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CASE REPORTOPEN ACCESS

Gallbladder Adenomyomatosis Mimicking Acute Cholecystitis in Young Male

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A B S T R A C T

Gallbladder adenomyomatosis (GA) is a benign, proliferative condition characterized by gallbladder wall thickening and the presence of Rokitansky-Aschoff sinuses. Though generally asymptomatic, GA can mimic acute cholecystitis, leading to diagnostic challenges. We present a case of a 32-year-old male with a year-long history of recurrent right hypochondriac pain radiating to the shoulder, accompanied by nausea and vomiting. Initial clinical and laboratory findings suggested acute cholecystitis. Imaging studies, including ultrasonography and magnetic resonance cholangiopancreatography, revealed diffuse gallbladder wall thickening with cystic spaces, indicative of adenomyomatosis. The absence of gallstones and the presence of a dilated cystic duct further supported the diagnosis. The patient underwent elective laparoscopic cholecystectomy, and histopathological examination confirmed diffuse adenomyomatosis with smooth muscle hypertrophy and Rokitansky-Aschoff sinuses. The postoperative recovery was uneventful, and the patient remained asymptomatic during a six-month follow-up. Gallbladder adenomyomatosis is benign and can be detected by imaging or during cholecystectomy. Theoretically, adenomyomatosis doesn't need any special care unless it presents symptoms, whether or not cholecystolithiasis is present.

Keywords: Gallbladder, Adenomyomatosis, Rokitansky-Aschoff sinuses, Cholecystectomy.

INTRODUCTION

Gallbladder adenomyomatosis is a prevalent and benign condition characterized by thickening of the gallbladder wall, observed in 2.0-8.7 percent cholecystectomy specimens, with a higher incidence in females compared to males [1].

The proliferation of epithelium, thicker muscle layers, and/or development of mucosal outpouchings into or through muscularis are all signs of adenomyomatosis. This could result in diverticula (Rokitansky-Aschoff sinuses), crypts, or ducts. This aberrant tissue may induce one or more segmental "strictures" around the gallbladder or appear as a nodule at the gallbladder's tip [2].

Case Report

A 32-year-old man without any known comorbidities complained of right hypochondriac discomfort that had been radiating to the right shoulder tip for a year, along with nausea and vomiting. The pain was of colicky which lasted for 1 hour with multiple attacks over the last 1 month. His vitals were normal. Physical examination revealed tenderness in the right hypochondriac region -murphys sign positive.

Laboratory analysis revealed normal levels of alkaline phosphatase as well as transaminases along with mild conjugated hyperbilirubinemia (total bilirubin level 2.0mg/dl; direct bilirubin level 1.1mg/dl). The patient was diagnosed with acute cholecystitis based on clinical and laboratory findings.

Ultrasound abdomen was taken and features were suggestive of diffuse adenomyomatosis of the gall bladder with a prominent cystic duct with minimal pericholecystic fluid collection.

The type of gall bladder lesion was determined by magnetic resonance cholangiopancreaticography, which showed diffuse thickening of the gall bladder wall with cystic spaces in its wall. There is minimal pericholecystic fluid collection. However, no gallstoneswere seen. The cystic duct is dilated measuring 7.8mm. Gallbladder measures approximately 6x1.8cmextra and intrahepatic bile ducts were normal. He was taken up for elective laparoscopic cholecystectomy.

INTRAOPERATIVE FINDINGS:

Elective laparoscopic cholecystectomy was done under general anaesthesia which revealed an enlarged gall bladder as shown in Figure 1.

Figure 1: Shows an enlarged gall bladder

HISTOPATHOLOGICAL EXAMINATION:

Histopathology of the cholecystectomy specimen showed diffuse smooth muscle hypertrophy of the gall bladder wall with several Rokitansky-Aschoff sinuses (RAS) consistent with adenomyosis of the gall bladder.

OUTCOME AND FOLLOWUPS:

The postoperative period was uneventful without any complications and was discharged on day 5 of the postoperative period. He was later followed up in the outpatient department for at least 6 months and is well.

DISCUSSION

Jutras initially identified gallbladder adenomyomatosis as a degenerative and proliferative disease of the gallbladder in 1960, and since then, there have been more and more reports of it [3].

Before 1960, the terms hyperplastic adenomyosis, intramural diverticulosis, cystic cholecystitis, proliferative glandular cholecystitis, adenomyosis, as well as adenofibromyoma were used [4].

