

Single Case

# Diffuse Gallbladder Adenomyomatosis with an Inflammatory Complication in an Adult

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## Keywords

Cholecystitis · Diffuse thickened gallbladder wall · Intramural diverticulosis · Gallbladder adenomyomatosis · Rokitansky-Aschoff sinuses

## Abstract

Gallbladder adenomyomatosis (GA) is a benign alteration of the gallbladder wall. There are three types involved: segmental, fundal, and diffuse pattern; the last type is very rare. Ultrasound is the imaging method of choice for diagnosing that shows Rokitansky-Aschoff sinuses with cholesterol deposition creating comet-tail artifacts. Asymptomatic GA does not require surgery in case there are no malignant lesions defined by imaging diagnosis. We present the rare case of a 51-year-old man who was admitted to hospital due to abdominal pain concomitant with inflammatory syndrome. Imaging and histological findings were appropriate to diagnose diffuse GA and complication of cholecystitis.

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## Introduction

Gallbladder adenomyomatosis (GA) is a benign acquired lesion whose feature is a thickened gallbladder wall containing small bile-filled cystic spaces. Jutras [1] described GA for the first time in 1960 and after that its case reports increased [2]. Previously, there were many terms used to refer to this condition, such as cystic cholecystitis, adenomyoma, cholecystitis glandularis proliferans, hamartoma, adenofibromyoma, intramural diverticulosis, and diverticular disease of the gallbladder, but currently GA is approved worldwide [3]. At present, the pathogenesis of GA is not yet clearly understood. It is less common in young people and females tended to be more commonly affected than males. The incidence is about 2.8–5% of cholecystectomy specimens [2, 4]. We present a rare case of diffuse GA type with a complication of cholecystitis.

## Case Presentation

A 51-year-old man presented to the emergency unit with right hypochondriac pain and abdominal distention happening 2 days before. There was no significant medical history. The additional symptoms consisted of fatigue and mild postprandial nausea. Reports showed no vomiting, jaundice, or weight loss. He was febrile with a temperature of 38°C; other vital signs were unremarkable. His abdomen was soft with tenderness noted in the right hypochondriac region; Murphy's sign was positive. Laboratory tests including AFP, CA19-9, CEA, SGOT, SGPT, bilirubin, transaminases, and alkaline phosphatase were within normal limits. White blood cell were increased slightly by about 12,000/mm<sup>3</sup>. Ultrasound features included an about 18–20 mm thickened gallbladder wall, near-total obliteration of the lumen, Rokitansky-Aschoff sinuses, calcification, and comet-tail artifacts (Fig. 1). No obvious color uptake was seen on Doppler evaluation. Computed tomography images showed a diffuse thickened gallbladder wall and inner layer enhancement. Accumulation of fluid and fat stranding surrounding the gallbladder suggested acute inflammation (Fig. 2). The patient underwent cholecystectomy. Gross images of surgical specimens showed diffuse thickening of the gallbladder wall. Histopathology confirmed GA accompanied by inflammation. This diagnosis was confirmed by the histopathologic features of a diffuse thickened wall, presenting Rokitansky-Aschoff sinuses, hyperplasia of collagen fibers, and hypertrophy of smooth muscle; no evidence of malignancy was detected. The patient recovered and was discharged after 5 days. He was followed up for 1 year and did not show any abnormalities.

## Discussion

GA is characterized by excessive epithelial proliferation which gives rise to epithelial infolding within the underlying muscular layer, resulting in gallbladder wall thickening with subsequent formation of intramural diverticulosis lined by mucosal epithelium [5]. Histologically, it is characterized by epithelial and smooth muscle proliferation. The muscular hypertrophy is considered to be caused by increased intraluminal pressure. On microscopy, the hyperplastic smooth muscle cells are accompanied by epithelial invaginations forming Rokitansky-Aschoff sinuses with cholesterol crystals or calcified nodules trapped inside because of chronically elevated intraluminal pressure. The gallbladder epithelium lines may penetrate

the subserosa and reach the serosa (Fig. 3). Three types of GA are described: fundal (localized), segmental (annular), and more rarely diffuse (generalized) [6]. The fundal pattern localizes at the fundus of the gallbladder, which presents as a central dimple. The segmental pattern is located in the body of the gallbladder; it separates the gallbladder into two communicating compartments like an hourglass. The diffuse pattern is extremely rare and manifests with thickening of the entire gallbladder wall (Fig. 4) [4–7].

