



Duplicated Gallbladder: an Incidental Anatomical Variation in a Patient with Symptomatic Cholelithiasis

Ludmila Baltaga¹, Dimosthenis Chrysikos², Spiros Delis¹, Charina Triantopoulou¹,
Dimitrios Filippou¹, Vasileios Protogerou¹, Theodore Troupis³

¹ National and Kapodistrian University of Athens, Athens, Greece

² Faculty of Medicine, National and Kapodistrian University of Athens, Athens, Greece

³ Department of Anatomy, School of Medicine, Faculty of Health Sciences, National and Kapodistrian University of Athens, Athens, Greece

Corresponding author: Dimosthenis Chrysikos, Faculty of Medicine, National and Kapodistrian University of Athens, Athens, Greece; Email: dixrys@yahoo.gr

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Abstract

Congenital malformations of the biliary tract represent a relatively rare entity with which surgeons, radiologists and clinicians are not adequately familiarized. We present a rare case of gallbladder duplication in a 40-year-old female, with the accessory cystic duct entering the left hepatic duct, which depicts the fifth reported case in the international bibliography. Our case illustrates the importance of detailed knowledge of anatomical malformations of the biliary tree, serving the purpose of a preoperative diagnosis of symptomatic cholelithiasis. It is also of paramount importance to take under consideration biliary tract malformations to avoid inadvertent complications such as biliary duct injuries in case of laparoscopic cholecystectomy.

Keywords

accessory gallbladder, anatomical variations, biliary tract, duplicated gallbladder, malformations

INTRODUCTION

Congenital malformations of the biliary tract depict a key surgical challenge in the form of accurate diagnosis and safe operative intervention during the most frequent operation of the biliary tree, which is laparoscopic cholecystectomy. Their occurrence affects up to 47% of the general population.^[1] Among these, gallbladder duplication carries an estimated incidence of 1 in 4000 to 5000 births.^[2] From 1926 through January 2022, a total of 62 instances were reported, with only a few of them being diagnosed preoperatively.^[3,4] Nevertheless, the rarest form of gallbladder duplication remains that of an accessory gallbladder emerging from the

left hepatic duct. Thus, to our knowledge, this represents the fifth reported case in the international literature of the forenamed malformation.

Our case illustrates the importance of a high index of suspicion in patients with recurrent symptoms of cholelithiasis who have been subjected to cholecystectomy, as well as a comprehensive knowledge of the biliary tract's anatomical variations, avoiding diagnostic and radiological imaging interpretations' errors, as well as intraoperative bile duct injuries. The present case is reported in line with the Surgical Case Report (SCARE) guidelines.^[5]

CASE REPORT

A 40-year-old Caucasian female presented with recurring symptoms of upper abdomen pain. Past medical history was unremarkable. Clinical examination revealed tenderness in the epigastrium and the upper right quadrant. Laboratory examinations were within the normal range. An abdominal ultrasound (US) depicted a gallbladder containing stones and another anatomical entity that fell under the differential diagnosis of either a cystic structure or an accessory gallbladder. Afterwards an abdominal computed tomography (CT) was performed that revealed a duplicated gallbladder. A separate artery was seen arising from the left hepatic artery and supplying the cephalic gallbladder. The duplicated gallbladder was situated directly above the left hepatic artery, from which the accessory cystic artery arose providing its blood supply. A magnetic resonance cholangiopancreatography (MRCP) was performed, which confirmed the existence of an accessory gallbladder (Figs 1-3), with its cystic duct originating from the left hepatic duct (Fig. 4). Due to the mild clinical image and the lack of symptoms' recurrence, no surgical intervention was scheduled. Conservative treatment with spasmolytics and lifestyle or dietary optimization was advised.

