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Anomalous radiological and clinical demonstration of gallbladder duplication in an asymptomatic pregnant female

Gallbladder duplication is an uncommon congenital anatomical deviation encompassing a number of variants. The morphological difference occurs during budding of the hepatic diverticulum. We report the case of an asymptomatic pregnant woman who was incidentally found to have a large subhepatic cyst on ultrasound. The case highlights the difficulty in diagnosing large gallbladder duplication and the different radiological imaging available to surgeons to aid in mapping such anatomical variants. We propose that this case holds valuable lessons for both radiologists and surgeons when considering the differential diagnosis and management of large hepatobiliary cysts.

KEY WORDS: Asymptomatic, Choledochal cysts, Duplication, Gallbladder, Pregnancy

Introduction

Gallbladder duplication is a rare congenital malformation with a predictable autopsy frequency of 0.02%¹. Historically, the first reports come from a sacrificial victim of Emperor Augustus in 31 BC².

The main symptoms of this anatomical malformation are vomiting, jaundice and abdominal pain. The symptoms mimic biliary colic, acute cholecystitis, and cholelithiasis and occur at the same rate as the normal variant³. Because there is no known risk of subsequent disease there is no indication for removal in completely asymptomatic patients^{4,5}.

We report, what is to our knowledge, a unique case of a pregnant woman who was incidentally found to have a large subhepatic cyst on ultrasound. Only after surgery and histology was it confirmed as duplication of the gallbladder.

Case Report

A 39-year-old postpartum woman (G1, P1) was referred to our tertiary centre for further investigations and specialist planning in surgical intervention after a cyst was originally diagnosed incidentally on ultrasound while the patient was being investigated for pre-eclampsia during her pregnancy. Previous to this she had been completely asymptomatic and continued her pregnancy to term, having an uncomplicated delivery. She had no past medical history and was on no regular medication. Pre-operative bloods were all within normal range.

The original ultrasound was suspicious of biliary cystadenoma and a subsequent MRI with pre and dynamic post Gadolinium was performed. This confirmed an 8.7cm cystic lesion which lay between the head of the pancreas and the liver Hilum (Fig. 1). The radiological report described a few smaller cysts surrounding the cyst inferolaterally, one of which may have represented the gallbladder and the others may have been intrahepatic. Intrahepatic duct dilatation was described with biliary compression at the inferior aspect of the cyst. Radiological findings supported a diagnosis of biliary cystadenoma arising from the inferior surface of the liver and causing extrinsic compression of the bile duct. The possibility of the lesion being a mixed intrahepatic and

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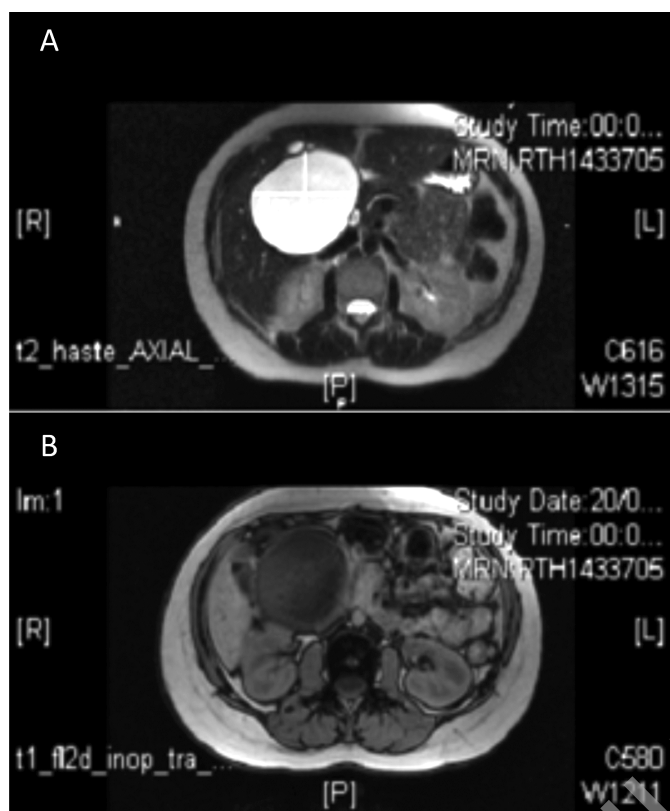


