

Case report

Double cyst duct in a young woman with a history of systemic lupus: A case report

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ABSTRACT

Introduction and importance: Cholecystectomy is the most common general surgery procedure, necessitating thorough knowledge of bile duct anatomy. Despite the bile duct's anatomical diversity, reports of double cystic ducts are rare. This case presentation aims to emphasize the importance of recognizing this unusual anatomical variant during surgical procedures to prevent complications and ensure patient safety.

Case presentation: This is a 22-year-old woman with a history of lupus. Initial clinical and ultrasound evaluations concluded she had cholangitis, choledocholithiasis, and cholecystitis.

A cholecystectomy was performed, during which a double cystic duct was identified and confirmed with intraoperative cholangiography.

Five days after surgery, the patient exhibited increased bilirubin levels, and a follow-up cholangiography showed dilation of the common bile duct with no passage of contrast medium into the duodenum. She underwent ERCP with endoprosthesis placement and is currently under follow-up with adequate progress.

Clinical discussion: Double cystic ducts are extremely rare anatomical variants. These variations pose significant challenges for surgeons during surgery, emphasizing the need for a universal culture of safety during cholecystectomy.

Although this anatomical variant is rare, surgeons must be aware of it. Given the uncertainty of anatomy, it is advisable to perform intraoperative imaging, such as cholangiography, before sectioning any duct.

Conclusion: Double cystic duct variations are rare, but when bile duct anatomy is unclear, intraoperative cholangiography is essential. This technique improves visualization of biliary structures, aiding informed decisions before duct ligation and reducing the risk of bile duct disruption.

1. Introduction

Anatomical variations of the extrahepatic bile duct are common, but double cystic ducts are extremely rare [1]. In 1998, Takahisa et al. reported the case of a 78-year-old woman with a double cystic duct. At that time, fewer than 10 cases had been reported worldwide [2]. Only 22 cases have been reported in the literature, the last one in 2022 [3]. Women have 75 % of the cases [4]. The importance of understanding anatomical variants, such as the double cystic duct, lies in their role in preventing complications like disruption of the biliary tract, as these variants increase the risk of biliary duct injury [5]. In this article, we present the case of a young female patient who was admitted with cholangitis and acute cholecystitis. During the transsurgical procedure,

the presence of a double cystic duct was detected, which was ligated, and the bile duct was explored, leaving a T-drain. She was referred for endoscopic retrograde cholangiopancreatography (ERCP). This work has been reported according to SCARE criteria [6].

2. Case presentation

2.1. Patient information

A 22-year-old woman with a history of systemic lupus sought medical attention due to a 3-day clinical picture.

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2.2. Clinical findings

She first presented with pain in the right upper quadrant, accompanied by nausea and vomiting on three occasions, made up of gastrointestinal content. She presented with a fever of 38.5 degrees, acholia, choloria, and generalized jaundice. Despite self-medicating with paracetamol, she did not improve, so she came to the emergency room.

There the patient was found to have generalized jaundice, a yellowish sclera, dehydrated oral mucosa, a fever of 38.1 degrees, tachycardia, and pain in the right upper quadrant, with a Murphy sign but no signs of peritoneal irritation. Given these clinical manifestations that suggested cholecystitis and cholangitis.

2.3. Lab tests and imaging

She underwent ultrasound of the liver and bile ducts, which revealed dilation of the intrahepatic bile duct of up to 6 mm. There was choledochus of 18 mm, with evidence of echogenicity 18×8 mm in the middle third in the interior, which cast an acoustic shadow. The gallbladder measured 13×7.4 mm and had multiple stones inside, as well as free perivesicular fluid. The laboratory results were as follows: total bilirubin 8.35 mg/dl, direct bilirubin 6.41 mg/dl, indirect bilirubin 1.94 mg/dl, aspartate transaminase 48 U/l, alanine transaminase 37 U/l, gamma-glutamyl transpeptidase 226 U/l, albumin 1.90 g/dl, hemoglobin 8.3 g/dl, hematocrit 26.5 %, leukocytes $47.46 \times 10^3/\mu\text{l}$, neutrophils 92 %, prothrombin time 17.5 s, time to peak 61.6 s, and international normalized ratio 1.65.

