International Journal of Surgery Case Reports 92 (2022) 106912

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

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

Case report

Recurrent cholangitis after choledochoduodenostomy: A case report

Julianus Aboyaman Uwuratuw [a,b,*], Bustaman Bakhtiar [c], Ibrahim Labeda [a], Erwin Syarifuddin [a], Robert Christeven [c], Muhammad Faruk [c]

[a] Division of Digestive, Department of Surgery, Faculty of Medicine, Hasanuddin University, Makassar, Indonesia
[b] Division of Digestive, Department of Surgery, Primaya Hospital, Makassar, Indonesia
[c] Department of Surgery, Faculty of Medicine, Hasanuddin University, Makassar, Indonesia

ARTICLE INFO

Keywords:
Cholangitis
Choledochoduodenostomy
Endoscopic retrograde cholangiopancreatography
Roux-en-Y
Case report

ABSTRACT

Introduction: Recurrent cholangitis is a long-term consequence of choledochoduodenostomy (CDD) that requires urgent treatment. The frequency of recurrent cholangitis ranges between 2.5 and 15.7%. This case demonstrated the importance of rapid and precise diagnosis through screening and therapeutic modalities in recurrent cholangitis.

Presentation of case: A male patient presented with a history of recurring right upper abdominal discomfort during the previous 3 years. The pain had been intermittent but had become more intense during the prior month. The patient was diagnosed with recurrent cholangitis following CDD. The therapeutic plan was closure of the CDD, which was identified as the cause of the recurrent cholangitis, and biliary drainage by modified Roux-en-Y choledochojejunostomy.

Discussion: Recurrent cholangitis was diagnosed based on clinical manifestations, including recurring right upper abdomen discomfort, jaundice, and fever accompanied with consistent laboratory and imaging findings. Drainage of bile into the distal common bile duct (CBD) is reduced in the side-to-side CDD arrangement. Consequently, the distal CBD becomes a reservoir with inadequate drainage, predisposing this so-called 'sump' to debris accumulation and cholangitis. The surgery was considered successful in preventing the recurrent cholangitis.

Conclusion: The recurrent cholangitis was occurred due to inadequate biliary drainage. The choledochojejunostomy procedure with modified Roux-en-Y might prevent the recurrent cholangitis by improving biliary drainage to the enteric.

1. Introduction

Acute cholangitis is a bacterial infection of the biliary system that is most often caused by blockage of the bile duct or hepatic duct. The diagnosis is based on characteristic clinical symptoms, abnormal laboratory values indicating biliary infection and blockage, and abnormal findings upon imaging investigations. When therapy is delayed, mortality increases. The mortality rate of acute cholangitis ranges from 5 to 10%, with a higher mortality rate in patients who require emergency biliary decompression or surgery [1,2].

Recurrent cholangitis manifests with symptoms such as right upper abdomen discomfort, jaundice, and fever. This disease results from long-term problems and seldom occurs after side-to-side choledochoduodenostomy (CDD). After CDD, the frequency of recurrent cholangitis ranges between 2.5 and 15.7%. Although endoscopic retrograde cholangiopancreatography (ERCP) has mostly supplanted CDD in recent years, it still has specific long-term effects and difficulties [3].

The side-to-side CDD design reduces bile outflow into the distal common bile duct (CBD). Consequently, the distal part of the CBD develops into a reservoir with inadequate drainage, making this so-called 'sump' susceptible to debris build-up, free air, and cholangitis. As a result, therapy begins with antibiotics and side-to-side CDD termination, followed by Roux-en-Y modification of the biliary-enteric bypass [4].

Here we report a case of recurrent cholangitis after CDD according to the 2020 Surgical CAse REport (SCARE) guidelines [5,6].