Fig. 1: MRI findings. T2 image (A) and T1 image (B) demonstrating a large cystic lesion near the hilum of the liver. The exact origin of this is not clear. It was impossible to identify two gallbladders.

extrahepatic choledochal cyst was also considered. A diagnostic aspirate of cyst fluid was performed, and this showed benign cytology but a high fluid CA 19-9. Surgery was planned due to the pre-malignant nature of both of the suspected differential diagnoses⁶ In general, the risk of cancer is 10-15% and increases with age⁷. Adenocarcinoma accounts for between 73%-84%, anaplastic carcinoma for approximately 10% and undifferentiat-



Fig. 2: Intraoperative cholangiogram showing intact biliary tree.

ed cancer and squamous cell carcinoma for 5% each^{8,9}, recommendations are to therefore remove any suspected choledochal cysts.

A right subcostal incision was extended to the left following discovery of a large subhepatic cyst compressing the liver and bile duct which was embedded in the head of the pancreas inferiorly. Extensive dissection and mobilization allowed clear visualization of the anatomy. A transcystic cholangiogram demonstrated a normal common bile duct but the contrast filled the cyst rather than flow into the duodenum (Fig. 2).

The cystic duct was transfixated and oversewn and the specimen completely excised. A second structure was found and was thought to be a 2nd duct duplication. This too was transfixated and oversewn (Fig. 3).

On gross examination, it was unclear as to whether this was a duplicated gallbladder or a mixed choledochal cyst. The specimen was sent for histology and the patient had an uneventful post-operative period and was discharged home successfully.

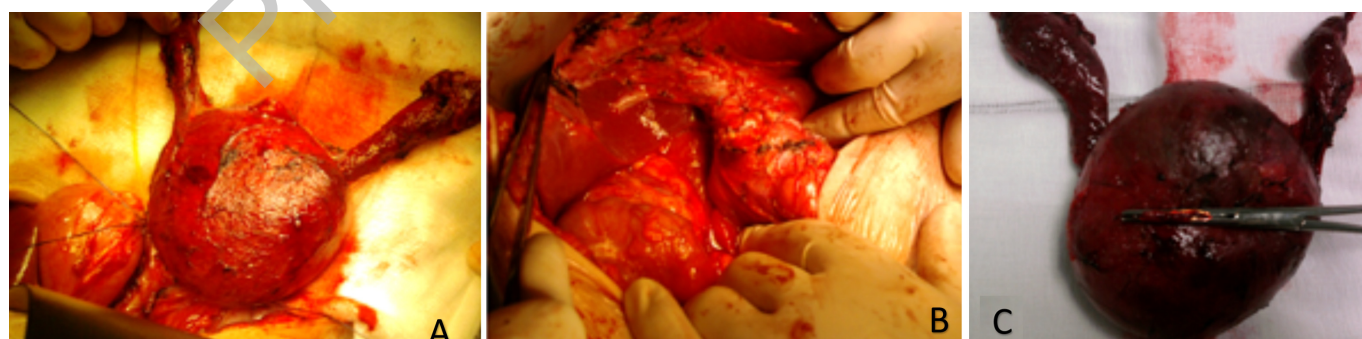


Fig. 3: Operative photographs (A, B, C).

Macroscopic evidence portrayed true duplication of the gallbladder, the larger of which (80mm x 45mm) appeared to communicate with a large cystic structure (75mm) attached to the cystic duct. The smaller (60mm x 35mm) gallbladder did not communicate with the cyst. Microscopically it was established that the cystic structure was in fact lined by biliary epithelium with underlying smooth muscle and therefore a cystically-dilated cystic duct. There was no difference in the gallbladders, but ducts of Luschka were seen in the larger gallbladder.

