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Duplication if the gallbladder in a patient with right colon cancer. A case report and review of the literature review.

AIM: *Duplication of the gallbladder is a very rare congenital variant, showing an incidence of about 1 in 4000 in the population, occurring twice as often in women than in men. In the literature only a few cases of prenatal diagnosis are reported. The knowledge of the existence of this anatomical variable is very important in order to avoid complications and iatrogenic damage during interventional and surgical procedures involving the biliary tract or adjacent organs.*
CASE REPORT: *A 79-year-old patient was admitted to our Hospital in May 2021 presenting abdominal pain. During hospitalization, a 5cm adenocarcinoma of the ascending colon was found. During surgery the known accessory gallbladder was found strongly adhering to the proximal transverse colon. The challenging viscerolysis maneuvers caused a lesion on one of the gallbladders, so we decided to proceed with cholecystectomy of both gallbladders.*
RESULTS AND DISCUSSION: *Duplication of the gallbladder is a rare congenital anatomical variant and requires special attention to the biliary and arterial anatomy in order to avoid iatrogenic damage. This variant can complicate surgical treatment for complications that need urgent treatment, such as cholecystitis. Currently the technique of choice for the evaluation of the biliary tree is magnetic resonance cholangiography. Laparoscopic cholecystectomy is the treatment of choice.*
CONCLUSION: *Surgeons should be aware of the different forms of presentation of gallbladder pathologies, also the ones that are not "the standard" of routine. Detailed preoperative study is essential to avoid a missed diagnosis.*

KEY WORDS: Gallbladder, Mininvasive surgery, Variant, Anatomical

Introduction

Duplication of the gallbladder is a very rare congenital variant, showing an incidence of about 1 in 4000 in the population, occurring twice as often in women than in men [1].

The diagnosis of this anatomical variation of the biliary tract is often incidental and is frequently highlighted during surgery.
In the literature only a few cases of prenatal diagnosis are reported. On prenatal ultrasound, the duplication can be seen as two cystic structures with fluid content in the upper right quadrant of the fetus. In patients with this anatomical variant, surgery may be proposed even if asymptomatic, given the high probability of complications [3].
Therefore, the knowledge of the existence of this anatomical variable is very important in order to avoid complications and iatrogenic damage during interventional and surgical procedures involving the biliary tract or adjacent organs. Preoperative anatomical evaluation of the biliary tree anatomy with instrumental examinations is crucial [2,4].

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ABBREVIATIONS

CT: Computed Tomography
ERCP: Endoscopic Retrograde Cholangiopancreatography
ESD: Endoscopic Submucosal Dissection
MRCP: Magnetic Resonance Cholangiopancreatography
PTC: Percutaneous Transhepatic Cholangiography

Case Report

We present a case of an accessory gallbladder in a patient who underwent video laparoscopic right hemicolectomy for cancer of the right hepatic flexure of the colon.
A 79-year-old Italian female was admitted to our institution with abdominal pain for a few days. On admission, she was afebrile and there was no jaundice. The patient presented a BMI of 31.2. During hospitalization she ran blood tests, colonoscopy and chest and abdomen CT scan, that showed a right colon cancer; as a collateral finding, a duplication of the gallbladder was diagnosed (Fig. 1).
The common hepatic, intrahepatic and extra hepatic biliary ducts had a normal caliber. The two gallbladders seem to share the same cystic duct.
The endoscopic examination revealed a 5 cm lesion of the distal ascending colon; on bioptic samples the lesion was found to be compatible with adenocarcinoma. Therefore, the clinical case was then discussed at the multidisciplinary oncologic meeting and surgery was advocated. During the right colonic flexure detachment, the accessory gallbladder was found to be strongly adhered to the proximal transverse colon.
The challenging viscerolysis maneuvers caused a lesion on one of the gallbladders, so we decided to proceed with cholecystectomy of both gallbladders (Figs. 2, 3).
The postoperative course was uneventful and the patient was discharged on the sixth postoperative day. Histopathological examination revealed an Y-type duplication of the gallbladders with adenomyosis; each gallbladder had a mucosal layer, tunica muscularis and subserosa, but they share a single tunica serosa.