DISCUSSION

The first case of a duplicated gallbladder was reported in 1675 by Blasius et al., and Boyden et al. proposed the first classification of a gallbladder duplication.^[6]

During the gestational period, extrahepatic bile ducts develop from the distal part of the hepatocystic diverticulum. By the end of the 4th week, both the cystic duct and the gallbladder bud are developed. Moreover, by the 5th gestational week, the common bile duct and the hepatic ducts appear, while the duodenum rotates to the right and the developing common bile duct becomes displaced dorsally, taking its final anatomical position.



Figure 1. MRCP image of the gallbladder in the normal anatomic location.

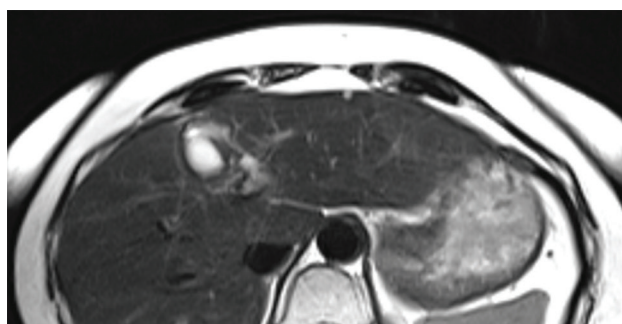


Figure 2. MRI-T2 axial image depicting a second gallbladder between liver segments II and IV.

In the rare case of gallbladder anlage duplication, elements of multiple gallbladders enter the bile duct through the same cystic duct. Thus occurs the anatomical malformations septate, V-shaped, or Y-shaped gallbladder. The V-shaped gallbladder consists of two distinctive bodies that

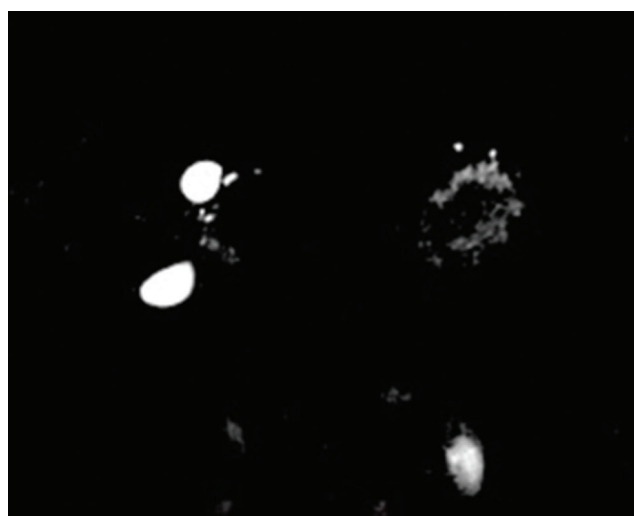


Figure 3. MRCP showing the two gallbladders.

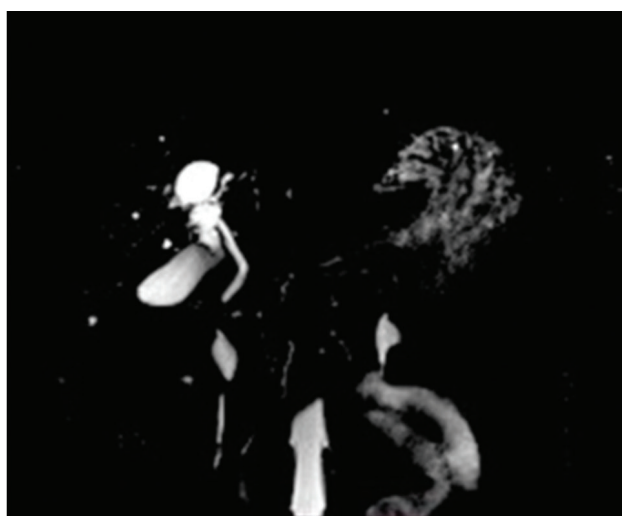




Figure 4. MRCP picture of the second gallbladder's cystic duct entering the left hepatic duct.

share the same cystic duct. In Y-shaped type, two distinctive gallbladders, with distinctive cystic ducts merge and end up as one structure in the bile duct.

