Radiology Case Reports 18 (2023) 1502–1506

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

Successful radiological detection and surgical management of type 3 choledochocele: A case report ☆,☆☆

Oadi N. Shrateh, MD[a,*], *Abdullatif khader, MD*[b,c], *Asil Musleh, MD*[d], *Khaled Abbadi, MD*[d], *Malvina Asbah, MD*[d], *Nihal Sawalha, MD*[b,d], *Afnan W.M. Jobran, MD*[a]

[a] *Medical Research Club, Faculty of Medicine, Al-Quds University, Jerusalem, Palestine*
[b] *Faculty of Medicine, Al-Quds University, Jerusalem, Palestine*
[c] *General Surgery Department, Palestinian Medical Complex, Ramallah, Palestine*
[d] *Department of General Surgery, Palestinian Medical Complex, Ramallah, Palestine*

ARTICLE INFO

Article history:
Received 29 December 2022
Revised 5 January 2023
Accepted 7 January 2023

Keywords:
Endoscopic retrograde cholangiopancreatography
Choledochocele
Operative surgical procedure
Choledochal cyst

ABSTRACT

According to Todani's classification, a choledochocele is a cystic dilatation of the distal section of the common bile duct that protrudes into the duodenal lumen. It is also known as a type III choledochal cyst. There are not many cases, and the cause is still unknown. Typically, it is misdiagnosed as a peptic ulcer, as was the case with the patient in this case study. For choledochocele diagnosis, multislice spiral computed tomography and magnetic resonance cholangiopancreatography may be equivalent to endoscopic retrograde cholangiography. Both endoscopic therapy and open surgical care are risk-free possibilities, and the cyst's size influences which strategy should be used. A 35-year-old woman admitted to the hospital with upper abdominal pain caused by a large choledochocele was successfully treated by open surgical management. In this case study, we outline the specifics of her situation and talk about the most recent research on similar cases and how to treat them therapeutically. There is ongoing debate regarding the best course of action. However, to achieve a successful outcome, open surgical care for choledochocele of large size (ie, > 3 cm in diameter) is advised.

☆ Funding: This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.
☆☆ Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.
* Corresponding author.
E-mail address: oadi.shrateh@students.alquds.edu (O.N. Shrateh).
https://doi.org/10.1016/j.radcr.2023.01.029

Introduction

Choledochocele is a cystic dilation of the distal section of the common bile duct protruding into the duodenal lumen, also known as a type III choledochal cyst according to Todani's classification [1]. Less than 2% of all choledochal cyst instances have been reported, making them rare [2]. The disease's rarity makes it difficult to diagnose and treat.

Case presentation

A married, nonsmoker 35-year-old female patient presented to our surgical outpatient clinic with the chief complaint of recurrent right upper quadrant and epigastric pain. The pain is gradual in onset, progressive, intermittent, radiating to the back, and related to fatty foods of years' duration. It is associated with nausea and occasional vomiting of regular gastric content. The patient has a free past medical history and claims a surgical history of laparotomy for a congenital upper GI anomaly at the age of 2 days, with no reports. The patient also had an open cholecystectomy 9 years ago and a cesarean section.

The patient sought medical advice in an outpatient clinic for the last 9 years following a cholecystectomy, with recurrent visits for the same complaint. Investigations over these years were all within normal range except for an upper GI endoscopy that showed severe duodeno-gastric (bile) reflux with moderate to severe mucosal erythema. A large bulb in D2 with mild luminal stenosis but a free passage into D3 and D4, soft in consistency, a positive pillow sign. Therefore, the patient was referred to our hospital for further evaluation. Laboratory tests at the time revealed hemoglobin levels of 12.7 g/dL (normal range 13.5-17 gm/dL), white blood cells of 6 k/μL (normal range 4.6-11 k/μL), platelets of 237 k/μL (normal range 150-450 k/μL), creatinine of 0.6 mg/dL (normal range 0.7-1.2 mg/dL), alanine aminotransferase of 16 IU/L (normal range 0-41 IU/L), aspartate aminotransferase of 13 IU/L (normal range 0-50 IU/L), alkaline phosphatase of 40 U/L (normal range 40-130 U/L), direct bilirubin of 0.2 μmol/L (normal range 0.0-0.3 μmol/L), total bilirubin of 0.3 μmol/L (normal range 0.0-1.2 μmol/L), and amylase of 42 U/L (normal range 40-130 U/L).

