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Semeret T. Munie  
*Henry Ford Health System*

Hassan Nasser  
*Henry Ford Health System, hnasser2@hfhs.org*

Pauline H. Go

Kelly Rosso

Ann Woodward  
*Henry Ford Health System, AWOODWA1@hfhs.org*

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**Recommended Citation**
Munie S, Nasser H, Go PH, Rosso K, and Woodward A. Case report of a duplicated cystic duct: A unique challenge for the laparoscopic surgeon. Int J Surg Case Rep 2019; 56:78-81.

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Case report of a duplicated cystic duct: A unique challenge for the laparoscopic surgeon

Semeret Munie a, Hassan Nasser b,⁎, Pauline H. Go c, Kelly Rosso d, Ann Woodward a

a Department of Surgery/Division of Trauma and Acute Care Surgery, Henry Ford Hospital, 2799 W Grand Blvd, Detroit, MI, 48202, USA
b Department of Surgery, Henry Ford Hospital, 2799 W Grand Blvd, Detroit, MI, 48202, USA
c Department of Cardiothoracic Surgery, University of Iowa, 200 Hawkins Drive, Iowa City, IA, 52242, USA
d Department of Surgical Oncology, Banner MD Anderson, 14416 W Meeker Blvd, San City West, AZ, 85375, USA

A R T I C L E   I N F O

Article history:
Received 2 February 2019
Accepted 19 February 2019
Available online 28 February 2019

Keywords:
Duplicated cystic duct
Double cystic duct
Laparoscopic cholecystectomy
Bile duct variation

A B S T R A C T

INTRODUCTION: Anatomical variants of the extrahepatic biliary tree are numerous, adding significantly to the risk of bile duct injury during cholecystectomy, especially when laparoscopic approach is employed. Duplicated cystic ducts draining a single gallbladder are extremely rare.

PRESENTATION OF CASE: A 34-year-old female presented with signs and symptoms of acute cholecystitis which was confirmed on imaging. She was found to have an accessory cystic duct on laparoscopic cholecystectomy requiring conversion to open laparotomy with intraoperative cholangiogram to delineate the anatomy.

DISCUSSION: In the English literature, there has been 20 reported cases of double cystic duct with a single gallbladder. Most of these cases were diagnosed intraoperatively despite the completion of a preoperative endoscopic retrograde cholangiopancreatography in a few of these patients.

CONCLUSION: The limited success of preoperative biliary tract imaging in demonstrating anatomic aberrancies prior to cholecystectomy clearly highlights the importance of maintaining constant vigilance for even the slightest anatomic abnormality at operation. Any uncertainty or concern for ductal injury mandates immediate operative cholangiogram with cannulation of all structures in question.

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1. Introduction

Biliary tree anomalies have been detected in up to 47% of the population based on operative, cholangiographic and autopsy studies [1]. The basis of bile duct injury is failure to identify biliary anatomy especially in the cases aberrancies. Thus, identification of these anomalies in biliary anatomy is crucial to avoid the morbidity and mortality associated with bile duct injuries. Unlike the more common variant in which two cystic ducts drain into distinct gallbladders or cavities [2], duplicated cystic ducts draining a single, unilocular gallbladder is extraordinarily rare, with fewer than 20 cases reported in the English literature. We report our experience with laparoscopic cholecystectomy in the setting of double cystic ducts identified intraoperatively. The case report has been reported in line with the surgical case report (SCARE) criteria [3].

2. Presentation of case

We report a case of a 34-year-old female who presented to the emergency department with three days of constant right upper quadrant and epigastric abdominal pain with associated nausea. The patient had similar pain two months ago that resolved and did not seek medical attention. On presentation she had normal vital signs. Physical examination demonstrated right upper quadrant tenderness without peritoneal signs with negative clinical Murphy’s sign. Her blood work revealed a white blood cell count of 13,500 /microliter, alanine aminotransferase of 318 U/L, aspartate aminotransferase of 259 U/L, alkaline phosphatase of 120 U/L, and total bilirubin of 1.7 mg/dL. Ultrasonography demonstrated new mild gallbladder wall thickening and negative sonographic Murphy’s sign with equivocal suggestions of acute cholecystitis. Hepatobiliary inminodiacetic acid scan was subsequently performed and showed non-visualization of the gallbladder consistent with acute cholecystitis.

