition, Mr. Lin did not exhibit typical manifestations of rickets such as skeletal deformities, growth retardation, or hypotonia. Therefore, the possibility of pseudohypoparathyroidism was considered.

DNA sequencing analysis did not reveal pathogenic mutations in the GNAS gene. Further laboratory collaboration identified abnormal GNAS gene methylation, leading to the diagnosis of pseudohypoparathyroidism type Ib.

He was subsequently treated with regular administration of active vitamin D3 and calcium supplements, with periodic follow-up.

Discussion

Epidemiologic studies have reported an estimated prevalence of 0.34 per 100,000 in Japan. The clinical manifestations of pseudohypoparathyroidism may include neurocognitive dysfunction, brachydactyly, short stature, ectopic ossification, parathyroid hormone resistance, thyroid-stimulating hormone (TSH) resistance and growth hormone resistance. Biochemical abnormalities typically include hypocalcemia, hyperphosphatemia and elevated parathyroid hormone levels. The clinical symptoms and severity of pseudohypoparathyroidism vary widely among individuals, and the diagnosis can be confirmed by molecular genetic analysis. Pseudohypoparathyroidism type Ia (PHP-Ia) is defined by resistance to multiple hormones (including PTH and TSH), features of Albright hereditary osteodystrophy (AHO), and reduced Gsα

activity. Pseudohypoparathyroidism type Ib (PHP-Ib) is defined by resistance to PTH, absence of AHO features, normal Gsα activity, and abnormal methylation of the GNAS gene. Management of hypocalcemia due to PTH resistance is similar to primary hypoparathyroidism, involving the use of active vitamin D metabolites or analogues in combination with oral calcium supplementation.

Conclusion

When evaluating the causes of hypocalcemia, in patients with normal renal function, elevated PTH, exclusion of vitamin D deficiency, and absence of rickets or typical Pseudohypoparathyroidism Ia manifestations, the possibility of pseudohypoparathyroidism type Ib due to renal-limited PTH resistance should be considered.

病例報告 114_C004

首例台灣耳-顎-指症候群第一型病例

First reported case of orofaciodigital syndrome type 1 in Taiwan: mimicker of autosomal dominant polycystic kidney disease

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Introduction

The orofaciodigital syndrome (OFDS) is a heterogenous group with presentations involving anomalies of face, mouth, digits, kidney cysts, and development disorders. Different gene mutations in variable inheritance patterns contribute to this syndrome. Orofaciodigital syndrome type 1 (OFDS1, also known as Papillon-Léage-Psaume syndrome) is the most common type of OFDS with X-linked inheritance. Abnormalities of body appearance in OFDS1 patients may be minimal. Multiple renal cysts are usually present as the first finding in many OFDS1 patients, which mimic autosomal dominant polycystic kidney disease (ADPKD).

Case Report

A 23-year-old previously healthy woman presented to our Nephrology Outpatient Department for bilateral renal cysts incidentally found in a health examination. Repeated renal echo showed bilateral mildly-enlarged kidneys (right kidney: 13.1cm, left kidney: 12.6cm). Bilateral multiple renal cysts looked different from ADPKD. The patient's renal cysts contained uneven distribution, irregular border, and not bulged out of the renal capsule. The following gene test revealed *OFD1* p.Ile229fs (c.686_689delTTGA), which is a pathogenic mutation of OFDS1. Tracing back her medical record, she received surgical intervention in her infant period for simple syndactyly of the right fourth and fifth finger, for polydactyly of the right fifth finger, and for cleft palate. In her twelfth year, another operation with Z-plasty and scar release was performed for webbing of her right fourth finger web space, and for clinodactyly (radial deviation) of the proximal interphalangeal joint of the right fifth finger. Based on above-mentioned anomalies of her face, digits, multiple renal cysts and gene test, the patient was finally diagnosed with OFDS1.

Discussion

There are many disorders present as mimickers of ADPKD with renal cysts, including Alagille syndrome (a development disorder, with small cystic kidneys and abnormal kidney function), collagen disorders, autosomal dominant tubulointerstitial kidney disease, recessive polycystic kidney disease (PKD), tumorous disorders, and syndromic ciliopathies. OFDS1 is one of the syndromic ciliopathies that mimics ADPKD. ADPKD is usually caused by gene mutations in *PKD1* and *PKD2*, while other mimickers of ADPKD contain different gene mutations, such as *JAG1*, *NOTCH2* in Alagille syndrome, and *COL4A3*, *COL4A4*, *COL4A5* in COL4A-related diseases (collagen disorders). OFDS1 is more common in female patients, with male lethality. OFDS1 contains malformations of the face (hypertelorism, broad nasal bridge, hypoplasia of nasal alae), mouth

(cleft lip, cleft palate, tongue nodules), and digits abnormality (syndactyly, polydactyly, clindactyly), PKD, and abnormal kidney function, etc.. To date, there is no definite treatment for OFDS1. Management of OFDS1 focuses on reconstructive surgery for the affected parts of the body. The treatment effect of the ADH-receptor antagonist, Tolvaptan, in OFDS1 patients is unknown.

Conclusion

When PKD and other presentations in different organs appear in a patient, other differentiated diagnoses besides ADPKD should be taken into consideration. Gene tests may help distinguish between these disorders. OFDS1 is one of the disorders that mimics ADPKD. Anomalies of the face, mouth, fingers, and other body parts can help identify this disease. Gene test is usually used to make confirmed diagnosis.

病例報告 114_C005

擬 Fabry 病理表現之膜性腎病變病例報告

Membranous Nephropathy with Mimicking Fabry Disease Pathology: A Case Report

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Introduction

Nephrotic syndrome and hematuria require comprehensive evaluation, with renal biopsy being the diagnostic gold standard when atypical findings are present. Fabry disease, an X-linked lysosomal storage disorder, is characterized by zebra-body inclusions in podocytes; however, Fabry-like changes can also be seen in mimics, such as drug-induced lysosomal dysfunction or genetic variants. Here, we report a case of PLA2R-associated membranous nephropathy with Fabry-like pathological findings, in which the distribution of inclusions was limited and localized, and neither enzymatic deficiency, drug exposure, nor LMX1B-associated features were identified.

Case Report

A 51-year-old woman with CKD stage 4, hypertension, hyperlipidemia, and right breast DCIS presented with hematuria and nephrotic syndrome. Laboratory tests showed BUN 29 mg/dL, creatinine 1.82 mg/dL, albumin 2.6 g/dL, UPCR 4645.5 mg/g, UACR 2999 mg/g, and 15–20 RBC/HPF, consistent with nephrotic syndrome and hematuria. She was admitted on July 4, 2024, for a renal biopsy.

Discussion

Laboratory Findings

- Serological tests for autoimmune diseases, including ANA, anti-dsDNA, ANCA (both p-ANCA and c-ANCA), and anti-GBM antibodies, were all negative. Viral markers for hepatitis B and hepatitis C were also non-reactive. Complement levels (C3 97.0 mg/dL, C4 44.1 mg/dL) were within normal ranges, while immunoglobulin levels showed no specific abnormalities (IgG 1074 mg/dL, IgA 197 mg/dL, IgM 970 mg/dL, IgE 104.3 IU/mL).
- PLA2R antibody: positive
- Fabry testing: α-galactosidase A activity and genetic testing were negative.

Pathology

- Light microscopy: chronic interstitial fibrosis, tubular atrophy, segmental glomerulosclerosis, and mesangial expansion.

Special stains: Silver stain showed spikes formation. PAS stain revealed thickened basement membrane.

- Immunofluorescence: PLA2R positive; consistent with primary membranous nephropathy.
- Electron microscopy: subepithelial deposits and myelin figures resembling zebra bodies.
- Final diagnosis: Primary membranous nephropathy with PLA2R positivity, mimicking Fabry disease.

Conclusion

Fabry disease results from α-galactosidase A deficiency with glycosphingolipid accumulation and typical zebra bodies. Similar Fabry-like changes may also appear in drug-induced lysosomal dysfunction (e.g., chloroquine, amiodarone) or in LMX1B-associated nephropathy. In this case, renal biopsy revealed Fabry-like inclusions with concurrent PLA2R-associated membranous nephropathy. Enzyme and genetic tests were negative, and there was no drug exposure or LMX1B-related features. Unlike true Fabry disease, the inclusions were more limited and localized. This case highlights the importance of careful clinicopathological correlation to guide proper management.

病例報告 114_C006

原發性腎上性功能不足病例報告

Primary adrenal insufficiency – A Case report

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Introduction

Primary adrenal insufficiency results from adrenal gland destruction, leading to impaired production of hormone. This case discusses a 57-year-old man received bilateral adrenalectomy and subsequently developed primary adrenal insufficiency.

Case Report

A 57-year-old male patient with history of right UPJ stone s/p Right URS-SM + JJ stent, arthritis. gout, hypertension, was found to have bilateral adrenal gland enlargement during routine outpatient follow-up. Surgical intervention was recommended, and the patient subsequently underwent bilateral adrenalectomy. Postoperatively, he was followed regularly in the outpatient clinic, with no significant signs or symptoms of adrenal insufficiency during the follow-up period.
The patient was later hospitalized for surgical treatment due to abdominal wall protrusion and testicular enlargement. Following discharge, he developed diarrhea, abdominal pain, and poor appetite, necessitating readmission. Laboratory evaluation revealed hyponatremia, hyperkalemia, low serum cortisol, and elevated ACTH levels. A diagnosis of primary adrenal insufficiency was made. After discharge, he continued regular follow-up at the outpatient metabolic/endocrinology clinic.
During follow-up, persistent electrolyte imbalances, low cortisol levels, and elevated ACTH concentrations were noted. Brain MRI was performed to evaluate for possible pituitary abnormalities; however, no significant abnormalities were identified. The patient remains under continuous outpatient follow-up for monitoring and management.

Discussion

Primary adrenal insufficiency most commonly occurs in the context of autoimmune diseases, infections, adverse drug reactions, or following adrenal surgery. In the present case, the patient had previously undergone adrenalectomy and initially remained asymptomatic without clinical evidence of adrenal insufficiency during postoperative follow-up. However, symptoms subsequently developed after a later hospitalization for surgery.
During outpatient follow-up, persistently elevated ACTH levels were noted. Despite adjustment of corticosteroid therapy, no improvement was observed. Ectopic ACTH production was suspected; however, imaging studies failed to identify any obvious tumor lesions. Considering the overall clinical course, the patient was suspected to have ectopic ACTH syndrome of unknown origin.

Conclusion

Patients at risk of adrenal insufficiency require long-term and close follow-up. When clinically indicated, biochemical testing should be performed to confirm the diagnosis, and early initiation

of glucocorticoid replacement therapy should be considered in the presence of suggestive symptoms. Furthermore, If the patient's clinical symptoms improve and there are no longer evident signs of adrenal insufficiency, a decline in ACTH levels would be expected. If ACTH levels do not decrease as anticipated, the possibility of ectopic ACTH syndrome should be considered.

病例報告 114_C007

革蘭氏陰性厭氧菌椎間盤炎合併骨水泥外漏及雙側腰大肌至右大腿膿瘍: 個案報告

Gram-negative Anaerobic Spondylodiscitis Associated with Cement Leakage and Bilateral Psoas Abscess Extending to the Right Thigh: A Case Report

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Introduction

Pyogenic spondylodiscitis caused by anaerobic organisms is uncommon and can present with extensive local and systemic complications.

Case Report

A 72-year-old woman with diabetes and dementia presented to the emergency department due to persistent low back pain with radiation to the right thigh after vertebroplasty for T12 compression fracture in a local hospital 6 days ago. She had a history of multilevel lumbar spondylolisthesis with spinal stenosis and prior posterior fusion from L2 to S2 four years earlier. Physical examination revealed painful swelling of the right thigh with impaired leg elevation. CT scan showed early L1/L2 spondylodiscitis with epidural abscess and cord compression (L1–L4), retroperitoneal and iliopsoas/soft tissue abscesses with cellulitis/fasciitis extending to the right hip, thigh and knee, L1 osteonecrosis and cement leakage. Spine MRI demonstrated multiloculated ventral epidural abscesses from T3 to L2 causing cord/cauda equina indentation, infectious spondylodiscitis at L1/L2, bilateral psoas abscesses, and a left paraspinal muscle abscess at T10–T11. She received antibiotic therapy and underwent emergency fasciotomy of the right thigh, which revealed massive malodorous pus at the deep fascia and inter-/intramuscular spaces of the anterior, medial, and posterior thigh compartments. Pus culture grew *Prevotella intermedia*. The following day, image-guided percutaneous drainage of the left psoas abscess was performed. One week later, she underwent repeat debridement of the right medial thigh and single-portal endoscopic discectomy with debridement, revealing necrotic intervertebral disc tissue at L1-L2. She improved clinically, regained the ability to raise her legs, and was referred for treatment of chronic periodontitis.

Discussion

This case illustrates the fulminant course of Gram-negative anaerobic spondylodiscitis, an uncommon but severe infection that may complicate vertebral procedures. Although cement leakage following vertebroplasty can predispose patients to secondary spinal infection through disruption of local tissues, the identification of *Prevotella intermedia*, an oral anaerobe, in this case suggests a hematogenous spread from a distant dental source rather than a direct consequence of the procedure. Its progression to epidural, psoas, and soft-tissue abscesses highlights the need for early recognition, broad-spectrum antimicrobial therapy, and timely surgical intervention.

Conclusion

Pyogenic spondylodiscitis should be considered in high-risk patients presenting with post-

vertebroplasty infection, while healthcare providers should remain aware of diverse possible etiologies. Aggressive multidisciplinary management combining antibiotics, surgical drainage, and endoscopic debridement is essential to improve outcomes.

病例報告 114_C008

胰臟癌誘發之第一型孤立性胃靜脈曲張：個案報告

Pancreatic Cancer-Induced Type 1 Isolated Gastric Varices: A Case Report

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Introduction

Gastroesophageal varices are dilated submucosal veins caused by portal hypertension. Type 1 isolated gastric varices (IGV1) account for a minority of cases but carry higher bleeding risk. Recognition of IGV1 without generalized portal hypertension warrants urgent evaluation for splenic vein disease and underlying malignancy.

Case Report

A 68-year-old man with a history of coronary artery disease (three-vessel disease status post stenting), heart failure with reduced ejection fraction, hyperuricemia, type 2 diabetes mellitus, hypertension, and hyperlipidemia presented with a 3-day history of melena and dizziness. He was on regular medications and routine outpatient follow-up. The patient denied recent use of analgesics, steroids, NSAIDs, or herbal medications.

Physical examination showed stable vital signs without pallor, icterus, or hepatosplenomegaly. Laboratory findings revealed mild anemia (hemoglobin 12.6 g/dL), thrombocytopenia (platelets $116 \times 10^3/\mu L$), and strongly positive stool occult blood (3+). Coagulation studies were normal.

Esophagogastroduodenoscopy (EGD) revealed type 1 isolated gastric varices (IGV1) without endoscopic features of portal hypertensive gastropathy (Figures 1A and 1B). No stigmata of active or recent bleeding were identified. The unexpected finding of IGV1 without evidence of portal hypertension led to further evaluation.

Based on the endoscopic finding of isolated gastric varices, abdominal computed tomography (CT) was performed to evaluate for regional venous obstruction. CT imaging revealed splenic vein thrombosis secondary to a 4.03 cm hypodense mass in the pancreatic tail with invasion of the splenic artery, splenic vein, and left adrenal gland (Figure 2). This finding explained the development of isolated gastric varices through sinistral portal hypertension.

Initial management focused on hemodynamic stabilization with terlipressin and a restrictive transfusion strategy. Afterward, endoscopic ultrasound-guided fine-needle biopsy of the pancreatic tail mass was performed without complication, and pathology confirmed adenocarcinoma. The patient was subsequently discharged and referred to oncology for staging and treatment planning.

Discussion

Unlike the majority of gastroesophageal varices arising from cirrhosis-related portal hypertension, IGV1 typically develops when the splenic vein becomes occluded, resulting in an increased pressure of these veins and the development of type 1 isolated gastric varices, reffered as "sinistral" portal hypertension secondary to splenic vein occlusion.

The management of pancreatic cancer-induced IGV1 encompasses dual therapeutic objectives:

stabilization of acute bleeding and definitive malignancy treatment. According to the 2016 AASLD guidelines, the management approach depends on the acuity of clinical presentation. For acute variceal hemorrhage, a restrictive transfusion strategy with a hemoglobin target of approximately 8 g/dL is recommended, along with the administration of empirical antibiotics. Non-selective beta-blockers remain the mainstay for primary prophylaxis against variceal bleeding. Nevertheless, curative treatment requires addressing the underlying pancreatic malignancy. For surgically resectable tumors, splenectomy, often combined with distal pancreatectomy, effectively resolves sinistral portal hypertension, thereby eliminating varices and preventing recurrent bleeding episodes.

Conclusion

This case highlights the importance of clinical suspicion for pancreatic malignancy when encountering IGV1. This vascular complication may represent the sole initial manifestation of occult cancer, making prompt recognition crucial for early diagnosis and timely intervention.

病例報告 114_C009

藥物誘發性膽汁鬱積後續出現敗血症相關性肝功能障礙，合併偶發含鐵血黃素沉著：病例報告

Drug-Induced Cholestasis Followed by Sepsis-Associated Liver Dysfunction with Incidental Hemosiderosis: A Case Report

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Introduction

Progressive jaundice in critically ill patients often involves multiple etiologies. We report a case of biphasic jaundice initially caused by drug-induced liver injury (DILI) and subsequently exacerbated by sepsis, with incidental hemosiderosis detected on MRI.

Case Report

A 58-year-old ventilator-dependent female with multiple comorbidities, including end-stage renal disease, developed progressive jaundice while being treated for pneumonia. Her total bilirubin escalated from 2.07 mg/dL to 10.53 mg/dL within five days of initiating cefoperazone/sulbactam. With negative viral and autoimmune markers and no evidence of biliary obstruction on CT, DILI was suspected, and the antibiotic was shifted to piperacillin/tazobactam and later discontinued. Despite this change, her bilirubin plateaued around 14 mg/dL, consistent with the known delayed recovery pattern of cefoperazone-induced cholestasis.

Twenty-one days following discharge to a respiratory care ward, the patient was readmitted with markedly worsening jaundice, presenting with total bilirubin of 32.10 mg/dL and direct bilirubin of 22.14 mg/dL. Initial vital signs demonstrated normal sinus rhythm at 96 beats per minute, blood pressure of 146/71 mmHg, and oxygen saturation of 93% on 40% FiO_2.

Comprehensive laboratory evaluation revealed significant abnormalities including leukocytosis (19,810/μL), anemia (hemoglobin 8.2 g/dL), coagulopathy with prolonged PT/INR and APTT, and elevated inflammatory markers (CRP 128.3 mg/L, procalcitonin 3.19 ng/mL). Hepatobiliary enzymes were markedly elevated, with gamma-glutamyl transferase at 384.3 U/L and alkaline phosphatase reaching 3,637 U/L. Chest imaging confirmed recurrent pneumonia, for which cefepime had been initiated seven days prior to admission.

Given the clinical presentation of progressive jaundice with suspected biliary obstruction, magnetic resonance cholangiopancreatography (MRCP) was performed. While this study again excluded biliary tract obstruction, it unexpectedly revealed previously undetected hepatic and splenic hemosiderosis. Subsequent iron studies and hematological evaluation were undertaken for comprehensive assessment.

Peripheral blood smear examination demonstrated target cells (2+), spherocytes (1+), and schistocytes (1+), with the predominance of target cells being characteristic of severe cholestasis. Laboratory findings included extreme hyperferritinemia >33,000 ng/mL and a reticulocyte count of 2.70%. Notably, the patient's transfusion history was minimal, having received only four units of packed red blood cells and ten units of cryoprecipitate in the period from CT to MRI imaging.

Discussion

This case illustrates the complex interplay between DILI and sepsis causing severe cholestasis. The initial five-fold bilirubin elevation within five days strongly implicates cefoperazone/sulbactam, which inhibits bile salt export pumps and causes prolonged cholestasis persisting weeks after discontinuation. Recognition of this delayed recovery pattern is critical to avoid unnecessary interventions.

The hemosiderosis discovered on MRI, despite minimal transfusion exposure and absence on prior CT, likely represents pre-existing secondary hemosiderosis from cholestasis-induced reticuloendothelial iron dysregulation and chronic transfusional siderosis. This finding underscores fundamental differences in imaging sensitivity: MRI's T2-weighted sequences exploit hemosiderin's paramagnetic properties, while CT substantially underestimates iron burden, potentially delaying recognition of this complication.

Conclusion

This case emphasizes recognizing the distinct patterns of drug-induced and sepsis-associated cholestasis, particularly cefoperazone's delayed resolution. Significant hemosiderosis in cholestatic patients may reflect underlying iron sequestration, for which MRI is the superior diagnostic modality.

病例報告 114_C010

頑固性低血鈉症伴肌纖顫—抗 LGI1 腦炎的診斷線索：病例報告

Refractory Hyponatremia with Myokymia as Diagnostic Clue in Anti-LGI1 Encephalitis: A Case Report

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Introduction

Anti-Leucine-rich glioma-inactivated 1 (LGI1) encephalitis is an autoimmune limbic encephalitis typically presenting with faciobrachial dystonic seizures and cognitive decline. Although hyponatremia occurs in over 60% of cases, its presentation as the primary severe manifestation remains uncommon and can delay diagnosis. We report a case where life-threatening hyponatremia preceded classic features, with myokymia providing the crucial diagnostic clue.

Case Report

A 56-year-old male presented with general malaise following one month of intermittent fevers, hiccups, and migratory arthralgia. Laboratory findings revealed severe hypo-osmotic hyponatremia (Na+ 104.4 mmol/L; osmolality 210 mOsm/kg) with low urine sodium (<20 mmol/L), refractory to standard treatment.

Progressive weakness and paresthesias prompted neurological consultation. Examination revealed preserved motor strength but subtle myokymia affecting perioral muscles and all limbs. Electromyography confirmed fasciculations, multiplets, and myokymic discharges, indicating peripheral nerve hyperexcitability.

Given these findings with refractory hyponatremia, autoimmune etiology was suspected. Serology confirmed anti-LGI1 antibodies, while cerebrospinal fluid analysis was negative, establishing the diagnosis.

Discussion

The severity of hyponatremia initially obscured the diagnosis, directing workup toward common etiologies and causing delay. The identification of myokymia proved pivotal. As a manifestation of peripheral nerve hyperexcitability strongly associated with voltage-gated potassium channel complex antibodies (including LGI1), myokymia should prompt consideration of anti-LGI1 encephalitis, particularly with concurrent limbic symptoms or unexplained hyponatremia.

Conclusion

Severe refractory hyponatremia can be the initial presentation of anti-LGI1 encephalitis. Clinicians should maintain high suspicion for this treatable condition and actively seek subtle signs of peripheral nerve hyperexcitability, such as myokymia, which may provide essential diagnostic clues.

病例報告 114_C011

頭頸部假性腫瘤作為 IgG4 相關疾病的臨床表現：病例報告

Head and Neck Pseudotumor as a Manifestation of IgG4-Related Disease: A Case Report

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Introduction

Immunoglobulin G4-related disease (IgG4-RD) is a chronic, systemic, immune-mediated condition that can affect multiple organs. Commonly involved sites include the salivary glands, lacrimal glands, periorbital tissues, pancreas, biliary tract, and retroperitoneum. Due to the lack of specific laboratory diagnostic markers, definitive diagnosis often requires histopathological analysis, which adds to the diagnostic challenge.

Case Report

An 82-year-old man who presented with a painless mass in the left neck detected during a routine follow-up at the gastroenterology clinic. Ultrasound imaging revealed multiple bilateral enlarged cervical lymph nodes, with the largest node measuring approximately 2.1 cm. Histopathological analysis revealed storiform fibrosis. Besides, Immunostaining showed dense infiltration of IgG4-positive plasma cells and IgG4+/IgG+ >60%. The patient had an elevated serum IgG4 level (315.9 mg/dL) and a background history of asthma, atopic dermatitis, and allergic rhinitis. Laboratory studies revealed centromere-pattern ANA positivity and p-ANCA positivity. Based on clinical, laboratory, and pathological findings, IgG4-RD was diagnosed. Treatment was initiated with prednisolone 5 mg twice daily and azathioprine 50 mg once daily.

Discussion

This case highlights the importance of including IgG4-RD in the differential diagnosis of cervical masses, particularly in patients with atypical histologic features or allergic predispositions. Early recognition is critical because appropriate immunomodulatory therapy can prevent unnecessary surgical or oncologic interventions. IgG4-RD is a systemic fibroinflammatory condition that can affect virtually any organ system. The absence of systemic symptoms and the presence of specific histopathologic features, including storiform fibrosis and an IgG4-rich infiltrate, are essential clues toward an accurate diagnosis. A hallmark feature is the infiltration of IgG4-positive plasma cells, typically with an $IgG4^+/IgG^+$ cell ratio >40% and an absolute count exceeding 10–30 cells per high-power field, depending on the tissue involved.

Imaging features are nonspecific but may show soft tissue masses, organ enlargement, or diffuse inflammatory changes. Involvement of multiple sites (e.g., pancreas, salivary glands, lymph nodes, retroperitoneum) is common in systemic disease, though localized presentations also occur.

Although the pathogenesis of IgG4-RD is not fully understood, it is believed to involve both Th2-dominant immune responses and regulatory T cell dysfunction. Many patients exhibit an atopic background such as history of asthma, atopic dermatitis, or allergic rhinitis. Sometimes, elevated serum IgE levels or eosinophilia were also noted. In the present case, the patient's allergic diathesis further supports the immunological basis of the disease.

The mainstay of treatment is glucocorticoids, which induce remission in most cases. However, relapses are common, and immunosuppressants such as azathioprine, mycophenolate mofetil, or rituximab may be used in refractory or relapsing disease. Close clinical follow-up is recommended to monitor for recurrence and assess for systemic involvement over time.

Conclusion

IgG4-RD may clinically and radiographically resemble malignant disease. Accurate diagnosis requires integration of clinical, radiologic, serologic, and histopathologic data. In this case, timely histological evaluation allowed for appropriate immunomodulatory therapy, sparing the patient from unnecessary treatment.

病例報告 114_C012

透析患者全瓣膜性心臟病合併進展性黃疸：不進行瓣膜置換的姑息療法能走多遠？

Pan-Valvular Heart Disease in a Dialysis Patient with Progressive Jaundice: How Far Can Palliative Measures Go Without Valve Replacement?

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Introduction

Pan-valvular heart disease involving both native and prosthetic valves is rare and challenging to manage, especially in frail patients with limited treatment options.

Case Report

A 74-year-old woman on dialysis with prior tricuspid and pulmonary bioprosthetic valve replacements presented with abdominal pain, diarrhea, dyspnea, and progressive jaundice. Echocardiography revealed severe aortic stenosis (AS) with pulmonary, and tricuspid bioprosthetic valve dysfunction. Right heart catheterization confirmed significant pressure gradients and low cardiac index. Due to financial constraints and the high surgical risk, transcatheter valve-in-valve procedures and surgical treatment were not pursued. Instead, balloon valvuloplasty was performed on all three valves, resulting in transient hemodynamic improvement and bilirubin reduction. The patient later died from complications.

Discussion

This case illustrates the balloon valvuloplasty as a palliative option in pan-valvular heart disease when definitive therapies are inaccessible. Such cases highlight a need for individualized, multidisciplinary care.

Conclusion

Evaluation of pan-valvular failure may be challenging. Balloon valvuloplasty can offer temporary relief in high-risk patients with pan-valvular failure.

病例報告 114_C013

淋巴水腫之謎：揭開下肢腫大的真相

Lymphedema's Enigma: Unraveling the Mystery of Leg Enlargement

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Introduction

Lymphedema is a chronic condition characterized by impaired lymphatic drainage, leading to progressive limb swelling. It commonly occurs following pelvic lymph node dissection and radiation therapy for gynecologic cancers, with an estimated incidence of 20%. Early recognition and diagnosis are critical to prevent irreversible tissue changes and functional impairment.

Case Report

A 60-year-old woman presented with progressive enlargement of the left lower extremity over six months. She had a history of left ovarian and endometrial adenocarcinoma diagnosed three years prior, treated with debulking surgery (including bilateral salpingo-oophorectomy, bilateral pelvic lymph node dissection, and omentectomy) followed by external beam radiation therapy (5040 Gy in 28 fractions) targeting pelvic and para-aortic lymph nodes, achieving complete remission.

One year after completing cancer treatment, she developed significant left leg swelling, approximately five times the size of the right leg. Physical examination revealed firm, non-pitting edema with intact distal pulses. Laboratory studies showed mildly decreased albumin, and echocardiography confirmed normal cardiac function. Duplex ultrasound excluded venous thrombosis. Lymphoscintigraphy demonstrated abrupt termination of tracer flow in the lower calf lymphatics and dermal backflow in the affected limb. Grade III lymphedema of the left lower extremity was diagnosed.

Discussion

Secondary lymphedema following gynecologic cancer treatment is a well-documented complication, often attributed to lymphatic disruption from surgery and radiation therapy. Early-stage lymphedema may present with soft, pitting edema, but chronic disease leads to fibrosis and non-pitting swelling. Lymphoscintigraphy remains a sensitive diagnostic modality, providing functional assessment of lymphatic flow, with characteristic findings such as delayed transport, absent nodal visualization, and dermal backflow.(2) Multidisciplinary management—including compression therapy, manual lymph drainage, exercise, and, in selected cases, microsurgical lymphatic reconstruction—is crucial to improving outcomes and quality of life.

Conclusion

This case highlights severe unilateral lymphedema as a late complication of gynecologic cancer treatment. Prompt recognition, imaging evaluation, and early intervention are essential to mitigate long-term morbidity.

病例報告 114_C014

克隆氏症患者以腸阻塞症狀為表現的迴腸腺癌：病例報告

Adenocarcinoma of the Terminal Ileum Presenting as Obstructive Symptoms in a Crohn's Disease Patient: A Case Report

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Introduction

Crohn's disease (CD) is a chronic inflammatory bowel disease with transmural involvement that may affect any part of the gastrointestinal tract. Its manifestations range from abdominal pain and diarrhea to fistulas and extraintestinal complications. We report a case of terminal ileal adenocarcinoma presenting with obstruction in a patient with longstanding CD history.

Case Report

A 69-year-old German engineer with a 30-year history of Crohn's disease, previously managed with segmental ileal resection, presented with recurrent intestinal obstruction characterized by postprandial abdominal pain, distention, and vomiting after years of asymptomatic remission without medication. Physical examinations revealed abdominal distension, previous surgical scar, normoactive bowel sounds, diffuse abdominal tenderness and rebounding tenderness. KUB showed dilated small bowel segments which was confirmed by CT scan. The patient's symptoms improved with nasogastric decompression. A Crohn's-related stricture was suspected, but initial colonoscopy was incomplete due to inadequate bowel preparation from obstruction. Subsequent ileocolonosocopy was repeated with evac enema preparation and waterjet cleansing during colonoscopy, and intestinal stenosis caused by circumflex inflammation was found at terminal ileum at about 5 cm proximal to ileocecal valve. As the scope could not pass through, an endoscopic biopsy was performed and histopathology disclosed intraepithelial high-grade neoplasia, consistent with invasive adenocarcinoma. MRI did not show other segments with stenosis or inflammation, or distant metastasis. The patient underwent right hemicolectomy with en bloc ileal resection, central vascular ligation, lymphadenectomy, partial bladder resection, and cholecystectomy. Pathology showed poorly differentiated mucinous adenocarcinoma (6.5 cm, pT2N0, G3, R0) with adjacent dysplasia and underlying Crohn's disease. Recovery was uneventful, and follow-up Magnetic resonance enterography revealed no residual disease or lymphadenopathy. The patient was in good condition 30 months after the operation. No small intestinal adenocarcinoma recurrence or active Crohn's disease was detected during follow-up.

Discussion

Ileal adenocarcinoma is clinically rare, but the risk increases in long-standing Crohn's disease, where strictures are more often due to inflammatory causes rather than malignancy. The strictures caused by chronic inflammation or malignancy are clinically indistinguishable without histopathology, making tissue-proven diagnosis essential. Symptoms do not always correlate with disease severity, as exemplified by patients with minimal Crohn's disease activity who present acutely with malignancy. Regular monitoring with biomarkers, endoscopy, and imaging—

including intestinal ultrasound—supports comprehensive Crohn's disease management.

Conclusion

Adenocarcinoma is a rare yet clinically significant complication of Crohn's disease, frequently presenting as intestinal obstruction. Given that inflammatory and malignant strictures are indistinguishable without histology, systematic surveillance with endoscopy and imaging is essential for early detection and improved outcomes.

病例報告 114_C015

小腸淋巴瘤以感染性腸炎為症狀表現：一例原發性空腸 T 細胞淋巴瘤

Small Intestinal Lymphoma Mimicking Infectious Enteritis: A Case of Primary Jejunal T-Cell Lymphoma

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Introduction

Primary gastrointestinal (GI) lymphoma is clinically rare but the most common extranodal lymphoma, with nonspecific clinical manifestations. Small intestinal involvement is uncommon and often mimics infection or inflammation, causing diagnostic delays. We present a case of primary small intestinal lymphoma initially presenting as acute abdominal pain resembling enteritis.

Case Report

A 64-year-old woman with a history of duodenal came to emergency room due to left lower quadrant (LLQ) pain for two days after seafood ingestion. Physical examinations showed normoactive bowel sounds, mild abdominal distension with LLQ tenderness, but without rebounding pain. No fever or chillness, no nausea or vomiting, no diarrhea or constipation, no body weight loss or fever were noted. Laboratory tests revealed leukocytosis with mildly elevated C-reactive protein. Abdominal CT was performed as an unusual clinical presentation of acute gastroenteritis and demonstrated segmental jejunal wall thickening with mild luminal narrowing. Empirical antibiotics were administered for suspected enteritis and she was admitted for further management. Fecal FilmArray GI panel was negative for Salmonella, Campylobacter, and E. coli. Though the patients' abdominal pain symptoms improved gradually, double balloon enteroscopy was suggested for evaluation of the small bowel abnormalities shown on the CT scan. Endoscopic evaluation disclosed a 4mm circumferential ulcer with stenosis at the jejunum, noted at about 50 cm from the pyloric ring, and biopsy revealed mature T-cell lymphoma, positive for CD2, CD3, CD7, TCR delta, CD20, and negative for CD4, CD5, CD8, CD56, TIA-1, BF1, TCL1, CD19. PAX5. Ki67 proliferation index was 50%. Further PET/CT showed involvement of the jejunum and mesenteric lymph nodes, consistent with Ann Arbor stage IIE disease. She was arranged with chemotherapy by hematologists and her lymphoma status improved after treatment.

Discussion

Gastrointestinal lymphomas are rare, with the stomach most commonly affected, followed by the small intestine and ileocecal region. Primary T-cell lymphoma of the small intestine is particularly uncommon. This case highlights the diagnostic challenge of differentiating intestinal lymphoma from infectious enteritis in patients with acute abdominal pain. Alarm symptoms such as weight loss, vomiting, hematemesis, melena, anemia, and perforation do not correlate with endoscopic findings. While most subtypes present with superficial or ulcerative lesions, aggressive forms like diffuse large B-cell lymphoma, mantle cell lymphoma, and T-cell lymphoma should be suspected when fungating or protruding lesions are seen. However, whether such lesions are caused by

inflammation or malignancy is still clinically indistinguishable without histopathology, making tissue-proven diagnosis essential. This emphasizes the need for timely biopsy and histopathologic confirmation for patients with suspicious endoscopic findings.

Conclusion

Primary gastrointestinal lymphomas are rare and may mimic infection or inflammation. Aggressive subtypes should be suspected when endoscopy shows fungating or ulcerative lesions, with histopathological analysis crucial for definitive diagnosis and timely treatment.

病例報告 114_C016

血清陰性型冷凝球蛋白腎炎：病例報告及文獻回顧

Seronegative Cryoglobulinemic Glomerulonephritis: A Case Report and Literature Review

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Introduction

Cryoglobulinemia is a disorder characterized by circulating immunoglobins which spontaneously precipitate at low temperature. It is classified into three types. Type I cryoglobulinemia consists of a single monoclonal immunoglobin and is associated with lymphoproliferative disorders. Type II and III cryoglobulinemia are referred as mixed cryoglobulinemia and caused by viral infection, autoimmune diseases, or lymphoproliferative disorders. It can affect multiple organ systems, including the skin, joints, peripheral nerves, and kidneys. Renal involvement is associated with a poor prognostic factor and occurs in 10-40% of patients.

Case Report

We report a case of a 59-year-old man with a history of hypertension, diabetes mellitus, anemia, leukopenia, dyslipidemia, strokes, and proteinuria, who presented with progressive edema at bilateral legs for 1 month. Nephrotic-range proteinuria was detected, along with hypocomplementemia and elevated serum rheumatoid factor levels , in the absence of detectable serum cryoglobulins. Renal biopsy and pathological examination revealed immune complex-mediated glomerulonephritis with a diffuse membranoproliferative pattern of injury and pseudothrombi in the glomerular capillaries. Immunofluorescence staining showed granular IgM and IgG staining in glomerular capillaries and mesangium without light chain restriction. The presence of subendothelial and mesangial electron-dense deposits was demonstrated by the electron microscopy. Despite proper sample handling and repeated testing, serum cryoglobulins remained negative. A diagnosis of seronegative cryoglobulinemic glomerulonephritis was made, and prednisolone was initiated at a dose of 30 mg per day.

Discussion

Due to complexity of sample preparation and analysis, false-negative cryoglobulin test results are common. Nevertheless, seronegative cryoglobulinemic glomerulonephritis remains a recognized entity in clinical practice. According to Javaugue et al., patients with seropositive or seronegative cryoglobulinemic glomerulonephritis may present with clinical and pathological features. Seropositive patients are more likely to exhibit extrarenal manifestations, such as skin involvement. They also demonstrate positive rheumatoid factor and abnormal serum free light chain ratio more frequently. Under light microscopy, 59% of seronegative patients have diffuse glomerular intraluminal pseudothrombi, compared to 27% of seropositive patients. Additionally, polytypic IgM-dominant deposits were observed in 70% of seropositive cases, whereas monotypic deposits were found in 52% of seronegative cases, with light chain restriction more common among the latter. Regarding outcomes, seronegative patients had a higher complete response rate following first-line therapy, although 10-year renal survival rates did not significantly differ (70% in

seropositive vs. 82% in seronegative patients).

Conclusion

In patients with suspected glomerulonephritis, renal biopsy and pathologic exam are essential, especially in cases of seronegative cryoglobulinemic glomerulonephritis. There are distinct differences in clinical and histological features between seropositive and seronegative forms. However, current evidence is primarily derived from non-Asian populations, highlighting the need for further research.

病例報告 114_C017

成人史迪爾氏症案例報告

A Case Report of Adult-Onset Still's Disease

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Introduction

Adult-onset Still's disease (AOSD) is a rare, multisystem auto-inflammatory disease clinically characterized by spiking fever (>39–40°C), pink–salmon transient skin rash, and arthralgia or polyarthritis. Laboratory examinations usually show leukocytosis with neutrophilia, an elevation in inflammatory markers such as ferritin, C-reactive protein (CRP) and erythrocyte sedimentation rate (ESR).[1,2] AOSD presents a diagnostic challenge due to its overlap with various inflammatory or infectious condition.

Case Report

The 54-year-old woman with type 2 diabetes mellitus presented with spiking fever up to 40°C along with generalized skin rash for over 1 month since July, 2025. She also experienced arthralgia and joint swelling over bilateral knees, elbows and wrists. The laboratory examinations revealed leukocytosis with neutrophilia, elevated CRP, ferritin and lactate dehydrogenase (LDH). At first, she received antibiotics but in vain. The whole body positron emission tomography (PET) at 2025/7/7 reported multiple hyper-metabolic lymph nodes at neck, bilateral diaphragm, axillae and pelvis. The lymph node excisional biopsy was performed and showed reactive hyperplasia. The bone marrow biopsy reported myeloid hyperplasia, favor inflammatory reaction. The skin biopsy was done at 2025/7/16, the pathology reported superficial perivascular eosinophilic and neutrophilic infiltrates with upper epidermal dyskeratosis, AOSD was suggested. She started treatment with methotrexate, cyclosporine, colchicine and steroid. Due to persisted symptoms and elevation in inflammatory markers, self-paid Tocilizumab was administered. Her clinical condition has improved after treatment.

Discussion

The Yamaguchi criteria are used to diagnose AOSD. At least 5 criteria need to be fulfilled and 2 of it must be major criteria. Any infection, malignancy or autoimmune disease which can mimic AOSD must be excluded.[3] The pathogenesis of AOSD is still unclear, with both genetic and infectious triggers being implicated. First line treatment consists of non-steroidal anti-inflammatory drugs (NSAIDs) and systemic corticosteroids. If inflammation persists or relapses despite steroid use, disease-modifying anti-rheumatic drugs (DMARDs) like methotrexate or biological agents such as anakinra, tocilizumab can be used.[1,2]

Conclusion

AOSD is a rare condition that can be difficult to diagnose due to the clinical presentation has a variable differential diagnosis. Recent advances in diagnostic criteria and treatment strategies have significantly improved quality of life of these patients.

病例報告 114_C018

以高血鈣為初始表現之類肉瘤病一病例報告

Sarcoidosis with initial presentation of hypercalcemia: a case report

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Introduction

Hypercalcemia is a common metabolic disorder usually implying an underlying problem. Sarcoidosis can be one of the potential causes of hypercalcemia, which results from an overproduction of 1,25-dihydroxyvitamin D due to the nature of granulomatous disease.

Case Report

An 85-year-old Taiwanese woman presented with general weakness for one month, accompanied by generalized pain and confusion. The biochemical profile showed moderate hypercalcemia (total serum calcium, 12.5 mg/dL) but low intact parathyroid hormone (PTH) level (below 1.2 pg/mL) and low 25-hydroxyvitamin D level (7.3 ng/mL). There was no evidence to support the possibility of multiple myeloma. However, both chest CT scan and whole-body PET scan revealed multiple lymphadenopathies over neck, mediastinum, retroperitoneum, and pelvic cavity. For tissue acquisition, she underwent video-assisted thoracoscopic surgery with wedge resection of right upper lobe of lung and lymph node sampling by chest surgeon. The final pathology reported non-necrotizing granulomas distributed along bronchovascular bundles and pleura, and in lymph nodes. The result of nested polymerase chain reaction for Mycobacterium tuberculosis complex was negative. Based on the findings and consultation with the pulmonologist, she was diagnosed as sarcoidosis with pulmonary involvement, and then treated with oral methylprednisolone. Her clinical condition got improved and the serum calcium level also declined to the normal range.

Discussion

We reported a case initially presenting with hypercalcemia, and made the diagnosis of sarcoidosis through a comprehensive evaluation. Apart from electrolyte correction, it is crucial to identify the underlying etiology of hypercalcemia, which can be classified roughly into PTH-dependent and PTH-independent causes. Additional tests may be required for further differential diagnosis. Sarcoidosis makes up 1% of cases of hypercalcemia. The mechanism for hypercalcemia is considered to result from the nature of granulomatous diseases, that the production of 1,25-dihydroxyvitamin D increases due to unregulated 1α-hydroxylase expression of active macrophages. The principle of diagnosis is based on compatible clinical characteristics, identification of non-necrotizing granulomas, and exclusion of other causes of granulomatous disease. The primary treatment is glucocorticoid, focusing on suppression of inflammatory process. Successful management can relieve the patient's discomfort and normalize the calcium level.

Conclusion

This case illustrates the importance of identifying the underlying etiology of hypercalcemia. Sarcoidosis is a multisystemic granulomatous disease with a wide spectrum of clinical manifestations. Hypercalcemia can be one of characteristics but rarely as an initial presentation. It requires a multidisciplinary approach to improve the care of patients.

病例報告 114_C019

Upadacitinib 治療 Cluster 5 型大血管巨細胞動脈炎之病例報告

Case Report: Upadacitinib in Cluster 5 Large-Vessel Giant Cell Arteritis

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Introduction

Giant cell arteritis (GCA) is classically characterized by cranial manifestations such as headache, jaw claudication, and visual loss. However, large-vessel involvement is increasingly detected using positron emission tomography/computed tomography (PET/CT), establishing large-vessel GCA (LV-GCA) as a distinct phenotype. Patients may present with systemic inflammation or ischemic symptoms without cranial features. We report a case of LV-GCA corresponding to Cluster 5 of the recently described arterial distribution patterns, successfully managed with upadacitinib as a novel steroid-sparing therapy.

Case Report

A 62-year-old woman with hypothyroidism and hypertension presented with persistent fever, chest discomfort, and thigh pain. Extensive infectious, autoimmune, and malignancy evaluations were unrevealing, but inflammatory markers remained elevated. PET/CT revealed increased uptake along the vascular bundles of the lower limbs, with only transient non-steroidal anti-inflammatory drugs (NSAIDs) response.

Nine months later, she developed neck tightness, left upper limb numbness, chest and shoulder pain, pelvic discomfort, and absent blood pressure in the left arm. Repeat PET/CT demonstrated fluorodeoxyglucose (FDG) uptake in the bilateral carotid and subclavian arteries, aortic arch, thoracoabdominal aorta, and iliac arteries. Magnetic Resonance Angiography (MRA) confirmed bilateral carotid stenosis, 50–60% stenosis of the left subclavian artery, and >70% stenosis of the left vertebral artery, while temporal artery ultrasound was unremarkable. Despite the absence of cranial symptoms, the findings supported LV-GCA.

She was treated with intravenous pulse corticosteroids, transitioned to tapering oral prednisolone, and initiated on methotrexate and upadacitinib. Follow-up showed resolution of systemic symptoms, normalization of blood pressure in the left arm, and marked improvement in inflammatory markers.

Discussion

This case illustrates the diagnostic challenge of LV-GCA, which may present without cranial manifestations and mimic infection or malignancy when systemic inflammation predominates. The vascular distribution corresponded to Cluster 5 of the international cohort analysis by Gribbons et al., characterized by diffuse aortic and arch vessel involvement and strongly associated with GCA rather than Takayasu arteritis. Treatment with upadacitinib led to marked improvement. Upadacitinib, a selective Janus kinase 1 (JAK1) inhibitor, demonstrated efficacy in the phase 3 SELECT-GCA trial, achieving sustained remission and reducing cumulative glucocorticoid exposure, highlighting its promise as a steroid-sparing option in LV-GCA.

Conclusion

This case emphasizes that LV-GCA can occur without cranial features, with Cluster 5 distribution supporting diagnosis, and highlights upadacitinib as a promising steroid-sparing therapy.

病例報告 114_C020

以突發性視力受損表現之巨細胞動脈炎：病例報告

Giant cell arteritis presenting with sudden visual loss: A case report

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Introduction

Giant cell arteritis (GCA) is a severe ischemic vasculitis that still leads to irreversible visual loss in about 20% of patients despite improved diagnostics. Recent advances, notably in temporal artery ultrasound, magnetic resonance angiography and immunosuppressants, aim to reduce diagnostic delay and improve ophthalmologic outcomes.

Case Report

A 76-year-old man with a history of hypertension, heart failure with preserved ejection fraction, atrial fibrillation, and type 2 diabetes mellitus presented with abdominal fullness, decreased appetite, and chronic intermittent diarrhea. He was admitted with suspected infectious diarrhea. Laboratory tests revealed leukocytosis, an elevated C-reactive protein (CRP) level (30.95 mg/dL), and a markedly elevated erythrocyte sedimentation rate (ESR) (92 mm/hr).

During hospitalization, his diarrhea gradually subsided, but he developed persistent fever, and both CRP and ESR remained elevated despite broad-spectrum antimicrobial therapy. On the 10th day of hospitalization, he experienced sudden vision loss in the right eye. Ophthalmologist was consulted, and further evaluation demonstrated disc edema and cilioretinal artery occlusion, while fluorescein angiography confirmed hypoperfusion consistent with arteritic anterior ischemic optic neuropathy. Ultrasonography showed bilateral temporal artery wall thickening with hypoechoic halos and positive compression signs. Magnetic resonance angiography revealed thickened walls and severe distal narrowing of the superficial temporal arteries, more pronounced on the right side.

Based on these findings, a diagnosis of GCA was established according to the 2022 ACR/EULAR classification criteria. High-dose intravenous methylprednisolone with 750 mg daily (1 mg/kg/day) for three days was initiated, followed by tapering to oral prednisolone. His inflammatory markers improved, fever subsided, and ophthalmologic follow-up demonstrated partial recovery of visual acuity with improvement of disc edema. He was discharged on oral corticosteroids and subsequently commenced subcutaneous tocilizumab every four weeks as maintenance therapy. At follow-up, he remained clinically stable without new ischemic complications.

Discussion

Visual loss in GCA remains a serious complication, with recent cohort studies reporting permanent vision loss in up to 13–20% of patients, most often due to arteritic anterior ischemic optic neuropathy. Recovery of vision after onset is rare, and profound initial deficits usually predict poor prognosis. Tocilizumab has reduced relapse and steroid dependence in GCA, but its impact on visual outcomes remains uncertain. In our patient, despite immediate corticosteroid pulse therapy and subsequent tocilizumab, only partial recovery of visual acuity was achieved, consistent with

existing literature. This underscores the need for prompt recognition of visual symptoms and rapid initiation of high-dose steroids to prevent irreversible optic nerve damage.

Conclusion

This case illustrates the diagnostic challenges of GCA presenting with nonspecific systemic symptoms before visual involvement. Multimodality imaging and ophthalmologic assessment were critical in confirming the diagnosis. Early corticosteroid therapy stabilized the disease, and tocilizumab was introduced for long-term control. Clinicians should remain vigilant for GCA in elderly patients with unexplained fever and inflammation, as timely intervention remains key to preserving vision.

病例報告 114_C021

雙重免疫檢查點抑制合併 EGFR 單株抗體用微衛星穩定型轉移性十二指腸腺癌治療達到完全緩解

Durable complete response with dual immunotherapy and EGFR monoclonal antibody in microsatellite-stable metastatic duodenal adenocarcinoma

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Introduction

Small bowel adenocarcinoma is a rare gastrointestinal malignancy with limited treatment options and worse outcomes than colorectal cancer. While immune checkpoint inhibitors (ICIs) showed impressive result in mismatch repair–deficient (dMMR)/microsatellite instability–high (MSI-H) tumors, microsatellite-stable (MSS) disease is traditionally considered resistant to immunotherapy.

Case Report

We describe a 74-year-old male with MSS duodenal adenocarcinoma who experienced recurrence after curative surgery and failure of subsequent chemotherapy. Molecular profiling by liquid biopsy confirmed MSS status with low tumor mutational burden. Given the lack of standard alternatives, he was treated with nivolumab, ipilimumab, and cetuximab. Tumor regression occurred within 3 months, with complete metabolic response confirmed by positron emission tomography (PET) at 12 months. immune-related adverse events included adrenal insufficiency, vitiligo, and grade 1 pneumonitis, were noted but all manageable. The patient has remained disease-free for 20 months after treatment cessation.

Discussion

This is the first reported case of the potential synergy of dual checkpoint blockade and EGFR inhibition in MSS duodenal adenocarcinoma. Nivolumab and ipilimumab enhance T-cell priming and restore exhausted effector cells, while cetuximab promotes antibody-dependent cytotoxicity and dendritic cell activation, facilitating immune infiltration. The development of vitiligo in this patient may reflect enhanced antitumor immune activity.

Conclusion

This observation provides a proof of concept that immunotherapy resistance in MSS gastrointestinal cancers may be overcome by rational combination strategies, underscoring the need for prospective clinical trials to confirm efficacy and safety.

病例報告 114_C022

異質性乳糜微粒血症候群之脂蛋白脂肪酶基因第五外顯子 C243G 錯義突變：病例報告

Missense Mutation C243G in Exon 5 of Lipoprotein Lipase Gene in a Woman with Multifactorial Chylomicronemia Syndrome: A Case Report

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Introduction

Severe hypertriglyceridemia (SHTG), defined as fasting triglycerides ≧500–1000 mg/dL, increases risk of pancreatitis. Multifactorial chylomicronemia syndrome (MCS) is a common cause of SHTG, and most commonly stems from defects in lipoprotein lipase (LPL) gene, combined with secondary factors such as diabetes, obesity, and alcohol use.

Case Report

We present a case of MCS with LPL gene mutation and diabetes.

Discussion

A 25-year-old woman with SHTG since adolescence, recurrent pancreatitis, and insulin-requiring diabetes presented with progressive abdominal distension. Imaging revealed cirrhosis, splenomegaly, portal hypertension, and bloody ascites. Extensive workup excluded infection and malignancy. Ascitic findings suggested old hemorrhage. During admission, triglycerides gradually reduced after adherence to a non-fat diet. Whole exon sequencing identified a novel, likely-pathogenic, missense mutation in the exon 5 of the LPL gene c.727T>G:p.Cys243Gly, supporting the diagnosis of MCS. This finding shed light on the underlying genetic component contributing to the patient's SHTG.

Conclusion

SHTG leads to recurrent pancreatitis and increases the risk of cirrhosis and diabetes. MCS is a common cause, often associated with LPL gene mutations. This case underscores the importance of comprehensive clinical, biochemical, and genetic evaluation in managing SHTG.

病例報告 114_C023

未癒合的瘰癧與隱藏的免疫缺陷：辨識抗干擾素-γ 自體抗體症候群

Unhealed Scrofuloderma and a Hidden Immunodeficiency: Identifying Anti–IFN-γ Autoantibody Syndrome

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Introduction

Nontuberculous mycobacteria (NTM) are important causes of lymphatic and cutaneous infections. Scrofuloderma, resulting from contiguous spread of mycobacterial lymphadenitis, typically presents with subcutaneous nodules that undergo necrosis and drainage, leading to ulceration and scarring. In immunocompetent hosts, prolonged multidrug therapy is usually effective, with surgery rarely required. When disease persists despite adequate therapy, an underlying immune defect should be considered. Anti–interferon-γ (IFN-γ) autoantibody syndrome is an emerging adult-onset immunodeficiency, particularly in Southeast Asia, in which neutralizing antibodies impair cellular immunity and predispose to refractory or disseminated NTM infection.

Case Report

A 51-year-old woman originally from Indonesia, residing in Taiwan, had no known underlying disease. Two years prior to the current presentation, she developed painful vesicular lesions on the left eyelid and a submandibular nodule. Biopsy showed suppurative inflammation, and cultures identified Mycobacterium avium complex (MAC). Despite treatment with clarithromycin, rifabutin, and ethambutol, along with intravenous amikacin, lesions persisted and progressed, presenting as multiple erythematous to violaceous indurated plaques and nodules with ulceration, crusting, and seropurulent discharge on the left periorbital and malar region. Repeated biopsies and acid-fast stains confirmed refractory MAC infection. Evaluation for immunodeficiency was initiated. A QuantiFERON-TB Gold test was indeterminate with a low mitogen response. Functional neutralization assay confirmed anti–IFN-γ autoantibodies, establishing the diagnosis of adult-onset anti–IFN-γ autoantibody syndrome. Rituximab was started in addition to multidrug antimycobacterial treatment.

Discussion

This case illustrates refractory NTM infection presenting as scrofuloderma, ultimately diagnosed as anti–IFN-γ autoantibody syndrome. In otherwise healthy hosts, scrofuloderma responds to multidrug therapy, with surgery only rarely required. Lack of improvement should raise suspicion of impaired host defense.

Anti–IFN-γ autoantibody syndrome is an increasingly recognized adult-onset immunodeficiency, especially in Asia. Patients are HIV-negative and previously healthy before developing severe, recurrent, or disseminated infections. Neutralizing antibodies block IFN-γ, impair JAK–STAT signaling, and compromise macrophage function, predisposing to NTM, Salmonella, and other opportunistic pathogens. Clinical features include lymphadenitis, pulmonary disease, osteoarticular involvement, and cutaneous lesions such as scrofuloderma.

Diagnosis requires a high index of suspicion. Microbiology identifies the pathogen but not the immune defect. An indeterminate interferon-γ release assay (IGRA) with low mitogen response may serve as an early clue, but functional assays are essential for confirmation. Demonstrating IFN-γ neutralization distinguishes pathogenic antibodies from incidental findings.
Management is challenging. Antimycobacterial therapy is indispensable but often insufficient. Immunomodulatory strategies, particularly rituximab (anti-CD20 antibody), have shown benefit in reducing antibody levels and relapse risk, whereas cyclophosphamide and corticosteroids yield less consistent results.

Conclusion

Refractory NTM infection should prompt evaluation for hidden immunodeficiency. Anti–IFN-γ autoantibody syndrome is an underrecognized cause of treatment-resistant disease. Functional neutralization assays provide definitive confirmation, while indeterminate IGRA results with low mitogen responses may serve as a screening clue. Recognition is essential, as management requires both prolonged antimycobacterial therapy and immunomodulatory treatment.

病例報告 114_C024

腎臟血管脂肪瘤併發感染性膿瘍：病例報告

Abscess Formation within Renal Angiomyolipoma: A Case Report

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Introduction

Renal angiomyolipomas (AMLs) are mesenchymal neoplasms composed of epithelioid cells, smooth muscle cells, and adipocytes. While most AMLs remain asymptomatic, some may present with hemorrhage, abdominal or flank pain, hematuria, or impaired kidney function. Infection is an exceedingly rare complication. To our knowledge, abscess formation within a sporadic renal AML has not been previously reported. We describe such a case here.

Case Report

A previously healthy 60-year-old man presented with progressive right flank pain and dysuria for two months, followed by fever during the preceding two weeks. He attended the nephrology outpatient clinic in July 2025. On physical examination, there was no tenderness in the costovertebral angle. He was admitted with the presumptive diagnosis of acute pyelonephritis.

On admission, laboratory tests revealed leukocytosis (white blood cell count: 11340/μL) with neutrophil predominance (85.1%), elevated C-reactive protein (12.05 mg/dL), hyperglycemia (blood glucose: 306 mg/dL), and preserved kidney function (serum creatinine: 0.77 mg/dL; estimated glomerular filtration rate: 102.49 mL/min/1.73m2). Urinalysis demonstrated pyuria (50-99/high-power field). Empirical intravenous ertapenem was initiated. Urine culture yielded a small colony count (10^3 CFU/mL) of gram-positive cocci. In view of his clinical improvement, ertapenem was continued. Hemoglobin A1c was markedly elevated at 15.7%, and treatment with insulin and oral hypoglycemic agents was commenced.

Given the atypical course of urinary tract infection, abdominal sonography was performed, revealing a 6.3-cm exophytic heterogeneous lesion arising from the right kidney. Further computed tomography (CT) demonstrated a 5.7 × 3.5 × 8.5 cm mass with rim enhancement and fatty components in the right kidney, suggestive of an infected AML. A CT-guided pigtail drainage was performed, yielding milky fluid, and the culture of the aspirated fluid demonstrated heavy growth of *Staphylococcus aureus*. Antibiotic therapy was switched to intravenous oxacillin.

Following the above treatment, the patient's fever, dysuria, and flank pain gradually resolved, and laboratory markers of infection and glycemic control improved. He was discharged with an indwelling pigtail catheter, oral dicloxacillin, and antidiabetic medication.

Discussion

Reports of infected renal AMLs are sparse in the current literature. In our case, the patient was found to have previously undiagnosed type 2 diabetes mellitus and presented with a renal abscess within a large sporadic renal AML. Poorly controlled diabetes mellitus may have been a

contributing risk factor for abscess formation within the AML. In addition, the patient worked as a plumber, and frequent skin abrasions associated with his occupation may have predisposed him to recurrent transient *Staphylococcus aureus* bacteremia, which could have seeded the renal abscess. Further follow-up will be required to assess the need for an extended course of antibiotic therapy, continued imaging surveillance, and potential requirement for surgical intervention.

Conclusion

Abscess formation within renal AMLs is a rare but clinically significant complication. Optimal management requires not only appropriate drainage and antibiotic therapy, but also correction of underlying conditions such as diabetes mellitus. This case highlights the importance of considering infection in renal AMLs and the need for comprehensive treatment strategies to achieve favorable outcomes.

病例報告 114_C025

反覆免疫性血小板低下紫斑症作為 IgG4 相關疾病的表現：一例病例報告

Recurrent Immune Thrombocytopenic Purpura as Manifestation of IgG4-Related Disease: A case report

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Introduction

IgG4-Related Disease (IgG4-RD) is a systemic fibroinflammatory condition that involves multiple organs, including the pancreas, salivary glands, biliary tree, kidneys, lungs, retroperitoneum, and lymph nodes. It often presents with organomegaly or mass lesions and can mimic malignancies. Hematological abnormalities are frequent and include polyclonal hypergammaglobulinemia, eosinophilia, and lymphadenopathy. However, manifestations like Immune Thrombocytopenic Purpura (ITP) are rare and not well-characterized.

Case Report

We describe a 55-year-old male with a history of chronic HBV infection, managed with antiviral therapy, initially presented with obstructive jaundice, with imaging suggesting autoimmune pancreatitis (AIP), complicated by splenic vein thrombosis and retroperitoneal fibrosis resulting biliary dilatation and right-sided hydronephrosis. However, an EUS-guided biopsy was inconclusive for IgG4-RD. A plastic stent and a double-J(DJ) stent were inserted to address the obstruction, and he was also treated with oral glucocorticoids due to high suspicion of IgG-RD. Subsequently, the patient developed isolated thrombocytopenia on two separate occasions, requiring hospital admission. The first episode was complicated by *H. pylori*-positive gastric ulcers. His platelet count was responsive to oral glucocorticoids and HP eradication treatment. However, the second episode was severe, presenting with petechiae and hematuria. He initially responded robustly to high-dose glucocorticoids and pulse therapy but relapsed upon tapering. He was eventually prescribed with other immunosuppressants including azathioprine and cyclosporine as well as thrombopoietin receptor agonists (eltrombopag), which successfully stabilized his platelet count.

Discussion

This case underscores the well-documented diagnostic challenges of IgG4-RD and highlights a rare but serious association with severe, relapsing ITP. The patient exhibited characteristic features including autoimmune pancreatitis and retroperitoneal fibrosis, yet definitive histological confirmation remained elusive—a common dilemma in IgG4-RD diagnosis due to the patchy nature of inflammatory infiltrates and sampling limitations. The concomitant splenic vein thrombosis further reflects the pro-fibrotic and vasculopathic tendencies of advanced disease.

The emergence of ITP represents an unusual hematologic manifestation, suggesting shared immunopathogenic mechanisms. Dysregulated T-cell and B-cell interactions, central to IgG4-RD pathophysiology, may facilitate loss of self-tolerance and generation of platelet autoantibodies. The clinical course, marked by steroid dependence and relapse upon tapering, illustrates the

aggressive autoimmune dynamics in this context.
Management required a multifaceted strategy: conventional immunosuppressants (azathioprine, cyclosporine) modulated the underlying systemic autoimmunity, while eltrombopag directly promoted megakaryopoiesis, mitigating bleeding risk. This combined approach successfully addressed both the fibroinflammatory pathology and the life-threatening cytopenia, emphasizing the need for integrated, multidisciplinary care in managing complex manifestations of IgG4-RD.

Conclusion

This case highlights ITP as a serious though uncommon hematologic manifestation of IgG4-RD, reinforcing the diagnostic challenges posed by this condition even when classic clinical features are present. The successful management of this complex presentation required a multidisciplinary approach combining conventional immunosuppression with targeted thrombopoietin therapy to address both the underlying autoimmune process and life-threatening cytopenia. Our case underscores the importance of recognizing ITP as a potential complication of IgG4-RD and demonstrates the efficacy of combined immunomodulatory therapy, thereby expanding our understanding of the diverse clinical spectrum of this systemic fibroinflammatory disorder.

病例報告 114_C026

一例 76 歲女性，高血鈣與明顯升高的 iPTH，並有飢餓骨症候群風險

A 76-Year-Old Woman With Hypercalcemia, Markedly Elevated iPTH, and Risk of Hungry Bone Syndrome

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Introduction

Primary hyperparathyroidism is an endocrine disorder characterized by excessive secretion of parathyroid hormone (PTH) from an overactive parathyroid gland. This condition can lead to a wide spectrum of clinical manifestations, including osteoporosis, nephrolithiasis, chronic kidney disease, and cognitive impairment.

Case Report

We report the case of a 76-year-old woman with primary hyperparathyroidism who presented with marked hypercalcemia (13.2 mg/dL), hypophosphatemia (1.5 mg/dL), and severely elevated intact PTH (681 pg/mL) at admission. Tc-99m sestamibi scintigraphy localized an adenoma in the right inferior parathyroid gland. During surgery, the 1.5-cm right inferior parathyroid gland was excised. Histopathological examination confirmed the diagnosis of adenoma. Three days postoperatively, both serum calcium and iPTH levels had returned to within the normal range (calcium 9.2 mg/dL, iPTH 14.8 pg/mL); however, her serum phosphate level remained low (1.9 mg/dL).

Discussion

This case highlights several risk factors for the development of hungry bone syndrome (HBS), including severely elevated preoperative iPTH, increased alkaline phosphatase, and the relatively large volume of resected parathyroid tissue. Although our patient did not develop frank hypocalcemia in the early postoperative period, her persistently low serum phosphate level suggested a state of increased bone remineralization. Vitamin D supplementation is recommended in such patients to reduce the risk of HBS, improve postoperative mineral homeostasis, and facilitate recovery. Early recognition of high-risk patients is crucial for prevention and timely management of this complication.

Conclusion

We present a case of primary hyperparathyroidism in a 76-year-old woman with hypercalcemia, hypophosphatemia, and markedly elevated iPTH, in whom a parathyroid adenoma was successfully excised. The patient demonstrated multiple risk factors for HBS, underscoring the importance of vigilant biochemical monitoring and consideration of vitamin D supplementation in the perioperative period. This case emphasizes the need for awareness of HBS risk when treating patients with severe biochemical derangements due to primary hyperparathyroidism.

病例報告 114_C027

92 歲女性胰島素瘤病例報告：以認知退化為初始表現

Insulinoma in a 92-Year-Old Woman Presenting with Cognitive Decline

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Introduction

Insulinoma is a rare functional pancreatic neuroendocrine tumor that presents with recurrent hypoglycemia due to inappropriate insulin secretion.[1] Diagnosis is often delayed in elderly patients, in whom neuroglycopenic symptoms may mimic cognitive decline or dementia.[23] We report a case of an elderly woman with an insulinoma in the pancreatic head, who initially presented with progressive cognitive decline.

Case Report

A 92-year-old woman with hypertension treated with amlodipine and olmesartan had experienced progressive weakness and cognitive decline over two months, during which she became unable to recognize family members and became disoriented to time and place, according to her son. She was subsequently brought to the emergency department after an episode of loss of consciousness. Her fingerstick glucose level was 21 mg/dL, and her consciousness improved to a Glasgow Coma Scale score of E4V4M5 after intravenous dextrose. Brain computed tomography (CT) revealed no acute abnormalities, whereas abdominal CT showed a calcified nodule in the pancreatic head. Recurrent hypoglycemia occurred despite continuous dextrose infusion, and she was transferred to our hospital under the impression of severe hypoglycemia.

There was no history of diabetes mellitus, hypoglycemic agents, insulin use, or gastric bypass surgery. During hypoglycemia (53 mg/dL), endocrine testing revealed the following results: immunoreactive insulin, 116 μU/mL (reference range, 2.6-24.9); C-peptide, 12.7 ng/mL (reference range, 1.1-4.4); ketone body, 0.1 mmol/L (reference range, <0.6); cortisol, 11.9 μg/dL (reference range, 4.82-19.5); adrenocorticotropic hormone, 21.7 pg/mL (reference range, <46). The inappropriately elevated insulin and C-peptide levels in the setting of hypoglycemia indicated endogenous hyperinsulinism.

For localization of a potential insulinoma, arterial stimulation and venous sampling (ASVS) revealed positive findings in the gastroduodenal and hepatic arteries. Angiography demonstrated a tumor blush in the pancreatic head supplied by branches of the gastroduodenal artery. Endoscopic ultrasonography (EUS) revealed a nodule adjacent to the calcified area in the pancreatic head. Fine needle aspiration cytology (FNAC) confirmed a neuroendocrine tumor with insulin immunoexpression.

The patient underwent robotic pancreaticoduodenectomy. No further hypoglycemia occurred during the remainder of the hospitalization. Final pathological examination revealed a well-differentiated neuroendocrine tumor in the pancreatic head, grade 2, pT1N0 (stage I). She was discharged in stable condition on the 23rd postoperative day.

Discussion

The diagnosis of insulinoma relies on demonstrating inappropriately elevated insulin and C-peptide levels during hypoglycemia, as in this patient. Instead of adrenergic symptoms, such as tremor or palpitations, neuroglycopenia manifested as cognitive decline and disorientation, which initially resembled dementia.[45] This underscores the importance of considering hypoglycemia in elderly patients with new cognitive changes.

Surgery remains the only curative treatment for insulinoma. Although pancreaticoduodenectomy is rarely performed in very elderly patients, advances in perioperative care and minimally invasive techniques may allow safe resection in selected elderly patients.[6] This case demonstrates that advanced age alone should not preclude potentially curative surgery when functional status permits.

Conclusion

This case highlights that insulinoma, though rare, should be considered in elderly patients with unexplained cognitive impairment and recurrent hypoglycemia. Careful biochemical evaluation, targeted localization studies, and timely surgical management can achieve cure even in advanced age.

病例報告 114_C028

偶發性髓樣甲狀腺癌的長期生化進展與晚期結構性復發

Long-Term Biochemical Progression and Late Structural Recurrence in Sporadic Medullary Thyroid Carcinoma

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Introduction

Medullary thyroid carcinoma (MTC) is an uncommon neuroendocrine malignancy derived from parafollicular C-cells, accounting for 1–2% of thyroid cancers. Surgery is the primary curative approach, yet recurrence or persistence is frequent, requiring lifelong surveillance. Conventional imaging modalities such as ultrasound, computed tomography (CT), and magnetic resonance imaging (MRI) may remain negative despite rising tumor markers, a condition termed biochemical incomplete response. Serum calcitonin and carcinoembryonic antigen (CEA) are central in MTC follow-up, and their doubling times (DTs) provide important prognostic value—short DTs suggest aggressive disease, whereas longer DTs reflect indolent behavior.

Case Report

A 77-year-old Taiwanese woman who underwent total thyroidectomy in 2003 for a 2-cm sporadic MTC (pT1N0M0). RET germline testing was negative. Although she remained clinically stable postoperatively, serial monitoring revealed progressive biochemical elevation. Calcitonin rose gradually from 18 pg/mL in 2008 to >1200 pg/mL by 2020, while CEA increased from baseline 2.0 to 6.8 ng/mL. Despite repeated conventional imaging—including I-131 scintigraphy, FDG-PET/CT, and CT scans—no structural disease was detected for nearly two decades. Biomarker kinetics calculated up to 2022 revealed calcitonin DT of 821 days and CEA DT of 2108 days, consistent with indolent progression. In 2023, ^68Ga-DOTATATE PET/CT finally identified a right thyroid bed lesion, confirmed by ultrasound. Subsequent neck dissection demonstrated metastatic MTC in 2/10 lymph nodes, reclassified as rpT0N1bM0.

Discussion

MTC requires lifelong surveillance, as recurrence or persistence is frequent even after complete resection. Conventional imaging modalities such as ultrasound, CT, and MRI often fail to detect minimal disease, particularly in patients with persistently elevated biomarkers, a scenario termed biochemical incomplete response. Biomarker kinetics, especially DTs of calcitonin and CEA, provide critical prognostic insight: shorter DTs predict aggressive disease, while longer DTs indicate indolent progression. Functional imaging plays an increasingly important role in this context. ^{18}F-FDG PET/CT is useful for aggressive or dedifferentiated tumors, whereas ^{68}Ga-DOTATATE PET/CT demonstrates higher sensitivity in indolent, SSTR2-expressing disease.

Conclusion

This case highlights the complementary role of biomarker kinetics and functional imaging in MTC follow-up. Calcitonin and CEA DTs offered prognostic insight, while ^{68}Ga-DOTATATE PET/CT

enabled localization of recurrence when conventional modalities were unrevealing.

病例報告 114_C029

乾癬性關節炎成人患者使用 Adalimumab 後誘發之急性瀰漫性腦脊髓炎--病例報告

Adalimumab-Induced Acute Disseminated Encephalomyelitis in an Adult Patient with Psoriatic Arthritis: A Diagnostic Challenge

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Introduction

Acute disseminated encephalomyelitis (ADEM) is a rare neurologic disorder that masquerades infectious meningitis or encephalitis. We demonstrate a 50-year-old man who developed ADEM after 14 years of adalimumab treatment, underscoring the diagnostic challenge in clinical practice.

Case Report

A 50-year-old man with psoriatic arthritis with ankle arthritis and Achilles enthesitis had been receiving adalimumab from 2011 to 2025. He experienced two unexplored febrile seizures in 2022 and 2024, followed by a one-year history of bradyphrenia, memory decline, tremors, and a wide-based bradykinetic gait. In May 2025, he visited the emergency department with high fever, disturbed consciousness, and recurrent seizures.

He received cerebrospinal fluid (CSF) studies and microbial infections were excluded by negative results of bacterial, viral, fungal, and tuberculosis polymerase chain reactions and cultures. Rare infections, such as Japanese encephalitis, were also excluded by virus-specific IgM antibody testing and next-generation sequencing. Due to persistent symptoms, associated with lymphocytic pleocytosis, markedly elevated protein, mildly reduced glucose in CSF, and normal IgG indices, meningitis still could not be excluded. Empirical ceftriaxone, vancomycin, acyclovir and a standard anti-tuberculous treatment were initiated. Further surveillance including positron emission tomography/computed tomography scans and a lymph node biopsy from the right neck disclosed negative results for possible infections. Due to suspicion of autoimmune or limbic encephalitis, he underwent a magnetic resonance imaging (MRI) of the brain on 27 May, 2025 which revealed multifocal T2/FLAIR hyperintensities within the basal ganglia, thalami, mesial temporal lobes, ventral pons, and the dorsal aspect of the midbrain, without diffusion restriction. A follow-up brain MRI showed progressive lesions with new bilateral cerebellar peduncle involvement. Autoimmune markers including ANA, ENA, antiphospholipid antibodies, anti- aquaporin 4, anti-MOG, and antibodies for limbic encephalitis including anti-Hu, anti-Ma2, anti-NMDAR, were all negative. Given the progressive polyfocal neurological deficits, bilateral multiple deep grey matter lesions, tumor necrosis factor-α inhibitor (TNFi)-induced ADEM was suspected. He received intravenous methylprednisolone pulse therapy of (1 g daily for 5 days). Fever and seizures subsided within 48 hours, although cognition improved only modestly. He was treated with antiseizures with gradually tapered steroids, with partial recovery of neurologic symptoms.

Discussion

Patients under TNFi have an increased risk of demyelinating diseases, particularly multiple sclerosis and related syndromes including ADEM.[2,3] Most reported cases of TNFi–related

demyelination arise within the first year of treatment.[3] Our patient illustrates that late-onset ADEM demyelination remains possible. ADEM can masquerade as infectious meningoencephalitis, particularly in Taiwan where empirical anti-tuberculous therapy was recommended.[1] However, poor response to antibiotics and the typical neuroimaging raised suspicion for ADEM in our case. Prompt cessation of TNFi and high-dose corticosteroids with gradual tapering are recommended.[1] Outcomes of ADEM are good in children but less favorable in adults. Approximately 47-50% adult patients experience persistent neurological sequelae, and the mortality rate ranges from 8-12% in population-based studies.[4]

Conclusion

Our case highlights the importance of structural evaluation for ADEM in patients undergoing TNFi if they present with encephalopathy, lymphocytic CSF, failure to antimicrobial therapy, and progressive deep-grey-matter lesions on MRI. Timely discontinuation of TNFi followed by steroid treatment may improve the outcome of ADEM in such patients.

