von Meyenburg complexes mimicking liver metastases at laparoscopic cholecystectomy

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DESCRIPTION

A 61-year-old male patient had four hospital admissions with *Escherichia coli* sepsis fever and abnormal liver function tests over an 8-year period of time (maximum aspartate aminotransferase 82 IU/L, maximum alkaline phosphatase 231 IU/L and bilirubin normal). On each occasion he was successfully treated with antibiotics. He was extensively investigated to determine the source of sepsis. Several CT scans of thorax, abdomen and pelvis were performed and were reported as showing multiple small liver cysts with no other abnormalities. An ultrasound scan showed that his gallbladder and bile ducts were normal and gallstones were not present. Hepatitis screen, echocardiography, gastroscopy and colonoscopy were normal and amoebic serology was negative. Liver MRI was unsuccessful due to claustrophobia. Endoscopic ultrasound scan showed foci of echogenic material in a thin walled gallbladder with a normal common bile duct. It was thought that he was having episodes of biliary sepsis and was advised to undergo laparoscopic cholecystectomy. At laparoscopy he was found to have multiple white liver lesions (figure 1). It was thought that these may be liver metastases. Laparoscopic cholecystectomy was performed and a liver lesion was biopsied. Intra-operative frozen section histology was not performed as pathology facilities were not on-site. The subsequent histology report showed that the liver lesion was a bile duct hamartoma (figures 2–4). His gallbladder showed features of chronic cholecystitis and no gallstones were present. He remains well with no further episodes of sepsis 3 months later.

Multiple bile duct hamartomas also known as von Meyenburg complexes (VMCs) were first described in 1918 and occur in 5.6% of the population.^{1 2} They are multiple small white nodular cystic lesions varying from 1 mm to 15 mm in size consisting of duct-like structures lined by biliary epithelium and

Figure 2 Histology of bile duct hamartoma, H&E x25 (low power magnification) with liver parenchyma containing a relatively well delineated lesion. The glandular lumina contain bile.

Figure 3 Histology of bile duct hamartoma, H&E x100 magnification showing small-to-medium sized, irregularly shaped bile ducts lined by bland cuboidal epithelium with intervening collagenous stroma.

surrounded by fibrous stroma. They result from ductal plate malformations of the smallest intrahepatic bile ducts due to disordered embryonic involution.³ They can clinically be confused with liver metastases, microabscesses and liver cysts, but have a characteristic appearance on MRI.⁴ Laparoscopic

Figure 4 Histology of bile duct hamartoma, H&E x400 (high power magnification) with dilated irregularly shaped glands lined by a bland cuboidal epithelium with no atypia or mitoses.

Figure 1 Operative photographs showing multiple white liver nodules during laparoscopic cholecystectomy.

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Images in...

biopsy of a nodule with intra-operative frozen section histology will give a rapid diagnosis. They are usually asymptomatic and found incidentally during laparoscopic surgery however there are several case reports of patients with VMCs presenting with fever and abnormal liver function tests, presumably where there has been secondary infection of the VMCs due to unrelated bacteraemia.^{5 6} Rarely malignant transformation can occur and long-term follow-up with MRI has been advocated.⁷ Time will tell whether our patient's episodes of sepsis were the result of biliary microcalculi or a rare presentation of recurrently infected VMCs.

Learning points

- ▶ Multiple bile duct hamartomas (von Meyenburg complexes) are caused by disordered embryonic involution of the smallest intrahepatic bile ducts. They present as multiple small white cystic nodules throughout the liver that can be misdiagnosed as metastases at laparoscopic surgery.
- ▶ When unusual liver lesions are found at laparoscopy biopsy with intra-operative frozen section histology is usually optimal management.

Contributors SP is the consultant surgeon responsible for this patient's care. He performed the laparoscopic cholecystectomy and found the unusual multiple white lesions throughout this patient's liver. His operative assistants were AY (registrar) and NK (CT1). All three surgeons (SP, AY and NK) thought that the liver lesions could be multiple metastases from an undiagnosed malignancy. They discussed

the case and decided to proceed with laparoscopic cholecystectomy to prevent future episodes of biliary sepsis and to also remove one of the white liver lesions for urgent histological analysis. Suboda Weerasinghe is the consultant pathologist who performed the histological analysis and diagnosed the liver lesions as benign bile duct hamartomas. All three surgeons thought that this case merited publication because of the rarity of the finding at laparoscopic surgery and because it could be misdiagnosed as malignancy. All three surgeons contributed to the literature search and found no previous publications of multiple bile duct hamartomas being reported at laparoscopic surgery. Suboda Weerasinghe agreed that this unusual case should be put forward for publication. All four authors (SP, AY, NK and SW) contributed to the writing of this manuscript.

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