* Corresponding author at: Division of Digestive, Department of Surgery, Faculty of Medicine, Hasanuddin University Makassar, Jalan Perintis Kemerdekaan KM 11, Makassar, South Sulawesi 90245, Indonesia.
E-mail addresses: boyuwuratuw@gmail.com (J.A. Uwuratuw), bustaman06@gmail.com (B. Bakhtiar), ibrlabeda@yahoo.com (I. Labeda), erwinsyarifuddin@yahoo.com (E. Syarifuddin), rchristeven.rc@gmail.com (R. Christeven), faroex8283@gmail.com (M. Faruk).

https://doi.org/10.1016/j.ijscr.2022.106912
Received 19 January 2022; Received in revised form 27 February 2022; Accepted 27 February 2022
Available online 1 March 2022
2210-2612/© 2022 The Author(s). Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

2. Presentation of case

A 34-year-old man was hospitalized with chief complaint of recurrent symptoms of fever accompanied with intermittent right upper quadrant pain, occasional mild jaundice, and vomiting, which he had suffered almost every month for 3 years. The symptoms had worsened and got more frequent since last three months, which he had general weakness, loss of appetite, and weight loss. He had been admitted to his hometown rural hospital due to the same symptoms for more than 4 occasions and was treated frequently by an internist. The symptoms were improved after several days of intravenous antibiotic administration in a rural hospital. After the symptoms had worsened, the patient was referred to the digestive surgery department in our hospital in the provincial capital city. For the last three months, jaundice had been more prominent. It was known that he had a history of a total bilirubin value of 4 mg/dL in his last hospital admission. He was treated with intravenous fluid resuscitation and vasoconstrictor to overcome his hemodynamic shock. Meropenem, a broad spectrum antibiotic, was administered thrice daily for 7 day to resolve the infection. We planned to perform a percutaneous transhepatic cholangiogram for diagnosis and also to perform emergency biliary drainage because the patient was unstable for major surgery. However, the patient was allergic to contrast agent. Nevertheless, he improved after 10 days of treatment. The patient was discharged from the hospital and was regularly examined in our outpatient department.

The patient had history of biliary surgery 32 years ago due to an unknown cause (when he was 2 years old). The detail information of prior operation history was unclear. The patient had no any other comorbidity.

The World Health Organization (WHO) performance status was grade 1 and the patient was hemodynamically stable upon physical examination. No jaundice was found. On abdominal physical examination, we found tenderness at epigastric and hypochondriac region. No palpable mass was discovered. From the laboratory test, we found the hemoglobin was 13.3 g/dL, white blood count (WBC) was 12.100/uL with Neutrophil 72.6%, thrombocyte count was 369.000/uL. His liver enzyme function test was 46 U/L and the coagulation was normal with the Prothrombin Time (PT) 14.5 s, international normalized ratio (INR)