病例報告 114_C030

Mikulicz 氏症作為主要臨床表現的 IgG4 相關性疾病：一個病例報告

Mikulicz's Disease as a Primary Manifestation of IgG4-Related Disease: A Case Report

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Introduction

Mikulicz's disease, a distinct phenotype of IgG4-related disease (IgG4-RD), is characterized by symmetric, persistent, and painless enlargement of the lacrimal and major salivary glands. Herein, we present a case of Mikulicz's disease, highlighting its characteristic clinical and radiological features.

Case Report

A 55-year-old female with an unremarkable medical history presented to our outpatient clinic with a one-year history of progressively enlarging, bilateral, painless upper neck masses. Laboratory investigations showed elevated serum IgG at 2410 mg/dL and markedly elevated IgG4 subclass at 1101 mg/dL. Cervical ultrasonography showed reactive lymph nodes and enlarged submandibular glands containing hypervascular, hypoechoic nodules separated by hyperechoic septa. A diagnosis of Mikulicz's disease was made given her clinical, serological, and radiological findings. The patient was started on oral prednisolone and azathioprine.

Discussion

IgG4-related disease is a systemic fibroinflammatory condition that can affect nearly any organ system. The diagnosis of Mikulicz's disease is based on the 2023 revised criteria for IgG4-related dacryoadenitis and sialadenitis, incorporating clinical, serological, and histopathological features. Importantly, the diagnosis requires exclusion of mimicking conditions such as Sjögren's syndrome, sarcoidosis, and lymphoma. First-line treatment involves oral glucocorticoids, which are associated with high initial remission rates. To reduce the risk of relapse, steroid-sparing immunosuppressants may be employed. Biologic agents also represent a promising option for patients with relapsing or refractory Mikulicz's disease.

Conclusion

This case highlights a classic presentation of Mikulicz's disease as a primary manifestation of IgG4-RD. Definitive diagnosis depends on clinical presentation, characteristic serology, and the exclusion of mimics like Sjögren's syndrome and lymphoma. While glucocorticoids remain the mainstay for inducing remission, the high relapse rate and risk of steroid dependence underscore the importance of early use of steroid-sparing agents to achieve sustained disease control. Clinicians should maintain a high index of suspicion to enable timely diagnosis and comprehensive management.

病例報告 114_C031

超音波導引腎臟切片術後形成之腎動靜脈瘻管：以延遲性血尿為表現

Delayed Hematuria as an Underrecognized Symptom of Renal Arteriovenous Fistula After Ultrasound-guided Renal Biopsy

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Introduction

Ultrasound-guided renal biopsy is widely used due to its safety, yet complications such as bleeding, infection, or arteriovenous fistula (AVF) may occur. Post-biopsy AVF typically presents with hematuria or flank pain within 24 hours. We report a case of intra-renal AVF presenting with hematuria on day 5 post-biopsy. Diagnosis was achieved through multimodal imaging, including sonography, microflow imaging, computed tomography, and angiography. Coil embolization was performed, emphasizing the importance of timely recognition and intervention for delayed complications following renal biopsy.

Case Report

An 83-year-old man with hypertension and nephrotic syndrome complicated by acute kidney injury underwent ultrasound-guided renal biopsy, revealing membranous nephropathy. No immediate complications occurred. On day 5 post-biopsy, he developed painless gross hematuria, resistant hypertension, oliguria, progressive anemia (hemoglobin 10.5→8.2 g/dl), and worsening renal function (creatinine 6.15 → 8.83 mg/dl). Doppler ultrasound showed abnormal cortical vascular formation with pulsatile flow and a relatively low resistant index; microflow imaging demonstrated redundant flow at the lower pole of the right kidney. CT revealed enhancing nodules with early venous drainage, and angiography confirmed an arteriovenous fistula (AVF) from a small branch of the right renal artery. Transcatheter arterial embolization with coil and N-butyl cyanoacrylate successfully obliterated the AVF. Blood pressure normalized and hematuria resolved, but renal function did not recover, requiring long-term hemodialysis after discharge.

Discussion

Renal biopsy is essential in nephrology for diagnosis. Post-biopsy AVF incidence ranges from 0.1% to 18%, with most asymptomatic and resolving spontaneously, though some present with hematuria, flank pain, or refractory hypertension. AVF may form immediately if an artery and vein are punctured, or develop days to months later. In our case, hematuria and hypertension appeared on day 5, likely from progressive hemodynamic shifts. Imaging is crucial for diagnosis. Ultrasonography with Doppler typically reveals turbulent, pulsatile flow and arterialization of draining veins; microflow imaging (MFI) further delineates AVF morphology without contrast.

Contrast CT may show early venous enhancement, while digital subtraction angiography (DSA) remains the gold standard, providing vascular mapping and guiding therapy. Treatment aims to control symptoms, reduce hemodynamic effects, and preserve renal function. Endovascular embolization is first-line due to its minimally invasive nature, while surgery is reserved for complex or refractory cases.

Conclusion

We report a post-biopsy AVF with delayed hematuria and hypertension, underscoring the need for physician vigilance, early imaging, and timely intervention to prevent complications and improve outcomes.

病例報告 114_C032

暴發型困難梭狀桿菌感染與偽膜性大腸炎：糞菌移植後的臨床改善

Fulminant Clostridioides difficile Infection and Pseudomembranous Colitis: Clinical Recovery after Fecal Microbiota Transplantation

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Introduction

Clostridioides difficile infection (CDI) is a common cause of healthcare-associated diarrhea, with clinical manifestations ranging from mild colitis to life-threatening fulminant colitis. Despite standard antibiotic therapy, recurrent or refractory cases remain a major clinical challenge. Fecal microbiota transplantation (FMT) has emerged as an effective alternative treatment, particularly in patients with severe or recurrent CDI.

Case Report

We describe the case of a 58-year-old woman with stage IV pancreatic body adenocarcinoma and lung metastasis, undergoing chemotherapy, who presented with fever, diffuse abdominal pain, and watery diarrhea for several days. She had a history of CDI one month earlier, treated with a 10-day course of fidaxomicin.

At presentation to the emergency department, she was febrile (38.5°C), tachycardia (112 bpm), and hypotension (85/52 mmHg). Physical examination revealed diffuse abdominal tenderness and hypoactive bowel sounds. Laboratory studies demonstrated bandemia (6.5%), an elevated C-reactive protein level (24.31 mg/dL), and a markedly elevated lactate level (26.5 mg/dL). Abdominal CT showed diffuse, long-segment edematous wall thickening of the colon, extending from the ascending colon to the rectum. Stool assays for *C. difficile* toxin A/B and nucleic acid amplification were positive, confirming the diagnosis of fulminant CDI.

The patient received intravenous metronidazole (Day 1–8), oral vancomycin (Day 1–4), and oral fidaxomicin (Day 5–8). However, she continued to experience profuse watery diarrhea (6–9 times/day). Colonoscopy on hospital Day 11 revealed multiple yellowish plaques and shallow ulcers consistent with pseudomembranous colitis. Histology demonstrated acute and chronic inflammation with exudates, although classic pseudomembranes were absent. Fecal Microbiota Transplantation(FMT) was performed on Day 11 via terminal ileum infusion, after which her stool consistency improved steadily, reaching formed stools by Day 14.

Discussion

This case highlights fulminant CDI in an immunocompromised patient with advanced malignancy receiving chemotherapy. Conventional antibiotic regimens (metronidazole, vancomycin, and fidaxomicin) proved insufficient, and colonoscopy confirmed pseudomembranous colitis. FMT was successfully performed, resulting in rapid clinical recovery.

The pathogenesis of CDI involves disruption of the gut microbiota, toxin production (toxins A and B), and host susceptibility. Fulminant CDI is characterized by hypotension, shock, ileus, or toxic megacolon, and is associated with high mortality. Current guidelines recommend fidaxomicin or

vancomycin as first-line therapy, yet recurrence remains common. For patients with multiple recurrences or refractory disease, FMT is strongly recommended, with reported cure rates exceeding 80%.

Conclusion

We report a case of fulminant CDI complicated with pseudomembranous colitis successfully managed with FMT after antibiotic therapy failure. FMT should be considered early in selected patients with severe or recurrent CDI, particularly in those with high risk of treatment failure.

病例報告 114_C033

第二型 Amiodarone 誘發性甲狀腺毒症併發甲狀腺風暴之病例報告：血漿置換與手術治療之經驗

Amiodarone-Induced Thyrotoxicosis Type 2 Presenting with Thyroid Storm: Successful Management with Plasmapheresis and Thyroidectomy

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Introduction

Amiodarone-induced thyrotoxicosis (AIT) is a rare but severe complication of amiodarone therapy that may precipitate thyroid storm, particularly in patients with underlying cardiac disease. Differentiating between type 1 and type 2 AIT is essential for timely diagnosis and appropriate management.

Case Report

A 43-year-old man with a history of heart failure post-mitral valve repair, chronic kidney disease, atrial flutter post-ablation on amiodarone, and coronary artery disease presented with 3 days of dyspnea. At a local hospital, he developed altered consciousness, bradycardia, hypoglycemia, and shock requiring intubation and vasopressors. Laboratory findings showed leukocytosis, hepatic dysfunction, elevated procalcitonin and lactate, and thyroid storm (TSH<0.015 μIU/mL, free T4>6.99 ng/dL, T3 537 ng/dL, Burch-Wartofsky Point Scale 80 points). Propylthiouracil, hydrocortisone, and antibiotics were initiated before transfer. On admission, he remained comatose and febrile with severe thyrotoxicosis(TSH <0.005μIU/mL, T3: 350 ng/dL, free T4 >7.77ng/dL) and marked hepatic dysfunction. Carbimazole, propranolol, and hydrocortisone and antibiotics were given. Due to refractory thyroid storm, plasmapheresis was initiated, improving consciousness and liver function. Thyroid autoantibodies were negative, and ultrasound revealed no nodules. After eight sessions, total thyroidectomy was performed. Pathology demonstrated follicular destruction with fibrosis, foamy macrophage infiltration, and multinucleated giant cells, consistent with type 2 AIT. Postoperatively, antithyroid drugs were discontinued and levothyroxine started. The patient recovered well, with cardiac follow-up arranged.

Discussion

AIT arises from amiodarone's high iodine content and direct cytotoxicity on the thyroid. Type 1 AIT: Increased thyroid hormone synthesis due to iodine load. Type 2 AIT: Destructive thyroiditis releasing preformed hormone. Mixed forms: Combination of both mechanisms.

Type 2 AIT typically occurs in previously normal thyroid glands. The pathophysiology involves direct cytotoxic effects on thyroid follicular cells, leading to release of preformed thyroid hormones into the circulation, rather than increased synthesis.

Diagnostic clues include absent or low radioiodine uptake and lack of hypervascularity on Doppler ultrasound.

Glucocorticoids are first-line therapy, but refractory cases may require plasmapheresis and thyroidectomy, particularly when cardiac status deteriorates or amiodarone withdrawal is not feasible. Early recognition and timely intervention are essential to reduce morbidity and mortality.

Conclusion

This case illustrates the importance of early recognition and aggressive management of type 2 AIT complicated by thyroid storm. In steroid-refractory cases, plasmapheresis followed by thyroidectomy can be life-saving.

病例報告 114_C034

假性腸阻塞於紅斑性狼瘡的表現

Intestinal pseudo-obstruction in systemic lupus erythematosus

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Introduction

Gastrointestinal manifestations are uncommon but can be clinically significant in systemic lupus erythematosus (SLE). Intestinal pseudo-obstruction (IpsO) is defined as presence of clinical features of intestinal obstruction without identifiable mechanical obstructive lesion. Early diagnosis could avoid unnecessary surgical intervention, as SLE-related IpsO can be treated well by medication including corticosteroid and immunosuppressants.

Case Report

A 39-year-old Taiwanese woman carried a diagnosis of SLE since 2007. She also has hydroureteronephrosis attributed to neurogenic bladder with vesicoureteral reflux and lupus enteritis, which led to frequent admission in the period. One week after tapering an intravenous methylprednisolone regimen for lupus enteritis, she developed progressive abdominal distension, anorexia, nausea, and mild diffuse tenderness. KUB radiograph demonstrated marked bowel diffusely gaseous dilatation; computed tomography excluded adhesive or neoplastic obstruction. She was then treated with intravenous methylprednisolone, but the clinical response was limited. Intravenous cyclophosphamide and rituximab were administered. Both her clinical condition and serial KUB films showed improvement afterwards.

Discussion

Intestinal pseudo-obstruction is an uncommon but important complication of systemic lupus erythematosus that is often underrecognized or misdiagnosed. Proposed mechanisms include lupus vasculitis with ischemic injury and fibrosis of the intestinal wall, autoimmune neuropathy of the enteric nervous system, and primary smooth muscle myopathy. SLE-IPO frequently coexists with other visceral smooth-muscle disorders, such as uretero-hydronephrosis, suggesting the possible mechanism could be smooth muscle dysmotility.

Conclusion

Intestinal pseudo-obstruction represents a rare but severe gastrointestinal manifestation of systemic lupus erythematosus . Without timely recognition and intensive therapy, patients face substantial risks of recurrence, unnecessary surgery and mortality.

病例報告 114_C035

34 歲男性急性腎損傷併腎臟腫大：非常見病因之病例報告

Acute Kidney Injury with Enlarged Kidneys in a 34-Year-Old Male: Beyond the Usual Suspects

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Introduction

Acute kidney injury (AKI) accompanied by renal enlargement is uncommon and warrants careful evaluation. While common etiologies include acute pyelonephritis, obstructive uropathy, renal vein thrombosis, and deposition diseases, malignant renal infiltration, though rare, remains a critical consideration, particularly in hematologic malignancies such as lymphoma or leukemia.

Case Report

A previously healthy 34-year-old male presented with progressive gastrointestinal discomfort and unintentional weight loss for one month. Laboratory evaluation revealed AKI with macroalbuminuria. Contrast-enhanced abdominal computed tomography demonstrated bilateral renal enlargement with poor corticomedullary differentiation. These findings were suggestive of an infiltrative renal process, prompting further investigation to establish a definitive diagnosis.

Discussion

Infiltrative renal diseases account for fewer than 5% of AKI cases but are associated with poor outcomes, especially when secondary to hematologic malignancy. Failure to recognize the condition early may lead to irreversible renal damage despite initiation of therapy. Imaging modalities such as ultrasound and computed tomography are crucial for excluding more common etiologies and raising suspicion for infiltration. Ultimately, renal biopsy is indispensable for histopathological confirmation and guiding appropriate treatment.

Conclusion

Although rare, infiltrative renal diseases should be included in the differential diagnosis of AKI with renal enlargement. Early recognition and timely intervention are essential to preserve renal function and improve patient prognosis.

病例報告 114_C036

災難性抗磷脂症候群病例個案與治療挑戰

Catastrophic Antiphospholipid Syndrome - Case report and therapeutic challenge

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Introduction

Catastrophic Antiphospholipid Syndrome (CAPS) is a rare but life-threatening disease that needs multidiscipline experts intervention. The presentation of this disease, by definition, would involve at least 3 vital organs, and may be precipitated by certain risk factors (mostly infection). Here we are presenting a CAPS case with clinical therapeutic challenge despite of prompt intervention.

Case Report

We reported a case of 55-year-old male with chronic kidney disease (stage 5), antiphospholipid syndrome for 10 years, history of multiple ischemic stroke with encephalomalacia. At first, seizure for 15 minutes was found, and new episode of ischemic stroke was later confirmed by radiography. Moreover, profound septic shock along with acute on chronic kidney disease (CKD) , bicytopenia(anemia and thrombocytopenia) was seen. Lab data later revealed low haptoglobin, high LDH, and hyperbilirubinemia. PB smear showed 1+ schistocyte. TMA was impressed and favored aHUS due to ADAMTS13 later showed within normal range. We first administrated 6 times of plasmapheresis and sent gene analysis, which later disclosed no gene mutation, and thus Eculizumab was not given. Haptoglobin was gradually increased but thrombocytopenia remained 20k - 50k. Due to the fact that CKD in progression , bone marrow suppression (APS related ITP) and several episodes was CVA during this admission course, the criteria of Catastrophic Antiphospholipid Syndrome was met. We therefore decided another 5 times plasmapheresis , IVIG 0.4g/kg administration and Prednisolone 5mg BID. Nonetheless, new episode of subacute ICH was seen and thrombocytopenia persisted.

Discussion

Catastrophic Antiphospholipid Syndrome (CAPS) has a high mortality rate, exceeding 30% even with aggressive treatment. Early detection and prompt therapy are critical for improving survival. The leading causes of death are cerebrovascular, cardiac, and infectious complications, with systemic lupus erythematosus (SLE) linked to higher mortality.

Conclusion

In our case, we first arrange diagnosis by excluding the possible TMA first, and then give the proper management (glucocorticoids, immune globulin, plasma exchange). However, the outcome still limited improvement.

病例報告 114_C037

肝細胞癌合併腦部轉移之病例報告

Hepatocellular Carcinoma with Brain Metastasis: Report of a Case

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Introduction

Brain metastasis of hepatocellular carcinoma (HCC) is a rare condition with possible severe neurological symptoms, indicating poor prognosis and advanced disease. In this report, we describe a case of HCC with brain metastasis complicated with consciousness disturbance, headache, and obstructive hydrocephalus.

Case Report

A 42-year-old gentleman living in Myanmar with a history of chronic hepatitis B related liver cirrhosis and HCC, Barcelona Clinic Liver Cancer classification stage C, was admitted to our hospital in June 2023. Initial staging revealed locally advanced HCC with right portal vein branch invasion but no distant metastasis. After initial staging, he received four courses of transarterial chemoembolization (TACE) and targeted therapy with lenvatinib. With viable tumors in liver after the third course of TACE, the patient shifted lenvatinib to immune therapy with atezolizumab plus bevacizumab. Immunotherapy was terminated after seven courses due to disease progression, with distant metastasis to left adrenal gland, left inguinal skin and bilateral lungs. Cryoablation therapy for left adrenal gland metastasis and excisional biopsy for left inguinal skin metastasis were performed. Cabozantinib was prescribed for rescue but terminated due to disease progression in left adrenal gland and bilateral lung metastases. The second course of cryoablation therapy for left adrenal gland metastasis was arranged after imaging diagnosis. Prior to this admission, the most recent anti-cancer therapy is tremelimumab and durvalumab after fully discussion, completed three courses.

The patient developed progressive headache and vomiting one week before admission. Drowsiness was observed upon arriving at our hospital. The computed tomography of brain revealed one 4.7 cm mass lesion at left cerebellar hemisphere with ventricular compression. We consulted Neurosurgery department and external ventricular drainage was immediately performed to alleviate obstructive hydrocephalus. The magnetic resonance imaging of brain revealed one heterogeneous enhancing mass, 3.3 x 5.2 x 4.8 cm, at left cerebellar hemisphere, compressing the fourth ventricle and left brainstem. He received a subsequent operation of suboccipital craniectomy for cerebellar metastatic tumor removal under neuronavigation technique. The pathological diagnosis was metastatic carcinoma from hepatic origin. The patient recovered well after the operations. Post-operative re-staging revealed no residual tumor in brain, no viable HCC within liver, but progressive metastases in both lungs and left adrenal gland. We arranged second-line targeted therapy with regorafenib and cryoablation therapy for left adrenal metastasis.

Discussion

HCC with brain metastasis is extremely rare. Previous studies disclosed poor prognosis in these patients, with median survival time less than twelve weeks. For these patients with obstructive hydrocephalus, external ventricular drainage is lifesaving, and subsequent operation for brain tumor removal should be considered in selective patients. Radiotherapy and systemic therapy are alternatives.

Despite aggressive combination with immunotherapy, targeted therapy, and multiple modalities of locoregional treatment, our case presents extensive organ involvement in disease progression. Although he tolerated most surgical procedures and systemic therapy, response to current combination therapy is the most important factor determining life expectancy.

Conclusion

HCC with brain metastasis is a rare condition with drastic neurological deficits. A careful consideration of surgical candidacy is necessary, followed by individualized plan of combinational therapy.

病例報告 114_C038

苦樂參半的診斷:猛爆性第 1 型糖尿病併布魯蓋達氏表現

The Bittersweet Diagnosis: A Rare Encounter with Brugada pattern in Fulminant Type 1 Diabetes

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Introduction

Fulminant type 1 diabetes (FT1D) is a distinctive subtype of type 1 diabetes, characterized by rapid onset of absolute insulin deficiency, a shorter duration of hyperglycemic symptoms, greater degree of ketosis, negligible C-peptide secretion and absence of islet autoantibodies. Here, we describe a rare case of fulminant type 1 diabetes with Brugada pattern electrocardiogram at presentation.

Case Report

A 17-year-old girl presented with 5-days history of nausea, diffuse abdominal pain and disorientation.

She did not have a recent weight changes, preceding symptoms of flu-like symptoms, medication use, history of pancreatitis, syncope or a family history of diabetes and sudden cardiac death.

On examination, her blood pressure was 123/58 mmHg, pulse rate of 100 bpm, afebrile, respiratory rate was 18/min. Her body mass index was 23. Investigations showed plasma glucose of 1013 mg/dl, increased blood ketone of 3.8 mmol/l, serum sodium 118 mmol/l, potassium 5.6 mmol/l, serum creatinine 1.79 mg/dl and metabolic acidosis with raised anion gap of 28 mmol/l. Pregnancy test was negative.

Her electrocardiogram showed ST-elevations in V1-2 (Figure A), with a Shanghai score of 3.5 points, suggesting probable Brugada syndrome. Echocardiography showed no significant structural changes and ejection fraction was 68%. She was treated with intravenous insulin infusion and fluid replacement. Her ECG returned to sinus rhythm with complete recovery of consciousness after resolution of diabetic ketoacidosis and correction of serum electrolytes.

Additional studies revealed that her HbA1c was 6.5% and undetectable glutamic acid decarboxylase antibody (GAD). After her glucose levels stabilized, glucagon stimulation test showed a very low fasting C- peptide (0.15 ng/ml) and post-glucagon C-peptide (0.28 ng/ml).

A diagnosis of fulminant type 1 diabetes was made, and the patient was discharged with multiple daily insulin injections. Three months after discharge, her HbA1c increased to 7.8%. Her glycemic profile on follow-up was assessed by using continuous glucose monitoring (Figure B).

Conclusion

FT1D is a rare diabetes subtype (type 1b), with episodes of diabetic ketoacidosis may trigger transient Brugada patterns, raising concerns about the risk of arrhythmias and sudden cardiac events.

The Japanese Diabetes Society 2012 diagnostic criteria for FT1D are as follow: (1) diabetic ketosis

or ketoacidosis in < 7 days after onset of hyperglycemic symptoms, (2) initial plasma glucose≧288 mg/dl and HbA1c < 8.5%, (3) fasting serum C-peptide < 0.3 ng/ml and <0.5 ng/ml after glucagon (or post-meal).

Patients with FT1D face unique challenges in diagnosis and management due to the rapid and complete loss of insulin production. Greater awareness of this rare and underrecognized condition is essential to improve timely diagnosis and treatment.

病例報告 114_C039

68 歲女性腹部超音波檢查結果假陰性的胰臟癌個案報告

DELAY DIAGNOSIS OF PANCREATIC CANCER IN A 68-YEAR-OLD FEMALE WITH FALSE NEGATIVE ABDOMNAL SONOGRAPHY FINDINGS- A CASE REPORT

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Introduction

Pancreatic cancer is difficult to detect due to a lack of specific symptoms. Diagnosis of pancreatic tail cancer is particularly difficult because it is less common, less likely to cause jaundice than cancer of the pancreatic head, and difficult to detect on abdominal ultrasound.

Case Report

A 68-year-old female presented with progressive back and abdominal pain. Left shoulder and low back pain developed gradually around 2 months ago. Abdominal sonography didn't reveal significant abnormality on pancreas. She visited orthopedic clinic where L spine compression fracture was noted. But the symptoms worsened gradually, poor appetite, body weight loss also developed. She came to out-patient clinic and emergency department due to aggravated pain. Lab data revealed WBC 8150/ul (4000-11000); Seg 76%, HBsAg: reactive, AST/ALT: 37/60 U/L (<32), ALKP: 426 U/L (35-100), r-GT: 317 U/L (6-42), CEA: 160 ng/ml(<5), CA19-9: 37857 U/ml(<27). CA 125: 376 U/ml(<35). Abdominal CT revealed a 4.3 cm pancreatic tail mass with multiple hepatic metastasis, and lymphadenopathy involving celiac, SMA, and paraaortic regions. Endoscopic ultrasonography and ultrasound guided biopsy was done and the pathology revealed adenocarcinoma.

Discussion

It takes 3 months for this patient to receive the diagnosis and initiate the treatment. Diagnostic delays are common among patients with pancreatic cancer and are significantly associated with presentation at more advanced stages of disease.

Conclusion

Pancreatic cancer carries poor prognosis. Improved understanding of practice gaps that lead to diagnostic delays can inform future targeted interventions to reduce potentially preventable diagnostic delays.

病例報告 114_C040

腹膜上皮樣間皮瘤以不明原因腹水及肺部纖維化為表現：病例報告

Peritoneal Epithelioid Mesothelioma Presenting with Unexplained Ascites and Pulmonary Fibrosis: A Rare Case Report

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Introduction

Malignant peritoneal mesothelioma is a rare and aggressive malignancy originating from the mesothelial cells lining the peritoneum. Its clinical presentation is often diverse and non-specific, commonly including abdominal pain, distension, weight loss, or an abdominal mass. Although asbestos exposure is a well-established risk factor for mesothelioma, obtaining a clear history of such exposure can sometimes be challenging. We describe a unique case of peritoneal epithelioid mesothelioma, initially presenting with pulmonary fibrosis and concurrent refractory ascites, ultimately diagnosed through exploratory laparotomy.

Case Report

A 71-year-old bedridden male with a history of intracerebral hemorrhage and epilepsy was admitted due to persistent fever, cough, and progressive abdominal distension. He was initially diagnosed with community-acquired pneumonia. A chest computed tomography (CT) scan, arranged for suspected pulmonary fibrosis, revealed subpleural interstitial changes and diffuse fibrothorax, suggesting a usual interstitial pneumonia pattern with possible asbestos-related changes. However, due to the patient's condition, information regarding prior asbestos exposure could not be obtained. Despite receiving antibiotics, his fever and abdominal distension persisted and progressively worsened. A Gallium scan showed increased uptake in the pelvic region, prompting further investigation. An abdominal CT scan subsequently revealed a 6.4 cm irregular mass at the rectosigmoid junction, diffuse peritoneal thickening, significant ascites, and regional lymphadenopathy. Analysis of the ascites showed exudative fluid but no malignant cells or specific infectious etiology, including tuberculosis. To rule out gastrointestinal or genitourinary malignancies, colonoscopy and cystoscopy were performed, both yielding inconclusive results. Given the persistent and unexplained ascites, an exploratory laparotomy was performed. Intraoperatively, extensive omental caking and diffuse peritoneal carcinomatosis were noted. Histopathological examination of peritoneal and omental nodules confirmed the diagnosis of epithelioid mesothelioma, with no evidence of colorectal malignancy. Due to poor Eastern Cooperative Oncology Group (ECOG) performance status, the patient subsequently received palliative treatment.

Discussion

Diagnosis of peritoneal epithelioid mesothelioma is challenging, particularly when presenting as unexplained ascites. This case is unique due to initial presentation solely with ascites, undiagnosed despite extensive conventional workup. Incidental pulmonary destructive lesions on chest CT, suggestive of asbestos-related changes, provided a crucial, albeit indirect, clue, as

asbestos exposure is a key risk factor for mesothelioma, even without clear occupational history. Abdominal CT findings, including rectosigmoid mass and peritoneal thickening, initially suggested common intra-abdominal malignancies. However, negative endoscopic findings (colonoscopy and cystoscopy) highlighted imaging's non-specific nature and the difficulty in differentiating peritoneal mesothelioma from other conditions. This case underscores the need for clinicians to consider peritoneal mesothelioma in patients with persistent, unexplained ascites when initial investigations are inconclusive. Early invasive procedures, such as exploratory laparotomy for tissue biopsy, are often indispensable for definitive histological diagnosis. Prompt and accurate diagnosis is paramount for timely management and improved patient prognosis.

Conclusion

This case report serves as a vital reminder for clinicians to include peritoneal malignant mesothelioma in the differential diagnosis for patients with refractory, unexplained ascites, particularly when standard diagnostic approaches fail to establish a clear etiology. Early consideration of exploratory surgery for histological confirmation is crucial for achieving a timely diagnosis and initiating effective treatment.

病例報告 114_C040

第二型糖尿病的女性年長者反覆空腹低血糖之案例報告：轉移性胰島素瘤的診斷與治療

Recurrent Fasting Hypoglycemia in an Elderly Woman with Type 2 Diabetes: A Rare Case of Metastatic Insulinoma

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Introduction

Insulinoma is a rare type of neuroendocrine tumor that results in excessive insulin secretion, leading to recurrent hypoglycemia. It is particularly uncommon in elderly patients with a long-standing history of type 2 diabetes mellitus (T2DM), where hypoglycemia is typically attributed to the effects of antidiabetic therapy. This can lead to misdiagnosis or delayed diagnosis, as endogenous hyperinsulinism may be overlooked. We present a rare case of metastatic insulinoma in an elderly woman with T2DM, whose persistent hypoglycemia was initially masked by oral hypoglycemic agents (OHAs).

Case Report

An 81-year-old woman who had T2DM on oral hypoglycemic agents (OHA) for decades presented with acute left-side weakness and slurred speech on January 23, 2025. Her clinical symptoms were initially suspected of transient ischemic attack or metabolic encephalopathy, prompting her admission to the neurology department. During her hospitalization, recurrent episodes of hypoglycemia were noticed with serum glucose levels between 40-50 mg/dL. Despite discontinuing all OHAs, the hypoglycemic episodes persisted even after discharge.

The patient experienced daily episodes of hypoglycemia, typically in the early morning or late afternoon, often without food intake. Interestingly, her HbA1c level had been decreasing since mid-2022, raising the suspicion of an underlying metabolic issue. Given the recurrent nature of hypoglycemia, the patient was referred to the metabolic service for further evaluation.

During a supervised fasting test, her serum glucose dropped to 39 mg/dL, with an elevated insulin level of 27.83 μIU/mL, confirming endogenous hyperinsulinemic hypoglycemia. Further examinations were arranged. Tumor markers revealed normal range of CA19-9 (32.73 U/mL) and mildly elevated CEA (9.1 ng/mL). Abdominal magnetic resonance imaging found a 5 cm tumor in the pancreatic tail, along with multiple hepatic lesions. To confirm the diagnosis of insulinoma, a liver biopsy was performed on February 12, 2025, which revealed metastatic insulinoma. The histological sections demonstrated liver tissue infiltrated by well-differentiated neuroendocrine tumor. The neoplastic cells exhibited a vaguely organoid arrangement with moderate eosinophilic cytoplasm and a salt-and-pepper chromatin pattern. Immunohistochemical analysis reveals positive for INSM1 and synaptophysin. The patient was started on octreotide therapy, leading to the improvement in her glycemic control.

Discussion

In this case, the patient’s long-term use of OHAs initially masked the presence of hypoglycemia, delaying recognition of the underlying cause. As her HbA1c progressively declined, coupled with

the recurrence of fasting hypoglycemia, a differential diagnosis of insulinoma became probable. The presence of hepatic metastasis in this patient is an unusual manifestation of this tumor. Imaging modalities alongside histopathological examination are critical for the diagnosis of insulinomas. Somatostatin analogues, such as octreotide, are essential in controlling hypoglycemia by inhibiting insulin secretion. This patient's response to octreotide highlights its importance in management for patients with metastatic insulinomas.

Conclusion

This case underscores the need for physicians to maintain a suspicion for insulinoma in elderly patients with T2DM, especially when they present with recurrent episodes of unexplained hypoglycemia even after discontinuing antidiabetic medications. It's crucial to early recognize this rare condition so that patients can get timely treatment with somatostatin analogues, which can greatly improve their outcomes.

病例報告 114_C041

陰性 ANCA 嗜酸性肉芽腫性多血管炎誤診為特發性嗜酸性粒細胞增多症：九年的診斷歷程

ANCA-Negative Eosinophilic Granulomatosis with Polyangiitis Masquerading as Idiopathic Hypereosinophilic Syndrome: A Nine-Year Diagnostic Odyssey

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Introduction

Eosinophilic Granulomatosis with Polyangiitis (EGPA) is a rare ANCA-associated vasculitis affecting small-to-medium vessels with an incidence of 1-3 per million annually. Cardiac involvement occurs in 40-60% of patients and represents the leading cause of mortality. Distinguishing ANCA-negative EGPA from Hypereosinophilic Syndrome (HES) remains diagnostically challenging, particularly when tissue confirmation is unavailable.

Case Report

A 45-year-old male presented with chest tightness in May 2015. Coronary angiography revealed triple vessel disease with diffuse aneurysmal dilatation, requiring multiple percutaneous interventions over nine years. In January 2024, he developed peripheral arterial occlusive disease with bilateral aneurysmal formation and left lower limb total occlusion, necessitating above-knee amputation after failed angioplasty. Persistent hypereosinophilia (>15,000 cells/μL) was noted throughout the clinical course.

Comprehensive evaluation in 2018 showed normocellular bone marrow with hypereosinophilia and negative autoimmune markers (ANA, c-ANCA, p-ANCA). Initially diagnosed with idiopathic HES, he received steroid therapy with immunosuppressive agents. Further testing revealed no PDGFR/FGFR mutations and initially normal nerve conduction studies, which later showed polyneuropathy in the ninth year. Pulmonary function testing demonstrated severely restrictive pattern with bronchodilator reversibility, suggesting asthma. Based on these findings, diagnosis was revised to EGPA. Treatment included methylprednisolone pulse therapy, cyclophosphamide, and mepolizumab for refractory disease.

Discussion

EGPA comprises two phenotypes based on ANCA status, with ANCA-negative cases representing the majority and showing more prominent eosinophilic features. This creates significant diagnostic overlap with HES, which is defined by persistent hypereosinophilia and organ damage without secondary causes. The key distinction lies in vasculitic involvement in EGPA. Cardiac complications occur in 16-62% of EGPA patients and account for up to 50% of deaths.

This case exemplifies diagnostic challenges of ANCA-negative EGPA, initially misdiagnosed as HES due to absent vasculitic evidence and negative ANCA serology. The severe progression to limb amputation represents an aggressive manifestation. The critical limitation was inability to obtain tissue pathology from coronary arteries due to procedural risk, and failure to preserve amputated tissue for examination.

Conclusion

This case demonstrates the diagnostic complexity of ANCA-negative EGPA and its overlap with HES, illustrated by a nine-year course resulting in limb amputation. The missed opportunity for histopathologic confirmation from surgical specimens underscores the importance of tissue preservation in suspected vasculitis. Clinicians should maintain high suspicion for systemic vasculitis in young patients with hypereosinophilia and progressive vascular disease, ensuring surgical specimens are preserved for definitive diagnosis and timely treatment guidance.

病例報告 114_C042

氣管脈絡球腫瘤：一例具有侵襲性特徵及惡性潛力的腫瘤

Tracheal Glomus Tumor：A Neoplasm with Aggressive Features and Malignant Potential

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Introduction

Tracheal glomus tumors are rare mesenchymal neoplasms originating from modified smooth muscle cells of glomus body. They can mimic carcinoid tumors both clinically and histologically. We present a case of upper tracheal glomus tumor with high-risk features and review the diagnostic, histologic, and treatment considerations.

Case Report

A 52-year-old male with history of hypertension and hyperlipidemia was referred for evaluation of persistent cough for six months. Initial bronchoscopy revealed a 1.5 cm protruding tracheal mass at upper third of the trachea, causing 90% airway obstruction. PET-CT showed a highly FDG-avid lesion without distant metastasis. On August 29, 2024, he underwent bronchoscopic tumor debulking under ECMO support. Pathology confirmed a benign glomus tumor. The patient was readmitted on November 5, 2024, for segmental tracheal resection with tumor excision and end-to-end anastomosis. Postoperative recovery was smooth. Immunohistochemical staining showed synaptophysin(-), CD34(-), desmin(-), collagen IV(-), β-catenin(-), SMA(+), H-caldesmon(+), and myosin(+), consistent with the glomus tumor profile. However, due to tumor size (>2 cm), deep location, vascular invasion, and cartilage destruction, malignant potential was considered. Follow-up imaging showed no evidence of local recurrence or distant metastasis.

Discussion

Glomus tumors are rare neoplasms derived from modified smooth muscle cells of glomus body, comprising less than 2% of soft tissue tumors. Tracheal glomus tumors are extremely uncommon, with only 82 cases involving the tracheobronchial tree reported up to 2019. Glomus tumors tend to occur in middle-aged males and are most often located in the lower third of the trachea. Symptoms include dyspnea, cough, and hemoptysis, although many patients are asymptomatic. Histopathologically, glomus tumors show positive staining for SMA and vimentin, and negative staining for neuroendocrine markers, aiding differentiation from carcinoid tumors and hemangiopericytomas. Although most glomus tumors are benign, some show aggressive behavior or malignancy. Malignant glomus tumors are diagnosed based on criteria including tumor size (>2 cm), deep location, nuclear atypia, and mitotic activity. The recommended treatment of malignant or bulky glomus tumors is surgical resection, with preferably negative margins. Incomplete excision may lead to local recurrence or distant metastasis, which are the primary causes of glomus tumor-related mortality. Bronchoscopic intervention may be used for diagnosis or airway restoration, but is insufficient as definitive therapy. Endobronchial treatments such as laser resection, high-frequency electrocoagulation, and argon plasma coagulation are typically reserved for patients who are not surgical candidates. Overall, the post-surgical prognosis is good,

but patients should have long-term follow-up due to the potential recurrence.
The role of chemotherapy is unclear due to limited data. Accurate diagnosis, which can be difficult due to the similarities to other tracheal tumors, depends on detailed histology and immunohistochemistry.

Conclusion

In summary, malignant tracheal glomus tumors are rare neoplasms. In this report, we describe the management of a typical patient presenting with a symptomatic glomus tumor of the proximal trachea. Complete surgical resection with negative margins remains the cornerstone of treatment. Early airway management and accurate histopathological diagnosis are essential for optimal outcomes and guiding further treatment strategies.

病例報告 114_C044

病例討論：急性腎損傷併移植腎特發性動靜脈廔管---賀癌平心毒性相關

Acute Kidney Injury Caused by Trastuzumab-related Cardiotoxicity in a Transplanted Kidney with Idiopathic Arteriovenous Fistula

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Introduction

Renal arteriovenous fistula (RAVF) is a rare vascular anomaly characterized by a direct connection between the renal artery and vein. It can be classified as acquired (70%), congenital (25%) or idiopathic (3%–5%). [1] Trastuzumab is a monoclonal antibody that selectively targets the human epidermal growth factor receptor protein (HER2). Cardiotoxicity is a well-known effect of trastuzumab, often leading to hypoperfusion and contributing to solid organ dysfunction, including acute kidney injury(AKI). Herein, we report a 56-year-old woman with RAVF in transplanted kidney who experienced a hypotensive episode and subsequent AKI while receiving trastuzumab for breast cancer.

Case Report

A 56-year-old woman complained about nausea and vomiting for two days after regular chemotherapy. She had chronic interstitial nephritis underwent deceased-donor kidney transplant in Oct 2011. She has never received kidney biopsy after transplant. There is also no history of cardiovascular diseases. She was diagnosed with HER2-positive invasive carcinoma of right breast, under Trastuzumab use. This time, she was hospitalized because of adhesion ileus over distal jejunum, resolved by exploratory laparotomy enterolysis. Acemetacin, a non-steroidal anti-inflammatory drugs(NSAID) was prescribed for pain relief. Trastuzumab was also resumed and blood pressure dropped twenty hours after infusion. Acute kidney injury(AKI) developed as well. Renal artery sonography showed a lobulated vascular mass lesion. Computed tomography angiography of abdomen showed arteriovenous fistula and the arteriovenous malformation nidus was noted. After the embolization, her renal function recovered and her serum creatinine level improved. Trastuzumab was administrated again after the procedure and no more episode of acute kidney injury was noted. Then followed-up in OPD was made.

Discussion

Renal arteriovenous fistula(RAVF) is a rare vascular anomaly. Non-traumatic RAVFs may result from aneurysm erosion or vascular degeneration [2]. While small RAVFs are usually asymptomatic, larger ones may lead to high-output heart failure (32%) and renal dysfunction[3]. In review of the medication history, the decline happened after a single dose of trastuzumab and a four-day course NSAID. Studies have reported an incidence of trastuzumab-related cardiotoxicity in 15-20% of treated patients. Notably, declines in cardiac function ≥10% has been observed in 40-45% of patients across consecutive trastuzumab-treated cohorts[4]. The resulting renal hypoperfusion may be contributing to acute kidney injury. NSAIDs are also known to inhibit prostaglandin biosynthesis. In conditions characterized by dysregulation of circulating arterial volume, such as

in states of increased renin-angiotensin-aldosterone system(RAAS), there is enhanced production of vasodilatory prostaglandins by the afferent arteriole endothelium. These compensatory mediators to preserve renal perfusion in the setting of decrease effective circulating volume. However, due to trastuzumab-induced hypoperfusion, this autoregulatory mechanism becomes insufficient. The combined effects of impaired prostaglandin synthesis and hemodynamic compromise can overwhelm renal autoregulation, leading to a consequent ischemic injury[5]. After an embolization was performed, subsequent follow-up showed no recurrence of acute kidney injury.

Conclusion

In summary, trastuzumab appears to increase susceptibility to other harmful factors, contributing to the development of acute heart failure related to cardiotoxicity, which manifests as hemodynamic instability. The presence of a renal AVF in the transplanted kidney led to hypoperfusion and subsequent renal function deterioration. Treatment such as embolization, may become necessary according to clinical presentation to preserve stable hemodynamic status.

病例報告 114_C045

靜脈硬化性結腸炎：腹痛的罕見病因與文獻回顧

Phlebosclerotic colitis: a rare etiology of abdominal pain and literature review

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Introduction

Phlebosclerotic colitis (PC) is a rare form of ischemic colitis caused by sclerosis and calcification of mesenteric and submucosal veins, resulting in chronic venous congestion and colonic ischemia. The condition is most often reported in East Asian populations and typically involves the right colon. Clinical manifestations are nonspecific, including abdominal pain, diarrhea, or gastrointestinal bleeding. Radiologic evidence of mesenteric venous calcification is crucial for diagnosis. Management ranges from conservative therapy to surgical resection in cases of progressive disease or complications.

Case Report

A 56-year-old woman with a history of peptic ulcer disease (on proton pump inhibitor therapy) and right forearm amputation presented to the emergency department in August 2019 with epigastric and right lower quadrant pain, nausea, and fever. Laboratory tests revealed leukocytosis (11,500/μL, 81.6% neutrophils) and elevated C-reactive protein (1.74 mg/dL). Abdominal computed tomography (CT) demonstrated mesenteric vascular calcifications along the ascending colon, wall thickening, pelvic ascites, and mild terminal ileal distention. She was diagnosed with phlebosclerotic colitis. Symptoms improved after antibiotic and supportive treatment, and she was discharged.

In January 2020, she returned with recurrent abdominal pain, epigastric fullness, low-grade fever, and diarrhea. Laboratory data showed leukocytosis (12,500/μL, 75.4% neutrophils), elevated D-dimer (855.2 ng/mL), and normal CRP. Abdominal CT revealed progression of phlebosclerotic colitis involving the right and transverse colon with acute ischemic changes. Stool tests were positive for *Clostridium difficile*. Diarrhea improved with antibiotics and supportive care, and she was discharged.

Despite initial improvement, her symptoms progressed. Colonoscopy before surgery revealed deep purple discoloration of the descending colon. In March 2020, she underwent subtotal colectomy with preservation of the sigmoid colon. Histopathology confirmed phlebosclerotic colitis with focal ischemic changes and arteriosclerosis. The postoperative course was uneventful.

Discussion

Phlebosclerotic colitis (PC) is a rare ischemic colitis caused by sclerosis and calcification of mesenteric and submucosal veins, most often affecting the right colon. The etiology is not established; possible contributing factors include chronic venous congestion, herbal medication use, and genetic predisposition. The disease usually follows an indolent course, presenting with abdominal pain, bloating, or diarrhea, but acute exacerbations may occur due to ischemia or superimposed infection, as in our patient with concurrent *Clostridium difficile* colitis.

Diagnosis relies mainly on radiology, with CT showing serpiginous mesenteric venous calcifications and colonic wall thickening. Colonoscopy may reveal bluish or purple mucosa consistent with chronic ischemia. Treatment depends on severity: conservative management may suffice in early stages, but progressive disease or complications require surgery. Subtotal colectomy is often effective and associated with good outcomes. Our patient initially improved with conservative treatment but developed progressive symptoms, ultimately requiring subtotal colectomy with confirmatory pathology.

Conclusion

Phlebosclerotic colitis is a rare chronic ischemic colitis characterized by mesenteric venous calcifications and recurrent abdominal symptoms. While conservative treatment may provide temporary relief, progressive disease often requires surgical resection. This case illustrates the natural progression of PC and highlights the importance of early recognition and timely surgical referral.

病例報告 114_C046

保守治療佯裝 Boerhaave 症候群之食道壁內剝離之病例報告

Conservative Management of Esophageal Intramural Dissection Mimicking Boerhaave Syndrome : A Case Report

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Introduction

We report a rare case of esophageal intramural dissection with mucosal ulceration mimicking Boerhaave syndrome. Timely endoscopy showed no transmural rupture, allowing conservative management. This case highlights diagnostic challenges and treatment decisions in atypical esophageal injuries resembling life-threatening conditions.

Case Report

A 67-year-old male with history of subtotal gastrectomy over 40 years ago for perforated peptic ulcer was admitted for sore throat, chest pain and dysphagia lasting one week, with a 9 kg weight loss. Initial panendoscopy revealed an esophageal mucosal defect in the upper third (22–26 cm from incisors), a false lumen dissection extending to 31 cm, and multiple longitudinal ulcers from 27–40 cm. Chest CT excluded obvious perforation. Nasogastric tube drainage was placed into the false lumen. Follow-up endoscopy showed mucosal healing with persistent but non-progressive dissection. Biopsy confirmed esophageal ulcer with low-grade intraepithelial neoplasia. The nasogastric tube was successfully removed 15 days post-procedure, and the patient resumed oral intake without discomfort. Recent panendoscopy showed no residual dissection or mucosal defect, with plans for surveillance every 3–6 months.

Discussion

Boerhaave syndrome is a rare, life-threatening condition caused by full-thickness esophageal rupture, often after forceful vomiting or increased intraesophageal pressure. It differs from Mallory-Weiss syndrome, which involves only mucosal lacerations.[1] Untreated Boerhaave syndrome can lead to mediastinitis, sepsis, and multi-organ failure. Diagnosis is difficult due to nonspecific symptoms; the classic Mackler triad (vomiting, chest pain, and subcutaneous emphysema) appears in few cases.[2] Most ruptures occur in the left distal esophagus. Risk factors include alcohol, overeating, and increased thoracoabdominal pressure. Early CT and contrast studies are essential for diagnosis.[2] Treatment varies from conservative care for contained leaks to urgent surgery or endoscopy. Delayed treatment worsens prognosis, with mortality exceeding 90% if untreated.[1,2] Some Boerhaave cases evolve from undiagnosed Mallory-Weiss lesions, underscoring the importance of early detection.[1] Rapid diagnosis, teamwork, and tailored management improve outcomes. Patient education is vital for early presentation and mortality reduction.

Conclusion

This case illustrates a rare esophageal injury mimicking Boerhaave syndrome, successfully treated conservatively. Accurate diagnosis through timely imaging and endoscopy is crucial to guide

treatment and avoid overtreatment in non-transmural lesions.

病例報告 114_C047

缺乏莢膜之隱球菌造成之腦膜炎案例

A Case of Capsule-Deficient Cryptococcal meningitis

Introduction

Cryptococcosis is a major worldwide disseminated invasive fungal infection. Cryptococcus species typically cause infection in immunocompromised patients, although infections in immunocompetent cases have been reported. In 2022, WHO listed Cryptococcus neoformans as a top fungal priority pathogen. The capsulated form of Cryptococcus neoformans is more common encountered and can be diagnosed by India ink stain or antigen detection. On the other hand, capsule-deficient strain is rare and raises the difficulty of diagnosis due to negativity of antigen detection. Herein, we present an immunocompetent case of cryptococcal meningitis which was infected by capsule-deficient Cryptococcus neoformans.

Case Report

A 70-year-old woman with underlying diseases of hypertension and dyslipidemia presented to our hospital with a 3-month history of intermittent fever and headache. She had 2 episodes of hospitalization respectively.

During the first admission, contrast brain MRI was arranged for evaluation of suspected chronic meningitis and showed multiple nodular and linear lesions over bilateral cerebral hemispheres. CSF laboratory parameters indicated chronic meningitis. No specific pathogen was noted in CSF analysis. Under the impression of TB meningitis, anti-TB treatment with oral HERZ and intravenous Methylprednisolone then shifting to oral Prednisolone were administered. Improving CSF laboratory parameters and clinical condition were noted. The patient was then MBD.

Nevertheless, she repeatedly presented to hospital due to recurrent low-grade fever and intermittent headache. Moreover, new-onset of glazing gaze and hand tremor were noted as well. We performed lumbar puncture and chronic meningitis was noted again. Brain MRI follow-up showed stable disease. Due to chronic meningitis with unknown pathogen, we consulted neurosurgeon for brain biopsy. However, CSF culture yielded *Cryptococcus neoformans*. Cryptococcal meningitis caused by *C. neoformans* was therefore diagnosed. Intravenous Amphotericin B and oral Flucytosine were prescribed. Improved headache and neurologic symptoms were noted under antibiotic treatment. Defervescence was also noted. The patient was then discharged from hospital.

Discussion

C. neoformans are frequently encapsulated. The capsule is not only the major virulence factor but the most commonly demonstrated part of the yeast over which most of the diagnostic modalities focus. The capsule can be detected by PAS, GMS, India ink stain, and mucicarmine stain or by targeting the cryptococcal antigen by latex agglutination assay, enzyme immunoassay, and lateral flow array. Fungal culture for Cryptococcus is considered the gold standard diagnostic method. Following growth in culture, methods such as MALDI-TOF MS and Sanger sequencing, can be

utilized for identification and differentiation between the various Cryptococcus species.

Conclusion

This case demonstrates a rare example of an immunocompetent patient who was found to have meningitis cause by a noncapsulated strain of *C. neoformans*. This case reminds the clinician of the importance of the conventional identification approaches such as CSF fungal culture.

病例報告 114_C048

80 歲糖尿病患者合併轉移性胰島素瘤

A 80-Year-Old Female with metastatic insulinoma

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Introduction

Insulinoma was a rare neuroendocrine neoplasm that caused endogenous hyperinsulinemia, over 90% of which were benign. The median age of diagnosis was in the fifth decade of life. We report an unusual case of insulinoma with liver metastasis in a female patient who was 85 years old.

Case Report

An 80-year-old female with underlying disease of type 2 DM for 20 years, under Janumet(sitagliptin 50mg/ Metformin 850mg) BID. and ischemic infarction in the left basal ganglion and left periventricular white matter, with right hemiplegia.
She was admitted for frequent hypoglycemia in the recent month.
2 weeks prior to the admission, she had experienced left side weakness and recovered spontaneously. Brain MRI revealed no evidence of acute ischemic infarction. She had been admitted with the impression of transient ischemic attack. During admission, she reported frequent hypoglycemic episodes in the past 1 month before this event. Thus, she was referred to the endocrinology department. She was admitted for a 72-hour fasting test to rule out Insulinoma. The supervised fasting test revealed markedly elevated serum insulin (27.83 μIU/mL) concurrent with severe hypoglycemia (serum glucose 39 mg/dL), suggesting endogenous hyperinsulinemia. a mass about 5 cm at the pancreatic tail, r/i pancreatic cancer with multiple liver nodules up to 4 cm, r/i metastasis. CA-199 (32.73 U/mL)were within normal range, CEA(9.1ng/mL) showed mild elevation in level. EUS(endoscopic ultrasound) biopsy was arranged but not done for gastric varices. Liver biopsy was done. The pathology report was compatible with Insulinoma. She received octreotide with less hypoglycemia episode noted. After being discharged , she was referred to a gastroenterologist and received Lanreotide and Everolimus treatment.

Discussion

This case described the diagnosis complexity of hypoglycemia in an elderly patient with multiple comorbidities including previous cerebral infarction, TIA and type 2 diabetes under OHA..
Insulinoma was a rare neuroendocrine neoplasm that caused endogenous hyperinsulinemia. The median age at diagnosis for insulinoma is between 47 and 56 years old. Less than 10% of insulinoma was metastatic, defined as malignant. A 72-hour fasting test is considered the gold standard for diagnosing insulinoma. Octreotide, a somatostatin analog, is effective in controlling hypoglycemia by inhibiting insulin secretion, and was well tolerated in this patient. Everolimus, an inhibitor of the mechanistic target of rapamycin (MTOR), was approved by the FDA in 2011 for the treatment of progressive pancreatic NE tumors.

Conclusion

Insulinomas is a rare disease and could cause neurological signs. It was difficult to make a diagnosis, especially in elderly patients with other neurological comorbidities. It should be kept in mind in elderly patients while significant recurrent hypoglycemic episodes occurred.

病例報告 114_C049

Enteritis cystica profunda 診斷必須依賴完整病理檢體：從不一致診斷中學到的課題

Diagnosis of Enteritis cystica profunda requires Complete Pathological Tissue Samples: Key Lessons Learned from Inconsistent Diagnoses

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Introduction

Enteritis cystica profunda (ECP) is a rare and nonneoplastic disease charactered with mucin-filled cysts surrounding epithelial cells in the submucosa of the small bowel[1]. The etiology is unclear but is surmised to be associated with certain conditions such as inflammatory bowel disease, Peutz-Jeghers syndrome, infection, ischemia, and even trauma[2,3]. Moreover, there is one hypothesis that epithelial cells migrated into the submucosal layer after mucosa ulcers progression or cystic dilation of glands takes place as healing process. Diagnosis and treatment of ECP in the reported cases is diagnostic laparoscopy followed by resection and anastomosis[4]. Here we presented a 72-year-old case of biopsy-proven adenoma receiving endoscopic papillectomy. However, the final diagnosis turned out to be this rare condition, Enteritis cystica profunda.

Case Report

This 72-year-old woman with history of hypertension presented to the emergency department (ED) with fever, jaundice and epigastric pain for three days. Other symptoms included anorexia, headache, and loose stool. She denied nausea, vomiting, tea-colored urine, clay stool. Due to computed tomography (CT) of abdomen from local hospital indicated hypodense nodule over pancreatic head with mild dilation of common bile duct (CBD) and the main pancreatic duct (MPD) and liver mass, she was referred to our hospital.

Blood tests showed leukocytosis with neutrophils predominant (WBC:15500/ul, Neutrophils:82.2%), normal range total bilirubin (T-bil:1.14mg/dl), mild elevated liver functions (AST:108 U/L, ALT:78U/L), and elevated hsCRP:15.24 mg/dL. Further survey revealed HbsAg: negative, HCV Ab: negative, and tumor marker were within normal range (AFP:2.62 ng/mL, CEA:1.13 ng/mL, CA19-9: 9.4U/ml).

Abdominal sonography as liver tumor survey favored liver abscess without obvious liquefaction, with later resolving after antibiotics treatment. Endoscopic ultrasonography (EUS) showed one ampulla of Vater tumor (17.9 x 14.1mm) without CBD or MPD invasion. The pathology of biopsy in this session showed pyloric gland adenoma.

Endoscopic papillectomy was scheduled on the following admission course four months later as management of adenoma. Pathology of operation sample showed enteritis cystica profunda, revealing mucosal glands with irregular dilation in submucosa. The pathological section of the re-biopsy from the ampulla of Vater lesion showed non-specific chronic inflammation.

Discussion

Adequate specimen collection is essential for a correct diagnosis[5]. However, forceps biopsy often makes it difficult to obtain sufficient specimens. In our case report, we confirmed that the

pathological samples obtained with forceps were insufficient to diagnose ECP. After discussing with the pathologists, they concluded that the biopsy from the first EUS examination could only yield a diagnosis of adenoma, as there were inadequate histological samples of the submucosa for confirmation.

Conclusion

Enteritis cystica profunda is a rare but benign disease which usually requires pathological diagnosis. Complete resected specimens are usually demanded for supporting the diagnosis. Despite ECP is generally not required treatment unless severe symptoms, surgical resection or endoscopic resection are usually needed to obtain sufficient specimens to confirm the diagnosis and to exclude possible malignant potentials.

病例報告 114_C050

創傷後巨大鈣化脾臟偽囊腫

Post-traumatic Giant Calcified Splenic Pseudocyst

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Introduction

Splenic calcifications are uncommon incidental findings on abdominal imaging. They may result from prior infections, infarctions, or trauma-related sequelae. Among these, post-traumatic splenic pseudocysts with calcified walls are particularly rare and often present years after the inciting event. Chronic organizing hematomas can progressively undergo fibrosis and dystrophic calcification, creating a characteristic rim-calcified mass. Most patients remain asymptomatic, and such lesions are typically detected incidentally during evaluation for unrelated complaints. Understanding their etiology and imaging characteristics is essential to differentiate benign calcified pseudocysts from malignant or infectious splenic masses, thereby preventing unnecessary interventions.

Case Report

A 75-year-old man presented to the emergency department with abdominal pain and was clinically diagnosed with constipation. Past medical history was notable for hypertension, type 2 diabetes mellitus, and chronic kidney disease. Abdominal radiograph unexpectedly revealed a well-defined, 10 cm round lesion in the left upper quadrant. Abdominal computed tomography demonstrated a large cystic splenic lesion with a thick, complete calcified wall, favoring a chronic post-traumatic hematoma with pseudocyst formation. No internal enhancement or septations were observed. On retrospective history review, the patient recalled a remote blunt abdominal injury that was never evaluated or treated. Given the absence of acute symptoms, hemodynamic stability, and lack of compressive effects, conservative management was recommended. The patient's abdominal pain improved with bowel regimen, and he was discharged for outpatient follow-up.

Discussion

Calcified splenic lesions encompass a wide differential, including granulomatous infections, vascular tumors, infarctions, and pseudocysts. Post-traumatic pseudocysts account for nearly 80% of nonparasitic splenic cysts, with approximately half demonstrating peripheral calcification. The pathophysiology involves unresolved intraparenchymal hematomas that evolve into fibrous-walled cysts with dystrophic calcification over time. Their imaging hallmark is a round, hypoattenuating mass with a thick curvilinear calcified rim, consistent with this case. Distinguishing these benign entities from neoplastic or infectious splenic processes is crucial, as unnecessary surgery may otherwise be considered. Management depends on size, symptoms, and risk of rupture. While large symptomatic cysts may warrant splenectomy or partial splenectomy,

asymptomatic patients with stable lesions are often managed conservatively. Our patient's presentation aligns with the natural history of a chronic, calcified post-traumatic hematoma incidentally discovered on imaging.

Conclusion

This case illustrates a rare giant calcified splenic pseudocyst arising from remote trauma. Recognition of its characteristic imaging appearance is essential for accurate diagnosis and avoidance of unnecessary intervention. Conservative observation remains appropriate in asymptomatic patients, underscoring the importance of integrating clinical history with radiologic findings.

病例報告 114_C051

腸道超音波診斷克隆氏症術後腸皮瘻管：一個病例報告

Intestinal Ultrasound Detects Postoperative Enterocutaneous Fistula in Crohn's Disease: A Case report

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Introduction

Fistulizing complications are a hallmark of Crohn's disease (CD), reflecting its transmural inflammatory nature. Enterocutaneous fistulae (ECF) are a rare but morbid postoperative complication, often requiring intensive management. While magnetic resonance enterography (MRE) and computed tomography enterography (CTE) are standard imaging modalities, intestinal ultrasound (IUS) has emerged as a rapid, non-invasive, and radiation-free alternative with comparable diagnostic accuracy. Contemporary guidelines increasingly endorse IUS for both initial diagnosis and longitudinal monitoring of CD complications. However, current evidence is scarce, as the existence of ECF has been reported in only a few case reports. This case report illustrates the pivotal role of IUS in identifying a complex postoperative ECF where conventional cross-sectional imaging was non-diagnostic.

Case Report

A 34-year-old woman with a long-standing history of ulcerative colitis underwent a left hemicolectomy for a presumed stricture. Postoperative histology revised her diagnosis to Crohn's disease. Her course was complicated by an anastomotic leak requiring a temporary ileostomy. Three months after ileostomy reversal, she presented with persistent, low-volume purulent drainage from the stoma scar. A contrast-enhanced CT scan was unable to delineate a fistulous tract due to severe postoperative inflammation. However, IUS clearly identified a linear, hypoechoic channel extending from the skin at the former stoma site to the distal ileum, confirming a mature ECF. IUS also revealed an associated subcutaneous abscess and a suspected secondary entero-enteric fistula. A follow-up IUS five weeks later demonstrated stability of the fistulous tract but an increase in the abscess size, highlighting its utility for disease monitoring and guiding subsequent management decisions.

Discussion

This case underscores the diagnostic value of IUS in complex postoperative settings. While CTE and MRE are established for detecting fistulae, their utility can be limited by artifacts, inflammation, and anatomical distortion, as seen here. IUS provided a definitive diagnosis by using high-frequency probes for detailed real-time visualization of the bowel wall and surrounding tissues. Its ability to dynamically assess structures without ionizing radiation or contrast agents makes it an ideal tool for repeated examinations. This aligns with recent guidelines recommending IUS as a first-line option for assessing CD activity and complications, offering superior patient comfort and

accessibility compared to other cross-sectional imaging techniques.

Conclusion

Intestinal ultrasound is a highly effective and feasible imaging modality for the primary diagnosis and longitudinal monitoring of postoperative enterocutaneous fistula in patients with Crohn's disease. This case demonstrates that IUS can succeed where conventional imaging fails, making it an indispensable tool in the multidisciplinary management of complex inflammatory bowel disease.

病例報告 114_C052

以反覆腸胃道出血為表現的 Waldenström Macroglobulinemia

Waldenström Macroglobulinemia Presenting as Recurrent Gastrointestinal Bleeding

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Introduction

A 70-year-old woman with melena, anemia, and ocular complications was diagnosed with Waldenström macroglobulinemia. Workup revealed high IgM, monoclonal spike, and marrow lymphoplasmacytic infiltration. She underwent plasma exchange and rituximab-based chemotherapy, later transitioned to rituximab-bendamustine, achieving clinical improvement.

Case Report

A 70-year-old woman with hypertension and type 2 diabetes presented with 1 month of melena. Despite panendoscopy and colonoscopy showing no bleeding source. Persistent melena led to ED evaluation, with blurred vision, headache, pallor, and dyspnea. Labs showed anemia (Hb 8.0 g/dL) and coagulopathy (APTT 47.9 s, INR 1.34, PT 16.3 s). CT revealed no active bleeding. Double balloon assisted enteroscopy revealed diffuse easily touching bleeding lesion including visible vessel over distal ileum and multiple colonic ulcers with adherent blood clots s/p endoscopic hemostasis.

Hematology evaluation revealed elevated free kappa light chain (26.2 mg/dL), abnormal kappa/lambda ratio (1.65), high IgM (4125 mg/dL), and β-2 microglobulin (4718 ng/mL). SPEP showed a 38.4% IgM M-spike. Bone marrow biopsy demonstrated aberrant B cells with kappa restriction (CD19+, CD20+, CD22+, CD27+, IgM+; CD5-, CD10-, CD11c-, CD103-), consistent with Waldenström macroglobulinemia (WM). Ophthalmology confirmed bilateral central retinal vein occlusion with macular edema and serous retinal detachment.

She underwent five sessions of plasma exchange followed by three cycles of rituximab, dexamethasone, and cyclophosphamide, then transitioned to rituximab-bendamustine. This case highlights WM presenting with GI bleeding, anemia, hyperviscosity-related ocular complications, and favorable response to immunochemotherapy.

Discussion

Waldenström macroglobulinemia (WM), first described by Jan G. Waldenström in 1944, is a rare B-cell malignancy arising from late-stage B-cell differentiation. WM cells secrete monoclonal IgM and occasionally express surface IgD. The annual incidence is about five per million, remaining stable over time.

Clinically, WM may present with gastrointestinal bleeding due to IgM deposition, acquired von Willebrand syndrome, hyperviscosity, or platelet dysfunction. Other features include lymphadenopathy, organomegaly, and cytopenias.

Diagnosis relies on both laboratory and histopathologic evaluation. Non-invasive testing includes serum and urine protein electrophoresis, immunofixation for IgM, and quantitative immunoglobulins. Bone marrow biopsy reveals intertrabecular lymphoplasmacytic infiltrates. Serum viscosity testing is indicated if symptoms of hyperviscosity are present or IgM >4000 mg/dL.

Treatment is not always required at diagnosis, but when indicated, rituximab-based regimens are preferred. Rituximab with cyclophosphamide and dexamethasone achieving median OS near 95 months. Rituximab-bendamustine is also widely used, offering efficacy, tolerability, and time-limited therapy without risk of IgM flare.
WM remains an indolent yet heterogeneous disorder requiring tailored diagnostic assessment and individualized therapy.

Conclusion

Waldenström macroglobulinemia is a rare but important differential diagnosis of gastrointestinal bleeding, particularly when endoscopic findings are inconclusive. Recognition of associated hyperviscosity and hematologic features is crucial. Early plasma exchange and rituximab-based therapy can effectively control disease and prevent irreversible complications, underscoring the need to consider WM in unexplained GI bleeding.

病例報告 114_C053

接受 Upadacitinib 治療之潰瘍性結腸炎患者合併貓抓病感染:一罕見病例報告

A Rare Case Report of Cat-Scratch Disease in an Ulcerative Colitis Patient under Upadacitinib

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Introduction

Cat-scratch disease (CSD) is usually self-limiting but may present with severe manifestations in immunocompromised hosts. Patients with ulcerative colitis (UC) receiving immunosuppressants like Upadacitinib are at increased risk of infections. We report a rare case of CSD in a UC patient treated with Upadacitinib, highlighting the importance of clinical vigilance for zoonotic infections in the setting of targeted immunosuppression.

Case Report

A 42-year-old man poultry farmer with a >10-year history of UC, maintained on Mesalazine 1000 mg BID and Upadacitinib 15 mg QD, was admitted for a routine colonoscopy follow-up. Following the procedure, he developed a fever of up to 38 °C, and a soft, palpable lesion measuring 3 × 3 cm was noted on the anterior neck. Empirical broad-spectrum antibiotic therapy with Brosym (cefoperazone–sulbactam) was initiated, as an intra-abdominal infection could not be excluded initially. Subsequent neck and chest computed tomography scan revealed a 3.0 × 2.3 cm well-defined focal lesion with internal low attenuation and peripheral enhancement, located inferior to the right sternocleidomastoid muscle near its sternal insertion, anterior to the thyroid gland—findings consistent with an abscess. The patient later recalled being scratched by a wild cat on the anterior chest wall approximately two weeks prior to admission. With suspicion for CSD, incision and drainage of the neck abscess were performed, and PCR testing confirmed Bartonella henselae. Azithromycin (500 mg on day 1, then 250 mg daily for 4 days) was given, and the patient's fever resolved after drainage and completing the antibiotic course. Upadacitinib was withheld during the acute infectious phase and resumed after recovery, with no recurrence noted to date.

Discussion

Cat-scratch disease (CSD), caused by Bartonella henselae and transmitted through cat scratches or bites, is generally mild in immunocompetent individuals but may become severe or disseminated in immunocompromised patients. Ulcerative colitis (UC) is a chronic inflammatory bowel disease frequently treated with immunosuppressive therapies—including corticosteroids, biologics, and small molecules—which predispose patients to opportunistic infections. A Croatian case report described a UC patient treated with infliximab who subsequently developed generalized CSD; immunosuppressive therapy was safely resumed following appropriate antibiotic treatment without recurrence [1].

Upadacitinib, a selective JAK1 inhibitor, has demonstrated efficacy in the treatment of Inflammatory bowel disease (IBD). However, its immunosuppressive effects may predispose patients to opportunistic infections. In the previous clinical trials, infections such as Pneumocystis

jirovecii pneumonia, cytomegalovirus, and herpes zoster were reported among IBD patients receiving Upadacitinib [2,3]. In our patient, CSD likely developed due to immunosuppression from Upadacitinib. Although rare in UC patients, this case highlights the importance of considering zoonotic infections like CSD in immunosuppressed individuals with atypical symptoms and relevant exposure histories.

Conclusion

This case highlights the risk of opportunistic infections like cat-scratch disease in UC patients on immunosuppressive therapy, especially JAK inhibitors. While rare, clinicians should suspect atypical infections in immunocompromised patients with animal exposure. Early recognition and antibiotics can lead to favorable outcomes without long-term interruption of treatment.

病例報告 114_C054

免疫健全成人因核梭桿菌引起的隱源性化膿性肝膿瘍

Cryptogenic Pyogenic Liver Abscess Caused by *Fusobacterium nucleatum* in an Immunocompetent Adult: A Case Report

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Introduction

Fusobacterium nucleatum is a gram-negative, anaerobic commensal organism found in the human oral cavity, where it contributes to periodontal plaque formation.[1] Although usually benign, it can act as an opportunistic pathogen under certain conditions. Reported risk factors include poor oral hygiene, chronic denture use, Lemierre's syndrome, and gastrointestinal translocation such as from diverticulitis.[2] Pyogenic liver abscess (PLA) is a potentially life-threatening condition, most often arising from biliary or gastrointestinal sources. We present a rare case of cryptogenic PLA caused by *F. nucleatum* in an immunocompetent adult with no identifiable primary focus.

Case Report

A 53-year-old previously healthy male presented with a one-month history of poor appetite, fever, cough, dyspnea, epigastric dullache, abdominal fullness, and a 3-kg weight loss. He worked as a compost truck driver and denied recent travel, known exposures, or dental procedures.

Laboratory tests showed leukocytosis with neutrophilia, normocytic anemia, thrombocytosis, and elevated CRP, ESR, ALP, and γ-GT. Tumor markers, autoimmune studies, and viral hepatitis panels were negative.

Contrast-enhanced abdominal computed tomography (CT) revealed a 77-mm liver abscess in the right hepatic lobe with transdiaphragmatic extension into the right lower pleural space, resulting in empyema. Amoeba tests in stool, blood, abscess cultures were negative. Percutaneous aspiration and drainage (PAD) were performed. Culture later identified *F. nucleatum*. Colonoscopy revealed no malignancy or structural lesion.

Empirical cefotaxime and metronidazole were initiated. Upon receiving culture results and antibiotic sensitivity, therapy was de-escalated to sulbactam–ampicillin plus metronidazole. Empyema was treated surgically with pleural decortication. The patient's condition improved significantly with antimicrobial therapy and drainage.

Discussion

Liver abscesses are generally classified as amoebic or pyogenic, both presenting with nonspecific symptoms such as fever, chills, right upper quadrant pain, and vomiting. On contrast-enhanced CT, the "double target sign"—a hypoattenuating core with peripheral enhancement and surrounding zone of low-attenuation edematous liver parenchyma—may appear in both, whereas the "cluster sign", representing coalescing microabscesses, is more specific to PLA.[3]

PLA usually arises from infections of the biliary tract, pelvic or intra-abdominal structures, or occult gastrointestinal malignancies.[4] Common causative organisms are *Klebsiella pneumoniae* and *Escherichia coli*, particularly originating from biliary infections. Hematogenous spread, particularly

from oral sources, is increasingly recognized, with *F. nucleatum* as an emerging pathogen.[1] *F. nucleatum*, an anaerobic gram-negative bacillus and oral commensal, may reach the liver through the portal venous system, either via gastrointestinal translocation or disruption of the oral–gut–liver axis.[5] While history, examination, and imaging guide diagnosis, definitive pathogen identification requires pus culture. Given the high mortality of untreated PLA, early recognition, culture-guided therapy, and appropriate drainage are critical. In this case, early empirical treatment followed by adjustment based on culture led to clinical improvement.

Conclusion

Pyogenic liver abscess carries significant morbidity and mortality. Although biliary and gastrointestinal origins are most common, clinicians should consider atypical or odontogenic pathogens in cryptogenic cases. This case underscores *F. nucleatum* as a rare but important cause of PLA, even in immunocompetent patients, and highlights the importance of broad differential diagnosis and timely microbiological confirmation.

病例報告 114_C055

鏈球菌引起之拉米爾症候群以海綿竇及頸靜脈血栓為表現之病例報告

A Case of Lemierre's Syndrome Caused by Streptococcus constellatus with Cavernous Sinus and Jugular Vein Thrombosis

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Introduction

Lemierre's syndrome used to be a not uncommon but severe disease, known for septic thrombophlebitis, classically involving the internal jugular vein after oropharyngeal infection. The incidence decreased a lot after antibiotics popped out and it was once called the forgotten disease because of its rarity.[1,2,3] Other sites of involvement, such as the external jugular vein and cavernous sinus, have been rarely reported.[4,5] While *Fusobacterium necrophorum* is the typical pathogen, other bacteria such as *Fusobacterium nucleatum*, streptococci, staphylococci, and *Klebsiella pneumoniae* have been reported.[3,6] Treatment comprises antibiotics, anticoagulant, and surgical treatment. The overall mortality rate was around 5%.[7] Here, we present a case of Lemierre's syndrome caused by *Streptococcus constellatus* with cavernous sinus, external jugular vein, and internal jugular vein thrombosis.

Case Report

A 62-year-old woman with no significant underlying disease presented to our hospital with a one-week history of headache, right-sided neck pain for 3 days, accompanied by mild dysphagia. CT and MRI demonstrated thrombosis of the right cavernous sinus, transverse-sigmoid sinus, and external jugular vein, as well as a suspicious fungal mycetoma in the left sphenoidal sinus. Peripheral vein echography after admission also revealed internal jugular vein thrombosis. She was diagnosed with Lemierre's syndrome and possible infective endocarditis, fulfilling the modified Duke criteria with one major criterion (positive blood cultures for *Streptococcus constellatus*) and two minor criteria (rheumatoid factor positive and fever). Antibiotic with Metronidazole was initiated for empirical covering anaerobic. Later, Vancomycin was used for blood cultures yielded *Streptococcus constellatus*, but was changed to levofloxacin after 4-week course of treatment, also for vancomycin-induced leukopenia. Liposomal amphotericin B was used for possible fungal sinusitis but discontinued after imaging excluded skull base osteomyelitis around the sphenoidal sinus. No anticoagulation was given as follow-up imaging showed thrombus regression. The patient improved clinically and was discharged stably after completion of antibiotics treatment.

