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RESEARCH ARTICLE

AGENESIS OF THE GALLBLADDER: CONTRACTED, OPERATED OR ABSENT?

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ABSTRACT

Gallbladder agenesis (GA) is an extremely rare condition. Some people with this rare condition can refer to hospital with biliary colic-like symptoms. This can lead to patients being misdiagnosed and unnecessarily operated. Here a 58-year-old female patient who presented with complaints of nausea, vomiting, abdominal pain is presented. Because of the pain in the back of the epigastric region, high serum amylase and lipase values, it was thought to be acute pancreatitis. Furthermore, gallbladder was absent, ductus choledocus was dilatated but intrahepatic bile ducts were normal in the sonographic examination and IV contrast enhanced abdominal computed tomography (CT) which were performed to investigate the etiology and the severity of pancreatitis. Upper abdominal Magnetic Resonance (MR) imaging and Magnetic Resonance Cholangiopancreatography (MRCP) examinations were performed to evaluate the biliary tract and gallstones more clearly on the patient who had no gallbladder operation history. Finally the patient was reported as gallbladder agenesis by MRCP. To avoid unnecessary surgical procedures, it is recommended that suspicious patients must to be evaluated with MRCP when gallbladder is not clearly identified via US or CT.

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INTRODUCTION

GA is a rare condition that affects women more frequently and is diagnosed on average at the second and third decades of life. The incidence varies from 13 to 65 per 100,000. It has shown that the incidence may be higher in some autopsy series (90 per 100,000) (Kasi, 2011; Mittal *et al*., 2011). Some of the patients are referred to the hospital with biliary colic symptoms eventhough they have no gallbladder (Kasi, 2011; 3). The etiology of GA is unknown, it is often a sporadic asymptomatic occurrence with no clear causes (Kobacker, 1950). Ultrasonography (US) is the first choice in the evaluation of gallbladder and bile ducts. However, it is difficult to distinguish GA, contracted pouch and stone fullfilled pouch (scleroatrophic or contracted lithiasic gallbladder) with US. It requires highly experienced ultrasound-maker (Fiaschetti *et al*., 2009). Endoscopic retrograde cholangiopancreatography (ERCP) also may be performed as a diagnostic method.

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However, significant mortality and morbidity with high rates of unsuccessful cannulation and interpretation of nonvisualization of the gallbladder as occlusion of the cystic duct, appear to be handicaps of ERCP (Fiaschetti *et al*., 2009). MRCP is a non-invasive and well-demonstrated imaging method in the evaluation of the biliary-tract. As it does not require contrast administration to visualize the bile, it is not compromised by biliary-stasis. It can also demonstrate an excluded and/or ectopic gallbladder. In this case report, an adult female patient who presented with acute pancreatitis and proved to be congenital GA by radilogical imaging techniques (US, CECT, MR imagingand MRCP), is reported. Besides etiology and the superiority of MRCP to other radiological imaging techniques are discussed.

Case report: A 58-year-old woman admitted to the hospital with complaints of abdominal pain, nausea and vomiting. There was no significant finding other than epigastric tenderness on physical examination. Amylase, lipase, ALT, AST, CRP, creatinine and direct bilirubin were found to be higher in hematological and biochemical studies. The patient was diagnosed as acute pancreatitis because of pain in the back of the epigastric region, high serum amylase and lipase values.

