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Case Report

A Rare Case of Honeycomb Gallbladder in a Patient of Chronic Calculus Cholecystitis

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ABSTRACT

Background: Honeycomb gallbladder (GB) is a rare condition characterized by multiple septations in the GB wall, giving it a honeycomb appearance. First described by Knetsch in 1952, this anomaly is typically congenital but can also be acquired. Patients with a honeycomb GB often present with a variety of symptoms, the most common being abdominal pain.

Case Description: We present the case of a 62-year-old female with a history of symptomatic chronic calculus cholecystitis. She underwent a laparoscopic cholecystectomy, which resolved her symptoms. Preoperative ultrasound imaging showed no evidence of multiple septations; however, post-operative examination revealed a multiseptated gallbladder.

Conclusions: Honeycomb GB is an unusual presentation. Among the theories explaining multiseptated GB, one suggests that chronic calculus cholecystitis leads to extensive denudation of the epithelial lining, causing fibrosis and calcification beneath, which results in contraction. Our case supports this theory, as these pathological changes likely contributed to the honeycomb appearance. While medical management typically focuses on symptomatic relief, cholecystectomy has been shown to completely resolve symptoms.

Keywords: Cholecystitis, Honeycomb gall bladder, Laparoscopic cholecystectomy, Multiseptated

INTRODUCTION

Honeycomb gallbladder (GB) is a rare condition characterized by multiple septations within the GB wall. This anatomical variant was first described by Knetsch in 1952[1], and since then, fewer than 150 cases have been reported worldwide.[2] The exact etiology of honeycomb GB remains unclear, with cases being either congenital or acquired. Several theories have been proposed to explain its development, including the embryonic, wrinkling, and Phrygian cap theories.[3-6]

A review of the literature suggests that ultrasound (USG) is the most effective imaging modality for diagnosis, while surgical intervention is recommended for patients with multiseptated GB (MGB). Recent studies indicate that contrast-enhanced computed tomography (CECT) can enhance the diagnostic accuracy of GB pathologies. Additionally, minimally invasive procedures such as laparoscopic cholecystectomy are preferred due to their shorter recovery times and reduced risk of complications compared to traditional open surgery.

CASE HISTORY

A 62-year-old female patient presented to our department with a chief complaint of abdominal pain persisting for 1-2 months. The pain was sudden in onset, non-radiating, and not associated with nausea, vomiting, or fever. It was relieved by taking analgesics. The patient's medical history included controlled type-2 diabetes mellitus for 20 years, managed with oral hypoglycemic agents, and hypothyroidism for 16 years, treated with oral Thyroxine. She had no history of hypertension, tuberculosis, asthma, or epilepsy. Her past surgical history included a caesarean section 40 years ago.

Initial investigations, as detailed in Table-1, revealed slightly decreased hemoglobin levels, a normal white blood cell count, elevated random blood sugar, and liver and renal function tests within normal limits. Viral markers, including HCV, HBsAg, and HIV, were non-reactive.

Table-1: Pre-operative investigations

Investigations	Value	Normal Range
Haemoglobin	10.5 g/dl	13.0-17.0
TLC	7.4 * 10^3/uL	4.0-10.0
RBS	165 gm/dl	80.0-140.0
Liver Function Tests		
SGOT	16 IU/L	0.0-35.0
SGPT	13 IU/L	0.0-41.0
Alkaline Phosphatase (ALP)	99 IU/L	40.0-129.0
Albumin	3.7 g/dl	3.5-5.2
Bilirubin	0.2 mg/dl	0.1-1.2
Renal Function Tests		
Serum urea	43 mg/dl	17.0-43.0
S. Creatinine	1.5 mg/dl	0.6-1.1
INR	0.83	0.8-1.2
S. Na+	139 mmol/L	136.0-145.0
S. K+	4.5 mmol/L	3.5-5.0
Viral Markers		
HCV	Non-Reactive	
HBsAg	Non-Reactive	
HIV	Non-Reactive	