Discussion

Lemierre's syndrome is classically caused by *Fusobacterium necrophorum* and characterized by an oropharyngeal infection, septicemia, internal jugular vein thrombosis, and metastatic foci such as pulmonary emboli.[2,7] Thrombus formation usually involves the internal jugular vein, while extension to the external jugular vein or intracranial sinuses is rarely reported.[4,5] In contrast, our patient developed Lemierre's syndrome due to *Streptococcus constellatus*, a pathogen of the

Streptococcus anginosus group known for abscess formation but seldom linked to this syndrome.[8] The case was further complicated by extensive thrombosis of the cavernous sinus, transverse-sigmoid sinus, and both jugular veins, along with suspected fungal sinus involvement. This highlights the broader spectrum of pathogens and thrombus sites in Lemierre's syndrome. Prolonged antibiotics remain the cornerstone of treatment,[7,9] whereas anticoagulation should be considered on a case-by-case basis depending on thrombus location, progression, and bleeding risk.[9,10]

Conclusion

Lemierre's syndrome caused by *Streptococcus constellatus* can present with extensive cranial and cervical venous thrombosis. Early recognition and tailored antibiotic therapy are essential for favorable outcomes. Usage of anticoagulation remains controversial and should be individualized.

病例報告 114_C056

無導線心臟節律器於大範圍燒燙傷併完全房室傳導阻滯患者之應用：病例報告

Leadless Pacemaker Insertion in a 27-Year-Old Male with 90% TBSA Burn and Complete AV Block

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Introduction

Burn injuries from explosions are complex and often lead to multisystem trauma including cutaneous burns, inhalation injury, internal organ damage, and possible cardiac conduction abnormalities. While cardiovascular complications are not uncommon in extensive burns, complete atrioventricular (AV) block is rare and potentially fatal. This case highlights the diagnostic challenges and successful use of a leadless pacemaker in a critically ill burn patient with AV block.

Case Report

A 27-year-old previously healthy Vietnamese male migrant worker sustained 90% TBSA third- to fourth-degree flame burns from an air compressor explosion on 2024/03/07. On arrival, he was in hypovolemic shock (BP 57/43 mmHg, HR 135 bpm) with signs of inhalation injury (hoarseness, soot in ears, oral and nasal cavities). Emergency management included fluid resuscitation, intubation, sedation, and norepinephrine infusion. Laboratory results showed leukocytosis, hyponatremia, severe lactic acidosis (lactate 41.5 mmol/L), elevated CPK, and hypoalbuminemia. Imaging ruled out internal bleeding but confirmed extensive burn injury.

During hospitalization, he underwent multiple debridement surgeries and tracheostomy. He experienced recurrent electrolyte imbalances and persistent tachycardia, likely related to sepsis and fluid shifts. On 2024/04/05, he developed a 13-second asystolic event without concurrent acidosis or electrolyte abnormalities. ECG confirmed complete AV block. Given the likelihood of conduction system injury from thermal or electrical trauma, an emergent temporary pacemaker (TPM) was inserted via the left femoral vein—the only remaining area of intact skin.

Transthoracic echocardiography revealed an incidental congenital bicuspid aortic valve with moderate aortic regurgitation. Due to widespread burns and high infection risk, a traditional permanent pacemaker was contraindicated as there was no viable chest wall tissue for generator placement. Over the next two months, the patient required three TPM revisions due to recurrent sepsis and bacteremia.

Once stabilized and infection was controlled, a leadless Micra™ pacemaker was successfully implanted via the right femoral vein on 2024/06/07. The device was deployed in the right ventricular septum with optimal electrical parameters and no complications.

Discussion

This case demonstrates a rare but serious complication of blast-related burn trauma: complete AV block, likely due to conduction system damage. Although not a classic electrical burn, the energy from the explosion may have disrupted the AV node. The absence of coronary ischemia and the spontaneous recovery from asystole without CPR suggest a primary conduction abnormality. Literature supports the possibility of delayed conduction blocks following electrical injuries, even

without myocardial necrosis.
In patients with extensive burns and high infection risk, leadless pacemakers offer a safer alternative to conventional devices by avoiding subcutaneous hardware and minimizing infection.

Conclusion

Complete AV block is a rare but life-threatening complication in burn patients. This case illustrates the feasibility and safety of leadless pacing in managing rhythm disturbances when conventional options are limited. Early recognition and a multidisciplinary approach are essential in optimizing outcomes for such complex cases.

病例報告 114_C057

Etanercept 治療複雜型史蒂文斯—約翰森綜合症併發血球低下與高凝血狀態：病例報告

Etanercept in Complex Stevens-Johnson Syndrome with Cytopenia and Hypercoagulability: A Case Report

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Introduction

Stevens-Johnson syndrome (SJS) is a rare, life-threatening mucocutaneous reaction with debated optimal treatment [1]. Its immunopathology involves drug-specific, HLA class I cytotoxic T-cell responses inducing massive keratinocyte apoptosis and systemic inflammation [2, 3]. Standard management requires prompt withdrawal of the suspected agent and supportive care [4]. The roles of systemic corticosteroids, cyclosporine, and intravenous immunoglobulin remain controversial[5]. Targeted therapies, such as CD80/86 or tumor necrosis factor-alpha (TNF-α) blockade, are also under investigation. We report a case of SJS complicated by cytopenia and hypercoagulability, and describe targeted anti-inflammatory therapy.

Case Report

A 38-year-old woman with right iliac bone osteosarcoma completed a second course of high-dose methotrexate 9 days before presentation. She developed fever (38.0 °C), a diffuse pruritic erythematous rash over her face, trunk and knees, painful oral/vaginal ulcers, and bullous eruptions on both soles. Gabapentin was prescribed for pain control a few days before symptom onset, without prior adverse drug reactions.

She received one dose of abatacept at the outpatient department and was subsequently admitted from the emergency department. Laboratory data showed pancytopenia: white blood cell count 0.9 x 10^3/ L (3.6-11.2) with neutrophils 22.4% (43.3-76.6), hemoglobin 7.8 g/dL (11-15); and platelets 71 × 10^3/L (130-400). C-reactive protein was 14.76 mg/L (<1), and D-dimer was 5,866 ng/mL (<500) with normal fibrinogen. HLA B*15:02 testing, herpes simplex virus culture, and anti-platelet antibody were all negative. No pemphigoid autoantibodies were detected.

Management included empiric antibiotics, granulocyte colony-stimulating factor, and methylprednisolone 1 mg/kg/day with supportive care including nutritional support, hydration, analgesia, and vitamins. Due to delayed healing of mucocutaneous lesions, a single dose of etanercept was administered on day 11. Enoxaparin 0.6 mg/kg once daily was prescribed for DIC. After 18 days, she was discharged with marked clinical improvement, normalized blood counts and coagulation parameters, and successfully tapered corticosteroids.

Discussion

SJS involves a destructive cascade from both adaptive and innate immune systems that amplifies skin damage [6]. Given the patient's severe pancytopenia, repeat administration of abatacept was avoided due to the elevated risk of infection associated with immunosuppression and compromised bone marrow function. Following treatment with etanercept and enoxaparin, the

patient demonstrated marked clinical improvement, including resolution of cytopenia and a decreased consumption of coagulation factors. Due to the patient's immunocompromised status and rapid clinical response, diagnosis was made based on clinical presentation without skin biopsy. Although a pharmacologic rechallenge could clarify the association with recent gabapentin exposure, it was deemed ethically inappropriate. While prompt withdrawal of the suspected agent and initiation of anti-inflammatory therapy are critical, the role and optimal timing of biologic agents remain uncertain and warrant further investigation. Blocking TNF-α dampened both adaptive and innate immune responses, contributing to the positive outcome.

Conclusion

The combination of etanercept and enoxaparin was associated with marked clinical improvement and normalization of laboratory parameters in a patient with SJS complicated by cytopenia and DIC. This case suggests that early administration of TNF-α inhibitors, such as etanercept, may be an effective therapeutic strategy for SJS, particularly in complex cases involving severe systemic complications.

病例報告 114_C058

Nivolumab 免疫相關不良事件：結腸炎之個案報告

Colitis as an Immune-Related Adverse Event Following Nivolumab Treatment for Advanced Esophageal Cancer: A Case Report

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Introduction

Immune checkpoint inhibitors (ICIs) targeting programmed cell death protein 1 (PD-1) have shown significant efficacy in advanced esophageal squamous cell carcinoma (ESCC). However, by enhancing immune activation, ICIs may cause immune-related adverse events (irAEs). Among these, gastrointestinal (GI) toxicities are clinically important, as immune-mediated colitis can progress rapidly and cause severe complications if unrecognized. We report a case of Nivolumab-induced colitis in a patient with metastatic ESCC, highlighting diagnostic challenges, treatment strategies, and potential future applications of bowel ultrasound in monitoring disease activity.

Case Report

A 64-year-old man with chronic obstructive pulmonary disease and hypertension was diagnosed with upper and lower ESCC in March 2025, staged as T1N2, T3N2, M1 (liver). He underwent a jejunostomy on March 8, chemotherapy with paclitaxel 60 mg and cisplatin 50 mg on March 26 and Nivolumab 120 mg on April 1.

Three days later, the patient developed watery diarrhea exceeding 10 episodes per day, accompanied by vomiting and abdominal pain. He was admitted on April 7 for dehydration and acute kidney injury. Laboratory findings included leukocytosis, hyponatremia, elevated BUN/creatinine, and metabolic acidosis. Stool studies were negative for infection etiologies. Abdominal CT showed no obstruction, while colonoscopy revealed diffuse erythematous and ulcerative colitis from the transverse to the sigmoid colon. Given the low likelihood of chemotherapy-induced diarrhea and absence of infection, a diagnosis of grade 3 ICI-induced colitis was established.

High-dose intravenous methylprednisolone was initiated, later transitioned to oral steroids by symptom resolution. He was discharged after one week. The following bowel ultrasound during Nivolumab rechallenge revealed colitis improvement.

Discussion

Gastrointestinal irAEs are among the most common toxicities of ICIs, occurring in 35–50% of patients[1]. Colitis occurs in 12–14% of PD-1 inhibitor users, typically onset within 4–8 weeks. The clinical presentation includes acute diarrhea, abdominal pain, and weight loss.

Diagnosis requires systematic exclusion of infectious etiologies and chemotherapy-induced diarrhea[2]. According to AGA, BSG, and NCCN guidelines, initial evaluation should include stool cultures, viral PCRs, and inflammatory markers, followed by colonoscopy with biopsy, which remains the gold standard for establishing diagnosis and grading severity[3,4].

Management depends on disease grade. While grade 1 colitis requires supportive care, patients

with grade ≧ 2 colitis requires corticosteroids. In steroid-refractory cases, infliximab or vedolizumab may be considered[5]. In this patient, early recognition and steroid initiation resulted in rapid improvement.

Future Clinical Implication:
As the use of ICIs expands, rechallenge after irAEs become an important clinical issue. Bowel ultrasound has emerged as a noninvasive bedside tool for dynamically assessing bowel wall thickness, vascularity, and peristalsis, offering real-time insights into inflammation. Compared to colonoscopy during anti-PD-1 rechallenge, the modality may detect worsening or recurrent colitis faster, safer and more patient-friendly. This approach may optimize patient selection and improve safety in future immunotherapy.

Conclusion

This case underscores the importance of early recognition and systematic evaluation of Nivolumab-induced colitis in ESCC. Prompt infections exclusion, endoscopic confirmation, and initiation of corticosteroids are essential for preventing life-threatening complications. Looking ahead, bowel ultrasound may aid in monitoring disease activity, especially for ICI rechallenge.

病例報告 114_C059

無抗磷脂抗體之 SLE 併腎病症候群發生肺栓塞：病例報告與治療挑戰

Pulmonary Embolism in Antiphospholipid-Negative SLE with Nephrotic Syndrome: A Case Report and Management Challenges

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Introduction

Thrombosis is a major complication of systemic lupus erythematosus (SLE), commonly associated with antiphospholipid syndrome (APS). However, APS-negative SLE patients may also develop severe thrombotic events due to immune dysregulation and nephrotic syndrome–related hypercoagulability.

Case Report

A 22-year-old woman previously healthy presented on 27 December 2024 with several days of bilateral leg swelling and pain. Labs revealed WBC 4,600/μL, Hb 9.3 g/dL, PLT 244×10^3/μL, albumin 1.4 g/dL, nephrotic-range proteinuria, hematuria, and strongly positive ANA, anti-Sm, anti-RNP; complement was low. She was diagnosed with systemic lupus erythematosus (SLE) with lupus nephritis and nephrotic syndrome, and treated with methylprednisolone pulse, prednisolone, MMF, hydroxychloroquine, plasma exchange, and belimumab. She got improved and was discharged on 10 January 2025.

On 24 January, she was readmitted for syncope and hypoxemia. Chest CT revealed bilateral pulmonary embolism with multifocal consolidations. She initiallly received catheter-directed thrombolysis, intravenous heparin, and intravenous thrombolysis for 11 days, followed by oral warfarin. Symptoms improved and she was extubated on 3 February 2025. She was discharged with warfarin, but INR was frequently subtherapeutic. Subsequently, she developed recurrent extensive DVT despite anticoagulation, reflecting the therapeutic challenges of venous thromboembolism in APS-negative SLE.

Discussion

Thromboembolism is a major complication of systemic lupus erythematosus (SLE). Although antiphospholipid syndrome (APS) is the strongest predictor, SLE itself confers a markedly increased risk of venous thromboembolism (VTE)]. Recent evidence shows that up to 30% of APS-negative patients develop thrombosis, particularly with nephrotic syndrome, hypoalbuminemia, complement consumption, and high disease activity. Recurrent events remain common despite warfarin, often due to subtherapeutic INR, while data on direct oral anticoagulants are limited. In extensive disease, catheter-directed therapy may be required. This case underscores that thrombotic risk in SLE extends beyond APS and highlights the need for vigilant monitoring and individualized anticoagulation strategies.

Conclusion

This case demonstrates that thrombotic events in systemic lupus erythematosus (SLE) may occur

even without antiphospholipid syndrome (APS), particularly in the presence of nephrotic syndrome, hypoalbuminemia, and complement consumption. Recent studies confirm that APS-negative or single-positive SLE patients with additional risk factors remain highly vulnerable to recurrence. Vigilant anticoagulation monitoring and individualized strategies are essential to reduce recurrence and improve outcomes.

病例報告 114_C060

末期腎病血液透析患者之意識障礙：一病例報告

Consciousness disturbance in an end-stage kidney disease patient under hemodialysis

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Introduction

Posterior reversible encephalopathy syndrome (PRES) is a rare but reversible disorder, often triggered by hypertension or renal failure. Hemodialysis patients are particularly vulnerable. We present a 57-year-old woman with ESRD who developed acute neurological disturbance during dialysis, diagnosed as PRES.

Case Report

A 57-year-old woman with a history of poor-controlled hypertension, type 2 diabetes mellitus and end stage renal disease under hemodialysis for three months presented with sudden onset upper limbs convulsive movements and altered mentation during dialysis. All her vital parameters were unremarkable, except systemic blood pressure was 224mmHg/92 mmHg. Neurological examination revealed loss of vision and presented bilateral Babinski extensor response. Blood examinations disclosed elevated levels of serum blood urea nitrogen (15.71mmol/L) and creatinine levels (680.83 μmol/L). Markers of autoimmune disease showed negative results. Computed tomography of the brain showed subtle loss of gray-white differentiation and effacement of cerebral sulci, without midline shift.

Discussion

In our case, computed tomography perfusion of the brain showed only small absolute mismatch volume in the brain parenchyma. Magnetic resonance imaging of the brain showed abnormal hyperintensity in the bilateral frontal-parietal-temporal-occipital lobes, bilateral thalamus, cerebellum and brainstem on T2-weighted imaging with fluid-attenuated inversion recovery, restricted diffusion on diffusion-weighted images, and diffuse microhemorrhages on susceptibility-weighted imaging. The impression was that of PRES. Differential diagnoses of PRES are broad. Infectious encephalitis is one of an important consideration. However, our patient remained afebrile. RCVS is another rare neurologic syndrome that share common risk factors with PRES. A thunderclap headache may be the initial clinical scenario of RCVS, which is not characteristic for the PRES. As in DDS cases, neurologic symptoms mostly appeared following initiation of dialysis treatment or after a sudden change of dialysis regimen. This made the conclusion of PRES more likely than DDS in this patient, who experienced a seizure attack and encephalopathy during dialysis. Brain CTP showed only a small area could be salvageable after reperfusion therapy, which was not the pattern of acute ischemic stroke.

PRES is defined as a rare disease of reversible subcortical vasogenic brain edema presented with acute neurological symptoms in the setting of blood pressure fluctuations, renal failure, autoimmune disorders, cytotoxic drugs, and pre-eclampsia or eclampsia. The typical imaging manifestations of PRES are findings of T2WI-hyperintense vasogenic edema. There is no

standardized management for PRES, but the disease is usually reversible when the precipitating cause is treated.

Conclusion

Diagnosis of PRES is not always straightforward, since there are several disorders, that presenting with an acute neurological deficit mimics PRES. Our case highlights the need for timely recognition of PRES to prevent devastating complications in a HD patient.

病例報告 114_C061

脾膿瘍併發破裂及術後繼發性血小板增多症

Splenic abscess complicated by peritonitis and secondary thrombocytosis after splenectomy in a 40-year-old male

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Introduction

Splenic abscess is a rare but serious infection that often occurs in immunocompromised patients. It can lead to life-threatening complications. This report presents a case with multiple comorbidities who developed splenic abscess complicated by rupture and secondary thrombocytosis following splenectomy.

Case Report

A 40-year-old male with history of type 2 diabetes mellitus, hypertriglyceridemia-induced pancreatitis with pseudocyst formation and gastric varices, treated by endoscopic sclerotherapy, presented with intermittent fever for one week.

Patient first experienced fever with chillness approximately one week before admission. Initially, he considered it to be a common cold. However, fever recurred 2 days before admission, accompanied with shortness of breath and left side chest tightness. COVID-19 and influenza rapid tests were negative, leading to the transfer to our ED. Vital signs were BP 104/62 mmHg, HR 125/min, RR 20/min, BT 40.5°C. Laboratory data revealed leukopenia (1,700/ul), anemia (9.2 g/dL), thrombocytopenia (121,000/ul), acute kidney injury, hyperbilirubinemia (D/T bilirubin: 1.03/ 2.09 mg/dL), elevated lactate and hs-CRP level (10.60mg/dL). CT scan showed a 55 mm splenic lesion with splenomegaly. Ceftriaxone, clindamycin and doxycycline were administered, while blood culture isolated 2 sets of Streptococcus anginosus and Parvimonas micra in one set. On the third day of admission, severe abdominal pain developed. Another CT indicatd free air formation outside of abscess and ascites; splenic rupture was suspected. Operation for drainage of intra-abdominal abscess revealed intra-abdominal abscess between spleen and cardiac of stomach, splenomegaly with varices but without cirrhosis. Due to adhesion of spleen with abscess cavity, splenectomy was performed. Afterwards, thrombocytosis was found (platelet: 1,248,000/ul) on POD8, which can be secondary thrombocytosis induced by surgery or infection, as genetic examination such as JAK-2 mutation, MPL, CALR was negative and vWF factor antigen and activity was within normal limit. Cytoreduction with hydroxyurea was prescribed. Afterwards, the patient's condition became stable, and platelet count decreased gradually. He was discharged to outpatient follow-up.

Discussion

Although splenic abscesses arise from hematogenous spread mostly. It can also result from contiguous contamination. Reports showed that pancreatic disease (such as chronic pancreatitis or pseudocyst) and bariatric procedures can lead to splenic abscess because of their anatomical proximity. Blood culture identified Streptococcus anginosus and Parvimonas micra, suggesting

bacterial translocation possibly related to his gastric varices. Postoperatively thrombocytosis could attribute to loss of splenic platelet regulation and surgical stress.

Conclusion

Splenic abscess should be considered in febrile patients with risk factors like pancreatic disease or portal hypertension. Typical symptoms such as abdominal pain or abdominal fullness didn't present in the patient initially. Timely diagnosis is the most important factor in successful treatment.

病例報告 114_C062

NK/T 細胞淋巴瘤以胰臟炎為最初表現：一個充滿挑戰的病例診斷

Acute Pancreatitis as the Initial Manifestation of Extranodal NK/T-cell Lymphoma: A Diagnostic Challenge

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Introduction

Extranodal NK/T-cell lymphoma is an aggressive EBV-associated non-Hodgkin lymphoma, most commonly presenting in the nasal cavity. Extranasal involvement included skin, gastrointestinal tract, testis. Here, we reported a rare case of acute pancreatitis caused by Extranodal NK/T-cell Lymphoma, which progressed to fatal multiorgan failure.

Case Report

This 61-year-old female without past medical history was admitted due to epigastric pain for one week. Previously, she had rhinorrhea and fever with night sweat for three months, along with oral ulcers and a 6-kg weight loss within six months. On arrival, her vital signs were stable. Lab data revealed elevated lipase 3468 U/L(8-58), triglycerides:197 mg/dL (<150). Abdomen computed tomography(CT) indicated acute pancreatitis without biliary obstruction. She denied alcohol consumption, medication, trauma, or abdominal procedure. Autoimmune workup showed negative. She later developed tender erythematous subcutaneous nodules on both legs, with skin biopsy confirming pancreatic panniculitis. Endoscopic ultrasound-guided fine-needle aspiration (EUS-FNA) of the pancreas showed hypoechoic and coarse parenchyma with several hyperechoic stranding and the pancreatic duct difficult to identify. The final pathology revealed extranodal NK/T-cell lymphoma. Ancillary study of IHC reveals CD3(+), EBER (+), CD8 (+), TIA-1 (+), CD20 (-), AE1/AE3 (-), CD5 (-), CD4 (-), TCR-delta (-), CD56 (-). However, the patient developed respiratory failure with pulmonary infiltrates. Bronchoalveolar Lavage was negative for pathogens but suggested Hemophagocytic Lymphohistiocytosis(HLH), supporting lymphoma. Later, septic shock likely from pancreatitis-related intra-abdominal infection (VRE bacteremia) occurred. Despite treatment, the patient expired from multi-organ failure.

Discussion

Gallstones and alcohol are the most common causes of acute pancreatitis; less common ones include medications, infection, hypertriglyceridemia, interventions, trauma, and rarely, tumors. Primary pancreatic involvement by extranodal NK/T-cell lymphoma is extremely rare, first reported in 2013. In our case, no clear etiology of pancreatitis was identified initially. Surprisingly, the final pathology via EUS-FNA revealed unexpected findings, even though a neoplastic etiology had been initially suspected. Hence, we hypothesize the pancreatitis resulted from pancreatic infiltration by lymphoma. Rapid disease progression precluded further imaging and biopsy to confirm the primary site.

Conclusion

Extranodal NK/T-cell lymphoma presenting as acute pancreatitis is rare and challenging to diagnose. Early recognition of atypical symptoms (rhinorrhea, oral ulcers) and B symptoms (fever, wasting, night sweats) may prompt timely tissue sampling when common causes are excluded. Malignancy, though rare, should be included in the differential diagnosis of pancreatitis.

病例報告 114_C063

心臟置入物黴菌感染：治療挑戰與無導線節律器之角色

Major Fungal CIED Infection in an Immunocompromised Host: Challenges and the Role of Leadless Pacemaker Therapy

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Introduction

Infection is a serious complication of cardiac implantable electronic device (CIED) therapy, with an incidence rate of 0.6% to 3.4% within 12 months post-implantation. Risk factors include advanced age, heart failure, diabetes, immunocompromise, anticoagulant or antiplatelet therapy, prior CIED infection, and hematoma. Mortality within 30 days or during hospitalization is approximately 5–8%. Management involves device removal, debridement, and antibiotic therapy. We present a case of CIED pocket fungal infection successfully treated with debridement and prolonged antibiotic use, followed by leadless pacemaker implantation.

Case Report

An 83-year-old man with permanent atrial fibrillation was admitted due to a CIED pocket infection. One month prior, he was diagnosed with atrial fibrillation accompanied by severe bradycardia and hypotension, and a pacemaker was implanted at another hospital. However, the wound over the pacemaker site healed poorly and developed a hematoma. Needle aspiration was performed, but purulent discharge and swelling persisted. The patient was subsequently transferred to our hospital for further management.

We performed removal of the generator and leads, accompanied by debridement of necrotic tissue and purulent material. Continuous intravenous isoproterenol infusion and oral theophylline were administered to maintain an adequate heart rate. Empiric antimicrobial therapy with piperacillin/tazobactam and teicoplanin was initiated for suspected pacemaker pocket infection, along with wet dressings of the open wound. Following debridement, the patient developed remittent fever. Intravenous fluconazole was subsequently introduced after Candida albicans was identified in the pus culture. The fever gradually subsided, and consultation with a plastic surgeon was obtained for negative pressure wound therapy, followed by wound closure. On hospital day 29, a leadless pacemaker (Medtronic Micra VR) was successfully implanted. The patient remained clinically stable and was discharged on hospital day 33.

Discussion

Complete device removal is essential for CIED infections. Isolated pocket infections should be treated with a 14-day course of antibiotics following device removal. Most CIED infections are monomicrobial, with fungal pathogens accounting for approximately 2% of cases. While empirical antifungal therapy may be considered, further evidence is required. Reviewing the patient's medical history, he had experienced COVID-19 pneumonia two months prior to this admission. Since then, he had been receiving oral prednisolone, which rendered him immunocompromised and predisposed to fungal infection as well as impaired wound healing. Preventive strategies for

CIED infections include administration of a single preoperative dose of cefazolin, use of antibiotic envelopes, and minimization of pocket hematoma formation. However, these measures would be insufficient in an immunocompromised host such as this patient. Therefore, implantation of a leadless pacemaker was chosen to minimize the risk of impaired wound healing and recurrent infection.

Conclusion

Major CIED-related fungal infections are rare but potentially fulminant complications of device therapy. We present this case to raise awareness among clinicians of this uncommon but serious entity. Furthermore, leadless pacemaker implantation may represent a valuable strategy to reduce the risk of recurrent pocket infections in susceptible hosts.

病例報告 114_C064

以 Linezolid 合併 Fosfomycin 成功治療對 Daptomycin 不具感受性且 Vancomycin 中間抗藥性之金黃色葡萄球菌菌血症之病例報告

Successful Treatment of Daptomycin Non-Susceptible, Vancomycin-Intermediate *Staphylococcus aureus* Bacteremia with Linezolid and Fosfomycin: A Case Report

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Introduction

Staphylococcus aureus remains a leading cause of severe infections, with treatment increasingly complicated by rising antimicrobial resistance. While vancomycin and daptomycin are key therapies for methicillin-resistant *S. aureus* (MRSA), strains with reduced susceptibility to both—specifically daptomycin non-susceptible and vancomycin-intermediate phenotypes—pose significant clinical challenges. Here, we report a case of invasive infection caused by a daptomycin non-susceptible and vancomycin-intermediate *S. aureus* (DNS-VISA) strain, highlighting diagnostic and therapeutic challenge in managing this rare but increasingly important resistance profile.

Case Report

A 55-year-old male with hypertension, diabetes mellitus, and end-stage renal disease (ESRD) on hemodialysis presented to the emergency department with several days of redness, swelling, and gangrenous changes in his right foot. He was admitted with a diagnosis of right leg cellulitis with superimposed osteomyelitis.

Right below-knee amputation was performed on the second day of admission. MRSA was repeatedly isolated from blood cultures. Spinal magnetic resonance imaging (MRI) revealed osteomyelitis with abscess formation at the C4–C7 and T1 vertebral levels. C6 corpectomy and C4/5, C7/T1 discectomy with abscess drainage was performed.

The patient was initially treated with teicoplanin, and later switched to daptomycin. Despite three weeks of daptomycin therapy, blood cultures grew DNS-VISA. Antibiotic therapy was then switched to a combination of linezolid and fosfomycin. Positron emission tomography-computed tomography (PET-CT) subsequently revealed an infectious process involving his arteriovenous fistula (AVF), leading to a partial AVF resection. Bacteremia was cleared by the 28th day of admission.

In the following hospital course, despite suffering several nosocomial infections and even an in-hospital cardiac arrest, all subsequent cultures from various sampling sites remained negative for *S. aureus* while on intravenous linezolid and fosfomycin. After 77 and 75 days of treatment, respectively, both agents were transitioned to oral linezolid plus rifampicin, based on a prior fosfomycin susceptibility result by disc diffusion. The patient was discharged after 112 days of hospitalization, without requiring mechanical ventilation or vasopressors.

Discussion

DNS-VISA, although rare, are increasingly reported in cases of prolonged or inadequately treated *S. aureus* infections. Clinical management of DNS-VISA infections is particularly challenging. Combination therapy—often involving a cell wall or membrane-active agent paired with a synergistic partner—is frequently necessary. While clinical experience remains limited, there is robust in vitro and preclinical evidence supporting the synergistic activity of linezolid and fosfomycin against VISA and other drug-resistant *S. aureus* isolates. Nevertheless, optimal antimicrobial therapy must be paired with prompt source control, which remains the cornerstone of successful treatment.

Conclusion

This case highlights the importance of early identification, aggressive source control, and a successful experience of linezolid plus fosfomycin to treat DNS-VISA bacteremia. Continued clinical vigilance and research are essential to optimize management strategies for DNS-VISA infection.

病例報告 114_C065

類似帕金森氏症的肝性腦病變的罕見病因：自發性門體分流引起的高氨血症

An Uncommon Cause of Hepatic Encephalopathy Mimicking Parkinsonism: Hyperammonemia From A Spontaneous Portosystemic Shunt

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Introduction

Ammonia is an endogenous neurotoxin mainly arises from the breakdown of amino acids and dietary protein. The liver converts ammonia into urea through urea cycle and the kidney excretes urea via urine under normal physiological conditions. Any impairment during the elimination of ammonia will lead to the accumulation of ammonia, hyperammonemia. Common etiologies of hyperammonemia include liver dysfunction, high-protein diet, severe infections, intoxication, and inherited urea cycle disorders. Neurological manifestations are the most common clinical presentation of hyperammonemia but are often misdiagnosed as neuropsychiatric disorders. Therefore, accurate differential diagnosis is critical for unknown origin of hyperammonemia.

Case Report

A 69-year-old female with medical history of type 2 diabetes mellitus, hypertension, and rheumatic heart disease status post mitral and aortic valve replacement complicated with severe stenosis of aortic prosthesis under warfarin presented to emergency department due to progressive drowsiness for several days.

According to medical records, this patient suffered from intermittent consciousness disturbance with behavior change, dizziness, confusion, disorientation, slurred speech, and memory impairment for years. She was previously diagnosed with Parkinsonism and was treated with antiparkinsonian medications.

At emergency department, laboratory data showed platelet was 150000 per microliter, ammonia was 264 μg per deciliter, aspartate amino transferase was 27 IU per liter, alanine amino transferase was 28 IU per liter, albumin was 3.7 g per deciliter, and total bilirubin was 1.1 mg per deciliter. The hepatic encephalopathy was diagnosed; however, abdominal ultrasonography revealed no definitive evidence of liver cirrhosis and the patient had neither hepatitis B or hepatitis C virus infection nor a history of alcohol consumption. Abdominal computed tomography was arranged and identified a portosystemic shunt between the inferior vena cava and the inferior mesenteric vein.

The patient underwent embolization of the portosystemic shunt. Followed laboratory data 5 days after the procedure demonstrated a marked decreased in serum ammonia to 86 μg/dL, accompanied by significant improvement of consciousness. She remained stable, and subsequent outpatient follow-up visits demonstrated no further abnormalities.

Discussion

The etiology of hyperammonemia can be broadly categorized into hepatic and non-hepatic causes. Ammonia, the neurotoxin, bypasses portal circulation through portosystemic shunt to systemic

circulation leading to the accumulation of ammonia and type B hepatic encephalopathy[1].
The causes of portosystemic shunt formation can be congenital vascular abnormality, hepatic parenchyma degeneration, anastomosis after trauma or abdominal surgery, or sometimes remained idiopathic. These shunts can be classified into five types based on anatomical location, with type 3 (extrahepatic type) being the most common type[2]. Females were predominant and median age at diagnosis was 53 years[3].
To make an accurate diagnosis of portosystemic shunt, imaging studies remain gold standard, especially abdominal CT and magnetic resonance imaging. Treatments for hyperammonemia secondary to portosystemic shunt includes symptomatic treatment and definitive interventions. The choice of treatment depends on the severity of symptoms. For patients with severe or refractory symptoms, intervention such as surgical ligation, occlusion, or embolization of the shunt are recommended[3].

Conclusion

Though the rare incidence,portosystemic shunt should be taken into consideration in patients presenting with unexplained hyperammonemia and neuropsychiatric symptoms. Early and accurate diagnosis is essential to avoid misdiagnosis and unnecessary treatments, thereby improves patients' outcomes and quality of life.

病例報告 114_C066

臨床醫生和實驗室工作人員之間溝通在 Bukholderia 菌種鑑定中的重要性

Importance of Communication between Clinician and Lab Staffs in Species Identification of *Burkholderia*

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Introduction

Burkholderia species is not a common isolate in clinical specimen. The most common species is B. cepacia complex. Other less common isolates are B. mallei and B. pseudomallei. Burkholderia pseudomallei is a category B critical biological agent and should be handled in biosafety cabinet. It is the etiology of melioidosis, which also a reportable infectious disease in Taiwan. Laboratory diagnosis of melioidosis can be difficult as the bacterium is often not readily isolated from clinical specimens and may not be correctly identified even when isolated. We report two cases of Burkholderia infection and demonstrate the importance of communication between clinicians and laboratory staffs in correct identification of B. pseudomallei.

Case Report

Case 1: A 67 y/o male patient with history of pulmonary TB, COPD, pneumoconiosis and adrenal insufficiency presented to emergency department with high fever and hypotension. Blood culture yielded GNB isolated in 3 sets of culture. He did not receive invasive procedure recently before this infection episodes. Though MALDI-TOF identified as B. pseudomallei, the morphology on culture plates did not show typical rough, flat and wrinkled appearance and API 20NE identified as B. cepacia. The patient was treated as B. cepacian sepsis.
Case 2: A 31 y/o pregnant woman with Sjogren's syndrome presented with left buttock cellulitis. Pus culture yielded GNB. The MALDI-TOF identified B. pseudomallei. There was rough, flat and wrinkled appearance on culture plate. But the identification with API 20 NE yielded B. cepacian. Wound care with topical agent was given.

Discussion

X

Conclusion

The case 1 had a relapse of sepsis with new complication of prostate abscess. Searching PubMed with key words, Burkholderia, bacteria and prostate abscess, the reported etiology revealed B. pseudomallei only. The morphology of B. psedomallei can be mucoid. Due to rarity of B. pseudomallei, and lack of clinical information, species identification just by API 20 NE can lead to misidentification. Further molecular identification may be necessary in those isolates with discordant results between clinical course and laboratory identification. Our experience highlights the importance of communication between clinician and microbiologic staffs in Burkholderia species identification.

病例報告 114_C067

案例討論: 心肌梗塞後心室中膈缺損

Post-Myocardial Infarction Ventricular Septal Defect: Case Report

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Introduction

Post-Infarction Ventricular Septal Defect (PIVSD) is one of the mechanical complications following Acute Myocardial Infarction (AMI). Mortality is high. Surgical timing remained debatable. Immediate surgical intervention is with surgical risk but preferable. Delayed surgery while hemodynamically stable showed favorable outcome in previous studies, but it is suspected to be associated with selection bias. This is a case of Non-ST segment elevation infarction (NSTEMI) progression into STEMI with RCA total occlusion, later diagnosed with PIVSD.

Case Report

This is a 69-year-old female with past medical history of Essential Hypertension. She was presented to our hospital due to acute chest pain with radiation to lower jaw and mandible with diaphoresis, started since 4:00. Chest X-Ray showed borderline heart size. Electrocardiogram revealed normal sinus rhythm progressive change of inferior leads to ST elevation and T wave inversion on day 2 after admission while she is under treatment with Dual Antiplatelet Therapy (DAPT) with Aspirin, Ticagrelor and Heparinization. Emergent coronary angiography and intervention was done. Total occlusion from mid Right Coronary Artery was managed by thrombosuction and balloon dilatation. The final angiogram revealed TIMI flow of 3. IABP was inserted as a support for coronary flow. A grade IV holosystolic murmur was most prominent on apex. Bedside echocardiogram revealed Ventricular Septal Defect (VSD) of size 2.24 cm on posterior septal wall. After cardiovascular surgeon consultation, an emergent VSD repair surgery was done. The patient developed Disseminated Intravascular Coagulation (DIC) with massive bleeding.

Discussion

PIVSD is a rare but high morbidity and mortality disease. First 24 hours after AMI intramural hematoma dissection showed up, hemorrhage flew into ischemic myocardium region. After 5 days would form coagulation necrosis. In the absence of reperfusion therapy, coagulation necrosis sets in, then neutrophils enter the necrotic zone, while neutrophils undergo apoptosis which may release lytic enzymes damaging necrotic myocardium and lead to VSD.

Echocardiogram is the first choice of diagnosis VSD, to access both VSD morphology and hemodynamic data.

- Parasternal long axis view : perimembranous outlet and muscular defects of mid anterior septum
- Parasternal short axis view
  Great vessels Level : perimembranous VSDs at 9–12 o'clock and outlet juxta-arterial VSDs at 12–3 o'clock
  Ventricles Level: Muscular VSD

- Apical 5-chamber : central perimembranous defect

Predictors for VSD including age, female, prior stroke, STEMI, anterior wall infarction, single vessel, high Killip class, and total occlusion without collateral support. Besides, people with STEMI for over 72 hours and RV involvement would increase the risk of VSD.

While ventricular septum is supplied by the septal perforating branches, which resulted from left anterior descending or posterior descending coronary artery. LAD artery supplies anterior wall of septum; LAD infarction may lead to anterior wall rupture. Posterior PIVSD may had serpiginous intramyocardial dissection tracts reaching the LV free wall and with poor prognosis.

Timing of MI-VSD closure remains ongoing debate. Current available evidence suggested that delayed surgical repair may be a survival benefit however still need prospective study to better determine optimal timing of intervention.

病例報告 114_C068

疑似 Methylphenidate 誘發之心肌炎：病例報告

Drug-Induced Myocarditis Following Methylphenidate Therapy: A Case Report

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Introduction

Myocarditis is most caused by viral or post-viral infections, but drug-induced hypersensitivity reactions are also recognized, involving agents such as anticonvulsants, antipsychotics, antibiotics, and catecholaminergic stimulants. Methylphenidate (Ritalin), a CNS stimulant that increases dopamine and norepinephrine activity, is used to treat ADHD and narcolepsy and may be prescribed off-label for performance enhancement. We present a case of methylphenidate-induced myocarditis

Case Report

A 33-year-old woman with a history of anxiety disorder on psychiatric medications presented with a three-day history of palpitations, progressive dyspnea, orthopnea, and orange-colored urine. She had no systemic illness but reported use of multiple medications, including cephradine, acetaminophen, diclofenac, and alprazolam. Furthermore, a methylphenidate overdose was suspected, as the patient reportedly ingested eight tablets within a single day to sustain work-related concentration. On arrival, her blood pressure was 87/60 mmHg and heart rate 67 bpm. Electrocardiography revealed sinus tachycardia with nonspecific ST-segment changes. Echocardiography revealed left ventricular ejection fraction of 45% with apical hypokinesia. Laboratory testing showed leukocytosis, elevated troponin-I (0.2956 ng/mL), CK-MB (8.5 ng/mL), markedly increased BNP (11,013 pg/mL), and elevated thyroid-stimulating hormone. NSTEMI was initially suspected; however, coronary angiography revealed no obstructive disease. Myocarditis was subsequently considered, and she received aspirin, furosemide, and colchicine. Viral PCR was negative, while serology indicated positive CMV IgG. During hospitalization, her cardiac biomarkers improved, pulmonary edema resolved, and no arrhythmia was documented. She was discharged on medical therapy including aspirin, bisoprolol, isosorbide, and ivabradine. At outpatient follow-up, she remained stable with symptom resolution.

Discussion

Myocarditis is an inflammatory disorder of the myocardium that can result from infectious, autoimmune, or drug-induced etiologies. Among pharmacologic causes, hypersensitivity myocarditis has been associated with antibiotics, antipsychotics, anticonvulsants, and several catecholaminergic stimulants. Methylphenidate, a central nervous system stimulant commonly prescribed for attention-deficit/hyperactivity disorder (ADHD) and narcolepsy, exerts its effects primarily by inhibiting dopamine and norepinephrine reuptake. Although its cardiovascular adverse effects—such as tachycardia, hypertension, and arrhythmia—are well recognized, myocarditis attributable to methylphenidate use is rarely reported.

In the present case, the patient developed chest discomfort, dyspnea, and elevated cardiac

biomarkers in the absence of obstructive coronary artery disease. The combination of reduced left ventricular systolic function, apical wall motion abnormalities, and biomarker elevation raised the suspicion of acute coronary syndrome; however, coronary angiography excluded significant stenosis. Viral testing was negative, and the patient's history of intermittent methylphenidate use suggested a potential drug-induced mechanism. The temporal association, exclusion of alternative etiologies, and subsequent clinical improvement after drug discontinuation and medical therapy strongly support a diagnosis of methylphenidate-induced myocarditis.

Conclusion

This case highlights the importance of considering drug-induced myocarditis in patients presenting with acute heart failure or myocardial injury without obstructive coronary artery disease, particularly when stimulant medications are involved. Clinicians should obtain a detailed medication history, including non-prescribed or intermittently used agents, to avoid misdiagnosis and unnecessary interventions. Early identification and cessation of the offending drug, combined with guideline-directed medical therapy, can lead to favorable outcomes.

病例報告 114_C069

非覆膜支架導致致命膽囊破裂：囊管評估與引流策略的臨床啟示

Gallbladder Rupture After Uncovered SEMS: Lessons for Cystic Duct Evaluation and Drainage Strategy

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Case Report

A 71-year-old fish salesman with alcoholism, hepatitis C virus infection treated with direct-acting antivirals (DAA) achieving sustained virological response, type II diabetes mellitus, hypertension, and benign prostate hypertrophy presented with intermittent epigastric pain for one month and fever. Physical examination showed epigastric tenderness. Laboratory tests revealed ALT 230 U/L, AST 193 U/L, alkaline phosphatase 336 U/L, total bilirubin 5.7 mg/dL, and direct bilirubin 2.99 mg/dL. CT showed bilateral intrahepatic duct (IHD) dilatation, common bile duct (CBD) wall thickening, and left lobe hypodense nodules. ERCP on day 2 showed a 2 cm stricture at the common hepatic duct (CHD), hepatic bifurcation, and dilated right IHDs (Fig. 1A), suggesting Klatskin tumor Bismuth IV. After sphincterotomy, biopsy, and balloon dilation, two plastic stents were placed in the right posterior and anterior IHDs (Fig. 1B). Recurrent fever and jaundice required repeat ERCP on day 17 with stent replacement. Liver biopsy confirmed mixed hepatocellular carcinoma and cholangiocarcinoma.

Five days post-discharge, he was readmitted for recurrent fever and right upper quadrant pain. ERCP showed a 2–3 cm stricture at proximal CHD and bilateral IHD dilatation. A Soehendra stent retriever was used to pass a tight right posterior IHD stricture, followed by 8 mm balloon dilation. Three uncovered SEMS (8 mm × 100 mm, Wallflex) were placed into the right posterior, left, and right anterior IHDs (Fig. 2). Four days later, acute cholecystitis with gallbladder rupture occurred (Fig. 3A), requiring percutaneous transhepatic gallbladder drainage (PTGBD) (Fig. 3B). He developed septic shock, delirium with self-removal of PTGBD, aspiration pneumonia, and required intubation. He died of multiple organ failure on day 34 of his second hospitalization.

Discussion

Gallbladder rupture following the placement of uncovered self-expandable metallic stents (SEMS) represents a rare but life-threatening complication. Acute cholecystitis occurs in approximately 3–12% of patients following uncovered SEMS placement and up to 25% following covered SEMS placement for malignant biliary obstruction [1,2]. Identified risk factors include tumor involvement of the cystic duct [2], inadvertent contrast injection into the gallbladder during ERCP [3], and multiple SEMS configurations across the cystic duct orifice[4].

Evidence-based preventive strategies can mitigate post-stenting gallbladder complications. First, pre-deployment imaging and careful cholangiographic assessment of the cystic duct are critical. Non-opacification should raise suspicion for obstruction risk. Second, avoiding unnecessary contrast injection into the gallbladder is recommended, as this has been linked to higher rates of post-ERCP cholecystitis3. Third, prophylactic gallbladder drainage, either endoscopic or percutaneous, may be considered in patients with hilar tumors involving the cystic duct. Finally,

early post-procedure monitoring is essential, as most episodes of cholecystitis occur within the first week following SEMS placement[4].

病例報告 114_C070

肝癌接受肝移植後侵襲性肺麴菌感染的病例報告

A case report of invasive pulmonary aspergillosis in a liver transplant recipient

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Introduction

Invasive pulmonary aspergillosis (IPA) is a major cause of morbidity and mortality among solid organ transplant recipients, particularly those with high-dose immunosuppression. Rare species such as Aspergillus lentulus pose additional diagnostic and therapeutic challenges due to reduced susceptibility to antifungals.

Case Report

A 53-year-old man with type 2 diabetes mellitus and alcoholic liver cirrhosis underwent deceased donor liver transplantation (DDLT) in 2013 and re-transplantation with splenectomy for graft failure in 2024. His post-transplant course was complicated by acute rejection requiring pulse corticosteroids, and CMV reactivation treated with valganciclovir. He was maintained on prednisolone, tacrolimus, and Mycophenolate mofetil.

He was sent to the emergency department in April 2025. He presented with fever, jaundice, cough, and oral candidiasis, which led to hospitalization. Lab data showed elevated CRP, hyperbilirubinemia, and impaired renal function. The Chest X ray and chest CT revealed bilateral infiltrates. Sputum smear showed yeast. Empirical cefotaxime and liposomal amphotericin B were started. Broncho-alveolar lavage grew Aspergillus fumigatus and Aspergillus lentulus, with fungal hyphae on gram stain. Anidulafungin and oral voriconazole were added for combination.

The patient developed acute respiratory distress syndrome (ARDS) and a left pneumothorax, necessitating chest tube drainage. With a combination of antifungal, antibacterial, and supportive treatments, the pneumothorax resolved, and the chest tube was successfully removed. The patient was discharged with oral Voriconazole. At the follow-up appointment in the outpatient department, the patient reported no further cough. A chest X-ray indicated improved patchy opacity in both lung fields. Tacrolimus was administered two weeks later, and Mycophenolate was resumed three months post-discharge.

Discussion

This case highlights IPA in a high-risk transplant recipient exposed to multiple immunosuppressive agents. A. lentulus infection is notable given its reduced azole susceptibility. Combination of antifungal therapy should be considered. Pneumothorax, though uncommon, can complicate IPA due to cavitary lung destruction. Careful adjustment of immunosuppression, antifungal choice, and supportive care were essential for clinical recovery.

Conclusion

IPA remains a life-threatening infection in liver transplant recipients. Early recognition, appropriate antifungal therapy, and management of complications such as pneumothorax are crucial to

improving outcomes.

病例報告 114_C071

[診斷挑戰]：透析無效之類肉瘤病相關高血鈣，對類固醇反應良好

Steroid-Responsive Hypercalcemia Secondary to Sarcoidosis, Refractory to Dialysis: A Diagnostic Challenge

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Introduction

Sarcoidosis is a multisystem granulomatous disorder of unknown etiology, most commonly involving the lungs and intrathoracic lymph nodes. Hypercalcemia occurs in approximately 2–20% of patients with sarcoidosis and is attributed to extrarenal production of 1,25-dihydroxyvitamin D_3 by activated macrophages within granulomas. This dysregulated vitamin D metabolism enhances intestinal calcium absorption and bone resorption, leading to hypercalcemia and subsequent renal impairment. Prompt recognition of sarcoidosis-induced hypercalcemia is crucial, as untreated hypercalcemia can result in significant morbidity, including acute kidney injury (AKI), altered mental status, and arrhythmias. Standard treatment of hypercalcemia includes intravenous hydration and loop diuretics, while in severe or refractory cases, hemodialysis may be required. Persistent hypercalcemia should raise suspicion for sarcoidosis, in which corticosteroids effectively suppress macrophage 1α-hydroxylase activity. Here, we present a patient with constitutional symptoms, hypercalcemia, and AKI, whose serum calcium levels remained elevated despite hemodialysis, but improved significantly following corticosteroid therapy, supporting the diagnosis of sarcoidosis.

Case Report

A 59-year-old previously healthy man, an ex-smoker and shoe repairman, presented with general weakness and poor appetite for one week. The family also reported diffuse musculoskeletal pain, followed by intermittent fever and altered consciousness. Initial laboratory studies revealed leukocytosis (WBC 16400/uL), hypercalcemia (16.2 mg/dL), AKI (BUN 61 mg/dL; creatinine 8.15 mg/dL), elevated CRP (19.56 mg/dL), and normal intact parathyroid hormone level (17.3 pg/mL). Chest X-ray showed prominent bilateral hilar shadows. Chest computed tomography (CT) revealed bilateral hilar lymphadenopathy, ground-glass opacities, and subcentimeter pulmonary nodules. Despite aggressive intravenous hydration and furosemide, the patient's hypercalcemia worsened, peaking at 17 mg/dL. Temporary hemodialysis was initiated on hospital day 2 for refractory hypercalcemia and worsening renal function; however, serum calcium levels remained elevated. Tumor markers and tuberculosis workup were unremarkable. Given persistent fever and altered mental status, brain CT and lumbar puncture were performed, both yielding negative findings. Empiric antibiotics were switched from cefotaxime to ceftriaxone for suspected central nervous system infection. Intravenous hydrocortisone was initiated concurrently. Following an increased dose of corticosteroids, the patient's fever subsided, consciousness improved, and serum calcium levels showed a marked decline to normal (9.8 mg/dL). His renal function gradually improved after discontinuation of dialysis, with a serum creatinine of 1.15 mg/dL at follow-up after discharge.

Discussion

The patient's hypercalcemia was refractory to standard therapies, including aggressive hydration, loop diuretics, and hemodialysis, but showed a rapid and sustained response following initiation of corticosteroids. This observation highlights the crucial role of glucocorticoids in inhibiting granuloma-mediated extrarenal 1α-hydroxylase activity, which leads to excessive 1,25-dihydroxyvitamin D production and subsequent hypercalcemia in sarcoidosis. Taken together, the clinical presentation, characteristic radiologic findings, and steroid responsiveness strongly support sarcoidosis as the underlying etiology.

Conclusion

This case underscores the importance of considering sarcoidosis in patients with severe hypercalcemia unresponsive to conventional treatments. In such refractory cases, a dramatic response to corticosteroids may provide therapeutic benefit and serve as a key diagnostic clue. Clinicians should remain vigilant for steroid-responsive hypercalcemia as a potential manifestation of sarcoidosis.

病例報告 114_C072

從皮疹到心臟：18 歲男性水痘帶狀疱疹病毒心包炎病例報告

From Rash to Heart: Varicella-Associated Pericarditis in a Young Male

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Introduction

Varicella-zoster virus (VZV) is a ubiquitous human herpesvirus that typically causes self-limited varicella or herpes zoster but may also result in severe complications. Among these, pericarditis is exceedingly rare, most often reported in immunocompromised hosts or individuals with autoimmune diseases. VZV pericarditis may mimic other infectious or autoimmune causes, creating diagnostic challenges, and can progress to life-threatening cardiac tamponade if not promptly recognized. Here, we present an unusual case of VZV-associated pericarditis in an adolescent with palindromic rheumatism receiving immunosuppressive therapy, underscoring the importance of early diagnosis and intervention.

Case Report

An 18-year-old male with a three-year history of palindromic rheumatism on sulfasalazine, hydroxychloroquine, prednisolone, and celecoxib presented with acute chest tightness and fever up to 38.9°C. Examination revealed tachycardia, an S3 gallop without murmurs, and multiple pustular nodules on his chest, back, elbows, ankles, and tongue. Laboratory data demonstrated leukocytosis (18.2 × 10^9/L, 80% neutrophils), anemia (hemoglobin 11.6 g/dL), and elevated hsCRP (16.4 mg/L). Electrocardiogram showed sinus tachycardia with diffuse ST elevation, chest radiography indicated cardiomegaly with left pleural effusion, and echocardiography revealed mild pericardial effusion with preserved ventricular function. VZV PCR from vesicular fluid and throat swab confirmed breakthrough varicella infection. He was started on intravenous acyclovir and ceftaroline, with continuation of corticosteroids. Symptoms improved initially; however, three days later he developed hypotension, tachycardia, fever, and recurrent chest pain. Bedside sonography demonstrated massive pericardial effusion with right ventricular diastolic collapse, consistent with cardiac tamponade. An emergent pericardial window was performed, leading to clinical stabilization. He completed antiviral therapy and was discharged with outpatient follow-up.

Discussion

This case highlights the uncommon but clinically significant complication of VZV pericarditis in immunosuppressed patients. Typical manifestations include chest pain, fever, dyspnea, diffuse ST elevation, and echocardiographic evidence of effusion. Diagnosis is confirmed by viral detection from vesicular or respiratory samples. Management centers on timely initiation of antiviral therapy, supportive measures, and pericardial drainage in tamponade. Literature review reveals only a few dozen reported cases worldwide, illustrating its rarity. The occurrence of pericarditis in this vaccinated adolescent raises concerns about waning immunity and breakthrough infections, particularly in patients with autoimmune disease and long-term corticosteroid exposure. These

findings emphasize the need for heightened clinical suspicion and awareness among physicians.

Conclusion

VZV infection should be recognized as a rare but important cause of pericarditis, especially in immunocompromised hosts or those with autoimmune conditions. Prompt diagnosis, early antiviral therapy, and appropriate pericardial drainage are essential to prevent severe outcomes such as cardiac tamponade. Breakthrough infections, even after vaccination, highlight the importance of ongoing vigilance and continued research into vaccine durability and long-term protective immunity.

病例報告 114_C073

成人肺炎黴漿菌感染併發急性心肌炎與心衰竭：病例報告

Acute Myocarditis and Heart Failure Secondary to Mycoplasma pneumoniae Infection in a Healthy Adult: A Case Report

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Introduction

Mycoplasma pneumoniae can cause myocarditis and, in some cases, heart failure. Multiple case series and reviews document the association between acute M. pneumoniae infection and cardiac involvement, including myocarditis, perimyocarditis, and pericarditis. Clinical presentations of patients include a spectrum from mild myocardial enzyme elevation to severe cardiac dysfunction and heart failure.

Case Report

A 24-year-old healthy female, as a playground staff, presented with high-grade fever, chills, cough, myalgia, rhinorrhea, and painful oral ulcers in 2025. Feb. Despite outpatient therapy, she developed progressive chest tightness, pleuritic pain, palpitations, and dyspnea. Chest radiography showed cardiomegaly, pulmonary edema, pleural effusion, and left lower lobe consolidation. Laboratory studies revealed elevated CRP, mild transaminitis, and hypokalemia. She was admitted with a tentative diagnosis of atypical pneumonia complicated by myocarditis and acute heart failure.

Lab data including Mycoplasma pneumoniae IgM, PCR, and cold agglutinin were positive. Initial ceftriaxone plus azithromycin was revised to cefotaxime and later doxycycline due to hepatitis and suspected macrolide resistance. She developed arrhythmias with elevated cardiac enzymes and NT-proBNP, requiring diuretics and cardiology consultation. Echocardiography showed preserved systolic function (LVEF 67%), though acute decompensated heart failure could not be excluded. Splenomegaly was noted on sonography. Corticosteroid therapy (methylprednisolone, prednisolone) was initiated for Mycoplasma-associated myocarditis.

She was diagnosed with Mycoplasma pneumoniae pneumonia complicated by myocarditis, acute heart failure with pulmonary edema, pericardial and pleural effusions, hepatitis, hemolysis, and splenomegaly. With doxycycline and steroids, she improved clinically and was discharged on 3/6.

Discussion

Evidence includes a case series in which 13 patients with active M. pneumoniae infection developed acute pericarditis or perimyocarditis; four of these patients experienced heart failure, and two died, with residual cardiac compromise attributed at least in part to the infection. Another report describes myocardial dysfunction mimicking acute myocardial infarction in a patient with M. pneumoniae infection, with marked clinical deterioration during the recovery phase. Additional literature reviews and case reports confirm that M. pneumoniae can cause myocarditis, sometimes with long-term sequelae or fatal outcomes.

In pediatric populations, elevated myocardial enzymes (e.g., hs-TroI, CK-MB) during acute M.

pneumoniae pneumonia correlate with clinical signs of myocardial damage, supporting the link between infection and cardiac injury. The pathogenesis is thought to involve both direct microbial effects and immune-mediated mechanisms.

While viral etiologies predominate in myocarditis, bacterial causes such as M. pneumoniae are recognized, especially in cases with concurrent respiratory symptoms and extrapulmonary manifestations. The American Heart Association notes that myocarditis can present with acute heart failure, and that non-viral pathogen, including bacteria, should be considered in the differential diagnosis.

Conclusion

This case highlights Mycoplasma pneumoniae infection can cause myocarditis, perimyocarditis, and heart failure. Clinical manifestations range from mild myocardial enzyme elevation to severe cardiac dysfunction. Cardiac injury results from direct microbial effects and immune-mediated mechanisms. Recognition of bacterial myocarditis is essential, particularly in patients with respiratory symptoms and extrapulmonary involvement. Early diagnosis and treatment can prevent more severe complications and improve patient outcomes.

病例報告 114_C074

腦部結核瘤模仿腦轉移瘤之案例報告與診斷挑戰探討

Intracranial Tuberculomas Mimicking Brain Metastases: A Case Report and Review of Diagnostic Challenge

Introduction

Tuberculosis, caused by *Mycobacterium tuberculosis*, can manifest as brain parenchymal tuberculoma, one of the most severe forms of extrapulmonary involvement. Tuberculomas may mimic various other conditions and present with a subacute to chronic course, ranging from asymptomatic cases to severe intracranial hypertension.

Case Report

We report the case of an 84-year-old man with a medical history of hypertension, cerebral palsy with right lower limb atrophy and hearing impairment, and benign prostatic hyperplasia.

At baseline, the patient was partially independent in activities of daily living (ADLs). He presented to the emergency department with progressive right-sided weakness over two days and a fall from bed while eating dinner, attributed to imbalance. On arrival at the emergency department, elevated blood pressure was noted during triage. Neurological examination showed bilateral upper limb weakness, more pronounced on the right side.

A brain CT scan revealed multiple ring-enhancing lesions with surrounding edema located in the left thalamus, left basal ganglia, bilateral temporal, frontal, parietal, and occipital lobes. He was admitted to the neurosurgery ward with a provisional impression of metastatic brain tumors. Dexamethasone and mannitol were administered for cerebral edema.

Brain MRI confirmed multiple ring-enhancing lesions with surrounding edema in the pons, right cerebellar hemisphere, left thalamus, left basal ganglia, and bilateral cerebral lobes. Differential diagnoses included multiple brain abscesses versus metastatic tumors.

Because of suspected brain abscess with ring-enhancing lesions, bacterial culture, TB culture, and Toxoplasma antibody testing were performed. Sputum acid-fast staining was negative; however, Mycobacterium tuberculosis was detected by quantitative PCR, and subsequent sputum culture confirmed the presence of M. tuberculosis. Additional evaluations for disseminated tuberculosis, including gastric aspirate, urine, and stool cultures, were negative. A diagnosis of pulmonary tuberculosis with possible CNS tuberculomas was established. Anti-tuberculosis therapy with Akurit-4 (isoniazid, rifampin, ethambutol, and pyrazinamide) along with dexamethasone was initiated.

Follow-up brain CT after one week of anti-TB therapy showed partial resolution of the ring-enhancing lesions and surrounding edema. The patient's level of consciousness gradually improved. Given the radiologic improvement and clinical response, CNS tuberculomas were favored over metastatic tumors. Brain tumor removal and biopsy was held by neurosurgeon as CNS tuberculoma was highly impressed and high risk of anesthesia.

Due to his inability to report visual symptoms, ethambutol was discontinued and replaced with levofloxacin; the regimen was adjusted to isoniazid, rifampin, pyrazinamide, and levofloxacin. He

was discharged from the isolation ward once pulmonary TB was controlled and serial sputum acid-fast stains were negative.

Post-discharge, anti-tuberculosis therapy was continued with rifampin, isoniazid, pyrazinamide, and levofloxacin for treatment of miliary TB. Serial brain CT scans demonstrated partial resolution of residual tuberculomas in the left frontal, parietal, and occipital lobes, and complete resolution of lesions in the pons, right cerebellar hemisphere, left thalamus, right temporal lobe, right frontal lobe, bilateral parietal lobes, and right occipital lobe after 16 months of treatment. Anti-tuberculosis therapy was discontinued after completing an 18-month course.

Conclusion

The diagnosis of tuberculomas is challenging and relies on clinical features, neuroimaging, and microbiological confirmation. Initial management typically involves a four-drug anti-tuberculosis regimen.

病例報告 114_C075

控制良好之 HIV 感染患者出現 Bartonella 腦膜炎合併顱內高壓

Bartonella Meningitis with Intracranial Hypertension in a Patient with Well-Controlled HIV: A Diagnostic Challenge

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Introduction

Bartonellosis is caused by Bartonella species and it can present with a wide variety of clinical symptoms. It can range from mild swollen lymphadenopathy to severe disseminated forms of disease like retinitis, endocarditis and CNS infection. Disseminated diseases occur most commonlyin immunocompromised patients such as persons with late stages of HIV or organ transplant recipients.

Case Report

A 49-year-old male with HIV under Biktarvy (recent viral load: undetectable) was admitted due to an hypercapnic respiratory failure due to acute asthma exacerbation triggered by influenza A infection. Initial management included sedation, bronchodilators, magnesium sulfate, and steroids. Hypercapnia gradually improved. Despite therapy, the patient's consciousness did not return to baseline status and showed agitation and dyspnea whenever tappering the sedation, prompted a central nervous system (CNS) evaluation.

Neurologic evaluation was then pursued due to fluctuating mental status. Lumbar puncture showed elevated opening pressure (40/27 cmH_2O) but negative cultures and PCR. Brain CT was unremarkable, while EEG demonstrated unstable cyclic state changes. Papular lesions along with lymphadenopathy on the right axilla raised suspicion for atypical infection. We reviewed the patient's social history which revealed his habit of raising stray cat, and we therefore sent CSF specimens for Bartonella PCR. Antibiotics were adjusted to Ciprofloxacin and Doxycycline to cover atypical infection and possible cat-scatch disease. Serial lumbar punctures showed gradual improvement in intracranial pressure (Feb 22: 30/13; Feb 25: 21/18 cmH_2O). Brain MRI and TEE were arranged to exclude intracranial lesions and endocarditis.

Finally, the patient was successfully extubated with clear consciousness, stable respiratory status, and normalization of EEG. Notably, cerebrospinal fluid PCR returned positive for Bartonella, supporting the diagnosis of Bartonella meningitis.

Discussion

Bartonella meningitis is rare, particularly among HIV-infected patients even when viral load is well controlled. Our patients still sufferered from disseminated form of bartonellosis. Diagnosis is challenging, as conventional methods—including blood cultures and serologic testing—often have limited sensitivity in immunocompromised hosts. In addition, cerebrospinal fluid (CSF) testing is frequently incomplete or not performed, which can obscure the recognition of meningitis caused by Bartonella even when clinical features are present. A thorogh history taking documenting cat or flea contact reasonably establish suspicion of Bartonella species.

Conclusion

The diagnostic process of CNS infection in this patient was particularly challenging. Despite persistent altered consciousness, neuroimaging was unremarkable, and both CSF culture and routine PCR failed to identify a pathogen. Without careful review of social history and clinical clues—such as papular lesions, lymphadenopathy, and cat exposure, the diagnosis of Bartonella could easily have been missed.

病例報告 114_C076

Sjögren 與類肉瘤病之罕見重疊：透過多種影像學工具鑑別出結節性肌病

From Sicca to Sarcoid: Multimodal Imaging Identifies Nodular Myopathy in Sjögren's Overlap

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Introduction

Although sarcoidosis is an exclusion in the 2002 and 2016 ACR/EULAR Sjögren's classification criteria, clinical overlap is increasingly recognized and diagnostically challenging. We report a rare case of nodular sarcoid myopathy with pulmonary sarcoidosis in long-standing Sjögren's syndrome, established non-invasively by multimodal imaging and controlled with a leflunomide-based, steroid-sparing regimen.

Case Report

A 59-year-old woman with a 10-year history of Sjögren's syndrome presented with multiple lumps in both calves over several months. The lumps were ~3 cm, located deep within the calf muscles, and mildly tender. Leg MRI demonstrated multiple ovoid/fusiform intramuscular lesions oriented along muscle fibers: on T1-weighted images, slight peripheral hyperintensity with central isointensity; on T2-weighted FSE (Fast Spin Echo), central low signal with hyperintense rims; and thick rim enhancement after gadolinium with mild perilesional edema and adjacent fascial fat stranding. DWI showed peripheral hyperintensity with reduced ADC, most pronounced in the left medial gastrocnemius. Sonography revealed hypoechoic linear-to-fusiform lesions within the medial gastrocnemius containing hyperechoic stripes and ill-defined ends; on the right, there was reduced fascicular caliber with increased power Doppler signal and patchy fatty atrophy. Chest CT demonstrated mediastinal and hilar lymphadenopathy. Gallium-67 SPECT/CT showed heterogeneous mediastinal uptake and abnormal uptake in the medial calves concordant with MRI, with additional activity in the lacrimal/sinonasal regions. Collectively, these multimodality findings supported nodular sarcoid myopathy. Serology showed positive anti-SSA/Ro60 antibodies and a positive antimitochondrial antibody (AMA > 1:320) despite normal γ-GT and alkaline phosphatase. Serum immunoglobulins were: IgG 1074 mg/dL, IgA 87 mg/dL, IgM 144 mg/dL, and IgG4 41 mg/dL. ANCA was negative. ESR, CRP, CK, and LDH were within normal ranges. The overall picture was highly suggestive of overlapping Sjögren's syndrome with nodular sarcoid myopathy and pulmonary sarcoidosis, despite the patient's refusal of biopsy. She achieved clinical remission of the nodules with hydroxychloroquine, leflunomide, and prednisolone, while Sjögren's features and pulmonary sarcoidosis remained stable.

Discussion

Sarcoidosis is listed as an exclusion criterion in Sjögren's classification due to clinical mimicry; however, recent reports challenge this paradigm and create diagnostic uncertainty. The chronic myopathic type is the most common form of sarcoid myopathy. To our knowledge, this is the first report of nodular sarcoid myopathy in a patient with Sjögren's syndrome. Evidence for managing sarcoid myopathy is limited, but leflunomide proved to be a well-tolerated, effective steroid-

sparing agent in this case. It induced clinical remission without hepatotoxicity—crucial given the positive AMA and concern for primary biliary cholangitis.

Conclusion

This case highlights the rare coexistence of nodular sarcoid myopathy, pulmonary sarcoidosis, and Sjögren's syndrome, challenging the notion of mutual exclusion in current classification criteria. Multimodal imaging enabled a non-invasive diagnosis, and combination therapy including leflunomide was a safe and effective steroid-sparing approach in this complex presentation.

病例報告 114_C077

使用 vedolizumab 治療罕見複雜重度潰瘍性結腸炎的病例報告與文獻回顧

R escue vedolizumab therapy in a rare case of complicated severe ulcerative colitis: A case report and literature review

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Introduction

Ulcerative colitis (UC) is a chronic inflammatory bowel disease sometimes associated with extraintestinal manifestations such as primary sclerosing cholangitis (PSC). Systemic lupus erythematosus (SLE) is a multisystem autoimmune disorder that rarely coexists with either UC or PSC. The simultaneous occurrence of UC, PSC, and SLE presents a unique diagnostic and therapeutic challenge. Vedolizumab, a gut-selective anti-α4β7 integrin antibody, has proven efficacy for UC, but its use in patients with overlapping autoimmune diseases remains largely unexplored.

Case Report

A 32-year-old man with a 10-year history of recurrent abdominal pain and fever presented with new-onset jaundice, diarrhea, hematochezia, and a chronic rash. Workup confirmed PSC, SLE, and severe UC. During hospitalization, he developed bacteremia, complicating management. Conventional UC treatments, including mesalazine and immunosuppressants (azathioprine followed by cyclosporine), offered limited improvement. Vedolizumab was subsequently introduced, leading to rapid clinical recovery and near-complete endoscopic remission of UC. Meanwhile, his PSC and SLE remained stable under existing therapies, although he continues to await liver transplantation for PSC.

Discussion

To date, no cases in Taiwan have documented vedolizumab use in patients simultaneously affected by UC, PSC, and SLE. This case demonstrates that vedolizumab can be effective and generally safe for UC, even in the setting of complex polyautoimmunity. However, close monitoring for infectious and autoimmune complications is essential, particularly when multiple immunosuppressive agents are used.

Conclusion

This report highlights vedolizumab as a promising option for UC in patients with concurrent PSC and SLE, emphasizing the importance of individualized treatment strategies and careful follow-up in rare, multifaceted autoimmune conditions.

病例報告 114_C078

莫見乎隱，莫顯乎微：急性鈎端螺旋體「肝腎風暴」下，低劑量類固醇是逆轉關鍵，還是巧合？

In an Acute Leptospiral "Hepatorenal Storm," Were Low-Dose Steroids the Decisive Factor—or a Coincidence?

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Introduction

Leptospirosis can present without fever yet progress rapidly to multiorgan dysfunction. We report a 59-year-old farmer with severe AKI and hepatic failure, initially confounded by acute hepatitis B. Empiric antimicrobials, dialysis, and later plasma exchange plus low-dose corticosteroids preceded biopsy-proven ATN and gradual recovery.

Case Report

This is a 59-year-old female farmer with a history of hypertension was admitted via the emergency department due to one week of progressive anorexia, nausea, generalized weakness, decreased urine output, and bilateral lower limb edema. She denied fever, upper respiratory symptoms, abdominal pain, diarrhea, rash, arthralgia, petechiae, or myalgia. Physical examination revealed jaundice, icterus, and bilateral pitting edema. Laboratory studies demonstrated acute hepatic dysfunction with hyperbilirubinemia, prolonged prothrombin time (INR 2.46), and markedly elevated aminotransferases, as well as acute kidney injury (serum creatinine 9.84 mg/dL, BUN 81 mg/dL) Abdominal computed tomography without contrast showed no intra-abdominal infection and kidneys of normal size. Initial management included hemodialysis and empiric antibiotics (ceftriaxone followed by levofloxacin and doxycycline, with consideration of leptospirosis and other atypical infection).

Serology revealed positive HBsAg and anti-HBc IgM with detectable viral load (1.03×10^3 IU/mL), acute hepatitis B infection was also suspected. Despite antiviral therapy (entecavir) and supportive measures, renal function continued to decline with anuria. Given the unexpected course, rapidly progressive glomerulonephritis (RPGN) and ATN (acute tubular necrosis) of leptospirosis were both suspected, prompting initiation of plasma exchange and corticosteroid therapy (prednisolone 10 mg BID) on hospital day 20. Renal biopsy revealed acute tubular cell injury without crescent formation, and the final serology showed positive result of leptospirosis. Following plasma exchange and corticosteroid therapy, the patient's clinical condition stabilized and renal function was also improving gradually. and she was discharged with outpatient follow-up. Finally, the patient's renal function returned to baseline in the OPD visits.

Discussion

Leptospirosis, a zoonotic bacterial infection, can cause severe multiorgan dysfunction, including liver failure, acute kidney injury (AKI), and ARDS. The disease's pathophysiology involves both direct effects of the bacteria and immune-mediated damage.[1] Corticosteroids have been suggested as adjunctive therapy in severe cases to mitigate harmful inflammation, particularly in refractory cases with immune-mediated kidney or liver failure.[2,3] However, clinical evidence

supporting the use of steroids is limited, with most studies showing low-certainty results. A recent meta-analysis of four randomized trials revealed no significant reduction in mortality or adverse events with corticosteroids. While corticosteroids improved renal function in the presented case, the optimal dosage and whether the recovery was due to steroids, antibiotics, or both remain unclear. Further, well-designed trials are needed to assess the true benefits and risks of corticosteroid use in leptospirosis.

Conclusion

In this case, clinical stabilization and renal recovery closely followed plasma exchange plus low-dose prednisolone, indicating corticosteroids likely played a pivotal adjunctive role in controlling immune-phase injury. Although high-quality evidence is limited, this case supports considering timely adjunct steroids in refractory hepatorenal leptospirosis unresponsive to standard therapy.

病例報告 114_C079

眾裡尋她千百度：瀰漫性非結核分枝桿菌揭露的免疫破口 - anti-IFN-γ autoantibody syndrome

A Thousand Searches, One Break in Immunity: Disseminated Nontuberculous Mycobacteriosis Unmasking the Anti–IFN-γ Autoantibody Syndrome

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Introduction

Anti-IFN-γ autoantibody syndrome is an immune-deficiency disorder associated with life-threatening infections, frequently leading to exhaustive yet unrevealing diagnostic workups. We will present a 36-year-old man presented with back and chest pain, marked inflammation, and osteolytic lesions mimicking hematologic malignancy. However, the final diagnosis defied everyone's expectations.

Case Report

This is a 36-year-old male with a 6-month history of back and chest pain, along with elevated inflammatory markers, was admitted to rule out aortic dissection and infection. Imaging revealed multiple osteolytic lesions, raising suspicion of multiple myeloma or lymphoma. His condition worsened with hypoxemia and bilateral lung infiltrates, requiring high-flow oxygen therapy. He was diagnosed with disseminated *Mycobacterium kansasii* infection, treated with meropenem, minocycline, moxifloxacin, clarithromycin, ethambutol, and rifabutin. Later, anti-IFN-γ autoantibody syndrome was identified, leading to the addition of prednisolone and hydroxychloroquine. The patient stabilized and was discharged with outpatient follow-up and delayed spinal surgery.

Discussion

Adult-onset immunodeficiency caused by neutralizing anti-IFN-γ autoantibodies (AIGA) disables the IL-12/IFN-γ axis, making immunocompetent adults, particularly in East Asia, vulnerable to disseminated infections like nontuberculous mycobacteria (NTM), such as *Mycobacterium kansasii*.[1] This patient's culture-proven infection, involving lung, bone, and marrow, initially mimicked hematologic malignancy, a diagnostic pitfall. AIGA testing is crucial in HIV-negative patients with disseminated or refractory M. kansasii infections.[2] Key points to consider: elevated D-dimer, leukocytosis, and CRP often indicate uncontrolled intracellular infection rather than malignancy, and cancer-focused work-ups delay microbiology and immune testing.[3] Antimicrobial therapy alone is often insufficient, requiring immunomodulation to reduce pathogenic antibody levels. Diagnosis of AIGA involves detecting anti-IFN-γ antibodies through ELISA and functional assays.[4] Treatment focuses on lowering antibody titers using B-cell-directed therapies like rituximab or cyclophosphamide, which restore IFN-γ signaling. Interferon-γ supplementation or IVIG alone is ineffective.[5,6] Genetic risk factors like HLA haplotypes strengthen pretest probability but do not alter management.[7]

Conclusion

Anti–IFN-γ autoantibody syndrome is a treatable, acquired immunodeficiency that masquerades as malignancy and severe community-acquired infection. This case gives us an enlightenment that clinicians should add AIGA to the differential for HIV-negative adults with disseminated NTM, necrotizing pneumonia with bone lesions, or paradoxical worsening on standard therapy.