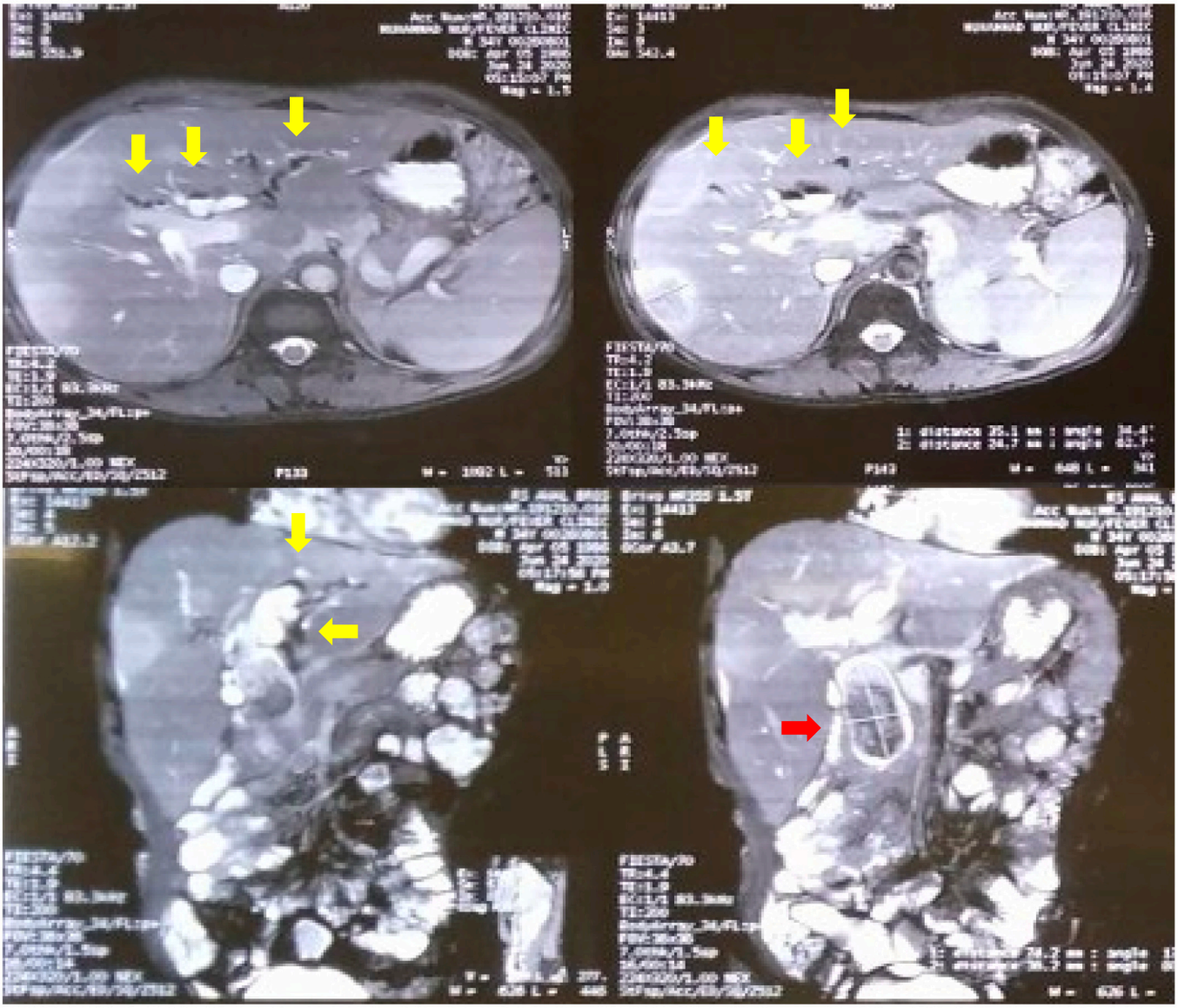

Fig. 1. MRCP findings of pneumobilia (yellow arrow), accumulation of debris, stones, and static bile mimicking a duodenal mass (red arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

1.19, activated partial thrombin time (aPTT) 35.9, total bilirubin 2.0 mg/dL, direct bilirubin 1.5 mg/dL, and albumin 2.5 mg/dL. Pneumobilia and a duodenal mass were revealed by magnetic resonance cholangiopancreatography (MRCP) (Fig. 1). Neither biliary three dilatation nor biliary stone was discovered. Biliary enteric fistula and duodenal tumor were suspected. Another sophisticated examination, Hepatobiliary Iminodiacetic Acid Scan (HIDA) was unavailable in our center.

This patient was diagnosed with recurrent cholangitis based on anamnesis, physical examination, and MRCP. As a result, the main goal of therapy was to keep the infection under control by providing the broad-spectrum antibiotic meropenem at 1 g every 8 h intravenously and underwent biliary drainage surgery. The exploratory laparotomy operation was conducted. Bilateral subcostal incision was performed to provide excellent operation field. During the operation, we discovered the absence of gallbladder and the appearance of a 2.5 cm side-to-side biliary enteric anastomosis between common bile duct and the second part of duodenum. No gallbladder was detected during an exploratory laparotomy. There was no any tumor in duodenum and biliary duct. Therefore, biopsy was not performed. We decided to terminate the side-to-side biliary enteric anastomosis between CBD and duodenum (Fig. 2). The anastomosis between duodenum and CBD was closed with polyglactin 910 suture 3/0. The surgery was continued to the creation of side to end anastomosis between jejunum and CBD with Roux en Y reconstruction manner (Fig. 3). The CBD was transected, and the distal stump was oversewn with 3–0 absorbable suture. The jejunum was transected at 40 cm from Treitz ligament, and a Roux limb of 50 cm was passed in a retrocolic fashion through the avascular portion of the transverse mesocolon to the right of the middle colic artery. The posterior wall of the duct was sutured to the jejunum with a continuous (simple running) polyglactin 910 suture. The anterior portion of the anastomosis was also completed using the continuous suture. The diameter of the new choledochojejunostomy was approximately 2 cm. Furthermore, the jejunojejunostomy was constructed with stapler at the level of 40 cm from the choledochojejunostomy anastomosis (Fig. 4). On the second day after surgery, the patient began a water diet. He was subsequently placed on an unrestricted diet on the sixth day. Because the outpatient's overall health was satisfactory, the drain output was low, and there were no indications of peritonitis, it was determined that there was no evidence of anastomotic leakage on the ninth day. One month following surgery, the patient was admitted to the polyclinic in good general health, without stomach discomfort, jaundice, or fever.

