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Case report and review of the literature

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Urgent cholecystectomy in patient with left-sided gallbladder. Case report and review of the literature

INTRODUCTION: *Sinistroposition of the gallbladder, or true left-sided gallbladder (LSG) without situs viscerum inversus, is a rare congenital anatomical variant where the gallbladder is located to the left of round/falciform ligament. It can be associated with anomalies of the biliary tree, portal system and hepatic vascularization. The surgical management of a LSG could be challenging even for an experienced operator, being usually an incidental intraoperative finding.*
CASE REPORT: *A 72 years old woman was admitted to our emergency department because of acute cholecystitis. There were no pre-operative indications of sinistroposition of the gallbladder and its aberrant position was discovered during the explorative laparoscopy; because of the unusual anatomy and chronic flogosis, the laparoscopic approach was converted to open surgery. The patient underwent a successful intervention and was discharged after 4 days without complications. Her family history revealed a daughter with biliary atresia.*
DISCUSSION: *LSG could remain undetected at preoperative imaging, but today, with advances in diagnostic imaging, the report of this condition has increased. Several hypothesis suggest the presence of an underlying embriologic mechanism for LSG and its associated anomalies, but its etiology is still unknown. The association with the daughter's biliary atresia makes reasonable a possible genetic correlation with this condition.*
CONCLUSIONS: *In case of LSG, laparoscopic cholecystectomy could be feasible and safe, but with an increased risk of injury to the major biliary structures, mostly in case of severe and chronic inflammation of the gallbladder. Surgeons have to know this variant because of its associated hepatic anomalies.*

KEY WORDS: Cholecystectomy, Emergency Surgery, Left-Sided-Gallbladder

Introduction

Left-sided gallbladder (LSG) is a rare congenital anomaly with a reported prevalence of 0,6% [1] and is usually an incidental intraoperative finding.
There are three main types of LSG, one is LSG with situs viscerum inversus (S-LSG), the other two types are without situs viscerum inversus: true LSG (T-LSG) or LSG with gallbladder in orthotopic position but to the left of an abnormally located right-sided round ligament (R-LSG).

T-LSG, also known as sinistroposition gallbladder, was first described by Hochstetter in 1886 [2] and defined as an anatomical variant where the gallbladder is located on the third hepatic segment to the left of the round ligament.
This is a rare anatomical finding that could remain undetected at preoperative imaging, especially when performed in emergency settings, even though advances in diagnostic techniques have allowed the condition to be assessed more frequently as reported in literature.
T-LSG can be associated with anomalies of the biliary tree, portal system and hepatic vascularization that could be of prior importance in case of surgery [3-5]. For these reasons, the surgical management could be challenging even for an experienced operator, because of the increased risk of bile duct injury, mostly in case of severe and chronic inflammation of the gallbladder.

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In this paper we describe the case of a woman with undetected T-LSG who underwent emergent cholecystectomy due to acute cholecystitis.

Case Report

A 72 years old woman, with a 2 month-history of chronic abdominal discomfort that got worse in the last 3 days, presented to the Emergency Unit at the Careggi University Hospital, Florence, with acute abdominal pain in the right upper quadrant, without nausea, vomiting or fever. She had no relevant medical history except for high blood pressure and chronic obstructive pulmonary desease under good medical control, and a previous surgery for uterine fibromatosis at the age of 35 yo. The only relevant event in her family history was the death of an 8-month daughter because of biliary atresia complications when she was 30 yo. The body mass index (BMI) was 35,8 kg/m^2 (class 2 obesity). On admission the vital signs were stable (heart rate 81 bpm, respiratory rate 15 breaths per minute, blood pressure 130/80 mmHg) with normal cardiopulmonary function. The physical examination revealed abdominal tenderness with positive Murphy's and Blumberg's signs. Laboratory tests revealed neutrophilic leucocytosis (white blood cells 13200/mm^3; neutrophil granulocytes 75%), increased serum levels of C reactive protein (123 mg/L) and bilirubin (1,3 mg/dL) with a normal liver function index. US examination revealed a distended gallbladder with thickened walls, completely filled with intraluminal stones, one of them located in the infundibulum. No dilatation of the biliary tree was observed. These findings were consistent with acute lithiasic cholecystitis and the patient was referred to the emergency surgery department for surgery. The intervention, performed by a surgeon with a large experience in emergency and laparoscopic surgery, started with the laparoscopic technique. After insertion of the camera into the umbilical port, 3 accessory ports were placed (according to the French technique in whereby the surgeon stands between the patient's legs): two operating trocars were placed in the right upper quadrant and left subcostal region and the reactor trocar was placed in the epigastric region. Intraoperatively, extensive adhesions between the greater omentum, the duodenum and the liver were found which made adhesiolysis particularly hard to perform. The gallbladder was not detected in its usual anatomic position, but was located on the left side of the falciform ligament, embedded within the liver parenchyma of the third hepatic segment. Due to the chronic flogosis, the hardened sclerotic tissue of the gallbladder and its aberrant anatomy, it was not possible to obtain a critical view exposure of the Calot's triangle. Hence a fundus first approach was attempted, still without achieving a good recognition of the vascular and biliary structures. At that point it was decided to proceed with a laparotomic convertion by a right subcostal incision. The dissection of the Calot's triangle revealed that the cystic duct crossed over the common hepatic duct before the confluence on its right side. No other anatomic anomalies were found (Fig. 1). This approach allowed to safely detect and ligate the cystic artery and duct. According to the treating surgeon, intraoperative cholangiography was not performed. The surgery time was 140 minutes. The following hospital stay was uneventful with the progressive normalization of the laboratory tests. The patient was discharged on the fourth postoperative day. The pathological examination confirmed the diagnosis of lithiasic cholecystitis.