病例報告 114_C080

高齡患者疑似中樞感染併發真菌血症致死病例

Fatal Fungemia Following Suspected CNS Infection in an Elderly Patient

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Introduction

Respiratory viral infections can weaken the body's defenses against fungi, making it easier for fungal infections to occur. Fungemia symptoms can be different for each person and may include confusion. If not diagnosed and treated early, it can lead to poor outcomes.

Case Report

An 89-year-old male with a history of skin tumor, sick sinus syndrome (post-pacemaker), and parkinsonism presented to the emergency department (ED) with flu-like symptoms and progressive deterioration in consciousness. He had received outpatient treatment for over three days without improvement. Despite negative influenza and SARS-CoV-2 screens, empirical antiviral therapy was initiated.

The patient was given empirical antibiotics, including meropenem and ampicillin, for suspected central nervous system infection. During the initial examination, a black necrotic lesion was noted on the hard palate following removal of a dental prosthesis. However, biopsy and otolaryngology consultation were deferred due to concerns over bleeding risk, the patient's fragile condition, and family preference.

After stabilization, vasopressor support was discontinued. The patient subsequently developed gastrointestinal bleeding, evidenced by positive fecal occult blood tests. He was treated with intravenous proton pump inhibitors and received peripheral parenteral nutrition.

On the following day, gingival bleeding was observed, accompanied by a high fever reaching 40°C despite antipyretic therapy. Later, the patient experienced recurrent hypotension and hypoxemia. Laboratory tests revealed rhabdomyolysis, acute renal failure, and myocardial ischemia. Supportive care, including oxygen therapy, was administered. Aggressive temperature control measures were taken due to suspected central nervous system–related malignant hyperthermia. Renal replacement therapy and advanced coronary arterial disease workup were withheld in consideration of bleeding risks and the patient's do-not-resuscitate status. With progressive hypoxemia and cyanosis of the limbs, the family elected for critical discharge.

A blood culture obtained during the second shock episode ultimately grew Yeast-like after 86 hours of incubation, while earlier blood cultures remained negative.

Discussion

Fungal bloodstream infections are challenging to detect early due to their slow growth and low initial burden. Repeating cultures often yield limited additional diagnostic value. In this case, early clues—such as the necrotic oral lesion—were present but not aggressively pursued, partly due to bleeding risk and partly in deference to the patient's frailty and previously expressed care preferences. Despite a low Candida score and lack of typical signs, fungemia developed and led to

rapid multi-organ failure. Even with early antifungal therapy, outcomes are often poor in critically ill elderly patients.

Conclusion

This case emphasizes the need for early clinical suspicion of invasive fungal infections in high-risk patients, even with negative initial tests. It also underscores the limitations of protocol-driven care in medically complex, end-of-life situations. While diagnostic certainty and aggressive treatment are often prioritized in acute medicine, there are situations—such as this one—where human-centered, comfort-focused care must take precedence.

病例報告 114_C081

特發性多發性卡斯爾曼氏病合併 TAFRO 症候群：病例報告與臨床治療回顧

Multicentric Castleman Disease with TAFRO Syndrome: A Case Report and Review of Clinical Management

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Introduction

Castleman disease (CD) is an uncommon lymphoproliferative disorder that often presents with non-specific complaints. Adult patients may initially seek medical attention for general symptoms, and a definite diagnosis usually requires a combination of clinical, laboratory, and histopathological evaluation. We describe a patient in whom multicentric Castleman disease (MCD) with TAFRO syndrome was identified during hospitalization.

Case Report

A 59-year-old man with a history of hypertension was admitted for progressive abdominal fullness, shortness of breath, and worsening bilateral leg swelling over two months. He also reported reduced appetite and decreased urine output. Physical examination revealed palpable cervical lymph nodes, ascites, and lower limb edema. Laboratory studies showed anemia, thrombocytopenia, elevated CRP, impaired renal function with proteinuria, and hypoalbuminemia. CT demonstrated bilateral pleural and pericardial effusions, marked ascites, splenomegaly, and lymphadenopathy involving cervical, axillary, mediastinal, and pericardial regions. PET scan confirmed FDG-avid mediastinal and hilar lymph nodes. Bone marrow biopsy showed a mildly hypercellular marrow without abnormal blasts or cytogenetic abnormalities. Excisional biopsy of a cervical lymph node revealed features consistent with multicentric Castleman disease, and together with clinical and pathological findings, the diagnosis of TAFRO syndrome was established.

Discussion

Castleman disease (CD) is a rare group of lymphoproliferative disorders, clinically classified into Unicentric CD (UCD) and Multicentric CD (MCD). MCD can be further subdivided according to HHV-8 infection status into HHV-8-associated MCD and HHV-8-negative MCD (iMCD). TAFRO syndrome represents an aggressive subtype of idiopathic MCD, characterized by thrombocytopenia, anasarca, renal impairment, bone marrow fibrosis, and organomegaly. MCD can occur at any age, and many symptoms are related to excess secretion of interleukin-6. It may present with peripheral lymphadenopathy, fever, night sweats, weight loss, and fatigue. The diagnosis of MCD requires: characteristic lymph node histopathology, involvement of multiple lymph node regions, associated clinical and laboratory abnormalities, and exclusion of other infections, malignancies, or autoimmune diseases. Management is guided by severity. Patients with mild disease can be monitored, whereas those with significant or rapidly progressive symptoms, should receive systemic corticosteroids combined with anti-IL-6 therapy. When anti-IL-6 agents are not available or ineffective, rituximab with chemotherapy regimens such as R-CHOP or R-CVP may be used. Once clinical stability is achieved, siltuximab is a reasonable maintenance option. Long-term treatment

may also involve siltuximab with short steroid courses or rituximab combined with thalidomide, particularly when immune-mediated cytopenia is present.

Conclusion

This case illustrates multicentric Castleman disease with TAFRO syndrome, presenting with anasarca, cytopenias, renal dysfunction, and splenomegaly. Prompt recognition and initiation of appropriate immunomodulatory therapy are critical, and individualized treatment can improve both outcomes and quality of life.

病例報告 114_C082

惡性瘧原蟲引起之重症瘧疾

Severe malaria caused by *Plasmodium falciparum*: a case report

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Introduction

Taiwan was officially issued for malaria elimination in 1965, with 10 to 30 cases imported annually from Southeast Asia, Africa and Oceania. Here, we presented a case of complicated *P. falciparum* malaria treated with standard Artemisinin-based therapy and adjunctive plasma exchange.

Case Report

A 66-year-old man presented to the emergency department with anorexia and dark urine for five days, accompanied with yellowing of skin color, and decreased urine output. He had returned from a 1-month trip to Africa, without antimalarial prophylaxis.

After admission, respiratory distress developed with hypoxia and metabolic acidosis, which resulted in intubation and machine ventilation. Vasopressor and continuous veno-venous hemofiltration were given for hypotension and oliguria. The blood films for malaria reported *Plasmodium falciparum*, with heavy parasitemia (20 Infected RBCs/100 RBCs). Antimalarial treatment with intravenous artesunate was initiated with a total of four doses and then shifted to oral artesunate-based combination therapy with artemether-lumefantrine after parasite density less than 1 %.

However, thrombocytopenia, anemia, and hyperbilirubinemia progressed even if the parasitemia was improved. Haptoglobin was less than 10 mg/dL and LDH was 599 U/L. Peripheral blood smear found schistocytes. The laboratory data suggested microangiopathic hemolytic anemia, and thrombotic microangiopathy-mimic condition related to malaria was suspected. Plasma exchange was therefore performed. His consciousness recovered dramatically on the next day of the first session of the plasma exchange, along with the resolution of hyperbilirubinemia and thrombocytopenia.

The multiple organ damage recovered gradually. However, the recurrence of malaria with parasite density 4.8% was confirmed eighteen days after the resolution of parasitemia. Another course of intravenous artesunate followed with oral artemether-lumefantrine was administered, with clindamycin and doxycycline completed for three weeks. He was discharged after 55 hospital days.

Discussion

Most cases of severe malaria were caused by *P. falciparum*, with high parasitemia and microvascular sequestration, which developed into multiorgan failure. Complications developed even after the initiation of prompt antimalarial therapy.

The adherence of the infected red blood cells to the endothelium of blood vessels, caused not only the sequestration of parasites, but also obstruction of tissue perfusion. With the expression of the parasite protein on the surface of the erythrocytes, *P. falciparum* erythrocyte membrane protein 1

(PfEMP1), the infected cells adhere to the endothelium of small blood vessels. The adherence to the microvasculature avoids the destruction of the infected erythrocyte being destroyed by spleen and even contributes to the recurrence of malaria after the appropriate anti-malarial treatment. Hemolytic anemia in severe malaria has a complex mechanism. In addition to the erythrocytes rupture while the release of merozoite, microangiopathic hemolytic anemia is also observed. Thrombotic thrombocytopenic purpura mimic presentation after the infection of *P. falciparum* had been reported. The similar case was noted in Taiwan reported in 2017, with artesunate and therapeutic plasma exchange used for the complicated *P. falciparum* with TMA-like syndrome.

Conclusion

Although the direct correlation between the plasma exchange and the therapeutic outcome was difficult to establish due to the complexity of clinical practice, plasma exchange may still contribute to the management of severe malaria complicated with microangiographic hemolytic anemia.

病例報告 114_C083

一個病例報告：以右側肋膜積液反覆發作表現之類澱粉蛋白質輕鏈型瀰漫性間質性類澱粉沉積。

Amyloid light-chain (AL) type diffuse parenchymal amyloidosis presenting as recurrent right side pleural effusion: a case report.

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Introduction

Amyloidosis is a pathological phenomenon referring to the extracellular tissue deposition of highly ordered fibrils composed of low molecular weight subunits of a variety of proteins, which are mostly normal constituents of plasma in their native form. Amyloid deposits may result in various clinical manifestations depending upon their type, location, and amount.

Case Report

A 73-year-old male retired officer, ex-smoker (1 pack per day for 40 years, and quitted for 20 years) with history of coronary artery disease, hypertension and type 2 diabetes was referred to chest-medicine (CM) out-patient department (OPD) on July, 2024 because of right pleural effusion found during pre-operation survey for retrograde intrarenal surgery (RIRS).

At CM OPD, mild dyspnea was mentioned, and spirometry favored COPD. Thoracic echo and pleural fluid analysis revealed simple right pleural effusion, clear yellow fluid, exudate (LDL: 164 U/L), lymphocyte predominant (76%), ADA<10 U/L, and no presentation of cytological malignancy. Without certain diagnosis according to obtained report, the patient underwent computer tomography scan (CT scan), which showed pericardial effusion, enlarged lymph nodes (LNs) in subaortic and right-side pericardial region, bilateral lung small solid nodules (~5mm, oval), and centrilobular micronodules in right upper lobe and lower lobe. To rule out malignancy and pericardium diseases, PET and cardiac echo were arranged, but there was no positive finding. However, the patient's dyspnea progressively worsened that he must rest after climbing few stairs. Follow-up chest X-ray (CXR) revealed progressive right pleural effusion compared to pre-operation CXR.

Thus, the patient was admitted for thoracoscopic exploration on August, 2024. During admission, lab data revealed ANA 1:80 with AC-10 pattern, and IgM 1149.00 mg/dL. Further survey showed less likelihood of rheumatic diseases. Thoracoscopy only noted some erythematous lesions on chest wall and the surface of lung, and pleura biopsy was done. Pathological report shows mixed lymphocytic infiltrates in the fibroadipose tissue, including CD20-positive B-lymphocytes and CD3-positive T lymphocytes, as well as nests of reactive mesothelial cells. The patient received pigtail insertion for drainage, and was discharged with the tentative diagnosis of mesothelioma, unspecific lymphoma and amyloidosis.

However, the disease showed recurrence on September, 2024, so the patient was referred to chest-surgery (CS) OPD for biopsy and pleurodesis. During the video-assisted thoracoscopic surgery (VATS), a vegetation on pleura near heart and a hard wedge on the oblique fissure were noted, and biopsy of these locations and pleurodesis of right pleural space were done. Pathological report

revealed amyloid deposits in the vascular walls, perivascular spaces with adjacent lung interstitial and pleural involvement, and immune stain showed polyclonal Kappa/lambda light chains for plasma cells. With the impression of diffuse parenchymal amyloidosis, suspect systemic amyloid light-chain type, the patient was referred to hematology & oncology specialist.

Conclusion

This is a rare case of recurrent pleural effusion, and was later diagnosed with AL type amyloidosis. We'll keep the patient in OPD follow-up until further evidence of multiple myeloma or leukemia is observed.

病例報告 114_C084

甲狀腺原發性淋巴瘤以黏液性水腫昏迷為初始表現之病例報告

Myxedema Coma as the Initial Presentation of Primary Thyroid Lymphoma

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Introduction

Myxedema coma represents the most severe, life-threatening state of hypothyroidism, which reported mortality remains between 20% and 60%[1]. It result from loss of the adaptive mechanism to maintain homeostasis[1], and be precipitated by acute stressors such as infection, myocardial infarction, cold exposure, and surgery in patients with poorly controlled hypothyroidism[2]. It is characterized by altered mental status, hypoventilation, hypothermia, and metabolic derangements. Primary thyroid lymphoma (PTL) is a rare thyroid malignancy, accounting for less than 5% of thyroid cancers and often arising in the setting of Hashimoto's thyroiditis[3]. Primary thyroid lymphoma initially impacts the thyroid gland, with subsequent spread to the lymph nodes and other organs occurring later[3]. The coexistence of myxedema coma and PTL is extremely unusual. We present a case in which coma was the first clinical manifestation of an underlying thyroid lymphoma.

Case Report

A 70-year-old woman with poorly controlled Hashimoto's thyroiditis was found obtunded at home after two weeks of progressive lethargy, cold intolerance, and a rapidly enlarging anterior neck mass. On admission, she presented hypothermia(31.5°C), bradycardia (38 bpm), and hypotensive (80/45 mmHg). Physical examination revealed periorbital puffiness, dry skin, delayed reflex relaxation, and a firm, immobile neck mass. Laboratory examination showed profound hypothyroidism (TSH >100 μIU/mL, FT4 <0.4 ng/dL), severe hyponatremia (115 mmol/L), and respiratory acidosis (pH 7.21, pCO_2 58 mmHg). CT demonstrated a bulky heterogeneous thyroid mass with tracheal compression. Core biopsy confirmed diffuse large B-cell lymphoma.

She was admitted to the intensive care unit and received intravenous levothyroxine, hydrocortisone, ventilatory support, passive warming, and cautious fluid management. Once stabilized, she was treated with R-CHOP chemotherapy. Her consciousness and hemodynamics improved gradually, and follow-up PET-CT revealed complete remission.

Discussion

Recent data reaffirm that myxedema coma remains uncommon but deadly; Contemporary data indicate mortality of approximately 25-50%[4]. On the oncologic side, multiple recent PTL series/case syntheses emphasize the strong association with autoimmune thyroiditis and the predominance of DLBCL histology, guiding clinicians to suspect PTL when a rapidly enlarging goiter occurs in Hashimoto's patients. These reports stress tissue diagnosis and favor chemo-immunotherapy (e.g. R-CHOP) as first-line, reserving surgery for select indications[5].

Conclusion

In patients with Hashimoto's thyroiditis and rapidly enlarging thyroid, PTL should be high in the differential. When altered mental status, hypothermia, and hypoventilation , myxedema coma need to be differential.

病例報告 114_C085

從脂肪炎到淋巴瘤：超音波跨科診斷的臨床啟示

From Panniculitis to Lymphoma: Cross-disciplinary Lessons with Ultrasonography

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Introduction

Panniculitis is an inflammation of subcutaneous fat that presents as painful erythematous nodules or plaques. It is often caused by infection, trauma, or drug reactions, usually responding to NSAIDs or corticosteroids. However, panniculitis may also indicate systemic diseases such as pancreatic disorders, lymphoma, autoimmune diseases, or vasculitis, requiring thorough investigation. We report a case of persistent panniculitis-like lesions ultimately diagnosed as lymphoma, highlighting the role of ultrasonography, cross-disciplinary collaboration, and comprehensive PET imaging that guided the final diagnosis.

Case Report

A 65-year-old man with no significant medical history presented with multiple painful nodules over the right calf and thighs, accompanied by ankle swelling for four weeks. Ultrasonography showed mixed lobular and septal panniculitis with increased Doppler flow and an enlarged right inguinal lymph node. Laboratory tests, including ANA, ANCA, ENA, and CRP were negative. CT revealed bilateral soft-tissue swelling with subcutaneous edema.

The patient initially received antibiotics but declined biopsy. Over the next three months, the lesions worsened with necrosis in the right leg and new nodules over the upper limbs and eyelid. PET revealed abnormal uptake in multiple subcutaneous sites. A forearm biopsy confirmed the diagnosis of extranodal NK/T-cell lymphoma. He was referred to hematology/oncology and subsequently improved after treatment with GELOX combined with nivolumab.

Discussion

Panniculitis can be classified as septal or lobular, each with characteristic ultrasonographic features. Septal panniculitis shows hypoechoic, thickened septa, hyperechoic lobules, non-compressible tissue, jigsaw-like appearance, and increased Doppler signal. Differential diagnoses include erythema nodosum, sarcoidosis, scleroderma, and cutaneous polyarteritis nodosa. Lobular panniculitis is characterized by diffuse hyperechogenicity, fat necrosis, minimal Doppler signal, blurred margins, and absence of septal thickening. Differential diagnoses include lupus panniculitis, pancreatic panniculitis, α1-antitrypsin deficiency panniculitis, and less commonly, gout panniculitis or post-steroid panniculitis.

Subcutaneous panniculitis-like T-cell lymphoma (SPTCL) is a lobular subtype, often presenting with fat thickening, homogeneous hyperechogenicity, poorly defined margins, linear hypoechoic areas, vascular signals, and infiltrative patterns. In this case, early sonographic findings and lymphadenopathy indicated a non-infectious etiology. With disease progression and the atypical features, biopsy became indispensable for establishing the diagnosis.

Conclusion

Histopathology remains the gold standard for diagnosing panniculitis, but ultrasonography provides a valuable, non-invasive tool to narrow the differential diagnosis and guide clinical management. Cross-disciplinary collaboration is essential when panniculitis mimics rare but serious conditions such as lymphoma.

病例報告 114_C086

免疫健全年輕女性水痘感染後併發 A 群鏈球菌臉部蜂窩組織炎與深頸部感染

Group A Streptococcal Facial Cellulitis and Deep Neck Infection Following Varicella Infection in an Immunocompetent Young Woman

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Introduction

Varicella, caused by varicella-zoster virus, is usually self-limiting in children, but may cause severe complications in adult. Secondary bacterial infection is common, and although rare, deep soft tissue infections from Streptococcus pyogenes can be life-threatening. We report a rare case of a young woman who developed deep neck infection and facial cellulitis following varicella.

Case Report

A 25-year-old previously healthy female teacher presented with a 5-day history of vesicular rash initially on the face, later spreading to trunk and extremities, with fever up to 40°C for 3 days. Examination showed skin lesions in different stages—macules, vesicles, pustules, and crusts—with pruritus and pain. Laboratory data revealed normal leukocyte count (6000 /uL) with neutrophil predominance (92.3%), elevated hsCRP (9.07 mg/dL), procalcitonin (0.52 ng/mL), and mild hypokalemia. Both VZV IgM and IgG were negative.

On admission, right facial swelling with submandibular lymphadenopathy was noted. On hospital day 2, she developed trismus and limited neck rotation. Contrast-enhanced CT showed facial cellulitis and deep neck infection without drainable abscess. Viral PCRs for VZV, EBV, EV, HSV, and CMV were negative. The diagnosis of varicella infection was made based on the clinical history and presentation. Empirical antibiotics (teicoplanin, clindamycin, amoxicillin-clavulanate) and acyclovir were given. Deep pus culture yielded Streptococcus pyogenes. No surgical drainage was performed due to airway stability and lack of abscess. With antibiotics, facial swelling and trismus improved, and no new varicella lesions appeared.

This case illustrates varicella-like illness complicated by group A streptococcal cellulitis and deep neck infection in an immunocompetent adult despite prior vaccination.

Discussion

Varicella complicated by invasive group A streptococcal disease in adults is rare but associated with high morbidity. The virus disrupts mucocutaneous barriers and induces transient immune suppression, allowing GAS colonization and spread into deep cervical spaces. Adults with vaccination but without natural infection may remain vulnerable. In this case, diagnosis was confounded by negative VZV serology and PCR, showing early tests may be unreliable and clinical recognition of rash is essential. Facial swelling, cervical edema, and trismus were warning signs of deep infection requiring urgent intervention.

Conclusion

This case highlights that even in healthy young adults, varicella can predispose to severe bacterial

complications. It underscores the invasive and potentially fatal nature of Streptococcus pyogenes infection following disruption of the skin barrier.

病例報告 114_C087

脫離葉克膜過程中因胸骨骨折引起延遲性心包填塞：罕見併發症病例報告

Delayed Cardiac Tamponade Caused by Sternal Fracture During ECMO Weaning: A Rare Complication and Case Report

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Introduction

Sternal fractures usually result from blunt chest trauma or chest compressions during cardiopulmonary resuscitation (CPR). Although rare, delayed cardiac tamponade secondary to sternal fracture can be a fatal complication. We report a patient with out-of-hospital cardiac arrest treated with extracorporeal cardiopulmonary resuscitation (ECPR) and percutaneous coronary intervention, who subsequently developed delayed cardiac tamponade on the fifth hospital day requiring urgent intervention during ECMO weaning process.

Case Report

A 59-year-old man presented with acute chest pain and subsequently developed out-of-hospital cardiac arrest (OHCA). Bystander CPR was performed, and he achieved return of spontaneous circulation (ROSC) after extracorporeal cardiopulmonary resuscitation (ECPR), followed by veno-arterial ECMO support. Coronary angiography revealed thrombotic occlusion of the left main coronary artery, left anterior descending artery (LAD), left circumflex artery (LCx). Primary PCI with balloon angioplasty to the left main and bare-metal stents to the LAD and LCx was performed, and he was admitted to the ICU with a diagnosis of STEMI complicated by cardiogenic shock and ventricular arrhythmia. After initial stabilization, ECMO weaning began on hospital day 3.

On hospital day 5, the patient developed hypotension and low ECMO flow. Echocardiography showed new pericardial effusion, and emergent pericardiocentesis followed by surgical repair revealed right ventricular injury caused by sternal fracture. Postoperatively, he required intra-aortic balloon pump reinsertion for pulmonary edema and persistent myocardial stunning. Despite these efforts, he developed progressive heart failure, new septic shock, and ultimately passed away on hospital day 15.

Discussion

Sternal fractures are typically caused by blunt chest trauma or chest compressions during CPR. Delayed cardiac tamponade after sternal fracture is rare but potentially fatal, usually resulting from bony fragment irritation or direct myocardial injury. Prolonged CPR increased the risk of sternal fracture. In this case, the patient stabilized after ECPR and revascularization, and ECMO weaning began on hospital day 3. During weaning, reduced venous drainage may have led to right atrial and right ventricular dilation and increased myocardial contractility, which could have enhanced mechanical contact between the fractured sternum and the underlying heart. This dynamic change may have contributed to delayed right ventricular laceration and tamponade.

Conclusion

Delayed cardiac tamponade due to sternal fracture is an uncommon but life-threatening complication, especially in patients undergoing prolonged CPR and ECPR. During ECMO weaning, reduced venous drainage and right heart expansion may increase vulnerability to sternal fragment injury. Vigilant hemodynamic monitoring, serial echocardiography, and prompt surgical intervention are essential when unexpected instability arises in these patients.

病例報告 114_C088

接受腹膜透析病患之米麴菌（*Aspergillus oryzae*）腹膜炎

Aspergillus oryzae Peritoneal Dialysis Peritonitis: A Case Report and Literature Review

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Introduction

Fungal peritonitis (FP) is a rare but serious complication of peritoneal dialysis (PD), accounting for 3–5% of cases and associated with high morbidity and mortality. *Candida* species are the most common pathogens, while *Aspergillus* peritonitis is less frequent but often fatal. Given the diagnostic and therapeutic challenges associated with *Aspergillus* peritonitis, we report a case of *Aspergillus oryzae* peritonitis in a patient undergoing continuous ambulatory peritoneal dialysis (CAPD).

Case Report

A 54-year-old man receiving CAPD for diabetic end-stage renal disease presented with turbid peritoneal dialysate and fibrinous debris. He had a history of peritonitis episodes caused by *Enterobacter cloacae*. On current admission, laboratory findings revealed elevated C-reactive protein (CRP) and an increased peritoneal white blood cell count. Despite empirical treatment with intraperitoneal ceftazidime and systemic fluconazole, his symptoms persisted. A positive serum galactomannan antigen test led to the initiation of voriconazole. Hemodialysis was resumed, and the Tenckhoff catheter was removed. Computed tomography showed no evidence of peritoneal adhesion. Fungal cultures later grew *Aspergillus* species, confirmed as *Aspergillus oryzae* by polymerase chain reaction. The patient responded well to oral voriconazole and was discharged in stable condition.

Discussion

To our knowledge, this is the second reported case of CAPD-related peritonitis caused by *A. oryzae*, with the first described by Schwetz et al., involving intravenous amphotericin B and caspofungin treatment, catheter removal, and transition to hemodialysis. Both *A. flavus* and *A. oryzae* belong to the *Aspergillus* section *Flavi*, which comprises eight clades and 33 recognized species. However, differentiating between these two species remains challenging due to their close genetic and phenotypic similarities; traditional methods such as MALDI-TOF MS and sequencing of Internal Transcribed Spacer (ITS), β-tubulin (benA), or calmodulin (CaM) genes are insufficient for distinction. Nargesi et al. demonstrated that *cyp51A* gene sequencing may aid in differentiation. We conducted a literature review of CAPD-associated peritonitis caused by species within *Aspergillus* section *Flavi*, identifying prior bacterial peritonitis treated with antibiotics and diabetes mellitus as common risk factors.

Diagnosis of *Aspergillus* peritonitis is often delayed due to the slow growth on Sabouraud dextrose agar, but non-culture-based biomarkers such as galactomannan (GM) and (1→3)-β-D-glucan (BDG) offer valuable diagnostic alternatives. GM and BDG, fungal cell wall components detectable in

serum or dialysate, have shown utility in cases of *A. fumigatus* and *A. niger* peritonitis. A cross-sectional study suggested diagnostic cutoffs for GM ≧0.5 and BDG ≧240 pg/mL in PD effluent, while another identified serum GM index ≧0.56 with 83% specificity. Per 2022 International Society for Peritoneal Dialysis (ISPD) guidelines, immediate catheter removal is recommended upon fungal identification, with antifungal therapy continued for at least two weeks thereafter. Treatment typically involves intravenous amphotericin B or azoles like voriconazole, with systematic review data indicating superior outcomes with voriconazole. Catheter removal occurred in 85.5% of cases and remains a cornerstone of effective management; nonetheless, overall mortality remains high at 38.2%, often due to delayed diagnosis, postponed catheter removal, or extensive peritoneal adhesions.

Conclusion

This case represents the second reported instance of *Aspergillus oryzae* peritonitis in a patient on peritoneal dialysis. It highlights the clinical features and management strategies for CAPD-related *Aspergillus* infections and underscores the potential role of non-culture-based biomarkers, such as galactomannan and β-D-glucan, in facilitating early diagnosis and improving patient outcomes.

病例報告 114_C089

原發性肝臟黏膜相關淋巴組織淋巴瘤：案例系列研究

Primary hepatic mucosa-associated lymphoid tissue lymphoma: A case series in a tertiary hospital in Taiwan

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Introduction

Primary hepatic lymphoma (PHL) is a rare extranodal lymphoma confined to the liver without extrahepatic involvement at diagnosis, accounting for 0.1% of liver malignancies and 0.4% of extranodal non-Hodgkin lymphomas (NHLs). Diffuse large B-cell lymphoma (DLBCL) is the most common subtype, whereas marginal zone B-cell lymphoma of mucosa-associated lymphoid tissue (MALT) represents a less frequent, indolent variant. Pathogenesis is linked to chronic infection and autoimmune conditions that promote lymphoid tissue formation and malignant transformation. Management strategies include surgery, chemotherapy, and immunotherapy. This study reports a case series of primary hepatic MALT lymphoma, focusing on clinical presentation, etiology, imaging, pathology, treatment, and prognosis.

Case Report

From 2015 to 2024, 70 patients at National Taiwan University Hospital were diagnosed with hepatic lymphoma by image-guided liver tumor biopsy. DLBCL accounted for 45 cases (64%), while 9 patients (12.8%) had low-grade B-cell lymphoma, including 4 cases (5.7%) of marginal zone lymphoma. Other subtypes included follicular, small lymphocytic, and hairy cell lymphoma. All four patients with hepatic MALT lymphoma were male, over 60 years of age (mean 63 years), and largely asymptomatic; only one reported epigastric discomfort. All tumors measured less than 3 cm. Two patients were hepatitis B carriers with HBV flare-ups treated with entecavir, one had resolved HBV with concurrent chronic HCV infection treated with ledipasvir–sofosbuvir, and one was negative for both HBV and HCV. All had metabolic disease, and three were active smokers. Tumor markers (AFP, CEA, CA19-9) were within normal ranges.

Imaging demonstrated nonspecific features: ultrasound showed ill-defined isoechoic lesions, while contrast-enhanced ultrasound revealed rapid arterial enhancement, portal-phase homogeneity, and late-phase washout. PET-CT detected hypodense lesions with increased glucose metabolism. MRI findings included T1 hypointensity, mild T2 hyperintensity, and restricted diffusion. Histopathology revealed CD20, CD43, and BCL-2 positivity, with negative CD3, CD5, CD10, and cyclin D1. No bone marrow involvement was noted. Treatments included two hepatic lobectomies, one radiofrequency ablation, and one active surveillance. Two patients received cyclophosphamide with corticosteroids, one received rituximab, and one had no adjuvant therapy but developed recurrence after 1.3 years. At final follow-up, all patients were alive, with a mean follow-up of 6.7 years.

Discussion

MALT lymphoma most commonly affects the stomach (around 50%) but rarely arises in the liver.

Literature reviews summarize 123 reported cases with median age 62 years and no sex predominance. One-third were associated with hepatitis B, 10% with hepatitis C, 10% with Helicobacter pylori, and 7% with autoimmune disease. Chronic infection may promote autoreactive B cells, lymphoid proliferation, and eventual malignant transformation. Regression following antimicrobial therapy has been documented. Genetic and immunologic dysregulation, including NF-κB activation and TNF-α–mediated inflammation, may further contribute.
No standardized treatment exists. Most patients undergo surgery and/or chemotherapy; some are managed with observation or radiotherapy. Rituximab-based regimens, particularly R-CHOP, remain common. Reported median follow-up is 23 months, with 80 out of 96 patients achieving remission, 10 relapsing, and 6 deaths. In our series, the median follow-up was 92 months, and all four patients survived, suggesting favorable long-term outcomes compared with prior reports.

Conclusion

Primary hepatic MALT lymphoma is rare, with nonspecific clinical and imaging features. Our series highlights a favorable prognosis with appropriate management. Prospective multicenter studies are needed to establish standardized treatment guidelines.

病例報告 114_C090

以不明原因腹痛為表現的陣發性夜間血紅素尿症：病例報告

Unexplained Abdominal Pain Leading to the Diagnosis of Paroxysmal Nocturnal Hemoglobinuria

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Introduction

Paroxysmal nocturnal hemoglobinuria (PNH) is a rare, acquired clonal hematopoietic stem cell disorder caused by somatic PIGA mutations, resulting in deficiency of GPI-anchored complement regulators (CD55, CD59). The incidence is 1–1.5 cases per million annually, with frequent diagnostic delays due to heterogeneous, nonspecific manifestations. This leads to complement-mediated intravascular hemolysis, thrombosis, and marrow failure. Clinical features include hemolytic anemia, dark urine, and elevated thrombotic risk. Eculizumab and ravulizumab (C5 inhibitors) remain first-line therapies, with proximal complement inhibitors under development.

Case Report

A 41-year-old man presented with intermittent epigastric pain for three weeks, occasionally localized to the left lower quadrant, cramping and sharp, 6–7/10. Pain occurred every 3–4 days, meal-aggravated, without nausea, vomiting, bowel changes, bleeding, weight loss, or urinary symptoms. He had no significant past or family history, and took no regular medications. Labs showed normocytic anemia, hyperbilirubinemia, and elevated CRP (16.27 mg/dL). Abdominal CT excluded lesions/thrombosis but showed jejunal wall thickening, suggesting segmental jejunitis. He received empirical antibiotics, though symptoms persisted.

Enteroscopy showed mucosal swelling and ulcer scar in the jejunum, suggesting Crohn's disease; prednisolone and mesalazine were started. However, biopsy was negative. Over five months, intermittent abdominal pain and progressive anemia persisted, with leukopenia. Iron deficiency was confirmed, though fecal occult blood tests were negative.

Further evaluation showed normocytic anemia (reticulocyte index 0.99), leukopenia, indirect hyperbilirubinemia, and elevated AST/LDH. Urinalysis revealed microscopic hematuria with positive occult blood. Hemolysis work-up showed normal folate, vitamin B12, and negative Coombs' tests, suggesting non-immune hemolysis. No thrombocytopenia, renal dysfunction, neurological deficits, or coagulopathy, making thrombotic microangiopathy unlikely.

Flow cytometry confirmed paroxysmal nocturnal hemoglobinuria (PNH) with large clone sizes (granulocytes 81.7%, monocytes 82.7%). He is under evaluation for eculizumab therapy.

Discussion

Paroxysmal nocturnal hemoglobinuria (PNH) is an acquired clonal hematopoietic stem cell disorder caused by PIGA mutation, resulting in deficiency of GPI-anchored proteins. The disease manifests with intravascular hemolysis, smooth muscle dystonia, thrombosis, and occasional marrow failure. Hemolysis-related free hemoglobin irreversibly scavenges nitric oxide, causing vascular dysregulation and smooth muscle symptoms such as abdominal pain, esophageal spasm, and erectile dysfunction. Thrombosis, particularly in large clones, remains the leading cause of

mortality.

In our patient, differential diagnoses included inflammatory bowel disease, infectious enteritis, and thrombotic microangiopathy. The absence of gastrointestinal bleeding, negative enteroscopic biopsy, and lack of thrombocytopenia or renal dysfunction made these less likely. Persistent anemia with hemolytic features redirected diagnosis toward PNH, confirmed by flow cytometry. Studies report 40–45% of PNH patients experience abdominal pain, often from smooth muscle dystonia and nitric oxide depletion. Given the large clone size (>80% in granulocytes and monocytes), this patient is at high thrombotic risk, and early complement inhibitor therapy is strongly recommended by guidelines.

Conclusion

Clinicians should maintain a high index of suspicion for PNH in patients presenting with unexplained abdominal pain and Coombs-negative hemolytic anemia, as timely diagnosis and treatment with complement inhibitors are crucial for improving prognosis.

病例報告 114_C091

HIV 相關類 Sjögren 氏症候群：一位 50 歲女性的不尋常表現

HIV-associated Sjögren-like Syndrome: An Unusual Presentation in a 50-year-old Woman

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Introduction

Human immunodeficiency virus (HIV) infection typically presents with acute retroviral syndrome or opportunistic infections in established high-risk populations, such as men who have sex with men, injection drug users, and individuals with multiple sexual partners or sexually transmitted infections [1,2]. We report an atypical case of HIV-associated acquired immunodeficiency syndrome presenting as Sjögren-like syndrome in a 50-year-old woman without traditional risk factors.

Case Report

A previously healthy 50-year-old woman developed progressive alopecia, xerostomia, and facial rash following mild COVID-19. Initial evaluation revealed elevated immunoglobulin E (IgE) levels and positive antinuclear antibodies, prompting referral to rheumatology. Clinical assessment and investigations were consistent with primary Sjögren syndrome; however, the patient demonstrated poor response to conventional immunosuppressive therapy. Subsequent monitoring revealed markedly elevated IgE (13,900 IU/mL) with new-onset diaphoresis and unintentional weight loss. Extensive workup excluding atopic disease, parasitic infections, malignancy, and eosinophilic granulomatosis with polyangiitis yielded negative results. HIV serological testing returned positive with severe immunosuppression (CD4+ count <35 cells/mm^3) and a high plasma HIV viral load (185,000 copies/mL). Diagnosis was established 15 months post-presentation. Following antiretroviral therapy (ART) initiation and cessation of autoimmune treatments, complete clinical resolution occurred within six months.

Discussion

This case exemplifies the diagnostic challenges associated with late HIV presentation, which affects 30-40% of newly-diagnosed HIV cases in Taiwan [3]. Risk factors for delayed diagnosis include advanced age, heterosexual transmission, female gender, injection drug use, and limited education—characteristics partially present in our patient [4-7]. Screening strategies focusing exclusively on high-risk populations may inadequately capture cases like ours, supporting universal opt-out testing approaches [8]. HIV-induced progressive CD4+ T-cell depletion triggers immune dysregulation characterized by hypergammaglobulinemia and CD8+ lymphocytosis, potentially manifesting as autoimmune-like phenomena including diffuse infiltrative lymphocytosis syndrome (DILS) [2,9]. While DILS and primary Sjögren syndrome share clinical features, such as sicca symptoms and parotid enlargement, DILS lacks disease-specific autoantibodies [10]. The American College of Rheumatology–European League Against Rheumatism (ACR-EULAR) classification criteria appropriately mandate HIV exclusion prior to

primary Sjögren syndrome diagnosis, given this clinical overlap [11].

Conclusion

This case underscores the importance of universal HIV screening to facilitate early diagnosis and treatment while reducing stigma. Clinicians should maintain heightened awareness for HIV infection when evaluating suspected autoimmune conditions, particularly in patients with atypical presentations or poor treatment responses. Comprehensive HIV testing should be considered before establishing definitive autoimmune diagnoses, regardless of perceived risk factors.

病例報告 114_C092

將藥物交互作用轉化為治療：Carbapenem 輔助清除於重度 Valproic Acid 中毒之病例報告

Harnessing a Pharmacokinetic Interaction: Carbapenem-Assisted Clearance in Severe Valproic Acid Overdose

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Introduction

Valproic acid (VPA) is a commonly prescribed broad-spectrum antiepileptic drug. However, acute intoxication can result in life-threatening complications. Clinical features include central nervous system depression, respiratory failure, and metabolic disturbances such as hyperammonemia and hepatotoxicity. We here-in report a case of severe VPA overdose successfully managed with a combination of hemodialysis, intravenous L-carnitine, and meropenem.

Case Report

A 69-year-old man with refractory epilepsy on multiple antiepileptic drugs was found unconscious at home with an empty valproate bottle at bedside. At the initial hospital, gastric lavage, activated charcoal, and naloxone were administered without clinical effect. He remained at E1V1M4 with worsening hypercapnia and respiratory acidosis, necessitating intubation and mechanical ventilation.

Upon transfer to our institution, his vital signs were stable and most laboratory results, including blood counts, biochemistry, and brain CT were unremarkable, except for mildly elevated ammonia (61 μmol/L). The serum VPA concentration was markedly elevated at 442.6 μg/mL. In the context of his history and presentation, the diagnosis of severe VPA overdose was made.

In addition to supportive care, multimodal therapy was initiated: hemodialysis to enhance elimination, meropenem to accelerate clearance via drug–drug interaction, and L-carnitine to mitigate hepatotoxicity and hyperammonemia.

After the first hemodialysis session with concurrent meropenem, his GCS improved to E3VTM6, and VPA level decreased to 165.1 μg/mL. The following morning, the VPA level remained at 143.5 μg/mL. Hemodialysis was withheld, but meropenem continued, leading to a progressive decline to 97.4 μg/mL within the therapeutic range. The patient was successfully extubated without neurological sequelae or major complications.

Discussion

Valproate toxicity arises through complex mechanisms, including increased GABAergic activity, sodium and calcium channel blockade, and carnitine depletion leading to impaired fatty acid metabolism and hyperammonemia. At therapeutic concentrations, VPA is highly protein-bound (>90%), limiting dialyzability. However, in overdose situations, protein-binding sites become saturated, increasing free drug fraction and enabling effective extracorporeal clearance. Besides, metabolic derangements, especially elevated ammonia, can also be corrected.

Carbapenems, such as meropenem, accelerate VPA elimination through a well-documented drug–drug interaction affecting its metabolism.

In our patient, rapid neurological recovery was observed after the initial HD session, with VPA level decreasing from 442.6 μg/mL to 165.1 μg/mL. Further reduction plateaued, reflecting the limited efficacy of HD at lower concentrations due to protein binding. Continued meropenem therapy sustained clearance and enabled normalization of VPA levels without rebound.
Adjunctive therapies play crucial roles in severe intoxication. L-carnitine supplementation can attenuate hyperammonemia and hepatotoxicity, and its use is recommended when patients present with coma, cerebral edema, or severe hepatic injury.

Conclusion

This case highlights the efficacy of a combined therapeutic strategy in severe VPA overdose. Integration of hemodialysis, carbapenem-induced clearance, and L-carnitine supplementation proved effective in achieving rapid recovery and preventing complications. Such a multimodal approach can optimize clinical outcomes and improve prognosis in otherwise life-threatening intoxication.

病例報告 114_C093

以惡性腸阻塞為表現之罕見同步性迴盲瓣胃癌轉移

Malignant Bowel Obstruction Caused by a Synchronous Ileocecal Valve Metastasis of Signet Ring Cell Gastric Cancer

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Introduction

Gastric cancer metastasis to the colorectum is exceptionally rare. These metastases typically arise from aggressive histologies, such as poorly differentiated or signet ring cell carcinoma, and can cause severe complications like bowel obstruction. Here, we report a highly unusual case of synchronous gastric adenocarcinoma presenting with malignant bowel obstruction from an ileocecal valve (ICV) metastasis in a patient with a complex hematologic history.

Case Report

A 75-year-old female with a history of multiple myeloma and intestinal MALT lymphoma presented with a one-month history of diffuse abdominal pain, nausea, vomiting, and melena. An esophagogastroduodenoscopy performed for the melena identified a Borrmann type III tumor in the gastric antrum, which was suspicious for either gastric adenocarcinoma or MALT lymphoma metastasis. An abdominal X-ray showed a small bowel ileus pattern, and a subsequent computed tomography scan revealed mild wall thickening of the ascending colon, dilated small bowel loops, ascites, and peritoneal nodules.

A colonoscopy revealed a stricture at the ileocecal valve (ICV), which was swollen and hyperemic, preventing passage of the scope into the terminal ileum. Two polypoid lesions were also noted in the cecum. Following biopsies and polypectomy, a comprehensive pathological analysis was conducted. The specimens confirmed metastatic signet ring cell adenocarcinoma. Immunohistochemical (IHC) profiling showed strong, diffuse positivity for Claudin-18 and weak immunoreactivity for CDX2, a profile that strongly supports a gastric primary. Further molecular analysis revealed a deficient mismatch repair (dMMR) status with isolated loss of PMS2 protein expression.

The final diagnosis was Stage IV signet ring cell gastric adenocarcinoma with peritoneal carcinomatosis and malignant small bowel obstruction, secondary to a rare synchronous metastasis to the ileocecal valve and cecum.

Discussion

This case of a rare colorectal metastasis from gastric cancer is exceptional for its synchronous presentation and exceedingly rare metastatic site. A recent systematic review identifying only 26 cases in the literature, with only 2 cases reported ICV involvement.[1,2,3] A 14-year case series estimated the incidence at a mere 0.1-0.2% among surgically treated gastric cancer patients.[4] In addition, this study found that 92.3% of such cases are metachronous, with a median disease-free interval of 34.5 months.[4]

The diagnosis was complicated by multiple challenges. The colonic lesions presented as mucosal swelling and polyps rather than a typical malignant mass.[5] Furthermore, the patient's complex history of MALT lymphoma created a broad differential diagnosis, with disease progression being a primary consideration.

Given these challenges, a definitive diagnosis was reliant on a modern IHC panel, where strong Claudin-18 positivity provided evidence of a gastric primary.[6,7] This molecular profiling also had profound therapeutic implications, shifting the management paradigm to systemic, molecularly-guided therapy.

Conclusion

This case highlights the rare and aggressive course of signet ring cell gastric cancer presenting as a synchronous metastasis to the ileocecal valve causing bowel obstruction. It underscores that metastatic disease must be a differential for new intestinal lesions, even in patients with complex oncologic histories. A comprehensive approach is crucial for an accurate diagnosis and guiding personalized systemic therapies in advanced-stage presentations.

病例報告 114_C094

本土日本腦炎併發雙側肺栓塞的病例報告

A 64-year-old confirmed case of Japanese encephalitis complicated with bilateral pulmonary embolism

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Introduction

Japanese encephalitis (JE) is one of the most important causes of epidemic viral encephalitis worldwide. In Taiwan, most reported cases reside in southern region. Since a universal JE vaccination program for children in Taiwan was started in 1968, most symptomatic cases occur in the elderly. Typical presentations include fever with abnormal motor behavior. Definite diagnosis requires viral isolation or presence of viral-specific IgM in cerebrospinal fluid (CSF) study.

Case Report

A previously healthy 64-year-old man who worked as a bank executive in Yunlin, Taiwan initially experienced right-hand clumsiness with progression to right hemiparesis in two days in September, 2024. High grade fever subsequently developed with productive cough and eventually presented with acute altered mental status. Laboratory tests revealed leukocytosis and thrombocytopenia. Serology was positive for herpes simplex virus (HSV) 1 and 2 immunoglobulin M (IgM). Intravenous acyclovir and empirical ceftriaxone and vancomycin were administered. CSF study revealed elevated total protein level without hypoglycorrhachia. However, HSV DNA, aerobic culture, tuberculosis polymerase-chain-reaction (PCR), varicella-zoster virus DNA, pan-enterovirus RNA PCR and cytomegalovirus viral load were negative in the CSF. The patient remained in poor consciousness level under antimicrobial agents, raising suspicion of an alternative causative pathogen. Based on the epidemiology, JE was suspected. CSF later reported positive for JE virus-specific IgM, confirming the diagnosis. During hospitalization, acute onset hypoxemic respiratory failure developed. Computed tomography disclosed bilateral pulmonary embolism. After days of anticoagulant treatment, respiratory failure resolved. Consciousness improved and the patient was eventually able to walk with assistance upon hospital discharge.

Discussion

We present a case who worked in a business area without identified contact history. Because the patient presented with fever and neurological symptoms in the seasonal peaking timing of the year in the relative prevalent subregion in Taiwan, JE was considered. Although positive serum HSV-1/2 IgM was reported initially, JE and HSV encephalitis differ in clinical features and regions of CNS involvement. Moreover, the presentation was complicated by bilateral pulmonary embolism, potentially associating with the sequelae of JE viral infection and concurrent cerebral venous sinus thrombosis reported in the literature. Possible aspects of the pathophysiology of thromboembolism formation in JE include immobilization resulting in hemostasis, systemic infection giving rise to hypercoagulability and viral penetration contributing to endothelial injury.

Conclusion

JE virus infected patients may present with variable neurological manifestations. Systemic infection of JE also promotes thrombosis formation. Thus, if new onset symptoms of dyspnea or neurological deficits occur, thromboembolic should be taken into account.

病例報告 114_C095

濾泡性甲狀腺癌合併雙側腎轉移導致末期腎臟病：病例報告

Follicular Thyroid Carcinoma with Bilateral Renal Metastases Leading to End-Stage Renal Disease: A Case Report

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Introduction

Follicular thyroid carcinoma (FTC) represents 6% of thyroid malignancies and typically metastasizes hematogenously to lungs and bones[1]. Renal metastases are exceedingly rare, with only sporadic case reports documented. We report a case of widely invasive follicular thyroid carcinoma that initially presented as bilateral renal masses, ultimately leading to end-stage renal disease requiring hemodialysis, and the subsequent challenges encountered in delivering RAI therapy.

Case Report

A 47-year-old Taiwanese male presented with several months of asymptomatic gross hematuria. He was initially seen in the outpatient urology clinic, where renal ultrasonography revealed bilateral kidney masses. Subsequent abdominal computed tomography (CT) scan revealed bilateral renal tumors and suspicious metastatic lesions at the T3, L2–L3, and L5 vertebral levels at a tertiary medical center. Bilateral radical nephroureterectomy showed follicular-like tumors breaching the renal surface; immunohistochemistry confirmed thyroid origin. Subsequent thyroid ultrasonography demonstrated irregular boundary left thyroid nodule with calcification, and total thyroidectomy confirmed FTC (negative *BRAF V600E*, positive *NRAS*, focally positive *Galectin-3*, positive *HBME-1,* negative *pan-TRK*). Initial staging was T3aNxM1.

Postoperatively, the patient received adjuvant reduced-dose radioactive iodine (I-131, 50 mCi) due to end-stage renal disease requiring lifelong hemodialysis after bilateral nephrectomy. Follow-up imaging and laboratory studies have shown no evidence of recurrence to date.

Discussion

This case illustrates several important points. First, bilateral renal masses as the initial manifestation of FTC is extraordinarily uncommon. The diagnosis of metastatic thyroid carcinoma in the kidney can be challenging, particularly when the primary thyroid tumor is small or occult. Diagnosis required immunohistochemical confirmation given the unusual presentation.

Second, the pathophysiology of FTC involves distinct molecular alterations compared to papillary thyroid carcinoma. NRAS mutations, occur in approximately 40-50% of FTCs, are associated with hematogenous dissemination and aggressive behavior. The NRAS mutation identified in our patient may have facilitated early distant metastasis and the unusual renal involvement [1, 2].

Management of RAI therapy in patients with ESRD necessitates dose adjustment due to impaired renal clearance and prolonged radiation exposure. Evidence from current literature indicates that empiric doses should be reduced by 13–28% in thyroid cancer patients on hemodialysis, while individualized dosimetric approaches are preferable whenever possible [3, 4]. Our empiric 50 mCi

administration aligns with these guidelines. Dialysis timing after RAI remains debated, ranging from immediate post-treatment dialysis to delayed initiation after 42-48 hours, underscoring the need for multidisciplinary collaboration[3, 4].

Conclusion

This rare case of widely invasive FTC initially presenting with bilateral renal metastases illustrates the diagnostic challenge of atypical metastatic patterns. NRAS mutation may drive aggressive behavior. In patients with ESRD, RAI therapy requires individualized dose adjustment and close multidisciplinary coordination. This case contributes to the limited literature on thyroid carcinoma with renal metastases and provides practical insights into the multidisciplinary management challenges encountered in this rare clinical scenario. Broader experience and standardized protocols are needed for optimizing therapy in such rare scenarios.

病例報告 114_C096

空腸彎曲桿菌與 Sapovirus 合併感染引起之迴腸末端炎與腸繫膜淋巴結炎：類似闌尾炎的臨床病例報告

Terminal Ileitis and Mesenteric Adenitis Imitating Appendicitis Due to *Campylobacter jejuni* and Sapovirus Co-Infection: A Case Report

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Introduction

Foodborne diarrhea is usually self-limiting, though causative pathogens are often difficult to identify. Empiric fluoroquinolone use is common, yet rising resistance is a growing concern. Multiplex stool PCR provides a rapid diagnosis that can reduce unnecessary interventions[1]. We describe a rare case of *Campylobacter jejuni* and sapovirus coinfection in an immunocompetent adult, which underscores the diagnostic utility of multiplex PCR and the need for tailored therapy.

Case Report

A 24-year-old healthy woman presented with acute abdominal pain and fever. Ten days prior, she traveled to Kyoto, Japan and consumed raw food including eggs, chicken, beef, and oysters. She denied rural travel or cluster history. Five days before admission, upon returning to Taiwan, she developed epigastric pain with fever up to 39°C, chills, nausea, and bilateral flank soreness. Initially, she took antipyretics and levofloxacin for presumed urinary tract infection. Her condition worsened with recurrent fever (38°C), lethargy, and progressive dull to cramping abdominal pain that migrating to the right lower quadrant, along with watery diarrhea containing mucus and blood streaks after 4 days.

Physical examination revealed McBurney point tenderness with mild rebound tenderness. Contrast-enhanced computed tomography showed inflammatory changes from terminal ileum to right ascending colon, with small but prominent para-aortic lymph nodes. She was admitted for hydration and empiric flomoxef for enterocolitis, as stool microscopy demonstrated >100 white blood cells per high-power field. FilmArray multiplex stool PCR detected *Campylobacter* spp. and sapovirus; former was confirmed by stool culture confirmed ciprofloxacin-resistant *C. jejuni*. Following a 3-day course of azithromycin, her symptoms gradually resolved and she was discharged uneventfully.

Discussion

This case illustrated terminal ileitis and colitis caused by *C. jejuni* and sapovirus co-infection mimicking appendicitis. *C. jejuni* occasionally causes severe disease with high fever and bloody diarrhea among Asians[2]. Sapovirus generally produces mild watery diarrhea[3]; however, co-infections can worsen illness through synergistic effects on immune activation, pro-inflammatory cytokine release, epithelial injury, and intestinal microbiota disruption[4-6]. Studies in pediatric patients[7] demonstrate that coinfection with multiple intestinal pathogens result in prolonged disease duration and more systemic symptoms compared to single-pathogen infections[7].

Most *C. jejuni* gastroenteritis cases are self-limiting that adequate rehydration remains the

mainstay of treatment. Due to emerging fluoroquinolone resistance, the Infectious Diseases Society of America recommends macrolides as the first-line antimicrobial agents for *C. jejuni* gastroenteritis, particularly in severe or prolonged cases[8]. The primary mechanism of fluoroquinolone resistance involves a point mutation in the *gyrA* gene, encoding the A subunit of DNA gyrase[9]. Besides, sapovirus, a relatively uncommon pathogen, has been reported in several cluster outbreaks in Taiwanese schools, typically causing mild gastroenteritis. Co-infection with these two pathogens has not been previously reported. Multiplex PCR stool panels provides substantial diagnostic value in enteric infections with excellent specificity and diagnostic accuracy across a wide range of notorious and difficult-to-diagnose pathogens[10].

Conclusion

C. jejuni and sapovirus co-infection can cause pseudo-appendicitis in healthy adults. Rapid diagnosis via stool multiplex PCR demonstrated that molecular testing is a timely and accurate method to detect uncommon pathogens, though conventional culture remains essential for determining amicrobial resistance.

病例報告 114_C097

一位肝細胞癌患者於免疫相關性肝炎接受類固醇治療期間之侵入性肺麴菌症

Proven Invasive Pulmonary Aspergillosis in Hepatocellular Carcinoma During Glucocorticoid Therapy for IrAE Hepatitis

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Introduction

Invasive pulmonary aspergillosis (IPA) is most commonly seen in immunocompromised individuals, including patients receiving prolonged courses of high-dose glucocorticoids, with prednisone daily equivalent more than 20 mg. We report a case of hepatocellular carcinoma (HCC) complicated by immune-related hepatitis requiring glucocorticoid therapy, diagnosed with proven IPA caused by *Aspergillus fumigatus*.

Case Report

A 71-year-old man with HCC and chronic hepatitis B presented with blood-tinged sputum and progressive dyspnoea for one week, with a fever and poor appetite. He was a lifelong non-smoker, employed as a civil servant, without history of botanic or zoonotic exposure. His baseline functional status was fully independent.

Two months before this evaluation, he was actively treated for stage C HCC, without metastasis, based on the Barcelona Clinic Liver Cancer classification with a combination of atezolizumab and bevacizumab, along with a monoclonal antibody targeting leukocyte immunoglobulin-like receptor B2 (LILRB2) as part of a clinical trial. His course was complicated by biopsy-proven immune-related hepatitis, for which he received glucocorticoids starting from methylprednisolone 1.1 mg/kg/day one month ago, remaining on prednisone at 0.5 mg/kg/day at presentation.

Physical examination revealed decreased breath sounds over the right lung and abdominal distension without tenderness. Laboratory studies indicated thrombocytopenia (6,600/μL), hyperbilirubinemia (3.11 mg/dL), elevated C-reactive protein (6.38 mg/dL), and liver enzyme (ALT 162 U/L). Chest computed tomography illustrated bilateral patchy opacities with halo signs, consolidation in periphery, and cavitary changes. Serum *Aspergillus* galactomannan antigen optical density (OD) was positive at 6.4, raising suspicion for IPA; therefore, isavuconazole was initiated.

Bronchoscopy with endobronchial ultrasound and transbronchial needle-aspiration identified concentric hypoechoic peribronchial lesions in RB9, and histopathology revealed suppurative necrotising inflammation with numerous acute-angle branching, septated hyphae highlighted by Grocott methenamine silver and periodic acid–Schiff stains. This finding fulfilled criteria for proven IPA and further supported by *Aspergillus* galactomannan antigen OD positive at 2.66 from bronchial washings. Targeted next-generation sequencing (tNGS) of bronchial washing identified *Aspergillus fumigatus*, confirmed by fungal culture eight days later.

The patient's symptoms gradually improved with isavuconazole, and the serum *Aspergillus* antigen OD decreased to 4.1 after one week of therapy. However, his condition deteriorated when developing *Klebsiella pneumoniae* bacteraemia, leading to refractory septic shock, lactic acidosis, and respiratory failure. Despite appropriate antibiotics, high-dose vasopressors, continuous renal replacement therapy, and mechanical ventilation, he expired because of multiorgan failure.

Discussion

According to the revised 2020 EORTC/MSGERC definitions of invasive fungal diseases, this patient satisfied host factors for IPA regarding prolonged glucocorticoid therapy against immune-related hepatitis. Glucocorticoid may compromise the host immunity and facilitate fungal growth. Although corticosteroid exposure is a well-recognised risk factor, the clinical features, demographics and incidence, and treatment outcomes of IPA in patients with immune-related adverse events remain unexplored. Moreover, the contribution of the LILRB2 inhibitor in this case is uncertain. Meanwhile, tNGS provided early recognition of the culprit fungi, offering advantages of precise and timely antifungal therapy.

Conclusion

This case highlights needs for further studies regarding IPA risk and management in immunotherapy-treated patients, as the role of immune checkpoint inhibitors is increasingly significant in cancer therapy.

病例報告 114_C098

以腎上腺結節增生做為初始表現之庫欣氏病：案例報告

Cushing disease initially presented as macronodular adrenal hyperplasia: A case report and review of literature

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Introduction

Cushing syndrome (CS) is characterized by elevated plasma cortisol levels resulting from exogenous exposure or endogenous overproduction. Common manifestations include moon face, central obesity, proximal myopathy, striae, hirsutism, and impaired glucose tolerance. Cushing disease (CD), a subset of Cushing syndrome, results from corticotropin (ACTH)-secreting pituitary adenomas. We report a case of recurrent CS, initially presenting with adrenal nodules and later diagnosed as CD. This case illustrates the diagnostic challenges of CD and highlights the importance of identifying the primary etiology of CS to guide appropriate treatment.

Case Report

A 61-year-old male presented in 2020, with bilateral adrenal nodules and left predominance incidentally discovered on abdominal computed tomography (CT). He reported a six-month history of weight gain, easy bruising, and muscle weakness. Physical examination revealed buffalo hump, central obesity, and purple striae. Initial laboratory testing showed elevated cortisol with loss of diurnal cortisol rhythm, while ACTH levels were within normal range. Pituitary magnetic resonance imaging (MRI) revealed no pituitary lesion. To confirm laterality, NP-59 adrenal scintigraphy was performed, which demonstrated more prominent enhancement of left adrenal nodules. CS due to hypercortisolism from left adrenal adenoma was favored, and left adrenalectomy was performed in February 2021, with gradual clinical and biochemical improvement.

In May 2023, the patient developed recurrent symptoms, including facial acne, oily scalp, and lower limbs weakness. Adrenal hormone testing showed recurrent CS with more markedly elevated ACTH. Adrenal CT revealed recurrent bilateral adrenal nodules. However, pituitary MRI identified a 4 mm microadenoma. Inferior petrosal sinus sampling (IPSS) in January 2024 confirmed CD. Transsphenoidal adenomectomy was performed in February 2024, with symptoms relieved and biochemical remission. Follow-up MRI in May 2024 revealed postoperative changes without residual tumor.

Discussion

The patient initially presented with CS and normal ACTH levels. Imaging studies revealed bilateral adrenal adenomas with left predominance, while pituitary MRI reported negative result, which led to left adrenalectomy. Two years later, recurrent CS with markedly elevated ACTH was diagnosed, and further MRI and IPSS confirmed CD. Literature review suggests the presentation compatible with ACTH-dependent macronodular adrenal hyperplasia (MAH), a form of CD characterized by chronic ACTH stimulation causing adrenal hyperplasia and autonomous cortisol production. This

autonomous secretion of cortisol suppresses ACTH, resulting in relatively low ACTH levels, a phenomenon referred to as “auto-suppression.” We postulate that a tiny, undetectable ACTH-secreting pituitary microadenoma initially stimulated adrenal growth and cortisol secretion in the present case. Adrenalectomy removed the source of auto-suppression, and led to pituitary tumor progression and recurrence. As up to 23% of CD patients have normal pituitary MRI findings, IPSS remains the gold standard of diagnosing CD. However, due to limited availability, procedural risks and costs of IPSS, shared decision-making is essential.

Conclusion

Adrenal tumors with laterality, such as MAH, can be initial presentation of CD. Therefore, identifying the primary cause of CS is essential. When clinical findings and test results are inconsistent, differential diagnosis should be reconsidered. A negative pituitary MRI does not exclude CD, and IPSS remains the most valuable diagnostic tool in challenging cases.

病例報告 114_C099

帶狀皰疹引起多發神經病變：罕見病例報告

Herpes Zoster–Associated Polyradiculopathy: A Rare Case Report

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Introduction

Herpes zoster results from reactivation of latent varicella–zoster virus in the dorsal root ganglia. It typically presents with a dermatomal vesicular rash and neuropathic pain. Neurological complications include postherpetic neuralgia, segmental motor weakness, and, rarely, polyradiculopathy. Polyradiculopathy involves multiple spinal nerve roots, leading to motor and sensory deficits that extend beyond a single dermatome.

Case Report

Herpes zoster with skin eruptions and neuropathic pain is common in clinical practice. We report a rare case involving weakness across multiple nerve root levels secondary to herpes zoster.

A 90-year-old woman presented after an accidental fall from her bed. She remained bedridden for one week due to low back pain. Subsequently, she developed a progressively painful vesicular rash over one week, extending from the left inguinal region to the medial thigh and down to the lower leg. Upon admission, she complained of inability to move her left leg in addition to the painful rash. Clinical examination revealed involvement of multiple dermatomes (L2–L5) (Figure 1). She was treated with oral acyclovir 200 mg every 4 hours and gabapentinoids for neuropathic pain. Nerve conduction studies and electromyography indicated axonal sensorimotor polyneuropathy. Given the diagnosis of herpes zoster–associated polyradiculopathy, dexamethasone 4 mg twice daily was initiated. After four days of steroid therapy, her left foot mobility improved, although she remained unable to lift her leg. A rehabilitation program was arranged to aid muscle recovery.

Discussion

Polyradiculopathy is an uncommon manifestation of herpes zoster, likely caused by viral spread within spinal cord segments. Clinicians should consider this diagnosis in patients with zoster who present with multifocal weakness and sensory deficits extending beyond a single dermatome. Early recognition and prompt antiviral therapy are essential to improve functional outcomes and prevent long-term disability.

病例報告 114_C100

以頑固性高血壓為表現之功能性腎上腺皮質癌：病例報告

Functional Adrenocortical Carcinoma Presenting with Refractory Hypertension: A Case Report

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Introduction

Refractory hypertension in young adults presents a significant clinical challenge that warrants a thorough investigation for secondary causes. While essential hypertension is the most common diagnosis, endocrine disorders, though rare, can be life-threatening if missed. Adrenocortical carcinoma (ACC) is one such rare cause. We present the case of a young woman whose refractory hypertension was the presenting sign of a functional ACC, highlighting the critical importance of a comprehensive diagnostic workup in this patient population.

Case Report

A 34-year-old female presented with refractory hypertension and Cushingoid features including moon face and weight gain, as well as signs of hyperandrogenism such as acne, a growing beard, and an irregular menstrual cycle. A workup for secondary hypertension revealed suppressed ACTH levels (<1.5 pg/mL) with normal cortisol level (14.7 μg/dL, normal range 6.02-18.4 μg/dL). A 1-mg overnight dexamethasone suppression test (ODST) failed to suppress cortisol (16.3 μg/dL), and late-night salivary cortisol was elevated at 0.69 μg/dL (normal <0.27 μg/dL), confirmed a diagnosis of ACTH-independent Cushing's syndrome. Elevated DHEA-S level (1192 ug/dL) was also noted. An abdominal computed tomography (CT) scan subsequently identified a large 16.8 cm right adrenal tumor, presumed to be an adrenocortical carcinoma, with attachment to the liver, right kidney, and direct invasion into inferior vena cava (IVC). Initial staging scans also revealed multiple pulmonary nodules and evidence of pulmonary embolism. Consequently, the patient underwent a radical en-bloc resection, which included a right adrenalectomy, right hepatic lobectomy, cholecystectomy, right nephrectomy, and IVC resection with reconstruction. The final pathology confirmed high grade ACC. Postoperatively, the patient was started on an adjuvant EDP-M (Etoposide, Doxorubicin, Cisplatin plus Mitotane) regimen. However, her disease continued to progress, and after failure of several subsequent lines of therapy, she ultimately opted for home hospice care.

Discussion

This case underscores that refractory hypertension in a young patient can be the initial manifestation of functional ACC; therefore, a thorough investigation for secondary causes is crucial in this clinical scenario. For localized ACC, complete surgical resection remains the only curative treatment and the most important determinant of long-term survival. However, the prognosis for metastatic ACC is poor. As this case illustrates, despite standard-of-care treatment with the EDP-M regimen and subsequent lines of therapy, the clinical outcome was unfavorable, reflecting the aggressive nature of ACC and the limited efficacy of current systemic therapies.

Conclusion

This case serves as a powerful reminder that patients with atypical or refractory hypertension require a high index of suspicion for secondary causes, for which a timely and systematic evaluation is essential. Although complete surgical resection is the cornerstone of management for localized ACC, the aggressive course of metastatic disease, as seen in this patient, highlights the urgent need for more effective therapeutic strategies to improve patient outcomes.

病例報告 114_C101

Ivabradine 成功應用於甲狀腺風暴合併乙型阻斷劑禁忌：病例報告

Successful Use of Ivabradine for Rapid Heart Rate Control in Thyroid Storm with Contraindication to Beta-Blockade: A Case Report

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Introduction

Thyroid storm (TS) is a rare but life-threatening endocrine emergency, with reported mortality rates up to 25% despite advances in therapy. Tachycardia is a hallmark manifestation of TS and often contributes to hemodynamic instability. Current guidelines recommend beta-blockers to control heart rate, but it may be hazardous in patients with acute decompensated heart failure or bronchospasm. Fatal circulatory collapse has been reported following their administration in TS. Thus, alternative approaches for rate control are of clinical importance.

Ivabradine is a selective inhibitor of the funny current (If) in sinoatrial node pacemaker cells, reducing heart rate without impairing myocardial contractility or bronchial tone. Herein, we present a patient with TS whose tachycardia was effectively managed with ivabradine when beta-blockers were contraindicated.

Case Report

A 62-year-old woman with a history of Graves' disease and asthma presented with one week of palpitations and progressive dyspnea. On admission, she was febrile, hypertensive, and markedly tachycardic in sinus rhythm at 130–150/min. Chest radiography revealed bilateral pulmonary edema, and thyroid function tests demonstrated suppressed TSH (0.12 μIU/mL) with elevated free T4 (2.68 ng/dL). Her Burch–Wartofsky Point Scale score was 65, consistent with TS complicated by acute decompensated heart failure.

She was treated with propylthiouracil, Lugol's solution, hydrocortisone, diuretics, and bronchodilators. Beta-blockers were avoided due to her underlying asthma and acute decompensated heart failure. Following cardiology consultation, ivabradine 2.5 mg twice daily was initiated on day 1, resulting in a rapid reduction of heart rate to <100/min within hours and improvement in respiratory status. Ivabradine was discontinued on day 4, and the patient was discharged on day 6 in stable condition with continued antithyroid therapy. Her thyroid function normalized within four weeks, and no recurrence of thyrotoxicosis or tachyarrhythmia was observed during follow-up.

Discussion

This case illustrates the potential role of ivabradine in managing sinus tachycardia during TS when beta-blockers are contraindicated. Preclinical studies suggest that thyroid hormone enhances If current density, supporting the rationale for ivabradine in thyrotoxicosis-induced tachycardia. By selectively inhibiting the If current, ivabradine reduces heart rate without impairing contractility or inducing bronchospasm, offering distinct advantages in patients with cardiopulmonary compromise. Our patient demonstrated rapid stabilization with ivabradine, consistent with

limited reports in the literature.

However, the efficacy and safety profile of ivabradine in TS remain insufficiently defined. For example, unlike beta-blockers, ivabradine does not inhibit the peripheral conversion of thyroxine to triiodothyronine, highlighting its limitation in addressing the underlying thyrotoxic state. Moreover, large-scale analyses in cardiovascular disease populations have associated ivabradine with an increased risk of atrial fibrillation, an arrhythmia that is already frequently observed in TS. These concerns underscore the need for careful patient selection and monitoring. Further clinical studies are required to establish the efficacy, optimal dosing, and safety of ivabradine in TS.

Conclusion

Taken together, this case demonstrates that ivabradine may represent a promising alternative strategy for heart rate control in TS when beta-blockers are contraindicated. Future studies are warranted to define its efficacy, safety, and evidence-based application in this high-risk population.

病例報告 114_C102

一例接受 Nivolumab 治療後出現猛爆性第一型糖尿病的轉移性胃腺癌病人之病例報告

Nivolumab-Induced Fulminant Type 1 Diabetes Mellitus in a Patient with Metastatic Gastric Adenocarcinoma: A Case Report

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Introduction

Nivolumab, a PD-1 blocking antibody, restores cytotoxic T-cell activity and enhances antitumor immune responses. In combination with chemotherapy, it is an established treatment for PD-L1–positive advanced gastric adenocarcinoma. However, excessive T-cell activation may trigger autoimmune destruction, and type 1 diabetes mellitus (T1DM) is a rare but serious immune-related adverse event (irAE) of the drug. Here, we present a case of a 75-year-old patient who developed fulminant T1DM during nivolumab treatment for metastatic gastric adenocarcinoma.

Case Report

A 75-year-old man with gastric adenocarcinoma underwent subtotal gastrectomy in May 2020. He had no history of diabetes, with-in-normal-range HbA1c (5.1% in May 2025), and no steroid exposure. His body shape (168cm, 67 kg), BMI (23.7 kg/m^2), and nutritional status remained stable after surgery. In June 2025, new liver metastases were detected. He received XELOX (oxaliplatin + capecitabine) plus nivolumab (PD-L1 CPS positive) from June 19, 2025, with dexamethasone 10 mg as part of the regimen. After four cycles (last on August 14, 2025), he developed dyspnea, epigastric pain, and decreased body weight (7 kg in 2 months), presenting to NCKUH emergency department on August 24, 2025. Laboratory evaluation revealed severe hyperglycemia (serum glucose: 1125 mg/dL), ketosis (β-hydroxybutyrate: 3.4 mmol/L), hyperosmolarity (measured osmolality: 349 mOsm/KgH2O), and high anion gap metabolic acidosis (Na: 124 mmol/L, Cl: 91 mmol/L, pH: 6.894, PCO2: 12.0 mmHg, PaO2: 155.0 mmHg, HCO3-: 2.3 mmol/L), consistent with diabetic ketoacidosis (DKA) and hyperosmolar hyperglycemic state (HHS). Further testing showed a negative GAD antibody, low C-peptide (<0.01 ng/mL), and mild elevated HbA1c (6.2%), and a glucagon stimulation test revealed unstimulated C-peptide level (0’ C-peptide: <0.01 ng/mL, 6’ C-peptide: <0.01 ng/mL) confirming the immune checkpoint inhibitor (ICI) –induced T1DM. He was treated with insulin infusion, transitioned to subcutaneous insulin, and discharged with close glucose monitoring.

Discussion

T1DM is a rare irAE of nivolumab, caused by autoreactive T-cell–mediated destruction of pancreatic β-cells, leading to rapid and irreversible insulin deficiency. Onset is typically abrupt and often presents as DKA with severe hyperglycemia and low or absent C-peptide levels. Long-term insulin dependence is common, as most patients fail to recover endogenous insulin secretion. Although the development of T1DM doesn’t appear to significantly affect overall survival, regular glucose monitoring and surveillance for coexisting endocrinopathies are essential.

Conclusion

Fulminant T1DM is a rare but serious complication of nivolumab that often presents with DKA. Early recognition, close glucose monitoring, and prompt insulin therapy are essential.

病例報告 114_C103

腹膜透析導管移除後持續發熱之培養陰性腹膜炎：Mycobacterium avium 複合群以分子方法確診並成功治療

Persistent Fever after Peritoneal Dialysis Catheter Removal: Culture-Negative Peritonitis Molecularly Diagnosed as Mycobacterium avium Complex and Successfully Treated

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Introduction

Nontuberculous mycobacteria (NTM) are uncommon causes of peritoneal dialysis (PD)–related peritonitis and are often underrecognized because cultures are negative and symptoms nonspecific. Mycobacterium avium complex (MAC) is particularly rare in non-HIV PD patients. We report MAC peritonitis with concomitant pleurisy, highlighting diagnostic pitfalls and the value of molecular testing and early targeted therapy.

Case Report

A 60-year-old man with end-stage kidney disease from type 2 diabetes had received PD for four months when he developed one week of turbid dialysate, abdominal pain, malaise, and intermittent fever. Dialysate effluent analysis showed neutrophil-predominant leukocytosis (600/μL; 85% neutrophils). Intraperitoneal ceftazidime and vancomycin were started for presumed PD peritonitis. Despite therapy, fever persisted and effluent leukocytosis worsened (1,346/μL, neutrophil 70%); serial blood and effluent cultures were negative. The PD catheter was removed and hemodialysis commenced, but fever continued despite piperacillin–tazobactam followed by doripenem, prompting transfer.

On hospital day (HD) 4, paracentesis revealed 894 WBC/μL with a mononuclear shift; Gram, acid-fast bacilli (AFB) stain, and bacterial/mycobacterial cultures were negative. Chest radiography suggested right pleural effusion, and contrast CT showed bilateral pleural effusions, ascites, and omental thickening. A gallium-67 scan on HD13 showed mild uptake at the hepatic dome and right lower lung. On HD17, temperature >39 °C and hypercalcemia developed (corrected calcium 11.68 mg/dL) with suppressed iPTH and low 25-hydroxyvitamin D, arguing against hyperparathyroidism or vitamin D intoxication.

Repeat sampling on HD19 (paracentesis and right-sided thoracentesis) yielded exudative fluids with lymphocyte/macrophage predominance; all cultures remained negative. On HD21, multiplex DNA microarray of both fluids identified MAC; HIV serology was negative. Laparoscopic biopsies on HD25 demonstrated non-caseating granulomatous peritonitis (AFB and GMS stains negative) and chronic pleuritis without granulomas. The final diagnosis was MAC peritonitis with associated pleurisy in a non-HIV patient previously on PD.

Targeted therapy was started on HD25: rifampicin 600 mg once daily, clarithromycin 500 mg once daily, and ethambutol 800 mg three times weekly. Fever abated within days; liver enzymes improved; chest radiography and abdominal ultrasonography showed resolution of pleural effusion and ascites. He was discharged on HD33 and, at 10-month follow-up, remained afebrile and clinically stable on maintenance hemodialysis without recurrence.

