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Sinistroposition: A case report of true left-sided gallbladder in a Vietnamese patient

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ABSTRACT

INTRODUCTION: Left-sided gallbladder without *situs viscerum inversus* (LSG-woSVI) is defined as a gallbladder located under the left lobe of the liver; to the left of the round/falciform ligament, but with all other viscera maintaining normal anatomical relationships. This is a rare congenital anomaly with a reported prevalence that ranges from 0.04% to 1.1%. It is usually an incidental intraoperative finding, and can be associated with anatomic abnormalities of the biliary tree, portal system and vasculature. LSG and associated variations may present significant challenges even for experienced surgeon.
PRESENTATION OF CASE: LSG-woSVI was unexpectedly discovered in a 49-year-old, Vietnamese female during laparoscopic cholecystectomy. There were no pre-operative indications of sinistroposition. The cystic duct joined the common hepatic duct on the right side, and the cystic artery crossed anterior to the common bile duct in a right-to-left direction. Antegrade cholecystectomy was performed without intraoperative or postoperative complications.
DISCUSSION: LSG is a rare anatomical variation that often remains undetected with ultrasound and pre-operative tests. Several hypotheses suggest underlying embryologic mechanisms for LSG and associated anomalies in ductal, portal and vascular anatomy, but the exact cause remains a mystery. Safe laparoscopic cholecystectomy can be done; however, there is an increased risk of injury to the major biliary structures compared to orthotopic gallbladder.
CONCLUSION: Laparoscopic antegrade cholecystectomy is feasible for LSG. However, surgeons need to be cognizant of anatomy, so that rapid modifications of surgical technique can ensure positive patient outcomes.

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1. Introduction

Sinistroposition (SPG) or left-sided gallbladder (LSG) without *situs viscerum inversus* is a rare congenital anomaly with a reported incidence that ranges from 0.04% to 1.1% [1,2]. This anatomical variant was first described by Hoochstetter in 1886 [3]. Normally, the gallbladder resides in the gallbladder fossa (or bed) between hepatic segments IV and V. In contrast, LSG is defined as a gallbladder that is located to the left side of the round ligament or ligamentum teres (or the thickened, free edge of the falciform ligament of the liver) [4]. Usually, it is diagnosed intraoperatively and is accompanied by anatomic variations that can prove quite challenging during cholecystectomy. These variations include portal vein anomalies, biliary system anomalies, segment IV atrophy, and variations in hepato-biliary vascular anatomy [4–6]. Even in the face of these dynamics, clinicians can safely perform laparoscopic cholecystectomy for LSG [6], but the risk of bile duct injury is greater than in case of the orthotopic gallbladder [7].

Historically, Gross [8] established a classification system that described 4 types of anomalous positions of the gallbladder: (1) intrahepatic gallbladder, (2) LSG, (3) retrodisplacement of the gallbladder, (4) transverse position of the gallbladder. In modern times, LSG is further classified with three recognized variants (Table 1): LSG associated with *situs viscerum inversus; (2)* True LSG, (3) Right LSG [2,4].

Secondary to the types of LSG and associated variations in hepatic, ductal and vessel anatomy, surgeons and anatomists must be cognizant of "hidden" presentations in patients that require excision of the gallbladder, and in the education of physicians,

Abbreviations: LSG, left-sided gallbladder; LT, ligamentum teres; RUQ, right upper quadrant; SPG, sinistroposition of the gallbladder; US, ultrasound/ultrasonography.

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Table 1
Classification of Left-Sided Gallbladder (LSG).

CLASSIFICATION	ABBREVIATION		DESCRIPTION
Left-Sided Gallbladder associated with *situs viscerum inversus*	**LSG-SVI**		a congenital condition in which the major visceral organs are reversed or mirrored from their normal positions. The normal arrangement of internal organs is known as situs solitus while situs inversus is generally the mirror image of situs solitus
True Left-Sided Gallbladder	**T-LSG**	***LSG-woSVI****	a gallbladder is positioned to the left of the ligamentum teres and falciform ligament and is located under the surface of the left liver lobe segments III (or II); *situs viscerum inversus is not present* most common type of *LSG-woSVI*
Right Left-Sided Gallbladder	**R-LSG**		a gallbladder located to the left of abnormally located right-sided round ligament/ligamentum teres (normally located at segment IVb); *situs viscerum inversus is not present*

*Left-Sided Gallbladder without situs viscerum inversus (LSG-woSVI).