3. Discussion

The recurrent cholangitis is a complication of a side-to-side choledochoduodenostomy (CDD), which may be caused by ascending infection. The lack of awareness of this condition and infrequent post operation follow-up may lead to sepsis. The side-to-side CDD is a prominent procedure for the drainage of the CBD and is usually performed in the setting of multiple calculi or biliary sludge in the CBD or dilated CBD (lumen diameter of more than 15 mm).

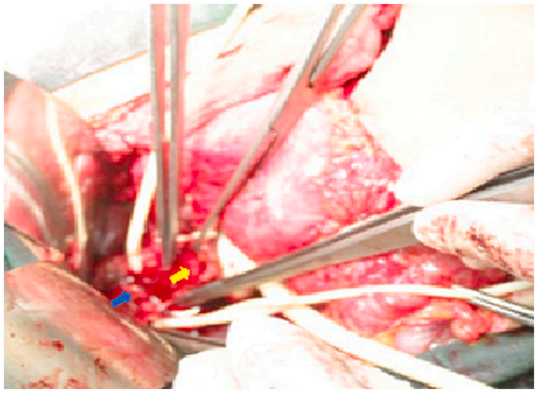

Fig. 2. Side-to-side anastomosis between CBD (blue arrow) and duodenum (yellow arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

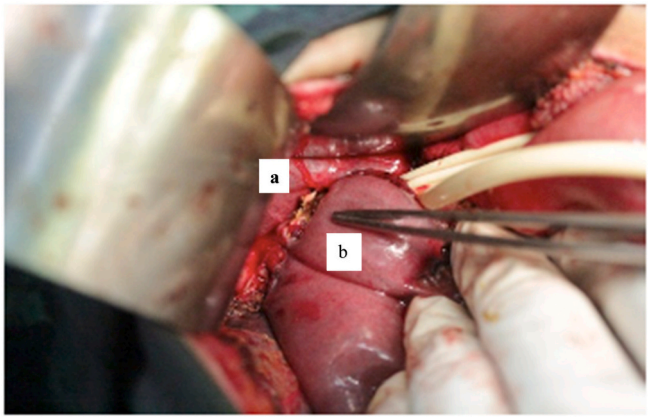

Fig. 3. Side-to-end anastomosis between the jejunum (a) and CBD (b).

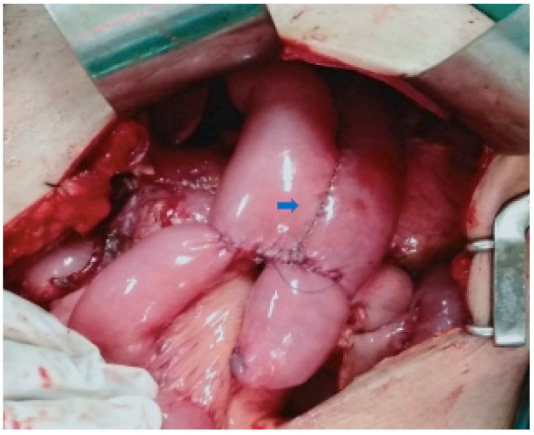

Fig. 4. Jejunojejunostomy (blue arrow) with a 40-cm Treitz ligament. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

In the configuration of a side-to-side CDD, the drainage of bile to distal CBD is decrease. Consequently, the distal part of the CBD alters into a poorly drained reservoir, causing this so called “sump” becomes prone to accumulation of debris [10]. Marbet et al. described that the most likely primary pathophysiological is the low filling pressure in the distal choledochus inflicted by the well-functioning anastomosis which interrupts normal distal peristalsis and drainage, thus predisposing to reflux of duodenal contents into the CBD through anastomoses, leading to the formation of a sump with debris, calculi, and bacterial overgrowth in the inefficiently drained limb [7].