The patient was taken to the operating room and the cystic duct and artery were dissected free from the cystic triangle laparoscopically. Both structures were secured proximally and distally and divided sharply. The gallbladder was dissected from the bed using electrocautery. Due to the contracted and intrahepatic nature of

Abbreviations: ERCP, endoscopic retrograde cholangiopancreatography; IOC, intraoperative cholangiogram.
⁎ Corresponding author.
E-mail address: hmnasser@hfhs.org (H. Nasser).

https://doi.org/10.1016/j.ijscr.2019.02.030
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and transected and the gallbladder was removed. The gallbladder aspect of the duct (Fig. 3) The second accessory duct was clipped, then ligated and divided. Under sedation, the common bile duct was transected and the biliary tract was opened from the cystic duct (Fig. 2). An attempt was made to cannulate the second duct, which was transected and divided. The cholangiogram confirmed a well-functioning right and left hepatic lobe. Intraoperative-technique was performed through the cystic duct. During the evaluation, the stenosis appeared to be a bile duct intussusception. The decision was made to convert to an open procedure. On further evaluation, the gallbladder was excised (Fig. 1), and this just prior to removal of the gallbladder. From then the bile ducts were identified and divided. A top-down technique of the gallbladder approach was switched to a top-down technique.

![Image 1](image1)

![Image 2](image2)

### Table 1

| Case | Author, Year | Country | Age/gender | Duplication type | Type | Preoperative ERC | Intraoperative ERC | Operative approach |
|------|--------------|---------|------------|------------------|------|----------------|-------------------|-------------------|
| 1    | Ferdman et al. 1991 | USA    | 70/Female  | Trabecular Type | Y    | Yes             | No                | Laparoscopic converted to open |
| 2    | Nakajou et al. 1995 | Japan | 57/Female  | Trabecular Type | Y    | Yes             | No                | Laparoscopic converted to open |
| 3    | Nakajou et al. 1996 | Japan | 60/Female  | Y Type           | Y    | No              | No                | Laparoscopic converted to open |
| 4    | Nakajou et al. 1996 | Japan | 60/Male    | Y Type           | Y    | No              | Yes               | Laparoscopic converted to open |
| 5    | Nakajou et al. 1996 | Japan | 65/Male    | Y Type           | Y    | No              | Yes               | Laparoscopic converted to open |
| 6    | Nakajou et al. 1996 | Japan | 65/Female  | Y Type           | Y    | Yes             | No                | Laparoscopic converted to open |
| 7    | Nakajou et al. 1996 | Japan | 70/Female  | Y Type           | Y    | Yes             | No                | Laparoscopic converted to open |
| 8    | Nakajou et al. 1996 | Japan | 70/Male    | Y Type           | Y    | No              | Yes               | Laparoscopic converted to open |
| 9    | Nakajou et al. 1996 | Japan | 70/Female  | Y Type           | Y    | No              | Yes               | Laparoscopic converted to open |
| 10   | Nakajou et al. 1996 | Japan | 70/Male    | Y Type           | Y    | No              | Yes               | Laparoscopic converted to open |
| 11   | Nakajou et al. 1996 | Japan | 70/Female  | Y Type           | Y    | Yes             | No                | Laparoscopic converted to open |
| 12   | Nakajou et al. 1996 | Japan | 70/Male    | Y Type           | Y    | No              | Yes               | Laparoscopic converted to open |
| 13   | Nakajou et al. 1996 | Japan | 70/Female  | Y Type           | Y    | No              | Yes               | Laparoscopic converted to open |
| 14   | Nakajou et al. 1996 | Japan | 70/Male    | Y Type           | Y    | No              | Yes               | Laparoscopic converted to open |
| 15   | Nakajou et al. 1996 | Japan | 70/Female  | Y Type           | Y    | Yes             | No                | Laparoscopic converted to open |
| 16   | Nakajou et al. 1996 | Japan | 70/Male    | Y Type           | Y    | No              | Yes               | Laparoscopic converted to open |
| 17   | Nakajou et al. 1996 | Japan | 70/Female  | Y Type           | Y    | No              | Yes               | Laparoscopic converted to open |
| 18   | Nakajou et al. 1996 | Japan | 70/Male    | Y Type           | Y    | No              | Yes               | Laparoscopic converted to open |
| 19   | Nakajou et al. 1996 | Japan | 70/Female  | Y Type           | Y    | Yes             | No                | Laparoscopic converted to open |
| 20   | Nakajou et al. 1996 | Japan | 70/Male    | Y Type           | Y    | No              | Yes               | Laparoscopic converted to open |

