

CASE REPORT

DOI: 10.4274/cjms.2020.2978

A Rare Gallbladder Anomaly Mimicking Choledochal Cyst; Hourglass Gallbladder

Güngör et al. An Hourglass Gallbladder

Feyyaz Güngör¹, Nihan Acar², İbrahim Cüneyit¹, Osman Nuri Dilek¹

¹Department of General Surgery, İzmir Katip Çelebi University, Atatürk Training and Research Hospital, İzmir, Türkiye

²Department of Anatomy, Pamukkale University Faculty of Medicine, Ankara, Türkiye

Abstract

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

Keywords: Hourglass gallbladder, anomalies, diagnosis, treatment.

To cite this article: Güngör F, Acar N, Cüneyit İ, Dilek ON. A Rare Gallbladder Anomaly Mimicking Choledochal Cyst; Hourglass Gallbladder.

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

26.01.2020

05.10.2020

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 as single case reports in the literature and its incidence is unclear. It is considered to be congenital in pediatric population and to develop after episodes of cholecystitis in adults. There is no typical finding 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 PRESENTATIONS

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 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 opened from the usual site to the distal common bile duct. (Figure 2-3). The extrahepatic biliary ducts were evaluated as completely normal. It was understood that the part which was interpreted as choledochal cyst was the body and neck part of the gallbladder shaped as hourglass. Cholecystectomy was performed (Figure 4) and the patient was discharged uneventfully on second postoperative day. The pathological examination was reported as chronic cholecystitis.

DISCUSSION

The hourglass gallbladder was first described in 1769 by Morgagni (3). Its incidence is unknown and there are very few case reports in the literature. In postmortem studies, the duplication rate of any part of the biliary system is 1 in 4000, and the hourglass gallbladder is a much rarer variant of congenital biliary malformation (3, 4). Although the pathogenesis is unclear, it is considered to be congenital in pediatric population and to occur due to chronic inflammation and fibrosis caused by cholecystitis (5). The cases may present with all common symptoms of biliary pathologies which makes the differential diagnosis difficult. Laboratory tests are usually normal unless there is an obstructive lesion. It can be detected with 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 the hourglass gallbladder is suspected or any sign of variation is detected, further evaluation should be carried on in order to confirm the diagnosis and determine whether there is a relation with the biliary tree. Subsequently, MRCP and computed tomography (CT) 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 that can not be diagnosed preoperatively such as here presented case, conventional surgery is also a safe and effective way to evaluate the biliary tract properly.

CONCLUSION

Gallbladder anomalies are rare entities that can hardly 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 the 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 that can not be diagnosed preoperatively such as here presented case, conventional surgery is also a safe and effective way to evaluate the biliary tract properly.

Acknowledgments: This manuscript was presented as a poster in 13th Turkish Congress of Hepatopancreatobiliary Surgery, 1–4 November 2017, Antalya, Turkey.

Informed Consent: Written informed consents were obtained from the patients who participated in this study for this study and also to publish photos of the patients.

Conflict of Interest: All of the authors declare that there are no conflicts of interest in connection with this paper.

Financial Disclosure: All of the authors declare that there are no financial disclosure in connection with this paper.

REFERENCES

1. Morteale KJ, Rocha TC, Streeter JL, Taylor AJ. Multimodality imaging of pancreatic and biliary congenital anomalies. *Radiographics*. 2006; 26: 715–31.
2. Turner MA, Fulcher AS. The cystic duct: Normal anatomy and disease processes. *Radiographics*. 2001; 21: 3–22.
3. St-Vil D, Luks FI, Hancock BJ, et al. Diaphragm of the gallbladder: a case report. *J Pediatr Surg*. 1992; 27: 1301-3.
4. Tsoraides SS, Cha AI, Crawford DL. Postcholecystectomy biliary symptoms. *J SurgEduc*. 2007; 64: 228-33.
5. Reddy M, Studentsov Y, Castronuova J. Acquired hourglass gallbladder. *ClinNuclearMed*. 2001; 26(2): 153.
6. Wong HYF, Lee KH. The hourglass gallbladder. *AbdomRadiol (NY)*. 2017, doi: 10.1007/s00261-017-1273-6.
7. Keil R, Snajdauf J, Rygl M, et al. Diagnostic efficacy of ERCP in cholestatic infants and neonates--a retrospective study on a large series. *Endoscopy*. 2010; 42: 121.

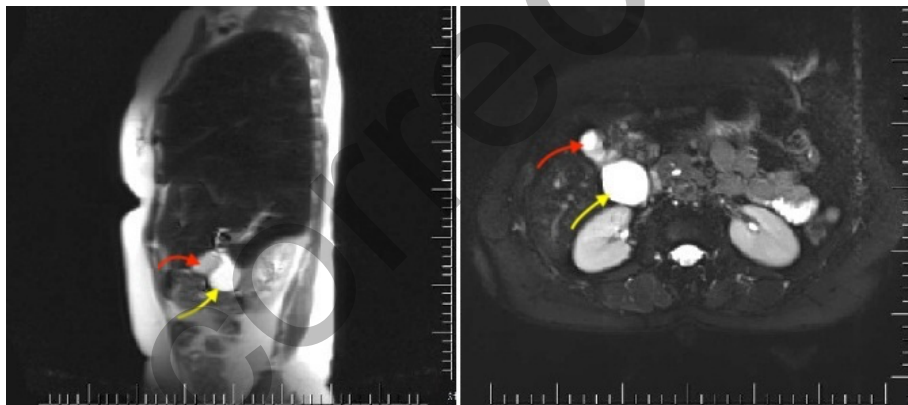


Figure 1. Sagittal and transverse section magnetic resonance cholangiopancreatography (T2 weighted). The yellow arrow was reported as choledochal cyst and the red arrow was reported as gall bladder