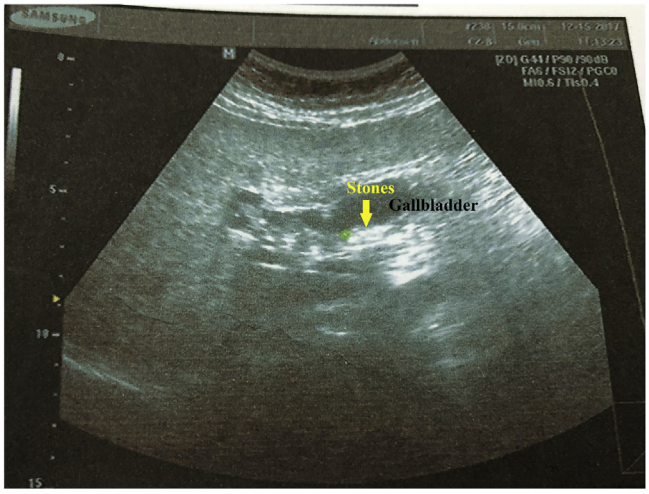

Fig. 1. Abdominal Echography. This abdominal ultrasound documents a normal liver. The gallbladder wall is not thickened. However, cholelithiasis (arrow) is present; with some stones measuring 15x7 mm. The common bile duct and intrahepatic ducts are of normal caliber and stone-free.

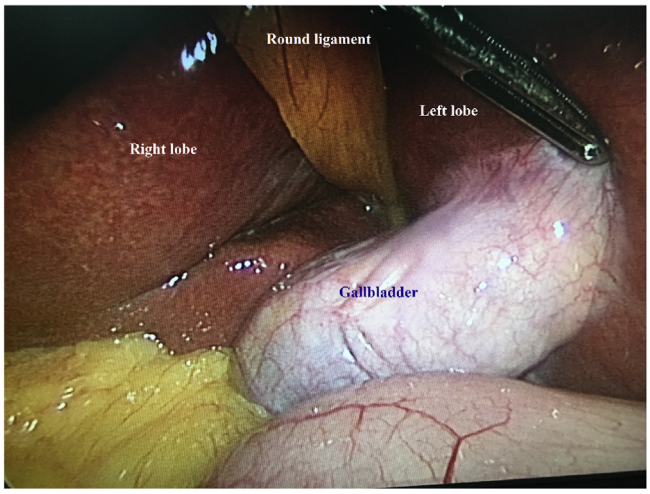

Fig. 2. Intraoperative Photograph. In this anterior view, sinistroposition, or true left-sided gallbladder, is observed. The gallbladder is located to the left of the round ligament under the left lobe of the liver. *Situs viscerum inversus* is not present.

respectively. The present report investigates the case of a 49-year-old female with LSG without *situs viscerum inversus* discovered incidentally during laparoscopic cholecystectomy, and discusses the present findings of this rare anomaly relative to data in the published literature. All work reported herein, follows the SCARE criteria for surgical case report guidelines [9].

2. Case report

Written/Informed consent was obtained from the patient for release of medical information and photographs, and for participation in this report. The study received ethical approval from the Ethics Committee of our institute.