Discussion

NTM peritonitis is diagnostically challenging because culture negativity and overlap with bacterial peritonitis delay recognition. Rapidly growing mycobacteria (e.g., M. fortuitum, M. chelonae) dominate PD cohorts, whereas MAC is uncommon; however, MAC is prominent among NTM pathogens in Taiwan. Concurrent pleural involvement in PD suggests trans-diaphragmatic spread driven by pressure gradients. In culture-negative presentations, applying molecular assays to sterile fluids can expedite diagnosis and justify early therapy, especially with supportive histopathology. Treatment for MAC in extrapulmonary disease is extrapolated from pulmonary/disseminated guidelines; macrolide-based triple therapy for ≧6 – 12 months is standard. Our patient's rapid, durable response to clarithromycin – rifampicin – ethambutol supports this approach.

Conclusion

MAC should be considered in PD patients with persistent, culture-negative peritonitis unresponsive to conventional antibiotics. Integrating molecular diagnostics with histopathology enables timely, targeted therapy and favorable outcomes, even when pleural involvement is present.

病例報告 114_C104

一例初期以氣喘表現之疑似第 3 型遺傳性血管性水腫病例報告

A Case Report of Suspected Type III Hereditary Angioedema Presenting Initially as Asthma

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Introduction

Hereditary angioedema (HAE) is a rare genetic disorder causing recurrent swelling of the skin, airways, and gastrointestinal tract due to bradykinin-mediated vascular permeability. It is classified into three types based on C1 esterase inhibitor (C1-INH) status. Types I and II result from SERPING1 mutations: Type I shows reduced C1-INH levels, while Type II has normal or elevated levels with impaired function. Type III features normal C1-INH, predominantly affects women, and is often estrogen-related. We report a 44-year-old woman initially presenting with asthma-like symptoms, later suspected to have type III HAE.

Case Report

A 44-year-old kindergarten teacher was diagnosed with asthma in her late thirties. She was initially treated with high-dose inhaled corticosteroid/long-acting β_2-agonist (ICS/LABA) but continued to experience dyspnea, chest tightness, and upper-airway wheezing. Oral corticosteroids were added after bronchodilator testing showed reversibility despite a normal FEV1/FVC ratio. Over time she developed facial swelling and easy bruising.

Trials of biologics including benralizumab and tezepelumab failed to improve her symptoms, which now included hoarseness and recurrent swelling. Because symptoms persisted despite therapy, angioedema was suspected. Laboratory evaluation showed normal IgE, tryptase, and mast-cell counts. Her edema was unresponsive to antihistamines and corticosteroids, suggesting a bradykinin-mediated process. Complement levels (C3, C4, and C1-INH) were normal. Whole-exome sequencing excluded *SERPING1* mutations but identified a single-nucleotide polymorphism in *KNG1*, raising strong suspicion for Type III HAE. Her exacerbations improved with fresh-frozen plasma, but omalizumab did not reduce the frequency or severity of attacks.

Discussion

Angioedema is a localized, non-pitting swelling that can involve the face, extremities, airway, or gastrointestinal tract. It may be mediated by histamine, as in allergic reactions, or by bradykinin, as in HAE. HAE has an estimated prevalence of 1 in 50,000 and is typically inherited in an autosomal dominant manner. While most cases are due to C1-INH deficiency (Types I and II), HAE-nC1INH presents with normal complement levels and is often linked to mutations in *F12*, *PLG*, *KNG1*, *ANGPT1*, and *MYOF*.

Diagnosis requires careful history and laboratory evaluation. In Types I and II, low C4 and C1-INH levels are diagnostic. In Type III, values remain normal, and genetic testing may be required. Imaging may demonstrate bowel wall edema during abdominal attacks, and laryngoscopy is useful in evaluating airway involvement. Because corticosteroids, antihistamines, and epinephrine are generally ineffective, acute management includes C1-INH replacement, bradykinin B2-

receptor antagonists, kallikrein inhibitors, or fresh-frozen plasma. Long-term prophylaxis can be achieved with lanadelumab or berotralstat.

Our patient showed the diagnostic difficulty of HAE-nC1INH, particularly when airway symptoms mimic asthma. The lack of response to corticosteroids, antihistamines, and biologics should prompt clinicians to consider a bradykinin-mediated process. Identification of a *KNG1* variant further supports genetic heterogeneity in this subtype.

Conclusion

This case illustrates the importance of considering HAE with normal C1-INH in patients with unexplained angioedema or asthma-like symptoms refractory to conventional therapy. Early recognition enables use of targeted treatments, including plasma-derived or bradykinin-directed therapies, which are essential to reduce morbidity and prevent life-threatening airway compromise.

病例報告 114_C105

Rituximab 治療因紅血球生成素引起之抗體導致的純紅血球再生不良：一名慢性腎臟病患者病例報告

Rituximab Treatment for ESA-Induced Pure Red Cell Aplasia in a Dialysis-Dependent CKD Patient: A Case Report

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Introduction

Pure red cell aplasia (PRCA) is a rare but serious complication of erythropoiesis-stimulating agent (ESA) therapy, occurring in 0.02-0.5% of chronic kidney disease (CKD) patients. ESA-induced PRCA typically results from neutralizing anti-EPO antibodies. This case report describes a Taiwanese man with CKD stage 5 who developed ESA-induced PRCA and demonstrated good hematologic response to rituximab therapy.

Case Report

A 76-year-old man with CKD stage 5 on maintenance hemodialysis and newly diagnosed hepatocellular carcinoma had received epoetin alfa and methoxy polyethylene glycol-epoetin beta for renal anemia over 11 months with initial good response. He presented with acute severe anemia (hemoglobin dropped from 8.0 to 3.9 g/dL) and reticulocyte count of 0.06%. Bone marrow biopsy revealed marked erythroid hypoplasia with M/E ratio >10:1 and markedly decreased erythroblasts, and neutralizing anti-EPO antibodies were detected, confirming ESA-induced PRCA diagnosis.

Rituximab was selected over conventional immunosuppressants due to his advanced age, active malignancy, dialysis dependence, and multiple comorbidities. He received four weekly infusions of rituximab (100 mg/week) from May to June 2024. Remarkable hematologic recovery was achieved: reticulocyte count increased from 0.06% to 2.5% within two months, hemoglobin stabilized from 3.9 to 7-8 g/dL, and transfusion requirements significantly decreased from biweekly to occasional needs. Iron overload developed with ferritin rising to 3303 ng/mL from repeated transfusions and was successfully managed with oral deferasirox 360mg daily.

Discussion

This case emphasizes the importance of recognizing ESA-induced PRCA in CKD patients presenting with sudden severe anemia despite prior stable ESA response. Early anti-EPO antibody testing is crucial for diagnosis. Rituximab could be a treatment option for patients intolerable with traditional immunosuppressants, providing acceptable efficacy with favorable safety profile. Moreover, management of transfusion-related iron overload is essential for comprehensive care.

Conclusion

This case demonstrates rituximab as an effective and safe treatment option for ESA-induced PRCA, particularly valuable in elderly dialysis patients with multiple comorbidities who may not tolerate

conventional immunosuppressive therapy.

病例報告 114_C106

高惡性度母細胞樣 B 細胞淋巴瘤以急性肝衰竭表現

High grade blastoid B-cell lymphoma presented with acute liver failure

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Introduction

High-grade blastoid B-cell lymphoma represents a rare and highly aggressive subset of high-grade B-cell lymphomas, characterized by lymphoblast-like cellular morphology. Advanced-stage disease presented nodal and extranodal involvement. This patient presented with extranodal involvement of the liver. The disease course is rapid and aggressive.

Case Report

A 74-year-old male with a history of hypertension and coronary artery disease presented with dark-colored urine, abdominal distension, poor appetite, generalized malaise, and tarry stool. Initial evaluation revealed leukocytosis (10630/uL), thrombocytopenia (54000/uL), elevated serum BUN (30mg/dL), Cr (1.57mg/dL), AST (264U/L), ALT (157U/L), Bilirubin-T/D (3.1/2.4mg/dL), ALKP (309U/L), GGT (394U/L) and LDH (1015U/L) levels. Endoscopy showed reflux esophagitis and duodenal ulcer bleeding. Abdominal CT showed bilateral adrenal masses. He received antibiotic treatment and supportive care but subsequently developed disorientation, aggravated hyperbilirubinemia (18.5/13.5 mg/dL), hyperammonemia (124 µg/dL), and acute respiratory failure requiring mechanical ventilation. He was transferred to our hospital after stabilization.

At our hospital, bone marrow biopsy revealed atypical cells suspicious for lymphoma. Despite initial stabilization, his condition rapidly deteriorated with lactic acidosis and multiple organ failure including lung, liver and kidney. Given the poor prognosis and rapid progression, the family chose palliative care. Final pathology confirmed high-grade blastoid B-cell lymphoma with positive CD20, BCL-2, CD10, BCL-6, and c-MYC expression, with a Ki-67 index of 95-100%.

Discussion

This case exemplifies the aggressive nature of high-grade blastoid B-cell lymphoma, which typically presents with rapidly progressive disease and widespread extranodal involvement. An elevated serum lactate dehydrogenase (LDH) level, in conjunction with high alkaline phosphatase (ALKP) and gamma-glutamyl transferase (GGT) levels, and the absence of a clear etiology for hepatitis, is highly suggestive of lymphoma with liver involvement. The patient's presentation with liver and bilateral adrenal involvement reflects the characteristic extranodal tropism of this malignancy. The high Ki-67 index (95-100%) and positive c-MYC expression are consistent with the aggressive biological behavior observed clinically.

Diagnostic workup requires bone marrow biopsy with comprehensive immunophenotyping (CD20, BCL2, BCL6, cMYC, Cyclin D1, SOX11, Ki67…etc.) and molecular studies (t(11;14), rearrangements in MYC plus BCL2 and/or BCL6…etc.) to distinguish this entity from other high-grade lymphomas and acute leukemias. A retrospective study revealed that high-grade B-cell lymphoma involved the liver in approximately 12% of cases. An article published in Frontiers in Immunology reported that

the 250-day overall survival rate in high-grade B-cell lymphoma was 19%. Early recognition is crucial, though therapeutic options remain limited given the poor response to conventional treatments.

Conclusion

High-grade blastoid B-cell lymphoma represents an extremely aggressive malignancy with poor prognosis and rapid disease progression. This case demonstrates the characteristic multiorgan involvement and fulminant clinical course. Early diagnosis through comprehensive pathological evaluation is essential, though treatment options remain limited. Family decisions for palliative care in such cases represent reasonable choices given the disease's natural history and poor therapeutic response.

病例報告 114_C107

免疫檢查點抑制劑誘導之糖尿病：三例自體抗體表現相異之病例報告

Immune Checkpoint Inhibitor-Induced Diabetes Mellitus: A Report of Three Cases with Contrasting Autoantibody Profiles

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Introduction

With the growing use of immune checkpoint inhibitors (ICIs), more attention has been drawn to related endocrine issues. Among these, immune checkpoint inhibitor-induced diabetes mellitus (ICI-DM) is a rare but serious complication. It leads to a sudden onset of severe hyperglycemia often complicated by diabetic ketoacidosis (DKA). The presence of islet autoantibodies varies, reflecting different underlying mechanisms. Here, we present three cases of ICI-DM with distinct autoantibody profiles.

Case Report

Case 1

A 75-year-old male with gastric cancer, status post subtotal gastrectomy, chemotherapy, and five cycles of nivolumab, presented with a 4-day history of malaise and poor appetite. Laboratory evaluation revealed glucose 1125 mg/dL, metabolic acidosis, ketonemia and HbA1c was 6.2%. C-peptide was undetectable (<0.01 ng/mL). Glutamic acid decarboxylase (GAD) antibody was negative. He was subsequently started on a basal-bolus insulin regimen.

Case 2

A 47-year-old woman with a history of endometrial cancer, status post surgery, presented with nausea, vomiting, and general malaise. She was enrolled in a clinical trial and had received 17 cycles of pembrolizumab. Her laboratory results were significant for a glucose level of 1549 mg/dL, β-hydroxybutyrate of 2.347 mmol/L, and an HbA1c of 7.4%. C-peptide level (<0.01 ng/mL) was undetectable; GAD antibody was negative. She was subsequently started on a basal-bolus insulin regimen.

Case 3

A 48-year-old male with esophagogastric junction adenocarcinoma after chemotherapy and the first cycle of pembrolizumab, presented with generalized weakness, epigastric pain, and dyspnea. Laboratory findings revealed glucose 573 mg/dL, pH 6.93, HCO3- 4.7 mmol/L, β-hydroxybutyrate 5.0 mmol/L, and HbA1c 7.4%. C-peptide was undetectable (<0.01 ng/mL), and GAD antibody was strongly positive (52.8 U/mL). He was stabilized with insulin therapy and discharged on a basal–bolus regimen.

Discussion

ICIs can trigger immune-related adverse events (irAEs). ICI-DM is a rare but serious complication, predominantly linked to PD-1/PD-L1 inhibitors. It is characterized by a sudden onset of severe hyperglycemia and frequent DKA (45–68%). Due to its rapid progression, HbA1c may be only modestly elevated, whereas C-peptide is typically absent, reflecting near total β-cell loss. Unlike

classic type 1 diabetes, autoantibody positivity in ICI-DM varies widely (0–71%), with anti-GAD most frequently detected. Our three cases—two GAD-negative and one GAD-positive—illustrate this variability and highlight the heterogeneous immunopathogenesis of ICI-DM. Previous studies have indicated that the latency from ICI initiation to the onset of diabetes is shorter in autoantibody-positive patients, suggesting a pre-existing autoimmune predisposition. These findings emphasize the importance of vigilance in patients receiving ICIs, regardless of autoantibody status, to facilitate early recognition and prompt management.

Conclusion

ICI-induced diabetes is a fulminant clinical entity that can occur at any point during therapy. The absence of islet autoantibodies does not exclude the diagnosis. A high index of suspicion for new-onset hyperglycemia or DKA in patients on immunotherapy is essential, as prompt diagnosis and insulin initiation are life-saving.

病例報告 114_C108

接受 Nivolumab 治療之晚期肝細胞癌患者發生免疫檢查點抑制劑肺炎：病例報告

Immune Checkpoint Inhibitor Pneumonitis in a Patient with Advanced Hepatocellular Carcinoma Receiving Nivolumab: Case Report.

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Introduction

Immune checkpoint inhibitors (ICIs) have become a cornerstone of first-line therapy for patients with unresectable hepatocellular carcinoma (HCC). Despite their clinical efficacy, these agents are associated with immune-related adverse events (irAEs), which reflect aberrant immune activation across diverse organ systems. Although most irAEs are mild and manageable without discontinuing ICIs, certain rare manifestations, particularly ICI-related pneumonitis, can be life-threatening, with reported mortality varying according to severity and response to prompt immunosuppressive therapy. Here, we describe a case of ICI-induced pneumonitis in a patient with unresectable HCC, highlighting the diagnostic challenges and management considerations inherent to this potentially fatal complication.

Case Report

A 62-year-old man with unresectable HCC who developed fatal ICI pneumonitis following the 4th cycle of nivolumab treatment. The patient presented with fever, fatigue, somnolence, and dry cough with scant white sputum. Laboratory studies demonstrated acute transaminitis, progressive hyperbilirubinemia and coagulopathy. Comprehensive respiratory pathogen testing returned negative results. Chest CT revealed bilateral ground-glass opacities and patchy alveolar consolidations. Despite intravenous methylprednisolone (2 mg/kg/day) initiation for the high suspicion of nivolumab-induced pneumonitis and hepatitis, the patient's hypoxemia continued to deteriorate, culminating in respiratory failure and death.

Discussion

ICI pneumonitis represents the most severe and potentially fatal immune-related adverse event, with all-grade pneumonitis incidence of 2.92% and high-grade pneumonitis of 1.53%. Clinical presentation includes dyspnea (53%), cough (35%), and fever (12%). Radiological manifestations include ground-glass opacity (96.1%) and consolidation (53.9%). Management follows common terminology criteria for adverse events (CTCAE) grading with prednisolone-equivalent dosing of 1-2 mg/kg/day for grades 2-4 pneumonitis. Steroid-refractory disease necessitates aggressive second-line immunosuppressive therapy including infliximab, IVIG, or mycophenolate mofetil.

Conclusion

Despite its low incidence, ICI pneumonitis presents significant mortality risk, necessitating early recognition and prompt intervention. For steroid-refractory pneumonitis, immediate second-line therapy is crucial for improving patient outcomes and reducing mortality rates.

病例報告 114_C109

人工主動脈瓣病人牙科處置後發生 *Lactococcus garvieae* 菌血症：病例報告

Lactococcus garvieae Bacteremia Following Dental Procedure in a Patient with Prosthetic Aortic Valve: A Case Report

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Introduction

Lactococcus garvieae is a gram-positive, catalase-negative, facultatively anaerobic coccus that was previously classified within the genus *Streptococcus*. This pathogen is well-known to cause infections in warm water fish, particularly rainbow trout. It has also been isolated from raw cow's milk, beef, poultry, goat cheese, and pork. In recent years, a small number of human cases of *L. garvieae* bacteremia have been reported, most of which were associated with infective endocarditis. Here, we presented a case of isolated *L. garvieae* bacteremia.

Case Report

A 59-year-old man with a medical history of aortic stenosis status post aortic valve replacement (May 2022), prosthetic valve endocarditis due to *Streptococcus mitis* complicated by cardiogenic shock requiring extracorporeal membrane oxygenation (February 2024), dyslipidemia, and type 2 diabetes mellitus presented with fever and myalgia for one day. He had recently undergone dental work but no antibiotic prophylaxis was prescribed. On admission, his temperature was 38.8°C and he was tachycardic. Blood cultures yielded Gram-positive cocci in (GPC) chains in all four bottles. Transthoracic echocardiography demonstrated no vegetation or significant valvular dysfunction. Empirical therapy with piperacillin/tazobactam was initially started, then de-escalated to penicillin for GPC, and subsequently changed to ceftriaxone following identification of *L. garvieae* bacteremia by MALDI-TOF. Further surveys for endocarditis were all negative: no vegetation detected on transesophageal echocardiography, no Roth spots on fundoscopic examination, and normal rheumatoid factor. Serial blood cultures turned negative after initiation of therapy. His clinical status improved with recovery of renal function and resolution of leukocytosis. He remained well without complications and was discharged o hospital day 8 with oral amoxicillin to complete the antibiotic course.

Discussion

L. garvieae infection has been reported in a variety of clinical presentations, including infective endocarditis, liver abscess, spondylitis, peritonitis, diverticulitis, urinary tract infections, and most commonly, bacteremia. A recognized route of transmission is the consumption of raw fish. Notably, many reported cases occurred in patients with underlying gastrointestinal conditions such as diverticulosis, gastric ulcer, chronic use of acid-suppresive therapy, or prior gastrointestinal surgery. In our patient, the recent dental procedure without antibiotic prophylaxis may have contributed to the development of bacteremia. Review of previously reported cases suggests that *L. garvieae* isolates are generally susceptible to ampicillin, ceftriaxone, and fluoroquinolones, consistent with the susceptibility profile observed in our patient.

Conclusion

L. garvieae is an emerging pathogen responsible for a range of human infections. Here, we reported a case of isolated *L. garvieae* bacteremia, possibly associated with a recent dental procedure without antibiotic prophylaxis. Further studies are warranted to elucidate the pathogenic mechanisms underlying *L. garvieae* infection.

病例報告 114_C110

瀰漫性類鼻疽伯克氏菌感染合併感染性關節炎與深部靜脈栓塞：病例報告

Disseminated Melioidosis with Septic Arthritis and Deep Vein Thrombosis: A Case Report

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Introduction

Melioidosis, caused by *Burkholderia pseudomallei*, is endemic in Southeast Asia and Northern Australia but remains relatively uncommon in Taiwan. Its clinical spectrum ranges from localized infection to fulminant sepsis, while musculoskeletal involvement is less frequently documented. We report an elderly man with disseminated melioidosis complicated by septic arthritis and venous thrombosis.

Case Report

An 84-year-old man with hypertension and a remote history of colon cancer presented with high fever, poor appetite, and progressive left knee pain. Laboratory studies revealed sepsis with impaired renal function and anemia. Abdominal computed tomography demonstrated sigmoid wall thickening with ileitis, for which empirical cefoperazone/sulbactam followed by flomoxef was administered. Blood cultures subsequently yielded *B. pseudomallei*. Arthrocentesis of the left knee confirmed septic arthritis, with synovial fluid culture also positive for *B. pseudomallei*. Targeted therapy with ceftazidime and doxycycline was initiated. During hospitalization, Doppler sonography revealed extensive thrombosis of the left femoral and popliteal veins, complicating progressive limb swelling. Anticoagulation with apixaban was attempted but discontinued due to bleeding tendency. The patient was also newly diagnosed with diabetes mellitus and developed features suggestive of adrenal insufficiency. His condition initially stabilized under antimicrobial therapy, and he was discharged on oral doxycycline. However, he expired shortly after discharge.

Discussion

Pulmonary disease represents the most common manifestation of melioidosis, whereas septic arthritis occurs in approximately 10% of cases, typically affecting the lower extremities. Thrombotic complications are rarely described but may result from a systemic procoagulant state driven by infection-related inflammation. This case underscores the diagnostic challenges of melioidosis in Taiwan, particularly in southern regions where clusters have been linked to rainfall, typhoon events, and riverbank disturbances. Diabetes mellitus is a major predisposing factor and was newly diagnosed in our patient. A thorough environmental and exposure history in this case—such as residence near swamps or rainwater collection—is essential for differential diagnosis. Standard treatment involves intensive therapy with ceftazidime or carbapenems, followed by prolonged eradication with trimethoprim-sulfamethoxazole or doxycycline.

Conclusion

This case highlights disseminated melioidosis with septic arthritis and deep vein thrombosis in an elderly man with newly diagnosed diabetes. Clinicians in Taiwan should maintain a high index of

suspicion for melioidosis in patients with sepsis and musculoskeletal involvement in endemic areas to facilitate timely diagnosis, targeted therapy, and appropriate long-term management.

病例報告 114_C111

頸動脈破裂症候群行血管內支架治療後發生中風併發症：病例報告

Stroke Complication After Endovascular Stent-Graft Placement for Carotid Blowout Syndrome: A Case presentation

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Introduction

A covered stent is a minimally invasive endovascular technique for carotid blowout syndrome (CBS) from head and neck cancer, providing rapid bleeding control by reconstructing the damaged artery. Although effective for immediate hemostasis, it carries notable risks, including rebleeding, graft infection, stroke, and even mortality.

Case Report

A 55-year-old Taiwanese man with right hypopharyngeal cancer (cT1N2aM0, Stage IVA) treated with surgery, chemotherapy, and radiotherapy in 2020, and synchronous esophageal cancer. On August 7, 2025, he experienced sudden massive hematemesis at home and was brought to Zhushan Show Hospital, where PEA occurred. After 15 minutes of CPR , return of spontaneous circulation was achieved. During intubation, profuse oral bleeding was noted. Neck CT at our emergency department showed extensive necrosis of the right parapharyngeal, hypopharyngeal, and visceral spaces with bony exposure of the thyroid cartilage and pseudoaneurysm formation along the medial wall of the right common carotid artery, consistent with carotid blowout syndrome. ENT found no active bleeder on fiberscope and surgical hemostasis was not feasible. The patient was intubated for acute hypoxic respiratory failure, started on sedation, broad-spectrum antibiotics, and antiepileptic therapy. EEG showed moderate to severe diffuse cortical dysfunction with generalized periodic epileptiform discharges. On hospital day 5, recurrent oral bleeding prompted endovascular management. He underwent transarterial embolization with stent-graft placement at the right ICA/CCA and coil embolization of the right ECA, followed by dual antiplatelet therapy. Immediate hemostasis was achieved. However, subsequent brain MRI revealed acute middle cerebral artery infarction without large-vessel occlusion, with cerebral edema and minimal midline shift, representing a stroke complication after covered stenting for CBS. Mannitol was added and neurology consulted, but serial EEGs consistently showed severe to extremely severe diffuse cortical dysfunction, indicating poor neurological prognosis. At present, the patient remains comatose and ventilator-dependent.

Discussion

This case illustrates that in post-radiation head and neck cancer, CBS can occur with extensive soft-tissue necrosis and carotid pseudoaneurysm many years after treatment. Although covered stent-graft and coil embolization provide rapid hemorrhage control, they carry significant risks of ischemic stroke and poor neurologic outcome, emphasizing the need for careful patient selection, vigilant post-procedural monitoring, and multidisciplinary management to optimize survival and minimize complications.

Conclusion

Post-radiation carotid blowout syndrome carries a poor prognosis, long latency, and high complication rates, especially after re-irradiation. Endovascular stent-grafting is the mainstream therapy and preserves carotid patency but entails significant risks of ischemia and rebleeding. It should be used selectively, particularly for acute CBS or patients with high stroke risk. Location-specific strategies and vigilant early post-procedural monitoring are essential to optimize outcomes and minimize complications.

病例報告 114_C112

肺栓塞併急性肺心症

Pulmonary embolism complicated by acute cor pulmonale: A case report

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Introduction

Pulmonary embolism (PE) is a potentially fatal manifestation of venous thromboembolism. The symptoms are non-specific, making the diagnosis more difficult. Due to its high mortality rate, early diagnosis is crucial. Diagnosis highly relies on clinical evaluation, D-dimer testing and image study. Acute cor pulmonale complicating PE without immediate treatment may cause severe morbidity. Early detection remains a tough barrier in daily practicing.

Case Report

A 46-year-old woman with no significant past medical history, presented with worsening dyspnea on exertion for one week. She had long-standing exertional intolerance since her first childbirth 17 years ago but recently developed orthopnea and was unable to ambulate without dyspnea. Her obstetric history includes G3P2SA1. One month prior, she experienced bilateral leg swelling and was found to have a high ANA titer (1:160, centromere pattern); hydroxychloroquine was initiated. Her symptoms worsened following a febrile respiratory infection recently, leading her to visit our emergency room for help.

Physical examination upon arrival revealed fever, oxygen desaturation, ranging from 85% to 94%, on ambient air, bilateral pitting edema, and jugular vein engorgement. No crackling breathing sounds were heard. Laboratory investigations showed severe microcytic anemia (hemoglobin 5.1 g/dL, MCV 58 fL), elevated NT-proBNP (5075 pg/mL), prolonged PT/APTT, and a high D-dimer. ABG was consistent with hypoxemia. The anemia survey indicated iron-deficiency anemia masked by chronic disease. Computed tomography (CT) of the chest revealed pulmonary thromboembolism over bilateral pulmonary artery. Acute cor pulmonale was highly suspected. Enoxaparin was prescribed immediately with catheter-directed thrombolysis afterwards. Her symptoms improved after the therapy.

Further evaluation, including echocardiogram, antiphospholipid syndrome (APS), tumor markers, paroxysmal nocturnal hemoglobinuria (PNH), systemic sclerosis and deep vein thrombosis, was conducted. Transthoracic echocardiography indicated estimated PASP = 69 mmHg and diastolic D shape of LV with McConnell sign. Other examinations revealed high CA125 (355.2 U/mL) and CA199 (175.8 U/mL) as well as Anti-centromere B antibody. The elevation of CA125 raised the suspicion of gynecological tumors, which was confirmed by vaginal ultrasound. Pelvic CT examination indicated enhanced solid and cystic mass at bilateral adnexal region, favoring ovarian carcinoma. Enoxaparin was later shifted to apixaban before discharge, with referral to the oncology department.

Discussion

Dyspnea is a common presenting symptom in the emergency department and warrants a broad

differential diagnosis. Pulmonary embolism should be considered in patients with unexplained hypoxemia, lower-extremity swelling, or risk factors for venous thromboembolism. This case illustrates the diagnostic challenge when clinical features are confounded by anemia, autoimmune markers, and suspected malignancy. The coexistence of pulmonary embolism, acute cor pulmonale, and a newly diagnosed ovarian carcinoma underscores the need for systematic evaluation, including screening for underlying thrombophilic states and occult cancer in patients presenting with unprovoked thromboembolism.

Conclusion

This case highlights the complexity of diagnosing pulmonary embolism in the presence of overlapping clinical features. Careful history-taking, physical examination, and judicious use of imaging and laboratory studies were critical to establishing the diagnosis. Recognition of underlying malignancy as a contributing factor to thromboembolism informed subsequent management.

病例報告 114_C113

以敗血性休克臨床表現的感染性肝囊腫合併下腔靜脈壓迫：病例報告

Giant Infected Hepatic Cyst Causing IVC Compression Syndrome Mimicking Septic Shock: A Case Report

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Introduction
Hepatic cysts are common asymptomatic lesions. However, it can sometimes have uncommon presentation.

Case Report
A 62-year-old female presented with fever, anorexia, and hypotension. The patient was initially diagnosed as septic shock. Image studies then identified presence of a huge infected hepatic cyst with IVC compression.

Discussion
We presents a case that demostrates a clinical presentation, which is rare but still compatible to its pathophysiology, of a common benign disease.

Conclusion
IVC syndrome secondary to a infected cyst is a rare condition that presents a significant diagnostic challenge, as its symptoms can mimic septic shock.

病例報告 114_C114

出血性膽囊炎-新型口服抗凝血藥物引起之少見併發症病例報告

Hemorrhagic Cholecystitis - An Unusual Bleeding Event Caused by Novel Oral Anticoagulants (NOACs)

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Introduction

Hemorrhagic cholecystitis is uncommon in clinical practice. Etiology of hemorrhagic cholecystitis could arise from antiplatelet/anticoagulant use, comorbidity with bleeding diathesis (e.g. renal failure, cirrhosis), blunt trauma, cystic artery pseudoaneurysm, malignancy. The aim of the report is to demonstrate a case of hemorrhagic cholecystitis after taking NOACs for superior vena cava syndrome and to illustrate the radiologic characteristics of hemorrhagic cholecystitis.

Case Report

A 66-year-old male patient with medical history of hypertension presented with progressive dyspnea on exertion for three months, significant weight loss (82 -> 57 kg) in one year, facial and bilateral lower limb edema. Chest CT scan disclosed highly suspected neo-growth such as lung cancer in right upper lobe with suspicious metastatic nodes in right lower neck with invasion upper mediastinum, invasion/compression SVC (superior vena cava). Right cervical lymph node biopsy proved poorly differentiated carcinoma with focal neuroendocrine differentiation.

Apixaban was initiated for SVC thrombosis on admission day 7, followed by chemotherapy (etoposide and cisplatin) for lung cancer treatment. The patient developed transient right upper quadrant abdominal pain on admission day 12. Apixaban was withheld due to intermittent hematuria and followed up laboratory test revealed progressive hyperbilirubinemia (T-bilirubin/D-bilirubin: 2.8/2.08 mg/dL) with markedly elevated liver enzymes (ALT/AST: 995/702 U/L).

The liver sonogram showed GB mild wall thickening with diffuse internal echo (hyperechogenic lesion size about 5cm). Further abdominal CT showed distension of gallbladder with wall thickness and non-dependent distributed hyperdense material in cavity, considering hemorrhagic cholecystitis. Surgical department was consulted, and no emergency surgery was indicated due to stable hemodynamics. The patient received 2 units of leukocyte-reduced packed RBCs for anemia and acute blood loss. The patient remained stable hemodynamics without recurrent abdominal pain or jaundice in the following hospital course.

Discussion

A total of 34 case reports of hemorrhagic cholecystitis were published during 1985-2018, including the cases published by M. Tarazi et al. Hemorrhagic cholecystitis carries a high risk of morbidity and mortality. Due to rare incidence, the proposed algorithm for hemorrhagic cholecystitis remains scarce. Patients are usually managed with exploratory laparoscopy, laparotomy, or percutaneous interventions. Hyperdensity in non-dependent portions suggests active or recent bleeding, as the blood has not yet had sufficient time to layer dependently. Since timely diagnosis of hemorrhagic cholecystitis was made in our case, the patient had already stopped anti-

coagulant use and received antibiotics administration. In patients with asymptomatic to mildly symptomatic hemorrhagic cholecystitis, conservative treatment may be considered if there are reversible causes and no hemodynamic instability.

Conclusion

Clinicians should be aware that abdominal pain with abnormal liver function tests and hyperbilirubinemia in these patients should raise suspicion not only for acute cholangitis and acute cholecystitis, but also hemorrhagic cholecystitis. Timely diagnosis, early withdrawal of anticoagulants, and antibiotic treatment may prevent the need for invasive procedures.

病例報告 114_C115

無結核性腦膜炎之顱內結核菌肉芽腫感染 – 個案報告

Intracranial tuberculoma without meningeal involvement – Case Report

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Introduction

Diseases infected by *Mycobacterium tuberculous* complex (MTB) are epidemic and endemic worldwide. Central nervous system (CNS) infections, most present with tuberculous meningitis. account for 5-10% of tuberculous infections, Clinical manifestations of CNS MTB include fever, headache, conscious disturbance, and focal neurologic deficits. Here we present a rare case of intracranial tuberculoma with tissue and microbiologic proofs but no meningeal involvement.

Case Report

A 77-year-old retired male was transferred from one community hospital to the medical center because of intermittent fever and conscious change for one more month. The medical history included hypertension, diabetes, atrial fibrillation, and chronic kidney disease (CKD) stage 3 for years under control. No contact or cluster history could be traced. He was admitted to one community hospital for one more month without definite diagnosis or finding except multiple-ring shaped lesions ranging from 0.2 to 0.7 cm over cerebrum, pons, and cerebellum noted by contrast-enhanced magnetic resonance imaging (MRI). The differential diagnoses included metastatic malignancy, tuberculoma, cryptococcoma, cerebral toxoplasmosis, and nocardiosis. At triage, relative stable vital signs (B.P.: 142/70 mmHg, P.R.: 70 beats/min, R.R.: 18 breaths/min) and confused consciousness ($E_3M_4V_3$) without focal neurologic deficit or meningeal irritation sign. Navigator-guided craniotomy for tissue specimen examination and external vetricular dranage (EVD) were done at the second day, which gray-colored nodules with central cheese-like materials were taken out. The mycobacterial cold stain of cerebral specimen showed many glial cells with few acid fast bacilli. The CSF test showed unremarkable findings (WBC: 0/mm^3, glucose/protein: 40/79 mg/dl). Both the mycobacterium PCR for MTB for biopsied tissue and CSF showed positive results (10^4-10^5 and 10^1-10^2 CFU/ml, respectively). Antituberculous medications including isoniazid, rifampin, pyrazinamide, and ethambutol and steroids were prescribed at the next day after craniotomy. The pathologic report of biopsied tissue showed caseating granulomatous inflammation with acid-fast bacilli. Six weeks later the mycobacterial cultures for tissue and CSF both showed MTB with susceptibility to the first line of antituberculous medications. After few episodes of aspiration complicated with pneumonia, the patient survived with confused state, and he was discharged from the hospital with scheduled antituberculous medications at home.

Discussion

The diagnosis of intracranial tuberculoma without meningeal involvement has been seldomly confirmed with tissue and microbiological proofs. Physicians should take into consideration with MTB infections especially stary-sky lesions with ring enhancement in crainal images, and early

diagnosis with histologic and microbiologic proofs are essential for appropriate and effective antituberculous treatment.

病例報告 114_C116

嚴重單純皰疹腦膜腦炎 – 個案報告

Severe herpes simplex meningoencephalitis – Case Report

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Introduction

Disease caused by *Herpesviridae* are occasionally seen in human lifetime, with clinical manifestations including mucocutaneous inflammation, lymphadenitis, hepatitis, and central nervous system (CNS) infection. Herpes simplex meningoencephalitis (HSME) is an acute or subacute illness associated with focal or global cerebral dysfunction caused by herpes simplex viruses (HSV I or II).

Case Report

A 62-year-old English teacher with Sjogren syndrome under hydroxychloroquine medication was sent to the Emergency Department (ED) because of intermittent fever for 5 days and subsequent altered mental status, visual hallucination, vomiting, and chest pain. No contact or cluster history could be traced. At triage, increase in blood pressure (160/96 mmHg), tarchycardia (119 beats/min), tachypnea (20 breaths/min), high fever (B.T.: 39.5 ^{0}C), and alert conscious state ($E_4M_6V_5$) were noted. An episode of clonic seizure with both eyes deviating to left side occurred during stay at ED. The CT scan of brain showed hypodense change of right temporal lobe, and lumbar puncture was done under the suspicion of CNS infection. The initial pressure was 150 mmH_2O, and the CSF analysis showed pleocytosis with lymphocyte predominance (WBC: 101 /mm^3, N/L:4%/86%) and mild increase in CSF protein (73.6 mg/dl) and normal CSF glucose (75 mg/dl). India ink, acid fast stain, and latex test for cryptococcal antigen all showed negative results. Multiplex PCR (BIOFIRE® FILMARRAY® Meningitis/Encephalitis Panal) displayed positive for HSV-1. The both /protein: 40/79 mg/dl). Intravenous acyclovir (10 mg/kg every 8 hours) with corticosteroids were prescribed immediately. Because of recurrent episodes of seizure attacks, intubation with endotracheal tube and ventilator support were done for for airway protection, and anticonvulsant with parenteral levetiracetam was used. Her consciousness returned to alert state after 48-hour treatment, and no more seizure attack developed. Successful weaning from ventilator support and extubation were accomplished at the 5th day at the intensive care unit (ICU). The followed magnetic resonance imaging (MRI) of brain demonstrated leptomeningeal enhancement over bilateral frontal base and right temporal lobe, which the findings were compatible with HSME. After 21-day of antiviral medication and corticosteroid with slow tapped dosing, she regained her health almost except mild degree of fatigue. She was discharged at the 25th day of hospitalizaiton with scheduled anticonvulsant medication at home.

Discussion

HSME exhibits with nonspecific manifestions and has significant morbidity and mortality if delayed diagnosis. Early recognition of HSME with nucleotide amplification methods, especially with commercial test kits, will help physicians reach the correct diagnosis early and treat with

appropriate antiviral medications.

病例報告 114_C117

Kartagener 症候群合併心肺衰竭之病例報告

Kartagener Syndrome complicated with Respiratory and Heart Failure: A Case Report

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Introduction

Kartagener syndrome, a rare autosomal recessive disorder within the spectrum of primary ciliary dyskinesia, is characterized by the clinical triad of situs inversus, chronic sinusitis, and bronchiectasis. Defective ciliary motility impairs mucociliary clearance, predisposing patients to chronic airway infection, progressive respiratory failure, and reduced quality of life. Although recurrent respiratory tract infections are typical, the development of pulmonary hypertension and subsequent right heart failure represents an uncommon but serious complication.

Case Report

A 44-year-old man with known Kartagener syndrome, bilateral bronchiectasis, and chronic hepatitis B carrier, suffered from repeated episodes of pseudomonas pneumonia, Clarithromycin had been prescribed for one year due to mycobacterium fortuitum pneumonia. He received intravenous ceftazidime and inhaled gentamicin with only temporary improvement during his previous admission.

He presented again with worsening dyspnea on exertion, unintentional weight loss, general weakness, peripheral edema, and copious purulent sputum two weeks after discharge. Bilateral crackles and wheezing breathing, jugular venous engorgement and bilateral leg pitting edema were noted by physical examination. Chest X-ray demonstrated situs inversus, severe bronchiectasis and lung destruction especially left lung area. Echocardiography demonstrated right ventricular dilatation, a D-shaped left ventricle, and markedly elevated pulmonary artery systolic pressure, consistent with pulmonary hypertension and right heart failure. Hypercapnic respiratory failure ensued, requiring non-invasive ventilatory support for both respiratory and heart failure. Sputum culture yielded mycobacterium fortuitum again. After broad-spectrum antibiotics for nosocomial pathogen and mycobacterium fortuitum, and medications for heart failure, general condition was improving, but still non-invasive ventilator dependent at night. He is currently participating in a structured pulmonary rehabilitation program.

Discussion

This case illustrates the progressive nature of Kartagener syndrome. Chronic bronchiectasis promotes recurrent infections, particularly with resistant pathogens such as Pseudomonas aeruginosa, which further accelerate lung destruction. The combination of recurrent pneumonia, persistent hypoxemia, and chronic inflammation contributes to pulmonary vascular remodeling and subsequent pulmonary hypertension. Right heart failure represents a late-stage complication associated with poor prognosis.

Previous reports had demonstrated that patients with Kartagener syndrome may develop pulmonary hypertension, which was associated with significant morbidity and potentially

worsened prognosis. When pulmonary hypertension develops, functional capacity and exercise tolerance decline rapidly. Management focused on airway clearance techniques, aggressive infection control, and supportive strategies to optimize cardiopulmonary function. Long-term macrolide therapy, inhaled antibiotics, and hypertonic saline inhalation had been shown to reduce exacerbations and improve quality of life. Furthermore, careful monitoring with echocardiography and cardiopulmonary testing was essential for early detection of right ventricular dysfunction.

For patients who progress despite maximal medical therapy, lung transplantation remained a viable option. Previous studies suggested that survival outcomes after transplantation in Kartagener syndrome were comparable to those in advanced lung diseases without situs inversus, although technical challenges may complicate the surgical. Early referral and multidisciplinary evaluation are therefore crucial to achieve the best possible prognosis.

Conclusion

This case emphasized the importance of aggressive pulmonary management, pathogen detection and treatment, and multidisciplinary management in patients with Kartagener syndrome to improve both quality of life and survival outcomes.

病例報告 114_C118

以急性譫妄表現之泌尿道感染併醫源性錐體外症候群導致之急性功能下降

Acute functional decline due to urinary tract infection presenting with acute delirium complicated with iatrogenic extrapyramidal syndrome

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Introduction

Delirium is a common acute health issue among elderly patients. Although delirium itself does not pose an immediate life-threatening danger, its occurrence often signals the deterioration of the patient and may be the first manifestation after an acute illness. We report an 82-year-old woman presenting with delirium due to severe urinary tract infection. Comprehensive geriatric assessment addressed physical, psychological, and social issues, with interdisciplinary support for functional decline. She developed temporary extrapyramidal symptoms from medication, later improved. After discharge, home-based post-acute care helped her maintain independence. No major rehospitalization occurred within one year.

Case Report

An 82-year-old female with stage 3 chronic kidney disease, previous fracture, and insomnia, was regularly followed at our outpatient clinic. Initially, she suffered from a fall with right knee laceration, managed conservatively. One months later, consciousness change with difficulty maintaining attention, nocturnal agitation, and talking to herself was noted at home and then brought to our emergency room. On arrival, she was drowsy with hypotension (75/48 mmHg), hypoxemia (SpO_2 93%). Septic shock due to urinary tract infection with acute delirium was impressed. She was admitted to our geriatric ward after hemodynamics stabilized for comprehensive geriatric assessment (CGA). CGA identified high fall risk, polypharmacy, and poor nutritional intake. For her delirium status, we managed with quetiapine, but later the patient experienced extrapyramidal symptoms(EPS), we prescribed diphenhydramine and EPS subsided gradually afterwards. Under medical therapy with rehabilitation and nutritional support, her infection resolved and consciousness improved, though functional dependence persisted. She was discharged with home-based post-acute care and remained free from major complications at follow-up.

Discussion

Delirium, often regarded as acute brain failure, is highly prevalent among hospitalized older adults, some studies even indicated that up to 50% of hospitalized patients are at risk of developing delirium. Delirium results from additive effects of predisposing (e.g., age, dementia, depression, malnutrition, chronic heart/kidney disease) and precipitating factors (e.g., infection, organ dysfunction, dehydration, hypotension, or medications such as benzodiazepines, opioids, anticholinergics, steroids, NSAIDs, polypharmacy). Although not directly fatal, delirium predicts poor prognosis. CGA helps identify risks, and can reduce delirium incidence by 24%, which can

help us deliver better quality of medical care and support.

Extrapyramidal symptoms (EPS) are drug-induced movement disorders linked to dopamine-receptor blocking agents. Manifestations include dystonia, parkinsonism, tardive dyskinesia, and akathisia. First-generation antipsychotics and antiemetics post a high risk of EPS. In this case, concurrent use of domperidone with quetiapine and mirtazapine likely contributed to EPS. Acute management includes airway stabilization with symptomatic relief with diphenhydramine or benztropine, and adjustment or substitution of causative medications.

Conclusion

We report an atypical case of urinary tract infection initially presenting with acute delirium, and developed extrapyramidal syndrome during symptomatic control of delirium. Delirium signals acute illness and poor prognosis of this case. For delirium status, while non-drug care is first-line, severe agitation may require cautious drug use due to EPS risk. We expect that this case may alarm us to pay more attention to delirious patient, and to utilize CGA to provide better medical care including delirium prevention.

病例報告 114_C119

一位高齡結腸癌併轉移性胃出口阻塞與癌症惡病質患者經十二指腸支架術治療

A Case report of an aged patient with colon cancer with metastatic gastric outlet obstruction and cancer cachexia post duodenal stenting

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Introduction

Cancer treatment in the elderly involves complex medical and ethical considerations. Fear of adverse effects often leads to refusal of aggressive therapy, yet disease progression can cause severe symptoms. We report a 92-year-old woman with metastatic colorectal cancer and gastric outlet obstruction, where treatment decisions weighed age, frailty, life expectancy, and family support. The case underscores balancing invasiveness and comfort in choosing endoscopic stenting over surgery for optimal palliative care.

Case Report

A 92-year-old woman with hypertension, hyperlipidemia, and prior hernia repair was diagnosed in 2020 with a 5 × 4 cm transverse colon adenocarcinoma. She underwent laparoscopic colectomy (grade II adenocarcinoma) followed by adjuvant mFOLFOX6, later switched to oral Tegafur, which was discontinued due to intolerance. In 2023, follow-up imaging revealed recurrence with metastases to the pancreas, peritoneum, lymph nodes, and duodenum. The patient declined further anticancer therapy and entered palliative care.

By October 2024, she presented with anorexia and vomiting. Endoscopy confirmed duodenal metastasis causing gastric outlet obstruction, and CT revealed disease progression with new lung, liver, and ureteral lesions. A palliative duodenal stent was placed after multidisciplinary discussion, while ureteral obstruction was managed conservatively. She developed and recovered from a urinary tract infection with bacteremia during hospitalization.

Post-stent, symptoms improved, allowing soft oral intake and cessation of parenteral nutrition. Appetite remained limited despite megestrol acetate. She was discharged in December 2024 under home hospice. This case underscores the importance of individualized, symptom-focused management and shared decision-making in very elderly patients with advanced metastatic cancer.

Discussion

Colorectal cancer usually spreads to the liver, lungs, or peritoneum, while pancreatic or duodenal metastases are rare and may cause gastric outlet or bile duct obstruction with symptoms such as vomiting, jaundice, and weight loss.

Malignant gastric outlet obstruction (MGOO) requires individualized care. Surgical gastrojejunostomy offers durable relief but carries significant risks in frail patients. Duodenal stenting provides a less invasive option with quicker recovery but shorter patency, while endoscopic ultrasound-guided gastroenterostomy (EUS-GE) offers a promising balance of efficacy and safety. [2-4]

Cancer-related malnutrition and cachexia worsen outcomes; management includes nutritional support and appetite stimulants like megestrol acetate, with new therapies such as anti-GDF-15 antibodies showing potential benefit. [5,6]

Conclusion

This case describes a frail nonagenarian with terminal cancer and malignant gastric outlet obstruction. Duodenal stenting was selected for palliation, successfully relieving symptoms and allowing home hospice care. Despite procedural risks, it was favored over surgery due to the patient's frailty and family preferences. The case highlights how minimally invasive palliation can ease suffering and preserve dignity in end-of-life care.

病例報告 114_C120

髓樣管型腎病引起的非少尿急性腎損傷：非分泌型多發性骨髓瘤的診斷挑戰、預後及兩年追蹤的個案報告

Cast Nephropathy-Induced Nonoliguric Acute Kidney Injury: Diagnostic Challenges, Outcomes, and 2-Years Follow-Up in a Case of Non-Secretory Multiple Myeloma

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Introduction

Acute kidney injury (AKI) is associated with progression to chronic kidney disease (CKD), end-stage kidney disease (ESKD), and elevated mortality. We report a case of unexplained renal failure in which renal biopsy revealed cast nephropathy, leading to the diagnosis of non-secretory multiple myeloma (NSMM)

Case Report

A 76-year-old male presented with progressive fatigue and poor appetite for one month. His medical history included hypertension. He denied reduced urine output. On admission, vital signs and physical examination were unremarkable.

Laboratory evaluations showed acute anemia and a decline of estimated glomerular filtration rate (eGFR) from 48.4 ml/min/1.73m^2 to 4.52 ml/min/1.73m^2. The urine protein creatinine ratio (UPCR) was 324.4 mg/gm. Autoimmune, infectious, and cryoglobulin workups were unremarkable. No M protein was detected on serum protein electrophoresis (SPEP). The serum free light chain (sFLC) assay disclosed mildly elevated kappa 681 mg/L, normal lambda 48.8 mg/L, and a kappa/lambda ratio of 13.95. Serum immunoglobulins were IgA 422 mg/dL, IgG 104 mg/dL, and IgM 21 mg/dL. The serum immunofixation electrophoresis (sIFE) detected monoclonal IgA restriction and suspect associated with kappa light chain restriction. The urine immunofixation electrophoresis (uIFE) discovered a faint kappa light chain signal, insufficient to confirm clonality. Bilateral renal parenchymal disease with mild atrophy was noted on renal sonography. The renal biopsy was performed for unexplained renal failure.

Histopathology revealed 23 glomeruli without globally sclerotic. Lightly eosinophilic fractured casts were observed in some tubules, weakly to negatively stained by periodic acid–Schiff (PAS). Immunofluorescence showed kappa light chain positivity in tubular casts. Electron microscopy showed intact glomerular architecture with no cast-like material in tubules. These findings were consistent with cast nephropathy.

Bone marrow biopsy confirmed plasma cell dyscrasia, establishing a diagnosis of NSMM. Hemodialysis initiated one month later. He received induction therapy with a triplet regimen of bortezomib, lenalidomide, and dexamethasone (VRd) for six months, followed by lenalidomide and dexamethasone (Rd) maintenance for one year. At the latest follow-up, the patient remained dialysis-dependent without evidence of myeloma progression.

Discussion

This case highlights the diagnostic challenge posed by NSMM, especially when screening for monoclonal gammopathy is inconclusive, and rare variant accounting for 1–5% of multiple myeloma cases.

Conclusion

In Taiwan, unexplained renal failure accounts for about 16.9% of renal biopsy indications. As biopsy is invasive, a comprehensive assessment should be completed beforehand. Given that AKI carries substantial risks of progression and mortality, prompt and accurate identification of the cause is important.

病例報告 114_C121

急性心肌梗塞與致命性心律不整的非典型嗜鉻細胞瘤病例

Pheochromocytoma Presenting as ST-Elevation Myocardial Infarction and Malignant Ventricular Arrhythmia: A Case Report

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Case Report

A 53-year-old female with a medical history of hypertension, type 2 diabetes mellitus, and dyslipidemia presented with a 2-day history of chest pain and palpitations. The electrocardiogram revealed ST-segment elevation in leads V1-V2, and serum troponin I was elevated to 1.31 ng/mL. Under the impression of ST-elevation myocardial infarction (STEMI), the patient underwent emergent percutaneous coronary intervention (PCI), which identified an 80% stenosis in the left anterior descending (LAD) artery. A drug-eluting stent (DES) was successfully deployed.

During PCI and the subsequent hospitalization, the patient exhibited extreme blood pressure fluctuations, ranging from 342/142 mmHg to 43/31 mmHg, accompanied by intermittent palpitations and nausea. She also developed sustained ventricular tachycardia (VT) during episodes of hypertensive surges. Given the combination of paroxysmal hypertension, palpitations, and arrhythmias, evaluation for secondary causes of hypertension was initiated. Urinary vanillylmandelic acid (VMA) was markedly elevated (194.74 mg/day), and abdominal computed tomography revealed a 7.2 cm hypervascular mass in the right adrenal gland, highly suggestive of pheochromocytoma.

A multidisciplinary team carefully weighed the risks of early non-cardiac surgery following recent myocardial infarction against the ongoing danger posed by catecholamine excess. Preoperative blood pressure was well controlled with alpha-adrenergic blockade using doxazosin, resulting in a marked reduction in blood pressure fluctuations. The patient subsequently underwent surgical resection of the adrenal mass, and histopathological examination confirmed the diagnosis of pheochromocytoma. Postoperative follow-up is planned to monitor recurrence and long-term cardiovascular outcomes.

Discussion

Pheochromocytoma is a rare catecholamine-secreting tumor, often characterized by varied and non-specific symptoms. The classic symptom triad includes headache, labile hypertension, and diaphoresis; however, up to 10% of patients may remain normotensive, complicating timely diagnosis. Atypical presentations can involve chest pain, palpitations, dyspepsia, or even neurological deficits.

In the present case, the patient exhibited typical features of myocardial ischemia, including chest pain, ST-segment elevation, and elevated cardiac biomarkers. Emergent PCI confirmed significant LAD stenosis secondary to atherosclerotic disease but did not demonstrate thrombus formation in culprit site. Notably, excess catecholamines can induce coronary vasospasm, microvascular dysfunction, and direct myocardial injury, leading to electrocardiographic changes and troponin

elevation in the absence of obstructive coronary thrombosis.

Furthermore, sustained VT in this patient was linked to catecholamine surges, highlighting the arrhythmogenic potential of pheochromocytoma. Excess catecholamines exert profound effects on myocardial excitability and vascular tone, predisposing patients to malignant arrhythmia. Certain pharmacologic agents may exacerbate this state by either increasing catecholamine levels or sensitizing adrenergic receptors. In this case, the administration of metoclopramide and propranolol—used initially for nausea and post-myocardial infarction management, respectively—may have amplified the risk of tachyarrhythmias by triggering additional catecholamine release.

Pheochromocytoma exhibits a broad spectrum of manifestations and frequently mimics other conditions. This case highlights an atypical presentation of pheochromocytoma manifesting as STEMI and life-threatening tachyarrhythmias. Clinicians should remain vigilant for pheochromocytoma in patients presenting with acute coronary syndrome accompanied by paroxysmal hypertension, hyperadrenergic symptoms, or malignant arrhythmia. Prompt biochemical evaluation and imaging are essential for early diagnosis, initiation of α-adrenergic blockades, and timely surgical resection, all of which are critical to optimizing outcomes.

病例報告 114_C122

一個加護病房中常被忽略的診斷：麴菌氣管支氣管炎

A Frequently Overlooked Diagnosis in the ICU: *Aspergillus* Tracheobronchitis

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Introduction

Aspergillus tracheobronchitis (ATB) is an uncommon but severe form of invasive aspergillosis. Because chest radiography (CXR) often shows subtle or absent abnormalities, recognition is frequently delayed, particularly among neutropenic, critically ill or post-influenza patients. Early bronchoscopy is crucial for timely diagnosis in these high-risk groups.

Case Report

We report a 62-year-old woman with newly diagnosed acute promyelocytic leukemia (PML-RARA–positive) and neutropenia who received induction therapy with all-trans-retinoic acid, dexamethasone, and hydroxyurea. Four days after the induction therapy, she developed hypercapnic respiratory failure and septic shock. Initial CXR revealed no definite pulmonary infiltrates, but serum galactomannan was positive (optical density index value of 5.31). Bronchoscopy demonstrated typical tracheobronchitis with black ulcerative lesions in the trachea and bilateral main bronchi. Biopsy confirmed tissue-invasive hyphae, consistent with ATB and invasive pulmonary aspergillosis. Isavuconazole was initiated for antifungal therapy. The FilmArray® pneumonia panel of bronchoalveolar lavage fluid was positive only for influenza A, for which antiviral therapy was initiated. With antifungal therapy, ventilatory support, and renal replacement therapy, her condition gradually improved.

Discussion

ATB can present as ulcerative, pseudomembranous, or obstructive subtypes. In this patient, the ulcerative form manifested as necrotizing eschar-like lesions with direct invasion of the airway mucosa. Notably, initial chest radiographs showed no focal consolidation, highlighting that radiographic imaging alone may not capture early or isolated tracheobronchial involvement. This case highlights the critical diagnostic role of bronchoscopy in high-risk patients with unexplained respiratory failure, especially when invasive fungal disease is suspected.

Beyond neutropenic hosts, invasive aspergillosis is increasingly recognized in post-influenza critically ill patients and is associated with poor outcomes. Influenza infection disrupts epithelial barriers and impairs neutrophil function, thereby creating a permissive environment for *Aspergillus* invasion. Prompt clinical suspicion, early bronchoscopy evaluation, and timely initiation of antifungal therapy are essential to improve outcomes.

Conclusion

This case underscores that ATB may present with minimal or absent radiographic abnormalities on chest radiography, leading to delayed recognition in high-risk patients. Bronchoscopy remains

indispensable for establishing a definitive diagnosis and guiding management. Clinicians should maintain a high index of suspicion for ATB, particularly in immunocompromised or post-influenza ICU patients, to enable early detection and optimize patient outcomes.

病例報告 114_C123

射頻消融治療術後併發膽道損傷之結腸癌肝轉移病患--個案報告與文獻回顧

Biliary Injury Following Radiofrequency Ablation for Liver Metastases of Colon Cancer: A Case Report and Literature Review

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Introduction

Approximately 25–30% of patients diagnosed with colorectal cancer develop liver metastases during the disease course. Radiofrequency ablation (RFA) is frequently used for patients with unresectable liver metastases, providing local tumor control and survival benefits in well-selected cases.

Case Report

We present the case of a 45-year-old woman with metastatic sigmoid colon adenocarcinoma (pT1N1bM1b, stage IVB), involving the liver, lungs and left ovary. She had previously undergone laparoscopic anterior resection, right lobectomy and cholecystectomy. On May 5, 2025, sonography revealed liver metastases in segments 1-4, with the largest lesion measuring 7 cm. She subsequently received palliative RFA and PEI (percutaneous ethanol injection) on May 19.

In late June, five weeks following RFA, she developed worsening abdominal pain. Computed tomography (CT) on June 28 showed calcified metastases (segment 1, left lobe) with left intrahepatic duct dilation. PTCD (percutaneous transhepatic cholangiography and drainage) was performed for hyperbilirubinemia and biliary obstruction, likely RFA-related iatrogenic injury. After stabilization, she received metallic stenting and balloon dilatation of the left intrahepatic and common bile ducts.

Despite these interventions, C-reactive protein (CRP) and bilirubin level remained elevated. Sonography identified lesions suspicious for biloma or abscess in segments 4a–4b. Repeated aspirations provided limited symptomatic relief.

She later developed worsening abdominal distension due to recurrent biloma/abscess causing mass effect and severe biliary tract infection. Despite PTAD (percutaneous transhepatic abscess drainage) revisions and antibiotics escalation, persistent bile leakage, hyperbilirubinemia, and recurrent sepsis episode ensued. Given her deteriorating condition, she was transferred to hospice ward for palliative care.

Discussion

RFA has emerged as a palliative treatment option for hepatobiliary malignancies but may injure adjacent structures, causing bile duct injury, biliary obstruction, biloma, abscess, or rarely, liver failure. The reported mortality and incidence rates of major complications following RFA range from 0.2% to 1.6% and 2.2% to 12%, respectively.

Risk factors comprise large tumor (>3 cm), proximity to central bile ducts, tumors located in hilar/segment 4a or 8, decompensated liver function (Child–Turcotte–Pugh class B or C), prior biliary interventions, use of multiple probes and repeated ablations.

Prevention strategies focus on patient selection, pre-procedural imaging (CT/MRI) to delineate the tumor–duct anatomy and technical modifications. According to previous systematic review, biliary cooling with cold saline or dextrose infusion reduces stricture rates to ~2% in high-risk cases. Application of lower energy, shorter ablation time, staged sessions, and early post-procedural monitoring (imaging, laboratory surveillance) help limit and facilitate prompt detection of complications.

Management involves percutaneous or endoscopic drainage of biloma or abscess, with antibiotics indicated for suspected infection. Endoscopic stenting is preferred in cases of biliary obstruction, while surgical intervention is reserved for refractory cases or major duct injuries.

Conclusion

RFA complications are uncommon but clinically significant, affecting prognosis and quality of life. Careful patient selection, cautious procedural technique and early recognition of warning signs are essential to prevent fatal outcomes. Future studies should focus on refining preventive strategies and optimizing monitoring measures to enhance patient safety.

病例報告 114_C124

以瀰漫性轉移性肺部鈣化為表現之高鈣血症危象：一例原發性副甲狀腺功能亢進病例報告

Primary Hyperparathyroidism Presenting with Hypercalcemic Crisis and Diffuse Metastatic Pulmonary Calcifications: A Case Report

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Introduction

Primary hyperparathyroidism (PHPT) is a common cause of hypercalcemia. Classic manifestations include bone loss, neurocognitive dysfunction, acute kidney injury, nephrolithiasis, and cardiac arrhythmias. Pulmonary involvement is exceedingly rare. In such cases, it may mimic pneumonia or acute respiratory distress syndrome (ARDS), which can easily lead to misdiagnosis.

Case Report

A 40-year-old woman without known prior medical conditions presented to the emergency department with a progressively enlarging bilateral neck mass for 4 months. Associated symptoms included fever, dry cough, constipation, and diffuse musculoskeletal pain involving joints and lower back. Laboratory findings were remarkable for leukocytosis, anemia (hemoglobin 10.7 g/dL), renal impairment (serum creatinine 4.09 mg/dL), elevated C-reactive protein (110.61 mg/dL), and severe hypercalcemia (10.65 mg/dL). Ionized parathyroid hormone (PTH) level was markedly elevated (1067.99 ph/mL). Initial chest radiography revealed tracheal deviation but no definite cardiopulmonary pathology. Non-contrast neck CT (computed tomography) demonstrated a multinodular goiter with the largest nodule located in the left thyroid lobe. Based on the presence of hypercalcemia and elevated iPTH, primary hyperparathyroidism was suggested and later confirmed, as parathyroid MIBI scintigraphy demonstrated uptake attenuation in right superomedial and left superior lobes.

During hospitalization, aggressive intravenous hydration, calcitonin, and bisphosphonates were administered. Furosemide was also given for suspected pulmonary edema as bilateral pulmonary infiltrates gradually developed. Empirical broad-spectrum antibiotics were initiated since superimposed pneumonia could not be excluded. Chest CT subsequently showed bilateral interstitial pneumonitis. Suspecting pneumocystis jirovecii pneumonia, systemic steroid and SMX/TMP were added. However, bronchoscopy and bronchoalveolar lavage analyses were unremarkable. Despite intensive treatment, refractory hypercalcemia, persistent cough, and worsening radiographic infiltrates remained. Emergent subtotal parathyroidectomy (right and left superior parathyroid glands) was performed. Postoperatively, the patient developed hypocalcemia, which was managed with calcium supplementation. Following surgery, rapid improvements in pulmonary infiltrates, respiratory symptoms, and renal function were observed.

Discussion

Pulmonary manifestations of PHPT are rarely reported but may occur in the setting of hypercalcemic crises. In our case, the bilateral pulmonary infiltrates were consistent with diffuse metastatic pulmonary calcifications (MPC). Importantly, the term “metastatic” in MPC is a

historical misnomer–it refers to calcium deposition on normal lung tissue due to metabolic derangements (e.g., hypercalcemia), rather than tumor metastasis. The proposed mechanism involves sustained elevation of PTH leading to persistent hypercalcemia, which promotes calcium salt deposition in lung tissue, possibly facilitated by the relatively alkaline environment of the pulmonary system. Clinically, symptoms are often nonspecific and may mimic pneumonia, ARDS, or cardiogenic pulmonary edema. Consequently, many patients initially receive broad-spectrum antibiotics or diuretics, delaying the correct diagnosis. In some cases, histopathological confirmation from lung biopsy may be required. While supportive managements such as hydration, calcitonin, and bisphosphonates are essential, the definitive treatment for PHPT-induced hypercalcemic crisis remains parathyroidectomy. As demonstrated in this case, both clinical symptoms and radiographic abnormalities can resolve rapidly once hypercalcemia is corrected.

Conclusion

Diffuse metastatic pulmonary calcifications represent a rare but important manifestation of PHPT. This condition should be considered in patients with severe hypercalcemia and unexplained bilateral pulmonary infiltrates refractory to conventional management of ARDS or pulmonary edema. Our case underscores the importance of early recognition and demonstrates the remarkable clinical recovery achievable following surgical intervention.

病例報告 114_C125

橋本氏甲狀腺炎轉化為葛瑞夫茲病的病例報告

Conversion of Hashimoto's Thyroiditis to Graves' Disease: A Case Report

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Introduction

Hashimoto's thyroiditis (HT) and Graves' disease (GD) are distinct autoimmune thyroid disorders that may represent a spectrum of thyroid autoimmunity. HT is a chronic inflammatory process characterized by anti-thyroid peroxidase antibodies, leading to follicular destruction and hypothyroidism. In contrast, GD results from thyroid-stimulating immunoglobulins and TSH receptor–stimulating antibodies, causing excessive thyroid hormone secretion and hyperthyroidism. Although mechanistically different, transitions between these disorders occur. The more common shift is from GD to HT due to glandular failure, while conversion from HT to GD is rare because chronic HT often leads to significant tissue damage. Nevertheless, this uncommon progression has been reported and carries important clinical implications.

Case Report

A 54-year-old woman with a history of Hashimoto's thyroiditis, maintained on levothyroxine since 2002 with an initial anti-TPO antibody level of 90.1 IU/ml. At that time, thyroid-stimulating hormone receptor antibody (TRAb) testing was not performed. She had been lost to follow-up for several years and presented with a two-week history of dyspnea, palpitations, diarrhea, and bilateral leg edema.

At emergency department, laboratory tests showed suppressed TSH (<0.0083 uIU/mL) and elevated free T4 (4.765 ng/dL), consistent with thyrotoxicosis. Shortly after admission, she experienced in-hospital cardiac arrest but was successfully resuscitated and transferred to the intensive care unit for thyroid storm management. Methimazole (20 mg every 6 hours), propranolol (40 mg every 6 hours), and hydrocortisone (100 mg every 8 hours) were initiated.

Further evaluation revealed a markedly elevated TSH receptor antibody (16.43 IU/L) and ultrasonography showing a bilateral thyroid goiter with a heterogeneous appearance, supporting autoimmune thyroid disease.

Despite intensive therapy, her course was complicated by persistent tachyarrhythmia, progressive hepatic dysfunction, and hospital-acquired pneumonia. Endocrinology consultation recommended thyroidectomy after iodine treatment once relative stabilized; however, hemodynamic instability precluded surgery. After family discussion, hospice care was pursued. The patient's condition deteriorated with worsening liver and heart failure, and she ultimately died in the intensive care unit.

Discussion

The conversion from hypothyroidism due to Hashimoto's thyroiditis (HT) to hyperthyroidism from Graves' disease (GD) is rare and not well described in the literature. A key mechanism involves a

shift in TSH receptor–binding antibodies: stimulating antibodies (TSAb) induce hyperthyroidism, whereas blocking antibodies (TBAb) contribute to hypothyroidism. A switch from TBAb to TSAb predominance may explain the observed transition. Another theory suggests partial recovery of thyroid tissue after autoimmune damage, enabling responsiveness to TSAb stimulation.

Clinically, vigilance is required when patients with HT develop features suggestive of hyperthyroidism. Warning signs include unexpected reduction or discontinuation of levothyroxine therapy. In such cases, testing for TSH receptor antibodies alongside TSH and free T4 monitoring can aid early recognition and management of GD.

Conclusion

In summary, this case illustrates the rare conversion of HT to GD, likely driven by dynamic changes in thyroid autoimmunity. Awareness of this possibility is essential to prevent delayed diagnosis. Early antibody testing and close monitoring should be considered, and further studies are needed to elucidate the underlying mechanisms.

病例報告 114_C126

IgG4 相關性疾病 – 從一個多處器官侵犯的案例上檢視早期診斷與偵測復發的重要性

IgG4-related disease - learning the importance of early diagnosis and early recognition of relapse in a case with multiple organs involvement

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Introduction

IgG4-related disease (IgG4-RD) is a disorder characterized by multi-organ fibro-inflammation. Organ damage can develop after the onset, and early diagnosis or recognition of relapse is essential. Here we reported a case whose diagnosis might have been delayed for years, and who experienced progressive organ involvement during relapses following the first remission.

Case Report

A 52-year-old woman presented to our hospital in May, 2025 with respiratory distress and shock. After emergent management, abdomen computed tomography (CT) scan was done and revealed severe pancreatitis, which was attributed to one of her underlying diseases, IgG4-RD.

Reviewing her history, a right submandibular tumor was excised by an otolaryngologist to exclude malignancy in 2012, and pathological examination revealed only lymphocytic infiltration. Six years later, a right breast tumor was noticed during the routine breast examination. Again, there was no evidence of malignancy on pathology. Lymphoplasmacytic infiltration and storiform fibrosis were found, which are classic features of IgG4-RD. High IgG4/IgG ratio (>40%) was confirmed on histological staining, and high serum IgG4 level (1000mg/dL) was disclosed. Furthermore, retroperitoneal fibrosis (RPF) leading to right hydronephrosis was found on CT scan. Treatment with Prednisolone was started, and the improvement was obvious initially; however, the IgG4 value rose again while Prednisolone was tapered. The patient was once lost to follow-up because of reluctance to resume medications, but was soon hospitalized for acute kidney injury (AKI) caused by exacerbated post-renal obstruction related to RPF. The renal function was improved after the obstruction relieved by percutaneous nephrostomy (PCN) and double-J stents. For IgG4-RD, the response to corticosteroids was less than expected, as the IgG4 value remained elevated at a high level (>1300mg/dL). We once suggested Rituximab to the patient, but she hesitated because the cost is not covered by the health insurance. Eventually, she ended up with a critical status.

Fortunately, the pancreatitis was controlled soon after admission. The patient finally agreed to receive Rituximab after her condition stabilized. An abrupt decline of serum IgG4 level was observed afterwards, and she has been under outpatient care with Mycophenolate mofetil (MMF) as well as low-dose Prednisolone.

Discussion

In our case, IgG4-RD might have emerged when the submandibular gland tumor was found, as isolated submandibular gland enlargement is a common finding of IgG4-RD. Unfortunately, further histological staining was not performed since the diagnosis was not thought of at that time. Additionally, it is reported that prolonged disease duration is a risk factor for relapse. Those

relapses leading to obstructive uropathy and pancreatitis could potentially have been avoided with an earlier diagnosis. Also, the reliability of serum IgG4 level for follow-up should be emphasized. Previous literature suggested that re-elevation of the marker typically heralds the return of active disease, which was proved again in our case.

Conclusion

Early diagnosis and detection of relapse of IgG4-RD are warranted not only because of potential organ damage, but also higher relapse risk with prolonged disease course. Serum IgG4 level is reliable for follow-up, and further workup should be considered once the value rises again.

病例報告 114_C127

醫師應該擔心估計腎絲球過濾率(eGFR)下降嗎？源自於一位服用 Crizotinib 病患的臨床經驗

Should Physician Worried About the eGFR Drop? Perspectives From a Lung Cancer Patient on Crizotinib

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Introduction

Crizotinib is an oral small-molecule tyrosine kinase inhibitor targeting ALK, MET, and ROS1 tyrosine kinases. In addition to common adverse effects such as gastrointestinal symptoms, visual disturbances, elevated transaminases, and loss of appetite, Crizotinib has been reported to inhibit tubular creatinine secretion. This results in elevated serum creatinine levels and a corresponding decline in creatinine-based estimated glomerular filtration rate (eGFR), potentially leading to misinterpretation of renal function. This case report highlights a reversible elevation of serum creatinine and pseudo-renal impairment following reinitiation of crizotinib.

Case Report

A 68-year-old man with a history of hypertension was diagnosed with right upper lobe lung adenocarcinoma, staged as pT2bN2M0 (Stage IIIA), in 2018. He initially received Platinum-based chemotherapy, followed by maintenance therapy with Pemetrexed for 34 cycles. Due to concerns regarding pemetrexed-induced nephrotoxicity and the identification of a ROS1 mutation on re-biopsy, the treatment regimen was changed to Entrectinib on September 2023, and later to Crizotinib on December 2023, due to worsening renal function under Entrectinib. His baseline kidney function was serum creatinine levels ranging from 1.3 to 1.6 mg/dL, corresponding to CKD-EPI creatinine-based eGFR 47-60 mL/min/1.73m2. No proteinuria was detected based on regular urine dipstick tests or albumin-to-creatinine ratio (UACR) assessments.

In March 2025, the patient was diagnosed with rectosigmoid cancer with invasion into the pericolonic space. He underwent a robotic-assisted anterior resection with an end-to-end double-stapled anastomosis on March 26, 2025. During hospitalization (March 23 to April 6, 2025), Crizotinib was unintentionally discontinued. Incidentally, a significant increase in renal function was observed, with a lowest serum creatinine of 0.99 mg/dL and a CKD-EPI creatinine-based eGFR of 83 mL/min/1.73 m^2. Crizotinib was reinitiated after discharge, and his serum creatinine increased to 1.4 mg/dL within two months, remaining stable thereafter. To further evaluate renal function, both serum creatinine and cystatin C were measured simultaneously. The eGFR calculated using the CKD-EPI 2021 creatinine equation was 55 mL/min/1.73 m^2, whereas the cystatin C-based eGFR (CKD-EPI 2012) was 72 mL/min/1.73 m^2. A similar cystatin C-based eGFR and creatinine-based eGFR difference was observed two months later. These findings are consistent with Crizotinib-induced inhibition of tubular creatinine secretion, resulting in a reduced creatinine-based eGFR without evidence of true nephrotoxicity.

Discussion

Creatinine is primarily eliminated by glomerular filtration, with approximately 10–20% secreted by the proximal tubule. Crizotinib interferes with creatinine secretion by inhibiting the activity of organic cation transporter 2 (OCT2) on the basolateral membrane and multidrug and toxin extrusion protein 1 (MATE1) on the apical membrane of renal tubular epithelial cells. This increases serum creatinine levels, especially in the first 2 weeks, without true impairment of glomerular function. The effect is reversible and shows minimal cumulative progression beyond the initial elevation. Therefore, non-creatinine-based methods should be considered when evaluating renal function in patients receiving Crizotinib.

Conclusion

Crizotinib may interfere with tubular creatinine secretion, potentially leading to a misinterpretation of renal function. In patients receiving Crizotinib, non-creatinine-based methods, such as cystatin C-based eGFR measurement, should be considered for more accurate assessment of kidney function.

病例報告 114_C128

接受免疫檢查點抑制劑治療後出現免疫性血小板減少症的肉瘤樣惡性胸膜間皮瘤病人：病例報告

Immune Checkpoint Inhibitor–Related Immune Thrombocytopenia in a Patient with Sarcomatoid Malignant Pleural Mesothelioma: A Case Report

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Introduction

Immune checkpoint inhibitors (ICIs), including combined blockade of programmed cell death protein-1 (PD-1) and cytotoxic T-lymphocyte–associated protein 4 (CTLA-4), have significantly improved survival outcomes in patients with advanced malignancies such as malignant pleural mesothelioma (MPM). However, these therapies can trigger immune-related adverse events (irAEs) affecting various organ systems, including the hematologic system. Immune thrombocytopenia (ITP) is a rare but potentially severe hematologic irAE, characterized by immune-mediated platelet destruction and impaired platelet production. Few cases have been documented in the literature. Herein, we describe a case of sarcomatoid MPM who developed severe thrombocytopenia following treatment with ICIs.

