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Duplication of the Gallbladder: A Rare Anomaly in the Surgical Practice Mostly Seen in Turkey; Review of the Literature and Report of a Case

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1. Abstract

Gallbladder duplication (GBD) is a rare biliary anomaly that can be diagnosed in the prenatal, pediatric and all other periods of life. Most bile ducts and vascular injuries can be encountered during laparoscopic cholecystectomy (LC) when performed without achieving clear definition of the biliary system anatomy. Therefore, it is quiet important to make a clear diagnosis of this rare variation before any planned elective surgical intervention. Although abdominal Ultrasonography (USG) is used liberally for the diagnosis of abdominal pain etiologies, the incidence of Gallbladder duplication discovered at this stage remains low. Abdominal Computerized Tomography (CT) and Magnetic Resonance Cholangiopancreatography (MRCP) provide better non-invasive tools for the precise description of biliary tree preoperatively. İt is of upmost importance to emphasize on meticulous dissection of the calot triangle, with complete removal of all connective tissues and fat before ligating any tubular structure, and not to hesitate to perform peroperative cholangiography in case of any suspected bizarre anatomy. İt is very interesting to note that more than one third of Gallbladder duplication cases have been reported recently from the middle east countries, especially Turkey.

2. Introduction

Duplication of gallbladder is a rare congenital anomaly resulting from abnormalities in embryogenesis during the fifth and sixth weeks of gestation [1,2]. Whether it is a duplication or triplication or separation of the gallbladder, those rare anatomical variations have been described 210 times in the literature in the form of case reports or short letters or papers presenting extremely small patient groups diagnosed with similar anomalies [3,4]. Although an incidence of 1 in 3000-4000 has been mentioned in the largest autopsy studies, and it is said that the incidence is twice more common in women than in men, the exact rate can never be told5,6. Latest advances in radiology with the widespread use of ultrasonography, Computerized Tomography and Magnetic Resonance Cholangiopancreatography helped discovering the duplication of gallbladder during routine evaluations of abdominal pain for both surgical and non-surgical conditions. This congenital malformation that may lead to disasterous bile duct injuries during cholecystectomy remains underestimated by many surgeons and keep to be a surprising challenge during surgery especially when not diagnosed preoperatively [7,8]. Recent studies emphasized the fact the gallbladder duplication can be diagnosed prenatally and can present in the form of jaundice or abdominal pain in the early childhood [1,9]. İt is crucial to remind the surgeons of this rare anomaly to ensure safer cholecystectomies and avoid making decisions before clarifying biliary anatomy in the Calot triangle.

3. Case Report

A 38 Years old female admitted to the hospital for elective Laparoscopic Cholecystectomy (LC). She had a history of several attacks of acute abdominal pain in the right upper quadrant associated with nausea, vomiting and anorexia. The patient was diagnosed six months earlier to be suffering from acute cholecystitis complicating a single gall stone measuring 25 mm without dilatation of both intra and extrahepatic biliary ducts as described by an upper

abdominal ultrasonography performed in a suburban hospital. Two months later the patient suffered a second attack where computerized tomography of the abdomen was performed to show a gallbladder duplication with the presence of one big stone occupying almost the whole space of one gallbladder cavity, while the other space was empty from any notable pathology (Figure 1). Although liver function tests proved always normal, an upper abdominal Magnetic Resonance series was performed to rule out any other anomaly of the biliary tree. Despite that double cystic duct could not be clearly visualized, biliary tree anomaly was certainly excluded. During laparoscopic cholecystectomy, two gall bladders looking fused externally and lined by a single peritoneal layer were noted to be sharing a single mesentery and a wide calot triangle. Meticulous dissection starting from the gallbladder infundibulum revealed a V shaped double cystic ducts emerging separately from both gallbladders and joining each other before draining into the common bile duct. The distal part of both cystic ducts was clipped twice after ligating the cystic artery to continue with retrograde dissection of the duplicated gallbladders from the liver bed (Figure 2). Single Jackson-Pratt drain was placed beneeth the liver and surgery was completed with the exteriorization of gallbladders and closing fascia and skin. The surgical procedure was smooth and uneventful and lasted for 90 minutes approximately. Patient was discharged the other day after removing her drain without any recorded pathology and was included as part of an outpatient's follow up plan.