In the cases of an accessory gallbladder, the main and the accessory cystic ducts join the bile duct separately. Likewise, cases of triple gallbladders have been observed.^[7,8] To highlight the importance of our case, to date there have been reported 17 cases of triple gallbladder, and only 4 of an accessory gallbladder entering the left hepatic duct.^[9]

Based on the above embryological development, Harlaftis, proposed a double gallbladder classification into two groups: the split primordium (*Vesica fellea divisum*) and the accessory gallbladder (*Vesica fellea duplex*), with a third miscellaneous group reserved to the malformations that do not fall into the two above-mentioned categories, such as a triple gallbladder (Fig. 5).^[10] According to this classification, our case falls into the left trabecular group.

Numerous other classifications for gallbladder duplications have been proposed except from Boyden et al. Gross et al. proposed a new classification by A-F type^[6,11], while Mochizuki and Makita introduced a different classification

based on types I-VI.^[12] Hence, our case corresponds to type H by Boyden et al., type E by Gross et al., and type VI by Mochizuki and Makita.

In addition, patients with gallbladder duplication are more likely to experience a higher incidence of gallbladder cancer.^[13]

In the differential diagnosis, entities that should be taken into consideration are choledochal cyst, focal adenomyomatosis, and gallbladder diverticula. The diagnostic algorithm, although not standardized, begins with abdominal ultrasonography, followed by MRCP, computed tomography, and/or endoscopic retrograde cholangiopancreatography (ERCP).^[14]

Due to its infrequent manifestation, there are no guidelines regarding the diagnosis and management of incidentally finding accessory gallbladders. Concurrent removal of both gallbladders at surgery is recommended, in order to minimize cholecystitis or biliary colic recurrence. The proposed treatment in symptomatic cases remains laparoscopic cholecystectomy with intraoperative cholangiography.^[15]

Thus, four more cases of a duplicated gallbladder with its accessory cystic duct arising from the left hepatic duct have been reported (Table 1). In most cases, ultrasonography raised the suspicion of an anatomical variation, yet failed to confirm the diagnosis. Subsequently, the definitive diagnosis was confirmed with either a laparotomy or ERCP. In all cases, cholecystectomy was performed. In our case, the diagnosis was confirmed with MRCP, and no ERCP or laparotomy was performed. Due to the non-recurrent character of the patient's symptoms, a cholecystectomy was not advised.

The aim of this rare case, is to shed light on anatomical variations or malformations of the biliary tree. Overall, the rarity of such cases may contribute to missed diagnosis. In order to depict biliary tree anomalies, imaging techniques that delineate the biliary anatomy of each case is of paramount importance.

CONCLUSION

To conclude, what is peculiar about duplicate gallbladder case reports is that it might be a risk factor for symptomatic or recurrent cholelithiasis. Preoperative knowledge of such biliary tract variations, can identify and thereby guide conservative or surgical decision with laparoscopic cholecystectomy. In case of surgical management, resection of both gallbladders is ideal to prevent recurrence of disease and avoid inadvertent complications such as hemorrhage or bile duct injuries.

Conflict of Interest

The authors declare no conflict of interest.

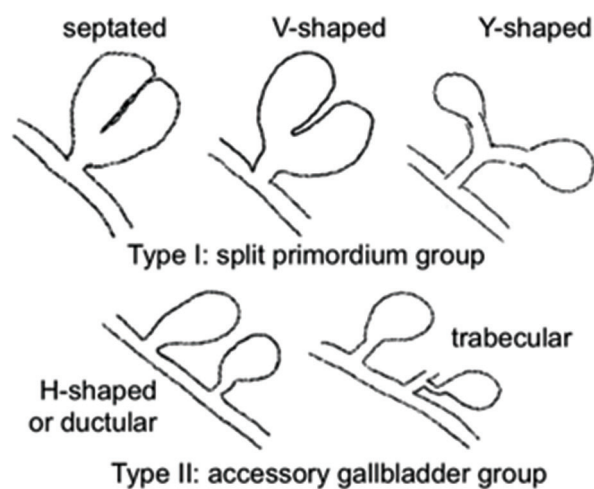


Figure 5. Classification of double gallbladder according to Harlaftis et al.^[10]

