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Choledocholithiasis Complicated by Cholangitis and Cholecystitis in A Patient with Anomalies of the Biliary Ducts (Case Report)

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Abstract

Background: Rapid development and broad implementation of modern imaging methods and diagnostic techniques have greatly contributed to more precise appreciation of the anomalous conditions and pathologies of the extrahepatic biliary system—one of the parts of the human body characterized with significant anatomical variability. **Case Report:** A 73-year-old female patient was admitted to The First Medical Center of Tbilisi with complaints of pain and a feeling of heaviness in the right hypochondrium, fever (38˚C), nausea, weakness, jaundice. Abdominal ultrasound revealed an enlarged gallbladder with thickened walls and a large stone incarcerated in the gallbladder neck. The diameter of the CBD was increased up to 4 cm, and large size stones present within the lumen. A CT scan has also revealed a cholecysto-duodenal fistula. Open cholecystectomy was decided as a treatment of choice. Intraoperatively was found a fistula between the fundus of the gallbladder and the duodenum, a gallbladder with thickened walls, and stones wedged into the neck, a common bile duct of significantly enlarged diameter (4 cm) with large size stones, and an accessory small diameter duct between the gallbladder and the CBD. After choledochotomy, 4 × 2 cm and 3 × 2 cm size stones were removed from CBD. During cholecystectomy, the Luschka duct was found within the gallbladder bed. **Conclusion:** We report on a rare case of anomaly presented in the form of an accessory bile duct between the gallbladder and the common bile duct, as well as with an accessory duct of Luschka. Additionally, significantly enlarged extrahepatic bile ducts with giant intraductal stones and cholecystoduodenal fistula were revealed. The combination of these pathologies and anomalies is extremely rare.

Keywords

Gallbladder, Cholecysto-Duodenal Fistula, Accessory Bile Duct, Duct of Luschka

1. Introduction

The most outstanding feature of the normal anatomy of the extrahepatic biliary system is its high degree of variability. Anatomic abnormalities associated with the opening of biliary system into the upper gastrointestinal tract have been increasingly recognized after more widely utilization of modern diagnostic methods in clinical practice. The data about the clinical implications of this anatomic variation are very limited and the treatment usually involves surgery [1].

2. Case Report

The patient, a 73-year-old female, was admitted to the First Medical Center of Tbilisi with complaints of pain and a feeling of heaviness in the right hypochondrium, fever (38˚C), nausea, weakness, and jaundice. According to the patient, complaints began about 6 hours before the admission. The patient was aware of gallstones. She had had periodic pain in the right hypochondrium, but has not agreed to surgery for many years. Seven years ago, a left nephrectomy was performed due to hydronephrosis. Thorough clinical and laboratory examinations were carried out. CBC test showed leukocytosis, neutrophilia with a shift to the left (WBC-15.85 × 10^9/L, NEUT-93%). Liver function tests showed elevated transaminases and bilirubin levels (TBIL-64.35 μmol/L, DBIL-45.12 μmol/L). CRP levels were elevated (CRP-232 mg/dl). Abdominal ultrasound revealed an enlarged gallbladder with thickened walls and a large stone incarcerated in the neck of the gallbladder. The diameter of the common bile duct (CBD) was increased up to 4 cm with large size stones visible within the lumen. A computed tomography scan was performed. In addition to the above-mentioned pathologies, cholecystoduodenal fistula was identified (**Figures 1-5**). Acute calculous cholecystitis, cholecystoduodenal fistula, choledocholithiasis, acute cholangitis, obstructive jaundice were diagnosed. It was clear that attempts to remove the stones endoscopically would not be successful due to their size. Surgical treatment via laparotomy approach was chosen.

After preoperative examination, consultation and preparation, urgent surgery was performed. Laparotomy was performed with a right subcostal incision. Intraoperatively was found a fistula between the fundus of the gallbladder and the duodenum, a gallbladder with thickened walls, with stones wedged into the neck, a common bile duct of enlarged diameter (4 cm) with large stones, and an accessory small diameter duct between the gallbladder and the CBD (**Figures 6-9**). The fistula was opened and dissected. Consequently, the duodenum was sutured with a double-row suture. The stones, incarcerated in the neck of the gallbladder, were pushed from the neck into the body of the gallbladder. After choledochotomy, 4 ×

2 cm and 3 × 2 cm size stones were removed from CBD (Figure 10, Figure 11). The accessory bile duct (AD) between the gallbladder and the CBD was cut. Bile was obtained from this duct. The proximal and distal ends were ligated, after