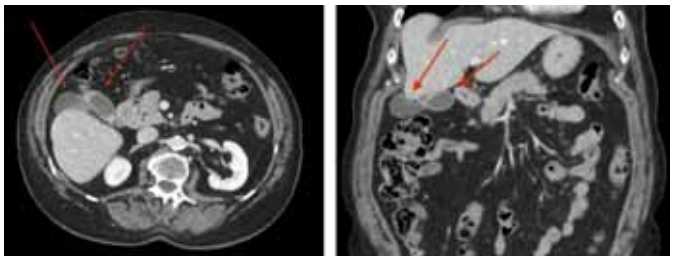

Fig. 1: Preoperative CT scan.

Discussion

Duplication of the gallbladder is a rare congenital anatomical variant and is established as a morphological abnormality. It is usually detected in 0.02% of autopsies or 0.03% of radiological examinations, with an incidence of 1 in 4000-5000 people. It requires special attention to the biliary and arterial anatomy in order to avoid iatrogenic damage [1].
The pathogenesis is unclear. Boyden in 1926 stated that numerous accessory outgrowths would already form during the 5th-6th week of embryological life, arising from a system of ducts called "hepatic antrum". They usually regress, but the persistence appeared to cause a duplication of the gallbladder [5,6]. The true incidence is not easy to estimate, as this type of anomaly can often be overlooked as it is asymptomatic and is often an occasional finding during surgery. In many studies the calculation of the incidence is therefore based on studies conducted on cadaver. Historically, the first case of accessory gallbladder dates back to 1674 and was found during an autopsy.
Only in 1911 did Sherren document for the first time an accessory double gallbladder in vivo [7].

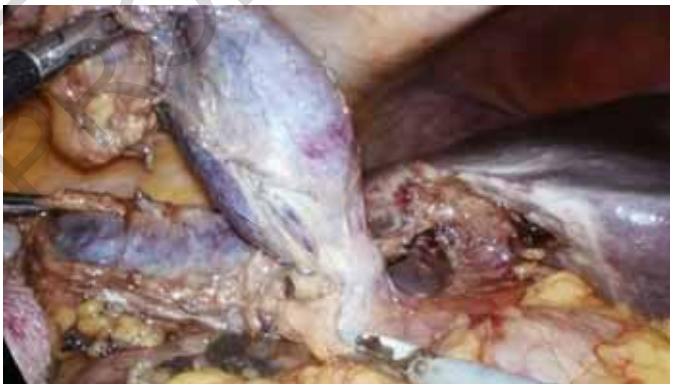

Fig. 2: Intraoperative finding.

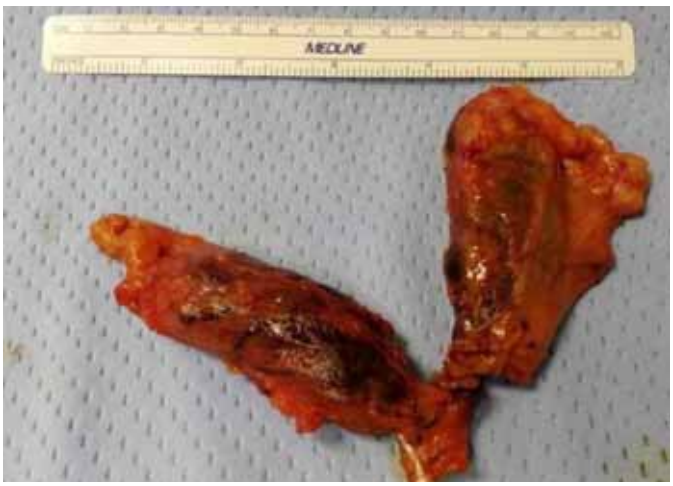

Fig. 3: The specimen shows the duplication of the gallbladder.

This abnormality is usually classified into two basic types by Harlaftis et al, according to the way that bile duct merged into the biliary system. Type I or split primordial group, with an incidence of 45.1%, is characterized by bilobed gallbladder that has the same embryological origin, a longitudinal septum separates the lumen into 2 chambers; type II or accessory gallbladder, with an incidence of 54.9%, when there are two completely separate gallbladders, each with an individual cystic duct for each gallbladder. Some Authors also identify a type III, that includes gallbladders with anatomical anomalies that do not fit either type I or II (Fig. 4). The triplication of the gallbladder is very rare. Only a few cases have been reported in the literature, one of which was described by Roeder et al, who demonstrate a case where two of the gallbladders were respectively affected by acute cholecystitis associated with cholelithiasis and papillary adenocarcinoma [1].

