

This article is licensed under the Creative Commons Attribution-NonCommercial 4.0 International License (CC BY-NC) (<http://www.karger.com/Services/OpenAccessLicense>). Usage and distribution for commercial purposes requires written permission.

## Single Case

# Robot-Assisted Laparoscopic Resection of a Todani Type II Choledochal Malformation

Anneke ten Hove<sup>a</sup> Ruben H.J. de Kleine<sup>a</sup> Maarten W. Nijkamp<sup>a</sup>  
Annet S.H. Gouw<sup>b</sup> Timco Koopman<sup>b</sup> Joost M. Klaase<sup>a</sup>

<sup>a</sup>Department of Surgery, Division of Hepato-Pancreato-Biliary Surgery and Liver Transplantation, University of Groningen, University Medical Center Groningen, Groningen, The Netherlands; <sup>b</sup>Department of Pathology, University of Groningen, University Medical Center Groningen, Groningen, The Netherlands

## Keywords

Choledochal malformation · Todani type II · Robot-assisted laparoscopic resection

## Abstract

Choledochal malformation (CM) comprise various congenital cystic dilatations of the extrahepatic and/or intrahepatic biliary tree. CM is classified into five different types. Our case describes a 58-year-old man presenting with acute abdominal pain. Further examination showed a Todani type II CM. Treatment for type II is complete cyst excision without the need for an extrahepatic bile duct resection. A robot-assisted laparoscopic resection of the CM was performed and the patient recovered without complications. Pathology results showed a Todani type II malformation in which complete squamous metaplasia has occurred. In this paper, we report, to the best of our knowledge, the first description of a robot-assisted laparoscopic resection of a type II CM.

© 2019 The Author(s)  
Published by S. Karger AG, Basel

## Introduction

Choledochal malformation (CM), previously called choledochal cyst, represents a congenital cystic dilatation of the extrahepatic and/or intrahepatic biliary tree. CM is rare in Western countries, with an incidence of 1:59,000 live births in The Netherlands [1]. However, this rate is remarkably higher in the Asian population with an incidence of 1:1,000 and CM occurs 3–4 times more frequently in females [2]. The reason for this higher incidence is still unclear, as is the etiology of the CM. The most serious and dangerous complication of CM is cholangiocarcinoma. According to the literature, the risk of developing malignancy is 11% and this rate increases with age [3]. CM were first classified by Alonso-Lej et al. [4] and further refined by Todani (see Fig. 1) [5]. According to this classification, CM is classified into five different types. Type II, the rarest of all with an incidence of 5.4% [6], appears as an isolated diverticulum protruding from the wall of the common bile duct. Nowadays the standard treatment for type I and IV is resection of the extrahepatic bile duct including cyst excision and the obligatory cholecystectomy [7]. This is followed by construction of a biliary-enteric anastomosis to restore continuity with the gastrointestinal tract through a Roux-and-Y loop. Type II is managed with only a complete cyst excision without the need for an extrahepatic bile duct resection. Extrahepatic bile duct resection is only advocated when the cyst has a large neck at its junction with the common bile duct or when there is an abnormal pancreatico-biliary junction, because these patients have higher risk of both gallbladder and bile duct cancer [8].

Using robotic surgery, it has become possible to perform complex minimal access liver, bile, and pancreatic procedures. Robotic surgery provides a steady three-dimensional view, articulated instruments, and magnification of the focused surgical area [9]. The literature reports many studies about robot-assisted procedures for pancreatic resection and hepatectomy [10]. However, studies of robot-assisted excision of the CM are scarce with incidental reports of Todani type I and IV CM resections only [11]. In this paper, we report, to the best of our knowledge, the first description of a robot-assisted laparoscopic resection of a type II CM.

## Case Report

A 58-year-old man (ASA 2, 182 cm, BMI 31.4) presented with acute upper abdominal pain, without vomiting, jaundice, or fever. Physical examination showed no positive findings, except tenderness over the epigastric area. Routine blood panel, blood amylase, liver enzymes, and the tumor marker CA 19-9 results were normal. Abdominal ultrasound showed a globular deviation near the gallbladder, suggestive of a cyst. Computed tomography and magnetic resonance cholangio-pancreatography showed a 25-mm-long, 26-mm-wide CM at the level of the hepatic hilum (Fig. 2). The CM seemed to have a connection with the common bile duct without affecting the duct itself, consistent with a Todani type II. After preoperative examinations, we decided to perform a robot-assisted laparoscopic resection of the cyst and a cholecystectomy.