Case Report

A 74-year-old man with a history of atrial fibrillation and moderate tricuspid regurgitation was diagnosed with sarcomatoid malignant pleural mesothelioma (MPM) in July 2024. He subsequently received immune checkpoint inhibitor (ICI) therapy with nivolumab (300 mg every 3 weeks) and ipilimumab (1 mg every 6 weeks) starting in September 2024. In April 2025, he was admitted for progressive dyspnea. Laboratory evaluation revealed severe thrombocytopenia with a platelet count of 17,000/μL. Chest radiography and bedside echocardiography demonstrated a massive left pleural effusion, and therapeutic thoracentesis yielded bloody fluid. Irradiated leukocyte-reduced single-donor platelet transfusion was administered, but thrombocytopenia persisted.

Peripheral blood smear analysis showed anisocytosis of red blood cells with increased reticulocytes, severe thrombocytopenia without platelet clumping, and scanty schistocytes. Immune thrombocytopenia (ITP) was therefore suspected. Additional laboratory work-up, including viral markers (anti-HCV), autoimmune profile (Anti-nuclear antibody, complement C3/C4, rheumatoid factor), and antiphospholipid antibodies, was performed to exclude secondary causes of thrombocytopenia. Hematology and rheumatology consultations supported the diagnosis of ICI-related ITP as an immune-related adverse event (irAE). High-dose corticosteroid therapy with intravenous methylprednisolone (40 mg every 12 hours) was initiated on July 14, 2025. Follow-up on July 23, 2025, revealed an increased platelet count (41,000/μL). The patient was discharged in stable condition and scheduled for outpatient follow-up. ICI therapy was withheld at the time of diagnosis and subsequently discontinued in accordance with guideline recommendations for grade 4 irAE.

Discussion

Immune-related thrombocytopenia is a rare hematologic adverse event of immune checkpoint inhibitors, posing diagnostic challenges due to overlapping causes such as infection, autoimmune disease, and marrow infiltration. In our case, persistent severe thrombocytopenia with hemorrhagic effusion required multidisciplinary consultations with hematology, rheumatology, and oncology to exclude secondary etiologies and confirm ICI-related ITP. The favorable platelet response to high-dose corticosteroid therapy further substantiates the diagnosis of irAE. According to ASCO (American Society of Clinical Oncology) and ESMO (European Society For Medical Oncology) guidelines, hematologic irAEs are rare but potentially severe, warranting high clinical vigilance and timely specialist involvement to ensure optimal management. This case highlights the importance of considering hematologic irAEs in patients presenting with unexplained cytopenia during immunotherapy, particularly when bleeding complications are present. Early recognition and prompt immunosuppressive treatment are crucial to prevent life-threatening outcomes.

病例報告 114_C129

以頑固壞死性遊走性紅斑與血糖控制惡化表現的升糖素瘤

Glucagonoma Presenting with Refractory Necrolytic Migratory Erythema and Poor Glycemic Control in an Elderly Patient

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Introduction

Persistent, treatment-resistant skin lesions often cause significant distress to patients and pose a diagnostic challenge to clinicians. Among the many possible etiologies, paraneoplastic dermatoses represent a rare but important cause that is frequently underrecognized or diagnosed late. Here we present a case of necrolytic migratory erythema (NME) associated with glucagonoma, highlighting the importance of considering paraneoplastic syndromes in patients with refractory dermatologic manifestations and metabolic disturbances.

Case Report

An 84-year-old woman with a history of hypertension, type 2 diabetes mellitus, dementia, paroxysmal atrial fibrillation, deep vein thrombosis, and a pancreatic tail mass presented with recurrent erythematous plaques with brownish discoloration and erosions over the back, perineum and both lower limbs for two years. She had multiple admissions at dermatologic wards, but the skin lesions often relapsed shortly after remission.

In December 2024, she was admitted to our dermatology ward due to worsening skin lesions. Skin biopsy revealed acute and chronic inflammation. During hospitalization, we noted her poorly controlled diabetes despite basal–bolus insulin plus linagliptin, with HbA1c increasing from 7.5% to 9.1% over the past year. At the same time, we reviewed her previous abdominal computed tomography from previous admissions, which reported a nodular lesion (around 2.4 cm) located between the gastric upper body lesser curvature and the pancreatic tail. The mass was not surgically managed nor biopsied previously due to the patient's advanced age. Taken together, these findings raised the suspicion of paraneoplastic dermatoses.

Endoscopic ultrasound-guided fine-needle biopsy (EUS-FNB) confirmed a pancreatic neuroendocrine tumor with positive glucagon staining. A diagnosis of glucagonoma with necrolytic migratory erythema (NME) was made. After discussion with the family, long-acting octreotide (given monthly subcutaneously) was initiated. After four months, glycemic levels improved dramatically (HbA1c reduced to 5.9% on linagliptin alone), and skin lesions resolved without recurrence.

Discussion

Glucagonoma classically presents with the triad of NME, diabetes mellitus, and sometimes with anemia, weight loss or thromboembolic events. Our patient fulfilled all three features, with recurrent NME, progressively worsening hyperglycemia, and persistent anemia (Hb 8–9 g/dL).

The standard diagnostic approach involves confirming elevated fasting plasma glucagon levels (>500–1000 pg/mL) followed by imaging localization and histologic confirmation. However,

plasma glucagon measurement was not commercially available in Taiwan. Given the presence of a pancreatic mass and typical clinical manifestations, we proceeded directly with EUS-FNB after discussion with the family and the gastroenterology team. Histopathology confirmed a glucagon-positive pancreatic neuroendocrine tumor, establishing the diagnosis of glucagonoma.
As the patient was elderly with multiple comorbidities, the family declined surgical resection. We initiated long-acting octreotide monthly as a palliative approach, providing effective control when surgery was not feasible.

Conclusion

In elderly diabetic patients with recurrent erythematous plaques and poor glycemic control, glucagonoma should be considered. Subcutaneous somatostatin analogues offer a safe and effective therapeutic alternative when surgery is not favored.

病例報告 114_C130

腘動脈瘤併慢性完全閉塞之罕見血管內治療成功案例報告

A Rare Case of Popliteal Artery Aneurysm Complicated with Chronic Total Occlusion Treated by Endovascular Therapy

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Introduction

Popliteal artery aneurysm (PAA) is the most common peripheral artery aneurysm, with its primary clinical significance lying in the risk of thromboembolic events, potentially leading to severe limb ischemia and even amputation. Although PAA can occur bilaterally in 50-70% of cases and is associated with abdominal aortic aneurysms in 30-50% of patients, PAA complicated by chronic total occlusion (CTO) is a relatively rare presentation and poses significant therapeutic challenges.

Case Report

We reported a rare case of a 58-year-old male car mechanic, whose occupation involved prolonged squatting, presenting with right popliteal artery aneurysm complicated by CTO, resulting in limb ischemia. He reported several months of right popliteal fossa pain, swelling, and right toe numbness and paresthesia.

Initial examination revealed a fixed, tender popliteal fossa mass with diminished distal pulses. Diagnostic imaging, including musculoskeletal ultrasound, MRI, and angiography, confirmed a 4.57 × 3.14 cm popliteal artery aneurysm with complete thrombotic occlusion and extensive collateral circulation. The endovascular therapy, chosen due to the complex nature of the CTO, proved highly challenging. Initial antegrade attempts failed to cross the densely occluded segment, even with various guidewires and supportive devices. A subsequent retrograde approach successfully entered the aneurysm sac, but the guidewire could not navigate beyond the occluded aneurysm into the superficial femoral artery. A breakthrough was achieved by using the retrograde guidewire as a precise anatomical marker, facilitating a second antegrade attempt which successfully traversed the CTO and entered the below-knee vessels. This was followed by balloon angioplasty and the deployment of two covered stents from the distal superficial femoral artery to the popliteal artery, effectively restoring antegrade blood flow. The patient experienced a remarkable recovery, with complete resolution of symptoms and a reduction in the popliteal fossa mass. At three-month follow-up, he had returned to his physically demanding job.

Discussion

Both open surgical repair (OSR) and endovascular therapy (EVT) are used to treat popliteal artery aneurysm (PAA), but they have distinct trade-offs. OSR, long considered the gold standard, offers superior long-term patency rates (80-90% at five years) and fewer re-interventions, but it is a more invasive procedure with longer hospital stays and a higher risk of complications. In contrast, EVT is a minimally invasive option, ideal for high-risk patients, associated with quicker recovery and shorter hospital stays. Yet, EVT has lower long-term patency (60-75% at five years) and a higher re-intervention rate due to the knee's constant motion, which can compromise the stent. For complex

cases like PAA with chronic total occlusion (CTO), the organized thrombus presents a significant challenge to guidewire navigation during EVT. However, a multi-strategy approach was successfully used in our case, leading to a rapid post-operative recovery for the patient.

Conclusion

Popliteal artery aneurysm complicated by CTO is a rare and extremely complex vascular condition posing a severe threat to limb viability. This case successfully demonstrates that despite the significant technical challenges in EVT of PAA with CTO, revascularization can be effectively achieved through the flexible application of combined antegrade and retrograde catheter techniques.

病例報告 114_C131

總體基因學於結核性腦膜炎的診斷應用

Integrating metagenomic sequencing into diagnostic pathways for tuberculous meningitis

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Introduction

Among the tuberculosis-associated diseases, tuberculous meningitis (TBM) is rare but very lethal, with limited diagnostic opinions. With advances in technology, emerging tools may improve clinicians' diagnostic accuracy. mNGS (Metagenomic next-generation sequencing) is a promising candidate. [1] Here, we presented a case of TBM revealed by mNGS.

Case Report

This 67-year-old man has a past medical history of coronary artery disease, diabetes, hypertension, dyslipidemia, and hyperuricemia. He started with a feverish sensation one week ago. Three days ago, he suffered from bilateral lower limb soreness and slow verbal output. Severe headache struck 2 days later and brought him to our emergency room. Laboratory data, brain CT (Computed Tomography), and brain MRI (Magnetic Resonance Imaging) showed unremarkable findings. Therefore, a lumbar puncture was performed. The result of the CSF (Cerebrospinal Fluid) study was illustrated in Table 1. Initially, Ceftriaxone, Vancomycin, and Acyclovir were given empirically for infectious meningitis, but final bacterial culture and Filmarray revealed no positive result and symptoms persisted under the treatment. Therefore, follow-up lumbar puncture was done on Day 7. (Table 1) According to the feature of monocyte-predominance and persistently high protein and low glucose in CSF, TBM was suspected, even though the negative result of acid-fast stain and GeneXpert. CSF mNGS was done on Day 8 and the final report came out on Day 9, revealing *Mycobacterium tuberculosis* (5 reads). Hence, we started Rifampicin, Isoniazid, Ethambutol, and Pyrazinamide with dexamethasone since Day 9. Headache improved and other neurological symptoms subsided under the treatment. Follow-up CSF profile was compatible with the treatment response on Day 28. (Table 1) He was then discharged with anti-tuberculosis medication. Finally, mycobacterial culture of CSF on Day 7 revealed a positive result on Day 34.

Discussion

mNGS is the sequencing method that can analyze microbial genomes without culture. Through increasing application in clinical diagnosis of the etiology of central nervous system infection, there are more and more studies about the diagnostic accuracy of mNGS in TBM. The meta-analysis by Xiang ZB et al found that mNGS showed comparable sensitivity (62%, 95% CI: 0.46-0.76) and similarly high specificity (99%, 95% CI: 0.94–1.00) compared with culture and Xpert-MTB/RIF. [2] Besides, it also possesses the rapid turnover time (average of 48 hours) and identification of co-infections. [3] In our case, mNGS confirmed our clinical suspicion of TBM one month earlier compared with the culture method.

mNGS still had some limitations and drawbacks in the application of pathogen identification. Among these, one of the important challenges is the data interpretation. [3] mNGS may identify

multiple irrelevant microorganisms. Therefore, clinical presentation and diagnosis were crucial in the mNGS data interpretation. [4] In our case, the mNGS report revealed not only *Mycobacterium tuberculosis* complex but also *Acinetobacter baumannii* (1870 reads). However, based on clinical presentation and course, and treatment response under anti-tuberculosis agent, *Acinetobacter baumannii* is not regarded as the causative pathogen.

Conclusion

mNGS offers significant potential to improve the diagnosis of infectious diseases, particularly when conventional methods fail to identify the pathogen.. Nevertheless, careful clinical judgment remains essential to avoid misinterpretation and overdiagnosis.

病例報告 114_C132

周全性老年評估個案報告：心臟衰竭病人在下肢感染後之心肺復健

Comprehensive Geriatric Assessment Case Report: Cardiopulmonary Rehabilitation for a Heart Failure Patients After Lower Limb Infection

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Introduction

Heart failure is a major cause of disability and hospitalization in the elderly. Functional decline may occur after acute illness such as infection or immobility. Comprehensive geriatric assessment (CGA) helps identify multidimensional problems, while cardiopulmonary rehabilitation provides a structured approach to improve exercise tolerance, lower rehospitalization rates and enhance quality of life.

Case Report

A 73-year-old woman with atrial fibrillation, chronic heart failure and hypertension was admitted due to poor healing of a right calf wound. She developed swelling, purulent discharge, fever and pain which led to acute functional decline. Her Barthel Index decreased from 100 to 80. CGA showed preserved cognition and mood, obesity with BMI of 42.7, moderate dependence in mobility, no delirium and no incontinence. She received antibiotics (piperacillin–tazobactam), optimized heart failure therapy and a structured rehabilitation plan. The program included aerobic exercise, resistance training, balance practice, nutritional support and functional retraining. After one month her exercise tolerance improved and she regained partial independence. She was discharged with a long-term rehabilitation plan and scheduled follow-up.

Discussion

This case demonstrates the usefulness of CGA in elderly patients with heart failure and comorbidities. The assessment revealed risk factors such as obesity, polypharmacy and reduced mobility which guided targeted interventions. Cardiopulmonary rehabilitation benefits both HFrEF and HFpEF patients by improving exercise capacity and daily function. It also reduces hospitalizations and improves survival. Multidisciplinary collaboration with physical therapy, occupational therapy and nutritional counseling was essential for recovery. Early initiation and individualized design ensured safety and effectiveness.

Conclusion

Comprehensive geriatric assessment combined with cardiopulmonary rehabilitation can improve outcomes in elderly patients with heart failure and infection. This case highlights the value of early detection, structured rehabilitation and teamwork in maintaining independence and preventing rehospitalization.

病例報告 114_C133

應用低劑量 Rivaroxaban 之雙途徑抑制於急性冠心症合併冠狀動脈擴張：案例報告

Combating High Thrombus Burden in Acute Coronary Syndrome with Coronary Artery Ectasia Using a Dual Pathway Inhibition with Low-Dose Rivaroxaban: A Case Report

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Introduction

Coronary artery ectasia (CAE) is defined as coronary dilation ≧1.5 times the diameter of an adjacent normal segment. CAE is observed in approximately 0.3% to 8% of patients undergoing coronary angiography and is associated with a high thrombus burden. Dual pathway inhibition (DPI) refers to the therapeutic strategy combining an antiplatelet agent with an anticoagulant. The ATLAS ACS 2–TIMI 51 trial demonstrated a favorable net clinical outcome with the addition of low-dose Rivaroxaban to standard therapy in patients with recent acute coronary syndrome (ACS). However, evidence supporting the acute-phase use of DPI in ACS patients with CAE remains limited.

Case Report

A 40-year-old female patient with chronic microcytic anemia was admitted with dyspnea and chest pain. Laboratory results revealed normal renal function, elevated Troponin-I 2.5 ng/mL (reference <0.015 ng/mL), creatinine kinase-MB 18.8 ng/mL (reference <3.4 ng/mL) and hemoglobin 7.7 g/dL (reference> 11.8 g/dL). Electrocardiography shows T-wave inversion in the inferior leads and ST-segment depression in V2-V5. With the impression of non ST elevation myocardial infraction, plain old balloon angioplasty (POBA) and thrombus aspiration were performed at right coronary artery (RCA) twice, at a two-days interval under the treatment of Aspirin, Ticagrelor, Tirofiban and Enoxaparin. However, much thrombus burden was still observed during the second catheterization, leading to unsuitability for stent implantation.

During hospitalization, the antithrombotic regimen was modified to Aspirin 100mg once daily (QD) plus Rivaroxaban 15mg QD, while heavy menstrual bleeding was noted. Therefore, the regimen was adjusted to aspirin 100 mg QD plus Rivaroxaban 2.5 mg twice daily (BID) at outpatient follow-up. After three months of DPI therapy, repeat catheterization revealed complete resolution of the thrombus in the RCA.

Discussion

Recent guidelines recommend/consider reasonable use of dual-pathway inhibition (Rivaroxaban 2.5 mg BID plus aspirin) for post-ACS patients after 12 months who have persistent high ischemic risk and no high bleeding risk, and for stable coronary artery disease patients at high risk of recurrent ischaemic events with low-to-moderate bleeding risk. However, the feasibility and safety of initiating DPI during the acute phase of ACS in patients with CAE remain uncertain. CAE is characterized by an enlarged coronary diameter, leading to slow flow and disturbed

hemodynamics that favor thrombus formation. Given this mechanism, traditional dual antiplatelet therapy (DAPT) may be insufficient to address the underlying pathophysiology. This case not only highlights the potential role of DPI in ACS patients with CAE, but also supports the safety of DPI with low-dose Rivaroxaban. Further studies are needed to elucidate the therapeutic mechanisms and to reinforce the role of DPI in ACS patients with CAE.

Conclusion

Our case illustrates the challenges of antithrombotic treatment in a patient with chronic anemia and heavy menstrual bleeding who presented with ACS and CAE. To balance antithrombotic efficacy against bleeding risk, the regimen was adjusted to aspirin 100mg QD plus Rivaroxaban 2.5mg BID and successfully eliminate the RCA thrombosis without increasing bleeding events. This case provides clinical insight into the potential application of DPI in ACS patients with CAE during the acute phase.

病例報告 114_C134

一例免疫性血小板低下紫斑症的診斷及治療經驗

A Case of Thrombotic Thrombocytopenic Purpura Caused by ADAMTS-13 deficiency

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Introduction

Thrombotic thrombocytopenic purpura (TTP) is a rare, life-threatening thrombotic microangiopathy(TMA) caused by ADAMTS-13 deficiency, often due to autoantibodies or, less commonly, genetic mutations. It presents thrombocytopenia, microangiopathic hemolytic anemia, and organ ischemia, frequently affecting the brain and kidneys.

Case Report

The patient with history of coronary artery disease post stenting 3 years ago had gross hematuria for a week and was treated as urinary tract infection at clinic, but in vain. He still felt general malaise with progressive disorientation noticed by his wife, so he went to emergency department where lab data showed thrombocytopenia, anemia, elevated lactate dehydrogenase and indirect type hyperbilirubinemia. Because blood smear showed schistocytes, thrombotic microangiopathy was suspected. After we excluded infection, methylprednisolone was given. Thrombocytopenia was still noted, so therapeutic plasma exchange(TPE) was arranged. We also checked ADAMTS-13 activity showing 0%. To exclude malignancy, we arranged abdomen CT, chest CT, colonoscopy, bronchoscopy showing no evidence of malignancy. Unfortunately, progressive conscious disturbance and bilateral lower limb poor endurance while ambulation was noted, and brain MRI showed acute infarctions at right cerebellar hemisphere and splenium of left corpus callosum. With continuous methylprednisolone use, plasma exchange, and hydration, his condition improved with increased platelet level. To sum up, immune TTP was diagnosed with thrombocytopenia, anemia with schistocytes, elevated LDH, indirect type hyperbilirubinemia, no activity of ADAMTS-13 after we excluded infection and malignancy；the patient was treated with TPE and corticosteroids.

Discussion

Immune TTP is driven by autoantibody-mediated severe ADAMTS-13 deficiency, resulting in accumulation of ultra large von Willebrand factor(VWF) multimers. These multimers promote microvascular platelet-rich thrombi, leading to ischemic organ injury, including acute cerebral infarctions and myocardial events. Neurologic symptoms and renal dysfunction are common, and patients with underlying cardiovascular disease are at increased risk for cardiac complications.

Immediate initiation of TPE and corticosteroids is the standard of care, rapidly removing autoantibodies and replenishing ADAMTS-13, which is associated with improvement in survival. Caplacizumab, an anti-VWF nanobody, is FDA-approved for acquired TTP and can be considered in the acute phase to accelerate platelet recovery and reduce early recurrence, but it increases bleeding risk. Unfortunately, Caplacizumab has not yet been introduced in Taiwan. Otherwise, Rituximab is also recommended for refractory cases and for relapse prevention, especially in

patients with low ADAMTS-13 activity after remission.

Conclusion

Rapid diagnosis and immediate initiation of therapy are essential to reduce mortality. Since iTTP is a form of TMA, it is essential to differentiate it from other TMAs like hemolytic uremic syndrome, disseminated intravascular coagulation, infections, and malignancy. Given the high risk of acute mortality associated with iTTP, short-term administration of corticosteroids is acceptable even before infections are ruled out. However, monitoring for any signs of infection is mandatory throughout the treatment course. After remission, monitoring of ADAMTS-13 activity is essential；patients with activity <20% during remission benefit from prophylactic rituximab to reduce relapse risk.

病例報告 114_C135

類風濕因子導致肌鈣蛋白檢驗偽陽性

Rheumatoid factor causes false positive of serum troponin level

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Introduction

Elevation of serum troponin level often indicates myocardial damage. However, several confounders may result in false positive of troponin level. Here, we reported a case of rheumatoid factor causes false elevation troponin levels.

Case Report

A 67-year-old man with history of hypertension, diabetes mellitus, coronary artery disease, benign prostatic hyperplasia, and rheumatoid arthritis, was admitted to urology ward due to his newly diagnosed urinary bladder infiltrating urothelial carcinoma, for radical cystectomy. Unfortunately, sudden onset of chest tightness developed. Non-ST segment elevation myocardial infarction (NSTEMI) was impressed. Coronary angiography revealed triple vessel disease, post drug-eluting stent (DES) in right coronary artery (RCA) and plain old balloon angioplasty (POBA) in left anterior descending coronary artery (LAD) and left circumflex coronary artery (LCX). Yet, followed-up cardiac enzymes remain increasing within three days despite relatively stable condition and no chest discomfort. We performed second coronary angiography, and no in-stent restenosis (ISR) or occlusion noted. Meanwhile, rheumatoid arthritis flare up was suspected according to the patient's clinical symptoms, with positive finding in serum rheumatoid factor. Confirmation with laboratory medicine department disclosed falsely elevation of serum troponin level.

Discussion

The blood sample was analyzed ten times using three different immunoassay analyzers: Beckman, Abbott, and Siemens. Elevated troponin I levels were observed exclusively with the Beckman analyzer, which is the routine platform employed in our institution. In contrast, results from both the Abbott and Siemens analyzers remained consistently negative. To further investigate the discrepancy, the sample was subjected to serial dilutions at 1:2 and 1:5 ratios, which were expected to yield proportionally reduced troponin I concentrations. However, the measured values did not exhibit a linear response to dilution, thereby confirming a false-positive result from the Beckman assay.

Troponins are the most valuable markers of cardiac disease. In addition, multiple factors may bring about troponin elevation, such as arrhythmias, aortic dissection, pulmonary embolism, sepsis, or renal failure. Regardless of cardiac or non-cardiac causes, laboratory error should be mentioned. As reviewing recent articles, several factors may lead to falsely elevation of troponin level, such as rheumatoid factors, heterophile antibodies, alkaline phosphatase (ALP), fibrin levels, or cross-reactions of diagnostic antibodies (anti-cTn, anti-cardiac troponin antibodies). Understanding the causes and mechanisms will help identify the diagnosis of cardiovascular diseases.

Conclusion

Myocardial damage was often considered as the cause of troponin elevation, but not the only. Other clinical manifestation, or even laboratory error should be carefully considered.

病例報告 114_C136

冠狀動脈支架感染併發巨大冠狀動脈瘤

Coronary Stent Infection Complicated with Giant Coronary Artery Aneurysm and Successful Endovascular Treatment

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Introduction

Coronary stent infection (CSI) is a rare but potentially fatal complication of percutaneous coronary intervention, typically caused by Staphylococcus aureus. Clinical signs are often non-specific, such as fever and chest pain. The most severe complications include mycotic aneurysm or pseudoaneurysm formation. We report a case of CSI involving a giant LAD pseudoaneurysm and right ventricular fistula successfully treated by endovascular therapy.

Case Report

A 41-year-old man with recurrent renal cell carcinoma (post-left nephrectomy), type A aortic dissection (status post valve repair and graft surgeries), and end-stage renal disease on dialysis presented on April 23, 2025, with recurrent fever, chest pain, and dyspnea. He had undergone LAD stenting for NSTEMI on January 27, 2025, complicated by in-stent thrombosis, pneumonia, and MRSA bacteremia treated with 8 weeks of antibiotics. PET/CT suggested possible graft infection. On this admission, chest CT revealed a fractured LAD stent with hematoma and pericardial effusion. ECG showed ST-segment elevation in leads I and aVL with elevated troponin I. Contrast CT demonstrated an extravasating saccular aneurysm at the stent site. Due to high surgical risk, intervention was postponed until blood cultures confirmed MRSA clearance. On May 7, 2025, coronary angiography confirmed a giant LAD pseudoaneurysm with RV fistula. Endovascular treatment involved seven stents, eight coils, and 17 balloon dilatations to successfully exclude the aneurysm and close the fistula. The postoperative course was complicated by empyema requiring thoracoscopic decortication, hemopneumothorax with hypovolemic shock, septic arthritis, and an intra-procedural cardiac arrest. The patient regained consciousness and was weaned from ventilatory support. Final diagnoses included CSI with pseudoaneurysm, stent fracture, RV fistula, MRSA bacteremia and empyema.

Discussion

Coronary stent infection remains a diagnostic and therapeutic challenge due to its rarity and non-specific presentation. Mycotic aneurysms or pseudoaneurysms occur in up to 62% of cases, while coronary perforation, fistula, or stent fracture is seen in about 41%. The “two-hit” hypothesis—vascular injury followed by infection—explains aneurysm formation. Drug-eluting stents may increase risk due to delayed endothelialization. Blood cultures are essential for identifying pathogens. Imaging tools like coronary angiography and PET/CT are crucial for diagnosis and monitoring. No standardized treatment guidelines exist. While surgical removal with CABG remains standard, both surgery and antibiotics carry high mortality. Endovascular therapy has

emerged as an alternative, especially for high-risk patients.

Conclusion

Coronary stent infection complicated by giant mycotic aneurysm and cardio-cameral fistula is an extremely rare but fatal condition. We report a case of MRSA-related LAD pseudoaneurysm with right ventricular fistula successfully treated by endovascular therapy after infection eradication, highlighting its role as a feasible alternative to surgery in high-risk patients.

病例報告 114_C137

未分化惡性肺動脈血管肉瘤：病例報告

Undifferentiated Pulmonary Artery Angiosarcoma – A Case Report

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Introduction

Primary cardiac sarcomas are extremely rare malignancies, often presenting with nonspecific symptoms that mimic more common cardiopulmonary conditions. Among them, pulmonary artery sarcomas represent a particularly aggressive subset, frequently misdiagnosed as pulmonary thromboembolism and typically identified only at an advanced stage.

Case Report

The 64-year-old woman with a history of hypertension presented with progressive exertion dyspnea for about 3 weeks associated with indistinct chest pain, poor appetite and weight loss. Physical examination revealed a grade III pan-systolic murmur at left sternal border. Electrocardiogram (ECG) showed non-specific ST-T changes while chest X-ray revealed a nodular lesion in the right upper lung. Echocardiography demonstrated a soft tissue mass with surrounding turbulent flow within the main pulmonary artery (MPA) associated with severe pulmonary hypertension and moderate tricuspid regurgitation. The patient had adequate left ventricular (LV) systolic performance and right ventricular (RV) systolic function. Chest computed tomography angiography (CTA) disclosed a pulmonary artery trunk mass with bilateral pulmonary nodules , raising the suspicion of angiosarcoma.

The patient was thus referred to cardiac surgeon for surgical intervention. Surgical exploration showed a fixed hard tumor extending from the right pulmonary artery to the right ventricular outflow tract (RVOT), involving pulmonary valve and RVOT muscle. The pulmonary valve, pulmonary artery, and part of the RVOT were excised. Histopathology demonstrated undifferentiated pleomorphic sarcoma infiltrating the arterial wall and surrounding tissue. Immunohistochemistry showed focal and moderate positivity for MDM2, faintly positive for cytokeratin (CK), but negative for SMA, SOX-10, CD31, and ERG. The diagnosis of pulmonary trunk sarcoma with lung metastasis (stage IV) was established and the patient was referred for further chemotherapy and immunotherapy.

Discussion

Pulmonary artery angiosarcoma is a rare and aggressive malignant vascular tumor originating from the endothelial cells of the pulmonary artery with fewer than 30 cases reported in the literature. Its nonspecific presentation often mimics pulmonary thromboembolism, complicating diagnosis. Imaging modalities such as CTA and Positron Emission Tomography/Computed Tomography (PET/CT) can aid in distinguishing sarcomas from thrombi. On chest CTA, pulmonary artery angiosarcoma appears as a low-attenuation filling defect with heterogeneous enhancement and possible extravascular spread, often indistinguishable from thromboembolic disease. PET/CT can increase diagnostic certainty by demonstrating intense FDG uptake and heterogeneous

enhancement, distinguishing them from bland thrombi. Histopathology and immunohistochemistry remain essential for definitive diagnosis, showing malignant endothelial proliferation and immunohistochemical positivity for markers such as CD31, CD34, and factor VIII-related antigen. Prognosis is poor due to rapid progression and early metastasis.

Conclusion

This case underscores the diagnostic challenges and dismal prognosis of pulmonary artery sarcoma. Early recognition and multidisciplinary management are crucial, though outcomes remain limited.

病例報告 114_C138

房室節迴旋性心搏過速伴房室 2 比 1 傳導比率：病例報告

Atrioventricular Nodal Reentrant Tachycardia with 2 to 1 Atrioventricular Ratio: A Case Report

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Introduction

Paroxysmal supraventricular tachycardia (PSVT) is a frequent arrhythmia. More than 60% of PSVT cases are due to atrioventricular nodal reentry tachycardia (AVNRT). The most common form is slow-fast AVNRT with a 1:1 atrioventricular (AV) ratio. Here, we present a case of AVNRT with 2:1 AV conduction and its distinctive electrocardiographic (ECG) features.

Case Report

An 11-year-old boy without medical history or family predisposition experienced recurrent palpitations for about a year. Each episode lasted about an hour, had no clear triggers, and terminated spontaneously. He presented to the emergency department with incessant palpitations. His ECG showed a regular ventricular rate of 189 bpm with visible pseudo S waves. An electrophysiology (EP) study was recommended.

In EP lab, his baseline sinus cycle length was 640 ms. Retrograde conduction occurred via the fast AV nodal pathway, whereas antegrade conduction occurred through dual pathways. Tachyarrhythmia was induced by programmed atrial pacing with a tachycardia cycle length of 290 ms, consistent with slow-fast AVNRT.

During the study, several non-sustained supraventricular tachycardia (SVT) episodes with 2:1 AV conduction block were observed. The atrial (A-A) interval measured 276 ms, and the ventricular (V-V) interval was 570 ms. Atrial signals without immediate ventricular conduction produced negative deflections after the T wave on surface ECG. An additional P wave was visible after T waves, creating the characteristic pattern described as “the kiss of the girl from Ipanema.” The tachycardia spontaneously reverted to 1:1 conduction. The occurrence of 2:1 conduction indicated that the ventricles were not part of the reentrant circuit, making atrioventricular reciprocating tachycardia (AVRT) unlikely. The tachycardia was initiated by programmed atrial pacing, and an AH jump preceded onset. Ventricular overdrive pacing showed a “V-A-V” response, with a post-pacing interval minus tachycardia cycle length of 133 ms. These findings made atrial tachycardia and AVRT less favored. The final diagnosis was slow-fast AVNRT with intermittent 2:1 conduction. Nine cryoablation lesions were applied to the low Koch’s triangle for slow pathway modification.

Discussion

AVNRT with 2 to 1 conduction is an uncommon presentation. The reentrant circuit within the AV node-maintained tachycardia, but only every second atrial activation is accompanied with ventricular activation. The differential diagnosis includes atrial tachycardia with 2:1 AV block, atrial flutter with 2:1 conduction, and junctional tachycardia. Electrophysiology studies reveal that the site of block may shift between supra or infra-Hisian regions, based on differential refractoriness and the involvement of upper or lower common pathways within the AV node.

AVNRT with 2:1 conduction is characterized by its distinct ECG pattern “The kiss of the girl from Ipanema”. It is mainly associated with slow-fast AVNRT due to the timing of retrograde atrial activation after the second run of reentry. The retrograde P wave follows T wave and resembles a kiss.

Conclusion

In summary, this case highlights an uncommon presentation of AVNRT with intermittent 2:1 AV conduction and its characteristic ECG pattern known as “the kiss of the girl from Ipanema” with successful slow pathway modification done via cryoablation.

病例報告 114_C139

由 HACEK 菌群所引起的感染性心內膜炎

A case of infective endocarditis caused by HACEK group organisms

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Introduction

Infective endocarditis caused by HACEK(*Haemophilus species, Aggregatibacter actinomycetemcomitans, Cardiobacterium hominis, Eikenella corrodens, Kingella kingae*) group organisms is rare but clinically significant due to its insidious onset and potential for embolic complications. We report a case of a 55-year-old male with *Aggregatibacter actinomycetemcomitans* bacteremia, complicated by aortic valve vegetation and splenic lesion, ultimately requiring surgical intervention.

Case Report

The 55-year-old man, with history of hypertension and independent activities of daily living, presented with intermittent fever for two weeks, accompanied by chills, productive cough, exertional dyspnea, poor appetite, and generalized weakness. Initial laboratory examination at the Emergency Department revealed leukocytosis, anemia, and elevated C-reactive protein. No obvious pulmonary lesion was noted on chest X-ray. He was admitted for further evaluation and treatment.

Empirical antibiotics (Amoxicillin/clavulanic acid 1000mg/200mg) every eight hours were initiated. Blood culture later identified *Aggregatibacter actinomycetemcomitans*. Dental evaluation revealed chronic periodontitis, and multiple extractions were performed. Chest computed tomography showed subsegmental fibrosis and a splenic lesion; abdominal Magnetic Resonance Imaging favored splenic hemosiderosis, splenic abscess can not be excluded. Echocardiography was arranged for bacteremia, revealed aortic valve sclerosis with regurgitation and stenosis, and Transesophageal Echocardiography confirmed vegetation on the non-coronary cusp. Under the impression of infective endocarditis, we kept antibiotics treatment with ceftriaxone and moxifloxacin. However, antibiotics regimen was adjusted for multiple times due to persistent fever, including Ceftriaxone, Cefepime, and Moxifloxacin.

Given persistent vegetation and clinical instability, the patient underwent single valve replacement surgery. Postoperatively, he was monitored in Intensive Care Unit, successfully extubated on the next day, and continued Ceftriaxone(2g every twelve hours) and Metronidazole(500mg every six hours). We de-escalated antibiotic to single line ceftriaxone according infection department suggestion. He was discharged after receiving 5-week ceftriaxone treatment course.

Discussion

Infective endocarditis is a microbial infection of the endocardial surface of the heart, most commonly affecting heart valves. It presents with fever, heart murmurs, and evidence of systemic

emboli or immunologic phenomena. Diagnosis relies on blood cultures and echocardiography. Recent studies have highlighted the distinct clinical features of HACEK infective endocarditis, which accounts for 1–3% of all cases. Patients are typically younger, with a subacute course and frequent prosthetic valve involvement. Embolic events like stroke are common, but heart failure and mortality rates are lower than in other forms of endocarditis. Ceftriaxone and fluoroquinolones remain effective treatments despite rising ampicillin resistance. Overall, prognosis is favorable with low relapse and surgical intervention rates.

In our case, we presented a patient with HACEK-related infective endocarditis, detailing the process from diagnosis to treatment selection. Antibiotic therapy alone proved insufficient, prompting consideration of surgical intervention. Following the operation, the patient underwent a prolonged course of antibiotic treatment.

Conclusion

This case highlights the diagnostic and therapeutic challenges of HACEK-related infective endocarditis, particularly *Aggregatibacter actinomycetemcomitans*. Odontogenic sources should be considered in culture-positive bacteremia without clear portal of entry. Multidisciplinary collaboration—including infectious disease, cardiology, dentistry, and cardiovascular surgery—was essential for successful management. Early recognition and timely surgical intervention are critical to improving outcomes in such rare but serious infections.

病例報告 114_C140

從挑戰到噩夢：發生在 CHIP-PCI 的罕見臨床併發症

An unimaginable nightmare in CHIP-PCI（Complex high-risk indicated percutaneous coronary intervention）

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Introduction

Percutaneous coronary intervention (PCI) is generally safe, but rare catastrophic device complications can occur. One of the complication is failure of a dilated stent balloon to deflate and subsequent entrapment within the coronary artery. This situation is extremely uncommon but potentially fatal.

Case Report

We present a case of HTN, DM, and ESRD patient, who suffered from respiratory failure due to heart failure and NSTEMI. And we did the Percutaneous coronary intervention for him and LM lesion was noted. During the procedure, however, after we deployed the stent, the stent ballon was stuck and fractured after we pulled, severe chest pain and hemodynamic unstable had happened then. We had tried several methods, and fortunately, finally we able to retrieve by snare without causing disaster with normal coronary flow.

Discussion

Balloon entrapment is an uncommon complication during PCI procedure. If entrapment occurs, it can be divided into failure to deflate or deflatable. If deflation is possible, for example, we can try gentle traction, in a back and forth motion, and also we can advance another guide extensor for retrieval. But If the balloon fails to deflate, plus, even worse, with balloon shaft fracture. This is even rare and one of the most dreaded complications, especially at the left main. The trapped balloon can occlude coronary flow, causing MI, shock, and even death. And only several case reports have been founded through literature.

Conclusion

undeflated balloon entrapment with shaft fracture is a very rare complication while doing PCI. It may cause severe catastrophe. If such an unfortunate event occur, we should always, first, stay calm, then stabilize the patient's hemodynamics (including vasopressor, IABP, ECMO). Several percutaneous maneuvers can be tried, and if fail, finally, do not hesitate to contact the surgeons for emergent operation.

病例報告 114_C141

轉移性泌尿腺癌以模仿潰瘍性腸炎臨床症狀做為表現

Colon metastatic urothelial carcinoma mimic presentation of ulcerative colitis

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Introduction

Ulcerative colitis (UC) is characterized by recurring episodes of inflammation limited to the mucosal layer of the colon. It commonly involves the rectum and may extend proximally in a continuous fashion to affect other parts of the colon. The major symptoms of UC include diarrhea, rectal bleeding, tenesmus, passage of mucus, and crampy abdominal pain. Due to its non-specific and variable clinical presentation, several differential diagnoses must be considered in such patients.

Case Report

We present the case of a 61-year-old male patient with a known history of ulcerative colitis, previously treated with Vedolizumab and Ustekinumab, and maintained on long-term Mesalamine therapy. He also had a history of recurrent vocal cord cancer following chemotherapy and immunotherapy.

In July 2024, the patient developed fever and progressive diarrhea. Colonoscopic biopsy confirmed CMV colitis. His symptoms, including diarrhea and bloody stools, improved with antiviral treatment consisting of intravenous ganciclovir followed by oral acyclovir. However, a few months later, the patient experienced recurrent abdominal discomfort despite ongoing Ustekinumab treatment for UC. A repeat colonoscopy was performed in March 2025 to evaluate disease status, revealing partial stenosis with severely hyperemic and swollen mucosa from the splenic flexure to the descending colon, and a colonic ulcer with similarly hyperemic and swollen mucosa in the sigmoid colon, noted after biopsy. Pathological examination revealed metastatic carcinoma of urothelial origin.

Further investigations, including abdominal compute tomography, revealed metastatic lesions in both hepatic lobes and suspected urothelial carcinoma in the left mid to lower ureter. Liver biopsy confirmed metastatic carcinoma. The patient opted for palliative care and passed away one month later.

Discussion

Inflammatory bowel disease (IBD) is a chronic inflammatory condition of the gastrointestinal tract with a broad differential diagnosis. In suspected ulcerative colitis (UC), clinicians must consider infectious causes (bacterial, mycobacterial, parasitic, fungal) and non-infectious conditions such as ischemic colitis, diverticulitis, and neoplasms.Secondary metastasis to the colorectum is rare, most commonly originating from the lung, ovary, or breast. Urothelial carcinoma, which arises from the urinary tract lining, typically spreads to lymph nodes, bones, liver, and lungs, but may occasionally metastasize to the rectum, potentially causing constriction. Gastrointestinal involvement is usually through direct extension and often presents with GI symptoms, although

up to one-third of cases may remain asymptomatic and are diagnosed only on autopsy.

Conclusion

Ulcerative colitis is characterized by recurring episodes of mucosal inflammation, most commonly involving the rectum and potentially extending proximally in a continuous manner. In patients diagnosed with UC who experience refractory flare-ups or inadequate response to monoclonal antibody therapies, alternative causes of colitis such as other malignancy or infection must be considered. This case underscores the importance of maintaining a broad differential diagnosis, including the possibility of metastatic from atypical malignancy such as ureter urothelial carcinoma.

病例報告 114_C142

瀰漫性大 B 細胞淋巴瘤相關的繼發性冷型自體免疫性溶血性貧血：一例病例報告

Secondary Cold Autoimmune Hemolytic Anemia Associated with Diffuse Large B-Cell Lymphoma: A Case Report

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Introduction

Autoimmune hemolytic anemia (AIHA) may occur as an idiopathic disease or secondarily in association with infections, medications, autoimmune conditions, and malignancies. We report a case of cold AIHA secondary to diffuse large B-cell lymphoma (DLBCL), highlighting the importance of early recognition and comprehensive evaluation in patients presenting with unexplained hemolysis.

Case Report

The patient was a 59-year-old woman with no significant medical history. She presented with progressive jaundice, exertional dyspnea, fatigue, and cough for 2–3 weeks. She denied fever, drenching night sweats, weight loss, chest pain, or abdominal discomfort. Initial laboratory investigations revealed severe macrocytic anemia (Hb: 3.9 g/dL) with pancytopenia, indirect hyperbilirubinemia (Toal bilirubin/Direct bilirubin: 2.15/0.79 mg/dL), low haptoglobin (< 8 mg/dL), and elevated lactate dehydrogenase (LDH: 403.9 IU/L). The direct antiglobulin test was positive, with negative DAT-IgG and positive DAT-C3d, consistent with cold autoimmune hemolytic anemia (AIHA). A bone marrow biopsy demonstrated abnormal lymphoid cells forming aggregates, which were positive for CD20 immunostaining. Abdominal computed tomography (CT) demonstrated marked splenomegaly with focal low-density lesions in the spleen. Subsequently, positron emission tomography/computed tomography (PET/CT) revealed findings suspicious for lymphoproliferative disease involving the bone marrow, spleen, multiple lymph node regions (bilateral cervical, external iliac, axillary, and mediastinal), as well as the right palatine and lingual tonsils. A biopsy of the right cervical and oropharyngeal lymph nodes ultimately confirmed the diagnosis of diffuse large B-cell lymphoma (DLBCL). After the first cycle of R-CHOP chemotherapy, the patient's cold AIHA showed rapid improvement with resolution of hemolysis. Upon completion of six cycles of R-CHOP, positron emission tomography (PET) demonstrated complete metabolic remission, and subsequent follow-up imaging revealed no radiologic evidence of tumor recurrence.

Discussion

AIHA is a hematologic disorder caused by autoantibodies and/or complement targeting erythrocytes, leading to varying degrees of hemolysis. It is broadly classified into two major types: warm AIHA, typically mediated by IgG, and cold AIHA, mediated by IgM and complement activation. Approximately 50-60% of warm AIHA cases are secondary to an underlying condition, most commonly lymphoproliferative disorders such as lymphoma or chronic lymphocytic leukemia (CLL). Cold AIHA accounts for roughly 15-20% of all AIHA cases. The most common secondary

causes are infections, particularly Mycoplasma pneumoniae and Epstein-Barr virus. However, clonal lymphoproliferative disorders must not be overlooked. In the review "How I Treat AIHA", Go et al. emphasize that, although cold AIHA is less frequently linked to overt lymphoid malignancy than warm AIHA, the clinician should maintain a high index of suspicion - especially in patients with atypical features (e.g., cytopenias, organomegaly, or systemic symptoms).

Patients with lymphoma-associated AIHA are usually older, at higher risk of relapse and mortality, and often have bone marrow involvement. Our case illustrates cold AIHA as the initial presentation of DLBCL, with pancytopenia and marrow infiltration, fitting this profile. Although she achieved remission, long-term follow-up is essential to monitor for both lymphoma recurrence and AIHA relapse. This case highlights the importance of thorough evaluation for secondary causes in newly diagnosed AIHA and addressing the underlying conditions through appropriate treatment.

病例報告 114_C143

肝硬化病患之不明原因低血氧

Unexplained hypoxemia in a patient with liver cirrhosis

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Introduction

In patients with underlying chronic liver diseases, intrapulmonary vascular dilatations and shunting impair gas exchange and lead to hypoxemia. Here, we report a rare case of hepatopulmonary syndrome diagnosed by an agitation saline test.

Case Report

This 59-year-old male patient had a history of liver cirrhosis, Child-Pugh score B, HCV and alcoholism related. He suffered from shortness of breath for a year prior to hospitalization. His SpO2 is 84% breathing ambient air. Laboratory tests revealed moderate hypoxemia, with PaO2 66.9 mmHg under oxygen 4 liter/minute supply through nasal cannula, which showed widened A-a gradient (129.9 mmHg). The pulmonary function test indicated small airway disease only. Chest CT scan showed mild fibrosis and emphysema of both lungs. Echocardiography demonstrated normal biventricular systolic function and excluded severe valvular diseases. According to the above studies, the cause of hypoxemia cannot be confirmed. To exclude pulmonary shunting under hypoxemia with widened A-a gradient, cardiac saline agitation test under echocardiography was done. Microbubbles appeared in the left chambers of the heart after 4 cardiac cycles, which indicated the presence of extra-cardiac shunting. Hepatopulmonary syndrome was diagnosed, based on his underlying liver cirrhosis, indirect evidence of pulmonary shunting, and hypoxemia. We recommended this patient receive liver transplantation and under evaluation.

Discussion

To approach hypoxemia, causes included (1) reduced inspired oxygen tension, (2) alveolar hypoventilation resulted from airway obstruction or lung parenchymal disease, (3) focal diminished V/Q ratio, (4) shunting, either alveolar filling processes, intra-cardiac or intra-pulmonary shunting, and (5) decreased oxygen diffusion capacity, such as interstitial lung disease. Among these mechanisms, V/Q mismatch, shunting, and decreased oxygen diffusion capacity all can result in widened A-a gradient. Assisted by pulmonary function test and chest CT scan, the differential diagnosis was narrowed down to shunting.

Hepatopulmonary syndrome (HPS) is characterized by presence of chronic liver diseases or portal hypertension, intra-pulmonary vascular dilatation that results in right-to-left shunt, and impaired gas exchange. In patients with HPS, prevalence of pulmonary vascular abnormality detected by chest CT scan is low, which makes chest CT scan an unreliable examination to detect intra-pulmonary shunting. In an agitation saline test, also named transthoracic contrast echocardiography, saline with microbubbles is injected into the venous system. Diameters of normal pulmonary capillaries are too small to let microbubbles pass through to the left side of the heart. Opacification on echocardiography will only be presented in right-side heart chambers. In

contrast, intra-pulmonary vascular dilatation allows microbubbles to pass through, thus opacification will appear in the left-side heart chambers after 3 to 4 cardiac cycles. The delay of this opacification also differentiates intra-pulmonary and intra-cardiac shunting. Liver transplantation remained the only curative management for HPS. Post liver transplantation 5-year survival is reported to be 76%.

Conclusion

In patients with underlying chronic liver diseases, hypoxemia with high A-a gradient should raise awareness of intra-pulmonary shunting and hepatopulmonary syndrome. Agitation saline tests can detect intrapulmonary shunting and confirm the diagnosis. Patients receiving liver transplantation have good survival. Early diagnosis of HPS and referral for transplantation evaluations are important.

病例報告 114_C144

孕婦以三維導航輔助無透視永久性心律調節器植入：病例報告

Fluoroless Permanent Pacemaker Implantation in a Pregnant Woman with Intermittent Complete Atrioventricular Block: A Case Report

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Introduction

Permanent pacemaker (PPM) implantation is a well-established treatment for advanced atrioventricular (AV) block. However, fluoroscopic guidance raises concerns in pregnant women, particually during the first trimester due to fetal radiation risks. Recent advances in electroanatomical mapping systems allow fluoroless implantation. We report the case of a pregnant woman with symptomatic complete AV block who successfully underwent fluoroless PPM implantation with the assistance of a 3D mapping system.

Case Report

A 31-year-old woman with hyperthyroidism under propylthiouracil presented with near-syncope while working. She experienced transient dizziness and a brief visual blackout. Holter monitoring revealed sinus rhythm with intermittent second- and third-degree AV blocks, including high-grade and 2:1 AV blocks, with the longest R-R interval of 2.18 seconds. Echocardiography demonstrated preserved left ventricular systolic function (LVEF 75.6%). She was 7 weeks pregnant (G1P0A1) and on progesterone for threatened abortion.

Given symptomatic intermittent complete AV block, PPM implantation was indicated. After obstetric consultation and informed consent, the procedure was performed under local anesthesia. Left subclavian vein access was obtained by anatomical landmarks. A 3D electroanatomical mapping system reconstructed venous and cardiac anatomy using a J-tip guidewire and decapolar catheter. A right ventricular screw-in lead was positioned at the mid-septum, and a St. Jude Endurity MRI VVI generator was implanted. Wound closure was performed with subcutaneous sutures and tissue adhesive. Radiation exposure was < 0.01 mGy. The postoperative course was uneventful, and she was discharged with cardiology and obstetric follow-up.

Discussion

Pacemaker implantation during pregnancy is rarely required but may be lifesaving in patients with symptomatic complete AV block. Conventional fluoroscopy poses risks of fetal radiation exposure, especially in the first trimester when organogenesis occurs. Although the estimated threshold for teratogenic effects is >50–100 mGy, minimizing exposure is critical.

Recent advances in 3D electroanatomical mapping systems allow near-zero or zero-fluoroscopy implantation of pacing devices, with comparable efficacy and safety to standard techniques [4]. In our case, the 3D system provided visualization of venous anatomy and cardiac chambers, facilitating safe lead placement without fluoroscopy. Our patient’s total radiation exposure was <0.01 mGy, essentially negligible.

Several case reports have also demonstrated successful fluoroless pacemaker implantation in pregnant women using electroanatomical mapping or intracardiac echocardiography [1–3]. This case adds to the growing evidence that fluoroless pacemaker implantation is feasible, safe, and effective, especially in high-risk populations such as pregnant women.

Conclusion

We report a successful fluoroless PPM implantation in a pregnant woman with complete AV block. This case supports the use of 3D mapping to achieve safe device implantation while avoiding fetal radiation exposure.

病例報告 114_C145

以噬血症候群表現的瀰漫性組織胞漿菌病：本土案例報告及文獻回顧

Disseminated Histoplasmosis Presenting as Hemophagocytic Lymphohistiocytosis: A Probable Indigenous Case Report and Literature Review

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Introduction

Histoplasmosis, an endemic mycosis caused by *Histoplasma capsulatum*, is prevalent in specific regions like North and Latin America but is considered rare in Taiwan, with only a few probable indigenous cases reported between 1977 and 2023[1]. The thermally dimorphic fungus exists as a mold in the environment and transitions to a yeast form within a host. In immunocompromised individuals, it can lead to a life-threatening disseminated infection. A severe complication of disseminated histoplasmosis is secondary hemophagocytic lymphohistiocytosis (HLH), a hyperinflammatory syndrome characterized by persistent fever, cytopenias, and hepatosplenomegaly[2]. We report the case of a patient with underlying myelofibrosis who presented with fever of unknown origin, digital cyanosis, and multi-organ failure, ultimately diagnosed as disseminated histoplasmosis-associated HLH.

Case Report

A 57-year-old male with a history of polycythemia vera presented with a one-week history of fever, malaise, and cyanosis of his right index finger. He reported a brief episode of similar symptoms a month prior. On admission, he was febrile to 38.1°C, and physical examination was notable for mild left upper quadrant tenderness. Initial laboratory results were significant for an elevated C-reactive protein (44.34 mg/L), mild transaminitis, and a markedly elevated D-dimer (17.22 mg/L mg/L). An abdominal CT scan confirmed splenomegaly with suspected focal infarction. The patient was admitted to the hematology service for presumed peripheral artery disease secondary to his underlying condition. Despite treatment with antiplatelet agents and vasodilators, which led to the improvement of his digital cyanosis, his fever persisted. Empirical antibiotic therapy with cefoxitin, later escalated to piperacillin-tazobactam, failed to elicit a clinical response.

The patient's condition rapidly deteriorated, progressing to multi-organ failure characterized by worsening pancytopenia, acute kidney injury, severe hepatitis, and coagulopathy. This clinical picture raised strong suspicion for HLH. The diagnosis was supported by key laboratory findings, including hypertriglyceridemia (397 mg/dL), extreme hyperferritinemia (>33,511.2 ng/mL), and hypofibrinogenemia (114 mg/dL), corresponding to an H-Score of 262 (>99% probability of HLH). With routine cultures remaining negative, a bone marrow aspiration was performed. The Giemsa-stained smear was pivotal, revealing hemophagocytosis and numerous intracellular yeast-like organisms within macrophages. Based on this finding, treatment with liposomal amphotericin B was promptly initiated. The diagnosis was definitively confirmed by serum metagenomic next-generation sequencing and subsequent bone marrow culture, both identifying *Histoplasma capsulatum*. Step-down therapy followed with isavuconazole 200 mg daily, subsequently transitioned to itraconazole 200 mg twice daily. The patient's condition steadily improved with

targeted antifungal therapy, and a later bone marrow biopsy also confirmed underlying myelofibrosis.

Discussion

This case highlights a rare instance of probable indigenous disseminated histoplasmosis in Taiwan, occurring in an immunocompromised host with myelofibrosis. The clinical presentation was particularly challenging, as the infection manifested as a severe, life-threatening HLH. Disseminated histoplasmosis is a significant fungal trigger for HLH, especially in immunocompromised states. While conventional diagnostics were initially unrevealing, direct bone marrow examination proved pivotal for rapid diagnosis by identifying intracellular yeasts and hemophagocytosis[3]. This allowed for the prompt initiation of targeted antifungal therapy, underscoring its diagnostic importance in patients with similar presentations, even in non-endemic areas.

病例報告 114_C146

5-FU 化學治療後少見併發症之病例報告

A rare complication after 5-FU chemotherapy

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Introduction

Hyperammonemia is a serious metabolic disorder that commonly occurs in patients with hepatic cirrhosis and portal hypertension. However, non-cirrhotic hyperammonemia can result from various etiologies, including increased ammonia production, decreased elimination, or drug-induced mechanisms. Among various medication, 5-fluorouracil (5-FU) is recognized as the second most common cause of drug-induced hyperammonemia after valproate, with an incidence ranging from 1-10% and typically occurring 0.5 to 5 days after administration. Here, we presented a case of hyperammonemia event occurred after second cycle of 5-FU chemotherapy.

Case Report

A 62-year-old female with no significant past medical history presented with 3 weeks of progressive jaundice, tea-colored urine, clay-colored stools, and 6 kg weight loss. Ultrasonography revealed a 3.8 cm pancreatic head mass with biliary obstruction and suspected liver metastases. Endoscopic Ultrasonography tissue biopsy confirmed pancreatic adenocarcinoma, staged as T4N2M1.The patient received first cycle of FOLFIRINOX chemotherapy (5-FU, Irinotecan, Oxaliplatin) in hospital without complication. Two days after the second cycle at outpatient clinic, she developed nausea, vomiting, and diarrhea, followed by acute consciousness change with upward gaze deviation and subsequent tonic seizures. Laboratory evaluation revealed severe hyperammonemia (1110 μg/dL, normal 31-123), metabolic acidosis (pH 7.31, anion gap 27.9), and elevated lactate (109 mg/dL). After reviewing her history, 5-FU is highly suspect the causative medication. Immediate management included 5-FU discontinuation, sodium bicarbonate, levetiracetam, and lactulose. The patient's ammonia levels normalized within 24 hours, consciousness fully recovered, and she was subsequently switched to alternative chemotherapy (Abraxane plus Gemcitabine) without recurrence.

Discussion

Ammonia metabolism primarily occurs through the urea cycle in the liver, which is the only organ capable of converting ammonia to urea for elimination. This system is often disrupted in cirrhotic patients. However, etiologies of non-cirrhotic hyperammonemia including GI bleeding, certain urea-producing bacteria, portosystemic shunting, medications, and congenital urea cycle disorders should not be forgotten. 5-FU is a pyrimidine analog widely used in the treatment of gastrointestinal, pancreatic, head and neck, and breast cancers. While its primary mechanism involves DNA and RNA synthesis inhibition, 5-FU can also interfere with the Krebs cycle and subsequently disrupt the urea cycle, potentially leading to hyperammonemia, particularly in patients with dihydropyrimidine dehydrogenase (DPD) deficiency. Immediate 5-FU cessation is a crucial step in managing this situation.

Conclusion

This case demonstrates the importance of recognizing 5-FU-induced hyperammonemic encephalopathy as a potentially reversible cause of altered consciousness in cancer patients receiving 5-FU chemotherapy. Early recognition and prompt discontinuation of the offending agent are crucial for patient survival and neurological recovery. Alternative chemotherapy regimens should be used in patients who experience 5-FU-induced hyperammonemia.

病例報告 114_C147

慢性砷曝觸併發急性骨髓性白血病與邊緣區淋巴瘤：個案報告與系統血液學模型

Chronic Arsenic Exposure and Development of Acute Myeloid Leukemia with Simultaneous Marginal Zone Lymphoma: A Case Report with Systems Hematology Model

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Introduction

The carcinogenic effect of arsenic exposure had been reported since the 1970s. Current literature supports a strong association between arsenic and the development of solid tumors, but no clear association is observed for the hematological malignancies clinically. Herein, we present a case of acute myeloid Leukemia with simultaneous marginal zone lymphoma who suffered from chronic arsenic exposure for more than twenty years.

Case Report

This is an 87-year-old farmer with a medical history of chronic kidney disease, diabetes mellitus and an abnormal urine arsenic level noted since 2014. He reported cutaneous contact to fulvic acid, a chemical compound used in soil conditioning that has trace amounts of arsenic for more than twenty years. Upon that, he also developed a habit of ingesting 1 gram of fulvic acid per day between 2017-2018. He received dimercaptosuccinic acid (DMSA) treatment in 2018 under the impression of arsenic poisoning.

The patient had lower gum biting difficulties and pain for three months from September 2022. He then underwent a tooth extraction procedure on January 3rd, 2023, after which he developed persistent postoperative gum bleeding.

After admission to the hematology ward, blood workup showed 8% of peripheral blast, and bone marrow revealed acute myeloid leukemia (AML) M2 with 22% blasts. Bone marrow biopsy revealed AML with lymphoid infiltrates, which later proved to be CD5-positive marginal zone lymphoma. Gingival tumor biopsy also revealed myeloid sarcoma and CD5-positive marginal zone lymphoma. Immunohistochemistry tests of small lymphocytic infiltrates revealed CD20+, CD5+, CD10-, CD23-, BCL6-and cyclin D1-. Chromosome analysis showed 47, XY, +8[29]/46, XY[1]. Molecular studies revealed ASXL1, RUNX1 mutations..

Despite receiving three cycles of venetoclax and low dose cytarabine, his bone marrow studies for treatment response still suggested non-remission. Unfortunately, he suffered from prolonged

pancytopenia, which led to multiple episodes of bleeding, renal function deterioration, and sepsis. The patient expired days after signing his will on do-not-resuscitate form.

Discussion

Cohort study by Ott et. al. in 1974 revealed increased incidence of leukemia and Hodgkin diseases in occupational exposure to arsenic. Risk factors for leukemogenesis include old age, environmental exposures, and genetic disorders. Through this case we will review how arsenic may contribute to the development of leukemia ranging from epigenetic dysregulation to cell development. At the molecular level, arsenic can interrupt some key mechanisms such as DNA methylation, histone modifications and creating excessive oxidative stress. Gene expression studies of lymphocytes from arsenic-exposed humans had strengthened the arsenic-associated inflammation model. ASXL1 mutation suggested a clonal hematopoiesis, which might also suggest an “inflammaging” process.

Conclusion

By elucidating possible mechanisms between arsenic and hematological malignancies, we can hopefully disentangle the niche of clonal hematopoiesis, inflammation, genomic instability, and leukemogenesis. This case might reveal possibilities to further explore the role of chelating agents in antileukemic therapy.

病例報告 114_C148

新冠肺炎急性呼吸窘迫症候群合併右心衰竭之治療挑戰與探討

Right Ventricular Failure as a Key Prognostic Challenge in Severe COVID-19 ARDS: A Case-Based Review

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Introduction

Right ventricular dysfunction (RVD) frequently complicates COVID-19-induced ARDS and is a critical factor associated with poor prognosis and increased mortality. RVD is primarily driven by mechanisms including increased pulmonary vascular resistance, hypoxic pulmonary vasoconstriction, pulmonary vascular obstruction and the effects of mechanical ventilation.

Case Report

A 68-year-old male with no significant past medical history was admitted due to severe ARDS progressing from COVID-19 pneumonia. The patient rapidly deteriorated, developing shock and multi-organ failure. Echocardiographic assessment revealed right ventricular dilation and reduced function, confirming RVD.

Discussion

The management presented significant challenges. We focused on optimizing right ventricular preload, afterload, and contractility through judicious fluid adjustment, precise use of vasopressors and inotropic agents, lung-protective ventilation strategies, and prone positioning.

Conclusion

This case highlights the imperative need for vigilance regarding RVD in the treatment of COVID-19 ARDS. Early diagnosis, continuous monitoring, and a comprehensive strategy for RVD management are crucial for improving the survival rate of these critically ill patients.

病例報告 114_C149

免疫治療誘發之潰瘍性大腸炎：晚期肝細胞癌免疫治療之罕見但可逆的併發症

Ulcerative Colitis Induced by Immunotherapy: A Rare but Reversible Complication in Advanced Hepatocellular Carcinoma

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Introduction

With the accumulation of results from various clinical trials, immunotherapy has gradually emerged as one of the therapeutic options for advanced hepatocellular carcinoma. However, treatment may also lead to associated adverse effects. Therefore, the timely recognition and appropriate management of these immune-related events has become a crucial clinical challenge.

Case Report

A 64-year-old man with stage IV hepatocellular carcinoma (HCC) and lymph node metastasis was treated with a combination of Atezolizumab (immune checkpoint inhibitor) and Bevacizumab (anti-angiogenic agent) starting April 2, 2025. Initial therapeutic response was favorable, with tumor regression and a decline in serum alpha-fetoprotein (AFP) from 1110 ng/mL to 733 ng/mL. However, after the fifth cycle of immunotherapy, the patient developed severe immune-related diarrhea—20 to 30 episodes daily—unresponsive to Loperamide, accompanied by dehydration, acute kidney injury, and electrolyte disturbances. Colonoscopy revealed left-sided colitis and sigmoid ulcers, raising concern for immune-related ulcerative colitis, a rare but serious immune-related adverse event (irAE). Oral Mesalazine (2 g BID) and Prednisolone (25 mg QD) were initiated on August 18, 2025. The patient's diarrhea improved dramatically—reduced to six episodes on Day 2 and resolved completely by Day 3. Immunotherapy was successfully resumed on August 20 (seventh cycle) without recurrence of colitis. The patient was discharged and remains stable under outpatient follow-up.

Discussion

Ulcerative colitis induced by immunotherapy posed a clinical dilemma: continuing immunotherapy risked worsening colitis, whereas discontinuation could allow HCC progression. Prompt recognition, colonoscopy evaluation, and early anti-inflammatory and corticosteroid therapy can effectively reverse immune-mediated colitis. Crucially, this enables the continuation of life-prolonging cancer therapy without interruption. Systematic literature review and timely immunosuppressive management play pivotal roles in navigating such therapeutic dilemmas.

Conclusion

This case highlights ulcerative colitis as a rare but reversible complication of immunotherapy in HCC patients. Timely diagnosis and appropriate treatment can improve the management of this complications.

病例報告 114_C150

電腦斷層顯示腦室腹腔分流管感染造成的腹膜端增強環 之病例報告

Peritoneal End Enhancing Ring on Computed Tomography Indicating Ventriculoperitoneal Shunt Infection: A Case Report

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Introduction

Ventriculoperitoneal (VP) shunt infection remains a serious complication with significant morbidity. Clinical manifestations are often non-specific, and imaging findings at the peritoneal end may provide an early diagnostic clue for timely intervention.

Case Report

A 47-year-old man with a history of traffic accident sustained bilateral subdural and subarachnoid hemorrhage, for which he later underwent cranioplasty. Progressive communicating hydrocephalus subsequently necessitated placement of a ventriculoperitoneal (VP) shunt. He later developed fever and abdominal pain without an obvious infectious focus and was treated with several broad-spectrum antibiotics, including vancomycin and ceftazidime, without improvement. Contrast-enhanced abdominal computed tomography demonstrated a ring-enhancing lesion measuring 9 mm in diameter and 35 mm in length surrounding the distal opening of the VP shunt. Adjacent fat stranding and mild peritoneal thickening were also noted, findings highly suggestive of shunt-related infection. After exteriorization of the shunt through the abdominal wall, cerebrospinal fluid culture yielded *Staphylococcus epidermidis*. Definitive resolution was achieved only after complete removal of the VP shunt followed by external ventricular drainage. The patient's fever subsided promptly, inflammatory markers normalized, and he was discharged home in stable condition without recurrence.

Discussion

A ring-enhancing lesion at the peritoneal end of a VP shunt should raise strong suspicion for shunt infection. This case highlights the limited efficacy of antibiotics alone and reinforces that complete shunt removal combined with targeted antimicrobial therapy is often necessary for clinical cure. Early recognition through imaging, together with microbiological confirmation, is essential for guiding optimal management.

Conclusion

Even a minor radiological sign on computed tomography can provide a critical clue for diagnosis.

病例報告 114_C151

一例華氏巨球蛋白血症的病人發生麴菌化膿性甲狀腺炎併短暫甲狀腺毒症的個案報告

Aspergillus suppurative thyroiditis in Waldenstrom marcorglobulinemia with transient thyrotoxicosis: a case report

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Introduction

Aspergillus suppurative thyroiditis is rare form of infectious thyroiditis, it may present with less acute symptoms and can mimic subacute thyroiditis. Definitive diagnosis is established by aspiration or biopsy, demonstrating hyphae on microscopy or positive fungal culture for Aspergillus. In this report, we describe a case of aspergillus suppurative thyroiditis in the patient of Waldenstrom marcorglobulinemia.

Case Report

A 71-year-old man had past history of Waldenstrom marcorglobulinemia, which was diagnosed on 2025/04 and he is regularly undergoing chemotherapy and targeted therapy. In early August, 2025, the patient felt gradual swelling and painful over left middle neck region. Thus, he came to our emergent department for evaluation. Blood test report indicated leukopenia and high level of CRP. Neck ultrasound showed bilateral multinodular goiter of thyroid gland, especially for left large thyroid nodule. Computed tomography showed an ill-defined mass lesion about 3.8 cm x 4.4 cm x 5.0 cm at left thyroid region. Therefore, needle biopsy was performed to left thyroid nodule after admission.

Biopsy result showed numerous hyphae and blood aspergillus galactomannan antigen is positive with index up to 18.57. The patient accepted left thyroidectomy and voriconazole was administered for weeks. Ultimately, after completion of treatment, the patient's condition stabilized and he was discharged.

Thyroid function tests were monitored during the hospitalization. In the admission day, thyrotoxicosis was shown to the value of free T4 4.32 ng/dL and undetectable TSH . Following left thyroid lobectomy and stabilization with antifungal therapy, the patient's thyroid function remained within normal limits without pharmacological treatment.

Discussion

Our patient initially presented with left throat region swelling and painful for one week. Ultrasonography and computed tomography revealed thyroid nodule and aspiration to the suspicious nodule provided critical diagnostic evidence, demonstrating fungal hyphae. Serum galactomannan assay was strongly positive, further supporting the diagnosis of invasive aspergillosis.

Thyroid function monitoring revealed transient thyrotoxicosis during the acute infectious phase. This likely resulted from follicular disruption and hormone release. Notably, after infection control and thyroidectomy, thyroid function normalized.

Conclusion

First, in immunocompromised patients presenting with neck swelling or painful, suppurative thyroiditis should be considered as a possible differential diagnosis. Second, diagnostic confirmation of aspergillus suppurative thyroiditis often requires a combination of pathology and fungal biomarkers, as imaging alone is insufficient. Third, integrated management with voriconazole and surgical resection can provide effective infection control. Finally, close monitoring of thyroid function is warranted, given the potential for transient dysfunction.

病例報告 114_C152

機械通氣期間發生嚴重支氣管痙攣，需進行 V-V ECMO 治療：案例報告

Severe Bronchospasm During Mechanical Ventilation Requiring V-V ECMO: A Case Report

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Introduction

Bronchospasm during invasive ventilation is rare but potentially life-threatening. Aerosol therapy and ventilator-nebulizer interactions may complicate detection and worsen patient outcomes.

Case Report

A 73-year-old man with systemic sclerosis, interstitial lung disease, and coronary artery disease was admitted for COVID-19 pneumonia on 2024/7/27. He was intubated for respiratory failure and placed on pressure-controlled ventilation with regular intermittent bronchodilator delivery via a Jet Nebulizer (JN) in the inspiratory limb. During hospitalization, he developed sudden severe bronchospasm with SpO_2 48%, hypotension, and decreased breath sounds. IV succinylcholine, inhaled epinephrine, terbutaline and ipratropium via JN were administered but hypoxemia persisted. The patient subsequently suffered asystole, achieving ROSC after 4 minutes of CPR. Due to refractory hypoxemia, V-V ECMO was initiated and oxygenation gradually improved. ECMO was later removed, but his course was complicated by sepsis, multi-organ failure, and he expired on 2024/8/20.

Discussion

Nebulizer aerosol can induce turbulent airflow, leading to ventilator flow sensor misdetection and autotriggering, causing ventilator waveforms to appear normal despite severe bronchospasm(1)(2). This may delay recognition until critical desaturation occurs. Furthermore, nebulizer flow increases airway resistance and auto-PEEP, exacerbating dynamic hyperinflation and patient-ventilator asynchrony(3)(4). Continuous bronchodilator delivery via a Jet Nebulizer (JN) carries additional risk, as JN generates higher circuit flow and is associated with lower aerosol deposition efficiency, potentially worsening these effects. According to recent consensus recommendations, Vibrating Mesh Nebulizers (VMN) and pMDI + spacer are preferred over JN during invasive ventilation because they produce more uniform particles, improve lung deposition, and introduce less additional flow(5). To optimize aerosol delivery and ventilator synchrony, clinicians should place the nebulizer before the HME, prefer VMN or pMDI+spacer when feasible, adjust ventilator trigger sensitivity, and closely monitor waveforms to avoid false tidal volume interpretation. Early ECMO should be considered when hypoxemia persists despite maximal intervention.

Conclusion

Nebulizer-induced flow interference may mask severe bronchospasm under mechanical ventilation. Optimized bronchodilator delivery, vigilant ventilator monitoring, and early ECMO support are essential for improving outcomes.

病例報告 114_C153

蛋白質流失性腸病合併重度低白蛋白血症病例報告

A Case of Protein-Losing Enteropathy with Severe Hypoalbuminemia

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Introduction

Protein-Losing Enteropathy (PLE) is a rare condition characterized by the loss of protein from the gastrointestinal tract, resulting in hypoalbuminemia and edema. It occurs due to various underlying causes, including lymphatic obstruction, inflammatory bowel diseases, infections, and malignancies. We present a case of protein-losing enteropathy in a 60-year-old man, who initially presented with bilateral lower limb edema.

Case Report

A 60-year-old male with a history of hypertension, hyperlipidemia, glomerular disease, and IgM monoclonal gammopathy of undetermined significance (IgM MGUS) presented with bilateral leg edema persisting for several years. His symptoms included dyspnea secondary to pleural effusion, with long-term use of loop diuretics. He experienced recurrent symptomatic hypotension, requiring frequent visits to the emergency department for albumin injections. The patient denied orthopnea, paroxysmal nocturnal dyspnea, jaundice, dark urine, gastrointestinal bleeding, or poor oral intake.

The most recent hospitalization was to clarify the cause. Laboratory results showed normal hematological and electrolyte parameters, but hypoalbuminemia was present (1.6 g/dL). Repeated urine protein-to-creatinine ratio (UPCR) measurements were not elevated (307.12 mg/g, 265.49 mg/g), thus ruling out nephrotic syndrome. MGUS-related hypoalbuminemia was suspected. A hematologist recommended a whole-body CT scan to evaluate for possible lymphadenopathy. Interestingly, abdominal CT revealed segmental thickening of the small bowel wall. Further investigation with capsule endoscopy revealed small bowel villus atrophy. Additionally, a low serum alpha-1 antitrypsin level (62.40 mg/dL) was noted. The final diagnosis was protein-losing enteropathy.

Discussion

The precise prevalence of PLE remains difficult to determine due to its diverse etiology and clinical presentation. However, it has become increasingly recognized in association with both systemic and gastrointestinal diseases.

Diagnosis typically involves a combination of clinical signs (such as edema, diarrhea, and abdominal distension), serum and fecal tests (e.g., alpha-1 antitrypsin clearance), and imaging studies (endoscopy) to assess the extent of protein loss and identify the underlying cause. It is crucial to exclude other causes of hypoalbuminemia. Liver diseases such as cirrhosis can impair albumin synthesis, while nephrotic syndrome results in albumin loss through the urine. Malnutrition and chronic inflammation associated with infections or autoimmune diseases can also lead to hypoalbuminemia.

At our institution, we faced limitations in performing diagnostic tests, such as stool alpha-1 antitrypsin and albumin scintigraphy, which provide diagnostic value for a comprehensive workup. Consequently, we referred the patient to a tertiary care center for more advanced evaluations, including biopsy and pathology reports, to confirm the diagnosis.

Conclusion

Given the complexity of diagnosing PLE and the wide range of potential underlying causes, it is crucial for clinicians to maintain a broad differential diagnosis and utilize a multidisciplinary approach to provide the best care for patients with this rare condition.

病例報告 114_C154

糞腸球菌菌血症導致腹膜透析患者次發性腹膜炎及動靜脈瘻管感染：病例報告

Enterococcus faecalis Bacteremia Leading to Secondary Peritonitis and Arteriovenous Graft Infection in a Patient on Peritoneal Dialysis

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Introduction

Peritoneal dialysis (PD)-related peritonitis is typically a primary infection originating from the catheter exit site or intraluminal contamination. However, Enterococcus species, especially E. faecalis, can cause secondary peritonitis following seeding from a distant focus via bacteremia, which is rare and carries higher morbidity. This case presents a crucial diagnostic challenge due to its atypical presentation and multifocal complications.

Case Report

A 65-year-old male on PD presented with fever and vomiting for 2 days, followed by turbid dialysate and abdominal pain. This sequence is atypical for primary PD peritonitis. Initial blood cultures yielded E. faecalis before dialysate cultures turned positive, supporting a primary bacteremia. After peritonitis treatment, a follow-up visit revealed left upper arm arteriovenous graft (AVG) swelling and abscess formation. Pus culture from the surgically debrided AVG also grew E. faecalis, establishing a simultaneous multi-site infection.