Recurrent cholangitis was diagnosed based on several symptoms, including recurring right upper abdomen discomfort, jaundice, and fever. The patient had experienced several occasions of mild acute cholangitis which improved after administration of broad-spectrum intravenous antibiotic. However, the acute cholangitis was severe on the last occasion which led the patient to septic shock. It was determined that this patient had acute cholangitis based on the 2018 Tokyo Guidelines criteria [8], which include the presence of indications of inflammation in leukocytes >10,000 per microliter (12,100 per microliter in this case) and jaundice total bilirubin ≥2 mg/dl (2.0 mg/dl in this case). In addition, pneumobilia and accumulation of debris were

discovered distal to the anastomosis (sump syndrome) on MRCP. The pneumobilia seemed to be caused by air insufflation from the duodenum through the preceding CDD anastomosis activity [3,9]. Thus, the diagnosis was based on clinical, laboratory, and imaging findings: namely, recurrent cholangitis following CDD, pneumobilia, and accumulation of debris. The severity of acute cholangitis is evaluated once a diagnosis of acute cholangitis is confirmed. Therefore, mild cholangitis was the diagnosis for this patient according to the TG18 criteria because only leukocytosis was found.

Antibiotic therapy and biliary drainage improvement are the primary treatment approaches. Antibiotic treatment is suggested for 4–7 days but may be extended to 2 weeks if endocarditis problems occur. For patients with specific comorbidities, post-CDD recurrent cholangitis is treated with piperacillin/tazobactam, ceftazidime + metronidazole, or meropenem with metronidazole after anastomotic surgery. Antibiotics were administered to this patient through intravenous meropenem 1 g every 8 h [10].

The principal treatment approach is to enhance the biliary drainage of the CBD distal to the anastomosis either by ERCP or by surgery. The initial treatment of cholangitis induced by sump syndrome usually begins with endoscopic sphincterotomy in order to decompress the CBD distally at the level of the Vater ampulla. If the ERCP management fails because of the presence of heavy gall stone burden, the existence of biliary stricture, the passage of food particles through the anastomosis into the CBD which cannot be cleared through a large enough sphincterotomy, the treatment of choice shifts to the surgical options include revision of the CDD to a Roux-en-Y hepaticojejunostomy, with resection of the distal portion of the CBD.

ERCP and open surgery may be used to conduct further procedures. ERCP is an endoscopic technique used to diagnose structures, most notably CDD anastomosis. It is most often performed on CBDs with a diameter < 15 mm. However, ERCP was not carried out in this instance since a diagnosis of sump syndrome was confirmed, and the treatment concepts include cleaning, removing, and reattaching the distal stump. Thus, open surgery, namely choledochojejunostomy, was the preferred method [11].

Biliary drainage with the Roux-en-Y technique is associated with a low incidence of morbidity and death and a success rate of 60–70%. In addition, numerous studies have indicated that patients had no postoperative complications. However, problems from anastomotic strictures have been reported in 10–25% of postoperative patients with recurrent cholangitis [12].

During our surgery, we discovered the side-to-side CDD anastomosis from prior surgery incidentally. However, the indication of prior CDD anastomosis surgery was unknown (the patient did not know the prior surgery procedure in his childhood). We decided to terminate the side-to-side CDD and created a new biliary-enteric bypass between the end of CBD and the anti-mesentric side of jejunum. The choledochojejunostomy procedure was chosen over hepaticojejunostomy due to its convenience, less invasiveness, and preservation of the more proximal bile duct (hepatic duct), so that a more proximal biliary-enteric bypass can be performed in the event of stenosis in choledochojejunostomy. The cause of cholangitis in this case was not related to any malignancy, so we did not resect the ductus choledochus.