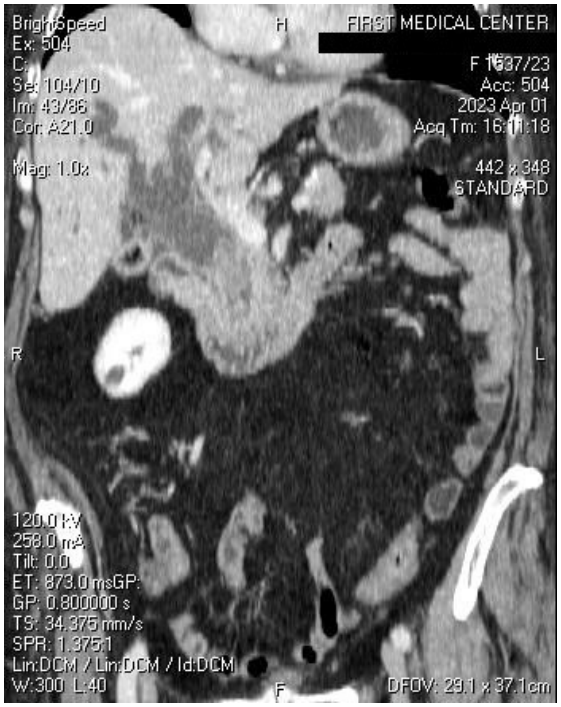


Figure 1. CT with contrast enhancement, coronary reconstruction. Dilated bile ducts and a large stone in the common bile duct are seen.

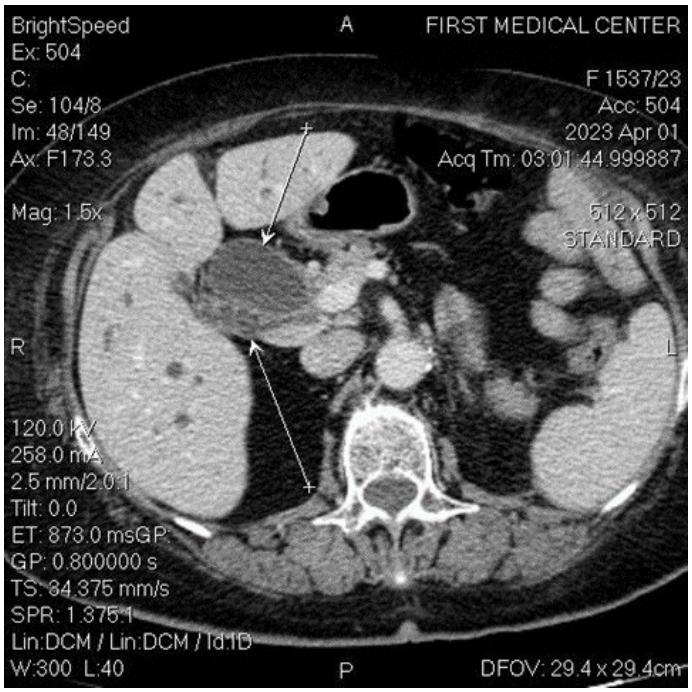


Figure 2. CT with contrast enhancement. Axial slice showing dilated bile ducts. Long arrow to cystic duct (CD), short arrow to common bile duct (CBD).

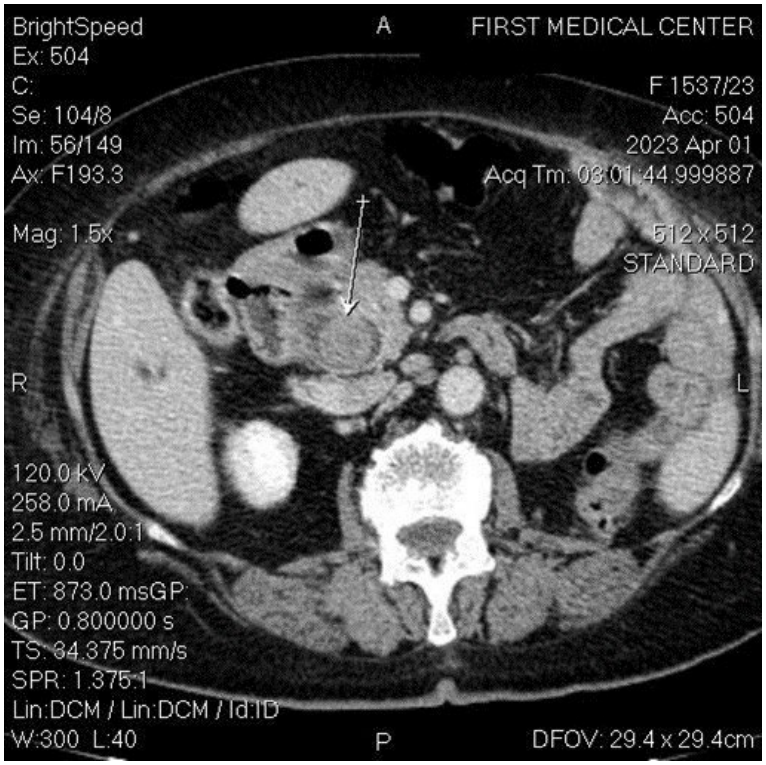


Figure 3. CT with contrast enhancement. Axial slice showing dilated bile ducts. A large stone in the CBD shown by the arrow.

