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Laparoscopic Cholecystectomy in Situs Inversus Totalis: A Case Report

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ABSTRACT

Situs inversus totalis is a rare condition characterized by the mirror image of the normal abdominal and thoracic viscera. Since the gallbladder is located in the left upper quadrant in this subset of patients, the diagnosis of symptomatic cholelithiasis poses a difficulty. Similarly, laparoscopic cholecystectomy in these patients is more demanding and presents technical challenges, especially for right-handed surgeons. Herein, we report a case of a 30-year-old female who was a known case of situs inversus totalis and was diagnosed with symptomatic cholelithiasis and subsequently underwent laparoscopic cholecystectomy with an uneventful post-operative period. The challenge while performing laparoscopic cholecystectomy in patients with situs inversus totalis calls for appropriate perioperative preparation and necessary modifications in the operative setup for the safe and successful operation of the patient, which is discussed in this report.

1 | Introduction

Situs inversus totalis (SIT) is a rare congenital abnormality, where there is transposition of both the thoracic and the abdominal organs, thus forming a left–right asymmetrical picture of the normal anatomical placement of the organs [1]. It is kept under an umbrella term, heterotaxia, which encompasses all congenital disorders associated with malposition about the left–right axis [2]. An important distinction is from simply situs inversus, where only some of the major abdominal and thoracic organs are reversed from their normal anatomical positions [3]. SIT is an even rarer condition, with an incidence of 1:10,000, and males being affected more than females (1.5:1) [1]. Due to this anatomical asymmetry, it may pose difficulties during diagnostic and therapeutic procedures [1, 3].

SIT appears to be familial, and laterality is established early on in the developmental process, during gastrulation. A protein, namely "Sonic Hedgehog" (Shh) is deemed to be responsible for the expression of two growth factors, Nodal and Lefty. If these proteins are released on the right side, the rotation of the heart occurs to the left side. However, if they are released on the left side, rotation occurs to the right. A gene named PITX2 has been identified, which controls the secretion of Shh and Nodal [4]. The exact mechanism behind this is yet unknown; however, it has been speculated that the mutation in the PITX2 and Nodal gene is responsible and is also associated with other conditions like primary ciliary dyskinesia (PCD) and Kartagener syndrome [1, 3–5].

Due to the unique transposition of the anatomical structures in SIT, the usual causes of abdominal pain may be a misdiagnosis. Similarly, these patients with cholelithiasis may have a presentation with left upper quadrant pain instead of the usual right, likely posing a diagnostic dilemma [6, 7]. Operative management in SIT patients also presents technical and ergonomic

Summary

- In patients with situs inversus totalis, the main challenge lies in the diagnosis itself.
- The surgical treatment for cholelithiasis in these patients can be a standard procedure if it is well planned.

challenges, especially for right-handed surgeons [8]. The mirror image of the anatomy causes difficulty in the dissection of Calot's triangle, thus requiring appropriate tweaks to the normal preoperative and intraoperative setting [8]. This report describes a case of a SIT patient with symptomatic cholelithiasis, managed by laparoscopic cholecystectomy.

2 | Case History/Examination

A 30-year-old female presented with colicky abdominal pain located over the epigastric and left hypochondrium regions for the last 4 months. The pain was intermittent in nature, non-radiating, and mild in severity. She complained of an increase in pain after the consumption of fatty food. She gave a history of occasional nausea without vomiting. There was no history of fever, jaundice, alteration in bowel habits, or recent significant weight loss. She was diagnosed with SIT while undergoing a medical check that was required for a job application. She underwent a lower segment cesarean section 3 years ago. There was no other significant medical or surgical history. Her family history was also non-significant. On examination, her general condition was fair, afebrile, and there was no icterus. Examination of the abdomen revealed mild tenderness on palpation over the left side with no guarding. Normal heart sounds were heard over the right side of the thorax.

