

Case Report

Khenchoul's Syndrome: An Unusual Clinicoradiological Entity Defined by an Ectopic Duplicated Gallbladder — A Case Report

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Abstract: Ectopic gallbladder (EGB) is a rare condition in which the gallbladder is located outside its normal anatomical position. Gallbladder duplication is an uncommon congenital anomaly, reported in approximately 1 in 4,000 individuals. Although usually asymptomatic, an unrecognized duplicated gallbladder may lead to significant complications, particularly when a second gallbladder is inadvertently left in situ following cholecystectomy. Ectopic gallbladders have been described in various abdominal locations, most commonly beneath the left hepatic lobe, intrahepatic, transverse, retrohepatic, retroperitoneal, within the lesser omentum, or embedded in the falciform ligament. We report the case of a 65-year-old woman who experienced recurrent episodes of abdominal pain and cholangitis for several years following a cholecystectomy performed in 2009. Multiple imaging investigations identified a cystic lesion within the right hepatic region. Interestingly, this lesion demonstrated an inconsistent appearance on liver magnetic resonance imaging, behaving as a “phantom cystic mass.” Surgical exploration, initially planned as a hepatic resection, revealed a cystic structure suggestive of a gallbladder located to the right of the falciform ligament and communicating with the left hepatic duct. Histopathological examination confirmed a true duplicated gallbladder. This finding explained both the patient's recurrent paroxysmal abdominal pain and the intermittent visualization of the cystic lesion on MRI, likely resulting from cyclical filling and emptying of the ectopic gallbladder. To our knowledge, this case describes a previously unreported clinicoradiological entity, for which we propose the term Khenchoul's syndrome, defined as an ectopic gallbladder located to the right of the falciform ligament. This case highlights the diagnostic challenges associated with gallbladder duplication after cholecystectomy and emphasizes the importance of meticulous intraoperative assessment and histopathological confirmation. Furthermore, it expands the spectrum of ectopic gallbladder locations and introduces a potentially novel clinicoradiological entity. This report was prepared in accordance with the SCARE 2023 guidelines.

Keywords: Gallbladder Duplication, Ectopic Gallbladder, Congenital Anomaly, Cholecystectomy, Cholangitis, Biliary Fistula, Case Report.

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INTRODUCTION

Gallbladder duplication is a rare congenital anomaly, with an estimated prevalence of approximately 1 in 4,000 individuals [1]. Although often asymptomatic, this condition may lead to significant clinical complications when one of the gallbladders is inadvertently left in situ following cholecystectomy. Such cases may result in persistent or recurrent biliary symptoms, including cholangitis, biliary fistula formation, and intra-abdominal abscesses [2].

Preoperative identification of this anomaly remains challenging, particularly in resource-limited settings or in the absence of detailed operative records. Furthermore, conventional imaging modalities, including ultrasonography and computed tomography, often lack sufficient sensitivity to reliably distinguish gallbladder duplication from other biliary abnormalities or postoperative changes, especially in the setting of chronic inflammation [3].

Ectopic gallbladder is another rare congenital anomaly characterized by an abnormal location of the gallbladder outside its usual anatomical fossa. A recent review [4], reported that the most common ectopic location was a left-sided gallbladder, accounting for 54.9% of cases. Intrahepatic gallbladders represented 15.5% of cases, whereas the transverse location was the least common, accounting for only 1.5%. No sex-related differences were observed. Ectopic gallbladders have also been reported in association with other syndromes and congenital conditions, including Mirizzi syndrome, Prune-Belly syndrome, and Churg–Strauss syndrome.

We report the incidental discovery of a duplicated ectopic gallbladder in a patient with a history of cholecystectomy and recurrent cholangitis. The diagnosis was established intraoperatively during exploration for a presumed cystic hepatic lesion. This rare entity underscores the diagnostic challenges of postoperative biliary symptoms and the need to consider congenital biliary anomalies in such settings. We further describe a previously unreported clinicoradiological entity, termed Khenchoul's syndrome, defined as a duplicated ectopic gallbladder located to the right of the falciform ligament.

CASE PRESENTATION

A 65-year-old woman with no relevant medical or surgical history underwent an open cholecystectomy in 2009 for symptomatic cholelithiasis.

