Gall bladder agenesis, pancreas divisum and undescended testes: a rare association

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Gall bladder agenesis is a rare congenital biliary anomaly that may be associated with other biliary and extra-biliary congenital anomalies. We report the association of gall bladder agenesis with pancreas divisum and undescended testes. [Indian J Gastroenterol 2001;20:71-72].

Key words: ERCP, gall bladder anomaly, ultrasonography

Gall bladder agenesis is a rare congenital anomaly occurring in 13-65 per 100,000 population.1 It is usually asymptomatic. Most of the symptomatic cases present with nonspecific biliary symptoms.1,2,3 Non-visualisation of gall bladder on ultrasonography may raise the suspicion, but often the diagnosis is made at laparotomy. We report a rare association of gall bladder agenesis with pancreas divisum and undescended testes.

A 45-year-old man complaining of nonspecific epigastric pain over the last 3 years was investigated by his family physician. Esophago-gastro-duodenoscopy was normal and abdominal ultrasonography was reported as showing chronic cholecystitis with an atrophic, contracted gall bladder. He was taken up for cholecystectomy but the surgeon could not locate the gall bladder. Intraoperative cholangiography was not performed due to lack of the facility at the center. He was subsequently referred to us for evaluation.

He had undescended testes on the left side; the rest of the physical examination was normal. Liver biochemistry was within normal limits. Abdominal ultrasonography corroborated the absence of gall bladder. To confirm gall bladder agenesis, we performed endoscopic retrograde cholangiopancreatography. The cholangiogram revealed normal intra- and extra-hepatic bile ductual system while the cystic duct and gall bladder were absent (Fig). There was an accessory hepatic duct, almost of the caliber of normal hepatic ducts, draining part of the right lobe of the liver into the common hepatic duct. The main pancreatic duct could not be opacified through the major papilla. Exsufflation of the accessory papilla revealed a dominant dorsal duct (pancreas divisum). Other members of his family were investigated and were found to be normal.

Due to the widespread use of abdominal ultrasonography, the chances of recognizing gall bladder agenesis preoperatively are increasing. It is important to be aware of this entity to avoid false positive diagnosis of chronic cholecystitis.3 Whenever suspected, a diagnostic endoscopic retrograde cholangiography would confirm the condition.2 Magnetic resonance cholangiography is a better, noninvasive alternative. Congenital anomalies of the gall bladder are rare and can be accompanied by other malformations of the biliary or vascular tree.

Gall bladder agenesis may be recognized in three situations: a) in the neonatal phase, often in association with other congenital malformations, b) in asymptomatic adults, usually detected incidentally during autopsy, and c) in symptomatic adults, in whom the diagnosis is often made at surgery.2 Richards et al4 found that biliary colic was the commonest presentation (54%), followed by dyspepsia (34%) and jaundice (27%). About two-thirds of patients with jaundice had associated choledocholithiasis. Positive familial association has been reported in some cases; a few others had associated congenital anomalies of the biliary as well as extra-biliary systems.1,2,5

In the reported case, gall bladder agenesis was associated with complete pancreas divisum; such an association has hitherto not been reported.

The suspected embryological basis for gall bladder agenesis is failure to develop or vacuolize the gall bladder bud during early intrauterine life.1 Both the gall bladder bud and the ventral and dorsal pancreatic anlage appear in embryos of about 4 mm in the 4th-5th week of intrauterine life.6 The ventral anlage is connected to the common bile duct during its origin while the dorsal anlage is in direct connection with the duodenum. The two anlagen come together due to the uneven growth of the duodenum and fuse by the 7th week of gestation. Failure of fusion results in pancreas divisum.6 Since both the gall bladder bud and the pancreatic buds develop during the same time in intrauterine development, whatever embryological event leads to the agenesis of one may also affect the other. Agenesis of the ventral pancreatic anlage, which is embryologically closely related to the gall bladder bud and the common bile duct, can explain the presence of only the accessory pancreatic duct in the reported case. The patient described in this report also had undescended testes, which
is difficult to correlate with the biliary-pancreatic anomalies. Since undescended testes is a more common anomaly and developmentally not related, it may be coincidental.

References

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Laparoscopic resection of liver metastasis using a harmonic scalpel
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We report successful laparoscopic resection of a solitary liver metastasis from a colorectal carcinoma in an obese man, using a harmonic scalpel. [Indian J Gastroenterol 2001; 20: 72-73]

Key words: Colorectal carcinoma, laparoscopy

Laparoscopic approach is fast taking over procedures that were deemed to be suitable only for open surgery. We report successful laparoscopic resection of a solitary liver metastasis, highlighting the advantage of this approach in selected patients.

This 54-year-old man underwent anterior resection and loop ileostomy for Duke’s C adenocarcinoma of the rectum. During follow up, CEA levels increased from 5 to 9.9 μg/L. CT scan revealed a calcified lesion in segment V of the liver adjacent to the gall bladder bed. CEA continued to rise to 22 μg/L. 18-fluorodeoxyglucose positron emission tomogram (FDG PET scan) showed focal uptake adjacent to the gall bladder. CT angiography (Fig) showed a solitary lesion. The patient weighed 120 kg and, due to his size, could not fit in the MRI scanner. Chest CT and bone scan were normal.

At laparoscopy, 12-mm ports were inserted above the umbilicus, below the xiphisternum and subcostally in the right midclavicular line. A 5-mm port was inserted in the anterior axillary line 8 cm from the right costal margin. Access was difficult because of adhesions to midline laparotomy and ileostomy scars. No other liver lesions were seen. The cystic duct and artery were divided between clips and the gall bladder was partially dissected off its bed. This was used as a handle for traction on the tumor to avoid direct handling. A 2-cm margin was marked around the tumor and parenchymal division was carried out with a harmonic scalpel (Auto Suture, Ascot, UK) using endoscopic Hemostasis was achieved with harmonic scalpel coagulation, clips and compression of the raw surface. The resected specimen measuring 9.5 cm x 5.0 cm x 5.0 cm, was placed in a plastic bag and delivered intact by enlarging the epigastric port to 5 cm. A suction drain was placed at the resection site. The procedure had to be interrupted once due to carbon dioxide retention. The operative time was 390 minutes and 2 units of blood were transfused.

The postoperative recovery was uneventful. The patient was discharged on the 5th post-operative day, requiring only occasional disconnexion for pain control. CEA level decreased to 0.3 μg/L. 15 days after the procedure. Pathologic examination showed a 4 cm x 3 cm x 3 cm moderately differentiated adenocarcinoma with the closest margin being 2 cm macroscopically, and 5 mm considering microscopic vascular invasion. He is well six months after the operation.

Laparoscopic liver resection has an advantage over open surgery because of a small incision, reduced wound pain and early discharge from hospital.1 Obese patients specially benefit from this approach as it lowers the risk of chest sepsis and deep vein thrombosis that result from prolonged immobilization following open surgery.2

Superficial location of the metastasis in our patient made it ideal for laparoscopic wedge resection. This