

Choledocho-Duodenal Fistula as a Complication of a Chronic Duodenal Ulcer

Kastriot Haxhirexha^{1,2*}, Labeat Haxhirexha³, Agron Dogjani³, Ferizat Dika – Haxhirexha¹, Aferdita Ademi^{1,2}, Blerim Fejzuli^{1,2}, Teuta Emini^{1,2}

Received: 24 May 2022 / Accepted: 19 June 2022 / Published online: 20 July 2022

This article is published with open access at <https://journal.astes.org.al>

© The author(s) 2022. © The Albanian Journal of Trauma and Emergency Surgery is an Open Access Journal. All articles are distributed under the terms of the Creative Commons Attribution Non-Commercial License: <http://creativecommons.org/licenses/by-nc/> which permits unrestricted non-commercial use, distribution, and reproduction in any medium provided the original work is properly cited.

Abstract

Choledocho-duodenal fistulas are rare disorders that can be of various aetiologies. Some of the most common causes of these fistulas are neoplastic processes in the extra-hepatic bile ducts, biliary stones or chronic duodenal ulcers. We will present the case of a patient with a choledocho-duodenal fistula who was treated in our clinic due to a severe episode of acute hepatitis.

We will present a case report as follow; a 52-year-old man was urgently brought to the abdominal surgery ward of the Tetovo Clinical Hospital due to severe pain in the right epigastric and subcostal region which also appears in the back region. It is accompanied by nausea, vomiting, sub-febrile fever, and weakness.

The patient reports that he has long been treated with proton pump inhibitors such as pantoprazole due to a chronic duodenal ulcer...

Conclusion; Choledocho-duodenal fistulas are very rare disorders and therefore pose a real challenge to surgeons and endoscopists in terms of their timely diagnosis. However, there are authors who prefer a conservative treatment, while some others suggest surgical intervention respectively fistulectomy, in order to prevent complications that may occur as a result of the regurgitation of duodenal contents in the bile duct.

Keywords: choledocho-duodenal, fistula, ulcer, ERCP, papillotomy

Introduction

Biliary-enteric fistula was first described by Bartholin in 1654, but so far biliary-enteric fistulas are still rarely reported, which are thought to be one or multiple pathological perforations between biliary tree and gastrointestinal tract [1, 2]. Choledochoduodenal fistula (CDF), the special type of biliaryenteric fistulas, is nearly 90% caused by cholecystolithiasis [3]. Increasing cases have been reported in the last 30 years since the progress in hepatobiliary

techniques, such as endoscopic retrograde cholangiography (ERCP), magnetic resonance cholangiopancreatography (MRCP), which have been applied to extensively reevaluate hepatobiliary diseases in the clinic, especially in Mainland China. However, the preoperative diagnosis of CDF is still difficult because of the nonspecific and/or minimal clinical symptoms [4]. Hence, CDFs are resulting in tough challenges for surgeons, especially for young surgeons.

CDF classification is important for diagnosis and treatment in the clinic, and the position of the fistula may suggest what the probable cause of CDF is.

CDFs are classified by Ikeda and Okada [5] and Gong et al. [6, 7] separately.

According to the location of the fistula, Ikeda divided CDFs into two types; Type I is located in the longitudinal fold of the papilla; type II is in the posterior wall of the duodenal bulb (Fig. 1A).

While Gong et al [6, 7] divided them into three types; the first type is type A, which is a CDF hole located more than 2 cm away from the papilla. The second play is type B, characterized by a CDF hole located less than 2 cm away from