An abdominal USG was performed, showing chronic calculus cholecystitis. The GB was contracted, measuring approximately 6 cm in length, and demonstrated a wall echo complex with multiple calculi. No peri-cholecystic fluid collection was observed. Intra-hepatic biliary radicals (IHBR) and the common bile duct (CBD) appeared normal. Based on these findings, surgery was planned. Proper preparation of the surgical field was conducted. Under general anesthesia, the patient was positioned in the reverse Trendelenburg position with the right side propped up. An infraumbilical incision was made to accommodate a 10 mm camera port. Additional ports were inserted under camera guidance: a 10 mm epigastric port and two 5 mm ports - one positioned in the midclavicular line 7.5 cm below the epigastric port, and the other in the midaxillary line 7.5 cm below and lateral to the former. During the procedure, the GB was identified, and meticulous anterior and posterior dissection was performed while staying superior to Rouvier's sulcus. The lower third of the gallbladder was carefully separated from the liver bed. The critical view of safety (CVS) was achieved, revealing the cystic duct and cystic artery entering the GB within Calot's triangle. Precise dissection ensued in this anatomical region [Figure-1].

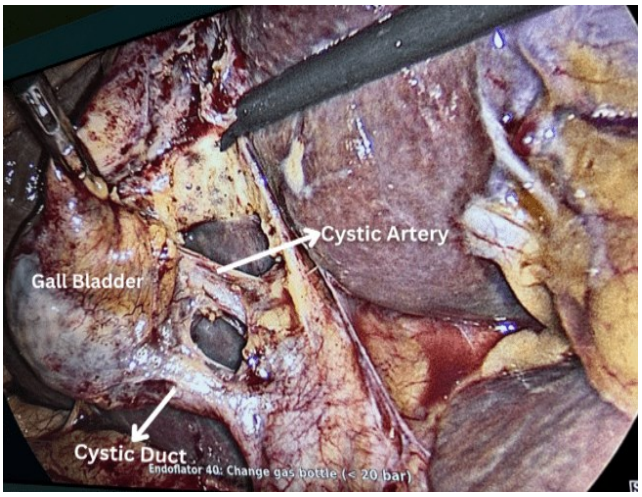

Figure-1: Critical View of Safety (CVS) Achieved - The cystic artery and cystic duct are clearly visualized connecting to the gallbladder, with the lower one-third of the gallbladder separated from the liver. Clear delineation of Calot's triangle is also evident

After dissection, the cystic duct and cystic artery were clipped using Ethicon Ligaclip 400 and 300, respectively, and then divided. The gallbladder was carefully separated from the liver bed. The specimen, which exhibited multiple septations and stones, was extracted through the epigastric port. Haemostasis was thoroughly ensured and rechecked [Figure-2].

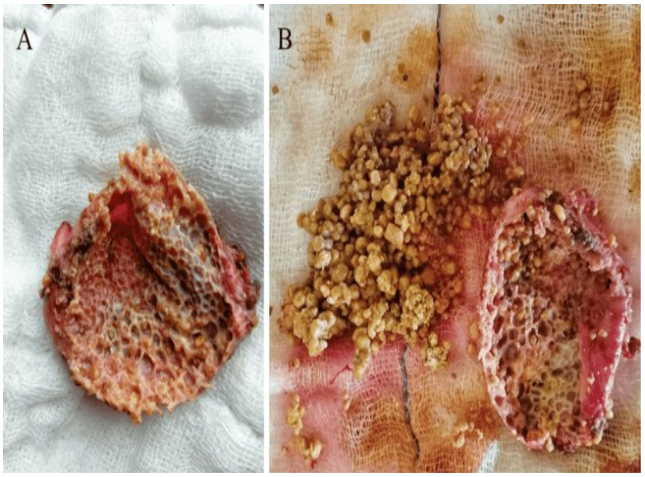

Figure-2: Cross section of Gall Bladder (a) Dissected gallbladder showing multiple septations. (b) Honeycomb appearance of the gallbladder along with multiple calculi

For postoperative pain management, a local anaesthetic agent, Bupivacaine, was administered in the subcutaneous plane. Closure of ports was performed using polyglactin 910

(vicryl) sutures, and the skin was approximated with a skin stapler.