Although GA is usually asymptomatic, it can manifest as abdominal pain or be associated with chronic inflammatory biliary tract disease. GA is mostly detected incidentally by imaging diagnostics and usually requires no treatment. On ultrasound, it appears as a localized or diffuse thickened gallbladder wall with intramural diverticula containing sinus cavities. There can be echogenic or anechoic luminal content in the gallbladder wall. Cholesterol crystals trapped in the sinuses manifest as comet-tail artifacts. Computed tomography and cholangiography are not routinely used in GA. Computed tomography images were useful in our case as they allowed visualization the contrast enhancement pattern and helped to exclude malignant disease of the gallbladder. In clinical routine, magnetic resonance imaging is an important reference examination. Pearl necklace sign, which reveals the presence of Rokitansky-Aschoff sinuses, is a specific finding of GA at magnetic resonance cholangiopancreatography [3, 7, 8]. The differential diagnosis includes all diseases that cause gallbladder wall thickening, such as xanthogranulomatous cholecystitis, chronic cholecystitis, or gallbladder cancer [5–9].

The relationship between GA and carcinoma is still controversial; some studies proposed that the segmental type of GA preceded the development of gallbladder cancer [9, 10]. Some more recent research has considered GA and gallbladder cancer to reveal different histological features: adenomyomatosis is characterized by epithelial and smooth muscle proliferation, whereas gallbladder cancer represents cell dysplasia [11]. Currently, GA cannot be considered a premalignant lesion based on available evidence. However, presence of stones and cholecystitis may lead to dysplastic changes and cancer [4, 10].

For the treatment of symptomatic GA, an indication for cholecystectomy is necessary. Asymptomatic GA does not require surgery, but the radiological diagnosis must exclude gallbladder malignancy. In our case, the patient presented with acute cholecystitis. Without timely treatment, this can progress to gangrene and perforation [12–14]. For these reasons, in our case, the indication for surgery is given.

### Conclusion

Diffuse GA is a rare benign lesion and can lead to inflammatory complications. It is an entity that can mimic cancer. Therefore, imaging studies play an important role in diagnosis and differentiation. Although we have contributed further knowledge about diffuse GA, it is not fully understood. Future insight studies will be needed to determine more advanced diagnosis techniques, effective treatments, and management methods.

### Statement of Ethics

We reported this case in compliance with the Declaration of Helsinki. Written informed consent was obtained from the patient for publication of his case, including images. The identity of the patient was protected.

## Conflict of Interest Statement

The authors have no conflicts of interest to declare.

## Funding Sources

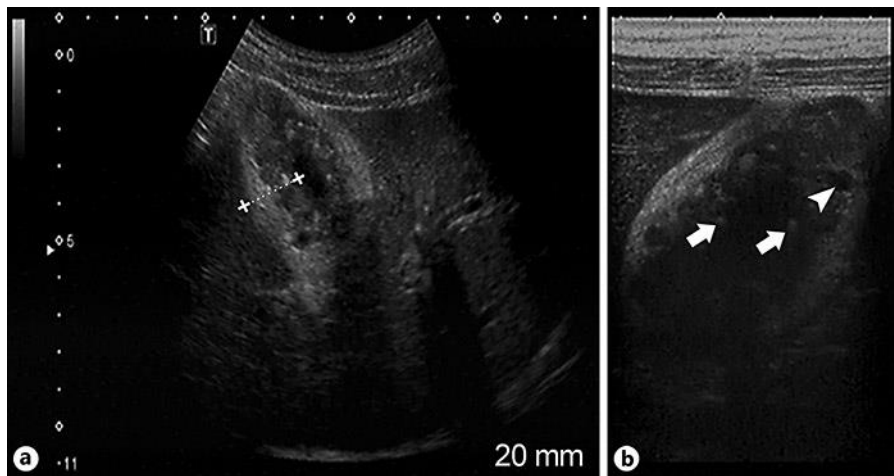
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## Author Contributions