## Surgical Method

With the patient in supine position, the 8-mm-diameter trocar was open inserted under the level of the umbilicus to establish pneumoperitoneum and for camera placement (arm 3). No significant abnormalities were detected in the abdominal cavity. Under visual guidance,

the 8-mm trocars of arm 1 and arm 2 were inserted into the right abdomen creating two left hands. Arm 4 was placed at the left abdomen. A 12-mm airseal® trocar was placed in the upper left of the abdomen. The peritoneum of the neck of the gallbladder and hepatoduodenal ligament were opened after maximum surgical exposure. An electric hook and scissors were used to dissect the peritoneal adhesions around the CM whilst moving it ventrally away from the common bile duct. The small stalk of the malformation was identified with its orifice into the common bile duct. An intraoperative view of the CM can be seen in Figure 3. After removal of the CM, a small opening in the common bile duct remained with evasion of bile. The small opening was closed with a cross-stitch PDS 5-0. Retrograde resection of the gallbladder was performed with clipping of the cystic duct and cystic artery after obtaining a critical view of safety. Both the gallbladder and the CM were removed through a surgical bag and were sent for pathology. One low vacuum abdominal drain was placed on the left at the level of the hepatoduodenal ligament. The abdominal cavity was confirmed to have no active bleeding, and the devices and gauzes were all counted and correct.

### Surgical Outcome

The total operation time including anesthesia was 4 h and 20 min without detected blood loss. Two days after the operation the abdominal drain was removed and the patient was discharged. No complications occurred.

### Pathology

Pathological findings are shown in Figure 4. Gross examination showed a smooth-walled cyst filled with bilious debris. Microscopic examination revealed a cystic lesion completely lined by uniform and normally maturing squamous epithelium, without columnar bile duct epithelium. This squamous epithelium lacked a granular layer. The cystic wall was fibrous and showed focal pre-existent peribiliary glands. The bilious debris within the cyst contained keratinized material mixed with bile. With immunohistochemistry, the squamous epithelium stained positive with CK5/6, p40 and p63, confirming the squamous nature of the epithelium. Caldesmon showed circularly arranged thin smooth muscle bundles in the cyst wall, compatible with a bile duct dilatation or biliary diverticulum, instead of a simple cyst or epidermal cyst. In conclusion, pathologic examination revealed a squamous cystic lesion with absence of bile duct epithelium and presence of keratinized material, which is likely to have had a connection with the bile duct because of the presence of bile within the cyst. Due to the lack of a granular layer in the epithelium and the presence of smooth muscle in the cyst wall, histology was not typical of an epidermal cyst. The lesion is therefore consistent with a Todani type II malformation cyst in which complete squamous metaplasia has occurred.

### Discussion

The largest western series of Todani type II reported surgical treatment in 19 patients. Surgery in 3 patients was attempted laparoscopically; however, the procedure required to be converted in all patients due to technical difficulties [6]. We report, to the best of our knowledge, the first successfully robot-assisted laparoscopic resection of a type II CM.

The most widely used robotic system is the Da Vinci Surgical System (Intuitive Surgical, Inc., Mountain View, CA, USA). Benefits of robotic surgery using this system are improved visualization due to three-dimensional view and magnification imaging system, articulated instrumentation, and advanced dexterity of the instruments. Due to these advantages, many surgeons may prefer robotic surgery over laparoscopic surgery. However, robotic surgery faces a considerable learning curve and the costs of the system (including instruments) are considerably high. Therefore, the role of robotic surgery continues to be a subject of evaluation.

### Pathology

Histological examination showed a unique case of Todani type II CM completely lined with squamous epithelium without bile duct epithelium. A similar finding has been reported in another Todani type II cyst by Kwon et al. [12] in 2017. This is in contrast to conventional CM, which is usually lined by columnar bile duct epithelium in which focal squamous metaplasia can occur [13]. The findings are not consistent with an epidermal cyst because of the presence of bile, the absence of a granular layer in the squamous epithelium, and the presence of smooth muscle bundles within the cyst wall. In conclusion, histological findings are consistent with a Todani type II malformation cyst in which complete squamous metaplasia has occurred.