Discussion

Gallbladder duplication is thought to occur in 1/4000 births¹ and arises when the caudal bud of the hepatic diverticulum divides during weeks five and six of gestation¹⁰⁻¹². Gallbladder duplication is classified by Boyden's classification and subdivides them into two main categories:

– Vesica fellea divisa (bilobed or bifid gallbladder with common neck);

– Vesica fellea duplex (or true duplication with two cystic ducts). This is additionally subdivided into Y (two cystic ducts uniting) shaped and H (two cystic ducts entering the common bile duct at different locations) shaped types. The locations of these entry points can be intrahepatic, subhepatic, within the gastrohepatic ligament or in the normal position^{13,14}. This form of true duplication tends to be more common¹⁵. Our case therefore falls under the vesica fellea divisa type and is unusual in that the cystic duct was itself cystically dilated.

Imaging consists of ultrasound in the first instance, which may identify a cystic lesion but rarely is able to identify gallbladder duplication unless the viscera are located separately. Statistically one paper showed that ultrasound will duplicate gallbladders in about 50% of cases^{16,17}. There is a classification designed for ultrasound diagnosis but there are only a handful of instances in which this has been used¹⁸⁻²¹. In a case such as our own, ultrasound is unhelpful in identifying the cystic duct for Boyden's classification and generally rarer variants are diagnosed at surgery^{16,17}.

Helical CT can be useful but tends to be less sensitive than MRI and is thus used less often in such situations²².

With the advent of non-invasive imaging like MR cholangiopancreatography many instances identified on ultrasound can now be successfully assessed pre-operatively but seldom assists in the pathology²³. In our case MRI provided little help in the evaluation pre-operatively due to the larger nature of one gallbladder encompassing the smaller, and the cystic nature of the cystic duct.

ERCP is still considered the gold standard for biliary tree appraisal but has the disadvantages of any invasive procedure²⁴.

Intraoperative cholangiography is recommended to confirm the relationship with the biliary system and integrity of biliary tree, it can also be useful pre-operatively if the anatomy is still not clear⁴. In our case this showed an unaffected biliary tree, ruling out the possibility of a choledochal cyst.

To our knowledge there is only one other case reported of an incidental finding on ultrasound²⁵ but the dimensions of gallbladder duplication in this case were much smaller.

Our case represents a unique anatomical alteration because of the size of the duplication and the finding in an asymptomatic pregnant woman. In addition, the difficulty in radiological diagnosis provides lessons for surgeons and radiologists alike when investigating asymptomatic hepatic cysts.

Based on this we suggest that for incidentally found hepatic cysts, in which the origin is radiologically difficult to ascertain, the differential diagnosis should include a duplicated gallbladder and considerations about further investigations such as MR cholangiography and ERCP should be fully discussed in an attempt to avoid surgery.

Conclusion

This case provides excellent lessons for surgeons and radiologists when considering asymptomatic hepatic cysts and the necessary surgical and radiological planning required. It allows us to suggest that gallbladder duplication should be more commonly thought of in the differential diagnoses of hepatic cysts.

Riassunto

La duplicazione della colecisti è una deviazione anatomica comprensiva di notevoli varianti. La differenza embriologica si verifica nel corso della formazione del diverticolo epatico. Riportiamo il caso asintomatico, di una donna in stato interessante che mostrava incidentalmente all'esame ecografico una notevole cisti subepatica. La peculiarità del caso è dovuta alla difficoltà di arrivare a diagnosi chiara di duplicazione della colecisti, nonostante la possibilità di ricorrere a notevoli esami diagnostico-strumentali nella mappatura delle varianti anatomiche. Riteniamo che questo caso possa essere di estrema utilità per radiologi e chirurghi nel considerare la duplicazione della colecisti nella diagnosi differenziale di estesa cisti epatobiliare.

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