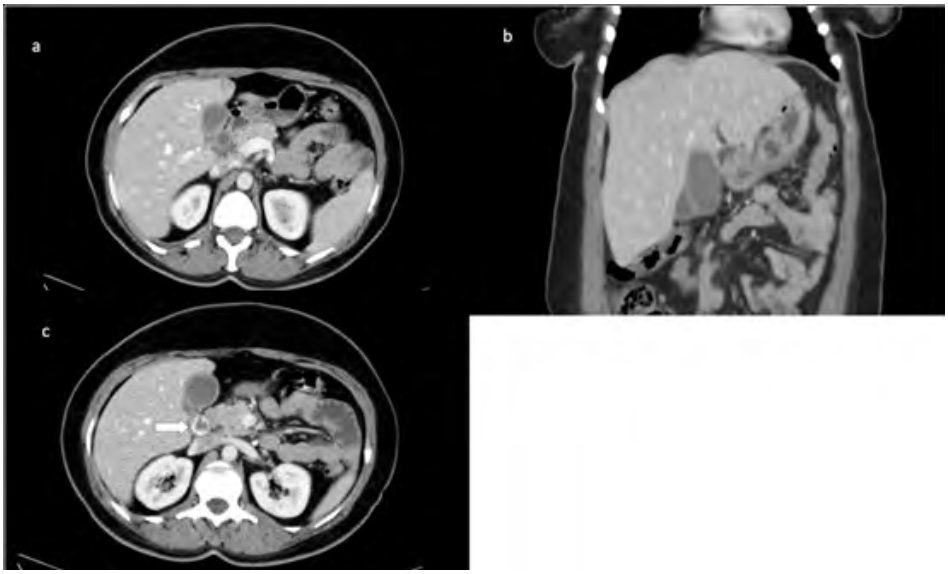

Figure 1: Computed tomography showing preoperative gallbladder duplication a: axial section b: coronal section c: gallbladder stone marked with arrow

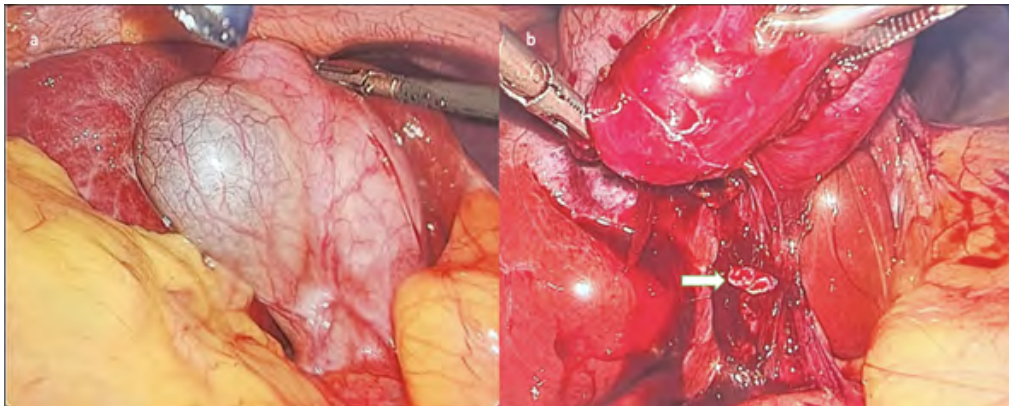

Figure 2: a: Double gallbladder covered with a single peritoneal layer and having wide calot b: V-shaped cystic duct marked with arrow

4. Discussion

Although an extremely rare biliary anomaly, gallbladder duplication has attracted the attention of obstetricians during routine prenatal examination with ultrasonography. Historically, gallbladder duplication was first reported in 1674 by Blasius at autopsy of 2 years old boy in Amsterdam [1]. In 2007, Sifakis et al. reported the first prenatal diagnosis of gallbladder duplication in 26 years old mother discovered with ultrasonography at 32 weeks of gestation, and not described by the same operator in the screening ultrasonography performed at 22 weeks of gestation [9]. It is worth to remind here that the fetal gallbladder is usually visualized in 65-82% of fetuses at 24-27 weeks of gestation [9]. In 2018, Maggi et al. repotred ductular duplicate gallbladder diagnosed with prenatal ultrasonography and confirmed on postnatal day 12 with 3D Magnetic Resonance Cholangiopancreatogaphy (MRCP) in 38 years old primipara mother [1]. Since 1953, different papers reported concurrent entities alongside gallbladder duplication. This includes duodenojejunal diverticulum, anteriorly displaced right hepatic artery, heterotopic gastric mucosa, ectopic thyroid, duodenal atresia, duodenal duplication cyst, gastrointestinal atresia and biliary cirrhosis. Most of the reports were relying on cadaveric and incidental surgical findings, or coincidentally discovered during radiological evaluation of abdominal pain of unknown etiology [10]. Szczech et al and Udelsman and Sugarbaker reported gall bladder duplication associated with biliary duct malformation or aberrant hepatic duct [11,12]. Gupta et al. reported two cases of gallbladder duplication in neonates associated with duodenal atresia in one patient and with pyloric, ileal and colonic atresia in the other [13]. İn 2021, Kumar S et al. reported an extremely rare case of choledochal cyst associated with gallbladder duplication in a 6 years old boy, and declared that it is the third case being reported in the medical literature [8]. In 1926, Boyden described his first classification of gallbladder duplication based on the anatomy of cystic duct and the way it drains into the biliary tree [10]. İn 1936, Gross described congenital anomalies of gallbladder and classified them into 6 types [10]. İn 1977, Harlaftis further modified the classification by describing two main types of duplications, type 1 which is a vesica fella divisa or bilobed gallbladder with single cystic duct, and type 2 named vesica fella duplex or true duplication of gallbladder with two different cystic ducts, with the respective incidence of 45.1% and 54.9% [7,14]. True duplication is further classified into Y shaped type cystic duct, where both ducts unite before entering the common bile duct, and H shaped type where each cystic ducts drain independently into the bilary tree [15]. More variations have been reported describing accessory cystic duct connecting to the right or left hepatic ducts. The most common variant found in the literature is the H type with an estimated incidence of 48.6% of the whole cases [10]. It was quite interesting to note that while 33 new cases of Gallbladder duplication have been reported worldwide between the years 2000 and 2022, the majority of case reports were from India (7 cases), USA (5 cases) and Turkey (5 cases), the highest prevalence of case per 100,000 population being in Turkey (Table 1).