A 49-year-old, Vietnamese female was admitted to the General Surgery Department with a history of intermittent pain in her right upper abdomen for 3 days. There was no prior medical history of jaundice, or biliary disease, and familial history was unremarkable. Physical examination revealed absence of tenderness in the right upper quadrant (RUQ) and epigastric region, and negative Murphy's sign. The patient's liver function tests were normal. Abdominal ultrasonography (US) documented cholelithiasis without dilatation of the bile ducts (Fig. 1). The patient underwent a laparoscopic cholecystectomy procedure. Upon visualization of the gallbladder bed, the gallbladder was observed to be absent from the normal location. Further inspection revealed a left-sided gallbladder, located to the left of the round ligament (Fig. 2) that was not detected using US. *Situs viscerum inversus* was not present. Intraoperative cholangiography was not preformed because of the lack of equipment; however, careful dissection revealed that the cystic duct joined the common hepatic duct on the right side. Antegrade cholecystectomy was performed, and there were no intraoperative or postoperative complications. The patient was discharged on the second postoperative day. Histological examination showed evidences of chronic cholecystitis.

3. Discussion

The gallbladder is usually located inferior to the liver on the von Rex-Cantlie line. Von Rex-Cantlie's line is a vertical plane that divides the liver into left and right lobes creating the principal plane used for hepatectomy. It extends from the inferior vena cava posteriorly to the middle of the gallbladder fossa anteriorly. It contains the middle hepatic vein that divides the liver into left and right lobes according to Couinaud's functional segmentation of liver. Segments II–IVa and IVb are on the left of the plane and segments V–VIII are on the right. The round ligament is normally present at the border between the left lateral and medial segments of the liver, with the faciform ligament in succession form the umbilical portion of the portal vein.

A true LSG is located to the left of the ligamentum teres (LT) and falciform ligament and under the surface of the left liver lobe segments III (or II) without *situs viscerum inversus* (LSG-woSVI; Table 1). It is accepted that for a diagnosis of a T-LSG, also referred to as sinistroposition, it must be located not only to the left of the LT and the falciform ligament but also under the surface of the left liver (3-degree hepatic segment), where the main middle hepatic

vein clearly runs to the right of the gallbladder, and the LT itself should originate from the left portal vein [4,10].

LSG is a rare congenital anomaly. Since the first published account of SPG in 1886 [3] less than 150 cases have been reported in the literature [4,11]. Hsu [5] reported a rate of LSG of 0.6% (9/1482) on CT scan. In cholecystectomy patients, LSG was found in 0.07 to 0.08% of cases [12,13]. In a retrospective study of published cases from 1996 to 2014, Abongwa, et al. [4], reported 55 cases of LSG-woSVI spanning 13 countries with an occurrence of 5:1, female-to-male ratio, respectively. The highest number of reported cases in the literature were from Australia (33%) and Japan (18%). Furthermore, more than half of the cases (28/55, or 51%) were reported in the last 5 years of the study period. The majority of LSG cases are discovered incidentally, intraoperatively [7,12]. In the present case, preoperative US revealed the presence of gallbladder stones but failed to demonstrate the abnormal position of the gallbladder and other associated biliary abnormalities. Hsu et al. [5], recommended that LSG should be suspected when abdominal ultrasound, CT scan, and angiography reveal no definite segment IV, absence of umbilical portion of the portal vein in the left lobe, and club-shaped right anterior portal vein.

There are three proposed, embryologic mechanisms for LSG-woSVI [8,14]. Normally, the endodermal lining of the foregut forms an outgrowth into the surrounding mesoderm called the hepatic diverticulum. The hepatic diverticulum gives rise to both the liver and gallbladder. The connection between the hepatic diverticulum and the foregut narrows to form the bile duct. Next, an outgrowth from the bile duct gives rise to the gallbladder and cystic duct, that intern divides the bile duct into the hepatic and common bile ducts. One hypothesis is that as the gallbladder develops from the hepatic diverticulum, it becomes attached to the developing left lobe of the liver and is carried across to the left side of the round ligament. Another hypothesis for development is that a second gallbladder develops directly from the left hepatic duct as an “accessory” gallbladder; and then the primary gallbladder either regresses or fails to develop [14,15]. The last suggested pattern of development of a LSG-woSVI may result from the failure of the quadrate lobe of the liver to develop as shown in operative findings [14].