Postoperative recovery was uneventful, and the patient was discharged the following day. Histological examination of the gallbladder revealed tissue lined by extensively denuded epithelium, with mild chronic inflammatory infiltrates comprising lymphocytes and plasma cells underneath. Areas of fibrosis, calcification, and congested blood vessels were noted [Figure-3].

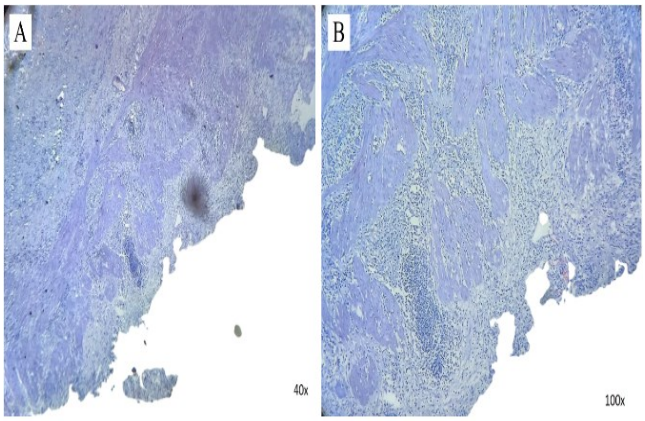

Figure-3: Histopathological Section. (a) Extensive denudation of the epithelium with lymphoid aggregates (40 X). (b) Fibrosis and mild chronic inflammatory infiltrates composed of lymphocytes and plasma cells (100 X)

All procedures performed in this study adhered to the ethical standards of the institutional and national research committee and the Helsinki Declaration. Written informed consent was obtained from the patient for the publication of this case report and accompanying images.

DISCUSSION

Honeycomb GB, also known as MGB, is a rare congenital condition first described by Knetsch in 1952.[1] In 1963, Simon and Tandon reported the first case along with the radiological finding of multiple, thin, crisscrossing septa that produced an irregular mosaic pattern in the GB on oral cholecystography.[4] Most cases are suggestive of a congenital anomaly, though some have an acquired cause, such as abdominal trauma, as described by Seider et al.[7] Many theories have been proposed to explain the aetiology of these presentations, including the wrinkling theory and the Phrygian cap theory. In 1970, Bhagwan et al.[6] proposed the wrinkling theory, suggesting that this rare presentation might occur due to the failure of the septations to disappear during embryological development. A systematic review by RS Terkawi et al.[2] demonstrated the development of multiple septations according to the wrinkling theory [Figure-4]. Intraepithelial clefts eventually fuse to form locular spaces. The presence of smooth muscles in the septations, continuous with the muscles of the gallbladder wall as seen in histopathology, further supports this theory.[6]

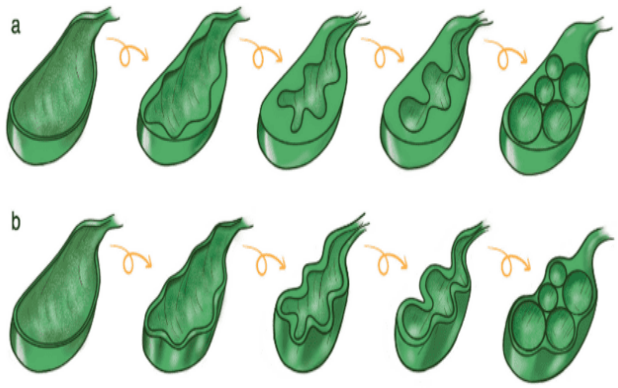

Figure-4: Illustration of the development process of multiseptated gallbladder (MSG). (a) Depiction based on the wrinkling theory. (b): Development of MSG through multiple cystic in-pouching of the gallbladder mucosa

Phrygian cap theory, proposed by Boyden in 1935, suggests that curling and flexures occur in the sac as the GB develops faster than surrounding structures, such as the sac lodge and peritoneum, during developmental stages.[6] Tan et al.[8] explored the development of MGB, based on their research on 11 human embryos, and found multiple cystic in-pouchings of the lumen into the GB mucosa, thought to be precursors to honeycomb GB [Figure-4b].

