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Case Report

A Case of Symptomatic Gallbladder Agenesis with Chronic Abdominal Symptoms

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Abstract

The anatomical area of the extrahepatic bile ducts exhibits plethora of anatomic variants. The detailed study and comprehension of anatomic variations of extrahepatic bile ducts is a prerequisite in order to avoid the intraoperative biliary or tract damages, but they are also necessary for the targeted treatment of any complications. Gallbladder agenesis is a rare congenital anomaly of the biliary tree with an estimated incidence of 0.007-0.027% in surgical series which is much lower compared to the incidence of other gallbladder anomalies. It may be asymptomatic, but sometimes is associated with symptoms such as upper quadrant abdominal pain, which may be mistaken for cholecystitis and can lead the patient to the operating room. We present a case of a 30-year-old male patient without any significant past medical history presented with a 2-year history of upper abdominal complaints, dyspepsia, epigastric abdominal pain and weight loss, normal laboratory workup and unclear radiological signs which led him to exploratory laparoscopy due to the patient's chronic symptoms, in order to exclude the presence of another underlying pathologic process. In addition to our case presentation, a relative review of literature was conducted. As a conclusion, examinations, such as transabdominal ultrasonography, may be misleading and MCRP should be the principal method of investigation to establish a presumptive diagnosis. However, in cases with a strong suspicion for a different underlying pathology, further investigation with exploratory laparoscopy may be warranted.

Keywords

agenesis, gallbladder, laparoscopy

INTRODUCTION

Gallbladder agenesis is a rare congenital anomaly of the biliary tree with an estimated incidence of 0.007-0.027% in surgical series (0.04-0.13% in autopsy series)[1], which is much lower compared to the incidence of other gallbladder anomalies, such as double or bilobate gallbladder that are found in up to 10% of autopsy cases.[2] Approximately half of all patients are symptomatic with an average age of 46 years at the time of presentation, but diagnosis is often difficult and requires a high degree of suspicion. Although the anomaly is equally encountered among males and females in autopsy studies, women are two times more likely to be symptomatic, which is concurrent to the patterns of symptoms for other biliary diseases.[3] Extrahepatic biliary atresia may be accompanied by gallbladder agenesis in an estimated 1 out of 6 cases,[1,4] while an underlying hereditary component may also be present.[5] Gallbladder agenesis may be associated with several other genitourinary, gastrointestinal or cardiovascular malformations, which often occur in similar patterns.[6] We report a case of a 30-year-old patient with gallbladder agenesis that underwent exploratory laparoscopy, with subsequent symptomatic relief at one year after surgery.

CASE REPORT

A 30-year-old male without any significant past medical history presented with a 2-year history of upper abdominal complaints, dyspepsia, epigastric abdominal pain and weight loss. Laboratory work-up did not reveal any increases in either total/direct bilirubin or liver functional tests. He had previously undergone esophagogastroduodenoscopy without any findings. Transabdominal ultrasonography revealed a hypo-echoic area in the right upper quadrant, which was presumed to be a dilated, overloaded gallbladder, without dilation of the intra- or extra-hepatic biliary tree ***(Fig. 1a)****. The patient then underwent magnetic resonance cholangiopancreatography, which did not visualize either the gallbladder or the cystic duct (****Fig. 1b****). At this point, the diagnosis of gallbladder agenesis was made, but the decision was made for the patient to undergo exploratory laparoscopy due to the patient's chronic symptoms, in order to exclude the presence of another underlying pathologic process.*

*The patient was subsequently taken to the operating room where the gallbladder and cystic duct were not visualized. The rest of the extra-hepatic biliary tree was present and no other macroscopic anomalies were visualized (****Fig. 2****). In addition, there were no other anomalies identified inside the abdomen. The patient had an uneventful recovery after the procedure and was discharged on the first postoperative day. After twelve months of follow-up, the patient did not have further abdominal complaints and did not undergo any additional therapeutic interventions.*