Table 1. Previous reported cases of a duplicated gallbladder with its accessory cystic duct arising from the left hepatic duct

Title	Year	Sex	Age	Presentation	Laboratory findings	US	CT	MRCP	ERCP	Laparotomy findings	Definitive diagnosis	Cholecystectomy	Remarks
Gross et al. ^[10]	1969	F	31	Fever, jaundice, weakness	NS	NS	NA	NA	NA	Duplicated GB	Laparotomy	Open	
Gorecki et al. ^[16]	1998	F	69	Right UAP, EP	Uneventful	Cystic structure	Accessory, partially intrahepatic GB		Accessory, partially intrahepatic GB	Accessory GB without a cystic duct	CT, ERCP	Open	
Kim et al. ^[17]	2009	M	78	Intermittent UAP, jaundice, anorexia	bil ↑, ALP ↑	Uneventful	Dilated CBD	Dilated CBD	Intrahepatic choledochal cyst with multiple stones, mildly dilated CBD	5 cm cystic structure resembling a GB, filled with stones	Laparotomy	Open (second)	10 years after an open cholecystectomy
Kawanishi et al. ^[18]	2010	M	75	EP, body weight loss	Uneventful	Cystic structure filled with stones	Cystic structure filled with stones	Cystic structure filled with stones	Accessory GB	Accessory GB	ERCP	Open	Adenocarcinoma

F: female; M: male; UAP: upper abdominal pain; EP: epigastric pain; NS: not specified; NA: not applicable; GB: gallbladder; CBD: common bile duct; US: ultrasonography; CT: computed tomography; MRCP: magnetic resonance cholangiopancreatography; ERCP: endoscopic retrograde cholangiopancreatography; bil: bilirubin, ALP: alkaline phosphatase.

Author contributions

L.B., D.C., and S.D.: conceptualization; L.B., D.C., D.F., and V.P.: manuscript writing, editing, and final revision; S.D., C.T., and L.B.: data collection; T.T., D.F., and V.P.: visualization and technical support; D.C. and T.T.: supervision. All authors approved the final draft of the manuscript.

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Удвоенный жёлчный пузырь: случайное анатомическое изменение у пациента с симптоматическим холелитиазом

Людмила Балтага¹, Димостенис Крисикос², Спирос Делис¹, Карина Триантопулу¹, Димитриос Филипу¹, Василейос Протогеру¹, Теодор Трупис³

¹ Афинский национальный университет имени Каподистрии, Афины, Греция

² Факультет медицины, Афинский национальный университет имени Каподистрии, Афины, Греция

³ Кафедра анатомии, Институт медицины, Факультет медицинских наук, Афинский национальный университет имени Каподистрии, Афины, Греция

Адрес для корреспонденции: Димостенис Крисикос, Факультет медицины, Афинский национальный университет имени Каподистрии, Афины, Греция; E-mail: dixrys@yahoo.gr

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Резюме

Врожденные пороки развития жёлчевыводящих путей представляют собой относительно редкое заболевание, с которым хирурги, рентгенологи и клиницисты недостаточно знакомы. Мы представляем редкий случай удвоения жёлчного пузыря у 40-летней женщины с переходом добавочного пузырного протока в левый печёночный проток, что представляет собой пятый зарегистрированный случай в международной библиографии. Наш случай иллюстрирует важность детального знания анатомических пороков развития жёлчного дерева для целей предоперационной диагностики симптоматической жёлчнокаменной болезни. Также крайне важно учитывать пороки развития жёлчевыводящих путей, чтобы избежать непреднамеренных осложнений, таких как повреждение жёлчных протоков в случае лапароскопической холецистэктомии.

Ключевые слова

добавочный жёлчный пузырь, анатомические варианты, жёлчные пути, удвоенный жёлчный пузырь, пороки развития