Patients with CM have a risk of 11% of developing malignancy [3]. The pathogenesis of carcinoma in patients with CM is unknown. A commonly accepted theory is that the carcinogenesis may be related to dysplasia and metaplasia of the epithelium [14]. It is also speculated that Todani types I and IV are both associated with an abnormal biliopancreatic junction and that the reflux of pancreatic secretions is responsible for malignant transformation [14]. Todani type II is not associated with an abnormal biliopancreatic junction and may have a different malignant degeneration process. This is supported by our pathological observation, where no columnar cells and focal squamous metaplasia were seen.

In conclusion, resection of Todani type II CM using robotic surgery is a safe and feasible option possibly enhancing the chance of saving the unaffected common bile duct, but still reducing the risk of development of a malignancy.

### Statement of Ethics

The authors have no ethical conflicts to disclose. The patient gave informed consent.

### Disclosure Statement

The authors have no potential conflicts of interest to declare.

### References

- 1 van den Eijnden MH, de Kleine RH, de Blaauw I, Peeters PG, Koot BP, Oomen MW, et al.; Nederlandse Studiegroep voor Choledochus Cysten/malformaties (NeSCHoC). Choledochal Malformation in Children: Lessons Learned from a Dutch National Study. *World J Surg.* 2017 Oct;41(10):2631–7.
- 2 Lipsett PA, Pitt HA. Surgical treatment of choledochal cysts. *J Hepatobiliary Pancreat Surg.* 2003;10(5):352–9.
- 3 Ten Hove A, de Meijer VE, Hulscher JB, de Kleine RH. Meta-analysis of risk of developing malignancy in congenital choledochal malformation. *Br J Surg.* 2018 Apr;105(5):482–90.
- 4 Alonso-Lej F, Rever WB Jr, Pessagno DJ. Congenital choledochal cyst, with a report of 2, and an analysis of 94, cases. *Int Abstr Surg.* 1959 Jan;108(1):1–30.

- 5 Todani T, Watanabe Y, Narusue M, Tabuchi K, Okajima K. Congenital bile duct cysts: Classification, operative procedures, and review of thirty-seven cases including cancer arising from choledochal cyst. *Am J Surg*. 1977 Aug;134(2):263–9.
- 6 Ouaïssi M, Kianmanesh R, Belghiti J, Ragot E, Mentha G, Adham M, et al.; Working Group of the French Surgical Association. Todani type II congenital bile duct cyst: european multicenter study of the French surgical association and literature review. *Ann Surg*. 2015 Jul;262(1):130–8.
- 7 Mabrut JY, Bozio G, Hubert C, Gigot JF. Management of congenital bile duct cysts. *Dig Surg*. 2010;27(1):12–8.
- 8 de Vries JS, de Vries S, Aronson DC, Bosman DK, Rauws EA, Bosma A, et al. Choledochal cysts: age of presentation, symptoms, and late complications related to Todani's classification. *J Pediatr Surg*. 2002 Nov;37(11):1568–73.
- 9 Lanfranco AR, Castellanos AE, Desai JP, Meyers WC. Robotic surgery: a current perspective. *Ann Surg*. 2004 Jan;239(1):14–21.
- 10 Milone L, Daskalaki D, Fernandes E, Damoli I, Giulianotti PC. State of the art in robotic hepatobiliary surgery. *World J Surg*. 2013 Dec;37(12):2747–55.
- 11 Han JH, Lee JH, Hwang DW, Song KB, Shin SH, Kwon JW, et al. Robot resection of a choledochal cyst with Roux-en-y hepaticojejunostomy in adults: initial experiences with 22 cases and a comparison with laparoscopic approaches. *Ann Hepatobiliary Pancreat Surg*. 2018 Nov;22(4):359–66.
- 12 Kwon DH, Johnson LB, Ozdemirli M. Primary Epidermoid Cyst of Biliary Duct Presenting as Choledochal Cyst. *Int J Surg Pathol*. 2017 Oct;25(7):619–22.
- 13 Katabi N, Pillarisetty VG, DeMatteo R, Klimstra DS. Choledochal cysts: a clinicopathologic study of 36 cases with emphasis on the morphologic and the immunohistochemical features of premalignant and malignant alterations. *Hum Pathol*. 2014 Oct;45(10):2107–14.
- 14 Jordan PH Jr, Goss JA Jr, Rosenberg WR, Woods KL. Some considerations for management of choledochal cysts. *Am J Surg*. 2004 Jun;187(6):790–5.