Since 2020, she developed recurrent episodes of acute cholangitis, typically triggered by copious meals, presenting as right upper quadrant pain and fever without

jaundice. She remained asymptomatic between episodes. These paroxysmal symptoms were suggestive of intermittent distension and collapse of an ectopic gallbladder located within the hepatic parenchyma.

She underwent two endoscopic sphincterotomies, the first in 2020 (not documented) and the second in January 2023, during which a common bile duct stone was extracted using a Dormia basket.

In April 2025, she was admitted with fever and right upper quadrant pain. Clinical examination revealed no jaundice. Laboratory tests showed total bilirubin of 6 mg/L (direct bilirubin 2 mg/L), with normal transaminases (AST 14 IU/L, ALT 19 IU/L).

Magnetic resonance cholangiopancreatography (MRCP) performed in January 2023 had demonstrated multiple intrahepatic abscesses in the left lobe and subphrenic region, associated with moderate intrahepatic bile duct dilatation, more pronounced on the left side. A fibrotic stricture at the biliary confluence and a 20 mm distal common bile duct stone were also identified. A 16 × 10 mm hilar lymph node was noted.

In 2024, a new episode of cholangitis occurred. Abdominal ultrasound revealed marked atrophy of the left hepatic lobe with a homogeneous liver parenchyma and a subphrenic cystic lesion measuring 31 × 25 mm in continuity with dilated bile ducts. A repeat MRCP confirmed a tight stricture at the biliary confluence involving the left hepatic ducts, classified as Bismuth type IIIb, associated with severe left hepatic atrophy, consistent with chronic biliary obstruction.

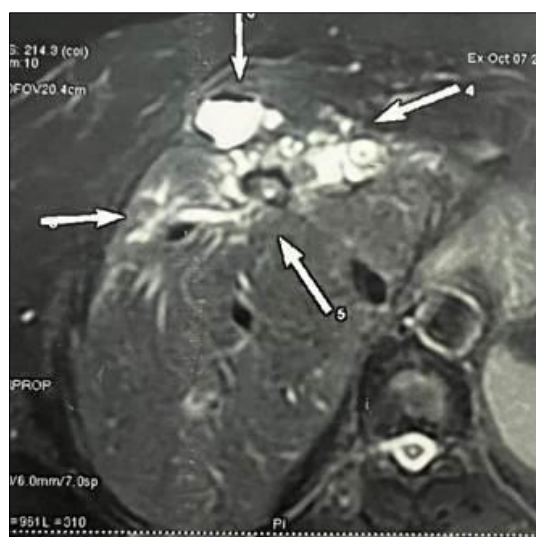


Figure 1: MRI showing a subphrenic cystic lesion

The subphrenic collection persisted despite endoscopic drainage. After multidisciplinary hepatobiliary tumor board discussion, a chronic biliary inflammatory process was favored and surgical

management was indicated, initially planned as a left hepatectomy.

Intraoperative exploration revealed a 4 cm fibrotic cystic structure communicating via a

transhepatic biliary fistula with the left hepatic duct. The lesion demonstrated macroscopic features consistent with a gallbladder-like structure, including characteristic

morphology and a cystic duct-like communication. The lesion was resected, and the fistula was ligated and divided. A drain was placed.



Figure 2: Operative specimen of the subphrenic cystic lesion

Postoperative recovery was uneventful. Drain output was minimal and non-bilious. The patient was discharged on postoperative day 7 in good condition. At one-month follow-up, she was asymptomatic with normal liver function tests and complete radiological resolution of the collection.

Histopathological examination revealed a tubular structure lined by biliary-type epithelium composed of non-atypical columnar cells. Focally, the mucosa was supported by a well-formed muscular layer, consistent with true gallbladder duplication rather than an inflammatory pseudocyst or fistulous tract. No dysplasia or malignancy was identified.

DISCUSSION

Gallbladder duplication is a rare congenital anomaly resulting from abnormal embryologic development of the biliary system, most commonly due to abnormal division or persistence of an accessory bud of the pars cystica, which normally gives rise to the gallbladder, cystic duct, and part of the extrahepatic biliary tree [1-5]. During normal embryogenesis, the hepatic diverticulum appears after the fourth week of gestation and subsequently divides into cranial (pars hepatica) and caudal (pars cystica) components. The pars hepatica gives rise to the liver and intrahepatic bile ducts, whereas the pars cystica forms the extrahepatic biliary system, including the gallbladder and cystic duct. Between the 5th and 12th weeks of gestation, these structures undergo elongation, migration, and recanalization. Disruption of this process may result in accessory or duplicated gallbladders.