A CT scan showed a distended duodenum due to a cystic structure measuring about 6.4 cm and filled with stones (Fig. 1). The complement evaluation by oral contrast confirms the absence of connection with the bowel loops. Magnetic resonance cholangiopancreatography showed an extrahepatic cystic structure filled with numerous stones not connected to Common Bile Duct (CBD) (Fig. 2). The first Endoscopic retrograde cholangiopancreatography (ERCP) showed a large bulge at the medial walls of D1 and D2. External compression caused displacement of the ampulla of Vater into an inaccessible and difficult-to-identify position.

The cholangiogram showed a large cystic dilation distally, most likely representing a cystic dilation of the distal bile duct (choledochal cyst?) full of stones. The relation of this cystic structure to the proximal biliary system couldn't be assessed. The proximal biliary system could not be accessed. To ensure adequate bile drainage, a 7*7 cm-long plastic stent was inserted into the dilated cyst. The second ERCP was done for a second opinion and showed a large cystic structure with many stones. The previous stent was in place with the diagnosis of choledochal cyst type I. EUS showed a large cystic structure filled with many stones. The biliary stent was in place in continuation with the dilated CBD. Most consistent with choledochal cyst type I. A multidisciplinary team opinion advised that the patient should undergo surgery. Intraoperative findings revealed severe adhesions, for which adhesiolysis was performed. It also showed disrupted anatomy due to the previous laparotomy in infancy. The upper GI tract was abnormal. All of the duodenum parts were completely intra-abdominal, with no retroperitoneal extensions. There was an absence of the Treitz ligament with the duodenojejunal anastomosis. Neither a cystic mass nor dilatation was discovered during CBD exploration and palpation. A cystic mass filled with stones was palpated in the duodenum, and a duodenotomy was done. The stent, which was inserted previously in the ERCP, was found protruding through a posteromedial orifice from a normal-looking duodenal mucosa, and surrounding it was a mucosal bulge filled with many stones. An opening was made around the stent, and stones were extracted.

Choledochocele type IIIB was diagnosed intraoperatively and managed accordingly. After inserting a new bile duct stent and a pancreatic stent, and then closing the duodenotomy, the

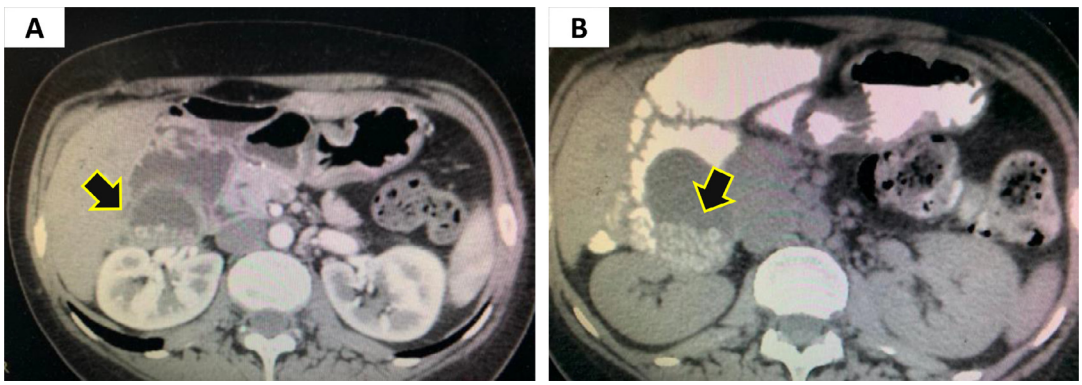

Fig. 1 – (A and B) A CT scan showed a distended duodenum due to a cystic structure measuring about 6.4 cm and filled with stones. The complement evaluation by oral contrast confirms the absence of connection with the bowel loops (arrows).

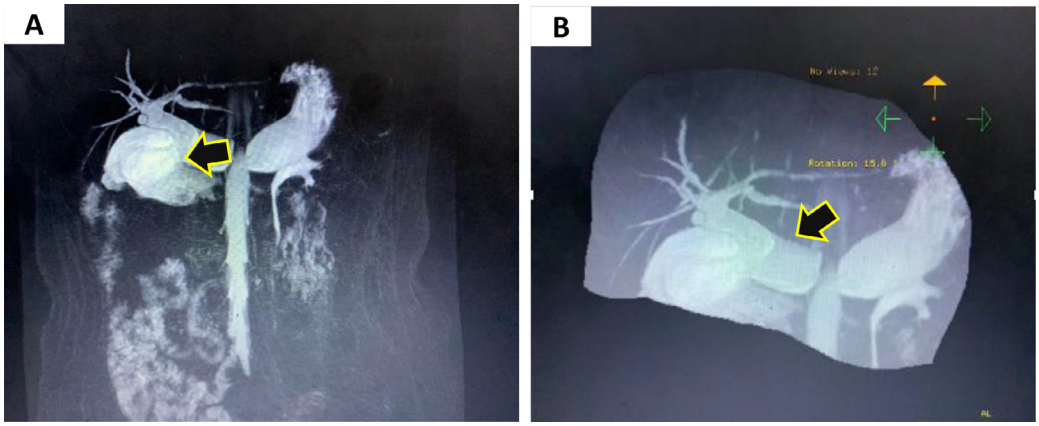

Fig. 2 – (A and B) An magnetic resonance cholangiopancreatography revealed an extrahepatic cystic structure containing numerous stones unrelated to Common Bile Duct (arrows).

