CASE REPORT

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A Rare Gallbladder Anomaly Mimicking Choledochal Cyst; Hourglass Gallbladder

Feyyaz Güngör[1], Nihan Acar[2], İbrahim Cüneyit[1], Osman Nuri Dilek[1]

[1]Department of General Surgery, İzmir Katip Çelebi University, Atatürk Training and Research Hospital, İzmir, Türkiye
[2]Department of Anatomy, Pamukkale University Faculty of Medicine, Denizli, Türkiye

Abstract

Biliary tract anomalies are surprises which may cause surgeons unexpected issues. Although biliary system anomalies are frequently seen (42-58%), an hourglass gallbladder is a rare condition which cannot be given a specific rate, since very few cases have been reported in the literature. In this study, a case in which an hourglass gallbladder was unexpectedly detected during an operation is presented in light of the literature data.

Keywords: Hourglass gallbladder, anomalies, diagnosis, treatment

INTRODUCTION

Anatomical variations of the biliary system are seen in 42-58% of the population.[1,2] The hourglass gallbladder, which is one of these variations, has been reported in single case reports in the literature but its incidence is unclear. It is considered to be congenital in the paediatric population and to develop after episodes of cholecystitis in adults. There are no typical findings in clinical presentation and preoperative laboratory tests. It may be detected on preoperative imaging if suspected. Although it is very rare, it is important in terms of morbidity and mortality in patients who develop complications in surgical treatment.

CASE PRESENTATION

A 54-year-old male patient presented with right upper quadrant pain provoked after high-fat meals for three months. His past surgical and medical history, physical examination and laboratory tests were unremarkable. Abdominal ultrasound (US) detected choledochal cyst and cholelithiasis. Magnetic resonance cholangiopancreatography (MRCP) also confirmed the diagnosis of choledochal cyst and classified it as type 2 according to Todani classification (Figure 1). Written informed consent was obtained from the patient and elective surgery was planned. Extrahepatic biliary tract resection was planned conventionally and the patient was operated on after preoperative preparations were completed. During the exploration, it was found that the gallbladder was in the normal position, the gallbladder narrowed from the fundus to the body and then widened again around the neck and the cystic duct was open from the usual site to the distal common bile duct (Figure 2). The extrahepatic biliary ducts were evaluated as completely normal. It was understood that the part which was interpreted as a choledochal cyst was the body and neck part of the gallbladder shaped like an hourglass. Cholecystectomy was performed (Figure 3) and the patient was discharged uneventfully on second day postoperative. The pathological examination was reported as chronic cholecystitis.

DISCUSSION

It is reported that the hourglass gallbladder was first described by Morgagni in 1769.[3] Its incidence is unknown and there are very few case reports in the literature. In post-mortem studies, the duplication rate of any part of the biliary system is 1 in 4,000, and the hourglass gallbladder is a much rarer variant of congenital biliary malformation.[3,4] Although its pathogenesis is unclear, it is considered to be congenital in the paediatric population and to occur due to chronic inflammation and fibrosis caused by cholecystitis.[5] Cases may present with all common symptoms of biliary pathologies which makes differential

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ORCID IDs of the authors: F.G. 0000-0002-4066-6072; N.A. 0000-0003-0720-3794; İ.C. 0000-0003-0555-2114; O.N.D. 0000-0002-6313-3818.

Address for Correspondence: Feyyaz Güngör
E-mail: feyyaz.gngr@gmail.com
ORCID ID: orcid.org/0000-0002-4066-6072

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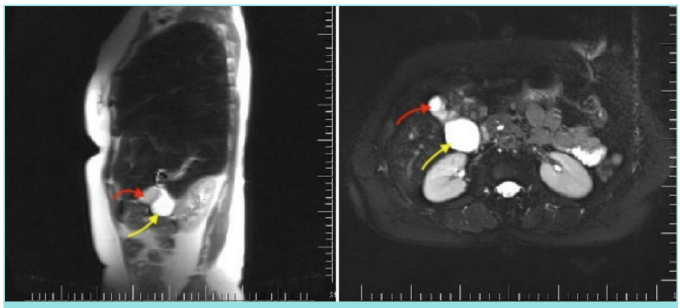

Figure 1. Sagittal and transverse section magnetic resonance cholangiopancreatography (T2 weighted). The yellow arrow was reported as choledochal cyst and the red arrow was reported as gallbladder.

