



Role of Hepatobiliary Scintigraphy in the Diagnosis of Gallbladder Agenesis; A Case Report and Brief Review of Literature

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ABSTRACT

Gallbladder agenesis (GA) is a rare congenital anomaly. Only 50% of the cases with GA are symptomatic, presenting mostly in the 4th or 5th decade of life. The clinical presentation of GA and imaging findings are non-specific and often misinterpreted as other diseases such as ectopic gall bladder. This can lead to unnecessary surgery when the final diagnosis is usually made. Although GA can cause an identical pattern to acute cholecystitis in hepatobiliary scintigraphy, in certain clinical settings, scintigraphy can be helpful as a confirmatory study, especially to rule out the possibility of ectopic gall bladder. In our case, the combination of imaging findings including ultrasonography, computed tomography, and hepatobiliary scintigraphy led to accurate diagnosis avoiding further surgeries.

KEYWORDS:

Gallbladder agenesis, Hepatobiliary scintigraphy, Ectopic gall bladder

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INTRODUCTION

Gallbladder agenesis (GA) is rare with an incidence of 10-65 per 100,000 population.^{1,2} Autopsy studies have shown a higher incidence.³ GA has been reported for the first time by Bergman back in 1702.^{1,2,4-6}

Only 50% of cases with GA will become symptomatic in the 4th or 5th decades of life.⁷ GA is almost always misinterpreted clinically as cholecystitis or as scleroatrophic gallbladder.⁸ Due to non-specific nature of the symptoms and imaging findings as well as being unexpected, it is usually difficult to diagnose GA correctly in symptomatic patients before surgery. Thus, many patients with GA finally underwent unnecessary surgery as a result of indefinite preoperative diagnosis. Although no specific surgical procedure has been introduced, most of the affected patients become asymptomatic for unexplained reasons after the surgery.⁷

CASE REPORT

A 60-year-old woman presented with a 3-year history of episodic colicky right upper and lower abdominal pain, which aggravated by fatty food, was referred to our nuclear medicine department for hepatobiliary

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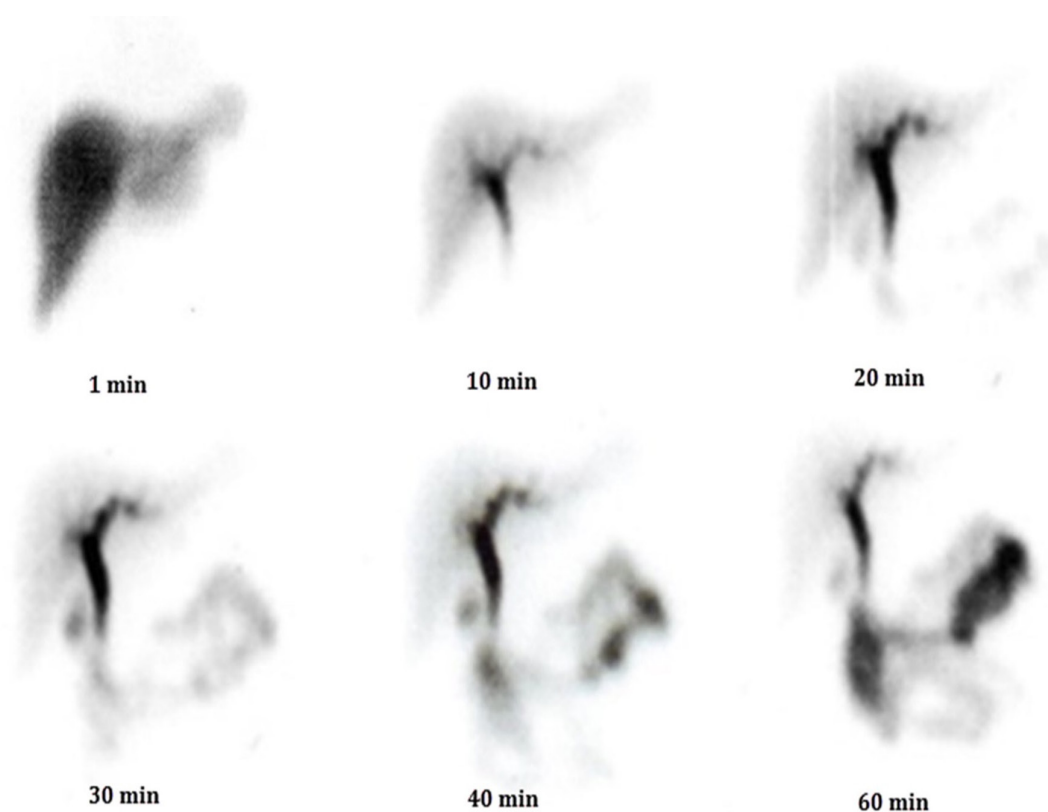