Gallbladder duplication has no specific symptoms related to the anatomical anomaly by itself, and most complaints are related to cholelithiasis and/or cholecystitis eventually. This anomaly can increase the incidence of gall stones because of inadequate drainage of bile from the gallbladder, and therefore is associated with higher incidence of related complications [14]. Although cholecystectomy is the most commonly performed operation in surgery, prophylactic cholecystectomy is never recommended for incidentally found gallbladder duplication [1,6,10]. On the other hand, omission of an accessory gallbladder can result in persistence of symptoms and need for reoperation. Therefore, an accurate preoperative diagnosis is crucial [3]. Lack of awareness with inadequacy of imaging methods are possible reasons of mostly reported complications as well as overlooking additional gall bladders before and during surgery [2]. Since most abdominal pain investigation starts with ultrasonography, most of the gallbladder duplication cases can be diagnosed at this stage, though ultrasound may not give sufficient anatomical information. It is very much surprising that although ultrasonography is used that frequent in the evaluation of abdominal pain and suspected gallbladder diseases, very few cases of gallbladder duplication have been diagnosed at this stage5. Computerized Tomography (CT) is considered to be more accurate and objective with fine cuts and the capability of making anatomic reconstructions with both coronal and sagittal views. In cases of suspected bile ducts anomalies, (MRCP) is accepted as a standard non-invasive imaging method that can provide a sensitivity of 95% and specificity of 85% compared to ERCP [16]. In case of suspicion of bile duct anomalies during cholecystectomy, an intraoperative cholangiography is highly recommended to minimize the risk of accidental biliary damage [15]. While in the past some authors encouraged open cholecystectomy to prevent the risk of overlooking the diagnosis of gallbladder duplication and variations of the biliary system, successful management of gallbladder duplication with laparoscopic cholecystectomy became the worldwide accepted gold standard of treatment [17,18].

Table 1: Duplication of gallbladder reported since 2000

Authors	Year reported	Cases/patients characteristics	Type of duplication	Geographic location
Haghighi et al.	2000	68 years old female	2	Australia
Shirahane et al.	2002	61 year old female	2	Japan
Goel et al.	2003	25 year old female	2-H type	India
Ozmen et al.	2003	34 year old female	1	Turkey
Valadez et al.	2004	44 year old male	1	Mexico
Barut et al.	2006	55 year old female	1	Turkey
Asbury et al.	2007	70 year old male	1	USA
Desolneux et al.	2009	61 year old female	1	France
Causey et al.	2010	15 year old female	1	USA
Akhilesh et al.	2011	42 year old female	2-H type	India
Bulus et al.	2011	56 year old male	2-Y type	Turkey
Hassan et al.	2012	83 year old female	2	UK
Shiba et al.	2014	38 year old female	1	Japan
Koszman et al.	2014	62 year old female	1	Denmark
Yasir et al.	2014	22 year old male	2-H type	İndia
Pillay	2015	56 year old male	1	Canada
Szczech et al.	2015	26 year old female	1	USA
Goh et al.	2015	28 year old male	1	UK
Gupta et al.	2016	2 day and 12 day old males (two cases)	1	India
Rawahi et al.	2016	42 year old female	1	Oman
Rajapandian et al.	2017	28 year old male	1	India
Ghaderi et al.	2018	38 year old male	2	USA
Romero et al.	2018	50 year old female	1	Ecuador
Umberto et al.	2018	Female newborn with prenatal diagnosis	2	Italy
Babur et al.	2018	47 year old female	2-H type	Turkey
Vezakis et al.	2019	63 year old female	2	Greece
Arif et al.	2019	38 year old female	1	İraq
Boukoucha and Dhieb	2020	58 year old female	1	Tunisia
Yilmaz et al.	2020	57 year old male	2-H type	Turkey
Singh et al.	2021	60 year old female	2-Y type	USA
Kumar et al.	2021	6 year old male	2-H type	İndia
Gadour et al.	2021	35 year old female	2-Y type	Sudan
Jefferys et al.	2022	52 year old female	2-Y type	England