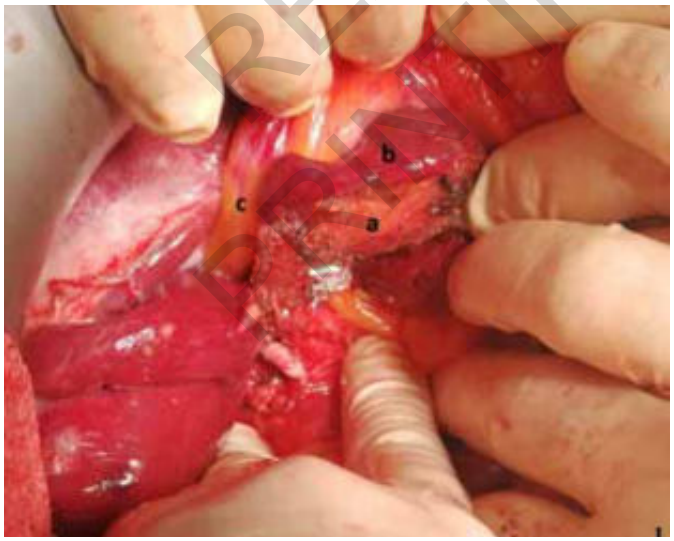

Fig. 1: Intra-operative photo showing the gallbladder bed (a) located on the third hepatic segment (b) to the left of the round ligament (c).

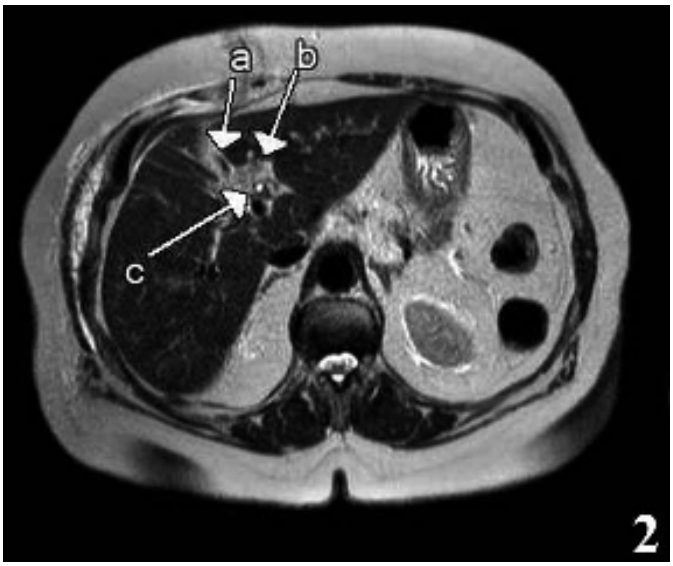

Fig. 2: Axial T2-wheighted MR showing the gallbladder bed (b) lying on the left of the falciform ligament (a) and the CD (c).

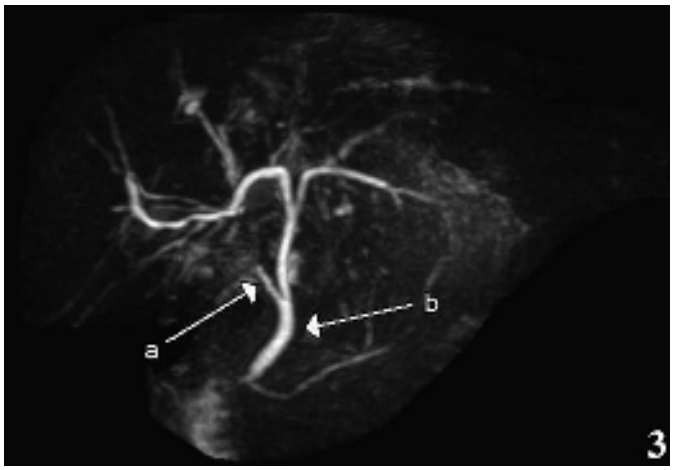

Fig. 3: Cholangio-MR showing the right implant of the CD (a) on the CHD (b).