These anomalies are usually asymptomatic and often discovered incidentally. In the present case, the diagnosis was delayed for more than 15 years despite recurrent biliary symptoms and multiples interventions.

The prevalence of gallbladder duplication varies according to different studies, ranging from

1/3,800 in the anatomical study by Boyden, to 1/600 in the French ultrasound-based study by Sénécaïl *et al.*, and up to 1/50 in the anatomical study by Gupta *et al.*, A systematic review by Darnis *et al.*, further highlights the heterogeneity of reported anatomical findings in multiple gallbladder anomalies [6].

Clinical presentation is generally non-specific, with most cases being diagnosed in the context of biliary lithiasis or its complications. In a large systematic review, 83% of multiple gallbladder anomalies were identified during evaluation or treatment of biliary stone disease (cholelithiasis, cholecystitis, choledocholithiasis, or acute pancreatitis) [6]. In asymptomatic patients, accessory gallbladders are most often detected incidentally during imaging studies performed for unrelated conditions [6].

In our patient, a complete open cholecystectomy had been performed in 2009. This was followed by recurrent cholangitis, segmental hepatic atrophy, and a persistent subphrenic collection. Radiological investigations (ultrasound, CT, and MRCP) suggested a chronic biliary fistula or abscess, without identifying a congenital biliary anomaly. Even MRCP, which is considered the most sensitive imaging modality for biliary anomalies (up to 96% sensitivity) [3], failed to demonstrate the duplicated gallbladder or the intermittent cystic structure described on previous imaging, likely due to cyclical distension and collapse of the ectopic gallbladder (“phantom cystic lesion”). This highlights the limitations of imaging in the setting of altered postoperative anatomy and chronic inflammation.

Boyden’s classification describes five types of ectopic gallbladder: intrahepatic, left-sided, retrohepatic, floating, and retroperitoneal [1]. Our case does not fit into any previously described category, as the gallbladder was located on the right hepatic side in an ectopic extrahepatic position. This atypical configuration further supports the uniqueness of this observation.

We therefore propose Khenchoul's syndrome, a clinicoradiological entity characterized by:

1. Paroxysmal biliary-type pain,
2. A “phantom” cystic lesion appearing on imaging and disappearing on subsequent examinations,
3. Cyclical distension and collapse of an ectopic duplicated gallbladder explaining both symptom variability and intermittent radiological visibility.

Surgical exploration revealed a well-formed cystic structure communicating with the left hepatic duct, initially interpreted as a chronic biliary fistula. Histopathological examination demonstrated a biliary-type mucosa with a well-developed muscular layer, confirming true gallbladder duplication rather than an inflammatory pseudocyst or fistulous tract. This lesion had remained undiagnosed for more than 15 years and mimicked acquired pathologies such as abscess or chronic fistula. Without accurate intraoperative identification, an unnecessary hepatic resection could have been performed.

Intrahepatic duplicated gallbladders are extremely rare, and their presentation after cholecystectomy is exceptional. The differential diagnosis includes Phrygian cap, Type II choledochal cyst (Todani classification), gallbladder diverticula, Caroli disease, and intraductal papillary neoplasms of the bile duct [7–9]. Ultrasonography and CT have limited diagnostic performance, with reported sensitivity around 60% for distinguishing these entities [10], which may lead to misdiagnosis and overtreatment.

This case emphasizes the importance of considering congenital biliary anomalies in patients with persistent or recurrent biliary symptoms after cholecystectomy, particularly when operative records are incomplete. Careful intraoperative assessment and systematic histopathological evaluation of any abnormal biliary structure are essential to avoid unnecessary surgical procedures.

CONCLUSION

Gallbladder duplication is a rare congenital anomaly that may remain undiagnosed for years, particularly when an accessory gallbladder is left in situ after cholecystectomy. This case highlights the limitations of imaging in the setting of altered biliary anatomy and underscores the importance of intraoperative vigilance and histopathological

confirmation. Accurate recognition of this entity is essential to avoid unnecessary hepatic resections and to ensure appropriate management.

This report further supports the description of a previously unrecognized clinicoradiological entity, Khenchoul's syndrome, characterized by a right paraligamentous ectopic gallbladder presenting with a distinct biliary clinical pattern.

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