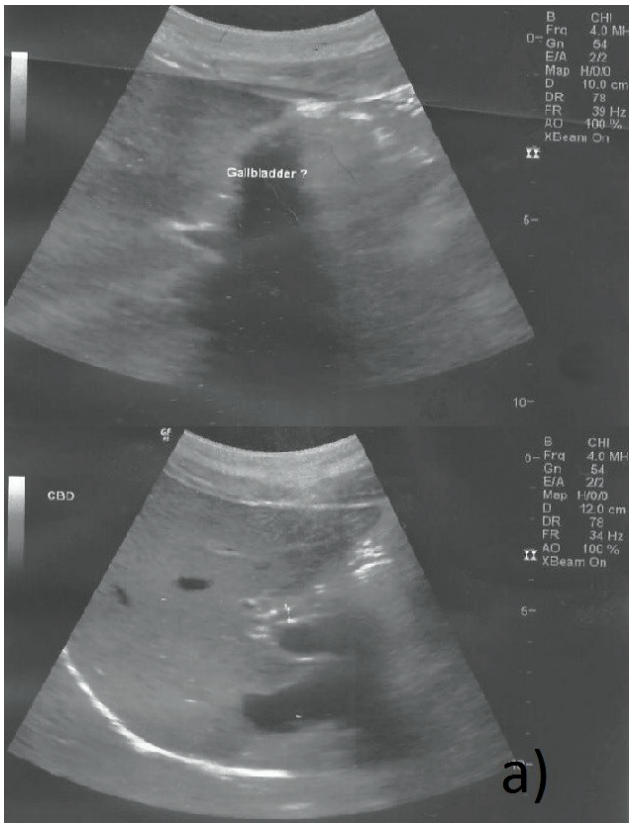

Figure 1a. Preoperative ultrasound showing a dilated gallbladder.

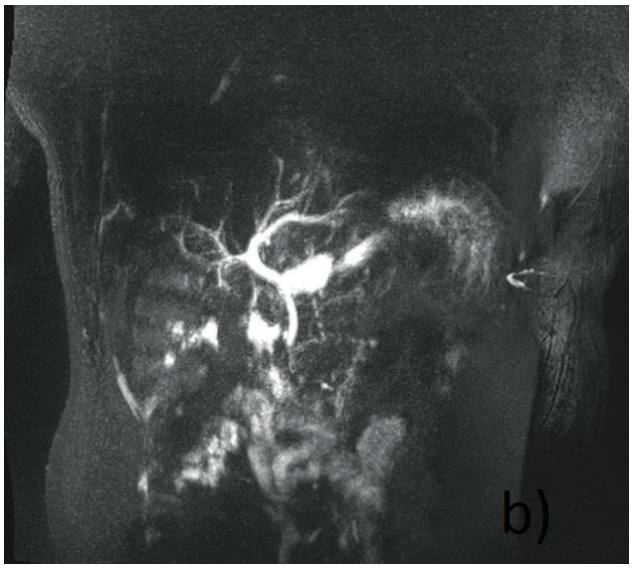

Figure 1b. Magnetic Resonance Cholangiopancreatography (MRCP) image not visualising the gallbladder.

DISCUSSION

Patients with gallbladder agenesis may present with right upper quadrant abdominal pain, which may be mistaken for cholecystitis. This symptomatology could be a result to Oddi Sphincter dysfunction[7,8], but may also be related to choledocholithiasis.[9] Diagnosis in symptomatic patients with gallbladder agenesis and upper abdominal complaints may be challenging, due to the fact that the gallbladder may falsely be visualized as atrophic and contracted, with potential presence of choledocholithiasis.

The WES triad (demonstration of gallbladder wall, echo of stone and acoustic shadow) has been proposed in order to differentiate between a contracted gallbladder with gallstones and bowel loops.[10] Ultrasonography may also be performed after fasting in order to reduce the amount of gas present in the bowel and assist in distinguishing bowel loops from an absent gallbladder. However, MRCP remains the best available method for diagnosing gallbladder agenesis[11], by demonstrating the non-visualization of the gallbladder and should be considered as a complementary imaging investigation.[12] The same examination may also assist in excluding the presence of an ectopic or intrahepatic gallbladder, which may be challenging to perform intraoperatively.[4] In contrast, the non-visualization of the gallbladder with either endoscopic retrograde cholangiopancreatography (ERCP) or hepatobiliary scintigraphy with ^{99}Tc hepatobiliary iminodiacetic acid (HIDA scan), may be misinterpreted as cystic duct obstruction or other conditions.[13] In some cases, however, combined with other imaging techniques, HIDA can be useful.[14]

