Benign Finding or Malignancy: von Meyenburg Complexes Found during Laparoscopic Cholecystectomy

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ABSTRACT

Von Meyenburg complexes are benign bile duct hamartomas that arise as cystic nodules of the liver. Von Meyenburg complexes are often asymptomatic and thus typically discovered incidentally on imaging or autopsy. They can also be encountered at the time of surgery where they often appear as scattered white liver lesions concerning for malignancy. Here, we present a case in which white hepatic nodules were found incidentally during laparoscopic cholecystectomy in a 36year-old female. Pathologic analysis confirmed the diagnosis of von Meyenburg complexes. The operating surgeon proceeded with laparoscopic cholecystectomy without complication. We report this case to encourage awareness of this benign entity. The finding of scattered hepatic lesions found intra-operatively can create concern for metastatic neoplastic processes. An awareness of von Meyenburg complexes and their gross appearance can better guide surgeons' intraoperative decision-making when encountering these characteristic hepatic lesions.

Key Words: Case Reports, Bile Duct, Hamartoma, Incidental Finding, Laparoscopic Cholecystectomy.

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INTRODUCTION

Von Meyenburg complexes (VMCs), also known as bile duct hamartomas, are benign cystic nodules of the liver.¹ First described by von Meyenburg in 1918, they result from malformations of the embryonic ductal plate and histologically appear as dilated bile ducts embedded in a fibrous stroma.²⁻⁴ Autopsy studies demonstrate a prevalence of 5.6% in adults and 00.9% in children.⁵ These nodules are typically asymptomatic and as such are typically discovered incidentally on imaging or at the time of surgery. Biopsy remains the most reliable means for diagnosis, although recent studies report that the diagnosis can be made on imaging findings alone.^{1,6,7} Here, we present a case report of a patient with no obvious radiographic evidence of VMCs who was then found to have innumerable scattered white hepatic nodules during laparoscopic cholecystectomy.

CASE PRESENTATION

The patient was a 36-year-old female with a past medical history of gastroesophageal reflux disorder, obesity, hypertension, and polycystic ovary syndrome who presented to general surgery clinic with complaints of one month of postprandial right upper quadrant pain. She denied complaints of jaundice or acholic stools. Her physical examination was unremarkable. Pre-operative computed tomography (CT) of the abdomen and pelvis with intravenous contrast demonstrated a large gallbladder calculous with associated gallbladder wall thickening. In regard to the liver parenchyma, a single pericentimeter with low attenuation focus in segment four was identified and thought to reflect focal fat infiltration. An ultrasound was ordered but never completed by the patient. There were no abnormalities in basic lab testing, including hepatic function panel and complete blood count. The patient was diagnosed with symptomatic cholelithiasis was offered a laparoscopic cholecystectomy.

Following laparoscopic entry and insufflation, the patient was noted to have diffuse sub-centimeter flat white lesions scattered throughout the liver (**Figure 1 A, B**). Given concern for neoplastic processes, three excisional biopsies of the liver nodules were performed and sent for permanent pathology as well as microbiology. At the time

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Informed consent: Dr. Darryl T. Hiyama declares that written informed consent was obtained from the patient/s for publication of this study/report and any accompanying images.

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Figure 1. Diffuse sub-centimeter flat white lesions scattered throughout the liver. (A) left lobe. (B) right lobe, including gallbladder.

of surgery, frozen section biopsies were not obtained opting instead for permanent fixation which we considered more accurate. The decision was made to proceed with the laparoscopic cholecystectomy as planned. The operation was uncomplicated. Pathology ultimately demonstrated bile duct hamartomas (von Meyenburg complexes) with no evidence of malignancy. The gallbladder pathology was consistent with chronic cholecystitis with cholelithiasis. The patient recovered well after surgery and required no further follow-up.

DISCUSSION

Bile duct hamartomas (von Meyenburg complexes) were first described by von Meyenburg in 1918.² VMCs arise from benign ductal plate malformations and have an estimated prevalence of 0.35% to 5.6% in adults^{5,8} and 0.9% in children.⁵ Histologically, they appear as dilated bile ducts embedded in a fibrous stroma.4 VMCs appear as multiple cystic multiple nodules that generally range from 5-10 mm. On ultrasound, they can appear as multiple small comet-tail echos. On CT, they can appear as multiple hypointense nonenhancing lesions. On magnetic resonance (MR) imaging or MR cholangiopancreatography, they appear as cystic structures associated with normal extra- and intrahepatic bile ducts.^{1,9} Although biopsy can offer a definitive diagnosis, some studies suggest MR imaging may be sufficient to diagnose VMCs without biopsy.1,6,7

VMCs can also be encountered intraoperatively, with most recent published literature documenting these incidental findings during open abdominal operations.¹⁰ These lesions have also been identified during laparoscopic procedures.^{11–15} They typically appear as numerous white scattered lesions, which can be disconcerting to the operating surgeon given the similar appearance of diffuse liver metastases. It is for this reason that we believe it to be important that surgeons are aware of this rare diagnosis and this potential red herring during surgery. If

encountered, we suggest performing 2–3 liver biopsies in different locations for pathological analysis. We also recommend a quick survey of all four quadrants to assess for metastasis. Unless there is concern for undiagnosed malignancy, it is reasonable to proceed with the intended surgery. After the procedure, the surgeon should discuss the findings with the patient and the need to biopsy. They should feel comfortably explaining one possibility is VMCs which are a benign finding. Once the diagnosis of VMCs is made, no further management is recommended, although there are rare instances of malignant transformation as well as cholangitis arising from these bile duct hamartomas.^{16–18}

CONCLUSION

This case report presents the benign clinical finding of VMCs incidentally found during a laparoscopic cholecystectomy. While the differential for hepatic lesions can be broad, the characteristic appearance of VMCs as scattered, white cystic nodules on the liver can help guide diagnosis. Biopsy with pathologic analysis can provide a definitive diagnosis and rule out more serious conditions such as metastasis or infection. For surgeons who identify similar lesions intraoperatively, we recommend biopsy with careful discussion with the patient afterwards. Despite VMCs rarity, surgeons should be aware of VMCs to guide proper intraoperative and perioperative management.

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