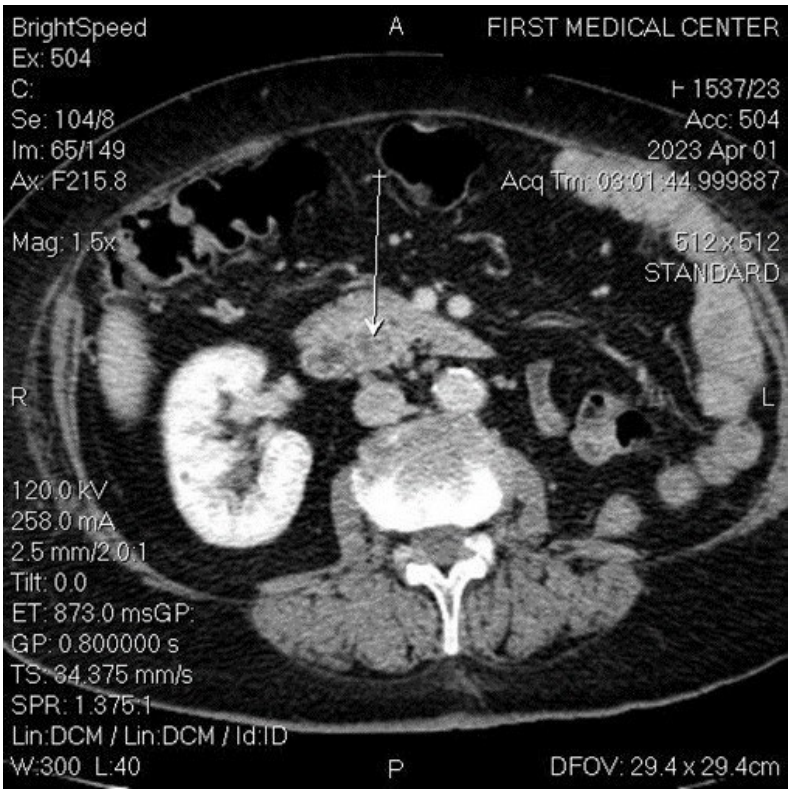


Figure 4. CT with contrast enhancement. Axial view showing another stone in the CBD adjacent to the ampulla of Vater.

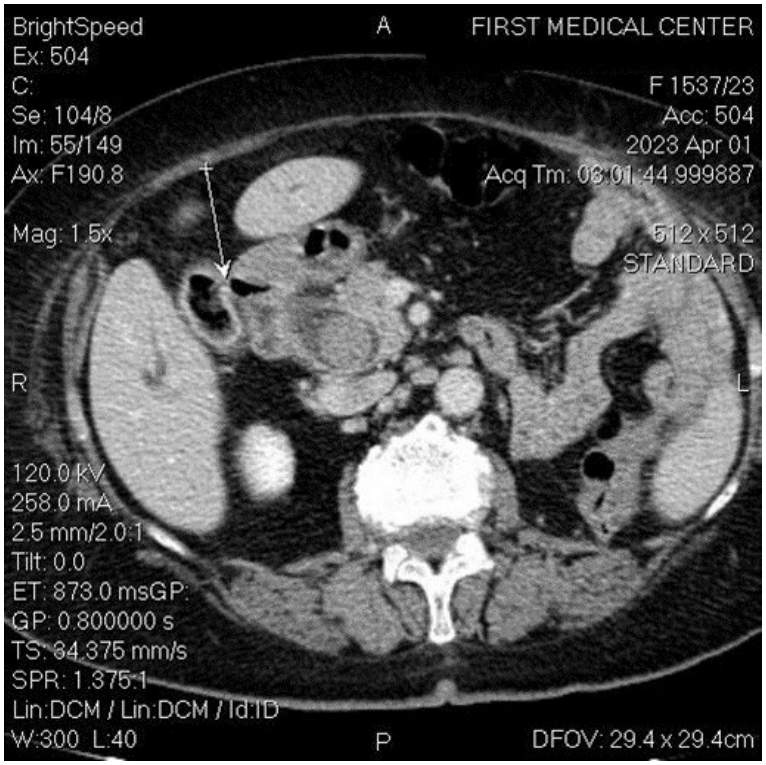


Figure 5. CT with contrast enhancement. Axial view shows the connection of the gallbladder with the wall of the duodenum. There are areas of gas density in the gallbladder.

