

Compression of Common Hepatic Duct (CHD) Caused by Dilated Cystic Duct, is it Another Variant or New Type of Mirizzi Syndrome: A Case Report

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ABSTRACT

Background: Narrowing or obstructing of Common Bile Duct (CBD) by gallstone impacted in distal cystic duct is known as Mirizzi Syndrome (MS). However, the compression of Common Hepatic Duct (CHD) because of the enlargement of cystic duct due to the impacted stone in the distal cystic duct has never been reported. **Case Report:** A 60 year old man, Indonesian Javanese ethnicity, came to the hospital with bile duct stone and obstructive jaundice due to the compression of Common Hepatic Duct (CHD) because of the enlargement of cystic duct. In this case, the stone just lies in the tip of the cystic duct causing obstruction of cystic duct but the stone does not directly compress the CBD. Long cystic duct with mid or low insertion into CBD possibly occurs in this case. The diagnosis of this case is common hepatic duct obstruction caused by the enlargement of cystic duct. Two surgeries were performed, the first surgery was only cholecystectomy, and the second surgery was taking out the remnant of cystic duct including the stone inside. The outcome of the surgery was good, and the patient was allowed to go home from the hospital without further complaints. This rare case cannot be grouped into the existing classification of Mirizzi syndrome. May this case be grouped as another variant or a new type of Mirizzi Syndrome? This case could be grouped as type I-a of Mirizzi syndrome according to the classification of Mc. Sherry, Beltran or Csendes. **Conclusion:** In Mirizzi syndrome the obstruction of CBD is not only caused directly by the stone in distal cystic duct, but also it can be caused by the enlargement of the cystic duct. This type of bile duct obstruction has never been reported before and this rare case cannot be grouped into the existing classification of Mirizzi syndrome. It is important for surgeons to recognize similar cases in the future, misinterpretation of imaging findings of bile duct obstruction makes surgery difficult and harmful for the patient.

Keywords: Common Hepatic Duct obstruction, Cystic duct dilatation, Mirizzi Syndrome.

INTRODUCTION

The misinterpretation of the imaging picture of bile duct obstruction makes the surgery difficult and harmful for the patient. Preoperatively, the diagnosis of the patient was CBD obstruction due to the characteristic of classic Mirizzi syndrome type-1 according to the classification of Mc Sherry, Beltrán and Csendes. Later it was known that the CBD obstruction was not caused by the stone in neck of the gall bladder but caused by the enlargement of cystic duct. The enlargement of cystic duct that causes compression to CHD has never been reported before, so misinterpretation in making diagnosis will occur. The wrong diagnosis made the surgery difficult but fortunately the outcome of the patient was good, although the patient had undergone 2 surgeries, 2 Magnetic Resonance-Cholangio-Pancreaticography (MRCPs) and 1 Endoscopic Retrograde Cholangio-pancreatography (ERCP), and this was a burden for the patient.

CASE PRESENTATION

A 60-year-old man of Indonesian Javanese ethnicity, owner of a car repair shop, came to the hospital complaining about 5 day-abdominal pain and icterus. There was no history of the same disease suffered by the patient's family. Upon the physical examination, we found the patient with

icteric, fever and tenderness at right upper quadrant of the abdomen. In the laboratory test, we found Total / Direct bilirubin 14.6 and 10.4, with white blood cells (WBC): 11.70 . On ultra sonography, gall bladder stones and dilatation of intra and extra hepatal bile duct because of impacted stone in distal CBD were revealed. Preoperative MRCP concluded distal CBD stone and dilatation of Intra Hepatic Bile Duct (IHBD) and Extra Hepatic Bile Duct (EHBD). Preoperative diagnosis was gall bladder stones and CBD stone with obstructive icterus and acute cholangitis. Open surgery was performed and because of the edema and heavy adhesion, the surgeon only made a cholecystectomy, the CBD stone was left in place with a plan of performing extraction of the CBD stone using ERCP the next day. However, on ERCP, there was no stone found inside the CBD, what was found there was the narrowing of CBD at the level of the confluence of CHD due to the compression from outside. The CBD stent was inserted passing through the narrow part of the CHD. MRCP was performed for the second time, it was known that the stone was located in distal cystic duct, contrary to the previous MRCP reporting that the stone was located in distal CBD. The compression of CHD was not directly caused by the stone in distal cystic duct as it commonly occurs in Mirizzi syndrome, but the compression of the CHD because of the enlargement of the cystic duct. After the CBD stent was placed, the bilirubin level