The ductal anatomy of SPG is of critical importance to the surgeon. The course of the cystic duct in SPG has been classically described as joining the hepatic duct on the right side in a right-to-left manner; resulting in a hairpin turn anterior to the hepatic duct [6,13,16]. Other reported ductal variations include duplication of the common bile duct; hypoplastic common bile duct, infraportal bile duct, and abnormal pancreatico-biliary junction have all been described [7,14,17,18]. In addition, although Less common, the cystic duct has been documented to join the common hepatic duct or left hepatic duct from the left side [10,22]. In the patient studied in this case, the cystic duct joined the common hepatic duct on the right side. Given this array of anatomical variation in ductal anatomy, accurate identification is critical for uncomplicated cholecystectomy when LSG is encountered in the surgical theater. Nojiri et al. [19] have shown that can be easily accomplished during laparoscopic cholecystectomy using fluorescent cholangiography.

Variations in vascular anatomy have also been reportedly associated with LSG. These include anomalies of the porta vein, hepatic vein, and hepatic artery [6,16,20,21]. For example, the right hepatic vein has been observed to course between the right anterior and posterior portal veins, the middle hepatic vein to the left of the right anterior portal vein, and the left hepatic vein toward left lateral section [20,21]. The left lateral hepatic artery has been documented with LSG with its origin at the celiac artery, and the left medial segment artery branched from the right hepatic artery [20]. Small (narrow) and long cystic artery, lack of a cystic artery, or a cystic artery crossing in front of the common bile duct or anterior to the cystic duct [7,8,14,22]. Still further, variations in the portal system have been noted to include a left lateral portal vein lacking an umbilical portion; as well as a right umbilical portion forming after branching of the left lateral portal vein [20]. No vascular anomalies were observed in the patient discussed in the present case, other than the cystic artery coursing anterior to the common bile duct from right to left.

Because routine pre-operative testing may fail to detect LSG, surgeons may be surprised during laparoscopy. When discovered during surgery, LSG must be promptly recognized by the physician, who must be aware of associated variations in ductal and vessel anatomy. Chrungoo et al. [22] suggest that diathermy be limited and division of structures not be done until a definitive picture of the bile duct and blood vessels is obtained. To avoid biliary injuries, many authors recommended antegrade dissection and intraoperative cholangiography [2,4,19,23–25]. Surgical techniques such as laparoscopic cholecystectomy have proved to be safe for the management of LSG [6,13,17,24]. Some authors suggested performing cholecystectomy by the antegrade approach to facilitate visualization of the anatomic structures and avoid injury of hepatic pedicle. In the surgery where anatomic structures remain difficult to identify, conversion to open surgery is recommended [6]. In the case presented herein, antegrade cholecystectomy was performed, and followed by an uneventful post-operative course.

4. Conclusion

LSG-woSVI is a rare congenital anomaly, that when incidentally encountered during intraoperative procedures can present significant challenges due to associated variations in ductal and vessel anatomy. In fact, LSG is usually diagnosed intraoperatively. Laparoscopic antegrade cholecystectomy can be performed safely for LSG.

Conflicts of interest

None of the authors have any conflict of interest; no financial or personal relationships.

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There are no sponsors or sources of funding or generosity for the current research/case described within the text of this manuscript.

Ethical approval

The study received ethical approval from the ethics committee of Hue University of Medicine and Pharmacy, Hue.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. All private information was kept anonymous.

Author contributions

TRI HUU NGUYEN – study design and data collection, writing the paper.

PHU DOAN VAN NGUYEN – data collection.

TUNG SANH NGUYEN – data collection.

THANH NHU DANG – interpretation, writing the paper.

ERNEST F. TALARICO, JR. – writing the paper, Editor; discussion/collaboration regarding findings.

Registration of research studies

This manuscript/study is that of a case report of an embryologic defect that was discovered during the course of surgery, and for which written consent was given by the patient. The present work did/does not involve basic science, clinical, translational, drug or mechanism research (or testing) on human subjects. Therefore, a UIN is not required.

Guarantor

TRI HUU NGUYEN, M.D., Ph.D.
ERNEST F. TALARICO, JR., Ph.D.

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