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CASE REPORT

The Odd Gallbladder—a Rare Case of Gallbladder and Lymph Node Sarcoidosis: a Case Report and Review of the Literature

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Introduction

Gallbladder sarcoidosis is exceedingly rare. [1] To date, there are only seven cases reported in the literature. We present a rare case of gallbladder and lymph node sarcoidosis and a review of the literature. Informed consent was obtained from the participating individual.

Case Report

A 67-year-old female was referred to our clinic 9 years ago for surveillance of fatty liver and incidental gallbladder polyps. Her past medical history includes hypertension, hyperlipidemia, diverticular disease and right breast carcinoma post wide local excision and radiotherapy.

The gallbladder polyp at presentation was 4 mm, and has grown to 7 mm in 2017 (Fig. 1). Concomitantly, she also developed dyspepsia and underwent a gastroscopy which revealed antral gastritis and a fundic gland polyp at the greater curve of the stomach. Patient was then treated conservatively for chemical gastritis with oral PPI. She was subsequently offered laparoscopic cholecystectomy in view of the size of the polyps and worsening biliary colic. Pre-operative liver function test was unremarkable.

She underwent laparoscopic cholecystectomy and peritoneal nodules excision biopsy. Intra-operative findings were a grossly fatty liver and multiple peritoneal inflammatory tissues, which were excised for histology. The operation was uneventful and she was discharged well and stable on post-operative day 1. Routine follow-up in a specialist outpatient clinic revealed a new complaint of loose stools but had since resolved with dietary modification.

Histology of the gallbladder returned as chronic cholecystitis with polypoid cholesterolosis. The gallbladder lymph node showed noncaseating granulomata (Fig. 2). Acid-fast bacilli (AFB) were negative. Histology of peritoneal nodules revealed fibroadipose tissue with multiple granulomata and no evidence of malignancy. Her overall histology findings are highly suggestive of sarcoidosis.

Subsequent outpatient rheumatology follow-up organised a myriad of autoimmune serology including angiotensin-converting enzyme (ACE) levels, CT imaging and TB screening test all of which returned within normal limits. Chest X-Ray and CT were normal without parenchymal lesions or hilar/mediastinal lymphadenopathy. Despite a normal echocardiogram in 2015, an ejection systolic murmur was auscultated, and hence, she was sent for a repeat echocardiogram to exclude myocardial sarcoidosis and is for further cardiology management.

Discussion

Sarcoidosis is a chronic, multisystem inflammatory disorder with unknown aetiology in the presence of noncaseating granulomas within involved organs. The condition affects people worldwide regardless of gender, ethnicity or age, with a global prevalence of 10–20 per 100,000 individuals [2]. The peak age

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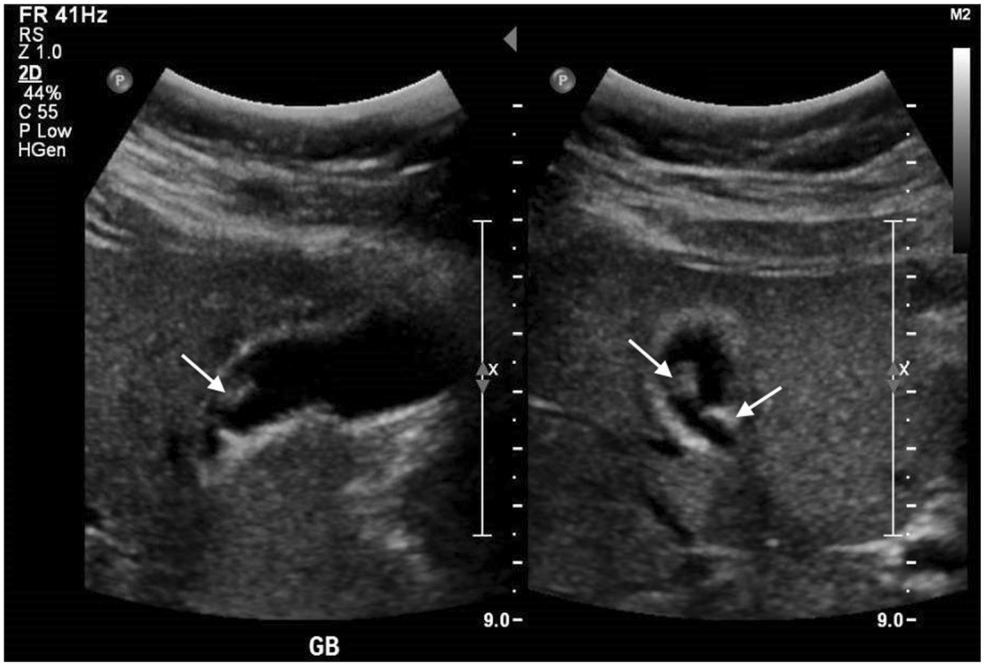