**Abbreviations:** ERC, endoscopic retrograde cholangiopancreatography; IOC, intraoperative cholangiogram.
was evaluated and showed the two cystic ducts with distal open lumens that communicated to the gallbladder.

Postoperatively, the patient’s liver function tests normalized. Patient was discharged home on postoperative day 3 and was tolerating diet. She was seen in the surgical clinic 2 weeks postoperatively and was doing well. The pathology report showed acute on chronic cholecystitis with mucosal ulceration and cholelithiasis.

3. Discussion

Intriguing with anomalous gallbladder anatomy and associated extrahepatic biliary duct aberrancy originates as far back as 1926 with Edward Boydens’s comparative report and classification of the various congenital anomalies of the gallbladder [4]. Thirty years later, Caster and Flannery categorized cystic duct duplication into 3 types: (1) Vype, wherein 2 cystic ducts join to form a single cystic duct that then enters the CBD, (2) Hype, in which each cystic duct independently joins the bile duct system at the CBD, right hepatic duct, left hepatic duct or common hepatic duct, and (3) trabecular type, in which one cystic duct enters the CBD while the other directly enters the liver parenchyma [5].

In the English literature, there has been 20 reported cases of duplicate cystic ducts draining a single gallbladder including our reported case (Table 1) [1,2,6–21]. Females constituted 75% of the reported cases. “H” type duplication with the cystic ducts joining the biliary system at the CBD, common hepatic duct, or right hepatic duct was reported in 11 cases (55%), representing the most common configuration. The “Y” and trabecular types represent 30% and 15% of the reported cases to date, respectively.

Double cystic duct was identified intraoperatively in 16 out of the 19 patients (84%) who were operated on. Despite the completion of a preoperative endoscopic retrograde cholangiopancreatography (ERCP) in 7 patients, the cystic duct anomaly was only identified in 3 cases (43%) [1,2,8–10,12,20]. This emphasizes the importance of being aware of this anatomic variant as even with invasive preoperative testing, the accessory duct was only identified intraoperatively. Choledectomy was performed and completed laparoscopically in 12 cases and intraoperative cholangiogram (IOC) was performed in 8 of these cases to delineate the anatomy when a second cystic duct was encountered [8–12,14,16–21]. Three other cases required conversion to laparotomy, one of which was our case, and was due to inability to clearly define biliary anatomy laparoscopically [2,13]. One case required reoperative laparotomy due to delayed recognition of the duplicated cystic duct, resulting in bile leak [9]. An IOC and preoperative ERCP was performed in that case but did not prevent the complication of a biliary leak.

4. Conclusion

The limited success of preoperative biliary tract imaging in demonstrating anatomic aberrancies prior to cholecystectomy clearly highlights the importance of maintaining constant vigilance for any slightest anatomic abnormality at operation. Any uncertainty or concern for ductal injury mandates immediate operative cholangiogram with cannulation of all structures in question. Although laparoscopic cholecystectomy is safe in experienced hands, a low threshold for conversion to laparotomy should be had when the anatomy cannot be deciphered.

Conflicts of interest

No conflicts of interest to be declared.

Funding

No source to be stated.

Ethical approval

The study is exempt from ethical approval in our institution.

Consent

Written informed consent was obtained for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Semeret Munie: Formal analysis; Writing – original draft.
Hassan Nasser: Writing – review & editing.
Pauline H. Go, MD: Writing – original draft.
Kelly Rosso, MD: Visualization.
Ann Woodward: Supervision.

Registration of research studies

Not applicable.

Guarantor

Ann Woodward, MD.

Provenance and peer review

Not commissioned, externally peer reviewed.

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