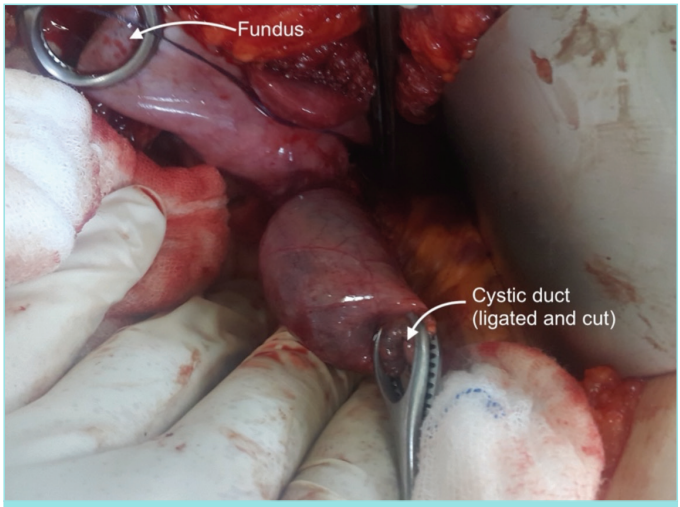

Figure 2. Preoperative image. Cystic duct (ligated and cut) and fundus marked.

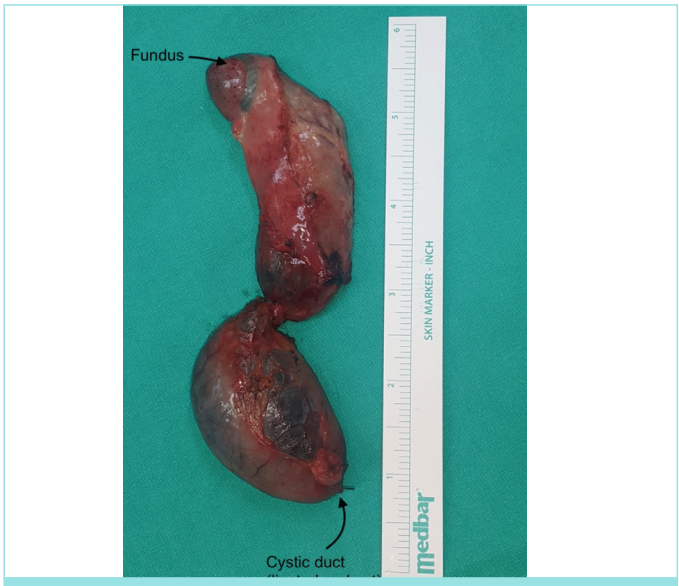

Figure 3. Specimen of hourglass gallbladder.

diagnosis difficult. Laboratory tests are usually normal unless there is an obstructive lesion. It can be detected via US by an experienced radiologist. In the literature, it is reported that similar images may also occur in the segmental type of gallbladder adenomyomatosis.[6] If hourglass gallbladder is suspected or any sign of variation is detected, further evaluation should be carried out in order to confirm the diagnosis and determine whether there is a relation with the biliary tree. Subsequently, MRCP and computed tomography should be performed and if there is still a conflict, hepatobiliary scintigraphy or endoscopic retrograde cholangiopancreatography (ERCP) can be preferred. Although it is an invasive technique, ERCP has a sensitivity of up to 100% in the diagnosis of biliary cysts.[7]

Laparoscopic cholecystectomy is the initial treatment in symptomatic cases. However, in cases which cannot be diagnosed preoperatively such as the case presented here, conventional surgery is also a safe and effective way to evaluate the biliary tract properly.

CONCLUSION

Gallbladder anomalies are rare entities which can rarely be detected with preoperative imaging. Therefore, the surgeon's attention during surgery is vital for diagnosis. In order to prevent possible complications, it is important to follow the rules for safe surgical dissection and to keep in mind potential variations of the biliary system.

MAIN POINTS

- Gall bladder anomalies are rare entities.
- Diagnosis is difficult and can usually be detected preoperatively.
- When detected in the preoperative period, it can be treated with laparoscopic surgery.
- However, in cases which cannot be diagnosed preoperatively such as the one presented here, conventional surgery is also a safe and effective way to evaluate the biliary tract properly.

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ETHICS

Informed Consent: Written informed consents were obtained from the patient who participated in this study and also for the publication of the photos used.

Authorship Contributions

Concept: N.A., Design: İ.C., Supervision: İ.C., Fundings: İ.C., Literature Search: F.G., Writing: F.G., N.A., O.N.D., Critical Review: O.N.D.

DISCLOSURES

Conflict of Interest: The authors have no conflicts of interest to declare.

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