ABSTRACT

The incidence of cysts of the biliary system is approximately 1 in 100,000 to 150,000 in Western populations. The pathophysiology of biliary tree cysts is believed to be a result of an anomalous pancreaticobiliary junction. Although surgery is the preferred treatment for biliary tree cysts, surgical resection of the cyst does not eliminate the risk of malignancy, but rather it reduces the risk. The main goal of surgery is complete resection of the cyst and reconstruction of the biliary tree. In addition to the complexity of the procedure, variations of the biliary tree and the hepatic hilum luminal structures make the surgery more difficult for the surgeon. In this report, the case of a 46-year-old male patient is described. He presented at the clinic with a previously diagnosed choledocal cyst of Todani class 1 and a history of peptic ulcer perforation surgery. Further evaluation verified a choledocal cyst measuring 6x4 cm. Robotic complete surgical resection of the cyst and reconstruction of the biliary tree with hepaticojejunostomy was performed. During the bladder dissection, a luminal structure localized near Calot’s triangle was observed and determined to be the left hepatic artery coursing over the anteromedial gall bladder and entering the liver at the level of the bladder fundus. The cyst and gall bladder were dissected while preserving the left hepatic artery. In conclusion, a minimally invasive approach for choledocal cyst resection should be performed at experienced centers because of frequent variations in both the hepatic arterial system and the biliary tract.

Keywords: Aberrant hepatic artery; choledocal cyst; robotic surgery.

Introduction

Cysts of the biliary system are seen in 1 in 100,000 to 150,000 in Western populations and 1 in 1000 in Asian populations. They are more common in women and are most frequently located in the extrahepatic biliary tract. [1] The pathophysiology of bile tree cysts is believed to be a result of an anomalous pancreaticobiliary junction. According to the Todani modification of the Alonso-Lej classification, biliary tract cysts are classified into five groups. Diffuse choledocal cysts are classified as type 1.

The most dramatic complication of choledocal cysts is malign transformation (in 10–30%). [2] Adenocarcinoma is the most frequently occurring malignancy of the extrahepatic biliary tract that originates from the cysts. Although
surgery is the preferred treatment for biliary tree cysts, surgical resection of the cyst does not eliminate the risk of malignancy but rather reduces the risk.\textsuperscript{[3]} Other clinical symptoms such as cholestasis, jaundice, pancreatitis, and cirrhosis may also be complications of choledocal cysts.

Management of choledocal cysts requires experienced surgeons specialized in hepatopancreaticobiliary (HPB) surgery. The main goal of the surgery is complete resection of the cyst and reconstruction of the biliary tree. The laparoscopic–robotic minimally invasive approach is a more complicated procedure that must be performed at experienced centers. In addition to the complexity of the procedure, common variation of the bile tree and the hepatic hilum luminal structures also make the surgery more difficult for the surgeon.

Many variations of the hepatic arterial system are described, whereas fewer are present in the portal venous system. The proper hepatic artery originating from the celiac axis and continuing as the common hepatic artery after the branching of the gastroduodenal artery, with branching to left and right hepatic arteries, is the most common arterial structure of the liver with an incidence of 60\%.\textsuperscript{[2]} However, in the case described here, we found that the left hepatic artery originating from the proper hepatic artery was aberrantly passing over the gall bladder, entering the liver at the level of the gall bladder fundus. Confusing the left hepatic artery with the cystic artery in our case may have had dramatic results.

Hepatic artery variations in specific cases, such as choledocal cyst surgery, may cause postoperative complications. We share our experience with robotic choledocal cyst excision and hepaticojejunostomy in a patient who had been previously operated for peptic ulcer perforation and had left hepatic artery variation.

**Case Report**

A male patient, aged 46 years, presented to our clinic with previously diagnosed choledocal cyst of Todani class 1. He had a history of peptic ulcer perforation surgery. Further evaluation of the patient with abdominal computed tomography (CT) verified a choledocal cyst measuring 6x4 cm (Fig. 1). The cyst was evaluated as proceeding into the pancreatic parenchyma at the head of the pancreas from the distal side of the cyst. Robotic complete surgical resection of the cyst with reconstruction of the bile tree with hepaticojejunostomy was planned for the patient.

The patient was taken into the operating theatre. After placing the 5 mm trocars, docking of the robot was completed. The patient had intra-abdominal adhesions as a result of previous peptic ulcer surgery. Immediately after adhesiolysis, the choledocal cyst was dissected. The dissection started from the hepatoduodenal ligament and advanced to the gall bladder over the cystic duct. During the bladder dissection, a luminal structure localized near Calot’s triangle was skeletonized and determined not to be perfusing the gall bladder. Further dissection of the luminal structure showed that it was the replaced left hepatic artery sailing over the anteromedial of the gall bladder and entering the liver at the level of the gall bladder fundus. The cyst and gall bladder were totally dissected from the environmental structures, including the portal vein, while preserving the replaced left artery. Choledoc was transected from 1 cm distal of the left and right hepatic duct bifurcation. The distal end of the choledocal cyst proceeding to the pancreatic head parenchyma then was dissected, sealed, and transected with the help of an endoscopic stapler, ensuring that the cyst was totally excised. A pathological specimen consisting of the cyst with gall bladder was removed with the help of an endoscopic bag (Fig. 2) (it is the Fig. 3 of the previous manuscript). After the resection of the specimen, reconstruction of the hepaticojejunostomy started with the transection of the jejunum from 40 cm distal of the ligament of Treitz with an endoscopic stapler. The distal stump of the jejunum was carried up to the liver.