If the septum is poorly represented, it is no longer defined as a double gallbladder. It is also necessary to differentiate the duplication of the gallbladder from the diverticulum of the gallbladder, which is instead a localized extroflection composed of all the layers of the gallbladder wall and usually presents a narrow neck[8]

Clinically, patients with this anatomic variant do not have any symptoms. Some authors have shown that these patients have no increased incidence of any biliary tract disease compared to the general population [9]. Pillay et al., hypothesized a possible increase in the incidence of gallbladder stones associated with this variant, suggesting that it may be due to inadequate biliary drainage. Furthermore, some authors have reported two cases of carcinoma in the accessory gallbladder [9].

Differential diagnosis is made with choledochus cysts, gallbladder diverticula, phrygian cap, a layer of pericystic fluid, and focal adenomyomatosis. There are no specific symptoms attributable to this anatomical variant. We can find cholelithiasis, cholecystitis (acute or chronic), cholecysto-cholic fistula, empyema, "porcelain" gallbladder and carcinoma as if it were a normal anatomical scenario [1,11].

According to the available literature (Table I), it is not very clear whether gallbladder duplication is associated with other congenital anomalies. Some Authors stated that in the literature there is no association between a duplicated gallbladder and other anatomical anomalies, while others have reported this anatomical variant in association with biliary tract malformations or with the presence of an aberrant hepatic duct. There are two reported cases of pediatric patients with gallbladder duplication in association with duodenal atresia [7].

The ultrasonographic evaluation for the gallbladder's diseases is the most useful diagnostic tool, it is sensitive in assessing gallstones, cholecystitis and detecting gallbladder anatomical abnormalities, but does not appear to play an important role in diagnosis in these patients. This anomaly is not easily overlooked on ultrasound. Magnetic resonance cholangiopancreatography (MRCP) seems to have a much better diagnostic capability with regard to the anatomical variables of the biliary tract and gallbladder. Both endoscopic retrograde cholangiopancreatography (ERCP) and percutaneous transhepatic cholangiography (PTC) are diagnostic, but they can't be considered routinary diagnostic methods given their invasiveness. Van Steenbergen et al. described a case of an accessory trabecular type gallbladder, diagnosed preoperatively with ERCP, which showed the accessory gallbladder intrahepatic, with biliary discharge into the right intrahepatic duct [1,11]. The possible diagnosis of these congenital malformations is important as they are considered a risk factor for iatrogenic damage of the biliary tract during cholecystectomy [7].

It is not yet clear what is the best approach, although in literature it seems more suitable to proceed with removal of the accessory gallbladder [1]. During surgery,

TABLE I - *Summary of gallbladder duplications reported from 1926 to 2020.*

Author	Year	Number of cases	Type of duplication
Boyden et al [14]	1926	20	/
Udelsman et al [15]	1985	60-years old male	II
Haghigi et al [16]	2000	68-years old female	II
Valadez et al [17]	2004	44-years old male	I
Barut et al [13]	2006	55-years old female	I
Delsoneaux et al [5]	2009	61-years old male	I
Szczech et al [18]	2015	26-years old female	I
Pillay et al [11]	2015	56-years old male	I
Goh et al [12]	2015	28-years old male	I
Ghaderi et al [19]	2018	38-years old male	II
Romero et al [6]	2018	50-years old male	I
Boukoucha et al [2]	2020	54-years old female	I

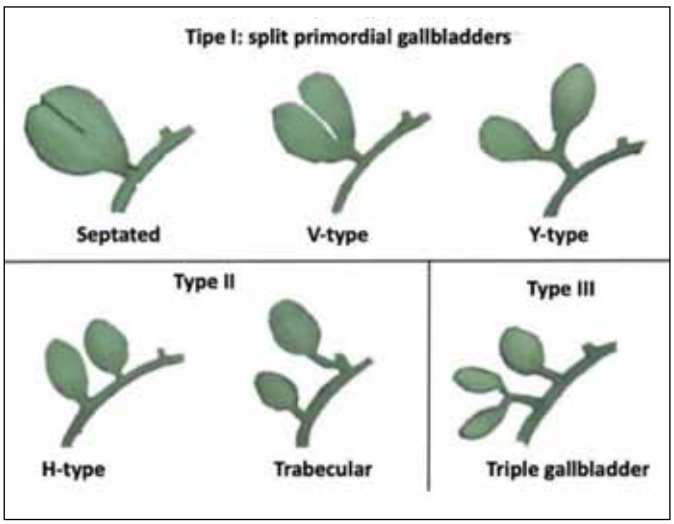

Fig. 4: Harlafti's classification of gallbladder anatomical anomalies.