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fell down and the condition of the patient was stable. Three months later, the second open surgery was performed to take the cystic duct stone, the remnant of cystic duct was cut as far as possible close to the CBD. The patient was allowed home in a good condition and the CBD stent was taken out three months later.

On the first MRCP, the enlargement of cystic duct was assumed as the enlargement of CBD because of the impacted stone in distal cystic duct due to Mirizzi Syndrome. During the surgery, the surgeon found and assumed that the enlargement of cystic duct as the enlargement of CBD. The surgeon did not find the stone and decided to finish the surgery. ERCP was needed to confirm the diagnosis and it was performed the next day. On ERCP, it turned out that the enlargement was CHD but not CBD, the enlargement of CHD was caused by the stone impacted in distal cystic duct. The distal cystic duct stone was identified later after the second MRCP had been performed. Preoperatively, on the first MRCP, it had actually been known that the compression of CBD occurred due to the compression caused by dilated cystic duct and not directly by the stone (Fig.1A), but it had not been thought of before because this type of CHD compression by dilated cystic duct has never been reported.

Twice surgeries were performed, the first surgery failed to identify and take the stone out because the surgeon was in doubt in identifying the CBD and moreover, inflammation and heavy edema made the surgery difficult, so only cholecystectomy was performed. After the location of the stone was identified by the second MRCP, the second surgery was easier to perform. The remnant of cystic duct together with the stone inside was taken out without opening the CBD. The condition of the patient was good and allowed home from the hospital without further complaints. The ERCP stent was taken out 3 months later. Follow-up visit for 3 years after surgery, the patient has no symptoms and lives without any problems. The patient realized that the case was difficult and never be reported before. The difficulty in making diagnosis and unexpected repeated surgical procedures had been understood and accepted by the patient. All the laboratory, imaging examinations and surgeries were performed with the patient's consent.

DISCUSSION

Mirizzi syndrome occurs when gallstones lodged in the neck of the gallbladder or cystic duct create mechanical pressure, leading to blockage of the common bile duct (CBD)^{1,2}. Anatomical variations of the cystic duct are common, seen in 18%-23% of patients. In these cases, the cystic duct typically merges with the middle third of the extrahepatic bile duct in 75% of instances, and with the distal third in 10%^{3,4}. Special consideration must be given to the distal insertion of a long cystic duct, as this anomaly can result in misdiagnoses during imaging, which can subsequently impact treatment options⁵⁻⁷. According to Zhou's findings, 5.9% of cases exhibited an abnormal confluence of the cystic duct into the CBD⁸, with the mid-posterior insertion being the most frequently observed variant⁹.

In this case, the preoperative diagnosis was CBD stone, obstructive jaundice and acute cholangitis. Open cholecystectomy and choledocholithotomy were planned. Intraoperatively, cholecystectomy was performed, cystic duct was cut and ligated. The remnant of cystic duct was assumed to be CBD, "choledochotomy" was performed but the stone was not found because of the heavy edema due to the inflammation. It was not known that there was a long cystic duct inserting into mid CBD. The surgeon decided to finish the surgery and planned to take the CBD stone out using ERCP the next day. However, on ERCP, there was no stone found inside the CBD, the CBD stent was inserted passing through the narrowing part at the level of Common Hepatic Duct (CHD).

The enlargement of long cystic duct compresses the CHD, it is possible when the infundibulum of Gall Bladder (GB) is fixed to the liver by

thick connective tissue. So the enlarged cystic duct moves closer to CBD and then compressing and bending out the CHD. Later, because the cystic duct continues to enlarge, the compression of CHD increases and makes the lumen of CHD narrowing and obstructing the CHD lumen (Figure 1B3).

