Cholecystitis in an intrahepatic gallbladder

A case report

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Summary

A case of cholecystitis in an intrahepatic gallbladder with concurrent choledocholithiasis is reported. The patient initially presented with pyrexia of unknown origin and subsequently with suppurative cholangitis; the diagnosis was resolved pre-operatively using contemporary techniques of gallbladder delineation. Simple drainage of the gallbladder with choledocholithotomy proved effective.

Case report

A 49-year-old man presented with pyrexia of unknown origin. A year before he had suffered a similar episode which had lasted a week and was accompanied by transient elevation of the ESR and alkaline phosphatase level. His present symptoms, of a month's duration, were malaise and weight loss with a week's history of daily fever associated with sweating and headache. There was no abdominal pain, nausea, vomiting, jaundice or change in colour of his excreta. He felt well between attacks. He had previously been treated for malaria and brucellosis in Central Africa.

He was distressed and had a temperature of 39°C. There was no jaundice. The abdomen was soft and not tender and the liver and gallbladder were not enlarged. There were no other abnormal findings. The ESR was 100 mm/h (Westergren), the white cell count 9600/μl with toxic granulation, the total bilirubin level 14 mmol/l (normal 2-17 mmol/l) with a conjugated fraction of 5 mmol/l, the alkaline phosphatase level 121 U (normal 30-85 U), and the γ-gluteryltransferase level 89 U (normal 0-50 U). Chest and abdominal radiographs, urinalysis and extensive screening for infectious diseases were negative.

On the 8th day in hospital he experienced severe, stabbing right upper quadrant pain, followed the next day by clinical and biochemical evidence of jaundice. Ultrasonography showed a dilated common bile duct and hypo-echoic areas in the right lobe of the liver, which were interpreted as metastases. Endoscopic retrograde cholangiography revealed a dilated common bile duct with two calculi at the lower end, but the gallbladder was not demonstrated (Fig. 1). A static 99mTc tin colloid liver scan revealed a large focus of reduced uptake within the right lobe of the liver (Fig. 2), and dynamic scanning sug-
gusted a cyst or metastatic deposit. Computed tomography (CT) confirmed a large cystic area within the liver confluent with the biliary tree and in keeping with an intrahepatic gallbladder (Fig. 3); in addition a dilated common bile duct with calculi was demonstrated (Fig. 4).

At laparotomy the presence of an intrahepatic gallbladder was confirmed; it appeared as a cystic swelling covered by liver tissue on the lower surface of the right lobe of the liver. This yielded pus on aspiration, and was then approached through the inferior surface of the liver; multiple gallstones were removed and the cavity was drained. No cystic duct was located. Two gallstones were removed from the common bile duct and this was drained with a T-tube which was removed at 15 days after T-tube insertion. A. SA. Computed tomography of the abdomen confirmed that the gallbladder — not seen at operation — was within the liver substance. In recent years there have been reports of pre-operative delineation of the intrahepatic gallbladder by newer techniques, including 99m Tc sulphur colloid, 99m Tc diethyl- acetylildil- iminodiacetate (HIDA) scans, ultrasonography and CT. 11 There has been no report of an association with other congenital abnormalities. While diverticula of the gallbladder and mesenteric abnormalities clearly predispose to cholecystolithiasis and infection, 4 it has also been suggested that the intrahepatic gallbladder predisposes to disease; 7 McNamee estimated that 60% of adult patients with this anomaly had gallstones. This, however, cannot constitute proof of predisposition, as patients present with cholecystitis rather than the anomaly.

Initial diagnosis in our patient, who presented with pyrexia of unknown origin and a misleading history of tropical infection, presented a problem, but once the jaundice became apparent, definitive investigation revealed the lesion fully. In this regard CT proved the most rewarding investigation. At operation, transhepatic removal of the gallstones and simple drainage of the gallbladder bed have proved effective, with no recurrence of symptoms at 10 months. We were unable to find adequate guidance on the technical aspects of the operation in the usual texts, but noted 2 reports of gallbladder removal accompanied by profuse haemorrhage, 12 and the suggestion by some that cholecystotomy is the treatment of choice. 3,13 Should our patient’s disease recur, we would consider elective cholecystectomy.

**Discussion**

While the intrahepatic position of the gallbladder is normal in a variety of animals, including the shark, many reptiles and most of the marsupials, 2 this location is rare in man. The gallbladder in man develops from the small caudal portion (pars cystica) of the hepatic diverticulum and is an intrahepatic structure during the 2nd month of development, thereafter emerging as an extrahepatic organ. 2 The intrahepatic gallbladder may therefore be regarded as an example of positional arrest during development. 1

There are various comprehensive classifications of gallbladder anomalies, 3, 4 and all authors describe two types of intrahepatic variant: partial or complete. Deve 7 first described the condition in 1903, finding 12 partial or complete examples in 130 infant autopsies. By 1936 McNamee 7 had found a further 15 cases, 3 at autopsy. Of the remaining 12 patients found to have this anomaly at laparotomy, 9 are known to have undergone surgery for cholecystitis. In 1 of McNamee’s own patients postoperative cholecystography confirmed that the gallbladder — not seen at operation — was within the liver substance. In recent years there have been reports of pre-operative delineation of the intrahepatic gallbladder by newer techniques, including 99m Tc sulphur colloid, HIDA scans, ultrasonography and CT. 11 There has been no report of an association with other congenital abnormalities. While diverticula of the gallbladder and mesenteric abnormalities clearly predispose to cholecystolithiasis and infection, 4 it has also been suggested that the intrahepatic gallbladder predisposes to disease; 7 McNamee estimated that 60% of adult patients with this anomaly had gallstones. This, however, cannot constitute proof of predisposition, as patients present with cholecystitis rather than the anomaly.

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**REFERENCES**