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Case Report

Benign biliary stricture associated with Castleman disease – a case report

Oscar I. D. Rodriguez[1], Carlos A. P. Muñoz[2]*, José Á. I. Juárez[2]

[1]Universidad Autónoma de San Luis Potosí, México
[2]Department of General Surgery, HRAE "Dr. Ignacio Morones Prieto", San Luis Potosí, México

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***Correspondence:**
Dr. Carlos A. P. Muñoz,
E-mail: dr.carlos.pemu@gmail.com

ABSTRACT

Castleman disease is a rare, heterogeneous group of lymphoproliferative disorders. It is classified as unicentric or multicentric. The unicentric type involves the growth of a single lymph node or a cluster of lymph nodes in the same region, which may present incidentally or cause obstructive symptoms depending on the anatomical location. We present the case of a 45-year-old male who developed abdominal pain in the epigastrium and upper right quadrant, pruritus, and jaundice for two months. He was diagnosed with a benign biliary stricture associated with Castleman disease, which was managed successfully with surgical intervention. We conclude that this condition is highly uncommon, presents a wide spectrum of differential diagnoses, and tends to recur; therefore, surgical resolution is suggested.

Keywords: Castleman disease, Benign biliary stricture, Jaundice, Case report

INTRODUCTION

Unicentric Castleman disease (UCD) is a heterogeneous disorder that may arise in any site containing lymphoid tissue. It may be clinically evident during physical examination, such as cervical or mediastinal lymphadenopathy, or incidentally detected on chest radiographs or computed tomography.[1]

However, when located in the abdomen, UCD represents a diagnostic challenge, frequently raising a broad spectrum of differential diagnoses.

We report a case of UCD located in the superior peripancreatic region that caused extrinsic compression of the common bile duct, leading to jaundice, pruritus, and a cholestatic pattern in laboratory tests.

We also consider that this condition triggered a local inflammatory reaction, resulting in a benign biliary stricture at this level.

CASE REPORT

We present the case of a 45-year-old male with a history of alcohol consumption, occasional smoking, appendectomy 15 years earlier, and a motor vehicle accident 20 years earlier without complications. He lived in precarious conditions with exposure to equines, goats, cattle and pigs.

The illness began in February 2024 with colicky epigastric pain radiating to the upper right quadrant generalized pruritus, and low-grade fever. Two weeks later, jaundice developed, prompting evaluation at a referral hospital. Laboratory findings showed a cholestatic pattern (total bilirubin 4.3 mg/dl, direct bilirubin 2.53 mg/dl, alkaline phosphatase 1540 U/l, γ-GT 482 U/l). Ultrasound and computed tomography revealed gallbladder sludge and dilatation of intra- and extrahepatic bile ducts. He was referred to our center with a presumptive diagnosis of choledocholithiasis but without complete imaging studies.

On admission, the patient presented with persistent colicky pain in the epigastrium and right hypochondrium, fever, dark urine, pale stools, and unquantified weight loss. Physical examination revealed jaundice and right upper quadrant tenderness with a positive Murphy's sign. Vital signs were stable. Admission labs: total bilirubin 7.36 mg/dl, direct bilirubin 5.95 mg/dl, AST 152 U/l, ALT 256 U/l, consistent with choledocholithiasis and mild cholangitis.

Laparoscopic cholecystectomy with transcystic cholangioscopy was performed, revealing a bile duct stricture (Figure 1). Conversion to open surgery was required, with cholecystectomy, T-tube placement, and biopsies. Subsequent cholangiography demonstrated a filling defect in the suprapancreatic portion of the common bile duct.

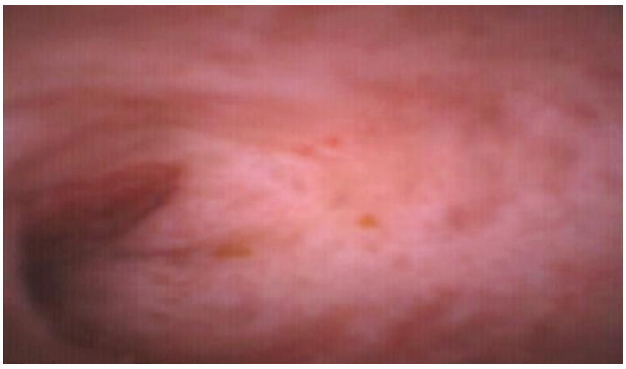

Figure 1: Cholangioscopic view of a common bile duct stricture.

Due to persistent cholestasis, magnetic resonance cholangiopancreatography (MRCP) was performed, revealing bile duct dilation 14.2 mm (Figure 2). ERCP with biliary stent placement achieved temporary resolution of the stricture. Histopathology reported lymphoid tissue with hyaline vascular features consistent with Castleman disease. Given the benign nature of the stricture and initial resolution with endoscopic management, a conservative approach with close follow-up (liver function tests, tumor markers, and imaging) was chosen.