2.4. Treatment and surgical intervention

Treatment included broad-spectrum antimicrobial therapy, fluid resuscitation, stabilization, maintenance in reserve blood products, and fresh-frozen plasma preparation for surgical procedures were performed. During the intervention, an open approach was taken through a Kocher-type incision. A gallbladder with a thickened wall and under tension, as well as perivesicular serous fluid and fibrin creams, was evident.

2.5. Intraoperative findings and procedure

We proceed to the dissection of the hepatocystic triangle, where the cystic artery is identified and a double duct is observed that connects the common bile duct with the gallbladder. These ducts and the artery were ligated using 3-0 silk (Figs. 1-3). Then we performed intraoperative cholangiography at the opening of the common bile duct with abundant

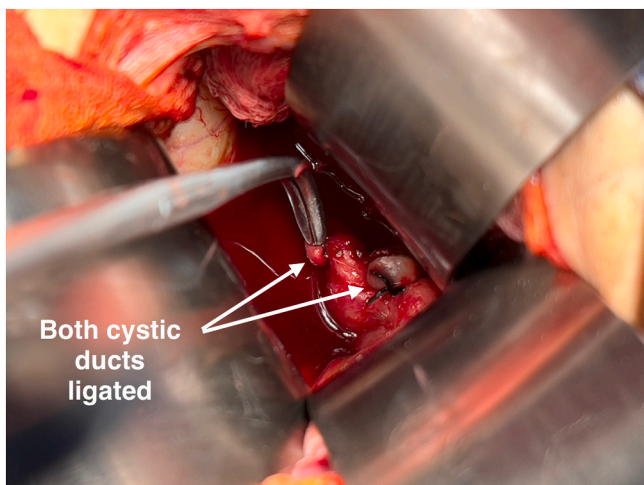


Fig. 1. Both ligated cysts Both cystic ducts ligated intraoperatively.



Fig. 2. Gallbladder with a 2nd cyst cannulated with a feeding tube.



Fig. 3. Postoperative gallbladder with both cysts ligated (2nd cystic cannula with a feeding tube).

discharge of purulent fluid, dilation of the intra- and extrahepatic bile duct, and 20 mm of the bile duct (Fig. 4). Next we explored the bile ducts and obtained multiple 2–8-mm stones. Saline washes were performed, and a T-tube was placed, followed by the insertion of a Penrose drain. Finally, the cavity was closed by planes.

2.6. Postoperative outcome and follow-up

After surgery, the patient had an adequate evolution: a decrease in leukocytosis and bilirubin levels, permeable T-drainage, no fever, and

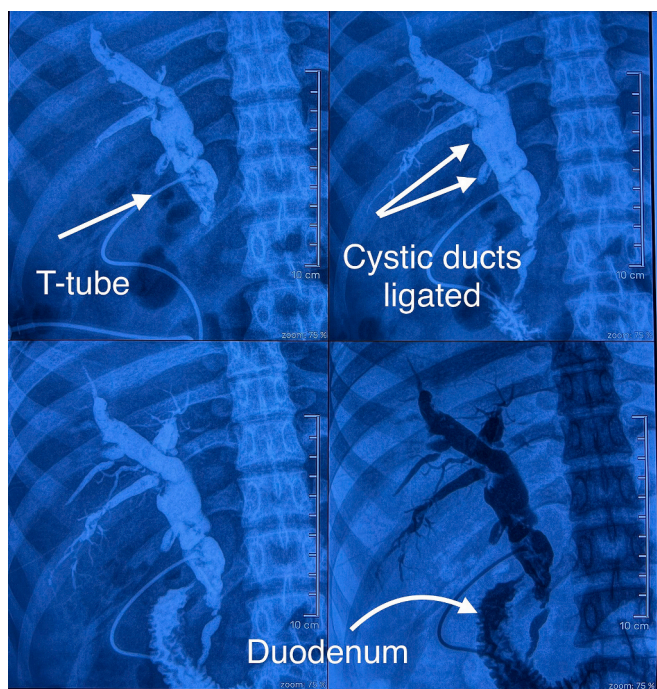


Fig. 4. Intraoperative cholangiography. Dilatation of the intra- and extrahepatic bile duct was observed, with a 20-mm common bile duct and passage of the contrast medium to the duodenum.