The choledochojejunostomy with the configuration of the end-to-side technique should be begun with cholecystectomy with cystic duct ligation is performed, followed by ligation of the CBD. The duct is opened at the level of the planned anastomosis. The remnant of the CBD is transected, and the distal stump is oversewn with 3–0 absorbable suture. A 50- to 70-cm Roux-en-Y limb of jejunum is passed through retrocolic, placed at the right side of the middle colic vessels, and positioned to reside adjacent to the proximal bile duct in a tension-free manner. The posterior wall of the duct is stitched to the jejunum with a running 3–0 or 4–0 absorbable suture. The tail of the suture and needle are left intact. A jejunotomy is fashioned along the duct, and single interrupted 3–0 or 4–0 absorbable sutures are used to approximate the mucosa of jejunum to the mucosa of duct with the knots tied on the inside of the lumen. The anterior portion of the anastomosis is then completed using the running suture used to construct the posterior row. With this kind of biliary-enteric anastomosis configuration, we believed the biliary drainage was improved and the sump syndrome has been resolved. During the surgery, we did not perform swab for culture to avoid ambiguous interpretation due to previous administration of broad spectrum antibiotic and contamination of normal flora from bowel content.

After the choledochojejunostomy surgery with modified Roux-en-Y, the patient did not complain of stomach discomfort, jaundice, or fever, and the laboratory results were improved as expected. However, long-term evaluation was needed to assess the outcome of our surgery.

The rate of postoperative problems after significant alterations in cholangitis with medication and surgical treatments is about 2.6–5%.

4. Conclusion

The recurrent cholangitis was occurred due to inadequate biliary drainage. Therefore, the principal treatment approach is to enhance the biliary drainage of the CBD distal to the anastomosis either by endoscopy or by surgery. The choledochojejunostomy procedure with modified Roux-en-Y could prevent the recurrent cholangitis by improving biliary drainage to the enteric.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Funding

No funding or sponsorship

Ethical approval

The study is exempt from ethical approval in our institution.

Consent

Written informed consent was obtained from the patient 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.

Registration of research studies

Not applicable – single case report.

Guarantor

Julianus Aboyaman Uwuratuw and Ibrahim Labeda

CRediT authorship contribution statement

Julianus Aboyaman Uwuratuw: study concept and surgical therapy for this patient. Julianus Aboyaman Uwuratuw, Erwin Syarifuddin, Robert Christeven, and Bustaman Bakhtiar: Data collection and Writing-Original draft preparation. Ibrahim Labeda: senior author and the manuscript reviewer. Robert Christeven and Muhammad Faruk: Editing and Writing. All authors read and approved the final manuscript.

Declaration of competing interest

Nothing to declare.

Acknowledgement

None.

References

[1] Y. Kimura, T. Takada, S.M. Strasberg, H.A. Pitt, D.J. Gouma, O.J. Garden, M. W. Büchler, J.A. Windsor, T. Mayumi, M. Yoshida, F. Miura, R. Higuchi, T. Gabata, J. Hata, H. Gomi, C. Dervenis, W.-Y. Lau, G. Belli, M.-H. Kim, S.C. Hilvano, Y. Yamashita, TG13 current terminology, etiology, and epidemiology of acute cholangitis and cholecystitis, J. Hepatobiliary. Pancreat. Sci. 20 (2013) 8–23, https://doi.org/10.1007/s00534-012-0564-0.

[2] M. Ahmed, Acute cholangitis - an update, World J. Gastrointest. Pathophysiol. 9 (2018) 1–7, https://doi.org/10.4291/wjgp.v9.i1.1.

[3] H. Abraham, S. Thomas, A. Srivastava, Sump syndrome: a rare long-term complication of choledochoduodenostomy, Case Rep. Gastroenterol. 11 (2017) 428–433, https://doi.org/10.1159/000477335.