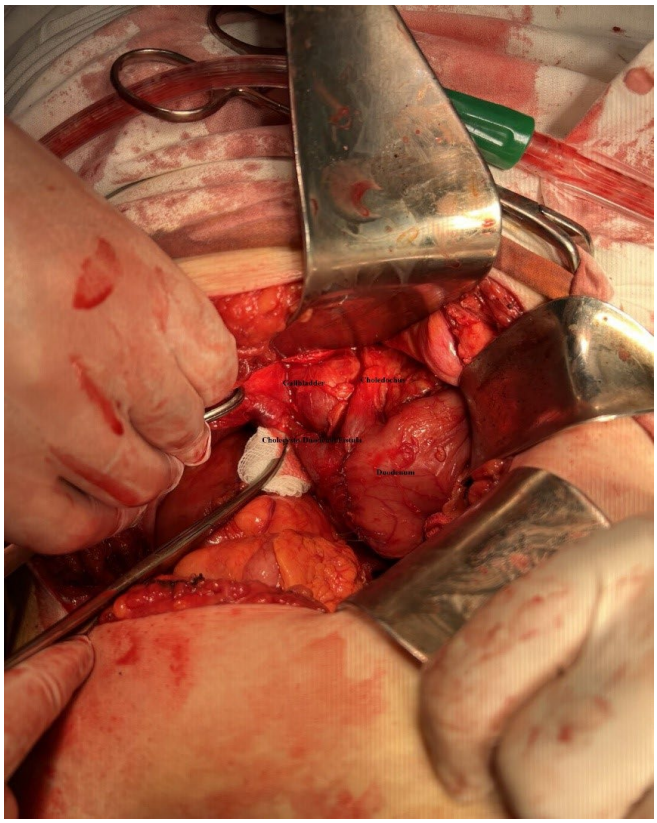
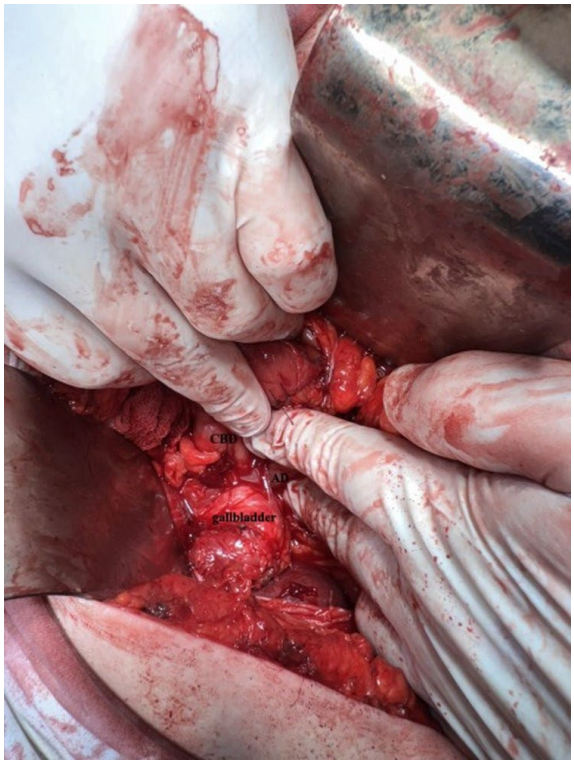
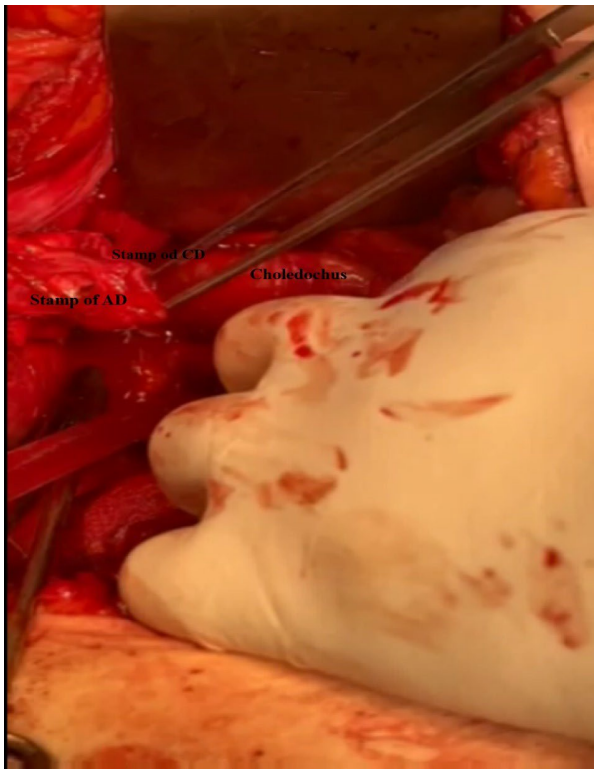


Figure 6. Cholecysto-duodenal fistula.