5. Conclusion

Gallbladder duplication with the probable associated anomalies are extremely rare entities that may be frequently overlooked during investigation and diagnosis of abdominal pain. Being unexpected during surgery, they may complicate its course and end up with high rates of morbidites. It is of upmost importance to define the anatomy of biliary tree in order to reduce the risk of iatrogenic biliary and vascular injury during even routine cholecystectomy. Laparoscopic cholecystectomy is always the surgical treatment of choice, with emphasis on meticulous dissection of calot triangle and intraoperative preformation of cholangiography to identify clearly the biliary anatomy whenever unclear. It is quite difficult to claim the exact incidence of gallbladder duplication in the normal population, and multicentric objective and prospective radiological studies should be done on asymptomatic individuals from different ethnicities to come up with results that reflect the reality of the situation.

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References

1. Maggi U, Farris G, Carnevali A, Borzani I, Clerici P, Agosti M, et al. Prenatal and accurate perinatal diagnosis of type 2H or ductular duplicate gallbladder. BMC Pediatr. 2018; 18(1):38.
2. Yasir M, Kapoor M, Aiman A, Suri A. Double gall bladder: A rare anomaly diagnosed during the laparoscopic cholecystectomy. J Sci Soc. 2014; 41(1):45-6.
3. Koszman B. Gall-bladder duplication - case report. Pol Przegl Chir. 2014; 86(9):433-5.
4. Causey MW, Miller S, Fernelius CA, Burgess JR, Brown TA, Newton G. Gallbladder duplication: evaluation, treatment, and classification. J Pediatr Surg. 2010; 45(2):443-6.
5. Goiney RC, Schoenecker SA, Cyr DR, Shuman WP, Peters MJ, Cooperberg PL. Sonography of gallbladder duplication and differential considerations. AJR Am J Roentgenol. 1985; 145(2):241-3.
6. Bulus H, Koyuncu A, Coskun A. Preoperative diagnosis of double gallbladder: A case report. Turk J Gastroenterol. 2012; 23(2):172-4.
7. Singh JP. Duplication of the Gallbladder as an Operative Surprise: A Case Report with Review of the Literature. Case Rep Surg. 2021; 2021:6668302.
8. Kumar S, Kumar A, Singh VP. Gallbladder duplication in a child with choledochal cyst: a rare surgical surprise. BMJ Case Rep. 2021; 14(7):e245109.
9. Sifakis S, Mantas N, Koumantakis G, Koukoura O. Prenatal diagnosis of gallbladder duplication. Ultrasound Obstet Gynecol. 2007; 30(3):362-3.
10. Vezakis A, Pantiora E, Giannoulopoulos D, Fontara S, Kontis E, Poludorou A, et al. A duplicated gallbladder in a patient presenting with acute cholangitis. A case study and a literature review. Ann Hepatol. 2019; 18 (1):240-5.
11. Szczech EC, Parikh SP, Beniwal J. Dual gallbladders with dual cholecystitis. Austin J Surg. 2015; 2(2):1055.
12. Udelsman R, Sugarbaker PH. Congenital duplication of the gallbladder associated with an anomalous right hepatic artery. Am J Surg. 1985; 149(6):812-5.
13. Gupta R, Gupta S, Sharma P. Gallbladder duplication associated with gastro-intestinal atresia. J Neonatal Surg. 2016; 5(2):14.
14. Pillay Y. Gallbladder duplication. Int J Surg Case Rep. 2015; 11:18-20.
15. Desolneux G, Mucci S, Lebigot J, Arnaud JP, Hamy A. Duplication of the gallbladder. A case report. Gastroenterol Res Pract. 2009; 2009:483473.
16. Kumar A, Mohanty NR, Mohanty M, Dash S. Comparison of MRCP and ERCP in the evaluation of common bile duct and pancreatic duct pathologies. Front Med Technol. 2023; 5:946555.
17. Babur T, Ozveri E, Ertem M. Gallbladder duplication: A rare case treated laparoscopically. ACU Saglik Bil Derg. 2020; 11(1):186-9.
18. Barut I, Tarhan OR, Dogru U, Bulbul M. Gallbladder duplication diagnosed and treated by laparoscopy. Eur J Gen Med. 2006; 3(3):142-5.