The follow-up MRI, performed 2 weeks after discharge, confirmed the presence of the gallbladder bed on the left hepatic segment (Fig. 2) with right-implantation of the cystic duct (CD) on the common hepatic duct (CHD) (Fig. 3). No other associated anomaly of the portal vein or biliary tree was found.

Discussion

Anatomically, the gallbladder is located on the visceral surface of the liver on the van Rex-Cantlie line, a vertical plane dividing right and left hepatic lobes (according to Couinaud's functional segmentation) which is the principal plane used for hepatectomy. The gallbladder lies on the fourth hepatic segment, to the right of the falciform ligament.
LSG is defined as the presence of the gallbladder to the left of the falciform ligament.
There are three variants of LSG: S-LSG, T-LSG and R-LSG.
In particular, for a diagnosis of T-LSG, also known as Sinistroposition, not only the gallbladder must be located to the left of the falciform ligament but also under the surface of the left liver (third hepatic segment), where the main middle hepatic vein clearly runs to the right of the gallbladder [6].
T-LSG is a rare congenital anatomic variant with a reported prevalence of 0,6%1 with the gallbladder located to the left of the falciform ligament, on the third hepatic segment. There are less then 160 cases of T-LSG reported in literature [7-13].
As this condition is usually asymptomatic and not easily detectable by diagnostic imaging, it might be underreported. However, thanks to advances in diagnostic imaging techniques, its prevalence has increased over time 1. The presence of a T-LSG should be suspected whenever the fourth hepatic segment is not clearly detected in US or CT10. This finding could indicate a fourth segment atrophy, often associated to T-LSG, as well as biliary tree and portal vein anomalies [4,14-17].
Even thought the exact developmental pathway of a T-LSG remains unclear, two possible embryological origins are suggested.
In the first modality, the gallbladder develops from the hepatic diverticulum in its normal location but is attached to the left lobe: as a result, the gallbladder migrates to the left side of the round ligament during the development of the left lobe (T-LSG). Then, the CD crosses the CHD from left to right, entering in its right-side, as in the typical anatomy [4,5,18-20]. This is the case described in this paper.
In the second modality, the gallbladder develops directly from the left hepatic biliary duct and the main gallbladder either regresses or fails to develop. In this case, the CD enters into the common bile duct (CBD) on the left or drains directly into the left hepatic duct [4,14,15,18,20,21].
However, even if the gallbladder is located in an aberrant position, the clinical presentation of an acute cholecystitis does not differ between a T-LSG and a gallbladder in orthotopic position. In both cases, in fact, the pain is referred in right upper quadrant and Murphy's sign can be present, probably due to the fact that the central nervous system does not transpose with the gallbladder's visceral pain fibers [8].
The presence of a T-LSG is usually a surgical finding during cholecystectomy or hepatic surgery.
In the case we described, the patient presented to the emergency room with acute and intense pain in the right hypocondrium, that might indicate acute cholecystitis. The US imaging, performed in emergent setting, confirmed the diagnosis of cholecistytis but did't reveal the presence of a T-LSG. No additional diagnostic imaging, such a CT scan, was required and the abnormal position of the gallbladder was an incidental finding during surgery.
T-LSG is usually associated with anatomical anomalies of the CD and cystic artery. There are three anatomical variants of the cystic duct in a T-LSG setting: the CD crosses the CHD to then converge on the right side, or the CD enters the CHD on its left side, or finally the CD reaches the left hepatic duct. A recent review [6] documented the right-side origin of the CD as the most common presentation, being present in 77% of T-LSG cases, as in this report.
Another frequent cause of difficulty is a relatively high-grade embedment of the gallbladder in the hepatic parenchyma, often observed in emergency settings, especially in cases of severe flogosis. Our patient presented with a 2 month-history of chronic pain in the upper right quadrant that got worse in the last 3 days, confirmed by the presence of hardened sclerotic tissue and adhesions around the gallbladder.
The conditions mentioned above, in addition to the patient's physical conformation and high BMI, led to