In Mirizzi syndrome, the stone that causes obstruction of CBD is located in the distal cystic duct and directly pushes the CBD or makes fistulae into CBD or intestine^{10,11}. However, in this case, the pressure of CHD is caused by the enlargement of cystic duct but not by the stone in distal cystic duct. This type of CHD obstruction cannot be grouped into the Mirizzi syndrome classification. So, can this type of CHD obstruction be classified as another variant or new type of Mirizzi Syndrome? Type I of Mirizzi Syndrome is the same between PL Mirizzi¹, Mc Sherry¹², Beltrán¹³, and Csendes¹⁴ classification, that is CBD obstruction because of external compression by the stone in distal cystic duct (table 1). However, in this case, it is possible to group the CHD obstruction into a new classification, that is Type I-a of Mirizzi Syndrome 2023 (Table 1). It is important for the surgeon to pay a special attention when performing the surgery in this type of case. Intraoperative cholangiography may be used to avoid misidentification of the CBD to prevent iatrogenic bile duct injuries¹⁵ or to insert CBD stent before surgery when freeing the long cystic duct which usually attaches to CBD especially in inflamed case^{16,17}.

Csendes et al. devised a classification for type II Mirizzi syndrome, dividing it into three subtypes (II, III, and IV) based on the extent of the common bile duct (CBD) involvement¹⁴. Type II involves a fistula affecting one-third of the CBD circumference; type III covers two-thirds; and type IV results in a completely unrecognisable CBD, merging entirely with the gallbladder¹². Beltran et al. added type V to the classification, specifically for cases with cholecysto-enteric fistula (CBF), subdividing them into (a) without gallstone ileus and (b) with gallstone ileus. Later, Beltran revised the classification to three types: (i) Type I, the "classic" Mirizzi syndrome; (ii) Type II, cholecystocholedochal fistula with subtypes (a) less than 50% and (b) more than 50% of the CBD diameter; and (iii) Type III, cholecysto-enteric fistula without (a) or with (b) gallstone ileus^{13,18}. Despite this, many authors prefer Csendes' original classification for its detailed specification of CBD wall involvement¹⁹. Though it accurately describes the types of Mirizzi syndrome, determining the degree of bile duct wall damage before surgery remains challenging²⁰.

CONCLUSION

In the conclusion, in Mirizzi syndrome the obstruction of CBD is not only caused directly by the stone in distal cystic duct, but also it can be caused by the enlargement of cystic duct. This type of bile duct obstruction has never been reported before and this rare case cannot be grouped into the existing classification of Mirizzi syndrome. May this case be grouped as another variant or a new type of Mirizzi Syndrome? This case could be grouped as type I-a of Mirizzi syndrome according to the classification of Mc. Sherry, Beltran or Csendes. It is important for surgeons to recognize similar cases in the future, misinterpretation of imaging findings of bile duct obstruction makes surgery difficult and harmful for the patient.

ABBREVIATIONS

CBD	Common Bile Duct
CHD	Common Hepatic Duct
EHBD	Extra Hepatic Bile Duct
ERCP	Endoscopic Retrograde Cholangiopancreatography
IHBD	Intra Hepatic Bile Duct

Table 1. Classifications of Mirizzi syndrome and Possible/New Classification 2024.

Pablo L. Mirizzi ¹	Mc Sherry ¹²	Beltrán ¹³	Csendes ¹⁴	Possible/New Classification 2024
Obstruction of CBD caused by gallstone in the neck of GB	Type I – Bile duct obstruction by external compression by gallstone in the neck of GB Type II – cholecystobiliary fistula (CBF)	Type I - Extrinsic compression CBD by gallstone in the neck of GB Type IIa - CBF affects < 50% of the CBD Type IIb - CBF affects > 50% of the CBD Type IIIa - CBF with CEF and without gallstone ileus Type IIIb - CBF with CEF and without gallstone ileus	Type I - Extrinsic compression CBD by gallstone in the neck of GB Type II - CBF affects < 33% of the CBD Type III - CBF affects 33-66% of the CBD Type IV - CBF affects > 66% of the CBD Type Va - MS with CEF and without gallstone ileus Type Vb - MS with CEF and gallstone ileus	Type Ia - Extrinsic Compression caused by dilated cystic duct

