

Suitability Of Robotic-assisted Surgery For Hepatic Peribiliary Cysts: A Case Report

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Title: Suitability Of Robotic-assisted Surgery For Hepatic Peribiliary Cysts: A Case Report

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Key Clinical Messages:

This study evaluates robotic-assisted surgery for hepatic peribiliary cysts, demonstrating its suitability through a successful minimally invasive subsegmentectomy on a symptomatic woman. Robotic surgery proved effective, offering less invasive options with reduced complications and recovery times, and may be preferable for managing potentially malignant hepatic cysts.

Summary:

Although hepatic peribiliary cysts are generally benign, surgery may be required if the cysts are growing or cause bile duct stenosis. However, few studies have reported the use of robotic surgery for removing hepatic peribiliary cysts. This case report describes a woman in her 50s with a cystic lesion in liver segment III that increased from 33 to 45 mm in 2 years. The patient was asymptomatic; however, robotic subsegmentectomy of segment III was performed owing to concerns about potential malignancy. The cyst was lined by a single layer of cylindrical epithelium without cytological atypia, and was pathologically diagnosed as a hepatic peribiliary cyst. Hepatic peribiliary cysts can be monitored if the cyst was diagnosed correctly and the patient has no symptoms. However, surgery is warranted if malignancy cannot be ruled out or the patient is symptomatic. Minimally invasive robotic surgery may be a good option for hepatic peribiliary cysts.

Keywords: Hepatic peribiliary cysts, Robotic surgery, Minimally invasive surgery, Hepatectomy, Case report

Background:

Hepatic peribiliary cysts are benign lesions that are thought to occur when inflammation causes obstruction of the secretory channels of the peribiliary glands around large bile ducts [1]. The age at the time of diagnosis of hepatic peribiliary cysts is in the 60s, and men are more frequently affected than women [1]. Although

hepatic peribiliary cysts are benign, surgery is sometimes indicated when the cyst obstructs the bile duct and causes jaundice or cholangitis [2, 3] or when the cyst increases in size and is difficult to distinguish from other malignant cystic tumors [4]. However, it is difficult to definitively diagnose hepatic peribiliary cysts as malignant, and minimally invasive surgery should be chosen when there is a possibility of benign disease. Furthermore, minimally invasive robotic surgery has recently become popular and has been shown to be safe for liver resection [5].

In the present case, we chose surgery as a treatment strategy for a growing hepatic peribiliary cyst that was difficult to distinguish from a tumor with malignant potential. The patient underwent robot-assisted hepatic subsegmentectomy with good outcomes.

Case Presentation:

A woman in her 50s was referred to our institution with a cystic lesion in her liver that had increased to 45 mm in diameter. The cyst initially measured 33 mm in diameter when it was first detected about 2 years ago and had been managed with the watchful waiting approach. She had no symptoms or relevant medical history, and was not taking any medications.

Investigations:

Laboratory data at the time of presentation to our institution showed almost no abnormalities, with an alkaline phosphatase concentration of 321 IU/L, gamma-glutamyl transpeptidase concentration of 48 IU/L, alanine aminotransferase concentration of 69 IU/L, and aspartate aminotransferase concentration of 35 IU/L. The bilirubin concentration was 0.7 IU/L, and she was negative for hepatitis serology. Ultrasound revealed a well-defined septated cystic mass in the left liver lobe with a maximum diameter of 45 mm (Fig. 1a–c). Subsequent abdominal computed tomography (CT), contrast-enhanced CT, and drip infusion cholecystocholangiography CT showed no contrast effect of the cyst, no calcification or enhanced nodules, and no bile duct traffic (Fig. 1d–f). Magnetic resonance imaging of the liver with cholangiopancreatography confirmed the presence of a multifocal cyst in liver section III (Fig. 1g, h). There was no traffic between the cyst and the bile duct. Bile cytology was negative.

Differential diagnosis:

A simple cyst was thought to be unlikely, and a provisional diagnosis of a complex cyst, mucinous cystic neoplasm, or hepatic peribiliary cysts was made. Given the patient’s age and symptomology, the growing size of the cyst, biliary compression, and risk of malignant transformation, we decided to perform surgical resection rather than to continue observation.

Treatment:

We performed a robotic subsegmentectomy of liver segment III via trocars placed in the lower abdomen (Fig. 2a). A resection map was preoperatively prepared (Fig. 2b). Surgery was performed using the intrahepatic glissonian approach in which the S3 Glisson’s sheath was dissected first (Fig. 2c). The liver was then dissected along the ischemic line (Fig. 2d, e). The operative time was 318 minutes and there was 63 ml of intraoperative blood loss. Macroscopically, there was a septated and loculated cavity within liver section III (Fig. 2f). The resection margin was 10 mm.

Microscopically, there were peribiliary glands located around the cyst. The cyst was lined by a single layer of cylindrical epithelium without cytological atypia (Fig. 3a, b). This epithelium was not underlined by an ovarian-like hypercellular stroma, and the cyst was pathologically diagnosed as a hepatic peribiliary cyst

(Fig. 3c–e).