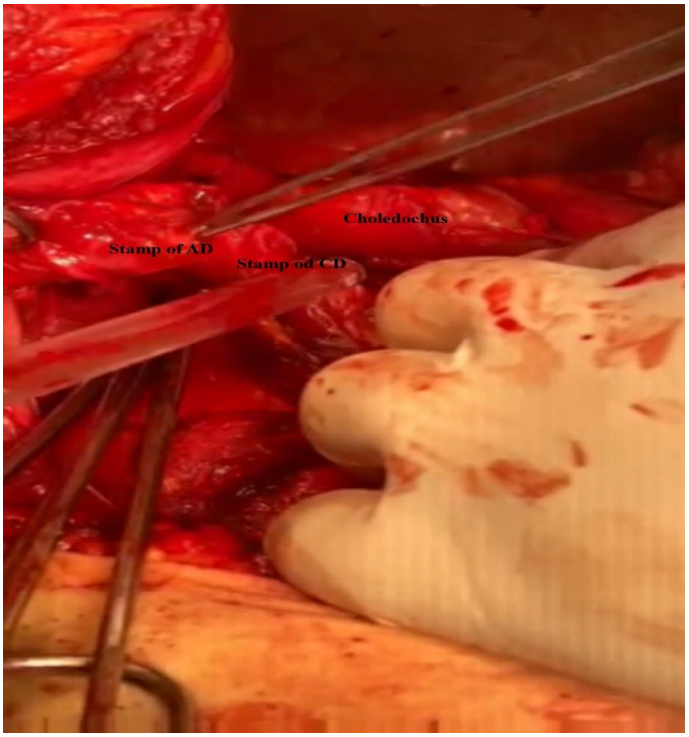
AD - Accessory Duct; CBD - Common bile duct.

Figure 7. Accessory duct between gallbladder and CBD.



CD - Cystic Duct; AD - Accessory Duct.

Figure 8. The stumps of the cystic and accessory ducts are seen.



CD - Cystic Duct; AD - Accessory Duct.

Figure 9. The stumps of the cystic and accessory ducts are seen.

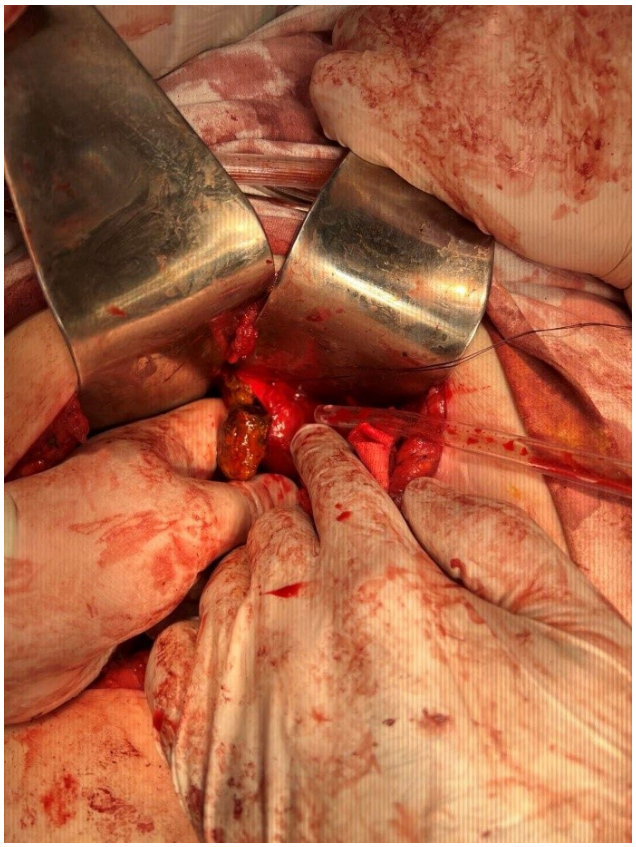


Figure 10. A large stone is removed from the CBD.