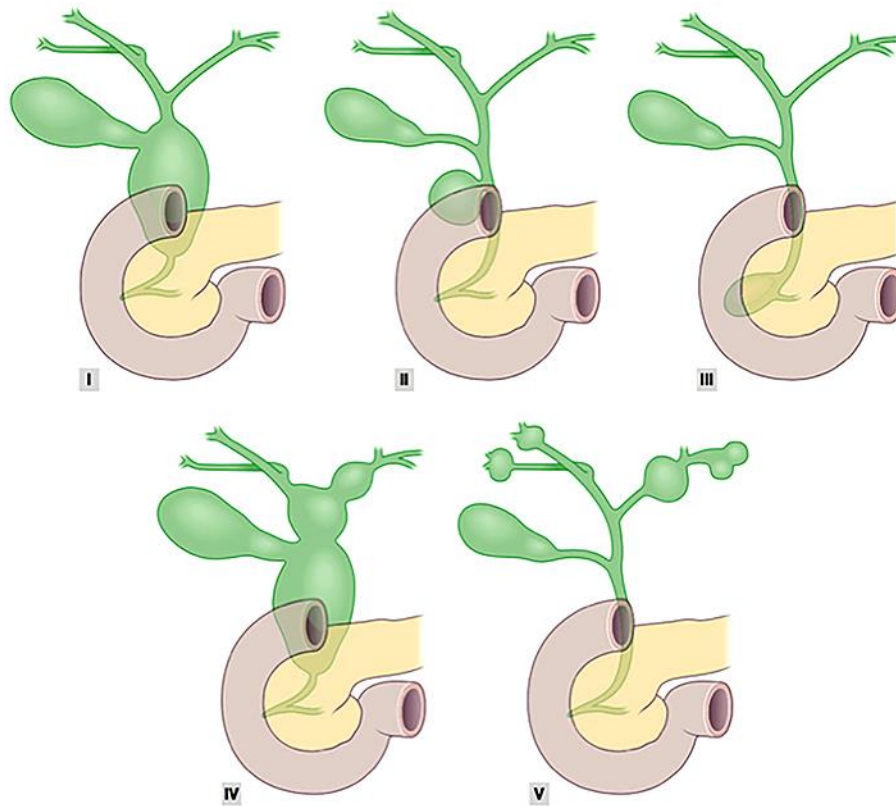


Fig. 1. Todani classification of choledochal malformations.