Original article, no submission or publication in advance or in parallel

* **Corresponding author:**

Prof. Dr. Kastriot Haxhirexha MD, PhD

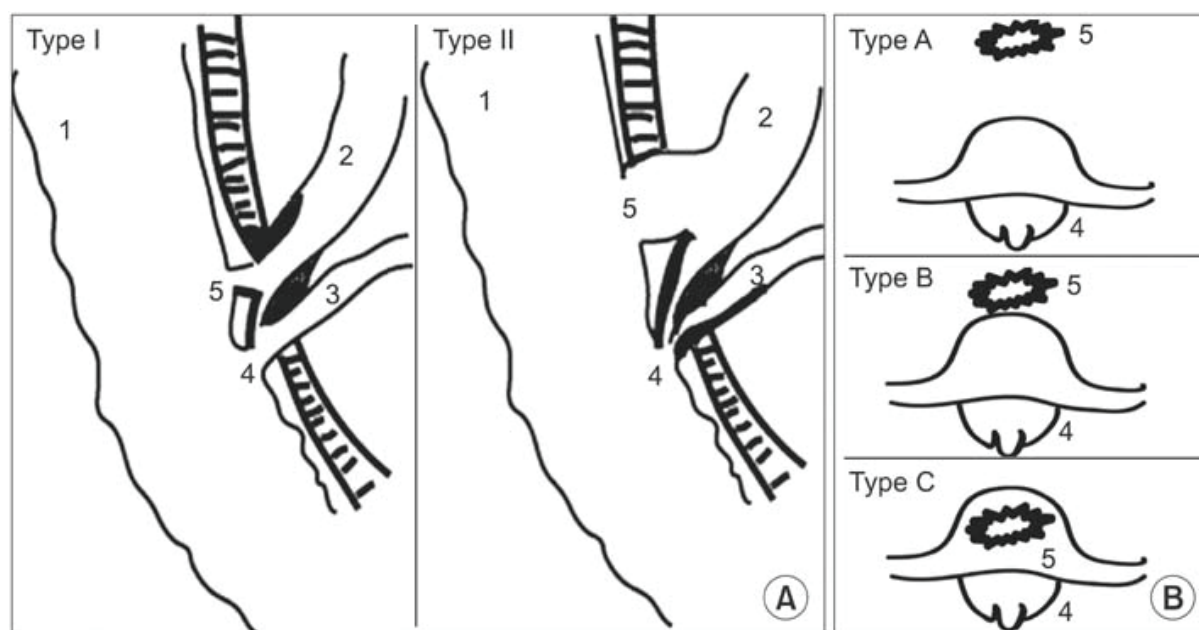
✉ dr.kastriot@gmail.com

1 Faculty of Medicine, the State University of Tetovo, RN of Macedonia.

2 Clinical Hospital – Tetovo, RN of Macedonia.

3 Faculty of Medicine, University of Medicine, Tirana, Albania





1- duodenum; 2 - CBD; 3 - pancreatic duct; 4 - major duodenal papilla; 5 - choledochooduodenal fistula - CDF.

Figure 1. The classification of choledochooduodenal fistula (CDF).

the papilla; and the third type is type C, also called PCDF, is a CDF hole located in the papillary fold (Fig. 1B).[8]

Fistulas between the biliary tract and the gastro-intestinal tract are very rare, especially those between the choledochus and the duodenum.

Although most of these fistulas are caused by the presence of stones in the biliary tract or choledocholithiasis (over 90% of them), sometimes the cause of their appearance can be chronic duodenal ulcers. [8, 9, 10]

Most of these disorders are associated with disturbances in the upper abdomen in the form of biliary colic, or in the form of chronic ulcerative pain.

The aim of our study is to show the case of a patient with acute hepatitis and severe abdominal pain as a result of a large choledocho-duodenal fistula from a chronic ulcerative process in the duodenum.

Case Report

A 52-year-old man was urgently brought to the abdominal surgery ward of the Tetovo Clinical Hospital due to severe pain in the right epigastric and subcostal region which also appears in the back region. It is accompanied by nausea, vomiting, sub-febrile fever and weakness.

The patient reports that he has long been treated with proton pump inhibitors such as pantoprazole due to a chronic duodenal ulcer. Immediately after hospitalization the patient underwent a native radiograph of the abdomen where no signs of pneumoperitoneum or any other changes were noticed. Also, an emergency abdominal ultrasonography did not show the presence of stones in the gallbladder or bile duct.

However, a thickening of the choledochal wall as well as a slight enlargement of the choledochus (8 - 9 mm) was visualized. At the same time this examination showed a steatotic liver with some small cystic formations on the right lobe.

The duodenum and stomach appeared slightly swollen. Laboratory examinations showed anemia expressed with hemoglobin valued at 11.2 g/dl and hematocrit of 32%. Moderate leukocytosis was also observed (WBC 14.1 mm³).