**Figure 1.** CT image of the choledocal cyst.
from the mesocolic opening and was anastomosed to the common hepatic duct by using the robot. A Roux-en-Y anastomosis was completed after the hepaticojejunostomy. Operation duration was 240 minutes and blood loss was 100 cc. The patient had no postoperative complications and was discharged on postoperative day 6. He had no complaints at follow-up.

**Discussion**

Those at the frontier of HBP surgery have not reached a consensus in evaluating variations in hepatic arteries in humans, but it was first discussed and classified widely by Michels\(^4\) in 1960; 26 different pathways for hepatic arteries were shown. The most common variation of the left hepatic artery is origination from the left gastric artery.\(^5\)

Biliary tract variations and hepatic arterial variations are the most complicating features of hapatipancreaticobiliary surgery. Hepatic arterial variations may be seen in up to 30–40% of patients, according to different references.\(^4-6\) Rong et al. studied hepatic artery variations in the CT scans of 2275 patients and introduced the term “vessel through strait sign (VTSS)”.\(^7\) They found an aberrant artery at the ligamentum venosum in which 89.4% of this aberrant artery consisted of the left hepatic artery. This variation is commonly seen, as in our case. The variation may be so extreme and complicating that the left hepatic artery may course around the esophagus and thenperfuse the liver.\(^8\)

In addition to hepatic artery variations, variation of the biliary tract is also a challenging problem for the surgeon. Even the variations of abnormal biliary tract anatomy, such as biliary tract cysts, are the most challenging of the biliary tract variations. Hathiramani et al.\(^9\) and Mantas et al.\(^10\) presented cases with aberrant bile ducts draining into a choledocal cyst. Fortunately, our case did not have a biliary tract variation coinciding with hepatic artery variation and choledocal cyst.

The only treatment for choledocal cyst is surgical resection because of the cysts’ increased potential for malign transformation. The pathogenesis of malign transformation is unknown; therefore, resection is the only way to decrease the malignant potential of these cysts. However, in 2017, Kwon et al.\(^11\) likely described the mechanism of malign transformation. They stated that the metastatic stratified squamous epithelium that they observed in one of the choledocal cysts they resected is the precancerous lesion of the adenocarcinoma that arises from the cyst. Another probable malign transformation mechanism is due to choledocal cyst papillomatosis. Malign transformation of these lesions is reported in around 35–41% of patients.\(^12\) Although choledocal cysts are more frequent in female patients, men more frequently experience papillomatosis and malign transformation.\(^13\) As stated before, complete resection of the cyst is the only treatment for biliary tract cyst complications, but the risk of malignancy cannot be eliminated. Patients still carry a higher
than average risk of malignancy, but the risk is currently lower than it was previously. The reason for malignancy after complete resection of the cyst is unknown.

In addition to the malign transformation of the cysts, biliary stones, bile stasis, and cholangitis are also complications of choledocal cysts. A very rare complication of choledocal cysts is spontaneous perforation, which may occur at the cyst or at the intact segment.\[14]\n
After the resection and reconstruction of the bile tract, one of the late complications of the surgery is duodenogastric regurgitation (DGR), which presents as non-specific abdominal complaints. The most common reason for DGR is hepaticoduedonostomy (HD). However, patients who undergo hepaticojejunostomy (HJ) do not experience DGR.\[15]\ For this reason, HJ anastomosis is superior to HD in external biliary tract reconstruction operations.

Actually the standard approach for biliary tract cysts are open resection of the cyst and reconstruction. As the new technologies in medicine improved, alternatives to open surgery such as laparoscopic approach started to be applied at daily use for HPB surgeons at biliary tract surgery. Robotic approach is the latest improvement and we believe that is the most feasible. Because of its 3D scope and ergonomic arms, robotic approach is superior to laparoscopic approach at biliary tract resection-reconstruction surgery.\[16]\n
In conclusion, complete resection of choledocal cysts and reconstruction of the biliary tract is a complicated surgery that must be performed by HPB surgeons. Otherwise, biliary tract adenocarcinomas are the probable long-term result for adult patients with choledocal cysts. A minimally invasive approach for choledocal cyst resection is applied at experienced centers, but because of the frequent variations in both the hepatic arterial system and biliary tract, surgeons may struggle with the procedure. We believe that robotic surgery as a minimally invasive approach to choledochal cyst resection increases the likelihood of minimally invasive completion of the procedure when compared to a laparoscopic approach in cases with extensive adhesions due to previous surgery and hepatic artery variations, as in our case.

Disclosures

**Informed Consent:** Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.

**Peer-review:** Externally peer-reviewed.

**Conflict of Interest:** None declared.

### References