Case Report

Two cases of ectopic liver attached to the gallbladder wall

John Griniatsos, Ali A Riaz and Alberto M Isla
Upper GI and Laparoscopic Unit, Ealing Hospital, Southall, Middlesex, UK

Background

Ectopic liver tissue is occasionally found either attached to the gallbladder or elsewhere in the upper abdomen.

Case Outlines

A 49-year-old man and a 39-year-old woman were found to have a tongue of liver tissue attached to the serosa of the gallbladder (but separate from the liver) during laparoscopic cholecystectomy for gallstones. The ectopic liver was removed with the gallbladder and was histologically normal in each case.

Discussion

Several embryological hypotheses have been advanced to explain the development of ectopic liver. The anomaly is usually discovered incidentally at operation. Although the tissue is histologically normal, it can develop the same conditions as orthotopic liver.

Introduction

Ectopic liver is a rare developmental anomaly in which liver tissue is situated outside the liver and has no hepatic connection [1]. Ectopic liver tissue can occur in several different organs such as the diaphragm, hepatic ligaments, omentum, stomach, retroperitoneum and thorax [2,3], but the gallbladder is the commonest site of origin.

Case Reports

Case no. 1

A 49-year-old man with a six month history of biliary colic was found to have gallstones on ultrasound scan and underwent a routine laparoscopic cholecystectomy. During the procedure, a smooth tongue of brown tissue measuring 15 mm x 5 mm x 2 mm was seen to be attached to the serosa of the gallbladder by a thin mesentery (Figure 1). It was excised with the gallbladder. Histological examination showed liver tissue containing normal tissue elements, i.e. portal tracts with bile ductules and vessels and normal hepatocytes with no disturbance in architecture (Figure 2), suggesting normal drainage. He made an uneventful postoperative recovery and was discharged home 24 hours later.

Case no. 2

A 39-year-old woman with a four month history of multiple attacks of biliary colic and acute cholecystitis secondary to gallstones (on abdominal ultrasound), underwent a laparoscopic cholecystectomy as a day case. A fragment of liver 10 mm x 5 mm x 2 mm attached to the serosa of the gallbladder was noted intraoperatively and was excised with the gallbladder (Figure 3). The histology report revealed normal architecture with normal hepatocytes.

Figure 1. Case no. 1: Laparoscopic view of the gallbladder showing the ectopic liver attached to the serosal surface.
arteries, veins and portal tracts but no bile ducts (ductopaenia), although there was no evidence of cholestasis. She made an unremarkable postoperative recovery and was discharged home a few hours later.

**Review of the literature**

A MedLine database search was conducted using the following key words: ectopic liver (tissue), heterotopic liver (tissue), developmental anatomy, developmental anomaly(ies), gallbladder. References were limited to papers of which the abstract at least was in the English language. Because of the confusion in the classification and terminology of developmental hepatic anomalies, reports of “ectopic liver” were only accepted if they fulfilled the criteria published by Collan and co-authors [1] namely that the anomalous liver tissue had no connection with the liver. According to these criteria, 81 previous cases of ectopic liver have been published, 33 of them arising in the gallbladder (Table 1) [1,3–12].

**Discussion**

The incidence of ectopic liver tissue attached to the gallbladder has been reported as low, but is likely to increase with the new diagnostic methods. Eiserth [13] found only 3 cases in 5500 autopsies (0.05%), but in the laparoscopic era Watanabe and colleagues [3] have reported 3 cases in 1060 laparoscopies (0.28%).

Several theories have been proposed to explain the development of ectopic liver at different sites: development

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**Table 1. Documented cases of ectopic liver attached to gallbladder**