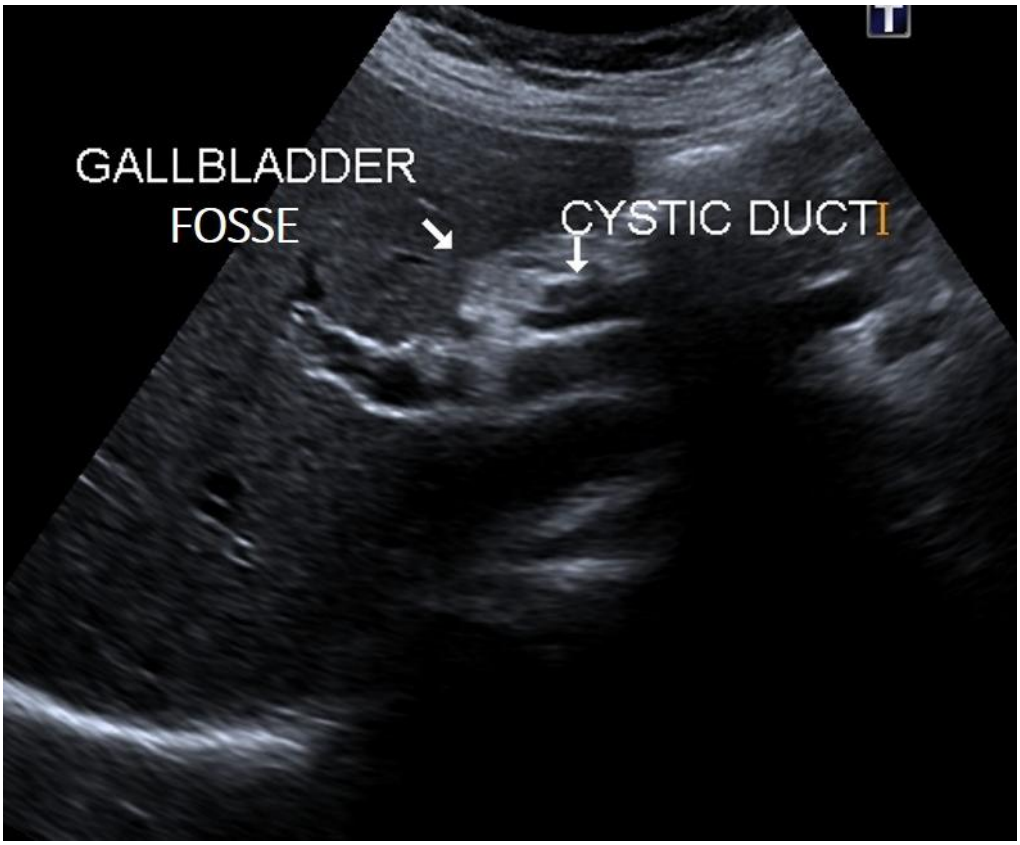

Figure 1. US image demonstrated a hyperechoic area in the gallbladder fossa. And a blind-ended cystic duct originating from middle part of common bile duct

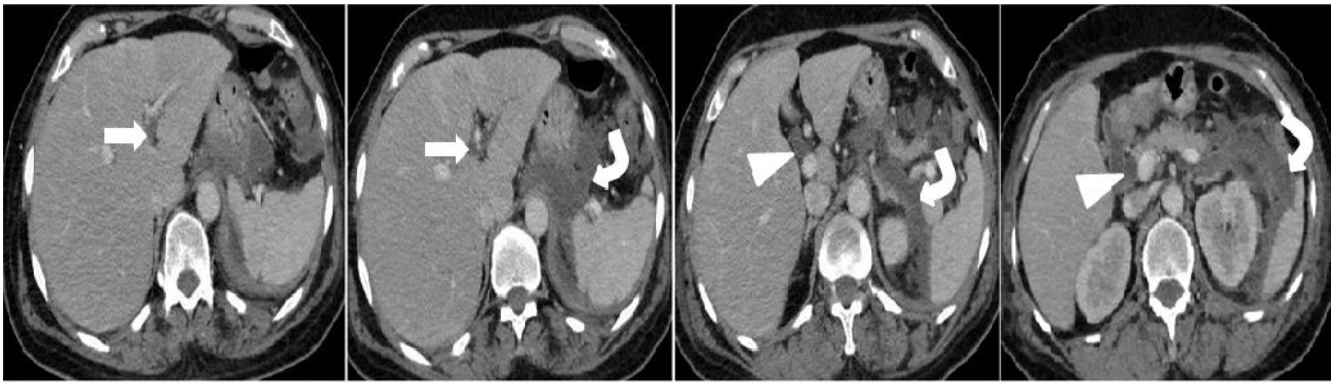

Figure 2. IV contrast enhanced Abdominal CT scan shows left main bile duct (arrow), choledoch (arrowhead) dilated. There is no material present that may be attributed to the operation performed in this section. Common fluid that supports acute pancreatitis extending from the peripancreatic area to the stomach, to the perisplenic area, and to the left kidney is present (curved arrow)

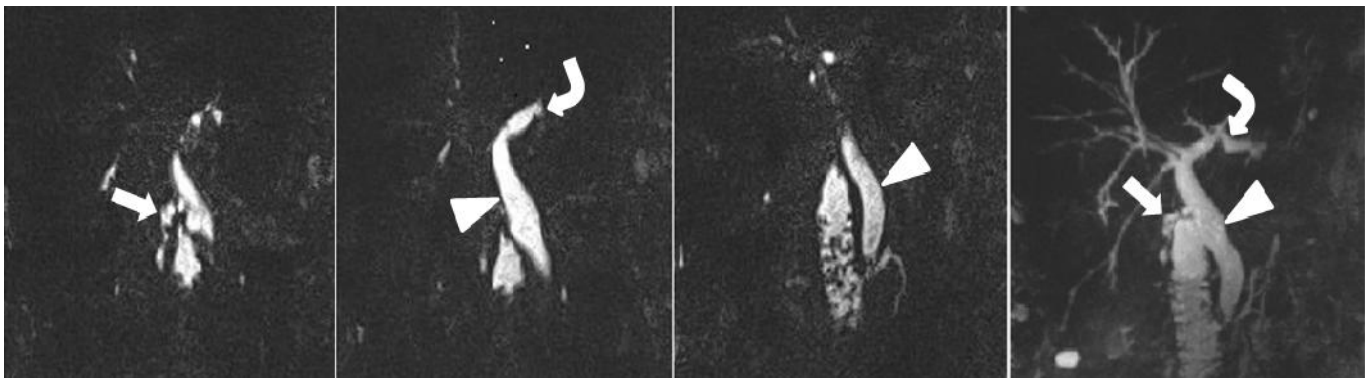

Figure 3. MRCP images demonstrated that there is a blind-ended cystic duct (arrow) originating from middle part of common bile duct. Left main bile duct (curved arrow) and choledoch (arrowhead) are dilated.