Note: CBD Common Bile Duct, GB Gall Bladder, CBF Cholecystobiliary Fistula, CEF Cholecystoenteric Fistula, MS Mirizzi Syndrome

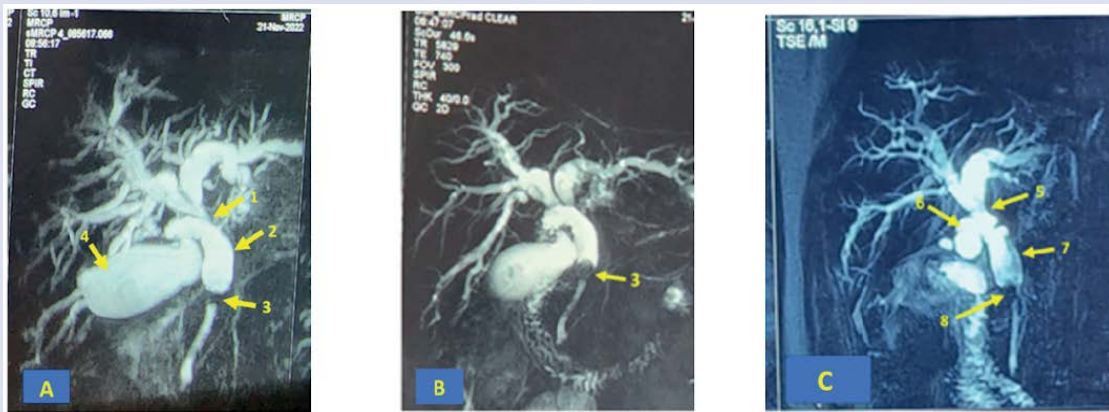


Figure 1. Preoperative MRCP (A-B) and Post cholecystectomy MRCP (C). (A&B): Narrowing CHD pushed by dilated cystic duct (A1), CBD dilatation on the first assumption (A2), the stone located at mid CBD on the first assumption (A3, B3), Gall bladder (A4). **(C):** The narrowing part of CHD (C5), remnant of dilated cystic duct (C6), part of non dilated CHD is bent by the dilated cystic duct (C7) and stone located at distal cystic duct (C8). CBD Common Bile Duct, CHD Common Hepatic Duct, MRCP Magnetic Resonance-Cholangio-Pancreatography.



Figure 2. Post cholecystectomy MRCP(A) and Illustration (B). (A) Cystic-duct stone does not directly compress the CBD. (B) Thick connective tissue in infundibulum of gall-bladder (1), Dilated cystic duct (2), CHD is compressed by dilated cystic duct (3), Part of non dilated CHD (4), Stone in distal cystic duct (5), Duodenum (6). CBD Common Bile Duct, CHD Common Hepatic Duct.

MRCP Magnetic Resonance-Cholangio-Pancreaticography

MS Mirizzi Syndrome

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I completely understand the patient's frustration with the problem, and I appreciate the cooperation of the patient with the subsequent procedures. I do my best to solve the problem as perfectly as possible. I also appreciate the patient's willingness to have his case reported in order to prevent other surgeons from experiencing the same unexpected problem in the future.

CONFLICTS OF INTEREST

There is no competing interests from the author.

FUNDING

This case report is only funded by the author

ETHICS STATEMENT

No ethical approval

DATA AVAILABILITY

Patient data, laboratory examination results and imaging reports were taken from the patient's medical record. I use the data of the patient, result of laboratory tests and imaging examinations based on the agreement of the patient. The patient agrees that his data will be used for publication.

AUTHORS' CONTRIBUTIONS

In this case, the author made the diagnosis and performed the surgeries, the decision to request MRCPs and ERCP was also made by the author.

CONSENT FOR PUBLICATION

Written informed consent was obtained from the patient's legal guardian for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

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