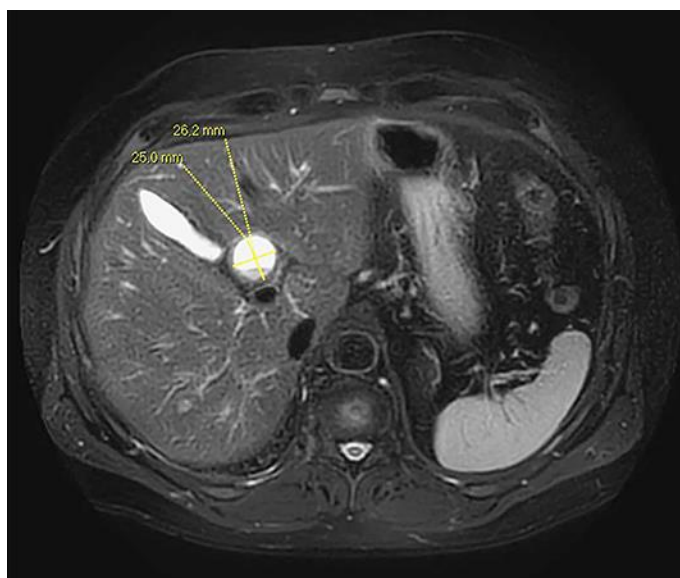
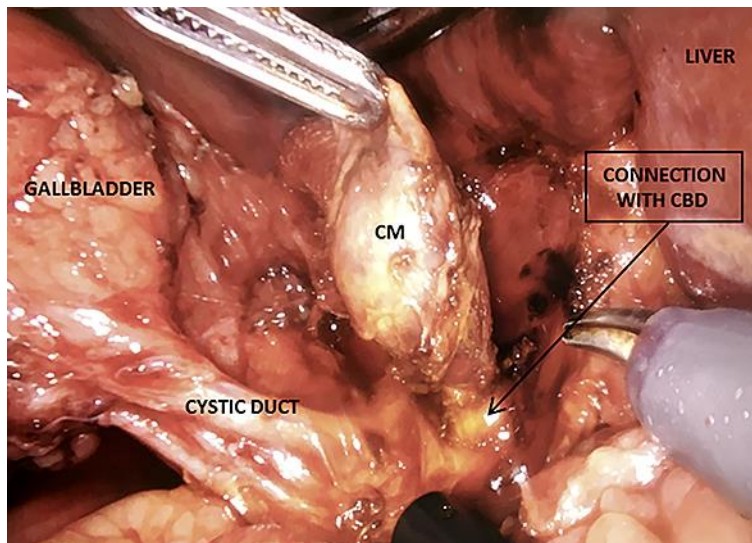
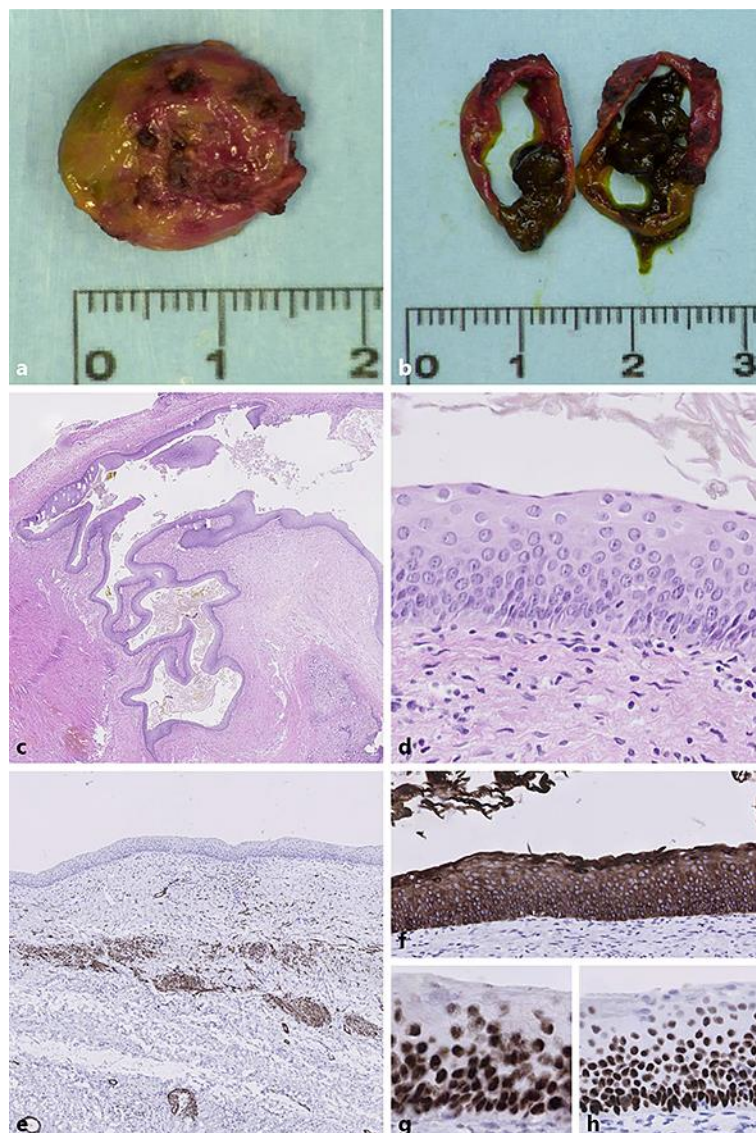


Fig. 2. Magnetic resonance cholangio-pancreatography showing type II choledochal malformation.



**Fig. 3.** Intraoperative view of a choledochal malformation. CM, choledochal malformation; CBD, common bile duct.



**Fig. 4.** Gross examination of the cyst (a), showing a thin-walled cyst filled with bilious debris on cut sections (b). c, d The cyst was filled with keratinous material mixed with bile and was completely lined by squamous epithelium. c Magnification,  $\times 20$ . d Magnification,  $\times 200$ . e Immunohistochemistry for caldesmon showed smooth muscle bundles within the cyst wall. CK5/6 (f), p40 (g), and p63 (h) confirmed the squamous nature of the epithelium.