Fig.1: The selected dynamic frames of ^{99m}Tc -BrIDA scintigraphy in anterior projection show normal early homogenous liver uptake with subsequent excretion of bile to the biliary tree and intestine with no evidence of accumulation of tracer in the region of gall bladder or other parts of the abdomen confirming the anatomical imaging findings of gallbladder agenesis.

scintigraphy. There was no history of jaundice or fever. The blood pressure and pulse rate were regular and body temperature was normal. Leukocyte count and liver function tests were within normal limits. The patient also had history of previous laparoscopic appendectomy last year. Ultrasonographic (US) examination 1 week before the hepatobiliary scan, could not visualize the gallbladder clearly. Hepatobiliary scintigraphy with 5 mCi ^{99m}Tc -BrIDA was performed and did not show filling of the gallbladder till 4 hours after injection despite the normal entrance of tracer into the bowels in early images (figure 1). Although this pattern is highly suggestive of acute cholecystitis, the patient was referred for further anatomical correlation with computed tomography (CT). Surprisingly, no evidence of gall bladder was revealed in abdominal CT as well (figure 2). Retrospective assessment of operation note revealed that during search in the undersurface

of the liver, the gallbladder was not seen in that time. She was effectively managed with conservative treatment with smooth muscle relaxants and became symptom-free after medication.

DISCUSSION

Although other anatomic anomalies of the biliary tract are considered to be not uncommon, GA is rare.⁹ As it is usually asymptomatic, it is often detected incidentally.¹⁰ While detected during surgery, there is a female predominance of 3:1; however, cases found in autopsies have an equal sex ratio.¹¹ Approximately 70% of the cases are usually isolated. Some familial cases, however, are associated with more severe anomalies.^{5,12-14} The most common associated anomalies involve the genitourinary tract, and gastrointestinal and cardiovascular systems. As the positive family history is just presented in some cases, a sporadic occurrence is suggested.¹⁵

Only 50% of cases will become symptomatic with no characteristic symptomatology. Right upper quadrant abdominal pain is the most common presenting symptom