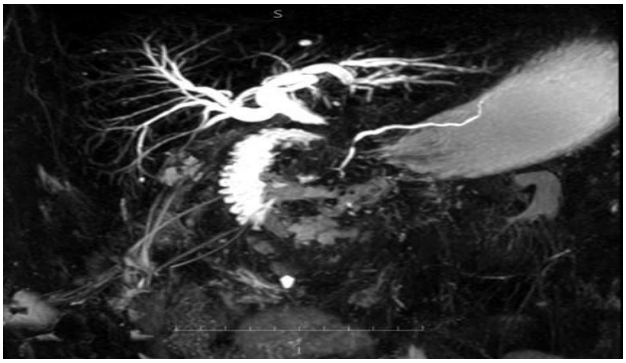

Figure 2: Common hepatic duct stricture visualized by magnetic resonance cholangiopancreatography (MRCP).

However, after 11 months, a follow-up MRCP revealed restenosis in the suprapancreatic and pancreatic segments of the bile duct. Tumor markers were negative, and a Roux-en-Y hepaticojejunostomy was performed (Figure 3). The patient subsequently showed favorable postoperative recovery with clinical and biochemical improvement. He was discharged after two weeks, with outpatient follow-up at one week and then monthly with imaging and labs, remaining asymptomatic to date.

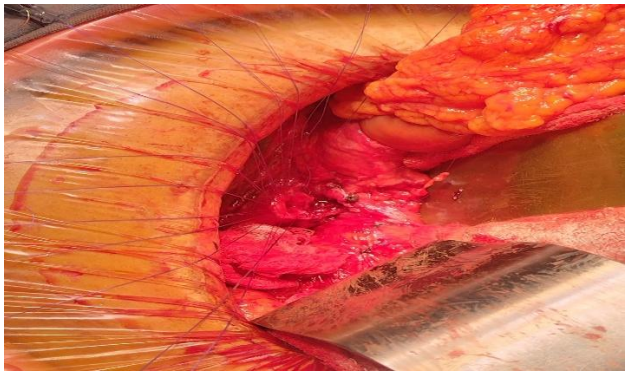

Figure 3: Roux-en-Y hepaticojejunostomy.

DISCUSSION

Castleman disease comprises a spectrum of rare, primarily benign lymphoproliferative disorders. It is mainly classified into unicentric and multicentric forms. Additional subtypes include idiopathic multicentric disease, HHV-8–associated multicentric disease, and multicentric disease associated with POEMS syndrome (polyneuropathy, organomegaly, endocrinopathy, monoclonal plasma cell disorder, and skin changes).[2] First described by Benjamin Castleman in 1954, several histopathological variants have since been identified.[4]

UCD refers to localized lymphoid enlargement of a single node or group of nodes.[3,5] Its association with benign biliary stricture is extremely rare, with only six cases reported to date. This makes the condition poorly understood in terms of diagnosis and management. The etiology of UCD remains unknown, with no established risk factors.[1]

The typical age of presentation is in the third to fourth decade of life, with a female predominance, particularly in Asian populations.[5] However, reported cases of UCD associated with biliary stricture occurred in patients aged 48–62 years, predominantly in Asian males.[6-10]

According to Hoffman, common anatomical sites include the abdomen (34.4%), thorax (33.6%), head and neck (22%), axilla (4.3%), inguinal region and pelvis (3.5%), with other sites being less frequent (2.3%). Clinically, Castleman disease may be present incidentally on imaging or with obstructive symptoms. Reported symptoms include pain, weight loss, night sweats, and fever.[3,5] In cases with biliary involvement, common manifestations

include epigastric/right hypochondrial pain, fever, jaundice, pruritus, nausea, vomiting, and weight loss—findings that overlap with many other biliary pathologies.[4,6-10]

Laboratory findings in UCD are often nonspecific. In approximately 20% of cases, elevated CRP, anemia, hypoalbuminemia, thrombocytopenia, or hypergammaglobulinemia have been described.[1,2,5] Our patient had no such alterations. In published cases, however, liver function tests revealed obstructive patterns.[4,6-10] One patient had anemia and leukocytosis, and another had markedly elevated CA 19-9 levels (900 U/ml).[4,8]

Imaging plays a central role in diagnosis. Ultrasound, CT, MRI, and PET-CT/PET-MRI may provide valuable information, though findings are often inconclusive.[3] Cholangioscopy has not been widely reported for these lesions, though we consider it valuable in confirming strictures.

Histopathology is essential for diagnosis and classification into hyaline vascular, plasma cell, or mixed subtypes.[1] Four published cases showed hyaline vascular type, as in our patient, while two demonstrated plasma cell infiltration.[4,6-10]

Diagnosis of UCD requires both imaging showing enlarged lymph nodes and compatible histopathology.[2] The treatment of choice is surgical resection, due to its high effectiveness and low recurrence. However, management depends on anatomical location, tumor size, and adjacent vascularity, with some cases suitable for close observation and favorable outcomes.[2,3]

Adjuvant options include monoclonal antibodies (rituximab), corticosteroids, or radiotherapy, which may be considered in selected patients depending on clinical course.[1]

CONCLUSION

The combination of Castleman disease and benign biliary stricture is exceedingly rare, with poorly understood pathophysiology. Clinical presentation overlaps with many biliary diseases, making diagnosis challenging. Based on our experience, given the recurrent nature of strictures in this setting and the limited long-term benefit of repeated endoscopic interventions, we suggest biliodigestive derivation (Roux-en-Y hepatico-jejunostomy) as the most viable definitive management option.

In conclusion, this rare association remains a diagnostic challenge that must be supported by both imaging and pathology.

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