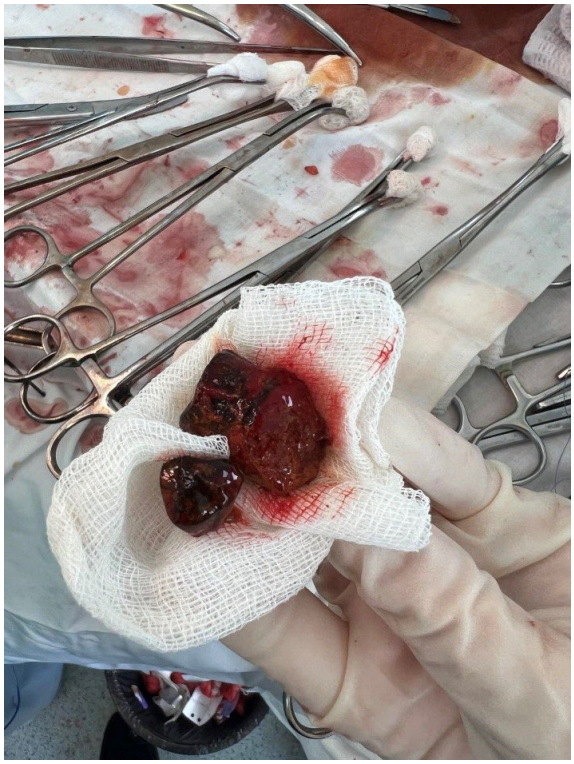


Figure 11. Stones extracted from the CBD.

CD - Cystic Duct; AD - Accessory Duct.

Figure 12. The opening of the additional duct of Luschka is seen.

which the AD was removed. Intraoperative cholangiography was performed through a T-tube. Passage of contrast into the duodenum was restored, no residual stones were observed. During cholecystectomy, Luschka duct was found in the gallbladder bed. The duct was ligated and cut (Figure 12). Cholecystectomy was performed. After irrigation of the subhepatic space, drainage was placed near the CBD and abdomen was closed in layers. Based on the intraoperative findings, the final diagnosis was established as acute calculous cholecystitis, cholecystoduodenal fistula, choledocholithiasis, acute cholangitis, accessory bile duct and duct of Luschka.

During the postoperative period, the patient developed biloma in the subhepatic area, which subsequently spread to the subdiaphragmatic area. Fever of varying grades was observed. CT imaging was done (Figure 13). Under CT control, percutaneous drainage of the biloma was performed using a "pig-tail" catheter. Intensive conservative therapy was carried out, which resulted in clinical and laboratory improvement. No fluid in the abdominal cavity was noted on CT scan. T-tube was removed on the 18th postoperative day after contrast cholangiography. The patient was discharged from the clinic in fully recovered condition on the 22nd day from admission. At follow-ups within one year after discharge, no complications were observed.

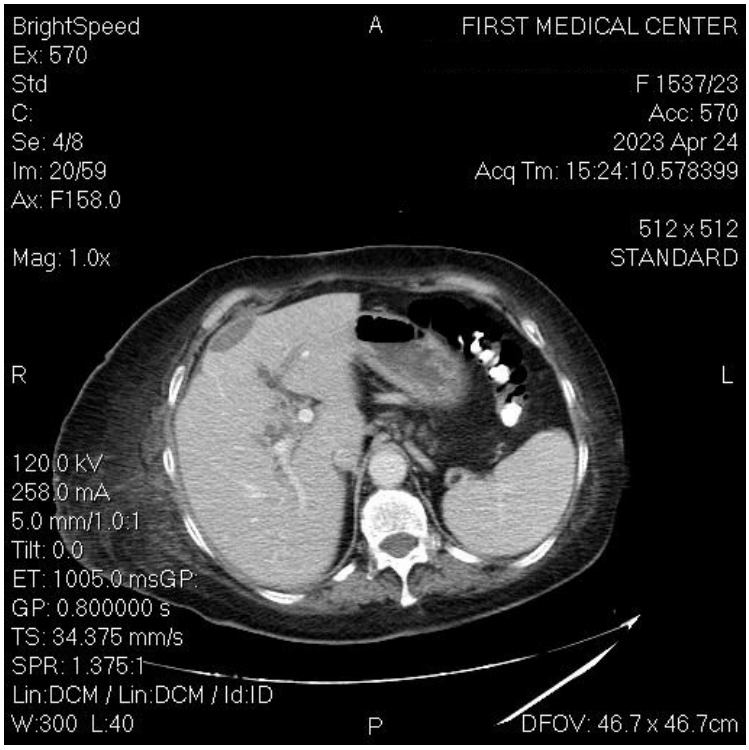


Figure 13. CT with contrast enhancement, axial projection, postoperative state. There is limited fluid along the edge of the liver—biloma.

3. Discussion

Surgical practice has long encountered anatomic variation in the biliary tree. The

notion of modal and aberrant anatomy was first described by Couinaud in 1957 through examination of liver corrosion casts on post-mortem specimens. Since, several imaging methods added to this technique to help visualizing the anatomy of the bile ducts. With the recent technical advancement of hepatic surgery, recognizing and considering anatomic variation in the surgical technique is now a must for success of surgery [2].