increased operatory risk, in particular bile duct injury. Our intervention was therefore converted from laparoscopic to open during surgery.
Althought a laparoscopic approach is feasible with T-LSG, there shouldn't be any hesitations to convert to an open procedure whenever the anatomic situation is unclear. In this case, the laparotomic approach allowed to better understand the anatomy and to safely perform the dissection of the Calot's triangle. Besides, the fundus-first approach seems safer in recognising biliary and vascular structures, both laparoscopically and during open approach [22,23].
Some Authors6 recommend to perform an intraoperative cholangiography to better understand biliary anatomy and perform a safer dissection. However, in this case, this wasn't necessary because the laparotomic approach permitted to have a complete control of all the anatomical structures.
The management of acute cholecystitis with T-LSG is challenging and requires an expert surgeon with skills in emergency and laparoscopic surgery. In case of severe inflammation, a cholecistostomy could be positioned to allow the transfer of the patient to a tertiary hepatobiliary center for later cholecistectomy [3].
One of the most peculiar aspects of this case, that could be interesting to investigate further, is the possible association between the mother's T-LSG and her daughter's biliary atresia, the commonest surgical cause of neonatal cholestasis [24], but still a rare congenital condition with an incidence of 1/8000-18000 live births [25].
The etiology of biliary atresia is still unknown; however, current literature stresses the interplay of both genetics and environmental factors [26].
To the best of our knowledge, this is the first time in literature in which this association is reported. Further researches are necessary as this finding might open new scenarios on the possible relationship between two rare conditions, that could suggest the same genetical pathway.

Conclusions

T-LSG is commonly an incidental intraoperative finding. Its presence should be suspected in case of unclear visualization of the fourth hepatic segment during preoperative diagnostic imaging.
Every surgeon should be aware of this condition to avoid bile duct injury or vessel damages. Althought laparoscopic cholecystectomy is feasible and safe, especially when performed by an experienced surgeon, laparotomic convertion and/or intraoperative cholangiography should be considered whenever the anatomy can not be promptly recognized, mostly in the presence of severe inflammation. Further researches are needed to better understand the ethiology of T-LSG and to shade a light on its possible association with other genetically determined biliary pathologies.

Riassunto

La collocazione a sinistra della colecisti consiste nella localizzazione del viscere sul terzo segmento epatico, a sinistra rispetto al legamento rotondo/falciforme del fegato e in assenza di situs viscerum inversus. Questa è una condizione congenita molto rara (la prevalenza riportata in letteratura è compresa tra lo 0,04 e l'1,1%) e può essere associata ad anomalie dell'albero biliare, del sistema portale e della vascolarizzazione epatica. Il management chirurgico non è semplice, anche per operatori esperti, perché spesso la diagnosi avviene intraoperatoriamente.
In questo articolo si riporta il caso di una paziente di 72 anni con la colecisti situata a sinistra che è stata sottoposta ad intervento di colecistectomia in regime di urgenza per un quadro di colecistite acuta. L'unico fatto degno di nota nell'anamnesi familiare della paziente era rappresentato da una figlia deceduta a pochi mesi di vita per atresia biliare.
L'ecografia dell'addome eseguita in urgenza aveva rilevato distensione della colecisti, con pareti ispessite e presenza di calcolo a livello infundibolare, senza tuttavia evidenziarne anomalie di posizione né ulteriori varianti anatomiche associate.
L'intervento è stato iniziato con tecnica laparoscopica, ma l'estesa sindrome aderenziale, nonchè la posizione anomala della colecisti, non rendevano possibile una corretta esposizione del triangolo di Calot, per cui l'intervento è stato convertito con approccio laparotomico mediante incisione sottocostale destra. Questo ha reso possibile un corretto riconoscimento delle strutture dell'ilo epatico, evidenziando il posizionamento a sinistra della colecisti con confluenza del dotto cistico sul lato destro del dotto epatico principale e quindi permettendo una più sicura legatura dell'arteria cistica e del dotto cistico.
La paziente è stata dimessa dopo quattro giorni, senza complicanze.
La Risonanza Magnetica eseguita due settimane dopo la dimissione ha confermato gli esiti della colecistectomia con il letto della colecisti situato a sinistra rispetto al legamento rotondo/falciforme e non ha evidenziato ulteriori anomalie anatomiche epatiche associate.
Questa anomalia nella collocazione della colecisti spesso non viene evidenziata alla diagnostica preoperatoria, anche se il suo riscontro risulta in aumento a causa degli avanzamenti tecnici dell'imaging diagnostico.
Sono state formulate alcune ipotesi eziopatogenetiche che potrebbero essere alla base di questa condizione e delle altre anomalie anatomiche ad essa associate. Un aspetto peculiare del caso descritto consiste nel dato anamnestico della figlia deceduta a pochi mesi di vita per atresia biliare, un'altra anomalia molto rara del sistema biliare la cui origine è ancora oggetto di studio. Benchè queste due condizioni siano molto rare e quindi difficili da studiare, la loro possibile associazione potrebbe aprire nuovi scenari eziopatogenetici per entrambe.

In conclusione, in caso di colecisti collocata a sinistra, la colecistectomia laparoscopica potrebbe essere una procedura sicura e fattibile, anche se in presenza di un aumentato rischio di complicanze, a causa delle possibili anomalie epatiche associate. La conversione laparotomica va sempre considerata nei casi in cui l'anatomia non sia ben riconoscibile, specialmente in casi di flogosi intensa.

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