Biochemical analyzes detected an increase of hepatic enzymes AST (163 U/L) and ALT (194 U/L). The analysis also found alkaline phosphatase (872 U/L), simultaneously the level of bilirubin in the blood was increased to (32 μ mol/L). CRP levels were elevated (76) and D-dimers (3100), while lipase levels were within normal range. Tumor markers Ca 16-9 was also slightly increased to (53 U/ml).

A hypo-albuminemia at a level of 2.2 g / dl was also present in the patient.

After stabilization of the general condition through antibiotics (ceftriaxone and phlagyl) accompanied by analgesics of the non-steroidal anti-inflammatory drugs (NSAID) group the patient underwent an abdominal CT with contrast.

On the CT aerobilia was observed along the ductus choledochus as well as a thickening of the duodenal wall in segments D1 and D2. In addition, in the right lobe of the liver three small cysts with diameters between 3 and 7 mm were visualized.

Due to the presence of air in the bile duct an Endoscopic retrograde cholangiopancreatography (ERCP) was also performed according to standard procedures. The ERCP showed the presence of an ulcerative process in segment D1

of the duodenum as well as an inflammatory process around it. After injecting a contrast liquid into the common biliary tract, the presence of a fistula between the choledochal and the duodenum above the level of the duodenal papilla was detected.

Since the ERCP did not indicate the presence of any pathological processes in the biliary tract, either neoplastic or choledocholithiasis, no intervention was undertaken other than a papillotomy by the endoscopist. (Fig. 2)



Figure 2 Papillotomy

A few days after treatment in our ward the stabilization of the general condition of the patient began. This was manifested by a return to normal levels of hepatic enzymes and bilirubin with an increase in the level of albumin serum. For this reason, ten days after the hospital stay, he continued outpatient treatment with the suggestion to continue regular check-ups for a certain period of time in our department. Six months after treatment the patient's condition is good and there were no signs of recurrence of the disease.

Discussion

Choledocho-duodenal fistula (CDF) is a rare disorder which can be the result of various pathological processes. Most of them are a consequence of bile stones or neoplastic processes in the biliary tract [11, 12]. Other disorders that may promote the appearance of these fistulas may include duodenal ulcers,[13] neoplastic processes in the bile ducts and dilatation of the choledochus during a choledochotomy. However, sometimes there may be no obvious cause of this disorder.

Choledocho-duodenal fistulas can be complicated by the development of acute hepatitis, severe pain in the upper abdomen that may mimic biliary colic, or with the appearance of liver abscesses.[14] However, sometimes these disorders may not be associated with obvious disturbances, and may be misdiagnosed during an examination for another patient disturbance.

Most of these fistulas are located in the periampullar region while those due to a duodenal ulcer are usually located above the papilla of the Vater. It is thought that the presence of these fistulas marks a greater risk both for the appearance of stones in the biliary tract, and for the development of neoplastic processes in said tract.[8]

Some studies show that both of these complications can occur due to infection of the biliary tree by the duodenal contents which easily regurgitates into the common bile duct. The passage of duodenal fluid into the choledochus can activate pancreatic enzymes which then become the cause of chronic cholangitis, neoplastic changes and the formation of bile stones.[9]

The best way to diagnose choledocho-duodenal fistulas is by an abdominal CT with contrast and ERCP.[15] Sometimes, a contrast gastro-duodenography enables the visualization of these fistulas due to a contrasting regurgitation from the duodenum to the choledochus. [16, 17]

Treatment of these fistulas remains controversial. If they are not complicated by frequent episodes of cholangitis, and show no signs of bile stone formation or the development of neoplastic processes, then conservative treatment and follow-up of the patient through ultrasonography and abdominal CT is recommended.[16]

This can sometimes be accompanied by an endoscopic sphincterotomy of duodenal papilla [18].

In cases with frequent episodes of cholangitis, formation of bile stones or the appearance of premalignant or malignant changes in the biliary epithelium a fistulectomy should be undertaken. However, there are authors who suggest surgical treatment of fistulas as soon as they are diagnosed in order to avoid the risk of complications mentioned above. [18]

Conclusion

Choledocho-duodenal fistulas are very rare disorders, and therefore pose a real challenge to surgeons and endoscopists in terms of their timely diagnosis. However, there are authors who prefer a conservative treatment, while some others suggest surgical intervention respectively fistulectomy, in order to prevent complications that may occur as a result of the regurgitation of duodenal contents in the bile duct.