Fig. 1 Ultrasound showing gallbladder polyps (arrows)

at incidence of sarcoidosis is usually between 20 and 40 years of age, affecting females more frequently than males [3].

Sarcoidosis manifests mainly in the pulmonary and lymphoid system, with lung and lymph node involvement in over 90 and 30% of patients respectively [3, 4]. Gastrointestinal and hepatic involvement in sarcoidosis is rare, occurring in less than 4% of cases [5, 6]. Even rarer is sarcoidosis of the gallbladder and its associated lymph node [1]. A review of the literature found five reported cases of sarcoidosis involving the gallbladder alone while the other two was involving the gallbladder's associated lymph nodes (Table 1). In our report, we are presenting another rare case of sarcoidosis of the accompanying lymph nodes of the gallbladder.

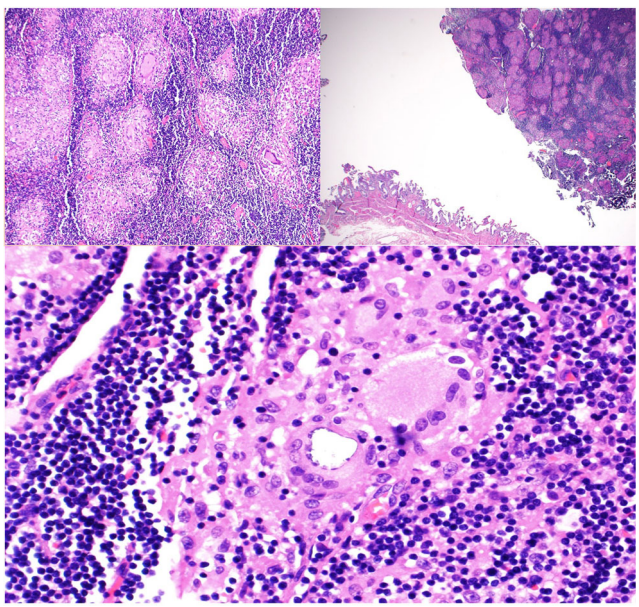

Fig. 2 Histopathological evidence of noncaseating granuloma within the lymph node

Sarcoidosis typically causes granulomatous inflammation of the gallbladder and/or accompanying lymph nodes. Although there were cases where patients only experience generalised vague abdominal pain, patients more often complain of biliary colic and symptoms of acute cholecystitis [3]. As shown in Table 1, four out of seven cases had presented with right-sided abdominal pain, nausea and vomiting. If left untreated, the condition might perpetuate and eventually become chronic cholecystitis, as demonstrated in half of the cases, including our patient. Further inflammation involving the biliary tree and lymph nodes can also lead to extrinsic compression of cystic duct, thereby precipitating obstructive jaundice. [3, 7, 8] Other more chronic presentation of biliary involvement includes portal hypertension, chronic cholestasis, biliary fibrosis/cirrhosis and Budd Chiari syndrome [5, 9]. Additionally, symptoms of sarcoidosis can often mimic that of malignancy in the system such as cholangiocarcinoma through strictures in the extrahepatic ducts [3].