The GB wall is often thicker (sometimes exceeding 10mm) due to the presence of diverticula, also known as RAS (Rokitansky-Aschoff sinuses), initially reported by Rokitansky in 1842, subsequently by Aschoff in 1905. It is an invasion of muscularis layer of gallbladder wall by mucosa. RAS is essential for diagnosis and may be observed, particularly in cases of xanthogranulomatous cholecystitis, although not being pathognomonic. However, RAS is especially significant in adenomyomatosis, related to wall thickening, as well as penetrating muscular layer (Figure 2).

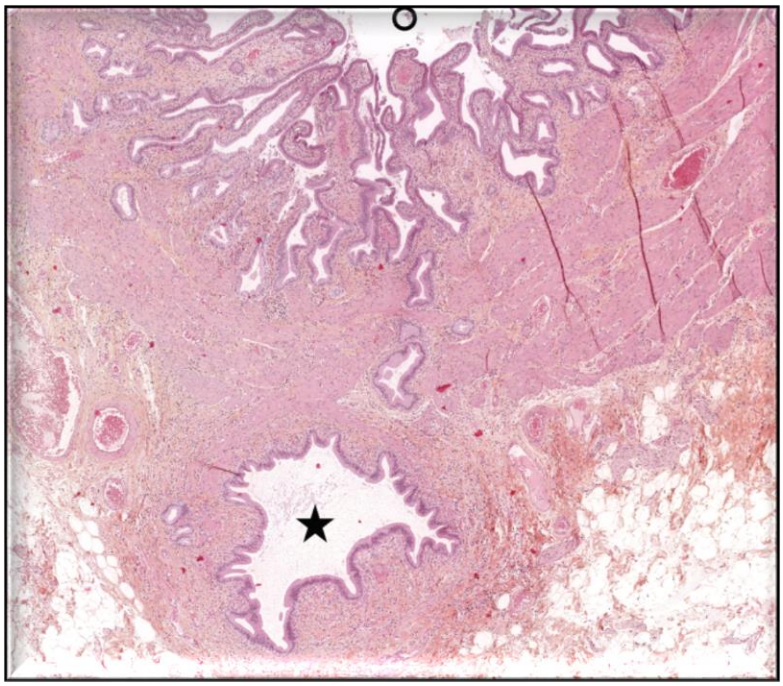

Figure 2: Histological section of gbadenomyomatosis: the presence of muscle thickening andRokitansky-Aschoff sinus(star), gb lumen (circle)

Beilby *et al*., indicated that neurogenic dysfunction caused elevated intraluminal pressure, which resulted in spasmodic closure of a "sphincter" in the proximal section of the cystic duct and intense contractions of the hypertrophied muscle in these gallbladder walls [5].

Similarly, Jutras and Levesque [6] concluded that neurogenic dysfunction as well as overdistention behind a spastic sphincter were the causes of pain in these individuals after observing distention of cystic along with common ducts in films taken soon after a fatty meal. Thus, in a process similar to that proposed in the diverticular illness of the colon, this neurogenic dyskinesia could be thought to be the cause of muscle hypertrophy as well asthe development of RAS.

There are four primary pathogenic forms of gall bladder adenomyomatosis (Figure 3).

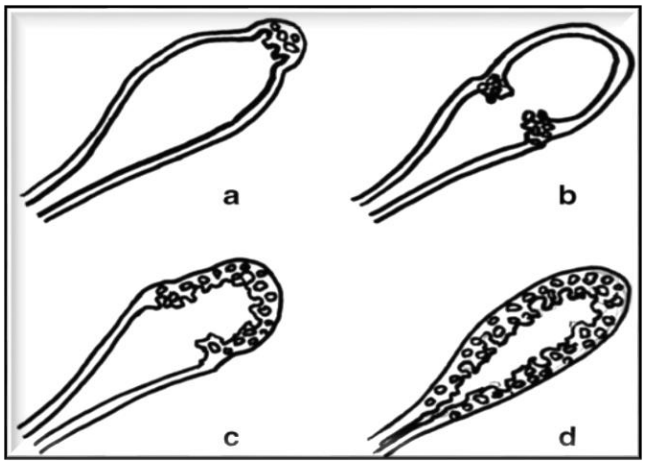

Figure 3: a-localized, b-annular, c-segmental, d-diffuse

A focused thickening that usually affects the fundal region distinguishes the most common pattern, referred to as localized or fundal type. The general form of the gallbladder is usually maintained, and the wall of the gallbladder that is not affected appears physiologically thin [7-9].

The distal part of the body and gallbladder wall's larger section, usually fundus, are involved in segmental form. While the unaffected part maintains its natural shape, the implicated part seems to be contracted.

The ring-shaped thickening of gallbladder wall, affecting center section, is a characteristic of the annular type. Only the affected area of the gallbladder appears contracted, taking on the form of an hourglass. The gallbladder lumen can also be divided into two distinct compartments by highly apparent epithelial growth. Biliary stones and sludge may so develop in isolated fundal compartments.

