

Biliary cancer risk should be kept in mind in laparoscopic surgery for adult choledochal cyst

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ABSTRACT

Choledochal cysts are a congenital pathology with a malignant potential that increases with age. The preferred treatment is surgical excision in all age groups, and is now often performed laparoscopically. The aim of this case report was to emphasize the importance of biliary tumor risk when laparoscopic excision of a choledochal cyst is performed in adults. A 46-year-old woman was referred to the clinic for a type 1 choledochal cyst with an accompanying gallbladder polyp with regular margins. She had not had previous abdominal surgery and there were no co-morbidities. The patient underwent a laparoscopic cyst excision, cholecystectomy, and hepaticojejunostomy. The hepaticojejunostomy failed and the procedure was completed by converting to open surgery. The gallbladder and choledochal cysts were removed through an abdominal incision. A specimen pathology report indicated gallbladder cancer (T2N0M0). One month later, a gallbladder bed resection and perihilar lymph node dissection were performed. No malignancy was detected in the second pathology specimen. Follow-up at the postoperative 16th month revealed no difficulties. A literature review of 231 adult laparoscopic choledochal cyst excisions yielded a 2.6% risk for biliary cancers. Especially in adults, biliary tumors are more commonly associated with choledochal cysts. The cancer risk should not be forgotten during laparoscopic surgery for congenital choledochal cysts.

Keywords: Bile-duct cancer; cholecystectomy; choledochal cyst; gallbladder carcinoma; hepaticojejunostomy; minimal invasive surgery.

Introduction

Choledochal cyst is a congenital disease with cystic dilatation of the bile ducts. It is often found in the first decade, but in recent years the incidence is increasing in older ages.^[1] It is seen about 10 times more in Asian societies than others. Todani et al.^[2] the cyst is divided into 4 types according to the location and dilatation type. While ultrasound is used as the first test at diagnosis, Magnetic resonance cholangiopancreatography is used as the most sensitive technique. Its complications

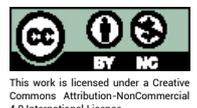
are cholelithiasis, hepatolithiasis, intrahepatic abscess, secondary bile cirrhosis, portal hypertension and biliary cancer. The biliary cancer can develop from the cyst (50%), gallbladder (48%) or intrahepatic bile ducts (2%).^[3] Surgical excision is the accepted treatment for most choledochal cysts.

In this case report, our aim was; to present an adult patient who underwent laparoscopic choledochal excision because of choledochal cyst and subsequently detected a gallbladder cancer.



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Case Report

A 46-year-old woman without any comorbidities was referred us because of the gallbladder polyp and type 1 choledochal cyst. There was no pathology in the physical examination of the abdomen and laboratory test. In MR cholangiography; a 3x3.5x5 cm benign looking like polyp in the gallbladder and a dilatation up to 26 mm in the choledochus was detected (Fig. 1). Laparoscopic surgery was planned and the patient was evaluated as ASA 1. In laparoscopic exploration, the gall bladder was in normal appearance and the polyp inside was felt as mobile. No lymph node or organ invasion were detected in laparoscopic exploration. Laparoscopic choledochal cyst excision, cholecystectomy and hepaticojejunomy were performed. However; a bilier leak was found in the hepaticojejunostomy line at the end of surgery and conversion to open surgery was thought to be necessary. The bile leak was seen posteriorly on anastomosis line, supportive sutures were placed and the operation was terminated. The specimen was removed from the laparotomy incision. On the macroscopic examination of the gall bladder, there was no problem with serosa and the polyp was palpable in the sac. Postoperatively, the pathologic report of the patient who was discharged without any complaints revealed a 6 cm sized, moderately differentiated adenocarcinoma in the gallbladder localization. It was reported that the tumor spread to the perimuscular tissue, there was no peritoneal perforation and lymph node spread (T2N0M0 - Stage 2). One month after the first operation, the patient underwent gallbladder bed resection and lymph node dissection. The patient was discharged without any problems on the eighth day after the second operation. No tumoral foci were detected in the second operative pathology. The patient continues to have a problem-free life at postoperative 16th month.