3 | Methods (Differential Diagnosis, Investigations and Treatment)

With the possible differential diagnosis of gastritis and biliary colic, the following investigations were done. Her laboratory investigations showed normal white blood cell counts of 9800 cells per cubic millimeters, and the liver function test was also found to be normal. Chest x-ray (Figure 1) revealed dextrocardia with the gastric bubble on the right side and the liver on the left. Ultrasonography of the abdomen showed multiple echogenic foci of 3 to 7 mm in size in the gallbladder and a common bile duct (CBD) of 6 mm. She was diagnosed with symptomatic cholelithiasis and planned for laparoscopic cholecystectomy. Routine preoperative workup was done and was found to be normal.

The operating theater was prepared such that the surgeon and camera assistant were on the right side, and the assistant surgeon and the laparoscopic tower were on the left side of the patient (Figure 2A). The surgeon in this case was right-handed. The patient was kept in a supine position with the left side slightly elevated. Four ports were created. The first port of 10 mm positioned supraumbilically was created, followed by the creation of pneumoperitoneum using carbon dioxide gas. After which, a 10 mm port in the epigastric region and two 5 mm ports were positioned in the left mid-clavicular line and left anterior axillary line, respectively (Figure 2B).

Diagnostic laparoscopy was performed, and the laterality was visualized (Figure 3). The fundus of the gallbladder was then retracted towards the right shoulder, and the retraction of Hartmann's pouch was done to the left by the assistant. The posterior dissection was then performed for the clearance of the cystic duct and artery. This maneuver prevented the crossing of hands of the surgeon. Thus, the Calot's triangle was identified, and dissection of the fibrofatty tissue was done to achieve the critical view of safety, such that only the cystic artery and duct were seen to be entering the gallbladder. Indocyanine Green (ICG) cholangiography was not done in this case to delineate the biliary anatomy. The gallbladder was then slightly separated from its attachment to the liver to expose a part of the cystic plate, following which the cystic artery and the duct were clipped and divided. The gallbladder was then dissected off its liver bed and removed through the umbilical port. On opening the retrieved gallbladder, two calculi were found inside, with its walls mildly inflamed. This was then sent for histopathological examination. The 10 mm ports were closed using polygalactin 1–0. The histopathological report concluded the diagnosis to be chronic calculous cholecystitis.

4 | Conclusion and Results

SIT is a rare condition, but for symptomatic cholelithiasis in this subset of patients, laparoscopic cholecystectomy is a rather

FIGURE 1 | Chest x-ray showing dextrocardia, right-sided gastric bubble, and shadow of the liver on the left.