When an entire organ is affected, even after fasting, the organ seems contracted, which is a characteristic of the diffuse type.

Numerous studies have shown that individuals with segmental-type adenomyomatosis, particularly in the elderly, have a greater incidence of gallbladder cancer than those either without GA or with different GA patterns [10].

Pauci-symptomatic or asymptomatic gall bladder adenomyomatosis is usually identified through the cholecystectomy specimen's pathology or imaging. In rare cases, the right hypochondrium may experience sensations similar to biliary colic. Patients are increasingly reporting non-specific digestive issues, such as flatulence, fatty food intolerance, nausea, and vomiting. Acalculous cholecystitis's initial eruption can also indicate it [11].

The first imaging modality used to diagnose GA was oral cholecystography (OC), which is now considered an outdated method (Figure 4).

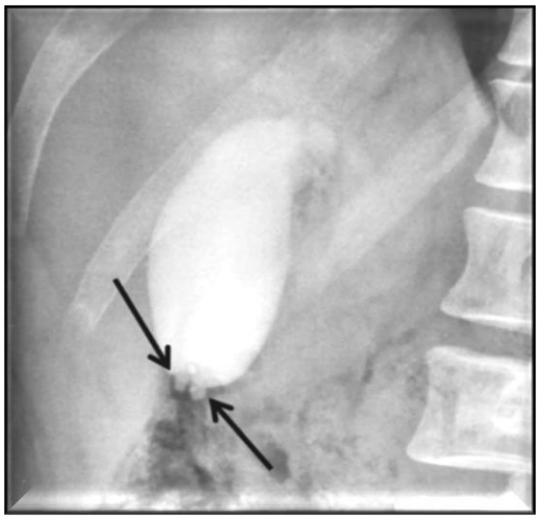

Figure 4: Oral cholecystography-fundal type Rokitansky Ashcoff sinus (arrow)

Trans-abdominal ultrasonography (US), which has an accuracy rate of 91.5-94.8 percentin distinguishing early-stage gallbladder cancer from GA, is the recommended imaging method for identifying and characterizing GA [12]. Rokitansky-Aschoff sinus identification is essential for adenomyomatosis imaging diagnosis [13, 14]. A thicker wall with tiny, anechoic intramural cystic spaces can be observed inthe US (Figure 5).

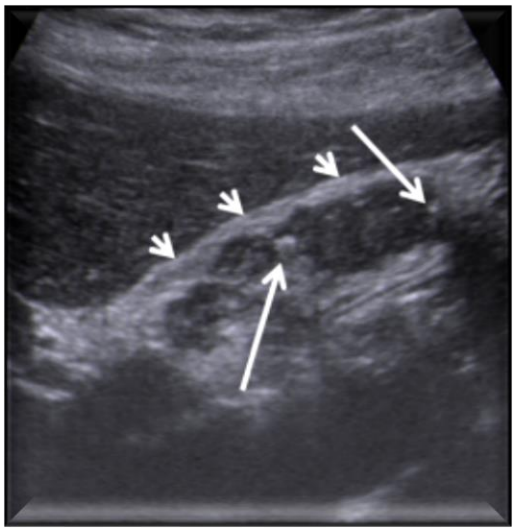

Figure 5: USG showing diffusely thickened gall bladder wall (arrowheads)with Aschoff sinuses (arrows)

Sludge, calculi, or cholesterol crystals in the sinuses can cause intramural echogenic patches known as comet-tail artifacts (Figure 6) or twinkling artifacts overcolor Doppler-US (Figure 7) [15].

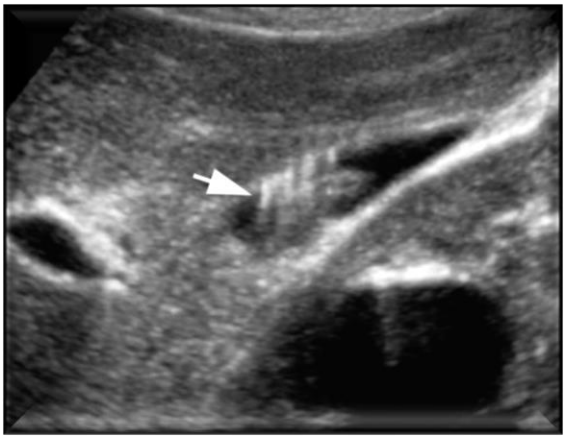

Figure 6: GB ultrasound showing segmental variety with comet tail artifact (arrows)