The most outstanding feature of the normal anatomy of the extrahepatic biliary system is its high degree of variability. The etiology of this anomalous opening is not known, but it has been argued that developmental errors formed during embryogenesis, which are not yet understood, could be a causative factor. The liver originates from the hepatic diverticulum, the cranial part (pars hepatica) of which gives rise to the intrahepatic and common hepatic ducts and the caudal part (the pars cystica) gives rise to the gallbladder and the cystic duct. [1] [3]. The pars cystica grows in length and represents the primordium of the gallbladder, the cystic duct, and common bile duct. For up to 8 weeks of gestation, the extra-hepatic biliary tree further develops through lengthening of the caudal part of the hepatic diverticulum. The pars cystica of the hepatic diverticulum begins initially from the anterior side of the future duodenum. At approximately the fifth week, the duodenum rotates to the right, so that the attachment of the developing common bile duct becomes displaced to its definitive position on the dorsal side of the duodenum. The hepatic duct develops from the cranial part (pars hepatica) of the hepatic diverticulum. In the 34-day embryo, the common hepatic duct is a broad, funnel like structure in direct contact with the developing liver, without a recognizable left or right hepatic duct. During the fifth week, a rapid entodermal proliferation takes place in the dilated funnel-shaped structure above the junction of common bile duct and cystic duct; this proliferation gives rise to several folds, resulting in several channels at the porta hepatis (transverse fissure of the liver). It is speculated that this remodeling at least partially explains the existence of the several variants in the configuration of the right and left hepatic ducts. The distal portions of the right and left hepatic ducts develop from the extrahepatic ducts and are clearly defined tubular structures by 12 weeks of gestation. The proximal portions of the main hilar ducts derive from the first intra-hepatic ductal plates. The ductal plate is the term given to the layer of cells surrounding the portal vein branches like a cylindrical sleeve. The extra-hepatic bile ducts and the developing intra-hepatic biliary tree maintain luminal continuity from the very start of organogenesis throughout further development [2] [4]-[6].

As opposed to congenital anomalies, the anatomical variations, as well as the modal type, are issued from a normal morphological development. The variations of division of extra-hepatic biliary ducts are very frequent. They are clearly explained by the sequence of embryological development in man, and also by compared anatomy. Lots of variations occur, some of them being more frequently encountered than others during cholecystectomy for gallbladder lithiasis. A cystohepatic duct draining a large hepatic territory is the most dangerous variation. As

a matter of fact, it can look as if the junction between the cystic duct and the common biliary duct was of the modal type. In surgical practice, the dissection of cystic duct must never go over the right side of the common biliary duct in order for it never to be injured. Radiological exploration of biliary tree during cholecystectomy for gallbladder lithiasis has to be routine, in order to discover the obviously unpredictable individual variations of division of extra-hepatic biliary ducts [7].

Biliary tract diseases represent a very common medical problem and often require emergent interventions. For example, cholelithiasis affects approximately 10% of the adult population, with the prevalence increasing with age. Approximately 35% of this population could be affected by complications and symptoms that could require cholecystectomy. Furthermore, the lack of knowledge in this surgical area could create many complications for patients, ranging from infections to definitive or even lethal injuries. Moreover, many other diseases could affect the biliary tree, representing very common causes of hospitalisation and surgical treatments [8] [9].

Variant extra-hepatic biliary anatomy has been reported to be related to gender, in particular, the maljunction of the pancreatico-biliary tract. In an Italian study, it was reported that a variant anatomy was significantly more common in females (45% vs. 26% in males; $p = 0.005$). This difference could probably be explained by a different embryologic development in the two sexes. Lack of data cannot though confirm this theory. In the present study, the majority of subjects were females: 68.6% of women versus 31.4 % of men, with a sex ratio of 0.45. Variant anatomy was not related to gender [2] [10]-[12].

Cholecystoenteric fistula, an abnormal communication between gallbladder and the gastrointestinal tract, is a rare complication of cholelithiasis. The cholecystoenteric fistula presents in variable symptoms, and an accurate diagnosis prior to surgery is rarely achieved [13]-[15]. Duodenal fistula is a rare disease, accounting for only 0.2% of cholecystolithiasis cases. The prevalence of internal biliary fistula is less than 0.3% in patients with gallstone disease. The relative frequency of fistula is cholecystoduodenal (60%), cholecystocolic (15%), cholecystogastric (5%), and choledochoduodenal (5%). The most common site of cholecy-stoenteric fistula is the duodenum, which accounts for about 80% of all fistulas of the gastrointestinal tract. Chronic cholecystitis with gallstones is reported to cause about 90% of cholecystoenteric fistulas. Preoperative diagnosis should be based on pneumobilia by abdominal CT scan, which should raise suspicion of a cholecystoenteric fistula or Mirizzi syndrome. Clinical symptoms of cholecystoduodenal fistula include gallstones, fever due to cholecystitis, abdominal pain, and jaundice. However, there are no characteristic symptoms, and diagnostic imaging is beneficial. If the diagnosis is not made preoperatively and is made intraoperatively, the fistula is recognized as an adhesion that is difficult to remove. Surgery is the first-line treatment for cholecystoduodenal fistula, although spontaneous fistula closure has been reported [16]-[18].