Our patient demonstrated symptomatic relief after exploratory laparoscopy without any additional therapeutic interventions. This observation has previously been reported

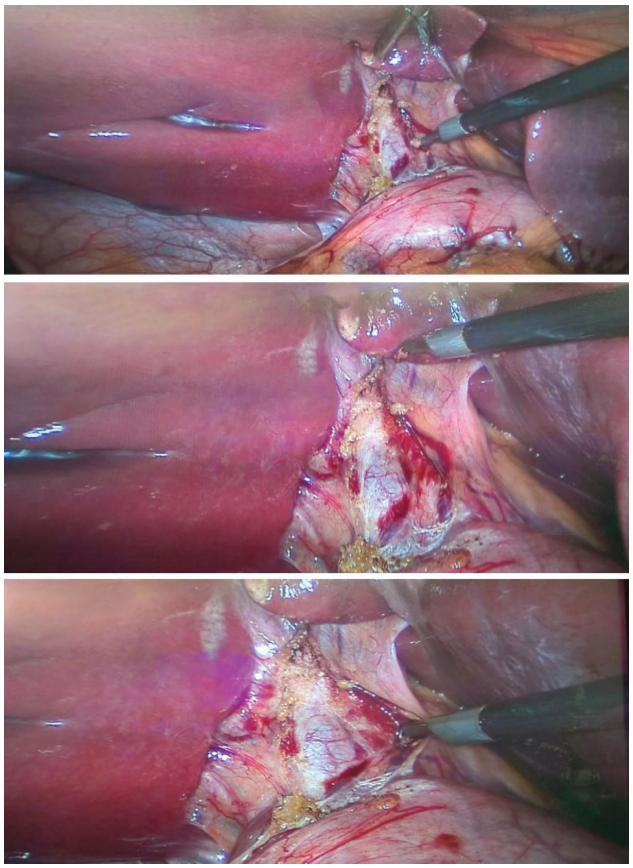

Figure 2. The gallbladder was not recognised intraoperatively.

in a case series by Bennion et al., where all 5 symptomatic patients were symptom-free postoperatively.[9] The underlying mechanism behind symptomatic relief is not known, but may be related to improved biliary kinetics. Exploratory laparoscopy may be challenging and may lead to biliary injuries during the intraoperative search for the gallbladder, especially during the dissection through the portal structures with absent anatomic landmarks.[15,16] For this reason, when a patient undergoes exploratory laparoscopy with a preoperative suspicion for gallbladder agenesis based on imaging tests, extensive intraoperative search for the gallbladder should not be pursued in order to avoid biliary injuries.

CONCLUSION

Patients with gallbladder agenesis may present with vague, non-specific complaints. Routine examinations, such as transabdominal ultrasonography, may be misleading and MCRP should be the principal method of investigation to establish a presumptive diagnosis. However, in cases with a strong suspicion for a different underlying pathology, further investigation with exploratory laparoscopy may be warranted. The latter may also be therapeutic by providing symptomatic relief, but the underlying mechanism behind this relief is not currently known.