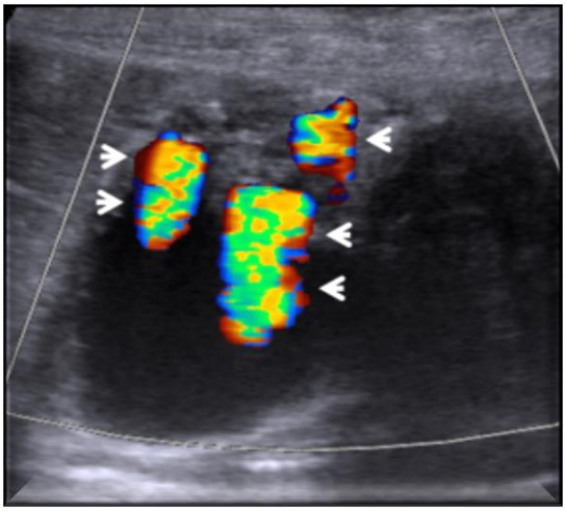

Figure 7: Colour Doppler showing twinkling artifact in Aschoff sinus (arrowheads)

Additional CT signs of adenomyomatosis include a thicker GB wall or a mass-like lesion with minute cystic spaces, which are indicators of bile-filled RAS. Fizzy gray enhancing spots in thicker GBwalls or dotted outside the

border of an augmenting inner wall layer in CT are referred to as the "cotton ball sign," which was only recently identified. It is highly sensitive in distinguishing malignancy of the gall bladder from adenomyomatosis (Figure 8) [17].

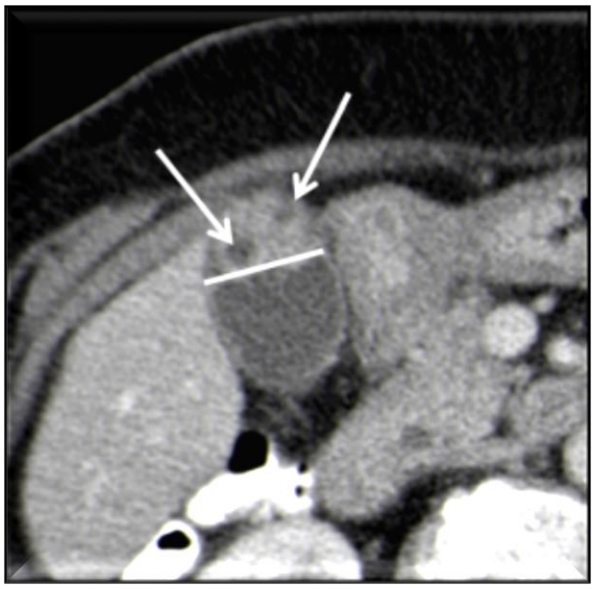

Figure 8: CT image of gbadenomyomatosis showing mural thickening (line) containing cystic spaces representing Rokitansky–Aschoff sinuses- cotton ball sign (arrows)

The "pearl necklace sign," or high signal-intensity foci of GB wall on a T2-weighted scan, provides basis for MRI diagnosis of adenomyomatosis (Figure 9) [18].

Figure 9: MRI of gallbladder adenomyomatosis: the pearl necklace sign (lot ofAschoff sinuses can be identified one next to the other around the gallbladder, leading to the so-called pearl necklace sign)

In cases of symptomatic ADM, cholecystectomy is recommended regardless of the presence of cholecystolithiasis. This is because cholecystectomy is an effective and long-lasting treatment in all documented cases [19].

The Haute Autorité de santé's 2013 recommendations state that asymptomatic ADM through imaging doesn't qualify as a cholecystectomy indication because there is a traditional zero risk of complications or degeneration [20].

Cholecystectomy should be recommended to exclude GB malignancy when adenomyomatosisdiagnosis is definitive even after a thorough imaging evaluation, encompassingcholangio-MRI.

If wall thickening results from adenomyomatosis, no additional treatment is required.; however, if gallbladder carcinoma is the cause, patient should receive professional intervention promptly. This is determined by performing an open cholecystectomy and obtaining a conclusive histological result. When there are symptoms of GA or radiological suspicion of GB malignancy, laparoscopic cholecystectomy is the gold standard [21].

CONCLUSION

Gallbladder adenomyomatosis is benign and can be detected by imaging or during cholecystectomy. The presence of a thicker wall with RAS serves as the basis for the pathological diagnosis. Cholangio-MRI is the best imaging test since it helps define the differential diagnosis (GB cancer) and identifies the location. Theoretically, adenomyomatosis doesn't need any special care unless it presents symptoms, whether or not cholecystolithiasis is present. Although adenomyomatosis and cancer have been implicated by certain authors, comprehensive prophylactic cholecystectomy is not advised.

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