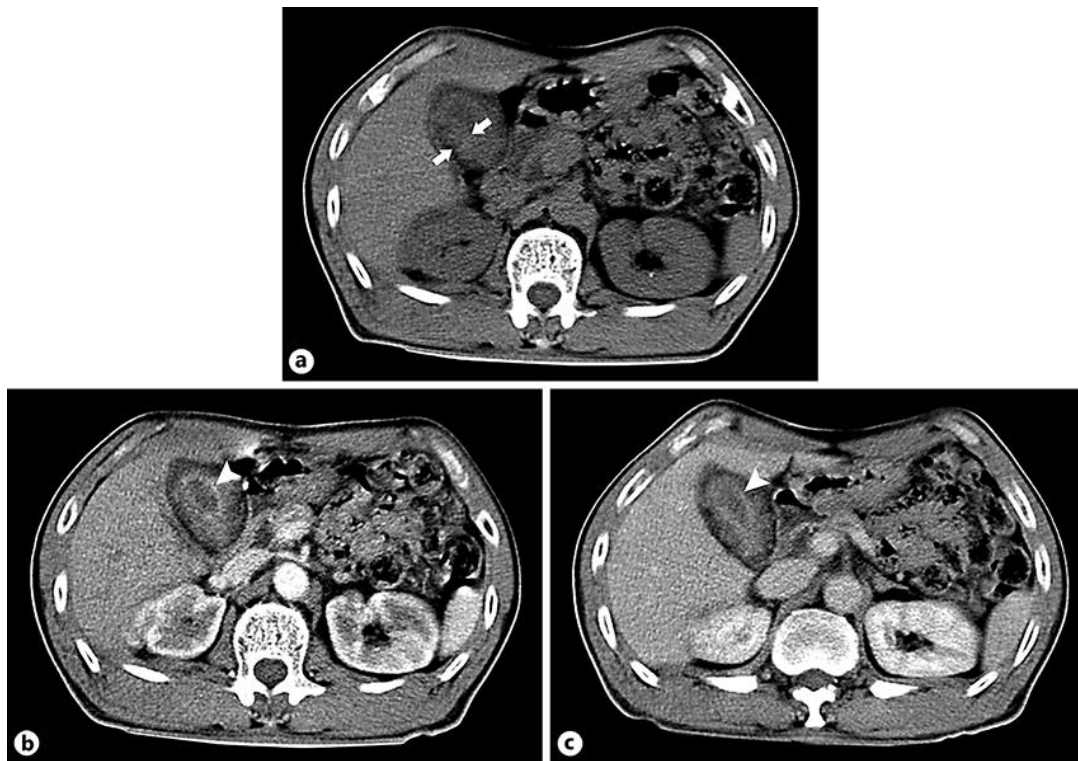
All authors contributed equally.

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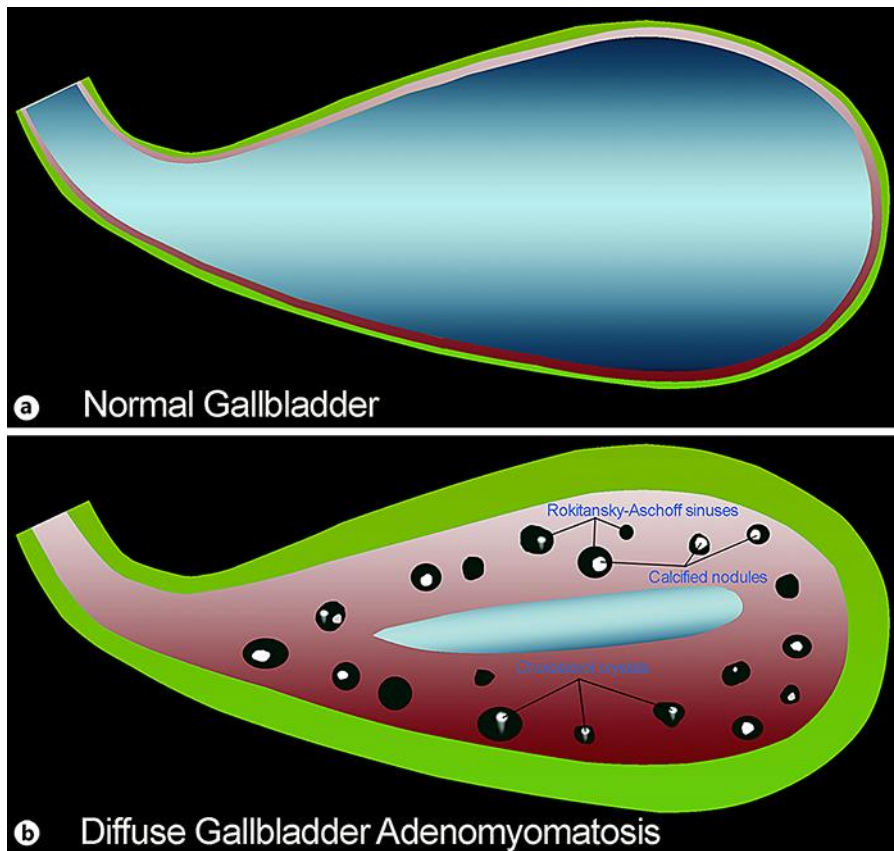