Therefore, the patient was sent to our radiology department to investigate the presence of stones in the bile ducts and other etiology of acute pancreatitis. US examinations were performed with a TOSHIBA Aplio 500 convex transducer (1.9-6 MHz; Toshiba Medical Systems Corporation, Tokyo, Japan). Pancreas was not able to be evaluated the because of the intense gas artifact in the abdomen US. There was mild free fluid in retro peritoneum. The intrahepatic bile ducts were normal. The proximal part of the ductus choledochus was dilatated. The gallbladder could not be visualized at the expected localization although the patient had no previous surgical history. There was an interrupted cystic duct originating from middle part of choledochus extending to the gallbladder fosse in the US (Figure1). So that further imagination techniques were recommended for the patient. İntravenous contrast enhanced CT was performed with a 64-detector row CT scanner (Brilliance CT system; Philips Healthcare, Cleveland, OH). There were fluid collections consistent with pancreatitis, which was thought to be originated from the pancreas tail segment, and dilatation in ductus choledochus in the abdominal CT which was performed to to evaluate the severity index of acute pancreatitis. The gallbladder could not be visualized in the expected localization and reported as probably status postcholecystectomy (Figure 2). Furthermore MRCP was recommended to evaluate bile ducts and gallbladder. MRCP was performed with 1.5-T Siemens MagnetomAera, Germany. Via MRCP a rudimenter cystic duct was apparent wheraeas a gallbladder was absent. Thus the diagnosis of GA was confirmed (Figure 3). Endoscopic retrograde cholangiopancreatography (ERCP) was performed because it was thought that there could be lower choledochal microstones in the patient who had continuing complaints; abdominal pain and high CRP and leukocytosis. Sphincterotomy was performed during ERCP and the patient was recovered from pain. CRP and leukocytosis receded and the patient was discharged with clinical recovery.

DISCUSSION

Although the pathogenesis of GA is not known precisely, it is accepted as a congenital malformation. 40-65% of cases are associated with other malformations such as cerebrotendinous xanthomatosis and the G syndrome, trisomy 18 and Klippel-Feil Syndrome (Peloponissios *et al*., 2005). Hepatic diverticulum leads to liver, gallbladder and bile ducts in a 4-week embryo. The end portion of the hepatic diverticulum is enlarged and separated in order to form caudal and rostral branches. The caudal branch gradually swells and extends, resulting in the shape of the gallbladder. In the meantime, the proximal narrow segment forms the cystic duct. Caudal branch hypoplasia causes gall bladder and cystic duct agenesis (Malde, 2010; Yener *et al*., 2015). In literature it has been reported that some patients with GA were referred to hospitals with biliary colic-like symptoms and were unnecessarily operated (Malde, 2010; Yener *et al*., 2015; Tang *et al*., 2015) Some postulate that an associated sphincter of Oddi dysfunction may be the cause of biliary colic in these patients (Wright, 1965). In other cases, associated development of common bile duct stones may be the cause (Malde, 2010). Although it can be demonstrated with US and other advanced imaging modalities, it is not easy to diagnose GA. US often misleads in the diagnosis of GA. Cases of GA have been reported as 'contracted/fibrotic gallbladder' on US. Misdiagnosis causes unnecessary operations which may legally expose physicians to certain sanctions.

The conditions that cause misdiagnosis are biliary colic-like symptoms such as right upper quadrant pain, jaundice, itching; reported as false-positive due to intestinal gas artefact, dilatated bile duct with calcified stone, ribbed fibrous tissue in the galbladder localization; physicians should not consider GA in differential diagnosis (Tang *et al*., 2015; Trompetas *et al*., 2004). Former situtions creates unnecessary costs, some operational-related risks for patients and malpractise sanctions for surgeons. Advanced imaging studies such as CT and MRCP have shown to increase the preoperative diagnosis rate (Fiaschetti *et al*., 2009; Peloponissios *et al*., 2005; Demir, 2008; Balakarishnan, 2016). Upper abdominal CT is usually basic imaging method to evaluate pancreatitis however it is often reported as status postcholecystectomy if gallbladder is not visualized, just as in ourcase. Besides CT gives poor information about intrahepatic and extra hepatic bile ducts.We recommend evaluation with advanced imaging techniques such as MRCP in case of doubtful cases to avoid unnecessary surgical procedures due to misdiagnosis and legal sanctions that may result. Currently it is still difficult to diagnose GA. Nevertheless, physicians should have suspicion of GA in differential diagnosis, and the demand for advanced imaging tests can help prevent unnecessary surgeries in these cases.

Conclusion

In conclusion, GA is a diagnosis that should be kept in mind in patients with no cholecystectomy anamnesis, who do not have ultrasonographic and CECT gallbladder vision in expected location. MRCP is a very effective and noninvasive method for diagnosing GA and evaluating other biliary pathways. Thus, the cost and morbidity of unnecessary surgical procedures can be avoided.

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