[4] U. Zeuge, M. Fehr, C. Meyenberger, M.C. Sulz, Mind the sump! - diagnostic challenge of a rare complication of choledochoduodenostomy, Case Rep. Gastroenterol. 8 (2014) 358–363, https://doi.org/10.1159/000369298.

[5] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, A. Kerwan, A. Thoma, A.J. Beamish, A. Noureldin, A. Rao, B. Vasudevan, B. Challacombe, B. Perakath, B. Kirshtein, B. Ekser, C.S. Pramesh, D.M. Laskin, D. Machado-Aranda, D. Miguel, D. Pagano, F. H. Millham, G. Roy, H. Kadioglu, I.J. Nixon, I. Mukhejree, J.A. McCaul, J.Chi-Yong Ngu, J. Albrecht, J.G. Rivas, K. Raveendran, L. Derbyshire, M.H. Ather, M. A. Thorat, M. Valmasoni, M. Bashashati, M. Chalkoo, N.Z. Teo, N. Raison, O. J. Muensterer, P.J. Bradley, P. Goel, P.S. Pai, R.Y. Afifi, R.D. Rosin, R. Coppola, R. Klappenbach, R. Wynn, R.L.De Wilde, S. Surani, S. Giordano, S. Massarut, S. G. Raja, S. Basu, S.A. Enam, T.G. Manning, T. Cross, V.K. Karanth, V. Kasivisvanathan, Z. Mei, The SCARE 2020 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230, https://doi.org/10.1016/j.ijsu.2020.10.034.

[6] A. Warsinggih, I. Fajar, J.A.Uwuratuw Labeda, M.Faruk Prihantono, Bilioenteric bypass stricture type II with hepatolithiasis: a case report, Ann. Med. Surg. (2020), https://doi.org/10.1016/j.amsu.2020.07.011.

[7] J. Virgile, R. Marathi, Cholangitis, in: Treasure Island (FL), 2021.

[8] S. Kiriyama, K. Kozaka, T. Takada, S.M. Strasberg, H.A. Pitt, T. Gabata, J. Hata, K.-H. Liau, F. Miura, A. Horiguchi, K.-H. Liu, C.-H. Su, K. Wada, P. Jagannath, T. Itoi, D.J. Gouma, Y. Mori, S. Mukai, M.E. Giménez, W.S.-W. Huang, M.-H. Kim, K. Okamoto, G. Belli, C. Dervenis, A.C.W. Chan, W.Y. Lau, I. Endo, H. Gomi, M. Yoshida, T. Mayumi, T.H. Baron, E. de Santibañes, A.Y.B. Teoh, T.-L. Hwang, C.-G. Ker, M.-F. Chen, H.-S. Han, Y.-S. Yoon, I.-S. Choi, D.-S. Yoon, R. Higuchi, S. Kitano, M. Inomata, D.J. Deziel, E. Jonas, K. Hirata, Y. Sumiyama, K. Inui, M. Yamamoto, Tokyo Guidelines, Diagnostic criteria and severity grading of acute cholangitis (with videos), J. Hepatobiliary. Pancreat. Sci. 25 (2018) (2018) 17–30, https://doi.org/10.1002/jhbp.512.

[9] J. Jones, F. Gaillard, Pneumobilia, in: Radiopaedia.Org, Radiopaedia.org, 2008, https://doi.org/10.53347/rID-1900.

[10] A. Sokal, A. Sauvanet, B. Fantin, V. de Lastours, Acute cholangitis: diagnosis and management, J. Visc. Surg. 156 (2019) 515–525, https://doi.org/10.1016/j.jviscsurg.2019.05.007.

[11] M.S. Suliman, M.M. Singh, K. Zaheer, S.U. Malik, A. Abu-Hashyeh, Is it really SUMP syndrome? A case report, Cureus (2019), https://doi.org/10.7759/cureus.5837.

[12] K.P. Croome, D.M. Nagorney, Bile duct cysts in adults, in: William R. Jarnagin (Ed.), Blumgart's Surg. Liver, Biliary Tract, Pancreas, Sixth Edit, Elsevier, Philadelphia, 2017, pp. 752–764.