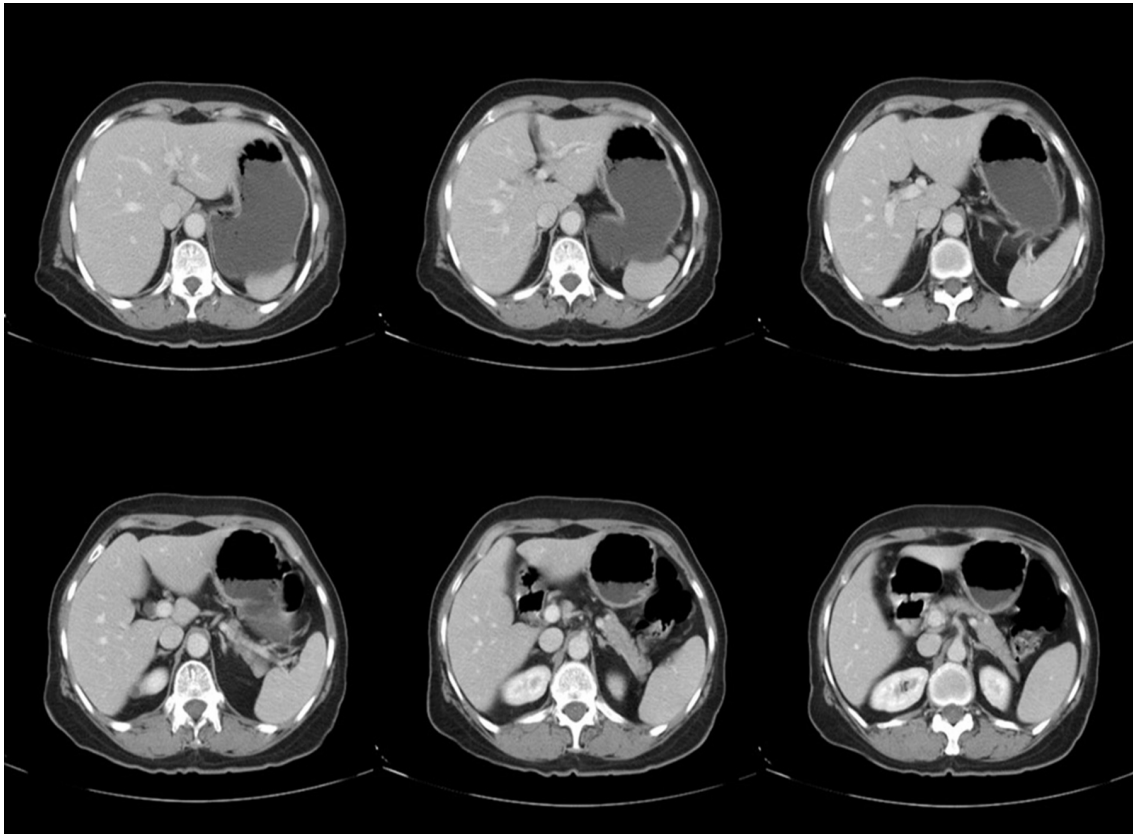


Fig.2: Transaxial abdominal tomograms show that gallbladder is not visualized in the expected anatomical region.

followed by nausea and vomiting, fatty food intolerance, dyspepsia, and jaundice.¹⁶ The jaundice is usually due to associated biliary stone with or without ascending cholangitis.^{10,17} The other symptoms may be related to other biliary pathologies such as biliary dyskinesia, or non-biliary causes such as esophagitis or duodenitis.¹⁸

The initial diagnostic method of choice for the evaluation of biliary system abnormalities is US. However, the conditions of examination and experience of examiners do not always permit an accurate appreciation. CT and magnetic resonance cholangiography are also other useful modalities to depict gall bladder anatomically.² Hepatobiliary scintigraphy is a functional imaging modality, which tracks the pathway of biliary clearance through the biliary tree.¹⁹ Despite the fact that hepatobiliary scintigraphy is a useful method for investigation of hepatobiliary abnormalities such as ectopic gall bladder, it is suggested that it has shortcomings in the diagnosis of GA.²⁰ In fact, in case of GA, hepatobiliary scintigraphy will not outline the gallbladder, and raise the suspicion of cholecystitis, which can produce the same pattern.²¹⁻²³ Chopra et al. reported a case of GA with non-identifiable gallbladder in sonography. In this case nonvisualization

of gallbladder in hepatobiliary scintigraphy considered as positive scan for acute cholecystitis and patient was referred for surgery.¹² However, it seems that for patients who had suspicious findings in favor of GA in other imaging modalities and when acute cholecystitis is unlikely upon the clinical presentation, hepatobiliary scintigraphy can play a confirmatory role in this regard especially by exclusion of ectopic gall bladder and thus will avoid unnecessary surgery for definite diagnosis. Malde et al. also have suggested to repeat imaging if gallbladder is not identified during radiological investigation and surgery reserved only for those who have confirmed evidence of gallbladder in preoperative imaging.² To prove the diagnosis of GA when gall bladder is not detected in the expected anatomical site during surgery, the surgeon must examine the most common sites for ectopic gallbladder including intrahepatic and retrohepatic regions, within the leaves of the lesser omentum or falciform ligament, or retroduodenal, retropancreatic, and retroperitoneal sites.²⁴ During laparoscopy, it will represent a difficulty for the surgeon while the biliary or portal structures can easily be injured during searches for a gallbladder that does not exist.^{25,26} In our patient we

could avoid such potential injuries on the strength of the evidence found in US, biliary scintigraphy, and CT of the abdomen.

For unknown reasons most symptomatic patients are reported to be symptom-free after the operation.⁷ For most of those patients who remain symptomatic, conservative treatment with smooth muscle relaxants can effectively manage their symptoms and sphincteroplasty may be reserved for non-responders.²¹

CONCLUSION

GA should be kept in mind whenever the gallbladder is not detected during imaging in patients with biliary-related pain. The correct diagnosis of GA, preoperatively, can help to avoid needless surgical exploration and its consequent risks. Our case demonstrates that in selected scenarios, hepatobiliary scintigraphy can guide physicians for evaluation of GA prior to the unnecessary surgery.

ETHICAL APPROVAL

There is nothing to be declared.

CONFLICT OF INTEREST

The authors declare no conflict of interest related to this work.

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