COI Statement: This paper has not been submitted in parallel. It has not been presented fully or partially at a meeting or podium or congress. It has not been published nor submitted for consideration beforehand.

This research received no specific grant from any funding agency in the public, commercial, or non-profit sectors. There are no relevant or minor financial relationships from authors, their relatives or next of kin with external companies.

Disclosure: The authors declared no conflict of interest. No funding was received for this case report

References

1. Marshall T, Kamalvand K, Cairns SR. Endoscopic treatment of biliary enteric fistula. *BMJ* 1990; 300:1176.
2. Liu TM, Chiu HH. Images in clinical medicine. Gallstone ileus. *N Engl J Med* 2010; 362:345.
3. Lin CT, Hsu KF, Yu JC, Chu HC, Hsieh CB, Fu CY, et al. Choledochoduodenal fistula caused by cholangiocarcinoma of the distal common bile duct. *Endoscopy* 2009;41 Suppl 2: E319–E320.
4. Luu MB, Deziel DJ. Unusual complications of gallstones. *Surg Clin North Am* 2014; 94:377–394.
5. Ikeda S, Okada Y. Classification of choledochoduodenal fistula diagnosed by duodenal fiberoscopy and its etiological significance. *Gastroenterology* 1975; 69: 130–137.
6. Gong JP, Zhou YB, Han BL. Diagnosis and classification of choledochoduodenal fistula. *Chin J Digest Endosc* 1999; 2: 42–43.
7. Gong JP, Zhou YB, Han BL. Diagnosis and surgical management of choledochoduodenal fistula. *J Third Mil Med Univ* 2000; 22: 287–289.
8. Wu MB, Zhang WF, Zhang YL, Mu D, Gong JP. Choledochoduodenal fistula in Mainland China: a review of epidemiology, etiology, diagnosis and management. *Ann Surg Treat Res.* 2015 Nov;89(5):240-246. <https://doi.org/10.4174/ast.2015.89.5.240>
9. Mohamed Ali Chaouch, Karim Nacef, Mossab Ghannouchi, Mohamed Ben Khalifa, Asma Chaouch, Molk Abdelkafi, Saida Jerbi, Moez Boudokhane. Choledochoduodenal fistula due to peptic duodenal ulcer diagnosed by X-barium meal study: interest of medical treatment. *Pan African Medical Journal.* 2018; 29:177
10. Jaballah S, Sabri Y, Karim S. Case report: choledochoduodenal fistula due to duodenal peptic ulcer. *Digestive diseases and sciences.* 2001; 46(11): 2475-2479.
11. Mallikarjunappa B, Ashish SR (2013) Choledochoduodenal fistula: a rare case report with review of literature. *JIMSA* 26(4):226
12. Dadzan E, Akhondi H (2012) Choledochoduodenal fistula presenting with pneumobilia in a patient with gallbladder cancer: a case report. *J Med Case Rep* 6:61
13. Xeropotamos N et al. Choledochoduodenal fistula: an unusual complication of penetrated duodenal ulcer disease. *Annals of Gastroenterology.* 2004; 17(1): 104-108
14. Staqnitti, F., Monqardini, M., Schillaci, F. Spontaneous biliodigestive fistulae: the clinical considerations, surgical treatment and complications. *G Chir* 2000; 21: 110–7.
15. Ho, G., Smith, L., Aitken, S. The diagnosis of choledochogastric fistula. *Gut* 2008; 57: A1–172
16. Akaydin, M., O. Demiray, E. Ferlengez, F. Eroztgen, Y. E. Ersoy, and M. Er. 2016. Importance of spontaneous choledochoduodenal fistulas detected during ERCP procedure. *Indian J. Surg.* 2016:1–5.
17. Shimono, T., K. Nishimura, and K. Hayakawa. 1998. CT imaging of biliary enteric fistula. *Abdom. Imaging* 23:172–176
18. Mallikarjunappa, B., and Ashish S. R. 2013. Choledochoduodenal fistula: a rare case report with review of literature. *JIMSA* 26:226.