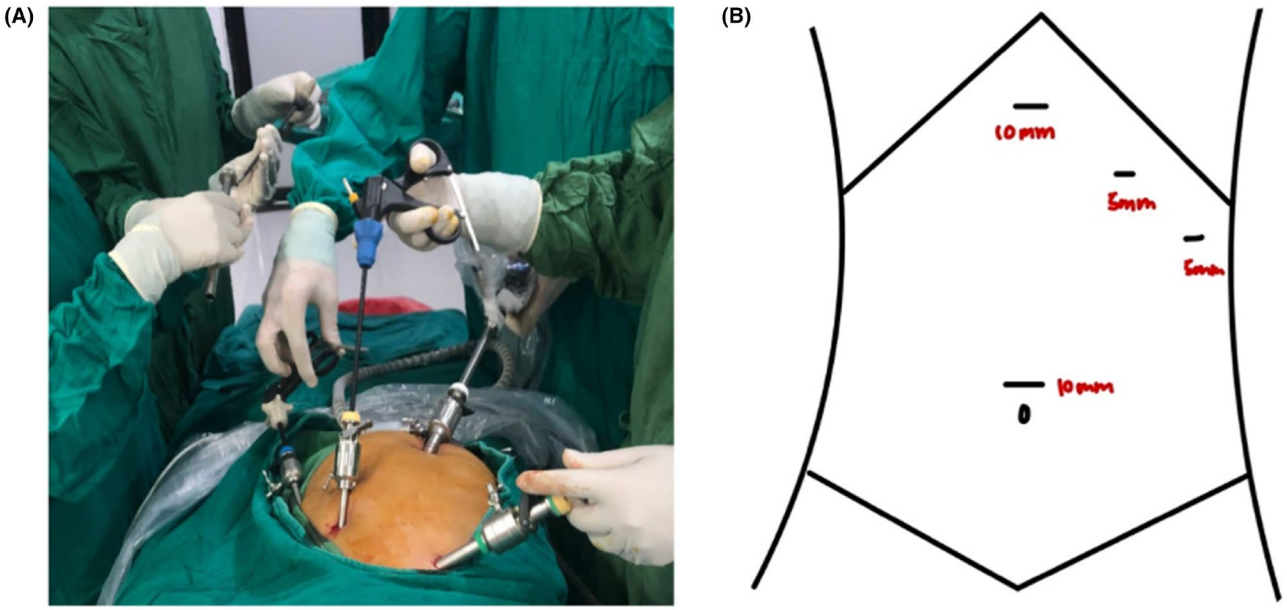

FIGURE 2 | (A) The operative setup demonstrating the primary surgeon present on the right side of the patient. (B) Incision sites for trocar placement.

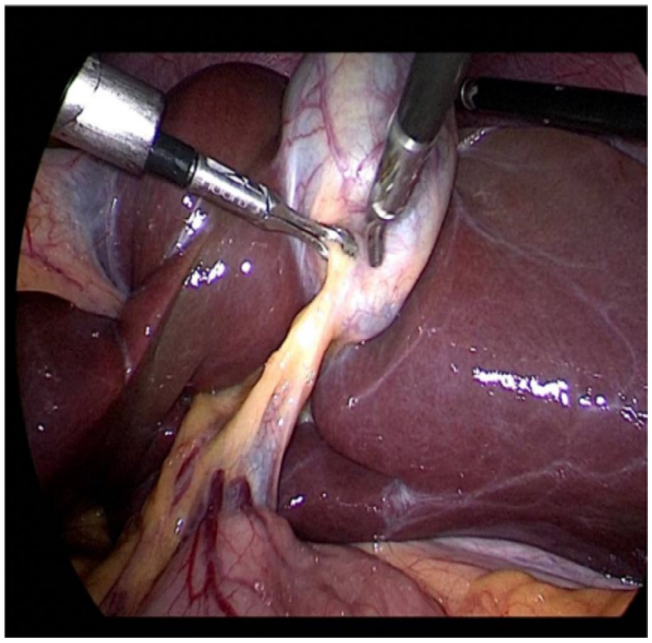

FIGURE 3 | Laparoscopic picture showing the gallbladder placed towards the left side of the falciform ligament.

standard procedure. However, special care must be taken while positioning the ports and placement of the surgeon and the assistants due to ergonomic and technical challenges surgeons face, especially right-handed ones. We presented a case where a right-handed surgeon was situated on the right side of the patient, with the assistant retracting the gallbladder while the primary surgeon dissected. This prevented the crossing of the hands of the surgeon. Her post-operative period was uneventful, and she was discharged on the second post-operative day. Follow-up on the seventh day showed a healthy surgical site. She was doing well at 1-year follow-up, with no recurrence of biliary symptoms or signs of infection.

5 | Discussion

SIT is a rare condition characterized by the complete transposition of both the thoracic and the abdominal viscera along the left–right axis, thus forming a mirror image of the normal anatomical picture [1]. SIT alone is asymptomatic, except for the accompaniment of cardiac anomalies associated with dextrocardia, such as atrial situs solitus, discordant atrioventricular (AV) connection, and to a lesser extent congenitally corrected transposition of great arteries (TGA) [6, 9]. Situs inversus is closely associated with a condition known as PCD. Fifty percent of the patients with PCD are associated with situs inversus, and together they are known as Kartagener's Syndrome [10, 11]. However, there isn't enough evidence that establishes the association between SIT and gallbladder disease [6, 12].