**Fig. 1.** Ultrasound revealed gallbladder adenomyomatosis using different frequency probes. **a** Sagittal plane image of the gallbladder with ultrasound transducer converts showed a thickened wall longitudinally, including numerous intramural echogenic foci. **b** A longitudinal section of the gallbladder with a linear ultrasound transducer showed comet-tail signs of cholesterol crystals (arrows) deposited in Rokitsky-Aschoff sinuses (arrowhead).

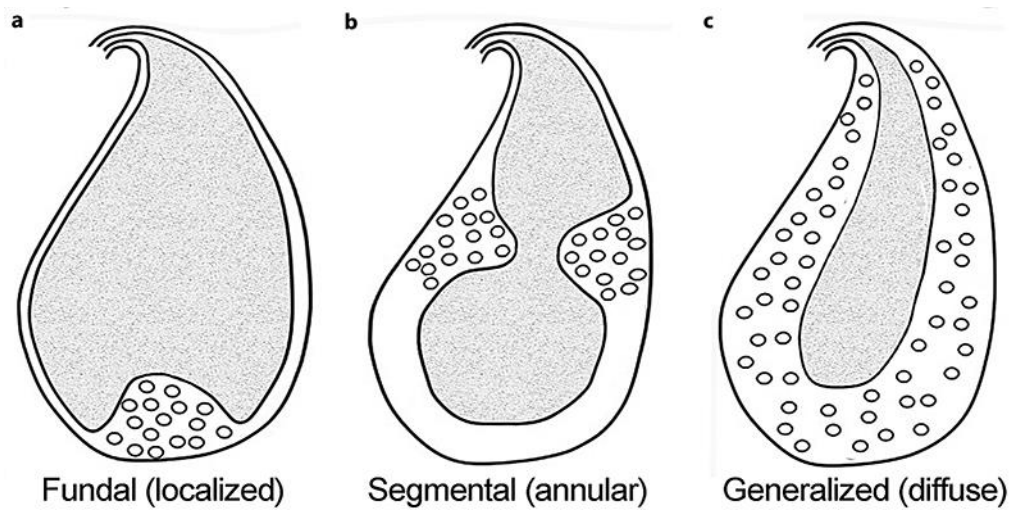


**Fig. 2.** Axial computed tomography images: unenhanced phase (a) and contrast-enhanced phases with arterial (b) and venous (c) gallbladder adenomyomatosis showing diffuse gallbladder wall thickening (arrows) with intact mucosal enhancement line (arrowheads).





**Fig. 3.** Illustration of a normal gallbladder (a) and of diffuse gallbladder adenomyomatosis (b).



**Fig. 4.** Illustration of three types of gallbladder adenomyomatosis. **a** The fundal type located in the fundus of the gallbladder. **b** The segmental type located in the body of the gallbladder which separates the gallbladder into two communicating compartments like an hourglass. **c** The diffuse type manifests thickening of the entire wall of the gallbladder.