Up to 50 % of patients with gallbladder sarcoidosis are often asymptomatic and are usually diagnosed incidentally through other modalities like chest X-rays and histopathological examination while investigating for another cause [10]. Based on the cases in Table 1, only one patient with gallbladder sarcoidosis had been diagnosed with sarcoidosis prior to the cholecystectomy while the remaining patients were found to have sarcoidosis incidentally only after surgery. Similarly, our patient was diagnosed with sarcoidosis only after histological diagnosis of the resected specimen. However, some patients may develope gastritis due to sarcoidosis. Retrospectively, our patient's symptom of gastritis could be related to sarcoidosis.

Table 1 Reported cases of gallbladder and lymph node sarcoidosis

	Gender	Age	Pre-operative diagnosis	Radiology	Histopathological result	Treatment
Case 1, 1965 [12]	Male	19	RHC pain and vomiting	Cholangiogram: chronic cholecystitis	Panparietal gallbladder and liver noncaseating granulomas Subacute cholecystitis AFB Negative	Cholecystectomy Corticotherapy
Case 2, 1978 [8]	Female	29	Symptoms of obstructive jaundice	(Not known)	Noncaseating granulomatous involvement of the common hepatic duct and surrounding lymph nodes	Temporary decompression by T-tube Corticotherapy
Case 3, 1988 [13]	Female	37	Jaundice and pruritus Treated sarcoidosis of thoraco-abdominal region (for 10 years)	(Not known)	Gallbladder neck noncaseating granulomas AFB negative	Cholecystectomy Corticotherapy
Case 4, 2004 [14]	Male	27	Biliary colic and biliary pancreatitis (2003); 7 months history of inguinal lymphadenopathy and exertional dyspnoea (Mantoux test negative)	Abdominal US: cholelithiasis	Noncaseating granulomas in liver, lymph node and gallbladder AFB negative.	Cholecystectomy Corticotherapy
Case 5, 2014 [10]	Female	20	10-day history of abdominal pain	US: 5.7-mm polypoid lesion in gallbladder	Chronic cholecystitis and non-necrotizing granulomatous lymphadenitis in one lymph node	Cholecystectomy
Case 6, 2015 [3]	Male	52	(Not reported)	(Not known)	Gallbladder sarcoidosis	Antrectomy Corticotherapy
Case 7, 2016 [1]	Male	70	Biliary colic	US and cholangiogram: chronic cholecystitis	Subacute and chronic lithiasic cholecystitis with several noncaseating epithelioid and giant cell granulomas AFB negative	Cholecystectomy Corticotherapy
Case 8, 2018	Female	67	Biliary colic	US HBS: 7-mm gallbladder polyps	Chronic cholecystitis with polypoid cholesterolosis Lymph node showed noncaseating granulomata AFB negative	Cholecystectomy

Differential diagnosis for patients with sarcoidosis includes infectious aetiologies, chronic granulomatous disease and malignancy. Infectious aetiologies include tuberculosis, *Mycobacteria*, histoplasmosis, toxoplasmosis, *Cryptococcus*, and schistosomiasis. All these conditions had been ruled out either by biochemical or histological examination in our patients.

Sarcoidosis is diagnosed primarily based on clinical and radiological findings and histopathological demonstration of noncaseating granulomas in more than one organ, in the absence of alternative diagnoses [2, 11]. Alternative diagnoses such as tuberculosis can be excluded by performing an acid-fast bacilli (AFB) test through Ziehl-Neelsen staining. In our case, her histology report revealed the presence of noncaseating granuloma in the gallbladder lymph node with negative AFB test.

The mainstay of systemic sarcoidosis is immunosuppressive therapy using prednisolone [5, 9]. However, there is limited literature on the specific treatment for gallbladder sarcoidosis. Based on our management of our patient and case reports previously published, in patients with symptomatic cholecystitis due to sarcoidosis, cholecystectomy should be the choice of treatment. Most patients would experience a resolution of most biliary symptoms post-cholecystectomy.

Conclusion

Sarcoidosis of the gallbladder and its associated lymph node is extremely rare. The condition causes granulomatous inflammation of the gallbladder wall and surrounding structures. Patients who are symptomatic should be treated with cholecystectomy as the definitive treatment.

Compliance with Ethical Standards

Informed Consent Informed consent was obtained from the participating individual included in the report.

Conflict of Interest The authors declare that they have no conflict of interest.

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