REFERENCES

1. Bianco G, Frongillo F, Agnes S, et al. Gallbladder agenesis: A case report and brief review. Ann Hepatobiliary Pancreat Surg 2018; 22(3):292–5.
2. Rabinovitch J, Rabinovitch P, Rosenblatt P, et al. Congenital anomalies of the gallbladder. Ann Surg 1958; 148(2):161–8.
3. Bahraini A, Odom JW, Talukder A. A case report of a patient with gallbladder agenesis resulting in a common bile duct injury. Int J Surg Case Rep 2018; 51:99–101.
4. Fiaschetti V, Calabrese G, Viarani S, et al. Gallbladder agenesis and cystic duct absence in an adult patient diagnosed by magnetic resonance cholangiography: report of a case and review of the literature. Case Rep Med 2009; 2009: 674768.
5. Nadeau LA, Cloutier WA, Konecki JT, et al. Hereditary gallbladder agenesis: twelve cases in the same family. J Maine Med Assoc 1972; 63(1):1–4.
6. Turkel SB, Swanson V, Chandrasoma P. Malformations associated with congenital absence of the gall bladder. J Med Genet 1983; 20(6):445–9.
7. Tagliaferri E, Bergmann H, Hammans S, et al. Agenesis of the gallbladder: role of clinical suspicion and magnetic resonance to avoid unnecessary surgery. Case Rep Gastroenterol 2016; 10(3):819–25.
8. Toouli J, Geenen JE, Hogan WJ, et al. Sphincter of Oddi motor activity: a comparison between patients with common bile duct stones and controls. Gastroenterology 1982; 82(1): 111–7.
9. Bennion RS, Thompson JE, Tompkins RK. Agenesis of the gallbladder without extrahepatic biliary atresia. Arch Surg 1988; 123(10): 1257–60.
10. MacDonald FR, Cooperberg PL, Cohen MM. The WES triad - a specific sonographic sign of gallstones in the contracted gallbladder. Gastrointest Radiol 1981; 6(1):39–41.
11. Pipia I, Kenchadze G, Demetrashvili Z, et al. Gallbladder agenesis: a case report and review of the literature. Int J Surg Case Rep 2018; 53: 235–7.
12. Molnar C, Sárközi T, Kwizera C, et al. Gallbladder agenesis - a rare congenital anomaly mimicking cholelithiasis in an adult woman. Orv Hetil 2019; 160(38): 1510–3.
13. Rajkumar A, Piya A. Gallbladder agenesis: a rare embryonic cause of recurrent biliary colic. Am J Case Rep 2017; 18: 334–8.
14. Haghighatafshar M, Amirkhani Z, Ghaedian T. Role of hepatobiliary scintigraphy in the diagnosis of gallbladder agenesis; a case report and a brief review of literature. Middle East J Dig Dis 2018; 10: 109–13.
15. Cabajo Caballero MA, Martin del Olmo JC, Blanco Alvarez JI, et al. Gallbladder and cystic duct absence. An infrequent malformation in laparoscopic surgery. Surg Endosc 1997; 11(5): 483–4.
16. Peloponissios N, Gillet M, Cavin R, et al. Agenesis of the gallbladder: a dangerously misdiagnosed malformation. World J Gastroenterol 2005; 11(39): 6228–31.

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Случай симптоматической агенезии жёлчного пузыря с хроническими абдоминальными симптомами

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Резюме

Анатомическая область внепечёночных жёлчных протоков обнаруживает множество анатомических вариаций. Детальное изучение и понимание анатомических вариаций внепечёночных жёлчных протоков является необходимым условием для предотвращения интраоперационного повреждения желчи или тракта, но они также необходимы для целенаправленного лечения любых осложнений. Агенезия жёлчного пузыря - это редкая врождённая аномалия жёлчного пузыря с частотой приблизительно 0,007-0,027% в хирургическом ряду, что намного ниже, чем частота других аномалий жёлчного пузыря. Это может протекать бессимптомно, но иногда проявляется такими симптомами, как боль в животе в верхнем квадранте, которая может быть ошибочно принята за холецистит и потребовать хирургического вмешательства. Мы представляем случай 30-летнего пациента мужского пола без каких-либо особенностей в истории заболевания, который в течение двух лет жаловался на боли в верхней части живота, диспепсию, боли в эпигастральной области живота и потерю веса, с нормальными результатами лабораторных исследований и неясными рентгенологическим доказательствами, что потребовало применения диагностической лапароскопии из-за хронических симптомов пациента, чтобы исключить наличие другого скрытого патологического процесса. Помимо презентации нашего случая, было проведено исследование доступной нам литературы. В заключение можно уточнить, что такие тесты, как трансабдоминальная ультрасонография, могут ввести в заблуждение, и магнитно-резонансная холангиопанкреатография (MRCP) должна быть ведущим методом исследования для установления предварительного диагноза. Однако в тех случаях, когда есть подозрение на другую скрытую патологию, может потребоваться дополнительное обследование с применением диагностической лапароскопией.

Ключевые слова

агенезия, жёлчный пузырь, лапароскопия