tolerated feeding. On the fifth postsurgical day, her bilirubin increased. T-probe control cholangiography revealed dilatation of the bile duct with a filling defect at the distal level without passage of the contrast medium to the duodenum (Fig. 5). She was referred for an ERCP with

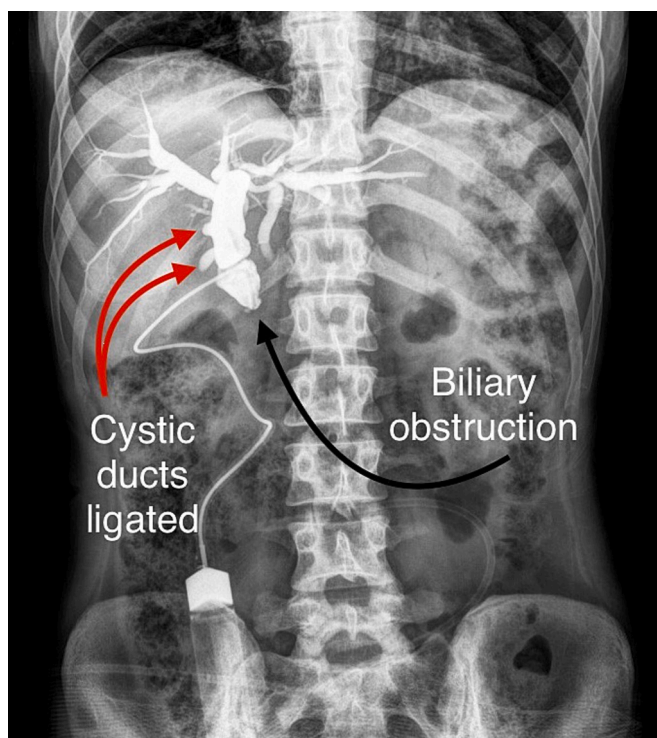


Fig. 5. Control cholangiography. The Red arrows: Both cysts ducts ligated. Note that the contrast medium did not pass to the duodenum. The patient was sent for ERCP. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

endoprosthesis placement. During the ERCP, a type 1 papilla was observed. During the sphincterotomy, multiple stones measuring 5 mm were identified, and the common bile duct was dilated up to its retro-pancreatic portion, reaching a diameter of 25 mm. The choledocholithiasis was successfully resolved, and an Amsterdam-type stent was placed. She continued to be followed by outpatient consultations and was also followed up by the rheumatology service for her lupus.

3. Discussion

Anatomical variants of the bile duct are common, with a reported prevalence of between 52.9 % and 58 % in the general population, underscoring its clinical relevance [1]. However, even more remarkable is the rarity of double cystic duct variants. According to a review of the literature, only 22 cases have been reported, as detailed in Table 1. It is reported more frequently in the female population, with an incidence of 78 % [4,7]. The age of presentation in 65 % of the patients is older than 50 years, and it has never been reported in anyone under 30 years old. In our case, the patient was a 22-year-old woman. Flannery and Caster classified the cystic doubles into three groups according to the configuration of the ducts, type Y, H and trabecular, type H being the most common, where the ducts drain separately to the right, left, or common liver [8]. Much like our case, however, a new case has recently been reported where a new configuration called inverse Y is described: a cystic duct of a single gallbladder diverges to form two cystic ducts that drain into the CBD separately [9].

These variations are a significant challenge for surgeons during surgical intervention since 86 % of patients are diagnosed intra-operatively, making it harder to dissect the hepatocystic triangle, thus highlighting the importance of maintaining a universal culture of safety during cholecystectomy [10]. In this way, the likelihood of intra-operative complications, such as disruption of the biliary tract, can be reduced, which undoubtedly contributes to a high rate of postoperative morbidity and mortality.