While the diagnosis of symptomatic cholelithiasis in patients with SIT is challenging, since our patient was diagnosed with it on routine health checkup prior to the development of illness, no particular challenge was faced in this case. The first successful laparoscopic cholecystectomy in a patient with situs inversus was performed in 1991 by Campos and Sipes [13]. In a similar report, Arya et al. performed the procedure by placing the ports and the instruments in a mirror-image fashion to the conventional laparoscopic cholecystectomy. The primary surgeon performed the dissection, while the first assistant retracted Hartmann's pouch throughout the surgery, thus preventing the crossing of the hands of the right-handed surgeon [14]. However, Alam and Santra reported conduction of the procedure with the first assistant holding the camera through the infraumbilical port and the second assistant retracting the Hartmann's pouch [8]. Further, the mirror image technique can be done via the American position, as described above with the primary surgeon positioned on the right side of the patient, or the French position (the Llyod-Davies Position), with the primary surgeon standing in between the abducted legs of the patient [14, 15]. These two positions are compared in Table 1. In our opinion, the

TABLE 1 | Comparison between the American and French positions for the mirror image technique of laparoscopic cholecystectomy in situs inversus totalis.

Parameter	Mirror image laparoscopic cholecystectomy in American position	Mirror image laparoscopic cholecystectomy in French position
Position of port	Ports are placed in a mirror image fashion to the standard laparoscopic cholecystectomy with 10 mm camera port placed via an infraumbilical incision, a 10 mm port in the epigastrium, below the xiphoid process and two 5 mm ports along the left subcostal line	Port placement is same as that in the American position
Position of primary surgeon	Primary surgeon stands on the right side of the patient, with dissection performed through the epigastric port	The primary surgeon stands in between the legs of the patient. The surgeon performs the dissection with their dominant hand and retracts the Hartmann's pouch with another
Position of assistant surgeon	First assistant stands on the right side of the patient and is retracting the Hartmann's pouch and the second assistant stands on the left side for the retraction of the fundus	The first assistant stands in the left side of the patient, holding the camera port. The second assistant stands on the right side of the patient, retracting the fundus
Advantages	Since most surgeons are trained in this position, they are more familiar with this technique	It is ergonomically favorable as both views of the Calot's triangle can be visualized and it is less likely that the instruments cross over
Disadvantages	It is ergonomically not favorable as only one view of the Calot's triangle is viewed comfortably and can also cause crossing of instruments	Many operating tables are not suitable for the French position and most surgeons are less likely to be trained in the French position

French position is ergonomically favorable as it provides both views of Calot's triangle. In this position, either the primary surgeon themselves or the assistant on the right side of the surgeon can retract the Hartmann's pouch. Cases of Single Incision Laparoscopic Surgery (SILS) have also been reported, with the aim of reducing invasiveness, improving cosmesis, and avoiding the crossing of the instruments [16, 17].

The use of Indocyanine Green (ICG) to delineate biliary anatomy is gaining popularity in laparoscopic surgery. Some surgeons use ICG fluorescence cholangiography to avoid bile duct injury. It is a safe, inexpensive, and useful modality to visualize the extrahepatic biliary tree, making the procedure easier in patients with SIT [18].

Author Contributions

Prapti Lakhey: data curation, writing – original draft. **Milan Shrestha:** data curation, methodology. **Rishav Sharma:** investigation, methodology. **Paleswan Joshi Lakhey:** conceptualization, supervision, writing – review and editing.

Ethics Statement

The authors have nothing to report.

Consent

Written consent has been taken from the patient for the publication of this case report.

Conflicts of Interest

The authors declare no conflicts of interest.

Data Availability Statement

Data available on request due to privacy/ethical restrictions:The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

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