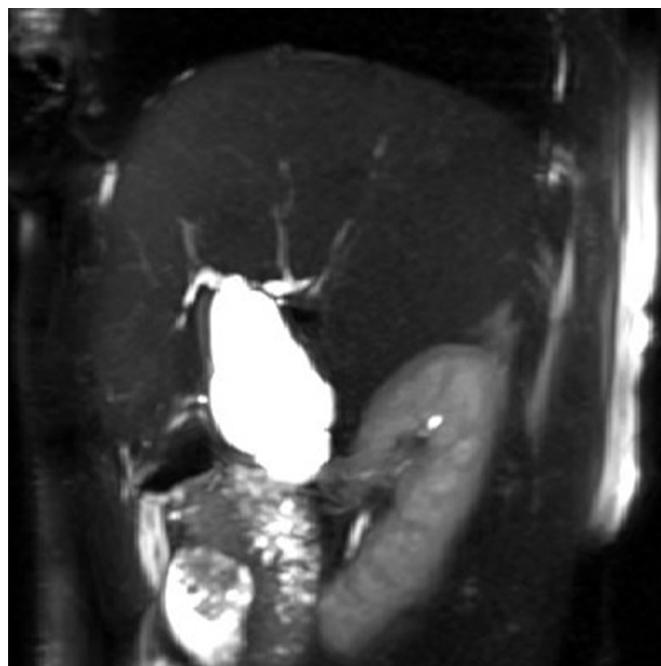


Figure 1. Dilatation of the choledochus (MR cholangiography).

Discussion

Although the choledochal cyst was first noticed early in the 18th century, the first case in the literature was published in the 19th century. In 1985, Todani et al.^[4] reported the classification of the lesions, which are defined as dilatation of the biliary tract. The most common types of choledochal cysts that are divided into five types are Types 1 and 4. Radiological examination is essential for both in the diagnosis and determination of the cyst type.^[5] Type 1 and type 4 choledochal cysts, require surgical treatment for prophylactic purposes. With the extend of minimally invasive methods, choledochal cyst excision and reconstruction has been started to be done by laparoscopic methods.^[6]

In the study of Liem et al.^[7] The rate of conversion to open surgery was only 0.6% in 616 laparoscopic and open group

Table 1. The details of literature review

Author	Year	Country	Period	Age	No	Biliary malignancy
Palanivelu	2008	India	1996–2008	26.5	35	3
Hwang	2012	South Korea	2009–2011	37.8±11.1	20	0
Jang	2013	China	2003–2011	32.7±13.9	82	1
Liu	2014	China	2007–2011	24.2±8.3	35	0
Duan	2015	China	2009–2013	23 (18–30)	31	0
Nag	2017	India	2011–2016	45.5±11.4	20	1
Gundogan	2018	Turkey	2013–1018	41.5 (22–49)	8	1
Total					231	6 (2.6%)

comparison study. In Diao et al.'s^[8] study; biliary leak was seen in 2% in laparoscopic group and 11% in open group. Contrary to our expectations, the biliary leak was more common in the open group. This difference is due to the fact that the increasing laparoscopic technical skills and the magnification advantage of laparoscopic surgery. Although our case were completed laparoscopically, we converted to open surgery to avoid an unsafe anastomosis. Comparative studies reported that; laparoscopic surgery did not have a disadvantage over open surgery, even the length of hospital stay and the need for reoperations were significantly lower.^[7,8] There were also publications showing that robotic surgery of minimally invasive surgical methods increases the safety of anastomosis of hepaticojejunostomy and reduces the rate of conversion to open surgery.^[9]

In choledochal cysts, the likelihood of developing biliary cancer increases with age. Cancer risk in under 10 years is below 1%, but 35% over 60.^[10,11] The most frequent site of malignancy is the cyst epithelium, the second is the gallbladder. The risk of malignancy at the gallbladder was previously considered a contraindication to laparoscopic surgery. The primary drawbacks were tumor spread due to gallbladder perforation and inadequate oncological surgery.^[12] With the development of minimally invasive surgery and the increase in experience, this contraindication has begun to disappear. In the last decade oncologic results with open surgery, let the laparoscopic technique as an acceptable alternative.^[13]

So far, we operated eighth adult choledochal cysts by laparoscopy and only in one case we identified a malignancy and we reported it here. The details of the remaining literature review was summarized in Table 1. Palanivelu^[14] reported three conversions all were done because there was suspicion of malignancy. Two patients had a mass at the junction of the cystic duct and the cyst wall. Cyst excision and removal of regional lymph nodes were performed. Histopathology revealed cholangiocarcinoma

with negative margins and nodes. These two patients had survived disease-free for 23 months. The third patient had a large choledochal cyst that extended up to the ampulla, replacing the entire pancreatic parenchyma. It was decided that the patient would need a Whipple's resection to remove the cyst in toto, so they performed it after conversion. Jang et al.^[15] reported a 37-year-old female presented for regular follow-up after the excision of a laparoscopic choledochal cyst (type Iva). A pancreatic head

mass was detected after 39 months and pancreaticoduodenectomy with SMV resection was performed. She got postoperative chemotherapy and was disease free for 36 months. Recently Nag reported^[16] one patient detected to have gallbladder cancer on a suspicious mucosal lesion on inspection of gall bladder specimen and this patient finally underwent radical cholecystectomy with choledochal cyst excision. The incidence of biliary cancer in 231 laparoscopic adult choledochal cyst was 2.6% (Table 1).^[14-19]

Conclusion

Choledochal cyst is a rare premalignant disease and it should be kept in mind that malignancy may accompany in laparoscopic resection of choledochal cyst, especially in adults.

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.

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