Although a tomographic study or magnetic resonance cholangiography would have been of great utility before the cholecystectomy [11]. They were not available in our unit during the intervention due to technical failures that were beyond our control.

As we have mentioned, it is rare to find this anatomical variant, but surgeons must keep it in mind, and given the uncertainty of the anatomy, before sectioning a canal, it is worth doing intraoperative imaging such as cholangiography, which is helpful and is used in 75 % of reported cases [12].

4. Conclusions

Although cystic duct variations are extremely rare, when the anatomical configuration of the bile duct is in doubt, it is valuable to resort to tools such as intraoperative cholangiography. This technique allows the biliary anatomy to be visualized more clearly and to make informed decisions before proceeding with the ligation of the ducts, avoiding any disruption of the bile duct.

Consent

Informed consent was taken from the patient for the publication of this report.

Ethical approval

Ethical review and approval were not required for the study the case report participants in accordance with the local legislation and institutional requirements.

The patients/participants provided their written informed consent to participate in this study. Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable

Table 1
Literature review of reported cases of double cysts [4].

Case	Author	Age/ gender	Country	Duplication type	Preoperative CPRE	Diagnosis	Operative approach	IOC
1	Perelman 1961	56/Female	USA	"H" type	No	Intraoperative	Not reported	Not reported
2	Senapati and Wolf 1984	56/Male	UK	Trabecular type	No	Intraoperative	Open	Yes
3		55/Female		"H" type	No	Intraoperative	Open	Yes
4	Nakasugi et al. 1995	50/Female	Japan	"Y" type	Yes	Preoperative ERCP	Laparoscopic	Yes
5	Ng et al. 1996	60/Male	Hong Kong	"H" type	Yes	Intraoperative	Laparoscopic converted to open	No
6	Momiyama et al. 1996	66/Female	Japan	"H" type	Yes	Postoperative	Laparoscopic	Yes
7	Hirono et al. 1997	74/Female	Japan	"H" type	Yes	Intraoperative	Laparoscopic	Yes
8	Fujikawa et al. 1998	70/Female	Japan	"H" type	Yes	Intraoperative	Open	Yes
9	Lobo et al. 2000	49/Female	Brazil	"Y" type	No	Intraoperative	Laparoscopic	Yes
10	Tsutsumi et al. 2000	74/Female	Japan	"H" type	Yes	Preoperative ERCP	Laparoscopic	Yes
11	Shivhare et al. 2002	46/Female	India	"H" type	No	Intraoperative	Laparoscopic converted to open	Yes
12	Huston et al. 2008	43/Female	USA	"H" type	No	Intraoperative	Laparoscopic	Yes
13	Aristotle et al. 2011	50/Male	India	"Y" type	NA	Postmortem	NA	NA
14	Shih et al. 2011	37/Male	Taiwan	"Y" type	No	Intraoperative	Laparoscopic	No
15	Shabanali et al. 2014	50/Female	Iran	"H" type	No	Intraoperative	Laparoscopic	No
16	Otaibi et al. 2015	54/Male	USA	"H" type	No	Intraoperative	Laparoscopic	Yes
17	Samnani et al. 2015	34/Female	Pakistan	"Y" type	No	Intraoperative	Laparoscopic	No
18	Fujii et al. 2017	57/Female	Japan	Trabecular type	Yes	Preoperative ERCP	Laparoscopic	Yes
19	Salih et al. 2017	33/Female	Iraq	"Y" type	No	Intraoperative	Laparoscopic	No
20	Semere et al. 2019	34/Female	USA	Trabecular type	No	Intraoperative	Laparoscopic converted to open	Yes
21	Suhyun Lim et al. [9]	82/Female	Japan	"H" type	No	Intraoperative	Laparoscopic	Yes
22	Zhi Liang et al. 2022 [3]	66/Female	China	"Reverse Y"	Yes	Intraoperative	Laparoscopic	Yes
23	Carlos J. 2024 Present Case	22/ Female	México	"H" type	No	Intraoperative	Open	Yes

Abbreviations: ERCP, endoscopic retrograde cholangiopancreatography.
IOC, intraoperative cholangiogram.

images or data included in this article.

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