Discussion

The clinical course, characterized by initial fever and bacteremia preceding peritonitis, suggests a hematogenous spread, leading to secondary peritonitis. E. faecalis often originates from the gastrointestinal or genitourinary tracts, which may serve as the primary source for both the bacteremia and subsequent seeding of the peritoneum and the AVG. This highlights the pathogen's high virulence and its propensity to cause infective endocarditis or distant abscesses.

Conclusion

This case underscores the importance of recognizing E. faecalis bacteremia as a cause of secondary peritonitis in PD patients, which may present atypically. Furthermore, it demonstrates a rare complication of simultaneous AVG infection from the same organism. Comprehensive workup for the primary source and aggressive treatment for multi-site infection are essential for optimal outcomes.

病例報告 114_C155

高齡壺腹腺癌造成膽道與十二指腸雙重阻塞：內視鏡金屬支架內支架技術之應用

Advanced Ampullary Adenocarcinoma with Biliary and Duodenal Obstruction: Successful Endoscopic Stent-in-Stent Revision in an Elderly Patient

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Introduction

Ampullary adenocarcinoma is a rare malignancy with a generally poor prognosis. Most elderly patients present at advanced stages and are not surgical candidates. Progressive tumor growth may result in simultaneous biliary and duodenal obstruction, posing a major challenge for palliation. Endoscopic metallic stenting, particularly the stent-in-stent technique, provides a minimally invasive option for symptom relief.

Case Report

A 92-year-old woman was diagnosed with adenocarcinoma of the ampulla of Vater and initially received a biliary metallic stent for obstructive jaundice. One year later, she presented with persistent nausea and abdominal distention. CT demonstrated tumor progression with biliary dilatation, pneumobilia, and duodenal obstruction. Endoscopy confirmed duodenal luminal narrowing and occlusion of the previously placed biliary stent. ERCP with metallic stent-in-stent revision was performed: the obstructed biliary stent was cannulated through the mesh, and a second metallic stent was deployed to restore drainage. The patient's symptoms improved markedly, and she was discharged in stable condition.

Discussion

Stent dysfunction after metallic stent placement occurs in 3–45% of patients, usually due to tumor ingrowth, overgrowth, or debris. In cases of concurrent biliary and duodenal obstruction, reintervention is particularly complex because of distorted anatomy, tight strictures, and overlapping wire mesh from prior stents. The stent-in-stent technique involves careful cannulation through the mesh of the occluded stent and deployment of a new metallic stent within the existing one. This procedure is technically demanding, requiring precise wire control and fluoroscopic guidance, but it enables re-establishment of drainage without resorting to surgical bypass. Compared with alternative strategies such as percutaneous drainage, stent-in-stent provides more durable patency, reduces recurrent obstruction, and allows repeat endoscopic access if needed. In this case, the successful use of stent-in-stent demonstrated that even in elderly patients with advanced disease, effective palliation can be achieved through minimally invasive means.

Conclusion

In elderly patients with unresectable ampullary adenocarcinoma, endoscopic stent-in-stent revision offers an effective palliative strategy for managing combined biliary and duodenal obstruction, improving quality of life despite poor overall prognosis.

病例報告 114_C156

免疫檢查點抑制劑誘發之第三型自體免疫性胰臟炎：一例食道鱗狀癌合併膽道阻塞之病例報告

Type 3 Autoimmune Pancreatitis with Biliary Obstruction Following Nivolumab Therapy in Esophageal Squamous Cell Carcinoma: A Case Report

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Introduction

Autoimmune pancreatitis (AIP) is an immune-mediated pancreatobiliary disorder, classically divided into type 1 (IgG4-related) and type 2 (idiopathic duct-centric). The recently recognized type 3 AIP is immune checkpoint inhibitor (ICI)-induced, with an incidence of 0.6–4% among patients on ICI therapy. It frequently mimics malignant obstruction, creating diagnostic and therapeutic challenges.

Case Report

A 48-year-old man with esophageal squamous cell carcinoma (cT3N2M0) underwent chemoradiotherapy (Dec 2023–Jan 2024) followed by nivolumab from March 2024. After nine cycles, incidental laboratory tests revealed liver enzyme elevation (AST 122 IU/L, ALT 96 IU/L, ALP 227 IU/L, GGT 169 IU/L, total bilirubin 0.45 mg/dL) and pancreatic enzyme elevation (lipase 165 IU/L; normal upper limit 82 IU/L). CT demonstrated pancreatic edema with biliary dilatation. ERCP excluded choledocholithiasis or tumor obstruction, and a plastic stent was placed for decompression. Serum IgG (828 mg/dL) and IgG4 (67.2 mg/dL) were within normal range. EUS-FNB excluded malignancy and IgG4-related disease. A diagnosis of ICI-induced AIP (type 3) was made based on temporal association with nivolumab, characteristic imaging findings and exclusion of alternative etiologies. Prednisolone (10 mg/day) was initiated, leading to normalization of enzymes and radiologic improvement within one month. The biliary stent was removed three months later.

Discussion

ICI-induced AIP is an emerging entity within the spectrum of immune-related adverse events. Its clinical presentation often overlaps with malignant biliary obstruction, particularly in oncology patients, making accurate diagnosis challenging. In this case, the absence of stones or tumor obstruction, normal IgG and IgG4 levels, and negative histopathology helped rule out alternative causes. The close temporal relationship with nivolumab exposure and rapid response to corticosteroids further supported the diagnosis of AIP type 3. Although the role of steroids in AIP-3 remains debated, our patient demonstrated a prompt biochemical and radiologic recovery, consistent with prior case reports. This emphasizes the need for early recognition, timely immunosuppression, and multidisciplinary collaboration to prevent irreversible pancreatic injury while maintaining optimal cancer care.

Conclusion

Type 3 AIP is an uncommon but clinically significant irAE. Early recognition and timely

immunosuppression are critical to prevent irreversible pancreatic injury and to optimize outcomes in cancer patients undergoing immunotherapy.

病例報告 114_C157

升結腸黏液性腺癌造成的成人腸套疊病例報告

Adult intussusception as an unusual presentation of ascending colon mucinous adenocarcinoma: a case report

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Introduction

Intussusception occurs when a portion of the intestine telescopes into an adjacent segment, usually due to abnormal intestinal motility or intraluminal lesions such as tumors. In children, most cases are idiopathic, whereas in adults, an organic etiology is usually identified. Benign and malignant tumors of the colon, as well as postoperative adhesions, are common lead points. Unlike the pediatric "classic triad" of sudden colicky abdominal pain, a palpable right-sided sausage-shaped mass, and currant-jelly stools, the clinical manifestations in adults are nonspecific, often delaying diagnosis. Here, we report a case of adult intussusception caused by an ascending colon mucinous adenocarcinoma.

Case Report

A 74-year-old male with a history of chronic hepatitis B, hypertension, diabetes mellitus, paroxysmal atrial fibrillation, and cerebral infarction under anticoagulant therapy presented with abdominal pain and fever for one day. His past medical history included choledocholithotomy and cholecystectomy for common bile duct stones, multiple endoscopic retrograde cholangiographies with stone extraction for recurrent choledocholithiasis, and hepatocellular carcinoma (Barcelona Clinic Liver Cancer stage 0) treated with radiofrequency ablation in December 2017.

On arrival at the emergency department, he reported persistent abdominal pain with vomiting. Physical examination revealed right-sided abdominal tenderness. Laboratory studies showed elevated liver enzymes: alkaline phosphatase (ALP) 253 IU/L and gamma-glutamyl transferase (GGT) 209 IU/L. Abdominal computed tomography demonstrated colonic intussusception at the hepatic flexure.

The patient underwent right hemicolectomy, which revealed a 4-cm ulcerated mass at the hepatic flexure. Histopathological examination confirmed moderately differentiated mucinous adenocarcinoma of the hepatic flexure, with invasion into pericolic tissue but no lymph node metastasis. Resection margins were free of tumor.

Discussion

Adult intussusception is rare and can be classified into enteric, ileocolic, colonic, or ileocecal types, depending on its location. Unlike pediatric cases with the characteristic triad of abdominal pain, vomiting, and bloody stools, adult presentations are usually vague, most commonly involving intermittent abdominal pain. This nonspecific presentation frequently delays diagnosis.

Colonic or ileocolic intussusception in adults is usually associated with tumors. Computed tomography is the most accurate diagnostic tool, with the characteristic "target" sign.

Surgical management is the treatment of choice for adult colonic intussusception. Reduction prior

to resection is generally avoided when malignancy is suspected, to prevent intraluminal tumor seeding or venous dissemination.

Conclusion

Colon intussusception is rare in adults and can present with vague symptoms. Abdominal pain can be the presentation. CT scanning is the most useful diagnostic radiologic method. Radical operation without preoperative reduction was suggested because of the high incidence of malignant lesions in the adult group.

病例報告 114_C158

一名微小病變腎病患者對類固醇治療反應良好，惟在劑量減少後出現復發。

A case of minimal change disease, good response to steroid but relapse after dose tapering

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Introduction

Minimal change disease (MCD) is a main cause of nephrotic syndrome in children (about 90%) and some adults (about 10%). Most MCD cases happen without a known cause (idiopathic), but some are secondary to drugs, cancers, infections, or allergies. In adults, a kidney biopsy is needed to confirm the diagnosis. Treatment aims to reduce protein in the urine and achieve remission, mostly by using immunosuppressive drugs like steroids (glucocorticoids).

Case Report

A 54-year-old woman with a history of minimal change disease (MCD) and resolved hepatitis B was admitted for persistent heavy proteinuria. About a year ago, she had leg and eyelid swelling. Tests showed very high urine protein levels, and a kidney biopsy confirmed MCD. Despite steroid and immunosuppressive treatment, proteinuria persisted.

Her condition worsened in March 2025, with increased proteinuria and swelling. Steroid doses were increased, and she received diuretics and albumin, but symptoms did not improve (UPCR: 31,088.14 mg/g by April 3, 2025). She was admitted on April 7, 2025, for high-dose intravenous steroids.

During admission, poor medication adherence was discovered. After treatment with high-dose steroids and albumin infusions, proteinuria (UPCR: 18,876.93mg/g by April 10, 2025) and serum albumin improved. No significant side effects occurred. Steroids were tapered gradually, and she was discharged in stable condition on April 14, 2025, with outpatient follow-up.

Discussion

Minimal Change Disease (MCD) usually has no known cause, but relapses can happen due to infections, allergies, some medicines (like NSAIDs, lithium, antibiotics), or cancers (like lymphoma). It can also be linked to autoimmune diseases, IgA nephropathy, vaccines, or viral infections. Younger patients and those with high protein levels in urine at diagnosis are more likely to relapse. Treatment often starts with steroids (glucocorticoids), but long-term use needs careful monitoring for side effects. If steroids don't work well, other medicines like cyclosporine, cyclophosphamide, mycophenolate mofetil, or rituximab may be used.

Conclusion

Minimal change disease (MCD) is a common cause of nephrotic syndrome in children (about 90%) and less common in adults (about 10%). Treatment aims to reduce protein in the urine and bring remission. Steroids (glucocorticoids) are usually the first treatment. However, relapses often happen. Managing causes, watching for steroid side effects, and using other immunosuppressive drugs can help control relapses.

病例報告 114_C159

產氣性膿胸在 A 型流感與 COVID-19 合併感染後的罕見病例

An Unusual cause of Emphysematous Empyema: *Peptostreptococcus micros* Following Influenza A and COVID-19 Coinfection

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Introduction

Pleural infection remains a serious complication of pneumonia, with mortality approaching 20% despite advances in antimicrobial therapy and surgical management. Anaerobic organisms, although less commonly reported, are increasingly recognized as significant pathogens in community-acquired empyema, particularly in elderly or debilitated patients. We describe a case of empyema thoracis caused by Peptostreptococcus micros in a patient with preceding Influenza A and SARS-CoV-2 infection, managed successfully with prompt chest drainage, video-assisted thoracoscopic surgery (VATS) with decortication, and tailored antimicrobial therapy.

Case Report

A 77-year-old woman presented with fever and progressive dyspnea 2 weeks after COVID-19 and a new Influenza A infection. She required high-flow nasal cannula for hypoxemia. Chest radiography revealed a large right pleural effusion, and thoracentesis yielded frank pus. Despite empiric piperacillin–tazobactam and linezolid, she developed respiratory failure. Computed tomography demonstrated hydropneumothorax with a lenticular shape of fluid collection and pleural thickening.

Urgent video-assisted thoracoscopic surgery with decortication revealed extensive pleural rind and purulent fluid. Pleural cultures grew *Peptostreptococcus micros*, an anaerobic Gram-positive coccus, while blood cultures were negative. By hospital day 13, the patient was ambulating with assistance and her chest radiograph showed satisfactory lung re-expansion with minimal residual space. She was discharged on oral amoxicillin-clavulanate for 14 days. At outpatient follow-up, she remained clinically stable.

Discussion

Post-viral bacterial infections are well described following Influenza and, more recently, SARS-CoV-2. The most common pathogens include *Streptococcus pneumoniae*, *Staphylococcus aureus* (often MRSA), and *Haemophilus influenzae*. These organisms account for the majority of secondary pneumonia and empyema cases. In contrast, anaerobes are less frequently identified but may be underrecognized due to culture challenges.

Peptostreptococcus micros is an uncommon cause of pleural infection, more often associated with dental, oropharyngeal, and aspiration-related disease. In large epidemiologic series, anaerobes account for up to one-third of community-acquired empyema, but *P. micros* specifically represents only a small fraction of isolates. Its identification in this patient underscores the importance of obtaining anaerobic cultures in pleural infection, especially in elderly patients with post-viral airway disruption and possible aspiration risk.

This case emphasizes that while typical post-viral empyema pathogens remain predominant, rare anaerobic bacteria such as *P. micros* can complicate the course. Recognition of these organisms is critical for appropriate antimicrobial selection and highlights the complex microbiology of post-viral pleural infection.

病例報告 114_C160

紅斑性狼瘡合併皮膚型 *Nocardia farcinica* 感染之病例報告與系統性文獻回顧

Cutaneous *Nocardia farcinica* in SLE and a Systematic Review of Reported Infections

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Introduction

Nocardiosis is a rare but potentially serious opportunistic infection, particularly in immunocompromised individuals. Patients with systemic lupus erythematosus (SLE) may be at increased risk due to underlying immune dysregulation and immunosuppressive therapy. We present a case of cutaneous Nocardia farcinica infection in an SLE patient and conduct a systematic review to better understand its clinical characteristics and possible risk factors.

Case Report

A 35-year-old woman with SLE and Sjögren's syndrome developed a progressive, painful swelling over her right forearm. Initial antibiotic therapy was ineffective. Fine-needle aspiration revealed gram-positive branching bacilli, later confirmed as Nocardia farcinica. Treatment with sulfamethoxazole/trimethoprim and a single dose of amikacin led to clinical improvement and resolution.

Discussion

A systematic search of PubMed and Embase was conducted through May 2025. Studies were included if they reported individual cases of nocardiosis in SLE patients. Data on demographics, immunosuppressive therapy, infection site, species, treatment, and outcome were extracted. Univariate and multivariate analyses were performed to identify factors associated with mortality. Sixty-two studies were included, totaling 69 patients. Most were female (76.8%) with a mean age of 39.4 years. Corticosteroid use was common (95.7%). Disseminated, pulmonary, and CNS infections predominated. The mortality rate was 14.5%. Male sex was the only variable significantly associated with mortality (OR 7.45, $p = 0.0104$). No specific drug or infection site showed a clear link to death.

Conclusion

Nocardiosis should be considered in SLE patients with atypical infections. Male sex may be associated with worse outcomes. Further studies are needed to clarify risk factors and guide management.

病例報告 114_C161

家族性 DSP 基因突變相關擴張型心肌病變：病例報告

Familial DSP Gene Mutation–Associated Dilated Cardiomyopathy: A Case Report

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Introduction

Desmoplakin (DSP) is a desmosomal protein essential for myocardial integrity and electrical conduction. Pathogenic DSP mutations were first reported in Carvajal syndrome and later in arrhythmogenic right ventricular cardiomyopathy, but growing evidence linked heterozygous variants to dilated cardiomyopathy (DCM) with LV dilation, fibrosis, and arrhythmia. This diversity presentation highlighted the genetic heterogeneity of DSP cardiomyopathy. Here, we reported a family with progressive heart failure and ventricular arrhythmia carrying a truncating DSP mutation (c.448C>T; p.Arg150*).

Case Report

A 53-year-old woman with no previous medical history developed exertional dyspnea in 2020, at which time echocardiography demonstrated mildly reduced left ventricular systolic function. No specific treatment was initiated, and she remained under observation. Over the subsequent years, she experienced progressive exertional dyspnea and intermittent palpitations. Electrocardiography revealed sinus rhythm with frequent premature ventricular complexes, and ambulatory Holter monitoring demonstrated more than 1,800 ventricular ectopic beats per day with non-sustained ventricular tachycardia. Laboratory testing revealed mildly elevated NT-proBNP levels (322 pg/mL). With further progression of dyspnea in 2024, serial echocardiography demonstrated progressive left ventricular dilation with a decline in systolic function, as the ejection fraction decreased from 45% in 2020 to 30% in 2024. Cardiac magnetic resonance imaging revealed biventricular dysfunction, with a right ventricular ejection fraction of 28.35%. Mid-wall late gadolinium enhancement was observed in the interventricular and posterolateral walls of the left ventricle, consistent with diffuse myocardial fibrosis. Guideline-directed therapy with bisoprolol, sacubitril / valsartan, and dapagliflozin was initiated, but spironolactone was avoided due to a hypersensitivity reaction manifested as rash, leading to partial symptomatic improvement. Family screening identified four additional carriers of the same variant, aged 28 to 59 years, all showing consistent cardiac involvement characterized by mild LV dilation, slightly reduced LV systolic function, and frequent PVCs. No cutaneous or hair abnormalities were observed in any case. Genetic analysis confirmed a heterozygous DSP p.Arg150* nonsense variant in all affected family members.

Discussion

DSP cardiomyopathy has been widely reported in the literature, with most cases demonstrating predominant LV involvement. Our case shared similar features, and notably some younger family members had already developed LV dilation or mild systolic impairment. While multiple DSP variants have been reported in Western cohorts, documentation from Asian populations remains

limited. To date, only one case report from Taiwan had been published, describing a different DSP mutation (c.6384delG) that presented with dilated cardiomyopathy but without cutaneous or hair abnormalities. An Italian case involving a 29-year-old male carried the same p.Arg150* variant as identified in our family and likewise exhibited DCM as the predominant phenotype. Our case expanded the geographic spectrum of DSP cardiomyopathy, provided evidence for p.Arg150* as a pathogenic variant, and highlighted how genetic confirmation could clarify etiology, guide risk stratification, and enable cascade screening in families with unexplained DCM.

Conclusion

We reported a 53-year-old woman with progressive dilated cardiomyopathy and ventricular arrhythmias associated with DSP variant, with four additional relatives carrying the same mutation and showing similar cardiac expression. This case highlighted the importance of desmosomal mutations in patients with unexplained DCM.

病例報告 114_C162

以橋本氏甲狀腺炎合併自體免疫萎縮性胃炎和維生素 B12 缺乏的第 IIIB 型自體免疫性多內分泌腺病症候群：案例報告

Autoimmune Polyglandular Syndrome Type IIIB Presenting with Hashimoto's Thyroiditis and Autoimmune Atrophic Gastritis with B12 Deficiency: A Case Report

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Introduction

Autoimmune polyglandular syndrome type IIIB (APS IIIB) is defined by the coexistence of autoimmune thyroiditis with other autoimmune diseases targeting the gastrointestinal system, such as autoimmune atrophic gastritis or autoimmune hepatitis. We report a case of APS IIIB in a patient with a known history of Hashimoto's thyroiditis who was subsequently diagnosed with autoimmune atrophic gastritis and vitamin B12 deficiency.

Case Report

A 67-year-old male with a history of Hashimoto's thyroiditis (anti-thyroid peroxidase antibodies: 524.36 IU/mL) has been on a stable daily dose of 150 µg levothyroxine for several years. During a routine outpatient visit, he complained of tongue pain with associated loss of papillae, as well as progressive numbness and pain in both soles. These symptoms prompted an evaluation that revealed a low blood vitamin B12 level of 180 pg/mL (normal range: 211-911 pg/mL). Subsequent esophagogastroduodenoscopy showed findings consistent with atrophic gastritis, localized to the gastric body. Further serological testing confirmed the diagnosis with a positive gastric parietal cell antibody (1:160). The patient was diagnosed with autoimmune atrophic gastritis and vitamin B12 deficiency. Following vitamin B12 supplementation, his tongue pain and limb numbness quickly improved.

Discussion

We describe a case of APS IIIB, which presented with both Hashimoto's thyroiditis and autoimmune atrophic gastritis, leading to vitamin B12 deficiency. APS III is defined by the coexistence of autoimmune thyroid disease with other autoimmune conditions, excluding Addison's disease. This syndrome is divided into four subtypes based on the organ systems affected. Coexisting autoimmune diseases are found in nearly 20% of patients with autoimmune thyroid disease. Specifically, the prevalence of anti-parietal cell antibodies in patients with Hashimoto's thyroiditis is reported to be as high as 25%, and autoimmune atrophic gastritis may occur in up to one-third of these patients. Atrophic gastritis can lead to non-specific symptoms such as weakness, unexplained weight loss, and signs of pernicious anemia or iron deficiency anemia. These symptoms can be easily mistaken for those of hypothyroidism, leading patients to question the adequacy of their levothyroxine dosage. Therefore, it is crucial to carefully evaluate persistent or new symptoms even after thyroid function has been normalized with treatment, as they may indicate the presence of a coexisting autoimmune disease.

Conclusion

Given the high prevalence of coexisting conditions, APS III may be more common than is currently recognized. We conclude that when treating patients with autoimmune thyroid disease, other autoimmune conditions should be carefully investigated, particularly when patients present with non-specific or persistent symptoms despite adequate thyroid hormone replacement.

病例報告 114_C163

內科藥物成功治療免疫低下病人之巨大前列腺膿瘍

Successful medical treatment of a huge prostate abscess in a 59-year-old immunocompromised host

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Introduction

Prostatic abscesses are uncommon but potentially life-threatening urologic infections. They typically arise as a complication of acute bacterial prostatitis, especially in immunocompromised patients such as those with diabetes or liver cirrhosis. The mainstay of treatment is prolonged antibiotic therapy combined with surgical or percutaneous drainage, especially for abscesses >2 cm. However, in select patients, conservative medical management alone may suffice. We present a rare case of a huge prostate abscess in an immunocompromised host that was successfully managed without drainage.

Case Report

A 59-year-old male with a history of untreated chronic hepatitis B virus (HBV) infection and newly diagnosed type 2 diabetes mellitus presented with a 2-week history of dysuria, frequency, fever, and chills. He also reported anorexia and weight loss (>5 kg). On presentation, he was febrile and hemodynamically unstable, with leukocytosis and pyuria. Abdominal CT revealed multiple ring-enhancing lesions in the right prostatic lobe (up to 37.4×56.2 mm), consistent with prostatic abscess. He was admitted to the medical ICU with septic shock secondary to complicated acute bacterial prostatitis.

Empiric IV Brosym was initiated. Blood and urine cultures yielded *Klebsiella pneumoniae*. He was hypoalbuminemic (serum albumin 1.6 g/dL) and malnourished, prompting daily IV albumin supplementation. A flare of chronic HBV was diagnosed based on markedly elevated HBV DNA and bilirubin (T-bil 25 mg/dL), and entecavir therapy was started. Due to severe jaundice and nutritional compromise, transurethral resection of the prostate (TURP) for drainage was deferred. In the second week, the patient developed severe leukocytosis (WBC 41,780/μL) and anemia (Hb 4.4 g/dL), with positive fecal occult blood tests. Endoscopy revealed erosive gastritis and esophagitis, managed with PPI therapy. Antibiotics were escalated to IV ertapenem, and transfusions were administered.

Repeat CT on Day 11 demonstrated interval reduction in abscess size (to 12.6×22.0 mm). Given clinical improvement and decreased abscess volume, surgical drainage was further deferred. Over the following week, the patient's jaundice and nutritional markers improved significantly (T-bil to 2.5 mg/dL, Alb to 2.5 g/dL). Urinalysis normalized, and leukocytosis resolved.

The patient was discharged on Day 19 with close outpatient follow-up planned with gastroenterology, nephrology, and urology teams. TURP was to be reconsidered following full nutritional recovery.

Conclusion

This case demonstrates that even large prostatic abscesses in immunocompromised patients may be managed conservatively with appropriate antibiotic therapy and close monitoring. Early identification of clinical risk factors, microbiologic confirmation, and radiologic follow-up are crucial in guiding management decisions.

病例報告 114_C164

De Winter 心電圖模式：罕見但致命的 STEMI 表徵

De Winter ECG Pattern: A Rare STEMI-Equivalent Not to Be Missed

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Introduction

The De Winter electrocardiogram (ECG) pattern was first described in 2008 by Dutch cardiologist Robert J. de Winter. It is characterized by the absence of ST-segment elevation, but instead shows 1-2mm upsloping ST-segment depression at the J-point in precordial leads (V1-V6), accompanied by tall, positive, symmetrical T-waves and 1-2mm ST elevation in lead aVR. This rare presentation occurs in approximately 2%-3.4% of patients with anterior myocardial infarction and is considered a ST-segment elevation myocardial infarction (STEMI)-equivalent, requiring immediate reperfusion therapy as per acute coronary syndrome guidelines.

Case Report

A 50-year-old male with no smoking history or known comorbidities presented to the emergency department with chest pain after exercise. The pain was described as progressive tightness lasting over 5 minutes, unrelieved by rest, and accompanied by dizziness and cold sweating. He denied previous similar episodes. On arrival, vital signs were normal, and physical examination revealed no significant findings apart from acute distress. An ECG obtained within 9 minutes showed sinus rhythm, tall symmetrical T-waves, upsloping ST-segment depression at the J-point in precordial leads, ST elevation in lead aVR, and no ST elevation in precordial leads. The cardiologist was promptly informed, and emergent percutaneous coronary intervention (PCI) was initiated based on the impression of De Winter ECG pattern. Angiography revealed 99% critical stenosis from the ostium to the proximal segment of the left anterior descending artery (LAD). Successful PCI and drug-eluting stent placement were performed. Post-PCI ECG showed normalized ST-segments, confirming successful revascularization. Cardiac biomarkers were later found to be elevated. Further workup revealed hypercholesterolemia. The patient was discharged on dual antiplatelet therapy, beta-blocker, ACE inhibitor, and statin.

Discussion

The De Winter ECG pattern is more commonly observed in younger, male patients with hypercholesterolemia compared to the classic STEMI pattern. In this case, recognition of the pattern as a STEMI-equivalent, without waiting for cardiac biomarker results, led to a door-to-wire time of 67 minutes, well below the recommended 90-minute threshold. Early identification was key to achieving optimal clinical outcomes.

Conclusion

The De Winter ECG pattern is a rare but critical marker of acute coronary artery occlusion, often involving the LAD. Prompt recognition in this patient led to successful intervention, preventing severe morbidity or mortality. This case underscores the importance of rapid identification of this

high-risk ECG pattern by clinicians.

病例報告 114_C165

病例報告：免疫療法的長尾效應在一位廣泛期小細胞肺癌病人接受 Atezolizumab 合併化療

Case Report: The Tail Effect of Immunotherapy in A Patients with Extensive-Stage Small Cell Lung Cancer Treated by Atezolizumab plus Chemotherapy

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Introduction

Small cell lung cancer (SCLC) is an aggressive cancer known for its rapid growth and early spread to distant sites in the body. When the cancer has extended beyond one side of the chest and cannot be effectively targeted with a single radiation field, it is classified as extensive-stage SCLC (ES-SCLC). The majority of SCLC cases are diagnosed as extensive-stage disease upon presentation, contributing to a poor prognosis. An analysis of nationwide data in Taiwan spanning from 2010 to 2016 revealed a median survival of 8.7 months and a five-year survival rate of less than 2% among patients with ES-SCLC.

Case Report

A 66-year-old male heavy smoker presented with a persistent cough, dyspnea, and swelling in the upper trunk and face. The chest radiograph revealed a mass in the right lower lung with hilar enlargement. Computed tomography (CT) identified a mass in the right middle lobe with extensive mediastinal lymphadenopathy. Metastatic nodules were discovered in the frontal lobe on brain MRI, and a whole-body bone scan showed multiple metastases in the thoracic and lumbar spines as well as ribs. A CT-guided lung biopsy confirmed the small cell carcinoma.

The patient underwent treatment with atezolizumab in combination with chemotherapy of etoposide and cisplatin for six 21-days cycles. This was followed by maintenance therapy with atezolizumab for a total of 30 cycles. After treatment, his symptoms and radiological abnormalities resolved. Subsequent chest CT scans, brain MRI, and bone scans indicated no evidence of disease. However, the patient experienced a grade II immune-related adverse event (irAE) of adrenal insufficiency, which was managed with daily cortisone supplementation. Following two years of immunotherapy, the patient received regular surveillance and remained disease-free for over five years.

Discussion

When platinum-based chemotherapy is combined with etoposide, ES-SCLC typically has a median overall survival of 8 to 10 months. The addition of immunotherapy has significantly enhanced outcomes and has become a standard treatment. Results from clinical trials such as IMpower133 have shown that the addition of atezolizumab with chemotherapy can prolong median survival to 12.3 months compared to 10.3 months, while in the CASPIAN trial, the addition of durvalumab to chemotherapy also increased median survival to 13.0 months from 10.3 months. Although the addition of immunotherapy has the potential to improve survival outcomes in ES-SCLC, challenges persist in managing this aggressive cancer. A subset of patients experience long-lasting benefits and disease control, leading to the observation of the “tail effect.” The mechanisms driving this

phenomenon are still under investigation, and it may involve the development of a sustained immune response against cancer cells. Unlike in non-small cell lung cancer where PD-L1 expression is a reliable biomarker for predicting response to immunotherapy, the role of PD-L1 expression in SCLC remains uncertain. Further research is necessary to determine effective biomarkers for identifying responders to immunotherapy in ES-SCLC.

Conclusion

This case underscores the potential of immunotherapy to achieve long-term survival in ES-SCLC. The patient has remained disease-free for over five years, demonstrating the presence of the tail effect in immunotherapy.

病例報告 114_C166

成人孤立性幼年型息肉：四例病例系列病例報告與文獻回顧

Solitary Juvenile Polyp: A Case Series of Four Adult Patients and Literature Review

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Introduction

Juvenile polyps are the most common benign hamartomatous polyps in children, usually presenting between ages 2–10 years with hematochezia or mucus in stool. Unlike Juvenile Polyposis Syndrome (JPS), solitary juvenile polyps occur sporadically without family history or genetic predisposition and carry negligible malignant risk. While frequently encountered in pediatrics, their occurrence in adults is exceedingly rare, often misinterpreted as adenomas or neoplastic lesions. Comprehensive case series in adults are lacking, and accurate recognition is crucial to avoid overtreatment or unnecessary genetic testing.

Case Report

From January 2013 to March 2025, 3,587 colonoscopies were performed at our institution, and four adult patients were diagnosed with solitary juvenile polyps. Case 1: a 42-year-old man with abdominal pain and hematochezia had a 1.8 cm pedunculated sigmoid polyp (NBI International Colorectal Endoscopic, NICE type 2). Case 2: a 63-year-old woman with bowel habit changes had a 0.6 cm pedunculated descending colon polyp with chicken skin-like mucosa (NICE type 2). Case 3: a 60-year-old woman with diarrhea had a 0.6 cm semi-pedunculated sigmoid polyp (NICE type 1). Case 4: a 36-year-old man with hematochezia had a 0.8 cm sessile rectal polyp (NICE type 1). All lesions exhibited hyperemic or eroded surfaces, were completely resected endoscopically, and confirmed histologically as juvenile polyps without dysplasia. None had family history or multiple polyps.

Discussion

Solitary juvenile polyps in adults are rare, with an estimated incidence of less than 0.1%. Their colonoscopic appearance, often erythematous, granular, or eroded with possible chicken skin-like mucosa, may mimic adenomas and complicate diagnosis. Histology typically shows cystically dilated mucin-filled glands, inflammatory stroma, and absence of atypia, confirming their benign nature. Advanced imaging modalities such as narrow-band imaging, chromoendoscopy, and pit pattern analysis may improve pre-pathologic recognition, although histology remains the diagnostic gold standard. Differentiation from Juvenile Polyposis Syndrome (JPS) is crucial, since solitary juvenile polyps are isolated, sporadic, and non-hereditary, without upper gastrointestinal involvement or increased cancer risk, and therefore do not require genetic testing or long-term surveillance.

Conclusion

This case series highlights four rare adult presentations of solitary juvenile polyps. These benign, sporadic lesions can be completely cured by endoscopic resection and should be distinguished

from adenomas and hereditary polyposis syndromes to prevent unnecessary interventions. Awareness of their clinical, endoscopic, and pathological features enables accurate diagnosis and optimal management, ensuring excellent prognosis without need for routine follow-up.

病例報告 114_C167

全身性硬化症相關間質性肺病的前上葉徵象：病例報告

Anterior Upper Lobe Sign in Systemic Sclerosis-Associated Interstitial Lung Disease: A Case Report

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Introduction

Systemic sclerosis (SSc) is an autoimmune disease characterized by immune dysregulation and progressive fibrosis affecting the skin and internal organs. Interstitial lung disease (ILD) is a frequent and serious complication in SSc patients. High-resolution computed tomography (HRCT) is essential for detecting SSc-ILD, typically revealing non-specific interstitial pneumonia or usual interstitial pneumonia patterns. The anterior upper lobe sign, demonstrated by upper lobe volume loss with anterior displacement of bronchi and vessels, is a significant radiologic feature indicative of early fibrotic changes in autoimmune diseases. Here, we present a case of SSc-ILD patient exhibiting this distinctive image finding.

Case Report

A 62-year-old woman presented with a productive cough, dyspnea, and tachycardia for 2 weeks. Laboratory tests indicated elevated level of brain natriuretic peptide. Chest radiograph showed cardiomegaly and diffuse bilateral lung infiltrates. Cardiac echocardiography revealed moderate mitral regurgitation, severe tricuspid regurgitation, reduced left ventricular ejection fraction (39.1%), and pulmonary artery hypertension. Thoracentesis demonstrated a transudative pleural effusion. Following diuretic therapy and intensive care, the patient's symptoms and pulmonary edema improved; however, a subsequent radiograph raised suspicion of pulmonary fibrosis. HRCT displayed fibrotic lesions, traction bronchiectasis, and honeycombing in both lungs, predominantly in the upper lobes, and with fibrosis extent exceeding 10%. The anterior upper lobe sign was also noted. Pulmonary function tests revealed a restrictive pattern with a forced vital capacity (FVC) of 52% of the predicted value. Physical examination revealed skin thickening of the fingers, fingertip lesions, and Raynaud's phenomenon. Rheumatologic blood tests showed positive antinuclear antibody and anti-Scl-70 antibodies, consistent with the American College of Rheumatology (ACR)/European League Against Rheumatism (EULAR) diagnostic criteria for systemic sclerosis. The diagnosis of SSc-ILD was confirmed, and the patient was initiated on treatment with Azathioprine and Nintedanib.

Discussion

Systemic sclerosis (SSc) is an autoimmune disease that affects multiple organs, including the lungs, where a significant number of patients develop progressive pulmonary fibrosis, a primary cause of mortality in this population. Early detection of pulmonary fibrosis in SSc is crucial for prompt treatment initiation and ongoing monitoring to prevent disease progression and enhance patient outcomes.

The anterior upper lobe sign is a distinctive radiological feature seen on HRCT in patients with

connective tissue disease-related ILD, such as rheumatoid arthritis, systemic sclerosis, and dermatomyositis. This sign is characterized by upper lobe involvement, especially in the anterior segments, exhibiting predominant ground-glass opacities, reticulation, and honeycombing. Its presence is highly specific for connective tissue disease-related ILD, aiding in the differentiation from other forms of ILD.

The SENSCIS trial, a phase 3 randomized controlled study, demonstrated that the antifibrotic agent Nintedanib significantly reduced the annual decline in FVC in patients with SSc-ILD. This underscores the importance of early therapeutic intervention and its potential to improve outcomes in these individuals.

Conclusion

The anterior upper lobe sign is a highly specific HRCT finding in connective tissue disease-related ILD, signaling early fibrotic lung involvement. Recognizing pulmonary fibrosis in conjunction with clinical and serologic information enables precise diagnosis and timely initiation of treatment for patients with SSc-ILD.

病例報告 114_C168

多發性骨髓瘤伴隨骨骼和肺部併發症：病例報告

Multiple Myeloma with Skeletal and Pulmonary Complications: A Case Report

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Introduction

Multiple myeloma (MM) is a malignant proliferation of plasma cells leading to monoclonal protein production, osteolytic bone lesions, anemia, renal impairment, and increased infection risk. We report a 67-year-old man with newly diagnosed MM complicated by extensive bone disease, myelomatous pleural effusion, and well responses to radiotherapy and VTd chemotherapy.

Case Report

A 67-year-old man with no underlying disease presented in February 2025 with severe chest and back pain following heavy lifting. Chest CT showed rib fractures and bulky osteolytic bone lesion of 6-8th ribs. And CT-guided rib biopsy confirmed plasma cell myeloma (CD138+, CD56+, cyclin D1+) and the following BM pathology favored multiple myeloma, CD138(+), atypical plasmacytoid cells in the marrow with lambda light chain restriction, compatible with multiple myeloma. After work up, under the diagnosis of multiple myeloma, IgA, lambda type, R-ISS stage III, FISH standard risk(No t(4,14); t(14,16) and TP53 deletion), complicated with multiple bone lesions and multiple pathological fractures (ribs, clavicle, T3, T7 vertebrae) causing severe pain and restrictive lung disease.

On March 1, cytology of right pleural effusion revealed myelomatous pleural effusion. In April 2025, the patient was admitted for progressive dyspnea. Thoracentesis yielded 450 mL turbid fluid. Cytology again demonstrated atypical plasmacytoid cells admixed with mesothelial cells and lymphocytes. Immunohistochemistry showed numerous lambda chain–positive plasma cells, scattered kappa-positive cells, and CD138 expression, confirming malignant pleural effusion. Radiotherapy was initiated for MM bone disease of ribs on May 6. And the follow of CT on August 14 showed decreased sizes and numbers as compared with previous CT and one deformity much relieved. Also the M protein level decrease from 4.6 g/dl to 0.1 g/dl.

Discussion

Myelomatous pleural effusion is a rare, ominous complication of MM, this entity occurs in approximately 1% of MM cases.Clinically, patients may present with dyspnea, chest pain, and respiratory compromise due to large or recurrent effusions. And radiotherapy of MM Bone Disease mainly used for osteolytic lesions in the spine, pelvis, and long bones, and our patient ribs lesion also showed well responses to the therapy.

Conclusion

This case highlights the rare occurrence of myelomatous pleural effusion in the setting of advanced multiple myeloma, underscoring its association with aggressive disease biology and poor prognosis. Despite the unfavorable presentation with extensive osteolytic lesions,

pathological fractures, and malignant pleural effusion, our patient demonstrated a favorable response to combined radiotherapy and VTd chemotherapy. This emphasizes the potential role of multimodal treatment strategies in achieving symptom relief and disease control, even in patients with high disease burden.

病例報告 114_C169

潛伏於泌尿道的殺手：Pseudomonas aeruginosa 造成眼內炎合併腦膿瘍的罕見病例

Case Report: A Silent Killer in the Urinary Tract: A Rare Case of Pseudomonas aeruginosa Endophthalmitis with Brain Abscess

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Introduction

Pseudomonas aeruginosa is an opportunistic pathogen capable of causing severe infections with high levels of antibiotic resistance. Although meningitis and brain abscesses due to P. aeruginosa are rare, their morbidity and mortality remain significant.

Case Report

A 78-year-old woman with retroperitoneal liposarcoma, invasive ductal carcinoma of the breast, and cervical carcinoma in situ presented with malaise and dysuria. She was diagnosed with urinary tract infection with suspected sepsis and was started on empiric antibiotics. However, she developed respiratory distress and hemodynamic instability during hospitalization.

During admission, bilateral yellow-red ocular discharge was noted, and ophthalmology diagnosed left endophthalmitis. Urine culture subsequently grew P. aeruginosa, leading to escalation to imipenem, vancomycin, and ciprofloxacin. MRI revealed a brain abscess, meningitis, and a left subdural hematoma. Neurosurgery advised conservative management.

The patient later developed sepsis, and cefepime was initiated for better CNS coverage. Fever improved, and follow-up MRI showed partial shrinkage of multiple abscesses, improved meningitis, and resolving endophthalmitis. Despite these improvements, she remained hypotensive and developed renal failure with oliguria. Despite high-dose vasopressors and diuretics, renal function worsened. Nephrology recommended initiating Continuous Venovenous Hemofiltration (CVVH) to correct acidosis and azotemia; however, the procedure was complicated by clotting. She deteriorated with refractory shock and coma and was pronounced dead.

Discussion

Our patient presented with endophthalmitis and a brain abscess. Given the positive urine culture for P. aeruginosa, which was the only organism isolated, we suspect that P. aeruginosa was the causative agent for both ocular and central nervous system (CNS) infections.

For serious P. aeruginosa infections, particularly in cases with a high risk of antimicrobial resistance or in patients where inappropriate therapy would significantly increase mortality, empiric treatment should involve two agents from different classes with demonstrated in vitro activity. The preferred regimen typically includes a beta-lactam as the primary agent and an aminoglycoside as the secondary agent. Once P. aeruginosa is confirmed and antimicrobial susceptibility results are available, therapy should be narrowed to a single active antipseudomonal agent. Exceptions where combination therapy might still be warranted include neutropenia with bacteremia, endocarditis, failure to respond to initial therapy, or cases of multidrug resistance. However, there is no clear evidence that combination therapy provides

additional benefit over monotherapy.

Conclusion

This case highlights the challenges of treating disseminated P. aeruginosa infections, especially in elderly patients with multiple comorbidities. Despite appropriate antibiotic therapy, prognosis remains poor due to intrinsic resistance, limited therapeutic options, and the patient's frailty.

病例報告 114_C170

肺炎克雷伯氏菌引起之肝膿瘍破裂併發腹腔積氣的案例報告

Case Report: Ruptured Liver Abscess Presenting as Pneumoperitoneum Caused by Klebsiella pneumoniae

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Introduction

Pyogenic liver abscesses (PLA) are life-threatening infections often caused by enteric bacteria, particularly Klebsiella pneumoniae in East Asia. While common complications include sepsis and metastatic infections, spontaneous rupture into the peritoneal cavity with resultant pneumoperitoneum is exceedingly rare. We present a case of a ruptured gas-forming PLA caused by K. pneumoniae, presenting as pneumoperitoneum and requiring delayed surgical management.

Case Report

A 67-year-old man with a history of treated gastroesophageal reflux disease presented to the outpatient department with a 10-day history of exertional dyspnea and fatigue. Associated symptoms included bilateral lower extremity edema and decreased urine output. He was tachycardic and had a distended, tender abdomen. Labs revealed acute kidney injury (creatinine 3.7 mg/dL), leukocytosis (WBC 48,100/μL), and elevated AST (133 U/L).

A plain chest radiograph showed a sub-diaphragmatic free air, consistent with pneumoperitoneum. Contrast-enhanced computed tomography (CT) of abdomen revealed a 145-mm multi-loculated fluid collection with internal gas in the left lateral segment of the liver, with evidence of rupture into the peritoneal cavity.

Blood cultures grew K. pneumoniae. He was diagnosed with pneumoperitoneum secondary to spontaneous rupture of a gas-forming PLA. Due to high surgical risk, he initially received antibiotics and percutaneous drainage, but this failed due to ascites and complex loculations. Surgical drainage was performed on day 42, and he was discharged on day 72.

Discussion

PLA typically presents with fever, right upper quadrant pain and tenderness, and elevated liver enzyme levels. However, in this case, the patient initially presented with symptoms mimicking heart failure, including dyspnea, bilateral lower extremity edema, and decreased urine output. Laboratory findings revealed acute kidney injury and marked leukocytosis, raising suspicion for severe sepsis as the underlying cause.

A retrospective analysis of 140 cases of K. pneumoniae PLA and identified several significant risk factors for spontaneous rupture, including diabetes mellitus (100% vs. 62.1%, $P = 0.003$), larger abscess size (mean diameter 7.8 cm vs. 6.1 cm, $P = 0.043$), gas formation within the abscess (87.5% vs. 23.5%, $P < 0.001$), and involvement of the left hepatic lobe (50.0% vs. 16.5%, $P = 0.018$). These findings align closely with our patient's presentation—a gas-forming abscess in the left lateral segment in a high-risk individual—highlighting the relevance of these parameters in predicting rupture risk.

Spontaneous pneumoperitoneum is commonly associated with perforation of hollow viscera such as the stomach, small intestine, or colon. Pneumoperitoneum secondary to rupture of a PLA is extremely rare, with only a limited number of cases reported in the literature. In such cases, CT is crucial for distinguishing between differential diagnoses and confirming the presence of gas within the abscess cavity, as was evident in our patient.

Conclusion

This case illustrates an unusual presentation of a ruptured gas-forming PLA manifesting as pneumoperitoneum. The absence of typical abdominal symptoms initially led to diagnostic uncertainty. This case emphasizes the importance of early imaging, maintaining a high index of suspicion for atypical presentations of PLA rupture.

病例報告 114_C171

晚期甲狀腺癌的跨領域治療

Multidisciplinary management of advanced thyroid cancer: a case report

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Introduction

Although most patients with thyroid cancer have a favorable prognosis, there remains a subset with advanced disease. For large tumors or those invading critical structures, total thyroidectomy may severely compromise quality of life. Recently, tyrosine kinase inhibitors (TKIs) such as lenvatinib and dabrafenib/trametinib have been explored as neoadjuvant therapies to improve resectability. In addition to systemic target agents, we have also incorporated minimally invasive procedures, such as transarterial embolization (TAE) and radiofrequency ablation (RFA), into our neoadjuvant armamentarium.

Case Report

A 74-year-old woman was referred for right thyroid mass. Fine-needle aspiration was classified as Bethesda III, and subsequent core biopsy revealed a carcinoma with oncocytic features. Molecular testing showed the absence of a BRAF mutation. Computed tomography (CT) demonstrated a right thyroid tumor measuring 5.3 cm with fine calcifications, irregular margins, and evidence of extrathyroidal extension to trachea. Positron Emission Tomography (PET) confirmed locally advanced thyroid carcinoma and suggested lymph node metastases in the central compartment, mediastinum, and right hilar regions. Clinical staging at presentation was cT4aN1bM1, stage IVB. After discussion at our KCGMH thyroid cancer committee, the patient was deemed to have locally advanced thyroid cancer that was considered unresectable or associated with significant surgical morbidity.

Given the locally advanced tumor, TAE was performed to reduce tumor burden. One month later, TKI with lenvatinib was initiated. During the lenvatinib treatment course, RFA was subsequently performed. Follow-up PET scan revealed significant regression of the thyroid tumor, with a small focus of suspected residual viable tumor in the right lobe. Throughout the entire lenvatinib treatment duration (4.5 months), only grade 1 hypertension was observed, and no other adverse events were noted. The patient tolerated the minimally invasive procedures (TAE and RFA) well, without any complications. Following this series of neoadjuvant therapies, the patient underwent total thyroidectomy, and got R0 resection, with final pathology confirming papillary thyroid carcinoma, staged as ypT4aN0a, harboring an NRAS Q61R mutation.

Discussion

Surgery followed by radioactive iodine (RAI) remains the gold standard for most thyroid cancers. For patients with large, locally invasive differentiated thyroid cancer (DTC) in whom an R0 or R1 resection is unlikely without significant morbidity, the use of systemic multikinase inhibitors or TKIs, with or without immunotherapy, in the neoadjuvant setting has been reported. Novel

techniques such as TAE and RFA can complement systemic therapy for locoregional disease control. In our case, the patient was initially considered unresectable; however, after multidisciplinary neoadjuvant treatment with TKI, TAE, and RFA, she subsequently underwent surgery smoothly without any sacrifice of the aerodigestive tract, and an R0 resection was achieved. Given these remarkable results, further larger prospective studies are needed to determine which patients may be appropriate candidates for such a strategy.

Conclusion

This case highlights the potential value of integrating targeted therapy with minimally invasive procedures such as TAE and RFA in the neoadjuvant management of advanced thyroid cancer initially deemed unresectable. Nevertheless, treatment must be individualized, balancing efficacy and toxicity, and involving patients in shared decision-making.

病例報告 114_C172

肺鱗狀細胞癌併腸轉移，案例報告與討論

A rare case of lung squamous cell carcinoma with intestinal metastasis

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Introduction

Lung squamous cell carcinoma is a common cancer in Taiwan (Between 2000 ~ 2022, lung cancer incident rate in Taiwan is about 51 per 100,000 people and lung squamous cell carcinoma incident rate is around 8 per 100,000 people). Lung squamous cell carcinoma is more commonly seen in elderly, and has strong association with smoking history. Lung squamous cell carcinoma can be resected in early stage but due to mild or even asymptomatic presentation, plenty of patients are diagnosed at advanced stages. The most common sites of metastatic disease are regional lymph nodes, adrenal glands, brain, bone, and liver. Intestinal metastasis of the disease is rare and mostly related to poor prognosis. Intestinal metastasis of the disease is mainly diagnosed by imaging examination result.

Case Report

A 64-years-old-male with history of hypertension and type 2 diabetes mellitus had been suffered from epigastric dullness with formed black stool, sometimes even melena, for 1 months. Patient did not seek medical assistance till progressive dizziness noted for 2 days. The patient came to our emergency department for help and initial exam found left upper mass about 70*90mm in size. Bronchoscopy biopsy for lung tumor was done and pathology told squamous cell carcinoma. Panendoscopy and colonoscopy for tarry stool was done but found no bleeder. Deep enteroscopy was done for evaluation and found tumor lesion at small intestinal which pathology told spindle cell sacroma with rhabdomyosarcomatous differentiation. Tumor resection of small intestine was done for recurrent gastrointestinal hemorrhage and pathology told metastatic poorly differentiated carcinoma with sacomatous change that favor origin from lung. Patient then received chemotherapy with regiment of Gemcitabine and Cisplatin.

Discussion

Intestinal metastasis of lung cancer is rare (varies from around 5% to 10%). This case report is a patient with small intestinal metastasis without respiratory symptoms, such as chronic cough or dyspnea, despite huge primary tumor size. Instead, the symptom that drove patient to hospital is chronic black stool that result from intestinal metastasis, somewhat a type of typical presentation of lung cancer patient that metastatic disease is the first sign of disease. Due to advanced stage, surgical resection is not indicated for disease treatment yet patient had jejunum resection for bleeding control. Unlike lung adenocarcinoma, lung squamous cell carcinoma is lack of effective molecular target thus we gave patient standard chemotherapy regiment with Gemcitabine and Cisplatin for primary squamous lung cancer.

Conclusion

Treatment for advanced lung squamous cell carcinoma remains challenging due to lack of effective molecular target leading to limited treatment choice. And we need to take patient's individually due to varies condition caused by metastatic disease. For example, our patient had duodenum resection and thus we need to be aware of patient's nutrition status more than those who without duodenum resection. For poor prognosis and survival rate of advanced lung squamous cell carcinoma, early diagnosis is an important task to save patient life. We should work with other teams and use available policy instruments to boost screening participation.

病例報告 114_C173

慢性糖尿病足潰瘍偶然診斷鱗狀上皮細胞癌

Incidentally Diagnosed Squamous Cell Carcinoma in a Chronic Diabetic foot ulcer

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Introduction

Type 2 diabetes mellitus (T2DM) is commonly associated with complications such as peripheral arterial occlusive disease, neuropathy, and chronic foot ulcers. Malignant transformation within diabetic wounds is rare but has been reported, including squamous cell carcinoma (SCC) arising in non-healing ulcers. We hereby reported a case of squamous cell carcinoma arising in chronic diabetic foot ulcer.

Case Report

The patient is a 55-year-old male with poorly controlled T2DM (HbA1c 12.3%), peripheral arterial disease, hypertension, dyslipidemia, alcoholic liver disease, and long-standing tobacco and alcohol use presented with a chronic non-healing ulcer at the right first toe amputated stump. The wound had persisted for four years despite multiple surgical debridement, amputations of the first to third toes for osteonecrosis and infection, and several endovascular procedures for severe arterial stenosis.

He was admitted this time for glycemic optimization and wound management. Duplex ultrasonography showed moderate-to-severe stenosis of the right tibial arteries, and bone scan suggested osteomyelitis. Following endovascular revascularization with angioplasty and drug-coated balloon placement, debridement and local flap reconstruction were scheduled. Intraoperatively, the ulcer base appeared hypertrophic and suspicious for malignancy. Frozen section revealed SCC. Wide excision with sequestrectomy was performed. Final pathology confirmed invasive SCC and osteonecrosis of the first metatarsal.

Tumor staging with chest and abdominal CT, and PET/CT, showed no metastasis. Two weeks later, a second definitive excision was undertaken due to suspected residual disease. Frozen pathology demonstrated focal residual SCC nests with clear margins. Wide excision and rotational flap closure were performed. The patient recovered uneventfully and was discharged 10 days postoperatively. Outpatient follow-up was arranged for ongoing surveillance.

Discussion

Cutaneous SCC risk factors include ultraviolet radiation, chronic immunosuppression, and chronic ulceration. T2DM confers a chronic inflammatory and immunocompromised state, increasing susceptibility to malignancy. Persistent nonhealing diabetic foot ulcers provide a local environment of chronic inflammation, favoring neoplastic transformation. In our patient, poor medical compliance attributes to long-standing poor sugar control. Chronic non-healing diabetic foot ulcer provides a local inflammatory state, which trigger further neoplastic growth.

Conclusion

This case underlines the potential for squamous cell carcinoma to arise in chronic diabetic foot wounds and the importance of vigilance during debridement procedures. Malignancy should be kept in mind on inspecting a chronic poor-healing wound. Comprehensive oncologic evaluation, proper surgical margins, and optimized diabetic and vascular management are key to favorable outcomes.

病例報告 114_C174

罕見病例:在急性腸繫膜缺血合併小腸壞死時，肝門靜脈氣體或門脈氣腫視為不良預後指標

Ominous Signs of Hepatic Portal Venous Gas or Portal Pneumatosis in Acute Mesenteric Ischemia with Small Bowel Necrosis

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Introduction

Hepatic portal venous gas (HPVG) is a concerning imaging finding typically observed on plain abdominal radiographs or computed tomography (CT) scans. It usually suggests intestinal ischemia, which necessitates urgent abdominal surgery, with a mortality rate of around 75%.

Case Report

An elderly female patient in her 80s had a medical background that experiencing a sudden and severe lower abdominal pain that radiated to the periumbilical area, accompanied by cold sweating, that afternoon.

A chest radiograph, conducted to evaluate the source of her ongoing abdominal pain, showed no evidence of free air beneath the diaphragm. An abdominal radiograph revealed significant portal venous gas and gaseous distension of the small bowel. Initial clinical symptoms raised suspicions of intestinal ischemia, prompting imaging.

An urgent CT of the abdomen without contrast showed a distended stomach and enlarged loops of the jejunum and proximal ileum. Additionally, a dilated ascending colon with thickened bowel walls was observed. Extensive portal venous gas in the liver was present, along with gas accumulation in the superior mesenteric, jejunum, ileum, and portal veins.

However, severe abdominal tenderness accompanied by a peritoneal sign was observed. A central venous catheter (CVC) was inserted, and norepinephrine, an inotropic agent, was given, lowering her blood pressure to 80/40 mmHg. Consequently, acute intestinal ischemia with septic shock was strongly suspected. This prompted the first consultation with a general surgeon, leading to a laparotomy. The surgery uncovered distended and gangrenous necrosis in a 180-cm segment of the small bowel. We intended to resect about 200 cm of the small intestin, which included 100 mL of exudative ascites, while preserving 8 cm of the terminal ileum. The specimen was forwarded to the pathology department. Intraoperative findings revealed embolic occlusion of the mesenteric arteries. Considering her history of atrial fibrillation and a previous cerebrovascular accident, a cardiac embolus may have caused the acute mesenteric ischemia secondary to a thromboembolic event.

Discussion

HPVG is recognized as a significant imaging indicator, particularly in cases of acute mesenteric ischemia, with reported mortality rates as high as 75% to 80% . Recent articles suggest that abdominal CT enables early detection and diagnosis. This capability can lead to timely interventions and potentially enhance clinical outcomes in specific cases.

Conclusion

This case is notable for the successful surgical outcome in an elderly patient with a high cardiovascular risk. These features have rarely been reported in combination, providing new insights into ischemic bowel pathology in older people.

病例報告 114_C175

瀰漫性新型隱球菌感染併多器官侵犯：臨床診斷挑戰

Disseminated Cryptococcus neoformans Infection with Multi-organ Involvement: Diagnostic Challenges in Clinical Practice

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Introduction

Cryptococcus neoformans is an opportunistic fungal pathogen that may cause disseminated infection in immunocompromised patients. Central nervous system, pulmonary, and cutaneous involvement are frequently reported, and the disease remains associated with high morbidity and mortality. Cryptococcal antigen (CrAg) testing is highly sensitive and specific, but diagnostic pitfalls, such as false-negative results due to the prozone effect, may occasionally occur and delay clinical recognition. Here, we report a patient with disseminated C. neoformans infection with multi-organ involvement, and we discuss diagnostic challenges in cryptococcosis, including the potential impact of the prozone effect.

Case Report

A 52-year-old man with history of mesangial proliferative glomerulonephritis was on prednisolone 5 mg BID for 4 months. He developed right thigh cellulitis for 3 weeks and received Augmentin and clindamycin. He presented with generalized weakness and dyspnea. Laboratory tests showed acute kidney injury with hyperkalemia, and emergent hemodialysis was arranged.

One week later, he developed persistent fever and cough. Chest X-ray revealed new right lower lobe infiltration, and tongue ulcers were noted. Antibiotics were shifted to ciprofloxacin, azithromycin was added for atypical pneumonia, and acyclovir was given for suspected herpes infection. CT of the thigh showed subcutaneous gas at the fascia, compatible with necrotizing fasciitis.

In the second week, he developed altered consciousness (E3M5V1) and right hand seizure. Lumbar puncture showed opening pressure 27 cmH_2O and low CSF glucose (11 mg/dL, serum glucose 95 mg/dL). India ink stain and cryptococcal antigen were positive. Liposomal amphotericin B (3 mg/kg) and fluconazole were started. Serum cryptococcal antigen titer was 1:4096. MRI revealed multiple T2/FLAIR hyperintense lesions at basal ganglia and supra- and infratentorial white matter. Culture of thigh pus and CSF both grew Cryptococcus neoformans. Cryptococcal meningoencephalitis was diagnosed.

One month later, skin biopsy also revealed fungal spores positive for PAS, GMS, and mucicarmine stains, confirming cryptococcosis. Repeat serum cryptococcal antigen titer decreased to 1:1024, but consciousness remained drowsy. Prozone effect was suspected, but further dilution of CSF specimen showed no higher titer, thus prozone effect was ruled out. After continued antifungal treatment and intracranial pressure control, the patient's consciousness improved to baseline.

Discussion

Disseminated cryptococcosis may present with multi-organ involvement. Although our patient's

antigen tests remained positive, clinicians should recognize potential pitfalls such as prozone effect, which may cause false-negative results and delay diagnosis.

Conclusion

Early recognition and treatment of disseminated Cryptococcus neoformans infection are crucial. Awareness of diagnostic challenges, including possible prozone effect, can improve timely diagnosis and patient outcomes.

病例報告 114_C176

左束支起搏器植入術後心房穿孔併發心包填塞之病例報告

Right Atrial Perforation with Cardiac Tamponade Following Left Bundle Branch Pacing: A Case Report

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Introduction

Cardiac implantable electronic devices (CIEDs) are essential for bradyarrhythmias and conduction disorders. Left bundle branch pacing (LBBP), a form of conduction system pacing, offers a more physiological alternative to right ventricular pacing and may reduce pacing-induced desynchrony and heart failure. Nevertheless, rare but life-threatening complications such as septal perforation and cardiac tamponade can occur. We report right atrial appendage (RAA) perforation with tamponade following LBBP.

Case Report

A 66-year-old man with no significant medical history had junctional bradycardia with LBBB on routine screening. A 24-hour Holter showed bradyarrhythmia (32–114 bpm) without long pauses. Transthoracic echocardiography (TTE) demonstrated preserved left ventricular systolic function and no structural abnormalities. Suspected sick sinus syndrome prompted an electrophysiological study, which revealed high-degree AV block with perinodal disease. Given LBBB and anticipated high pacing burden, an LBBP pacemaker was planned. During implantation, the initial right atrial (RA) lead exhibited a high threshold and provoked chest tightness with transient hypotension that improved after hydration; the RA lead was replaced with normalization of threshold. After wound closure, chest pain, hypotension, and diaphoresis recurred. Emergent TTE showed a large pericardial effusion; pericardiocentesis drained about 1300 mL of dark, bloody fluid. Despite fluids and inotropes, hypotension persisted, prompting urgent cardiothoracic surgery consultation. Emergency surgery identified RAA perforation, which was repaired. Postoperatively, he remained hemodynamically stable; follow-up TTE showed no recurrent effusion. He was extubated the next day, drains were removed on postoperative day 3, and he was discharged on day 8. At follow-up, Holter showed sinus rhythm with intermittent pacing, and device interrogation was normal.

Discussion

Cardiac tamponade after pacemaker implantation is uncommon (≃0.1–2%) and usually results from lead perforation, most often at the right ventricular apex or free wall; septal perforation is more typical of LBBP given its target. RAA perforation is less frequent owing to low-pressure of RA, but clinically significant. Reported risk factors include chronic heart failure, operator inexperience, dual-chamber or CRT-P systems, active-fixation RA leads, prior temporary pacing, and steroid or anticoagulant use. Intraoperative chest pain during lead manipulation may signal irritation or early perforation. When symptoms persist or recur, immediate echocardiographic reassessment is warranted; implanting a new lead at an alternative site while leaving the suspicious lead in place

until definitive evaluation may be safer than simple repositioning.

Conclusion

RAA perforation is a rare but serious complication of LBBP that may not be fully resolved by pericardiocentesis alone. Early recognition of warning signs, such as intraoperative chest pain, and timely surgical intervention are crucial. Clinicians should maintain vigilance during lead placement when unexpected symptoms occur.

病例報告 114_C177

新冠肺炎後合併頸靜脈壓迫症候群之 Lemierre’s 症候群：一個病例報告

Lemierre’s Syndrome After COVID-19 Infection Complicated by Jugular Vein Compression Syndrome: A Case Report

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Introduction

Lemierre's syndrome is a rare condition characterized by septic thrombophlebitis of the internal jugular vein, often as a result of an oropharyngeal infection. Common related pathogens include Fusobacterium species, Enterobacteriaceae and Streptococci.

Case Report

We reported a case of a 65-year-old woman with a history of stage IV chronic kidney disease and atrial fibrillation on warfarin therapy, who was admitted to the intensive-care unit due to E. coli-related urinary tract infection, acute kidney injury and septic shock. A pulsatile, painful swelling mass over the right side of neck was noted after admission. Bedside echocardiography demonstrated turbulent flow in the right internal jugular vein. Contrast-enhanced neck CT was performed to exclude an arteriovenous fistula, which revealed a fistulous connection between the right subclavian artery and the right internal jugular vein at the T3 level, accompanied by aneurysmal change of right internal jugular vein. Review of her medical history showed that she had Covid-19 infection in 2022, and experienced right neck pulsatile sensation afterwards. A chest CT performed in 2022 had also demonstrated the same fistulous image. Based on the working diagnosis of Lemierre’s syndrome, metronidazole was added to meropenem to provide further coverage against oropharyngeal anaerobes. Angiography was not performed due to high risk of contrast-induced nephropathy at that time. Instead, neck MRA was arranged and showed filling defects within the internal jugular vein. Further venous Doppler sonography of the upper extremity revealed thrombosis involving the right internal jugular, subclavian, brachial, and basilic veins. Warfarin therapy for atrial fibrillation was continued, and the antibiotic regimen was later shifted to cefixime plus metronidazole for a total course of four weeks to treat septic thrombophlebitis. Unfortunately, the patient developed irreversible acute kidney injury with persistent oliguria during follow-up, thus long-term hemodialysis was arranged. Subclavian angiography and jugular angiography were performed consequently, which revealed compression of the right jugular vein by the subclavian and vertebral arteries, rather than the presence of an arteriovenous fistula. Following the completion of antibiotic therapy and the continued use of anticoagulation, the patient remained afebrile, with complete resolution of neck pain.

Discussion

With the widespread use of antibiotics for upper respiratory tract infections, the incidence of Lemierre's syndrome has declined substantially. Nevertheless, cases have still been reported nowadays with possible severe complications, especially after the covid-19 pandemic, which had raised concern among experts. Covid-19 has been associated with endothelial dysfunction,

hypercoagulability, and vascular inflammation, all of which may predispose individuals to thrombosis in the presence of vascular abnormalities. In addition, the role of anticoagulation in Lemierre's syndrome remains debated. In our case, anticoagulation with warfarin was continued for atrial fibrillation, which may provide additional benefit in managing extensive venous thrombosis.

Conclusion

Lemierre's syndrome should be considered in patients presenting with fever, a tender or swollen neck, and symptoms suggestive of embolism. Appropriate antibiotic therapy should be initiated, and the use of anticoagulation should be individualized.

病例報告 114_C178

Aveir 無導線心律調節器於高齡室上性心搏過速合併房室傳導阻斷患者植入

Aveir Leadless Pacemaker in an Elderly Patient with SVT and AV Block

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Introduction

Leadless pacemakers represent a significant advancement in pacemaker technology. It offered a minimally invasive alternative traditional transvenous pacemaker with advantages such as the absence of surgical pocket and avoidance of lead-related complications, including dislodgement, perforation, and wound infection. The Aveir leadless pacemaker(Abbott) is a new generation device that can provide atrioventricular synchronous pacing, retrial available, and long battery duration. We provided a successful implantation of the Aveir leadless pacemaker in an elderly patient with supraventricular tachycardia who developed atrioventricular block after using a minimal dosage of beta-blocker and was successfully treated with leadless pacing.

Case Report

A 72-year-old female with hyperlipidemia experienced her first episode of syncope with palpitations in November 2020, resulted vertebral fracture. Holter monitoring at cardiology clinic revealed 2.5 hours of supraventricular tachycardia (SVT). She started on bisoprolol 1.25 mg daily, which relieved her palpitations.

In February 2024, she experienced a syncopal episode while traveling. Two Holter studies and neurological evaluation that year were unremarkable while syncope recurred. Finally, Holter study performed on May20, 2025, revealed 6.2 seconds of paroxysmal complete atrioventricular block. Bisoprolol was discontinued. A repeat Holter study one month after demonstrated an episode of SVT lasting 27 minutes. However, the patient declined radiofrequency ablation due to personal reasons. After discussion, she opted for self-paid implantation of a leadless pacemaker, considering its advantages of a smaller wound and shorter hospitalization.

On June 23, 2025, she underwent implantation via the right femoral vein, with the device positioned in the low septum of right ventricle. The 52-minutes procedure, with 24 minutes of fluoroscopy and 68 mL contrast, was completed without complications. She discharged two days later, and follow-up two weeks post-procedure confirmed good device function. EKG showed pacing rhythm. She continues regular outpatient follow-up with stable device performance.

Discussion

Leadless pacemakers are a safe alternative to traditional devices, reducing risks of lead fracture, venous obstruction, and pocket-related complications such as infection or hematoma. In our case, the patient experienced minimal postoperative discomfort including no surgical scar and wound. Notably, given the patient's age of 72 and the battery longevity of 20 years noted in the recent check, raises the reasonable expectation of lifelong pacing support. The system also allows retrieval and replacement, and an additional atrial device can be added if AV synchrony becomes necessary.

Conclusion

This successful experience with the Aveir leadless pacemaker demonstrates its safety and clinical utility, while also paving the way for future applications in patients requiring more complex pacing support. Our case contributes to growing evidence supporting its role as a foundation for broader adoption, including future atrial and dual-chamber strategies.

病例報告 114_C179

法布瑞氏症的「偽裝術」：肥厚阻塞性心肌病變面具下的真兇

Fabry Disease Masquerading as Hypertrophic Obstructive Cardiomyopathy

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Introduction

Hypertrophic obstructive cardiomyopathy (HOCM) is characterized by asymmetric septal hypertrophy, systolic anterior motion (SAM), and dynamic left ventricular outflow tract (LVOT) obstruction. Fabry disease, an X-linked lysosomal storage disorder caused by α-galactosidase A deficiency, may mimic this phenotype. Differentiation is essential because Fabry disease is treatable with enzyme replacement or chaperone therapy, while sarcomeric HOCM is managed symptomatically.

Case Report

A 54-year-old male presented with progressive exertional dyspnea. He had no history of hypertension, diabetes, or significant coronary risk factors. Physical examination revealed a systolic murmur along the left sternal border, accentuated with Valsalva maneuver.

Transthoracic echocardiography demonstrated marked left ventricular hypertrophy with interventricular septum 19 mm and posterior wall 16 mm. Left ventricular ejection fraction was preserved at 73%. Doppler assessment revealed impaired relaxation with E/A ratio 0.6, septal E/E' 18 indicating elevated LV filling pressure, SAM of the mitral valve, and a resting LVOT gradient of 42 mmHg. These findings were highly suggestive of HOCM.

To exclude ischemic or infiltrative causes, myocardial perfusion SPECT was performed, showing homogeneous perfusion without evidence of ischemia or infarction. Tc-99m pyrophosphate scintigraphy revealed a heart-to-contralateral ratio of 1.18, below the threshold for transthyretin cardiac amyloidosis.

Right heart catheterization demonstrated near-normal hemodynamics with right atrial pressure 4 mmHg, pulmonary artery mean pressure 14 mmHg, pulmonary capillary wedge pressure 8 mmHg, cardiac index 3.45 L/min/m^2, and pulmonary vascular resistance 1.05 Wood units. These data suggested preserved global hemodynamics despite LV hypertrophy.

Given the discrepancy between typical HOCM morphology and negative ancillary testing, right ventricular endomyocardial biopsy was obtained. Histopathology revealed hypertrophied, vacuolated myocytes containing lamellar inclusion bodies (“zebra bodies”), consistent with Fabry disease. This established the final diagnosis of Fabry cardiomyopathy masquerading as HOCM.

Discussion

This case demonstrates the diagnostic challenge when Fabry disease presents with typical HOCM features. Echocardiographic findings of concentric hypertrophy, preserved systolic function, and LVOT obstruction are not pathognomonic for sarcomeric HOCM. Without careful evaluation, Fabry disease may be overlooked, leading to delayed initiation of disease-specific therapy.

Fabry disease can be suspected when patients show hypertrophy without conventional risk factors,

especially if systemic clues such as neuropathic pain, corneal verticillata, or family history are present. However, in some cases, cardiac manifestations dominate, as in this patient. Here, myocardial biopsy provided the definitive diagnosis when noninvasive imaging failed to discriminate.

Therapeutic implications are substantial. HOCM management includes β-blockers, non-dihydropyridine calcium channel blockers, disopyramide, and septal reduction therapies for severe obstruction. By contrast, Fabry disease requires enzyme replacement or chaperone therapy, which can slow or even reverse organ involvement. Early recognition thus has the potential to alter prognosis significantly.

Conclusion

This case highlights that typical HOCM echocardiographic findings may conceal Fabry disease, a treatable systemic disorder. A comprehensive diagnostic strategy combining echocardiography, nuclear imaging, catheterization, and myocardial biopsy is critical in patients with unexplained hypertrophy. Early identification enables initiation of disease-specific therapy and avoidance of inappropriate interventions, ultimately improving long-term outcomes.

病例報告 114_C180

Takotsubo 症候群合併左心室血栓：心室功能改善與血栓完全消退

Takotsubo Syndrome Complicated by Left Ventricular Thrombus: Complete Functional Recovery and Thrombus Resolution

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Introduction

Takotsubo syndrome (TTS) is an acute stress-induced cardiomyopathy characterized by transient left ventricular (LV) dysfunction. Although typically reversible, it can be complicated by LV thrombus formation, which carries a significant risk of systemic embolism. Anticoagulation is indicated in such cases, and resolution of thrombus often parallels recovery of systolic function.

Case Report

A 65-year-old woman with diabetes mellitus and schizophrenia, residing in a long-term care facility, presented with acute onset of dizziness, generalized weakness, and vomiting for one day. Laboratory tests demonstrated normocytic anemia and impaired renal function without evidence of infection. Initial transthoracic echocardiography showed LV ejection fraction (EF) of 44% with hypokinesia involving the mid-to-distal anterior, anteroseptal, and inferoseptal walls, as well as apical dyskinesis, consistent with Takotsubo syndrome. Global longitudinal strain (GLS) at this stage was -7.9%. Coronary angiography excluded obstructive disease but revealed myocardial bridging. The patient was admitted and treated for acute heart failure.

Close echocardiographic monitoring was arranged after catheterization. One week later, EF had improved to 54.6% with GLS of -8.0%. However, new apical akinesis with aneurysm formation was detected, accompanied by a large LV apical thrombus measuring 4.25 × 1.29 cm. Anticoagulation with warfarin was initiated, and INR was carefully titrated to therapeutic range. Serial follow-up imaging demonstrated progressive systolic recovery: after approximately three weeks of anticoagulation, EF had further improved to 62.7% and GLS normalized to -15.6%. Wall motion returned to normal, and the LV thrombus had completely resolved. No systemic embolic events occurred. The patient was subsequently discharged in stable condition with outpatient follow-up.

Discussion

Takotsubo syndrome typically follows a self-limiting course with recovery of LV systolic function within days to weeks. In this patient, EF improved from 44% to 62.7%, accompanied by normalization of GLS from -7.9% to -15.6% during follow-up, reflecting both functional and contractile recovery. LV thrombus is a recognized but relatively uncommon complication, occurring in 2–8% of TTS cases, particularly in patients with apical ballooning and significant systolic dysfunction. Thrombus may not be evident at initial presentation, underscoring the importance of vigilant echocardiographic surveillance. In this case, repeat imaging one week after catheterization was critical in identifying the thrombus, which emerged despite partial recovery of EF. Prompt anticoagulation with warfarin prevented embolic complications and facilitated complete thrombus resolution within three weeks.

Conclusion

This case illustrates the dynamic course of Takotsubo syndrome, characterized by reversible LV dysfunction, progressive EF and GLS recovery, and the occurrence of an LV apical thrombus that resolved under timely anticoagulation. Careful imaging follow-up, early recognition of complications, and tailored management with anticoagulation are essential to optimize outcomes in patients with Takotsubo syndrome complicated by LV thrombus.

病例報告 114_C181

血壓忽高忽低的診斷挑戰：一例嗜鉻細胞瘤病例報告

Intermittent Hypotensive Shock and Hypertensive Crisis: A Diagnostic Challenge of Pheochromocytoma

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Introduction

Pheochromocytoma is a rare catecholamine-secreting tumor of the adrenal medulla with diverse clinical manifestations. While sustained or paroxysmal hypertension is typical, presentations may vary widely, leading to delayed recognition. Some patients experience extreme fluctuations in blood pressure, ranging from profound hypotension to hypertensive crisis, complicating diagnosis. Recognizing this hemodynamic instability is essential, particularly in patients with cardiovascular comorbidities. We report a case of recurrent near-syncope and alternating hypotensive and hypertensive episodes, ultimately diagnosed as pheochromocytoma.