<table>
<thead>
<tr>
<th>Author</th>
<th>Year of publication</th>
<th>No of cases</th>
<th>Histology</th>
</tr>
</thead>
<tbody>
<tr>
<td>Cited by Tejada &amp; Danielson⁴</td>
<td>before 1975</td>
<td>17</td>
<td>Normal liver</td>
</tr>
<tr>
<td>Collan et al¹</td>
<td>1978</td>
<td>2</td>
<td>Fatty change</td>
</tr>
<tr>
<td>Torchio &amp; Maconi³</td>
<td>1978</td>
<td>1</td>
<td>Cirrhosis</td>
</tr>
<tr>
<td>Tamuar et al⁴</td>
<td>1985</td>
<td>1</td>
<td>Normal</td>
</tr>
<tr>
<td>Natori et al⁵</td>
<td>1986</td>
<td>1</td>
<td>Haemosiderosis</td>
</tr>
<tr>
<td>Tejada &amp; Danielson⁴</td>
<td>1989</td>
<td>1</td>
<td>Cirrhosis</td>
</tr>
<tr>
<td>Watanabe et al³</td>
<td>1989</td>
<td>3</td>
<td>Microhamartoma</td>
</tr>
<tr>
<td>Iaconi &amp; Masoni⁸</td>
<td>1990</td>
<td>2</td>
<td>Normal</td>
</tr>
<tr>
<td>Svane &amp; Knudtzon⁹</td>
<td>1991</td>
<td>2</td>
<td>Cholestasis</td>
</tr>
<tr>
<td>Hamdani &amp; Baron¹⁰</td>
<td>1994</td>
<td>1</td>
<td>Normal liver</td>
</tr>
<tr>
<td>Arakawa et al¹¹</td>
<td>1999</td>
<td>1</td>
<td>Alcoholic cirrhosis</td>
</tr>
<tr>
<td>Djuricic et al¹²</td>
<td>1999</td>
<td>1</td>
<td>Normal liver</td>
</tr>
<tr>
<td>Present article</td>
<td>2002</td>
<td>2</td>
<td>Normal liver</td>
</tr>
</tbody>
</table>
of an accessory lobe of the liver with atrophy or regression of the original connection to the main liver [3], migration or displacement of a portion of the cranial part (pars hepatica) of the liver bud to other sites [4], dorsal budding of hepatic tissue before the closing of the pleuroperitoneal canals [10], trapping of hepatocyte-destined mesenchyma in different areas [14] and entrapment of nests of cells in the region of the foregut following closure of the diaphragm or umbilical ring [15]. The close relationship between the cystic portions and the parenchymal cell cords of the primitive liver may explain why ectopic liver arises in the gallbladder wall [4,10], as in the two present cases.

Ectopic liver is sometimes associated with other congenital anomalies such as biliary atresia [15], agensis of the caudate lobe, omphalocele, bile duct cyst [16] or cardiac and conotruncal anomalies [17], but not when the heterotopic tissue is in the gallbladder. Although the ectopic tissue is usually attached to the serosa of the gallbladder or lies within its wall, it can also occur in the gallbladder lumen [7]. As in the present cases, it may have its own mesentery [4] and its histological architecture resembles normal liver with regular lobules, central veins and portal areas [9,10]. Depending on its location, ectopic liver tissue can drain into the biliary tract [6], into another organ [18] or have no drainage system [19]. Drainage into the gallbladder seems likely in the first of our cases because of the absence of bile duct dilatation or cholestasis, but in the second case the ectopic liver tissue was without a drainage system.

Intra-, retro- and extra-peritoneal ectopic liver tissue usually remains asymptomatic, although obstructions of the oesophagus [18], pylorus [20] and portal vein [21] have all been reported. With ectopic liver attached to gallbladder some authors [8] described right sided abdominal discomfort, but it is unclear if this is related to ectopic liver itself or to co-existing gallstone disease.

The natural course of ectopic liver tissue is unpredictable. The anomaly is relatively common in the perinatal period but disappears during postnatal remodelling [22]. Hepatocytes in an ectopic liver behave like normal hepatocytes and show the same pathological findings as those of the main liver [11]. Thus ectopic liver in the gallbladder can undergo fatty change [13], haemósiderosis [4], cholestasis [9] or cirrhosis [3]. Ectopic liver tissue is also at increased risk of carcinogenesis. Of 48 cases (excluding those with a gallbladder location), 22 developed hepatocellular carcinoma [11] whereas only one of 33 cases of ectopic liver attached to the gallbladder developed cancer (p < 0.001, Student’s t-test). A possible explanation for this difference is that ectopic liver attached to gallbladder is an anomaly occurring later during late embryogenesis and is therefore well differentiated.

Preoperative diagnosis of this anomaly is difficult because of lack of symptoms and difficulties in imaging. It is usually an incidental finding during a laparoscopy, laparotomy or autopsy performed for unrelated reasons. The diagnosis should be considered when a soft-tissue mass is seen to arise from the gallbladder wall on abdominal ultrasound or CT scan [10]. The combination of intravenous injection of indocyanine green with laparoscopy [3] may increase the diagnostic accuracy of this type of developmental anomaly.

There are too few case reports for definitive treatment to be clear-cut. It would be sensible to resect the ectopic tissue if encountered during cholecystectomy for gallstones, but to leave it alone if seen incidentally during other procedures.

References

4 Tejada E, Danielson C. Ectopic or heterotopic liver (choristoma) associated with the gallbladder. Arch Pathol Lab Med 1989;113:950–2.
10 Hamdani SD, Baron RL. Ectopic liver simulating a mass in the gallbladder wall: Imaging findings. AJR 1994;162: 647–8.

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