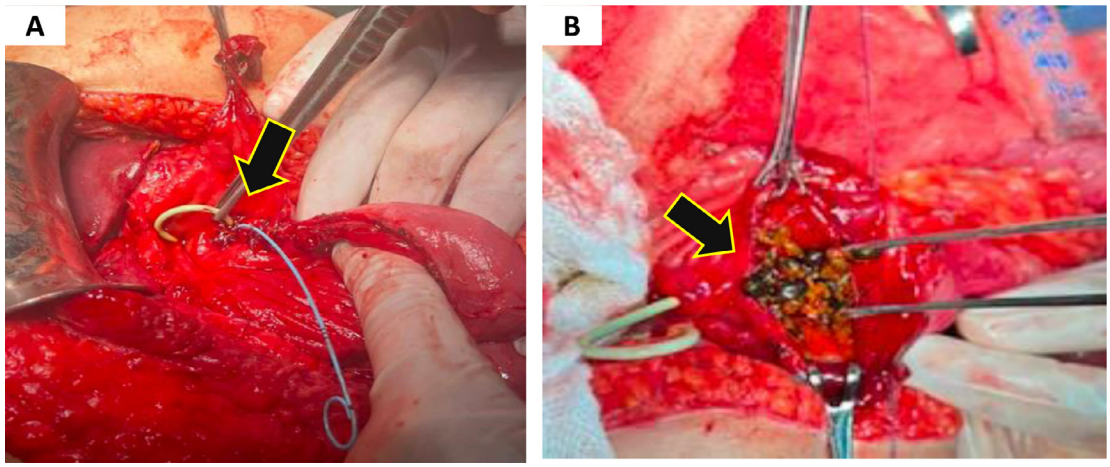

Fig. 3 – (A and B) Intraoperative images. The diagnosis of choledochocele type IIIB was made intraoperatively and treated accordingly, with complete cyst excision and marsupialization of the distal biliary orifice after inserting a new bile duct stent and pancreatic stent and then closing the duodenotomy.

cyst was completely excised and the distal biliary orifice marsupialized, as shown in Figure 3.

On follow-up, the patient was doing well, asymptomatic, with excellent outcomes and rapid resolution of her symptoms. The patient reported passage of the previously inserted stent intraoperatively with stool. A follow-up ERCP was done and showed a normal cholangiogram. Histopathology reported a diagnosis of choledochocele with no evidence of malignancy.

Discussion

Wheeler [3] reported the first choledochocele case in 1915. According to the report, the anomaly was a little, tight cyst that was located in or close to the common bile duct's orifice [3]. Since then, the number of papers on choledochocele has progressively expanded with the advancement of medical tools like endoscopy and imaging.

Choledochocele's etiology is still unknown. Tanaka [4] proposed that the failure of a bile duct to retreat during development was the cause for creation. Even though some choledochocele appears to be acquired, Sterling [5] hypothesized that papillary stenosis or sphincter of Oddi failure may obstruct bile flow, causing a rise in distal bile duct pressure, which may then evaginate into the duodenum. In their analysis of 24 patients, Sarris and Tsang [6] found that the choledochocele's inner epithelium was duodenal mucosa in 63% of the cases and biliary tract epithelium or unclassifiable glandular epithelium in 37% of the instances.

It is debatable in histology whether choledochocele is a particular kind of duodenal duplication cyst or a distinct entity because of the presence of intestinal mucosa. Because the majority of cases involve duodenal mucosa, congenital intraluminal duodenal diverticulum has been proposed as a possi-

ble factor in the pathophysiology of choledochocele in young infants. Contrary to congenital forms, papillary stenosis may lead to ampulla dilatation in elderly patients through an inflammatory response brought on by bile and pancreatic juice stasis. As a result, undifferentiated and biliary tract epithelium predominate later [7].

The most typical symptom of choledochocele is abdominal pain. Other symptoms include nausea, vomiting, and jaundice. The diagnosis depends heavily on the image examination. Cystic masses in the duodenum cavity can be found with a noninvasive B-ultrasound examination; however, intestinal gas may cause the image to be distorted. As in our case, there were typically no specific findings. Endoscopic ultrasound may be used to classify the lesion and direct treatment choices [8]. It may also highlight the cyst's wall layers and its link to the pancreaticobiliary tree. Some believe that ERCP is the most effective way to confirm the presence of a choledochocele.