Case Report

A 66-year-old man with coronary artery disease, hypertension, diabetes, and dyslipidemia presented with near-syncope after coughing. Initial evaluation showed hypotension (84/49 mmHg) and sinus tachycardia. He was hospitalized for pneumonia and presumed vasovagal syncope. Cardiac studies were unremarkable. After discharge, he continued to experience fluctuating blood pressure, with hypotension down to 83/46 mmHg and hypertensive crises up to 240/118 mmHg, sometimes with dizziness and headache. These episodes persisted despite antihypertensive adjustments. Given the remarkable variability, secondary hypertension was suspected. Abdominal computed tomography revealed a 5-cm right adrenal mass with central necrosis. Biochemical testing confirmed catecholamine excess. Preoperative management included alpha-blockade with doxazosin and supportive antihypertensives. The patient underwent robotic-assisted laparoscopic right adrenalectomy, and pathology confirmed pheochromocytoma (pT1, negative margins). Postoperatively, his blood pressure stabilized without antihypertensive therapy.

Discussion

This case illustrates the diagnostic challenge when pheochromocytoma presents atypically. Instead of persistent hypertension, the patient exhibited alternating hypotension and hypertensive crisis, delaying recognition. Clinically, physicians should suspect secondary hypertension when patients have unexplained blood pressure fluctuations, especially when extreme values coexist in the same individual. Initial surveys should include careful history, medication review, and biochemical testing with plasma or urinary metanephrines and catecholamines. Imaging with CT or MRI is then performed to localize the lesion. Cardiology is often the first specialty consulted because of syncope or fluctuating blood pressure. However, once biochemical evidence suggests catecholamine excess, endocrinology input is crucial for diagnostic confirmation and preoperative optimization. Referral to urology is required for definitive surgical management. Multidisciplinary teamwork among cardiology, endocrinology,

radiology, anesthesiology, and urology is essential to ensure accurate diagnosis and safe perioperative care. Preoperative alpha-blockade and volume expansion reduce intraoperative cardiovascular risk, while postoperative surveillance is necessary to confirm biochemical remission and detect recurrence. Delay in diagnosis may lead to recurrent hypertensive emergencies, cardiomyopathy, or even sudden death, highlighting the need for vigilance.

Conclusion

Pheochromocytoma should be considered in patients with unexplained and extreme blood pressure fluctuations, even without the classic triad of headache, palpitations, and diaphoresis. Early recognition, appropriate diagnostic survey, and timely referral are critical to avoid misdiagnosis and prevent life-threatening hypertensive crises or hypotensive shock. This case emphasizes the importance of systematic evaluation and multidisciplinary collaboration in patients with atypical presentations of secondary hypertension, reinforcing that pheochromocytoma remains a curable condition when identified and treated appropriately. Long-term follow-up is recommended to monitor for recurrence, and clinicians should always maintain suspicion for secondary hypertension in patients with inconsistent responses to standard therapy.

病例報告 114_C182

子宮肌瘤壓迫靜脈致深部靜脈栓塞並以手術前後期間抗凝劑成功治療：一病例報告

Uterine Leiomyoma-Induced Deep Vein Thrombosis Successfully Managed with Perioperative Anticoagulation Bridging: A Case Report

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Introduction

Although both deep vein thrombosis (DVT) and uterine leiomyoma (UL) are common conditions encountered in clinical practice, DVT secondary to venous compression by UL is rare. When myomectomy is required to restore venous circulation in the lower limbs, the optimal perioperative strategy to prevent DVT and pulmonary embolism (PE) with either pre-myomectomy inferior vena cava (IVC) filter placement or anticoagulation bridging alone remains controversial. Herein, we report a case of DVT caused by a large UL compressing both common iliac veins, which was successfully managed using a perioperative anticoagulation bridging strategy.

Case Report

A 38-year-old woman presented to the emergency department with a one-day history of progressive swelling, pain, and numbness in the left lower extremity. Her estimated glomerular filtration rate was 93 ml/min/1.73m^2 and liver biochemistry tests were normal. Contrast-enhanced computed tomography revealed a large UL (dimensions: 14cm × 14cm × 8cm) that completely compressed and occluded the left common iliac vein. The right common iliac vein was also severely compressed, but venous flow remained patent. Duplex ultrasonography confirmed thrombosis involving the left common femoral, great saphenous, and popliteal veins.

The patient was initiated on parenteral anticoagulation with enoxaparin (60 mg, 1 mg/kg) every 12 hours for 7 days, followed by a transition to oral dabigatran 150 mg twice daily. Owing to limited regression of left lower limb swelling, IVC filter placement followed by myomectomy was scheduled after one month of anticoagulation, in consultation with the gynecology team. However, IVC filter insertion was unsuccessful because of severe obstruction of the common iliac veins. Dabigatran was discontinued 3 days before surgery, and an uneventful laparotomy myomectomy was performed. Postoperatively, continuous heparin infusion was initiated on day 1, followed by a transition to oral dabigatran. After a 7-day hospitalization, the patient was discharged with complete resolution of lower limb edema and no major bleeding events. The dabigatran dose was subsequently reduced to 110 mg twice daily. Follow-up computed tomographic angiography at 9 months showed no residual thrombus.

Discussion

Prophylactic IVC filter placement is sometimes considered in patients at high risk of PE, particularly those undergoing surgery who cannot safely receive anticoagulation. However, IVC filters are associated with procedure-related complications, including DVT, IVC penetration, and guidewire entrapment. Moreover, their role in PE prevention remains controversial due to limited supporting evidences.

Myomectomy is a procedure with high bleeding risk, and our patient concurrently faced a high thromboembolic risk due to recent DVT. In this dilemma, preoperative IVC filter placement would have been a reasonable option. Nevertheless, when IVC filter insertion failed, perioperative anticoagulation bridging was shown to be a safe and effective alternative in this case.

Conclusion

DVT can occur secondary to venous compression from UL, a rare but clinically important condition. Clinicians should consider UL as a potential underlying cause in patients presenting with unexplained DVT. Furthermore, this case highlights that myomectomy can be safely performed under an anticoagulation bridging strategy when IVC filter placement is not feasible. Careful perioperative anticoagulation is essential to balance the competing risks of bleeding and thromboembolism.

病例報告 114_C183

鼻腔手術之成功正面效應對於合併慢性鼻竇炎和嚴重阻塞性睡眠呼吸中止症及內科治療無效患者

Successful positive health impact of nasal surgery in comorbid difficult-to-treat chronic rhinosinusitis and severe obstructive sleep apnea with failed adequate response to medical therapy

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Introduction

Identification of promising candidates for either primarily combining CPAP and nasal surgery, or as salvage treatment of it in co-morbid severe OSA and CRS with CPAP failure, is vital to successful positive surgical impact and improved clinical outcome.

Case Report

A 56-year-old woman with DM and allergic rhinitis had chief complaint of disruptive sleep and loud snoring with witnessed sleep pauses for about 4 years. Nasal blockades have become worse in the last two years. ESS 8, BMI 30, neck circumference was 37 cm. Local PE showed lateral peritonsillar narrowing, MPS 2 and grade 2 palatine tonsillar hypertrophy. She also had moderate COPD of Gold stage I, group B of mixed allergic eosinophilic endotype and treated with regular therapy but still experienced frequent moderate AE of COPD together with acute flare episodes of CRS. Very Initial AHI was 45/h, ODI of 52/h, and snoring index of 255. AHI down to 15 on CPAP pressure 8 cmH2O. But owing to intolerance CPAP use with Oro-nasal interface very well as intranasally increased impeding pressure, and failed response to oral antibiotics, IN-CS and OCS, she performed bilateral pan-sinusotomy and stereotactic procedure for paranasal and facial sinus washing and biopsy, and partial inferior nasal turbinectomy. Biopsy report showed chronic inflammation of rhino-sinuses. Post-surgery AHI on CPAP use improved to 5, ODI to 7, and CPAP intolerance symptoms diminished with much better comfort.

Discussion

OSA is characterized by airway obstruction at either single or multiple levels of upper airway during sleep due to either anatomical narrowing or/and physiological collapse in different anatomic sites, proportions and severities in individual patients. Mutually interactive pathophysiology in comorbid CRS-OSA is due to late phase reaction secondary to chronic allergen exposure, leading to dilated nasal mucosal capacitance vessels, local congestion, retained nasal secretion, chronic nasal soft tissue swelling with decreased nasal diameter, therein inhibiting nasal mechanoreceptors, decreasing activation of nasal-ventilatory reflex and muscles tone of upper airway. Chronic arterial O2 desaturation by impaired nasal airflow and frequent sleep apneic events causes increased inflammatory cytokines and then contributes to development of both higher disease burden, disease sequelae, higher oral antibiotics and OCS use, CPAP intolerance, and worse clinical outcomes such as reduced productivity, cognitive impairment and decreased quality of life. The role of nasal surgery as salvage treatment in such patients with CPAP failure is to enlarge nasal passage and improve smooth nasal airflow, AHI, RDI, CT90% (Cumulated % of the

time spent with SaO2 of below 90%), NCV (nasal cavity volume), MCA (minimal cross-sectional area), and tolerate CPAP therapy.

Conclusion

In patients with co-morbid severe OSA and difficult to treat CRS, and impaired CPAP tolerance, we should consider possibility for intervening with surgical treatment for either CRS or OSA if exact site of upper airway problem was well detected in order to alleviate diseases burden, sequalae and chronic medication side effects, for enhancing upper airway (UA) airflow, arterial oxygen (O2) level, and improving AHI, ODI, daytime function, nighttime sleep, cardiorespiratory health and quality of life.

病例報告 114_C184

肺炎演變肺膿瘍,嚴重的克雷伯肺炎菌感染

Empyema with Klebsiella pneumoniae after Severer necrotizing pmeumonia

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Introduction

Although the mechanism of progression into infection in K. pneumoniae strain remains unclear, evidence shows that certain plasmids and chromosomal influence virulence.Prompt treatment is needed.

Case Report

This 50 y/o male patient had a history of diabetes mellitus for many years with regular medication and insulin control , Chronic Hepatitis B and alchoholism
Admitted for necrotizing pneumonia and emergent endotracheal tube was
Inserted. Chest X ray follow up with left lung collapse and drainage with fluid.
Pleural effusion (PH:7.0 Sugar:<2 mg/dl Protein :3.6 g/dL LDH :8812 IU/L)
Thus chest surgeon was consulted and operation arranged. Bronchoscopy was
also arranged for possible infectious survey. Blood culture yield Klebsiella pneumonia. After series antibiotics and aggrasive chest care above 2 weeks. He was extubated and transfer to ward.

Discussion

status of host defense, such as cancer, diabetes mellitus, and alcoholism, influencing the host susceptibility to K. pneumoniae infection

Conclusion

Empyema have multiple elective treatment
Klebsiella pneumonia may take long hospitalization
Delays to treatment are key contributors to poor outcomes
Early drainage and intervention save lifes

病例報告 114_C185

嚴重手部軟組織感染與金黃葡萄球菌相關併水泡症狀

Hand cellulitis relates to S.aureus, severe blister formation

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Introduction

Extracorporeal toxin removal (ECTR) techniques play a pivotal role in the treatment of various types of acute poisoning.7

Case Report

42 years old male patient, prisoner has past history of Hypertension,
Hypertensive Cardiovascular Disease, Brain intracerebral hemorrhage s/p operation on 16 years, Epidural hemorrhage (left frontal) on 2024/09/10 post craniotomy and cranioplasty at left F-T, Chronic kidney disease
Hypothermia with BT 34~35 degree, sent to ER on 2/24
Emergent endotracheal tube insertion for poor resp pattern
urine culture yield MRSA on 2/24.Antibiotics with Teicoplanin give. Seizure occured 2/25, consulted Neurologist and Keppra 500mg IVD QD & Depakine 400mg IVD Q8H. Due to Bradycardia , cardiologist was consulted and high dose inotropic agent was adopt.
Hypothyroidism thus Eltroxin 1# QD with hydrocortisone after endocrinologist consultation. Acute kidney injury (Cre 2.4->3.7 mg/dl) and acidosis, CVVH arranged 2/26~3/31.
On 2/28, blisters around the right arm and severe swelling, thus GS was consult and suggested observation
Rheumatologist was consulted and autoimmune titer was surveyed
but no active finding (HBsAg Positive, Anti-HBc-IgG Positive, IgG 832.0 ANA ＜1:80X(-) Anti-dsDNA: Negative
IgE 389.700 Anti-cardiolipin :negative
β2-GPI IgG/IgM Negative, ENA anti-Sm/anti-RNP Negative
HR , urine output and resp pattern improved. Healing condition on skin was noted. Extubation on 3/25. He was transferred to ordinary ward

Discussion

Due to the high heterogeneity in the sepsis population, individualized treatment may be more beneficial[1]

Conclusion

Blister disease and soft tissue may reflect severe infection and impact risk
Specific are distiguished by target antigens, clinical manifestations, and immunopathological profiles.[6]

CVVH and steroid can modestly reduce cytokine levels and elimated risk[3,4]
Multidisciplinary work and adequate support, resuscitation improved Outcomes

病例報告 114_C186

兩側下肢水腫. 病例討論

Bilateral Lower Leg Edema

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Introduction

Clinically, bilateral lower leg edema is one of the occasionally found symptoms, the causes of which include thyroid function disorder, congestive heart failure, liver cirrhosis, chronic glomerulonephritis, hypoalbuminemia, and drug induced, etc. Generally speaking, the illness is most prevalent in the old age population. Some of the cases have a simple clinical presentation; however, some are more complicated with other comorbidities. The latter need more time and effort to make a correct diagnosis and provide proper treatment.

Case Report

From 2021 to 2025, we collected 14 cases that presented clinically with bilateral lower leg edema. Through detailed history taking, physical examination, laboratory blood tests, X-ray study, EKG, and abdominal ultrasound examination, etc., the final diagnosis was made, and proper treatment was given accordingly, as well as follow-up evaluation.

Among them, one case showed a special clinical manifestation with both pitting edema and myxedema simultaneously. This special case study is included in this report.

Discussion

A) Special Case Report: A case of liver cirrhosis, complicated with chronic glomerulonephritis and hypothyroidism

- A patient, male, aged 49, had a chronic alcoholic drinking history. He suffered from both leg edema and ascites and was hospitalized for further management. Chronic alcoholic hepatitis, liver cirrhosis, and chronic glomerulonephritis were diagnosed. The clinical course showed improvement after treatment; however, the lower leg edema did not disappear completely. He even felt his legs became stiffer, with walking difficulty.
- He visited our clinic for medical help. On physical examination, thickened skin with edema was noted in both lower legs, and myxedema due to hypothyroidism was suspected (Fig. 1). Abdominal ultrasound revealed liver cirrhosis with ascites (Fig. 2).
- Laboratory blood test showed the following:
  - Liver function test: GOT/GPT 31/16; BT/BD 4.5/3.7; γ-GT 505 (0.5–79)
  - Renal function test: Cr 1.45; eGFR 55 ml/min
  - Thyroid function test: TSH 8.5 (0.5–4.7)
- Thus, alcoholic liver cirrhosis, chronic glomerulonephritis, and hypothyroidism were diagnosed, and Levothyroxine (Eltroxin) was added to the medication. The myxedema improved gradually, and he could walk without difficulty thereafter.

Conclusion

This is a case of mixed pitting edema and myxedema due to comorbidities of liver cirrhosis, chronic glomerulonephritis, and hypothyroidism. It is necessary to identify all comorbidities and provide treatment accordingly. In this way, the bilateral lower leg edema can be resolved.

病例報告 114_C187

骨髓纖維化合併紅斑肢痛症與慢性肢體威脅性缺血：案例報告

Post-ET Myelofibrosis Presenting with Erythromelalgia and Chronic Limb-Threatening Ischemia: A Case Report

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Introduction

Erythromelalgia is a microvascular thrombotic manifestation of myeloproliferative neoplasms (MPNs), most commonly essential thrombocythemia (ET). Because symptoms may mimic lower extremity arterial disease (LEAD), early recognition of MPN together with vascular evaluation is crucial to prevent tissue loss. We report a JAK2 V617F–positive patient with post-ET myelofibrosis (MF), complicated with chronic limb-threatening ischemia (CLTI).

Case Report

A 51-year-old woman with diabetes and CKD3b presented with a one-year history of progressive bilateral lower-limb burning pain and dependent erythema; during the preceding 3–4 months she became unable to stand. There was also gangrenous change of her second toe, for which she presented at the cardiology clinic to rule out LEAD. She also reported marked weight loss (≃40→28 kg/6 months) and chronic watery diarrhea. Laboratory studies showed leukocytosis (WBC 21.6×10^3/μL; ANC 18.1×10^3/μL), thrombocytosis (718×10^3/μL) with giant hypogranular platelets, elevated LAP (274), and splenomegaly. HBsAg was positive.

Angiography demonstrated a discrete 90% right popliteal artery stenosis with otherwise patent inflow/outflow; sequential balloon angioplasty to the SFA–popliteal segment was successful. Given the discrepancy between her severe symptoms and relatively mild macrovascular disease, other prothrombotic causes were suspected. She was thus started on dual-pathway inhibition with aspirin and vascular dose of rivaroxaban, after which her symptoms partially improved.

Bone marrow biopsy revealed hypercellular marrow with megakaryocytic hyperplasia and myelofibrosis grade 2; JAK2 V617F mutation allele burden was 87%. Given symptomatic splenomegaly and constitutional/ischemic symptoms, ruxolitinib 5 mg twice daily was initiated. Over four months, erythromelalgia and dependent rubor resolved, gait recovered, and the ischemic toe wound healed.

Discussion

This case illustrates two complementary mechanisms of limb pain in MPN: (1) erythromelalgia from platelet activation and arteriolar microthrombosis, and (2) superimposed large-artery disease causing CLTI that required revascularization plus antithrombotic therapy. Clinical clues to an underlying clonal process included marked thrombocytosis with giant platelets, a high LAP score, splenomegaly, and a positive JAK2 V617F mutation.

The marrow finding of grade-2 myelofibrosis with massive splenomegaly is diagnostic of post-ET myelofibrosis (MF) according to IWG-MRTcriteria. Therapeutic goals shift from ET-style cytoreduction to MF-directed control of splenomegaly and symptom burden. Ruxolitinib was

therefore selected as disease-modifying therapy, as it is the standard of care for symptomatic intermediate/high-risk primary or post-ET MF and is preferred over hydroxyurea or interferon once overt MF is established. Importantly, it is also the standard approach for managing microvascular complications such as burning pain and dependency rubor, where ruxolitinib has demonstrated greater efficacy than alternative JAK inhibitors and remains the first-line in this setting.

Conclusion

Post-ET MF may present with erythromelalgia and CLTI. When dependency-induced burning erythema accompanies thrombocytosis and splenomegaly, clinicians should evaluate for MPN evolution (including JAK2 testing and marrow staging) and for LEAD. A combined strategy—revascularization, antithrombotic therapy, and ruxolitinib for MF—can relieve pain, prevent progression to gangrene, and restore function.

病例報告 114_C188

以冠狀動脈非阻塞性心肌梗塞為表現的副神經節瘤：病例報告

Paraganglioma presenting with myocardial infarction with non-obstructive coronaries: a case report

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Introduction

Paragangliomas are rare neuroendocrine tumors that arise from extra-adrenal paraganglia and share many features with pheochromocytomas. Their clinical manifestations vary depending on location, but those along the sympathetic chain often secrete catecholamines, which may lead to a spectrum of cardiovascular complications, including arrhythmias, hypotension, and myocardial ischemia. Here we describe a case of paraganglioma presenting with myocardial infarction with non-obstructive coronary arteries (MINOCA), highlighting diagnostic and management challenges.

Case Report

A 52-year-old man reported chest pain for three days. He was seen at another hospital where non-ST segment elevation myocardial infarction was diagnosed according to symptoms, ST segment depression in lead V4-6 on ECG and elevated troponins. He was then transferred to our hospital for further management. He denied prior similar episodes, cardiovascular history, or significant comorbidities. Coronary angiography revealed non-obstructive coronary arteries but identified severe spasm in the patient's right coronary artery, which resolved after intracoronary administration of nitroglycerin. He was thus diagnosed with MINOCA, coronary spasm-induced, and was discharged with bisoprolol and diltiazem to prevent coronary spasm.

However, after discharge, the patient returned to the clinic with intermittent headaches, nausea, and fluctuant blood pressures, with systolic readings exceeding 200 mmHg. These symptoms raised suspicion for pheochromocytomas, possibly elicited by unopposed alpha-adrenergic activity after starting bisoprolol. Blood tests revealed increased plasma free metadrenaline levels at 0.55 nmol/L (normal value < 0.5 nmol/L) and free normetadrenaline levels at 3.44 nmol/L (normal value < 0.9 nmol/L). CT demonstrated a 4.8x4.0 cm well-defined marginal enhancing cystic mass at inferior mesenteric artery orifice level of the left periaortic space. No apparent adrenal nodules, enlarged regional lymph nodes or metastases were found. His plasma chromogranin A level was 88.8 ng/mL (normal value < 101.9 ng/mL). He then underwent laparoscopic retroperitoneal tumor excision, and pathology confirmed the diagnosis of a paraganglioma. The surgery was uncomplicated, and the patient fully recovered. All antihypertensive drugs were discontinued and his blood pressure normalized. He has been asymptomatic since then and continues follow-up in the clinic.

Discussion

MINOCA is clinically defined by the fulfillment of acute myocardial infarction criteria but without significant coronary artery obstruction. Potential causes include coronary artery spasm, microvascular dysfunction, plaque erosion, coronary embolism, and spontaneous coronary

dissection etc. Literature has reported that pheochromocytomas or paragangliomas can trigger myocardial oxygen demand and supply mismatch, catecholamine-driven tachycardia, and coronary vasospasms that may lead to myocardial ischemia in the absence of coronary atherosclerosis, further complicating the diagnosis.
Additionally, patients with acute myocardial infarction are often treated with β-adrenergic blockers according to standard of care. However, in cases of catecholamine-releasing tumors, blocking β-adrenergic mediated vasodilation can lead to a paradoxical increase in blood pressure due to unopposed α-adrenergic receptor stimulation, which can worsen the patient's symptoms. This underscores the sore need for an accurate diagnosis.

Conclusion

Our case illustrates that pheochromocytomas or paragangliomas can mimic the clinical presentation of acute myocardial infarction. When investigating the underlying causes of MINOCA, particularly in patients with unexplained and fluctuant blood pressures, pheochromocytomas and paragangliomas should be considered as a possible differential diagnosis.

病例報告 114_C189

肺部鱗狀細胞癌患者接受免疫治療後出現噬血性淋巴組織球增生症: Tocilizumab 成功治療案例

Hemophagocytic Lymphohistiocytosis Induced by Pembrolizumab in Lung Squamous Cell Carcinoma: Effective Treatment with Tocilizumab

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Introduction

Hemophagocytic lymphohistiocytosis (HLH) is a rare but life-threatening immune-related adverse event (irAE) of immune checkpoint inhibitors (ICIs). Its nonspecific manifestations often mimic infection, delaying diagnosis and treatment.

Case Report

A 44-year-old man with advanced squamous cell carcinoma of the right upper lung (cT3N2M1b, stage IV, PD-L1 TPS 55%) received first-line cisplatin, paclitaxel, and pembrolizumab. After the third pembrolizumab cycle, he developed ICI-induced HLH, fulfilling HLH-2004 criteria with fever, cytopenias, splenomegaly, hyperferritinemia, and hypofibrinogenemia. Corticosteroids failed to induce remission, but tocilizumab led to rapid improvement. Chemotherapy was resumed without immunotherapy, and subsequent surgical resection achieved pathological complete response. No recurrence of lung cancer was observed during three years of follow-up.

Discussion

This report presents a rare case of pembrolizumab-induced HLH in advanced lung squamous cell carcinoma. HLH may mimic infection or cytokine release syndrome, yet diagnostic features such as marked hyperferritinemia, hypofibrinogenemia, splenomegaly, and cytopenias are characteristic. While corticosteroids remain first-line therapy, this case was steroid-refractory. Tocilizumab, an IL-6 receptor inhibitor, provided rapid disease control and long-term benefit. Emerging data also suggest IL-6 blockade may potentiate antitumor activity. With prompt recognition and tailored management, patients with ICI-related HLH can still achieve durable cancer remission.

Conclusion

This case highlights that early recognition of HLH in patients receiving ICIs is essential to prevent life-threatening complications. Tocilizumab offers a potential treatment for steroid-refractory HLH, and long-term lung cancer control remains achievable despite this complication.

病例報告 114_C190

Denosumab 與 Romosozumab 治療妊娠及哺乳相關骨質疏鬆症：病例報告

Treatment of Pregnancy- and Lactation-Associated Osteoporosis with concurrent Denosumab and romosozumab : a case report

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Introduction

Pregnancy- and lactation-associated osteoporosis (PLO) is a rare condition characterized by fragility fractures occurring in late pregnancy or early postpartum, most often affecting the vertebrae.The pathogenesis of PLO is unclear, and there is no accepted consensus regarding the treatment of PLO. Although treatments with drugs such as bisphosphonate, strontium ranelate, denosumab, and teriparatide were reported, there is no report of a patient with PLO treated with denosumab and romosozumab.

Here, we present the first case of a patient with PLO treated with a combination of denosumab and romosozumab.

Case Report

A 34-year-old woman presented in January 2024 with sudden-onset back pain two weeks postpartum. She had a history of low BMD and autoimmune thyroid disease. After a spontaneous abortion in August 2022 with positive ANA (1:1280), she was treated with hydroxychloroquine and low-dose prednisolone. During her second pregnancy, she also received aspirin (100mg daily) and prophylactic enoxaparin.

She underwent cesarean delivery at 39+3 weeks in January, 2024 with a healthy boy. She breastfed 600mL/day. Two weeks later, she developed sudden severe back pain. Image revealed compression deformities at T8, T12, L2. Dual-energy X-ray absorptiometry (DXA) confirmed markedly reduced bone mineral density. MRI later showed new T11-T12 fractures. A tentative diagnosis of pregnancy- and lactation-associated osteoporosis (PLO) was made.

Nonpharmacologic measures included discontinued breastfeeding, adequate calcium and vitamin D supplement, corset support, and gradual weight-bearing activity. Denosumab was administered on February 1, 2024, followed by the initiation of romosozumab on April 11, 2024. The combination therapy was continued for one year, resulting in significant improvement in BMD.

Discussion

PLO, first reported in 1955, presents in the early postpartum with vertebral fragility fractures, severe back pain, and height loss. Although rare, recurrence risk is high. Management aims are the prevention of new fractures, the relieve of pain, and an increase BMD through supplementation of calcium/vitamin D, cessation of breastfeeding, weight-bearing activity. Several drugs that are used to treat osteoporosis have also been used for PLO. Most reports describe single-agent therapy, but our case demonstrates that combined denosumab and romosozumab can be effective in improving BMD and stabilizing fractures.

Conclusion

Our patient's BMD values increased by 17.3% at the lumber spine compared to the baseline without any adverse events. Further studies are needed to confirm long-term efficacy and safety.

病例報告 114_C191

單核球增多性李斯特菌（Listeria monocytogenes）感染之腦膿瘍，罕見案例報告

Listeria monocytogenes brain abscess: a rare case report

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Introduction

Brain abscess is a focal infection within the brain parenchyma, commonly occurring in immunocompromised patients or following neurosurgical intervention. Rapid diagnosis and treatment are crucial to improve outcomes. Listeria monocytogenes, an uncommon but important cause, especially affects vulnerable populations such as neonates, the elderly, and immunocompromised hosts.

Case Report

An 87-year-old female with a history of colon cancer and cardiac arrhythmia, was admitted to our hospital for a 3-week history of cough, accompanied with shortness of breath and fever up to 38.4°C one day prior to admission. Initial workup at emergency room showed pneumonia, urinary tract infection, and sepsis.

On the fourth day after admission, the patient developed new left-sided weakness, left central facial palsy, and tongue deviation. Subsequent contrast CT showed a 1.2 cm ring-enhancing lesion in the right striatocapsular region with marked perifocal edema consistent with brain abscess. Besides, blood cultures taken during hospitalization grew Listeria monocytogenes. Antimicrobial therapy was adjusted with intravenous ampicillin/sulbactam 3 g every 4 hours and Ceftriaxone 1g every 12 hours. This regimen was continued for 6 weeks with close monitoring. Follow-up brain CT approximately 4 weeks after admission demonstrated resolution of the previously seen ring-enhancing lesion and surrounding edema, with no midline shift or herniation detected. The patient showed clinical improvement and neurological symptoms stabilized.

Discussion

Listeria monocytogenes is a Gram-positive, facultative intracellular bacterium that causes listeriosis, a severe foodborne illness primarily transmitted by ingestion of contaminated food such as unpasteurized dairy, soft cheeses, and deli meats. It is a Gram-positive intracellular bacterium causing severe infections primarily in immunocompromised, elderly, and pregnant patients. Brain abscess due to Listeria is rare and presents with nonspecific neurological symptoms. MRI is the preferred diagnostic tool, showing characteristic ring-enhancing lesions. Prompt blood culture diagnosis is critical as Listeria is resistant to cephalosporins but sensitive to ampicillin, which is the first-line treatment, often combined with gentamicin. The recommended treatment duration is a minimum of 6 weeks, especially in CNS infections.

In our case, early identification of Listeria monocytogenes from blood cultures allowed timely adjustment to targeted antibiotic therapy with intravenous ampicillin and sulbactam, aligned with first-line treatment recommendations. The prolonged course of antibiotics (over 6 weeks) contributed to successful resolution of the brain abscess without need for surgical intervention.

However, initial delay in considering Listeria infection may have postponed prompt CNS-directed therapy, potentially affecting prognosis. Neurosurgical intervention was not performed despite the abscess size; while medically successful here, surgical drainage is often advised in larger or refractory lesions.

Conclusion

Listeria monocytogenes brain abscess is an uncommon but life-threatening infection predominantly affecting vulnerable immunocompromising patient populations. Its nonspecific clinical course and diagnostic challenges require high suspicion, especially in the elderly and immunocompromised. Early blood culture diagnosis and neuroimaging with MRI guide timely and appropriate treatment.

病例報告 114_C192

一名 70 歲男性混合型胰臟導管與神經內分泌癌的案例

A case of pancreatic mixed ductal-neuroendocrine carcinoma in a 70-Year-Old Male

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Introduction

Pancreatic mixed neuroendocrine-non-neuroendocrine neoplasm (MiNEN) is an extremely rare malignancy, accounting for less than 2% of all pancreatic tumors. This hybrid tumor demonstrates aggressive behavior, often driven by the poorly differentiated neuroendocrine component. Due to its rarity and overlapping features with more common pancreatic pathologies, prompt diagnosis and optimal treatment remain challenging.

Case Report

A 70-year-old male presented with intermittent epigastric pain and unintentional weight loss of 3 kg over two months. Laboratory assessment revealed elevated CA-199 (250 U/mL). Abdominal CT and MRI demonstrated a pancreatic body mass with main pancreatic duct dilatation and suspected splenic vein thrombosis. Transabdominal echo was performed with biopsy but not sufficient tissue was obtained for diagnosis. After multidisciplinary discussion, the patient underwent laparoscopic subtotal pancreatic-splenectomy. Gross pathology showed a 7.5x4.0x2.5 cm ill-defined tumor. Histological examination confirmed mixed ductal and neuroendocrine carcinoma, with tumor involvement at the resection margin, but no regional lymph node metastasis. Immunohistochemistry revealed CK7 and CEA positivity in the ductal component, and synaptophysin/chromogranin A positivity in the neuroendocrine component. Bone metastasis was detected at one month post-diagnosis, while abdominal metastasis was identified at one year post-diagnosis. The patient received sequential chemotherapy regimens (gemcitabine, cisplatin, irinotecan/5-FU) for the disease accordingly. Lung metastasis was noted at the second year with presenting episodes of shortness of breath. Regimens of abraxane and gemcitabine, and FOLFOX were administered by progression disease. Family meetings were held multiple times due to disease progression and clinical deterioration. Palliative care was initiated and the patient expired 3.5 years after diagnosis.

Discussion

Pancreatic MiNEN is an uncommon entity with a generally poor prognosis and limited treatment guidelines. Its diagnosis requires adequate surgical tissue for both histological and immunophenotypic confirmation. The two tumor components may expand separately, complicating both local and distant disease management. Surgical resection is the primary modality for non-metastatic disease, but recurrence is frequent. The most effective chemotherapy protocol is not standardized and often targets the more aggressive component. According to published data, the median survival for pancreatic MiNEN remains less than three years, with only a minority of patients surviving beyond two years.

Conclusion

Pancreatic MiNEN represents a rare and challenging diagnosis, requiring accurate histopathological evaluation and multidisciplinary management. Further case collection and long-term follow-up are needed to advance therapeutic strategies and improve patient outcomes.

病例報告 114_C193

卡波西肉瘤相關之免疫重建症候群：一名加護病房長期住院病人的案例報告

Kaposi sarcoma-associated immune reconstitution inflammatory syndrome: A case report of prolonged admission in intensive care unit

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Introduction

AIDS-related Kaposi sarcoma (KS), an AIDS-defining illness per CDC, is the most common tumor in HIV and KSHV carriers. Its incidence has declined with widespread potent antiretroviral therapy (ART), but KS may be “masked” at AIDS onset and later flare after ART initiation as immune reconstitution inflammatory syndrome (IRIS). We present a male patient with KS-associated IRIS (KS-IRIS) whose prolonged hospitalization allowed direct observation of a classic “unmasking IRIS” presentation.

Case Report

A 39-year-old man with poorly controlled HIV presented in November 2024 with upper abdominal pain, fever, chills, dry cough, and dyspnea. Labs showed anemia, thrombocytopenia, neutrophilia, hypoalbuminemia, and elevated procalcitonin. No petechiae or skin lesions were noted. His CD4 was 66/μL and HIV viral load was 943 copies/mL, which was then diagnosed as AIDS. Chest X-ray and KUB were unremarkable. Abdominal CT demonstrated hepatosplenomegaly and multifocal lymphadenopathy (neck, axillae, supra-/infra-clavicular, mediastinum, paraaortic/aortocaval, iliac, inguinal). The patient subsequently received endotracheal intubation due to respiratory failure caused by septic shock.

Bone marrow biopsy showed normal cellularity without evidence of aplastic anemia, multiple myeloma, or myelodysplastic syndrome; Immune thrombocytopenic purpura (ITP) was then suspected. IVIG, methylprednisolone, HLA-matched single-donor platelet transfusion, romiplostim were prescribed, but thrombocytopenia still worsened. Splenectomy was then performed to reduce platelet clearance because of hepatosplenomegaly, but benefits was limited. Echo-guided lymph node biopsy revealed absent germinal centers to rule out lymphoma, but was incidentally positive for KSHV immunostaining. mNGS of blood also detected high titers of KSHV, EBV, and CMV. Gastrointestinal endoscopy did not show Kaposi-like lesions.

After months of treatment for AIDS and sepsis, multiple red, purple, brown, and black cutaneous lesions appeared on the neck, arms, thighs, buttocks, and back. Skin biopsy confirmed Kaposi sarcoma. Pegylated liposomal doxorubicin (Lipo-Dox) was initiated; further chemotherapy planned post-discharge.

Discussion

KS-IRIS arises from restoration of immune responses against KSHV after ART and may present as worsening pre-existing (paradoxical IRIS) or new-onset KS (unmasking IRIS), typically within days to six months of ART initiation. Risk factors include advanced tumor stage (T1), high pre-treatment HIV viral load (>5 log10 copies/mL), and detectable KSHV DNA. KS-IRIS carries substantial

morbidity and mortality, especially with visceral involvement. Management entails continuation of ART and systemic chemotherapy when indicated; glucocorticoids are generally contraindicated due to risk of severe KS exacerbation.
In this case, early KSHV evidence existed but KS could not be confirmed until clinical improvement; retrospectively, multifocal lymphadenopathy, hypoalbuminemia, and refractory thrombocytopenia likely reflected occult KS later "unmasked" by immune recovery.

Conclusion

KS remains a critical KSHV-related complication in AIDS. Cutaneous manifestations can be masked and then unmasked or acutely worsen with IRIS. Accurate identification and combined ART plus systemic therapy are essential for KS-IRIS management.

病例報告 114_C194

IgG4 相關性胰臟炎之案例報告

A Case Report of IgG4-Related Pancreatitis

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Introduction

IgG4-related pancreatitis, also known as type 1 autoimmune pancreatitis, is histologically defined by dense lymphoplasmacytic infiltration, storiform fibrosis, and elevated serum IgG4 levels. Clinically, it frequently mimics pancreatic malignancy, presenting with obstructive jaundice, pancreatic masses, or main pancreatic duct dilatation. Accurate distinction between autoimmune pancreatitis and pancreatic cancer is crucial to prevent unnecessary surgical intervention.

Case Report

A 70-year-old man with a medical history of type 2 diabetes mellitus and Barrett's esophagus presented with unintentional weight loss of 6 kg over five months. He reported no abdominal pain, jaundice, or other systemic symptoms. Abdominal ultrasonography revealed a 22.7-mm nodule in the pancreatic head with mild dilatation of the main pancreatic duct. Magnetic resonance cholangiopancreatography (MRCP) demonstrated two lesions: a 57-mm mass in the pancreatic tail and a 27-mm lesion in the head/uncinate process, both associated with ductal dilatation. Tumor markers were within normal limits. Endoscopic ultrasound-guided fine-needle biopsy (EUS-FNB) revealed chronic inflammatory changes, stromal fibrosis, and numerous crushed cells that stained strongly positive for IgG4. Malignancy was excluded. Serum IgG4 level was markedly elevated (1,570 mg/dL), confirming the diagnosis of IgG4-related pancreatitis. The patient was started on oral prednisolone 10 mg three times daily. After two months, abdominal ultrasonography demonstrated complete resolution of the pancreatic head nodule. A follow-up MRCP is scheduled. Notably, prior to corticosteroid therapy, his glycemic control was poor, with fasting plasma glucose levels of 170–180 mg/dL. After one month of prednisolone therapy, fasting glucose improved significantly to 106 mg/dL.

Discussion

In this case, the presence of two pancreatic lesions with unexplained weight loss initially raised strong concern for malignancy. However, histopathological findings together with a markedly elevated serum IgG4 level confirmed the diagnosis of IgG4-related pancreatitis. Corticosteroids remain the standard first-line therapy, and our patient showed a favorable response with regression of the pancreatic head lesion after two months of treatment. Interestingly, in patients with type 2 diabetes mellitus and autoimmune pancreatitis, corticosteroid therapy may paradoxically improve glycemic control by reducing pancreatic inflammation and partially restoring β-cell function, despite their well-recognized hyperglycemic potential. In our patient, fasting glucose improved substantially following prednisolone therapy, suggesting that the anti-inflammatory effects outweighed its diabetogenic risk.

Conclusion

This case highlights the importance of considering autoimmune pancreatitis in the differential diagnosis of patients with pancreatic masses and poorly controlled diabetes. Early recognition and timely corticosteroid therapy can result in rapid resolution of pancreatic lesions and improved glycemic control. Nevertheless, close long-term follow-up is warranted due to the high risk of relapse, and prompt retreatment is essential for favorable long-term outcomes.

病例報告 114_C195

近二十年台灣首例諾氏瘧原蟲感染一病例報告

The First Case of *Plasmodium knowlesi* Malaria in Taiwan in Past Twenty Years: A Case Report

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Introduction

Plasmodium knowlesi (*P. knowlesi*), a zoonotic malaria parasite endemic to Southeast Asia, can be as life-threatening as *Plasmodium falciparum* (*P. falciparum*) and requires prompt anti-malarial treatment. With climate change and globalization, *P. knowlesi* malaria poses an emerging threat to Taiwan. We report the first case of severe *P. knowlesi* malaria diagnosed in Taiwan in the past 20 years, occurring in a previously healthy adult traveler returning from the Philippines.

Case Report

A 33-year-old Caucasian man from the United States traveled to Palawan, Philippines, for three weeks, where he was exposed to contaminated stream water, seawater, and numerous mosquito bites in the rainforest. He arrived in Taiwan after the trip and, on the fifth day of his stay, developed one to two febrile episodes to 39 degree Celsius per day, accompanied by chills, headache, lethargy, tea-colored urine, and generalized myalgia. The illness progressed with dyspnea and oliguria, prompting presentation to the emergency department. Physical examination revealed mild scleral icterus without skin rash or eschar. Initial laboratory tests showed thrombocytopenia, elevated liver enzymes, and direct hyperbilirubinemia. Peripheral blood smear demonstrated a few ring forms and a suspected schizont in red blood cells, with a parasitemia of 0.1%. *P. knowlesi* infection was later confirmed by species-specific PCR at the CDC. Artemether–lumefantrine was administered for three days, leading to defervescence, clinical improvement, and clearance of parasitemia by the second day of treatment. Liver enzymes gradually returned to normal on follow-up.

Discussion

Plasmodium knowlesi, recognized as the fifth human malaria parasite, primarily affects residents of Southeast Asia but is increasingly reported among travelers. It is the most prevalent zoonotic malaria, with long-tailed and pig-tailed macaques as natural reservoirs and *Anopheles* mosquitoes as vectors. Although most malaria-related deaths are attributed to *P. falciparum* globally, severe *P. knowlesi* infections also occur and often require intensive management. Its short 24-hour erythrocytic life cycle contributes to severe disease by enabling high parasitemia, leading to impaired microvascular sequestration, reduced erythrocyte deformability, endothelial activation, glycocalyx degradation, and hemolysis, which lead to impaired perfusion and organ injury. Risk factors for severe disease include advanced age and manifestations such as respiratory distress, hypotension, and acute kidney injury. However, microscopic diagnosis is challenging due to its morphological similarity to *P. falciparum* and *P. malariae*, prompting the need for molecular alternative methods. Artemisinin-based combination therapy is recommended as first-line treatment, with intravenous formulations indicated in severe cases. The patient usually achieves

rapid recovery once timely treatment is initiated. With globalization and proximity to endemic regions such as Sabah and Sarawak in Malaysia, the Taiwan CDC has updated its malaria protocol, especially for *P. knowlesi* this year, highlighting the importance of awareness and prophylaxis.

Conclusion

P. knowlesi malaria can cause severe and life-threatening malaria as with *P. falciparum*, and the disease requires more recognition and early treatment in febrile travelers from endemic countries returning to Taiwan.

病例報告 114_C196

胰島素瘤合併胸腺增生及格雷夫斯氏病：類生長抑素受體配體治療經驗之病例報告

Concurrent Insulinoma, Thymic Hyperplasia, and Graves' Disease: Therapeutic Insights from Somatostatin Receptor Ligand Treatment

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Introduction

The simultaneous occurrence of insulinoma, thymic hyperplasia, and Graves' disease represents an exceptionally rare clinical entity with unique diagnostic and therapeutic implications. We report a case demonstrating this triad and the unexpected resolution of thymic hyperplasia following somatostatin receptor ligand (SRL) therapy.

Case Report

A 58-year-old woman with a history of Graves' disease presented with acute chest pain and was diagnosed with non–ST-elevation myocardial infarction (NSTEMI). She was treated with percutaneous coronary intervention (PCI) and dual antiplatelet therapy. Initial laboratory testing revealed significant hypoglycemia (blood glucose 2.6 mmol/L), and Whipple's triad was confirmed during admission. A supervised fasting test demonstrated endogenous hyperinsulinemic hypoglycemia. Imaging localized a 2.3 cm hyper vascular lesion in the pancreatic body, consistent with insulinoma, and incidentally revealed a 6.9 cm anterior mediastinal mass.

Surgery was deferred due to recent PCI and antiplatelet therapy, and long-acting octreotide (20 mg monthly) was initiated. This resulted in effective glycemic stabilization, shrinkage of the insulinoma (from 2.3 to 1.9 cm), and resolution of hypoglycemic symptoms. Remarkably, repeat imaging at ten months demonstrated complete disappearance of the mediastinal mass.

Discussion

The concurrent presentation of insulinoma, thymic hyperplasia, and Graves' disease represents an unprecedented clinical triad, potentially reflecting overlapping autoimmune and endocrine pathophysiology. This coexistence, complicated by significant cardiovascular comorbidity, poses distinctive diagnostic and therapeutic challenges.

Hypoglycemia constitutes a recognized adverse prognostic marker in acute coronary syndromes, particularly in non-insulin-treated patients [1]. Proposed mechanisms include catecholamine surge, proarrhythmic effects, endothelial dysfunction, and platelet activation impairing coronary perfusion [2]. Our patient's endogenous hyperinsulinemic hypoglycemia secondary to insulinoma may have contributed to the cardiovascular event, though the temporal relationship remains indeterminate.

Insulinoma, occurring in 1-4 cases per million annually, necessitates surgical resection as definitive therapy [3]. However, recent percutaneous coronary intervention requiring dual antiplatelet therapy precluded immediate surgical intervention, mandating medical management. Somatostatin receptor ligands effectively controlled hypoglycemia while unexpectedly achieving complete thymic resolution.

Thymic hyperplasia associates with autoimmune thyroid disease through thyroid-stimulating hormone receptor expression and thyroid-stimulating antibody stimulation in thymic tissue [4]. Characteristically, thymic regression follows achievement of euthyroidism. Remarkably, our patient demonstrated complete thymic resolution without significant thyroid functional changes, suggesting direct or indirect SRL effects on thymic tissue.

Somatostatin receptors (sst1, sst2A, sst3) are expressed in normal and hyperplastic thymic tissue [5]. Clinical evidence demonstrates SRL efficacy in advanced thymic tumors through antiproliferative and antiangiogenic mechanisms, including apoptosis induction and mitogenic pathway inhibition [6]. Our case substantiates potential broader therapeutic applications of SRLs in selected thymic lesions beyond glycemic control.

Conclusion

This case emphasizes that hypoglycemia warrants evaluation for coexisting endocrine or autoimmune disorders. In inoperable insulinoma, SRL therapy can be an effective and well-tolerated alternative and may impact other lesions. Notably, thymic hyperplasia regressed with SRL, suggesting potential therapeutic effects beyond glycemic control. This rare triad highlights the need for further study to elucidate underlying mechanisms and potential broader applications of SRL therapy.

病例報告 114_C197

肺部實變表現下之邊緣區淋巴瘤：85 歲男性病例報告

From Consolidation to Revelation: Marginal Zone Lymphoma in an 85-Year-Old Man

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Introduction

Primary pulmonary marginal zone lymphoma (MZL) of mucosa-associated lymphoid tissue (MALT) is a rare form of extranodal B-cell lymphoma, accounting for less than 1% of all primary pulmonary malignancies. Its radiologic appearance often mimics infectious consolidation or primary lung carcinoma, making accurate diagnosis particularly difficult. Because MZL tends to follow an indolent course and responds differently to therapy compared with lung carcinoma, accurate identification is crucial for proper management and prognosis.

Case Report

An 85-year-old man with a history of hypertension and dementia presented with progressive weakness, poor oral intake, and altered consciousness. Chest computed tomography revealed right middle lobe consolidation with total collapse, initially suggesting pneumonia or malignancy. Bronchoscopy demonstrated copious whitish secretions, raising suspicion for fungal infection. However, transbronchial biopsy showed monotonous small to medium-sized lymphoid cells with lymphoepithelial lesions. Immunohistochemical staining was positive for CD20, BCL-2, CD43, and MNDA, confirming the diagnosis of marginal zone lymphoma. Positron emission tomography (PET) demonstrated FDG uptake confined to the right middle lobe without systemic involvement, consistent with Ann Arbor stage IE disease.

Discussion

Pulmonary MZL typically presents as localized consolidation, nodules, or masses that lack the spiculated margins and rapid progression commonly observed in lung carcinoma. It is often misinterpreted as pneumonia or mucus impaction, particularly in elderly or immunocompromised patients. Histopathologic examination with immunohistochemistry remains the gold standard for diagnosis. The differential diagnosis includes infectious processes, organizing pneumonia, and other low-grade lymphoproliferative disorders. In localized disease, treatment options may include watchful waiting, surgical resection, or systemic therapy such as rituximab-based immunochemotherapy, depending on the clinical condition and disease extent.

Conclusion

This case serves as a reminder that primary pulmonary marginal zone lymphoma should be considered when pulmonary consolidation fails to resolve despite adequate therapy. In elderly patients with unusual or persistent radiographic findings, obtaining tissue confirmation through bronchoscopy is crucial for accurate diagnosis. Awareness of this uncommon disease helps clinicians tailor management appropriately and prevents overtreatment based on an incorrect assumption of primary lung cancer.

病例報告 114_C198

疑似 Afatinib 相關胰臟炎之 EGFR 突變肺癌患者

Suspected Afatinib-Associated Pancreatitis in a Patient with EGFR-Mutated Lung Cancer

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Introduction

Afatinib, an irreversible EGFR tyrosine kinase inhibitor, is widely used to treat EGFR mutation-positive non-small cell lung cancer. Although it is effective in cancer therapy, rare and serious adverse events, such as drug-induced pancreatitis, have not been previously reported. We describe a rare suspected case of Afatinib-associated pancreatitis in a patient with advanced lung adenocarcinoma.

Case Report

A 63-year-old woman with advanced lung adenocarcinoma underwent right upper lobectomy and wedge resection of the right lower lobe. Postoperative pathology confirmed an EGFR exon21 mutation, leading to initiation of Afatinib therapy. After starting Afatinib, she developed progressive weakness, poor appetite, and oral ulcers, with elevated serum lipase. Imaging studies—including KUB, abdominal CT with and without contrast, and abdominal sonography—showed no evidence of pancreatitis. Common causes of pancreatitis such as gallstones, bile duct obstruction, metabolic disorders, anatomical anomalies, infections, and trauma were all excluded.

Discussion

Given the temporal association with Afatinib administration and the absence of alternative etiologies, Afatinib was suspected as the likely trigger. Supportive care including fluid management and electrolyte correction led to symptomatic improvement and normalization of lipase. The patient was subsequently switched to an alternative EGFR inhibitor without recurrence of symptoms. This case highlights the challenges in identifying rare drug-induced adverse events when imaging does not provide confirmatory evidence.

Conclusion

Afatinib-associated pancreatitis is an extremely rare and previously unreported adverse event. Clinicians should remain vigilant for unexpected toxicities during targeted therapy, even in the absence of imaging findings, and consider prompt drug discontinuation and alternative treatment when necessary.

病例報告 114_C199

特發性肺纖維化患者之肺膠樣腺癌：罕見病例報告

Primary Pulmonary Colloid Adenocarcinoma in a Patient with Idiopathic Pulmonary Fibrosis: A Rare Case Report

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Introduction

Pulmonary colloid adenocarcinoma is a rare histological subtype of invasive adenocarcinoma, characterized by abundant extracellular mucin and relatively few malignant cells. Idiopathic pulmonary fibrosis (IPF) is a chronic and progressive interstitial lung disease that has been associated with an increased risk of lung cancer. Here, we present a rare case of pulmonary colloid adenocarcinoma in a patient with IPF.

Case Report

An 85-year-old man with idiopathic pulmonary fibrosis receiving antifibrotic therapy with nintedanib was found to have a 4.5-cm mass in the right lower lobe on non-contrast chest computed tomography (CT) during routine follow-up. The carcinoembryonic antigen level was 7.51 ng/ml with no other remarkable findings. The initial CT-guided transthoracic biopsy was inconclusive and complicated by pneumothorax. Positron emission tomography (PET) demonstrated 18F-fluorodeoxyglucose (FDG) uptake in the right lower lobe lesion, while contrast-enhanced brain magnetic resonance imaging and bone scintigraphy revealed no evidence of metastatic disease. A subsequent wedge resection was performed, and histopathological examination confirmed colloid adenocarcinoma. The tumor was staged as pT2bN0M0. The patient declined adjuvant chemotherapy and has remained alive under surveillance for approximately nine months after surgery.

Discussion

Pulmonary colloid adenocarcinoma was classified as a rare variant in the 2015 WHO classification of lung tumors. It is characterized by alveolar wall destruction with extensive extracellular mucin pools and relatively few malignant cells. Owing to its high mucin content and low malignant cell density, pulmonary colloid adenocarcinoma typically presents as an intrapulmonary mass with poor contrast enhancement on CT and low ^{18}F-FDG uptake on PET scan. Biopsy diagnosis is often challenging, and surgical resection is frequently required to establish a definitive diagnosis.

IPF is a well-recognized risk factor for lung cancer, with squamous cell carcinoma being the most common histological type. However, coexistence with pulmonary colloid adenocarcinoma has not been reported in the available literature. The presence of emphysema and honeycombing increases the risk of pneumothorax, while fibrotic tissue surrounding the lesion often leads to non-diagnostic biopsy results. These factors further complicate the diagnosis of colloid adenocarcinoma in patients with IPF. Surgical resection carries a significantly higher risk of acute exacerbation, which is associated with poor prognosis. In contrast, complete resection of localized colloid adenocarcinoma has been associated with favorable outcomes. For the limited number of

reported cases, the overall prognosis remains uncertain and warrants further investigation.

Conclusion

This case highlights the rare occurrence of colloid adenocarcinoma in patients with IPF. The coexistence of IPF and colloid adenocarcinoma makes diagnosis and management more challenging. Fibrotic tissue surrounding the lesion, together with the mucinous content of the tumor, increases the probability of a nondiagnostic biopsy, while invasive diagnostic procedures and surgical interventions carry an elevated risk of acute exacerbation of IPF, which is associated with poor prognosis. Our case demonstrates a successful surgical resection and underscores the importance of vigilance regarding potential complications in this high-risk population.

病例報告 114_C200

以多發性肺部結節為表現之轉移性副神經節瘤：個案報告

Metastatic Paraganglioma Presenting As Multiple Pulmonary Nodules: A Case Report

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Introduction

Multiple pulmonary nodules are common seen in malignancy lung cancers or metastatic malignancy, especially in patients with known solid organ tumors. Other differential diagnosis also includes sarcoidosis, infectious diseases, autoimmune diseases and interstitial lung disease. Diagnostic approach requested history taking, biochemistry profile, image study, microbiology result and finally with pathological confirmation.

Case Report

We reported on a 71-year-old female who was admitted for unexplained lower limb edema and shortness of breath, and imaging studies revealed multiple bilateral pulmonary nodules. After a series of diagnostic workups, the patient ultimately underwent video-assisted thoracoscopic surgery (VATS) with wedge resection, as the initial CT-guided biopsy tissue was insufficient for a definitive diagnosis.

Discussion

Pathological findings of VATS revealed paraganglioma with metastasis. Paraganglioma typically occurs in the head and neck or adrenal glands, and pulmonary metastasis is extremely rare, which significantly increases diagnostic difficulty. The nonspecific cardiovascular symptoms in this case were likely related to this type of tumor. Immunohistochemical staining of chromogranin A, synaptophysin, GATA3 would be helpful in confirming the differentiation of paraganglioma and assessing the difference between primary and metastatic paraganglioma.

Conclusion

This case serves as a reminder to clinicians to consider this rare entity such as paraganglioma in the differential diagnosis when faced with similar presentations and inconclusive routine examinations, especially in patients with multiple pulmonary nodules.

病例報告 114_C201

藥物性肺損傷與感染造成間質性肺病的診斷

Diagnostic Challenge in Distinguishing Drug-induced Lung Injury from Infection-related Interstitial Changes

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Introduction

Influenza A pneumonia frequently reveals diffuse ground-glass opacities, centrilobular nodules, or tree-in-bud patterns. Such radiologic features are associated with severe pneumonia, and episodes of intense inflammation may predispose to subsequent interstitial lung disease. Gemcitabine, a nucleoside analog, is widely used in the treatment of breast cancer, non-small cell lung cancer and other malignancies. Although gemcitabine is generally well tolerated, pulmonary toxicity—including interstitial lung disease—has been recognized, with higher-grade adverse events (grade III/IV) occurring infrequently.

Case Report

A patient who recently commenced gemcitabine for recurrent cholangiocarcinoma presented with a new usual interstitial pneumonia (UIP) pattern at both lung bases on computed tomography, in the absence of notable preceding respiratory symptoms. Gemcitabine-induced pulmonary toxicity was initially considered the likely cause. However, subsequent bronchoalveolar lavage unexpectedly identified an influenza A infection, creating a diagnostic dilemma: was the interstitial lung disease attributable to the drug or the viral infection? Following antiviral treatment, the patient was re-challenged with gemcitabine, and the previously observed interstitial pneumonia did not recur.

Discussion

There is limited discussion in the medical literature regarding the potential additive risk of interstitial lung disease when influenza infection coincides with gemcitabine therapy.

Conclusion

While gemcitabine-induced pulmonary toxicity was initially considered, subsequent bronchoalveolar lavage identified influenza A infection, complicating attribution of the interstitial lung disease to either etiology.

病例報告 114_C202

內視鏡超音波在鑑別鼻咽 IgG4 相關疾病與鼻咽癌中的角色

The role of EUS in differentiating nasopharyngeal IgG4-related disease from nasopharyngeal carcinoma

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Introduction

IgG4-related disease is uncommon, with nasopharyngeal involvement being particularly rare in literature. We present a case of an IgG4-related nasopharyngeal mass that resembled malignancy.

Case Report

A 50-year-old man, who denied any underlying chronic systemic diseases, reported experiencing left-sided tinnitus, nasal and aural fullness, and headaches for three months. A brain magnetic resonance imaging scan revealed left nasopharyngeal carcinoma. Biopsies obtained via nasopharyngoscopy and trans-nasal ultrathin gastroscope both indicated chronic inflammation. Notably, the combination of trans-nasal ultrathin gastroscope and endoscopic ultrasound revealed a swollen left nasopharynx with an intact mucosal lining and a thickened submucosal layer. A subsequent computed tomography-guided biopsy of the left nasopharynx indicated IgG4-related disease.

Discussion

To our knowledge, this was the first report using endoscopic ultrasound to evaluate the submucosa inflammatory process of nasopharynx. Treatment for IgG4-related disease usually includes corticosteroids, which have been shown to reduce inflammation and improve symptoms. Early recognition of IgG4-related disease with nasopharyngeal involvement is essential for effective management and differential diagnosis to prevent potential complications.

Conclusion

Nasopharyngeal EUS is essential for distinguishing between mucosal and submucosal processes, aiding subsequent biopsy procedures and ultimately enabling the definitive diagnosis of nasopharyngeal IgG4-RD.

病例報告 114_C203

內視鏡肌間剝離術對於早期直腸癌病人之治療效果：一例病例報告

A case report of endoscopic intermuscular dissection in a patient with early rectal cancer

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Introduction

Endoscopic intermuscular dissection (EID) is a novel endoscopic approach for a patient with early rectal cancer, whose lesion locates near the anal verge. We aim to determine the treatment effect of EID for such patients.

Case Report

A 70-year-old male received colonoscopy due to positive fecal immunochemical test, and it showed one 3cm tumor at rectum, which located at 4cm above anal verge. Abdominal computed tomography showed rectal tumor 2.2cm, and AJCC 8th edition staging status was T2N0M0. In addition, initial laboratory survey revealed intact renal function and liver function, and no anemia nor leukocytosis was found. Endoscopic ultrasound (EUS) revealed that the tumor invaded at least deep submucosa and superficial muscularis propria layer. We performed EID for en-bloc resection in view of the rectal tumor, and no abdominal pain, bloody stool nor peritoneal sign was noted after the procedure. The final pathological report showed rectal adenocarcinoma, which invaded the submucosa layer (pT1), and the horizontal margin and vertical margin were 5mm in distance, respectively. Furthermore, no lymphovascular invasion was reported. Regarding the invasion depth of the rectal cancer, the patient received adjuvant concurrent chemoradiotherapy (CCRT) after the EID, and no specific post-EID complication was reported at out-patient clinic follow-up so far.

Discussion

The risk of lymph node metastasis associated with deep submucosal invasion should be balanced against the mortality and morbidity of total mesorectal excision (TME). Dissection through the submucosa hinders radical deep resection, and full-thickness resection may influence the outcome of completion TME. EID in between the circular and longitudinal part of the muscularis propria could potentially provide an R0 resection while leaving the rectal wall intact.

Conclusion

EID for early rectal cancer appears to be feasible and safe, and the high R0 resection rate creates the potential of rectal preserving therapy.

病例報告 114_C204

以難治性肺炎為表現之肺部淋巴瘤：病例報告

Non-resolving Pneumonia Revealing Extranodal Marginal Zone Lymphoma: A Case Report.

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Introduction

Pulmonary lymphoma is a rare entity, classified as primary pulmonary lymphoma (PPL) in the absence of extrapulmonary disease within 3 months of diagnosis, or secondary pulmonary lymphoma (SPL) when systemic involvement is present. PPL accounts for only 0.4% of all lymphomas, with mucosa-associated lymphoid tissue (MALT) lymphoma as the most common subtype. By contrast, SPL is more often diffuse large B-cell lymphoma (DLBCL). Clinical and radiologic features are often nonspecific, and histopathological confirmation is essential for diagnosis.

Case Report

We present a 62-year-old male university teacher, an ex-smoker with a 20 pack-year history, who was admitted with chest tightness and bilateral pulmonary consolidations unresponsive to 1 week of levofloxacin. He reported only mild exertional dyspnea, and examination revealed bilateral crackles. Chest CT demonstrated diffuse ground-glass opacities and consolidations. Autoimmune markers were negative, and microbiologic work-up, including Mycobacterium tuberculosis PCR and Pneumocystis jirovecii PCR, was unremarkable. Bronchoalveolar lavage showed B-cell predominance (88.5%), and CT-guided biopsy confirmed low-grade B-cell lymphoma, consistent with pulmonary MALT lymphoma. PET-CT revealed intense gastric uptake, and endoscopy confirmed synchronous gastric MALT lymphoma. The patient was diagnosed with extranodal marginal zone lymphoma (MZL, Ann Arbor stage IV, IPI 3, ECOG 1). He was started on rituximab plus bendamustine with concurrent tenofovir prophylaxis for chronic hepatitis B. Treatment was well tolerated except for grade I fatigue, and follow-up chest imaging after four cycles demonstrated disease regression.

Discussion

This case highlights the diagnostic challenge of pulmonary lymphoma, which often mimics infection. PPL with indolent histology, such as MALT lymphoma, generally carries an excellent prognosis, whereas aggressive subtypes and SPL are associated with poorer outcomes. Importantly, pulmonary MALT lymphoma retains its indolent nature irrespective of primary or secondary presentation. Histopathological confirmation and comprehensive staging, including PET-CT and gastrointestinal evaluation, are essential to establish diagnosis. Our case underscores the importance of considering lymphoma in non-resolving pneumonia and supports rituximab–bendamustine as an effective treatment for advanced MZL.

病例報告 114_C205

嚴重咳嗽引起意外之外的卡倫征(Cullen)以及葛雷一特那氏(Grey-Turner)徵象

Unexpected Cullen and Grey-Turner Signs Caused by Severe Coughing

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Introduction

Cullen sign and Grey-Turner's sign, known as ecchymosis over peri-umbilicus and bilateral flanks, have been classically described as retroperitoneal hemorrhage with most commonly associated with hemorrhagic pancreatitis. Both have been seen in <1% of individuals with acute pancreatitis that implies bad prognosis of the disease. Yet, they can appear in other diseases. Rare associations include decompensated liver cirrhosis with portal hypertension, hepatocellular carcinoma rupture and neuroendocrine lung carcinoma with liver extension. (1,2,3) On rare situation, it might be in association with severe coughing as like ours.

Case Report

An 82-year-old man with medical history of ischemic stroke, end-stage renal disease on hemodialysis, and long-term aspirin therapy presented with right flank pain for 2 days. He reported severe bouts of coughing due to upper respiratory tract infection 5 days earlier. Examination revealed ecchymoses over the umbilicus (Cullen sign) and the right flank (Grey-Turner sign). Laboratory tests included normal amylase, lipase, and coagulation profile. Computed tomography revealed no retroperitoneal hemorrhage but demonstrated a hematoma in the right internal oblique muscle, consistent with a cough-induced abdominal wall hematoma.

Discussion

Cullen and Grey-Turner signs are classically associated with hemorrhagic pancreatitis or retroperitoneal bleeding. This case highlights that cough-induced hematoma may be a less common cause of these signs but important to recognize. The cause of cough-induced hematoma is unknown, but it can be speculated that increased intra-abdominal pressure from severe coughing may have caused shear stress on the muscle that eventually leads to the rupture of small intramuscular vessels at internal oblique muscle. Hence, not every occurrence of Cullen and Grey-Turner signs indicates retroperitoneal hemorrhage, as illustrated in this case.

Conclusion

We reported an unexpected finding of Cullen sign and Grey Turner sign caused by severe coughing.

病例報告 114_C206

料想不到的 Stabler Sign

The Unexpected Stabler Sign

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Introduction

Ecchymotic cutaneous signs, such as Grey Turner, Cullen, Fox, and Stabler signs, are well-recognized markers of underlying internal hemorrhage. Among these, Stabler sign—ecchymosis of the inguinal region—classically indicates retroperitoneal bleeding, most frequently associated with ruptured abdominal aortic aneurysm, but also reported in trauma, anticoagulation therapy, and portal hypertension. (1,2) These signs reflect blood tracking along fascial planes to superficial cutaneous sites and therefore provide important diagnostic clues.

While Stabler sign has been consistently attributed to retroperitoneal sources, extraperitoneal etiologies are rarely described. We report a rare case where Stabler sign was produced by extraperitoneal extension of a breast hematoma, highlighting the mechanism behind the cause of the ecchymosis.

Case Report

A 33-year-old woman presented with ecchymosis over the right inguinal region for one week. She denied alcohol use or history of pancreatitis. She had undergone right breast surgery for a fibrocystic nodule three days before onset of bruising. On examination, swelling and ecchymosis were observed over the right breast and right inguinal area—resembling a Stabler sign. Laboratory tests revealed normocytic anemia (Hb drop from 14.0 to 11.2 g/dL) without elevated amylase or lipase. CT excluded retroperitoneal hemorrhage and showed a large right breast hematoma with blood tracking along chest, abdominal, and inguinal fascial planes.

Discussion

Cullen and Grey-Turner signs are classically associated with hemorrhagic pancreatitis or retroperitoneal bleeding. This case highlights that cough-induced hematoma may be a less common cause of these signs but important to recognize. The cause of cough-induced hematoma is unknown, but it can be speculated that increased intra-abdominal pressure from severe coughing may have caused shear stress on the muscle that eventually leads to the rupture of small intramuscular vessels at internal oblique muscle. Hence, not every occurrence of Cullen and Grey-Turner signs indicates retroperitoneal hemorrhage, as illustrated in this case.

Conclusion

We demonstrate that Stabler sign can result from extraperitoneal blood tracking along fascial planes from a breast hematoma to the inguinal region. To our knowledge, this is the first reported case illustrating the pathophysiologic mechanism linking extraperitoneal bleeding to cutaneous ecchymosis.

病例報告 114_C207

以黃疸為初始表現的轉移性前列腺癌：病例報告

A Rare Initial Presentation of Metastatic Prostate Adenocarcinoma with Cholestatic Jaundice: A Case Report

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Introduction

Cholestatic jaundice is most commonly attributed to hepatobiliary obstruction, viral hepatitis, or drug-induced liver injury. Rarely, it can present as a paraneoplastic manifestation of extrahepatic malignancies in the absence of hepatic metastasis, a phenomenon described as Stauffer's syndrome. First recognized in renal cell carcinoma, similar hepatic dysfunction has occasionally been reported in other solid tumors, including prostate adenocarcinoma. We report a patient with advanced prostate adenocarcinoma who developed severe cholestatic jaundice, representing a paraneoplastic manifestation mimicking Stauffer's syndrome.

Case Report

A 69-year-old male farmer presented with progressive jaundice for two weeks. He had a history of hypertension, dyslipidemia, and gout, managed with olmesartan, fenofibrate, and benzbromarone. He also reported hematuria and voiding difficulties for one month. He denied alcohol, tobacco or betel nut use. On admission, the patient was alert, thin (BMI 19 kg/m^2), and icteric but afebrile and hemodynamically stable. Abdominal examination revealed no hepatosplenomegaly or tenderness. Laboratory studies showed marked direct hyperbilirubinemia (total bilirubin 23.4 mg/dL, direct bilirubin 16.9 mg/dL), elevated alkaline phosphatase (312 U/L), but only mild AST/ALT elevation. Albumin was slightly decreased (3.6 g/dL), and tumor markers including AFP, CEA, CA19-9, and SCC antigen were within normal limits. Viral and autoimmune hepatitis serologies were negative. Imaging by abdominal ultrasound revealed no bile duct dilatation. PSA was profoundly elevated at >2000 ng/mL. Prostate biopsy confirmed adenocarcinoma, Gleason score 4+5 (Grade Group 5), stage pT2c with nodal involvement and bone metastases. The patient was initiated on androgen deprivation therapy with leuprorelin and bicalutamide. After treatment of prostate cancer, following data showed improved bilirubin level. He was diagnosed with metastatic prostate adenocarcinoma complicated by paraneoplastic cholestatic jaundice resembling Stauffer's syndrome.

Discussion

This case illustrates a rare paraneoplastic manifestation of prostate cancer. Stauffer's syndrome, classically described in 3–20% of renal cell carcinoma, is characterized by intrahepatic cholestasis, elevated alkaline phosphatase, hypoalbuminemia, prolonged prothrombin time, and hypergammaglobulinemia without hepatic metastasis. Although most commonly associated with renal tumors, similar cholestatic presentations have been reported in prostate and other malignancies. The proposed mechanism involves tumor-derived cytokines, particularly IL-6, inducing systemic inflammation and downregulation of hepatobiliary transporters. Elevated IL-6

correlates with higher ALP and anemia, and normalization of hepatic dysfunction after tumor-directed therapy supports its paraneoplastic nature. Our patient's laboratory profile of predominant cholestasis, absence of biliary obstruction or hepatic metastasis, and advanced prostate cancer fulfills the clinical spectrum of this entity. Temporal association with prostate cancer progression and partial improvement after systemic therapy suggests a paraneoplastic contribution. Recognition of this syndrome is critical, as misdiagnosis may delay appropriate oncologic management.

Conclusion

We present a rare case of metastatic prostate adenocarcinoma complicated by cholestatic jaundice. This paraneoplastic hepatic dysfunction, though classically associated with renal cell carcinoma, can also occur in prostate cancer. Clinicians should maintain awareness of this unusual presentation, especially in patients with unexplained cholestasis and elevated PSA, as timely recognition and initiation of androgen deprivation therapy may improve both hepatic function and oncologic outcome.

病例報告 114_C208

經橈動脈行腎交感神經阻斷術及生物可吸收冠狀動脈支架植入後 11 年之血管攝影

Renal Denervation for Resistant Hypertension and Coronary Angiography After 11 years of Bioresorbable Vascular Scafford Implantation via Radial Artery

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Introduction

Renal denervation (RDN) has been adopted as a treatment option for resistant hypertension (rHT). The procedure is usually performed via the femoral artery route, which requires hemostasis in bed afterward. Bioresorbable vascular scaffolds (BVS) were once regarded as a new generation of coronary stents. Here, we report a case of RDN and coronary angiography after BVS implantation via radial artery access.

Case Report

An 83-year-old male with a longstanding history of hypertension. He underwent coronary angiography (CAG) in 2014 due to suspected unstable angina, during which BVS were deployed in the proximal to mid-left anterior descending artery (LAD) and the mid-left circumflex artery (LCX). Despite optimal medications, his blood pressure remained poorly controlled, and renal function deterioration was noted. Ambulatory blood pressure monitoring (ABPM) revealed a 24-hour average BP of 138/67 mmHg. Given the high-risk profile and organ damage, resistant hypertension (rHT) was diagnosed, and RDN was considered.

We used a proximal left radial artery approach to avoid prolonged lying and hemostasis in the ICU. CAG showed patent coronary arteries after 11 years of BVS deployment, with no restenosis. Bilateral renal artery angiography revealed suitable anatomy for RDN. The procedure was successfully performed with standard pre-medications for pain control at the ablation sites and points. Hemostasis at the radial artery access site was achieved, and the patient was able to mobilize immediately after the procedure. He was discharged the next day. Three months later, ABPM showed a blood pressure of 134/72 mmHg after discontinuing one antihypertensive medication, and his renal function had also improved.

Discussion

According to domestic guidelines, RDN can currently be considered for hypertensive patients with high-risk profiles, those intolerant or nonadherent to antihypertensive medications, or with features indicative of neurogenic hypertension. The procedure is generally safe and simple, with rare complications. However, the need to use the femoral approach in most cases may be a barrier. We chose the left radial artery with a more proximal puncture level to ensure the length of the RDN device was sufficient to complete the procedure, in addition to providing greater comfort and fewer access site precautions.

BVS was once popular due to the assumption that coronary stents would be absorbed within two to three years, preserving vessel elasticity without leaving foreign implants. However, it was withdrawn from the market due to issues with stent thrombosis. Recent trials have shown that the

risk of adverse events is comparable to that of ordinary drug-eluting stents once the BVS has been fully absorbed. Our patient received the intervention 11 years ago and has remained free from clinical events; CAG also showed acceptable vessel patency.

Conclusion

RDN can be performed via the radial artery approach with careful selection and strategy.

病例報告 114_C209

肺炎黴漿菌感染誘發幼年型系統性紅斑狼瘡：病例報告

Mycoplasma pneumoniae Infection Triggering Juvenile-Onset Systemic Lupus Erythematosus: A Case Report

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Introduction

Systemic lupus erythematosus (SLE) is a complex autoimmune disease with multifactorial etiology. Infectious triggers are well recognized in amplifying disease activity, yet Mycoplasma pneumoniae–associated lupus onset remains rarely reported. Awareness of such associations is crucial in pediatric and adolescent populations, where initial manifestations may mimic infection alone.

Case Report

A 15-year-8-month-old girl with prior juvenile idiopathic arthritis was admitted with high-grade fever, vomiting, diarrhea, and respiratory complaints. Serology was positive for Mycoplasma pneumoniae IgM and group A streptococcal antigen. Despite Doxycycline and Ampicillin-sulbactam, her fever recurred, accompanied by worsening cytopenias, elevated inflammatory markers, and hemodynamic instability. Laboratory evaluation revealed ANA 1:10,240, anti-Sm/RNP positivity, Coombs-positive hemolytic anemia, hyperferritinemia, hypertriglyceridemia, and hypocomplementemia, fulfilling 2019 EULAR/ACR criteria for SLE. She required intensive care and received methylprednisolone pulse therapy plus intravenous immunoglobulin, followed by oral corticosteroids and Hydroxychloroquine, with marked improvement.

Discussion

This case illustrates the pivotal role of infection in triggering autoimmune activation. Mycoplasma pneumoniae is a common respiratory pathogen but may also act as an initiator of SLE flare or even new disease onset through molecular mimicry and B-cell activation. Clinicians focusing solely on pneumonia risk overlooking underlying autoimmunity. Early recognition of red flags—persistent cytopenias, complement consumption, evolving autoantibodies—can distinguish refractory infection from infection-triggered lupus activation.

Conclusion

Mycoplasma pneumoniae infection may precipitate juvenile-onset SLE and lead to fulminant disease activity if unrecognized. Careful evaluation of persistent systemic abnormalities in post-infectious settings enables timely immunosuppressive therapy and improved outcomes.