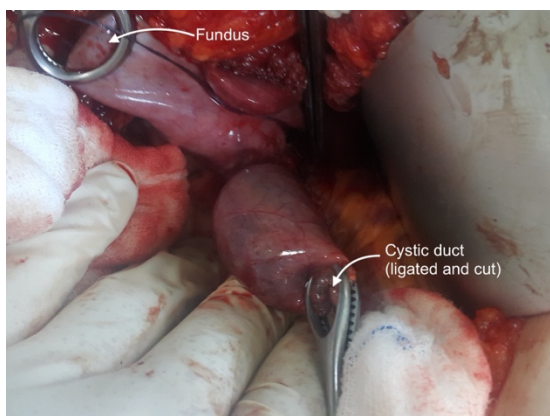


Figure 2. Peroperative image. Cystic duct (ligated and cut) and fundus marked

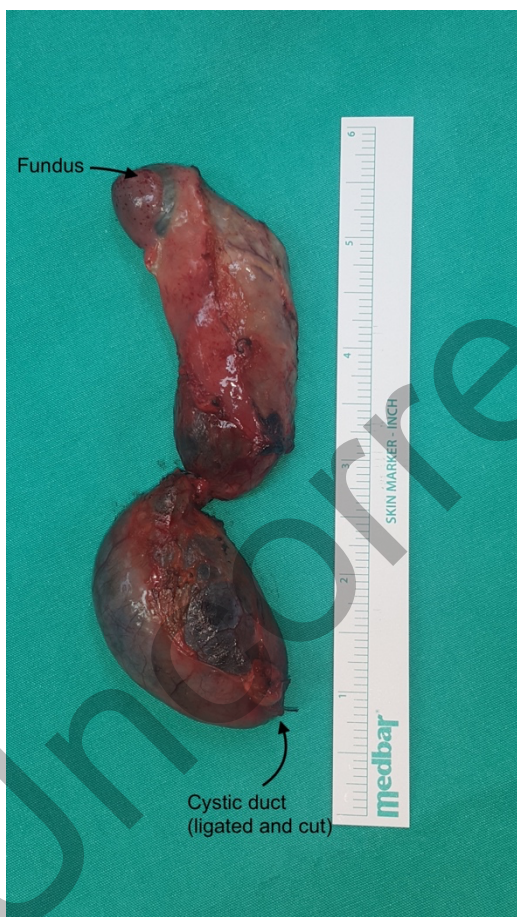


Figure 3. Demonstrative view of the hourglass gallbladder

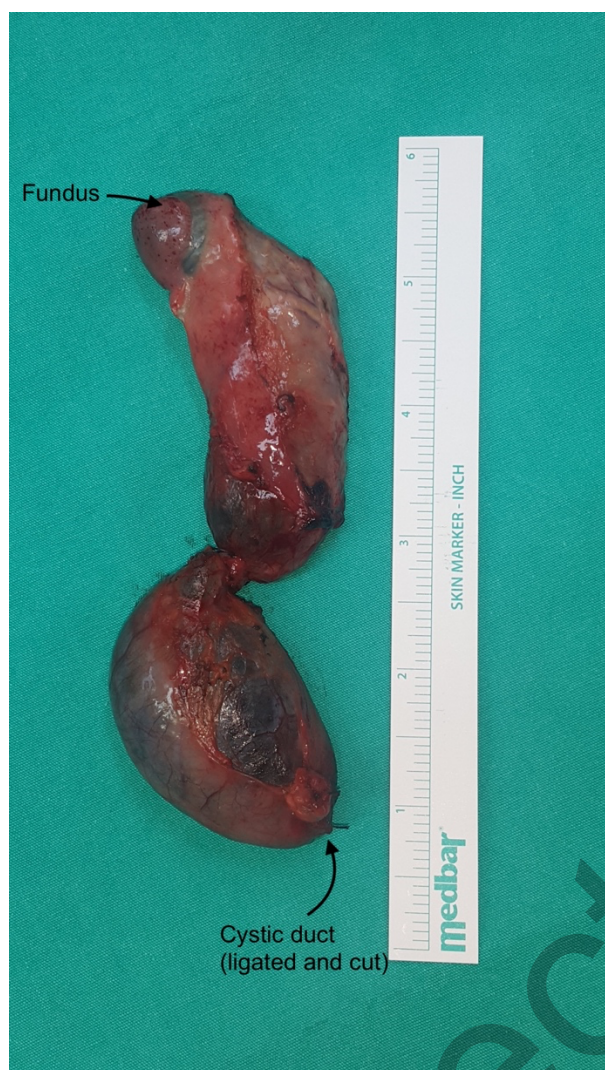


Figure 4. Specimen of hourglass gallbladder