Outcome and follow-up:

The postoperative course was uncomplicated, and the patient was discharged on postoperative day 17. She was recovering well by the time of the first follow-up at 2 weeks postoperatively, with further review planned for 6 months postoperatively.

Discussion:

Hepatic peribiliary cysts are reported to occur due to obstruction of the outflow tract of the peribiliary glands caused by alcohol, inflammation, or bile duct blood flow disorders [1]. A review of 1,000 autopsy reports showed that hepatic peribiliary cysts predominantly affect men and have a prevalence of 5% in normal liver [6]. As the peribiliary glands develop along the left branch of the intrahepatic bile duct, serous cysts are characteristically present in the left liver lobe near the porta hepatis and around the large bile ducts in a beaded pattern [6]. Therefore, to diagnose a hepatic peribiliary cyst, it is important to demonstrate the absence of bile duct traffic to the cysts by endoscopic retrograde cholangiopancreatography or drip infusion cholecystocholangiography CT, and to rule out malignancy by cytological diagnosis. Histopathologically, hepatic peribiliary cysts are lined by a single layer of columnar epithelium without atypia; however, there is a report of hepatobiliary cystadenoma and cystadenocarcinoma in the peribiliary gland being mistakenly followed up as hepatic peribiliary cysts

[7].

Hepatic peribiliary cysts are benign lesions that can be monitored without intervention after being accurately diagnosed. However, in Japan, surgery is performed in cases in which malignancy cannot be completely ruled out, in patients with jaundice, and in patients with repeated episodes of cholangitis [2]. If the cyst is located around a large bile duct near the porta hepatis and is impossible to distinguish from a malignant tumor, open hepatic resection is generally performed [2]; however, there is one reported case in which laparoscopic surgery was performed in Japan [2]. Open surgery is considered highly invasive, and minimally invasive surgery should be chosen if possible, especially for benign tumors. In the present case, it was difficult to rule out malignancy, so we decided to perform minimally invasive robotic surgery in accordance with recent reports of the effectiveness of minimally invasive robot-assisted liver resection [5].

In our case, the patient was female and the imaging findings were somewhat atypical because the cyst did not form a series of beads, making it difficult to distinguish from hepatic mucinous cystic neoplasm. In addition, the cyst had increased in size during 2 years of follow-up, so the possibility of malignancy could not be ruled out. Therefore, surgery was chosen. We decided to perform robotic-assisted hepatectomy because it is less invasive with less intraoperative bleeding and less postoperative pain than open surgery [5].

The perihilar accessory glands are also of interest as a source of intraductal papillary neoplasms of the bile duct and cholangiocarcinomas, and there are reports of cholangiocarcinoma thought to have originated from hepatic peribiliary cysts [6]. Therefore, as more cases are accumulated and a relationship between hepatic peribiliary cysts and cholangiocarcinoma is suggested, surgery may become the treatment of choice for hepatic peribiliary cysts.

In conclusion, the diagnosis rate of hepatic peribiliary cysts is expected to improve with advances in imaging technology. However, there are cases, such as the present case, in which surgery is warranted. In the present case, robot-assisted hepatic subsegmentectomy was performed safely and achieved good results. Our results suggest that robotic surgery may be useful for the treatment of hepatic peribiliary cysts for which surgery cannot be avoided.

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Figure:

Figure 1. Imaging of the hepatic peribiliary cyst. (a) Ultrasound shows a multi-septated cyst measuring 33 mm in diameter at 23 months preoperatively. (b) The cyst measures 39 mm at 9 months preoperatively. (c) The cyst is 45 mm at 3 months preoperatively. (d) Computed tomography (CT) demonstrates a 45 mm multi-septated cyst predominantly within segment III with no calcification. (e) There is no contrast effect on contrast-enhanced CT, and (f) no bile duct traffic on drip infusion cholecystocholangiography CT. (g, h) Magnetic resonance imaging and magnetic resonance cholangiopancreatography reveal cysts in close proximity to B3.

Figure 2. Surgical resection. (a) Trocar placement. (b) Location of the cyst and planned resection line. (c) Glisson's sheath of S3 is dissected from the porta hepatis. (d) The liver is then resected along the demarcation line. (e) After completed resection. (f) Macroscopically, there is a septated and loculated cavity within section III. The cyst runs alongside the bile duct.

Figure 3. Histopathological findings. (a) Microscopically, peribiliary glands are located around the cyst (HE staining, $\times 200$). (b) The cyst is lined by a single layer of cylindrical epithelium without cytological atypia (CK-19 staining, $\times 100$). This epithelium is not underlined by an ovarian-like hypercellular stroma as stained by (c) estrogen receptor, (d) progesterone receptor, and (e) inhibin ($\times 100$).

Conflict of Interest Statement:

The authors declare no conflict of interest.

Funding Statement:

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Ethics Statement:

Informed consent was obtained from the patient for the surgical procedure and for publication of this case report, including the ultrasonographic, CT, MRI findings, and intraoperative observations along with other clinical details. All identifying information has been appropriately anonymized to maintain patient confidentiality. All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation and with the Helsinki Declaration of 1975, as revised in 2013.

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