cholangiography can be a valid aid to perform a careful dissection and recognition of the accessory cystic artery and the approach should be laparoscopic [1]. In the management of a duplicated gallbladder, some Authors established that surgery is not mandatory for the accessory gallbladder discovered accidentally, only if the patient is symptomatic, cholecystectomy should be performed for both gallbladders [6] while other Authors stated that if a double gallbladder is detected during surgery, removal of both gallbladders is indicated in order to avoid complications in the remaining organ. Correct identification of the outlet area of the infundibulum in the cystic and biliary tract is necessary during cholecystectomy so laparoscopic cholecystectomy associated with intraoperative cholangiography is the most suitable intervention in the case of symptomatic gallbladder. The indications in case of double gallbladder in the absence of symptoms remains controversial [7]. The final diagnosis is made with histopathological evaluation, to exclude other lesions that may affect the biliary tract 1.

Conclusion

Duplication of the gallbladder is a rare congenital abnormality which could be associated with other anomalies of the bile duct and vascular systems. Surgery is considered as a critical predisposing factor for iatrogenic bile duct injuries during cholecystectomy and it is only indicated for symptomatic duplication. Surgeons should always be aware of the different forms of presentation of gallbladder pathologies, also the ones that are not "the standard" of routine. Detailed preoperative imaging is essential to avoid a missed diagnosis and MRCP is now the ideal choice of non-invasive imaging modality.

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Thanks to Cozacov et al [1], who made the graphic classification in Fig. 4, from the patient for publication of this case report and accompanying images. A copy of the written.

Riassunto

La duplicazione della colecisti è una variante congenita molto rara, con un'incidenza di circa 1 su 4000 persone ed una frequenza doppia negli individui di sesso femminile.
In Letteratura sono riportati solo pochi casi di diagnosi prenatale. La conoscenza dell'esistenza di questa variabile anatomica è fondamentale per evitare complicanze e danni iatrogeni durante le procedure interventistiche e chirurgiche coinvolgenti le vie biliari o gli organi adiacenti.
In questo lavoro riportiamo il caso di un paziente di sesso femminile di 79 anni ricoverata presso il nostro Ospedale a maggio 2021 per comparsa di dolore addominale da alcuni giorni. Durante il ricovero è stata fatta diagnosi di adenocarcinoma del colon ascendente. Dopo esecuzione degli esami di staging viene confermata la diagnosi di adenocarcinoma del colon senza ripetizioni a distanza; inoltre viene fatta diagnosi incidentale di duplicazione della colecisti. Sottoposto il caso clinico alla valutazione del Gruppo Oncologico Multidisciplinare, viene posta indicazione ad eseguire l'intervento chirurgico in regime di elezione. In sala operatoria la nota colecisti accessoria risultava fortemente adesa al colon trasverso prossimale e durante le complesse manovre di viscerolisi, è stata lesionata una delle colecisti. A questo punto abbiamo deciso di procedere eseguendo la colecistectomia di entrambe le colecisti.
La duplicazione della cistifellea richiede un'attenzione particolare all'anatomia biliare e arteriosa per evitare danni iatrogeni. Questa variante, se misconosciuta, può portare a complicanze importanti durante l'intervento chirurgico, soprattutto se questo viene condotto in regime d'urgenza (per esempio in caso di paziente affetto da colecistite acuta). Al giorno d'oggi risulta utile studiare adeguatamente l'albero biliare prima dell'intervento e la metodica migliore a tal fine è la colangioRM. A questo punto si può procedere con l'intervento che viene seguito preferenzialmente con metodica mininvasiva.
È fondamentale quindi, considerare sempre le possibili varianti anatomiche e le diverse forme di presentazione delle patologie della colecisti, poiché non sempre si interviene in un "quadro standard". Soltanto un'accurata valutazione preoperatoria può aiutare per evitare danni iatrogeni al momento dell'intervento chirurgico.

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