Normal biliary anatomy of the intra-hepatic ducts is present in 58% - 64.5% of the population. Many anatomic variations of the intra-hepatic ducts have been described in the literature, particularly intrahepatic right posterior biliary duct that is more often described in its different insertions. In addition, the described normal anatomy is present in only approximately 53% of the population. The anatomical variations of cystic duct (CD) insertion are historically less investigated than those of other biliary tracts [9].

CD insertion occasionally may have an unusual presentation. Reported uncommon anatomic variations include a high fusion of the cystic duct with the common hepatic duct, aberrant fusion of the cystic duct with the right or left hepatic duct, and similarly, drainage of an aberrant or accessory right posterior duct into the cystic duct [19]-[21].

The entry point of the CD relative to the common hepatic duct (CHD) is highly variable. The CD courses parallel to the CHD in 10.6% of patients. There is insertion from the medial aspect in 18.4% of patients and distal insertion in 10%. In a study of 198 patients who underwent MRCP, 4% were found to have a low medial insertion. However, low insertion in this study was defined as the CD joining the CHD at its lower third. The particular variation of a medial insertion at the level of the ampulla is an exceedingly rare finding. Other variations in cystic duct anatomy include anterior or posterior spiral insertion, proximal insertion and more rarely joining the left or right hepatic duct high in the porta hepatis or directly into the duodenum [22] [23].

A single gallbladder with a double cystic duct is a relatively rare congenital malformation. Up until late 2017, only 20 patients had been reported with duplicated CD in the English literature. The preoperative diagnosis of a duplicated CD is difficult, and the majority of the cases are found intraoperatively. In 1956, Caster and Flannery studied 101 cases with congenital abnormalities of the gallbladder and categorized double cystic ducts into three types according to the site of the drainage. The extreme rarity of this variation poses a challenge to surgeons, and missing the case may lead to unwanted consequences such as bile duct injury and postoperative morbidities. The anatomical distinctions of CD and gallbladders were first reported by Edward Boyden in the early 1920s. Later, Caster and Flannery classified CD duplications into three subgroups: H type, wherein cystic ducts join the CBD at two different sites; Y type, wherein cystic ducts join each other and then drain into the CBD; and the trabecular type, wherein one CD joins the CBD and the other enters the liver parenchyma. Duplicated CDs are more common among females. This could be because of the higher incidence of symptomatic cholelithiasis and cholecystectomy in the female gender. It has been reported that in 80% of such patients, a double gallbladder is also present. The age at the diagnosis of a double CD varies between newborns and about 75 years [24]-[28].

Double CD is associated with a double gallbladder 80% of the time. The Harlaftis classification divides these variants into 2 groups. Type 1 has a single CD

and a septum that divides a gallbladder into two. Type 1 can further be divided into 3 subgroups: septated, V shaped, and Y shaped. Type 2 describes accessory gallbladders, each with individual CDs. In 2010, Causey *et al.* described a case of septated type 1 with 2 CDs that are attached to each other, and they postulated that the septation extended all the way down to the level of the CBD. Because of the aberrant anatomy, double CD cases predispose patients to higher risks of complications and converting laparotomy. Laparoscopic cholecystectomy has been done successfully with very few instances [28]-[31].

Anatomical variations occurring in the bile ducts, including the duct of Luschka, are very common. Anatomical variants of the biliary system are the secondary causes of bile leakage. They are classified into four types: the Luschka duct, also referred to as subvesicular or supravesicular duct; cystohepatic canal, also called the cholecysto-hepatic duct; segmental or sectoral bile duct variations; and aberrant bile ducts. The Luschka ducts are the most common abnormalities, and their incidence rate varies from 12% to 50%. In most cases, small bile ducts originate from the right hepatic lobe, and they do not open into the gallbladder. This differentiates them from the actual cholecystohepatic ducts. Cystohepatic ducts drain a part of the right lobe into the cystic duct or the gallbladder. Several studies have shown that the combined incidence rate of cystohepatic and cholecystohepatic canals varies from 0.2% to 2.3%. Aberrant bile ducts are a rare anatomical variant comprising the bile ducts within the vesicular fossa's connective tissues [32] [33].

4. Conclusion

We report on the case with a rare anomaly presented in the form of an accessory bile duct between the gallbladder and the common bile duct, as well as with an accessory duct of Luschka, concomitant with extrahepatic bile ducts of significantly enlarged size with large intraductal stones and cholecystoduodenal fistula. The combination of these pathologies and anomalies is an extremely rare case.

Informed Consent

The patient has provided informed consent.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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