In 67% of the cases that have been reported in the literature, ERCP has been done, and this technique has the advantage of simultaneous treatment implementation. Due to their similar specificity and sensitivity, multislice spiral computed tomography and magnetic resonance cholangiopancreatography may take the place of an ERCP for diagnosis [9]. For the best image of our patient, endoscopic and radiologic methods were both used.

Wheeler successfully performed open surgical therapy on the original choledochocele case (a 65-year-old male patient), and it is still regularly done today. This includes transduodenal total cyst removal with sphincteroplasty. Mane et al. [10] performed the first endoscopic sphincterotomy procedure on a patient in 1974 who was a 21-year-old girl. Endoscopy has since gained recognition as a workable alternative therapy with positive outcomes. Pediatric patients have effectively received both treatments [4].

The size of the cyst influences the clinical decision-making process, but the choice of therapeutic approach is still up in the air. The threshold at which transduodenal cyst excision should be done may be 3 cm in diameter [11]. However, the primary goal of choledochocele treatment should be to preserve normal pancreatic and bile excretion, with the secondary goal of reducing the risk of cancer. We chose to undergo open surgery because of the patient's enormous cyst lesion's diameter. The majority of patients were alleviated following therapy, whether it was endoscopy or surgery. Rare reports of choledochocele malignancy [12] have been made. However, pancreaticobiliary maljunction may accompany or cause choledochocele; in this case, biliary tract cancer is more common, according to Horaguchi's report [9]. Those who are asymptomatic or unintentionally discovered, on the other hand, may benefit from additional monitoring.

Conclusion

In conclusion, choledochocele is a rare condition, and there is ongoing debate regarding the best course of action. According to the successful outcome of our case, we advise open surgical care for choledochocele of large size (ie, >3 cm in diameter).

Author contribution

Writing the manuscript: Asil Musleh, Afnan W.M. Jobran, Oadi N. Shrateh; Imaging description: Nihal Sawalha, Khaled Abbadi, Malvina Asbah; Reviewing & editing the manuscript: Oadi N. Shrateh, Abdullatif khader.

Authorship

All authors attest that they meet the current ICMJE criteria for authorship.

Patient consent

Written informed consent to publish this case and use anonymized radiologic material was obtained from the patient.

REFERENCES

[1] 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;134(2):263–9.
[2] Yamaguchi M. Congenital choledochal cyst: analysis of 1,433 patients in the Japanese literature. Am J Surg 1980;140(5):653–7.
[3] de Courcy Wheeler W. An unusual case of obstruction to the common bile-duct (choledochocele?). Br J Surg 1940;27(107):446–8.
[4] Yang J, Xiao G-F, Li Y-X. Open surgical treatment of choledochocele: a case report and review of literature. World J Clin Cases 2018;6(14):842.
[5] Sterling JA. Diverticula in the terminal portion of the common bile duct. Am J Pathol 1949;25(2):325.
[6] Sarris G, Tsang D. Choledochocele: case report, literature review, and a proposed classification. Surgery 1989;105(3):408–14.
[7] Schimpl G, Sauer H, Goriupp U, Becker H. Choledochocele: importance of histological evaluation. J Pediatr Surg 1993;28(12):1562–5.
[8] Law R, Topazian M. Diagnosis and treatment of choledochoceles. Clin Gastroenterol Hepatol 2014;12(2):196–203.
[9] Horaguchi J, Fujita N, Kobayashi G, Noda Y, Ito K, Takasawa O. Clinical study of choledochocele: is it a risk factor for biliary malignancies? J Gastroenterol 2005;40(4):396–401.
[10] Deyhle P, Schnaars P, Meyer HJ, Nüesch HJ, Akovbiantz A. Electrosurgical removal of a choledochocele through an endoscope introduced by mouth (author's transl). Dtsch Med Wochenschr (1946). 1974;99(3):71-2.

[11] Lobeck IN, Dupree P, Falcone RA Jr, Lin TK, Trout AT, Nathan JD, et al. The presentation and management of choledochocele (type III choledochal cyst): a 40-year systematic review of the literature. J Pediatr Surg 2017;52(4):644–9.

[12] Ladas SD, Katsogridakis I, Tassios P, Tastemiroglou T, Vrachliotis T, Raptis SA. Choledochocele, an overlooked diagnosis: report of 15 cases and review of 56 published reports from 1984 to 1992. Endoscopy 1995;27(03):233–9.