Some studies suggest that this condition might result from incomplete cavitation of the solid embryonic GB, as there is an absence of muscle layer in the septa of MGB.[4,5] Regardless of age, upper right quadrant pain is the most common symptom (54.5%), followed by epigastric pain (33.3%) and nausea/vomiting (27.2%) [9,10]. There is a significant female predominance, and most patients present with abdominal colic pain, often of long duration.[11] The pain may be attributed to biliary stasis and increased intraluminal pressure, leading to painful sensations. This can be visualized as delayed passage of bile under biliary manometry and scintigraphy.[12] Evidence suggests that USG is the investigation of choice for diagnosing MGB, although other imaging techniques such as CT, ERCP, and MRCP have also been employed. On USG, the GB typically appears to have multiple intercommunicating compartments, a finding not observed in our case. Differential diagnoses include desquamated GB mucosa, cholesterolosis, and GB pseudodiverticulosis.[13–16] ERCP and MRCP can also visualize the honeycomb pattern. Nakazawa et al.[17] suggested that MRCP is a superior imaging modality compared to ERCP, due to its non-invasive nature and ability to simultaneously identify pancreatico-biliary pathologies. They also reported the

visualization of grape-like clusters throughout the GB on MRCP.

Literature suggests that cholecystectomy is the optimal treatment modality for MGB and has been shown to relieve symptoms. In our case, the patient underwent a laparoscopic cholecystectomy and experienced symptom relief post-operatively. While cholecystectomy remains the best treatment option for symptomatic patients, it should also be considered for asymptomatic patients where MGB is an incidental finding.[3,18] Pery M et al. reported a case of MGB with a coexisting choledochal cyst. It was the first instance where MGB was diagnosed pre-operatively using ultrasound, which showed multiple linear echoes within the GB. The patient underwent cholangiography, confirming the presence of multiple septations and diffuse dilation of the CBD suggestive of a choledochal cyst. Based on these radiological findings, the patient underwent cholecystectomy along with choledochoduodenostomy.[19] Similar cases reported by Tan et al.[8] and Bahadir B et al.[20] involved coexisting choledochal cysts, for which the patients underwent Roux-en-Y hepaticojejunostomy. There have also been cases where patients were symptomatic but chose not to undergo surgery, with symptoms resolving over time. However, the majority of cases required surgical intervention for symptom resolution.[21–24]

Diagnosing MGB can be a formidable challenge and may require various radiological imaging modalities. Although USG can effectively diagnose this condition, it can sometimes be missed. Therefore, clinicians should be vigilant during specimen dissection and confirm the diagnosis through histopathology. A thorough review of the literature suggests that cholangiography is the gold standard investigation for this condition.[19] Laparoscopic cholecystectomy is regarded as the optimal surgical intervention due to its advantages, including a lower complication rate, shorter hospital stays, less pain, and faster recovery compared to conventional open cholecystectomy. In our case, the patient underwent laparoscopic cholecystectomy, was discharged the following day, and had an uneventful post-operative period with follow-up for six weeks. However, limitations include the report being based on a single case, which restricts generalizability. Future studies should aim to delineate the association between GB cancer and honeycomb GB. Due to the case report nature of this study, this association is beyond its scope.

CONCLUSIONS

In conclusion, Honeycomb GB is a rare condition characterized by multiple septations within the GB wall. While USG is the most commonly used diagnostic tool, CECT and MRCP can also be effective. Cholangiography remains the gold standard for diagnosis. Although no specific medical treatment exists for honeycomb GB, minimally invasive laparoscopic cholecystectomy is the preferred treatment, as it effectively alleviates symptoms and improves quality of life. This case report highlights the significance of thorough dissection and histopathological evaluation in diagnosing this condition.

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