Laparoscopic cholecystectomy in situs inversus totalis: Case report with review of techniques

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1. Introduction

Situs inversus (SI) is rare congenital disorder with an autosomal recessive mode of inheritance [1]. The estimated incidence varies from 1 in 5000 to 20,000 live births [1]. Situs inversus refers to spectrum of transposition of the body viscera, which can be complete (totalis) where both the thoracic and abdominal organs are reversed resulting in mirror image of the normal anatomical structures, or it may be partial (partialis) where either thoracic or abdominal organs are reversed [2]. Whereas the presence of an abnormally positioned organ known as situs ambiguous [3]. Dextrocardia refers to right sided heart which also can be found as different isolated entity [3]. Situs inversus (SI) is associated with various congenital anomalies including congenital heart diseases, renal dysplasia, and biliary atresia [2]. Kartagener’s syndrome represents a triad of situs inversus totalis (SIT), bronchiectasis, and sinusitis [2]. Diagnosis and management of symptomatic gallbladder stones in patient with SIT is challenging. Minimal invasive surgery is the preferred option however it carries within many technical difficulties due to the anatomical variation. We present a case of symptomatic gallbladder stone in a patient with situs inversus totalis requiring laparoscopic cholecystectomy (LC), discussing its feasibility and reviewing the surgical techniques in the literature. the work has been reported in line with the SCARE criteria [4].

2. Case report

A 40-year-old male not known to have any chronic medical illness, presented complaining of epigastric and left upper quadrant pain for 1 month, associated with intermittent nausea and vomiting, and aggravated by fatty meals, with no other associated symptoms. He had frequent visits to the emergency department where he was managed with analgesia and antacids with mild symptomatic improvement. Clinical examination was unremarkable with no evidence of jaundice or abdominal tenderness. His blood test results showed a normal complete blood count, kidney function, and liver function. Chest X Ray revealed dextrocardia with stomach fundic gas shadow on right side (Fig. 1). Abdominal ultrasonography revealed transpositioning of the solid organs with a
left sided liver and gallbladder with a solitary stone and mild wall thickening. We elected to perform a Magnetic Resonance Cholangiopancreatography to delineate the anatomy and to rule out any anomalies within the biliary tree. It confirmed the previously noted findings, showed no evident anomaly within the biliary tree, and confirmed the diagnosis of situs inversus totalis (Figs. 2 and 3). The patient was scheduled for an elective laparoscopic cholecystectomy.

The operating room equipment arrangement was adjusted as Mirror Image of Routine Laparoscopic cholecystectomy (Fig. 4). The Monitor was placed on left side of the patient. The surgeon with the camera assistant were on right side of the patient and the first assistant was on left side of the patient. The abdomen was scrubbed and draped in the standard aseptic technique. The first infraumbilical 11 mm trocar introduced pneumoperitoneum induced using the open technique. Three 5 mm trocars were placed, at the xiphisternum which was used for the surgeon’s left hand, at the left midclavicular line 2 cm below the costal margin which was used as working port for the surgeon’s right hand and at left anterior axillary line 5 cm from the costal margin which was used for retraction of the gallbladder fundus by the second assistant, respectively. Inspection of the abdominal cavity confirmed the presence of situs inversus totalis, with the liver and the gallbladder positioned in the left side (Fig. 5). The Calot’s triangle was identified. The peritoneum overlying the gallbladder infundibulum was then incised and the cystic duct and cystic artery identified and circumferentially dissected, till the critical view was obtained. The cystic duct and cystic
Table 1
Summary of reported cases of laparoscopic cholecystectomy in patients with situs inversus totalis.

| Serial no. of cases in each series | Author | Year of publication | Diagnosis | Gender | Age |
|------------------------------------|--------|---------------------|-----------|--------|-----|
| 1                                  | Campos and Sipes et al. | 1991 | Chronic Cholecystitis | Female | 39 |
| 2                                  | Takei et al. | 1992 | Cholecystitis | Female | 51 |
| 3                                  | Lipschutz et al. | 1992 | Cholangitis / CBD calculi | Female | 80 |
| 4                                  | Goh et al. | 1993 | Empyema | Male | 62 |
| 5                                  | Drover et al. | 1992 | Chronic Cholecystitis | Female | 29 |
| 6                                  | Huang et al. | 1992 | Chronic Cholecystitis | Male | 36 |
| 7                                  | Schiffino et al. | 1993 | Chronic Cholecystitis | Female | 53 |
| 8                                  | Mc Dermott and Causah et al. | 1994 | Cholangitis / CBD calculi | Male | 66 |
| 9                                  | Malataniet al. | 1996 | Acute Cholecystitis | Female | 25 |
| 10                                 | Crosher et al. | 1996 | Cholecystitis | Male | 63 |
| 11                                 | D’Agata and Boncompagni et al. | 1997 | Chronic Cholecystitis | Female | 72 |
| 12                                 | Habib et al. | 1998 | cholecystectomy | Female | 45 |
| 13/14                              | Demetriades et al. | 1999 | Acute Cholecystitis / Chronic Cholecystitis | Female | 61/37 |
| 15                                 | Djohan et al. | 2000 | Chronic Cholecystitis / appendectomy | Female | 20 |
| 16                                 | Wong et al. | 2001 | Chronic Cholecystitis / CBD calculi | Female | 68 |
| 17                                 | Dorthi et al. | 2001 | Chronic Cholecystitis | Female | 43 |
| 18                                 | Nursal et al. | 2001 | Chronic Cholecystitis | Female | 42 |
| 19/20                              | Yaghan et al. | 2001 | Chronic Cholecystitis / Chronic Cholecystitis | Female | 48/38 |
| 21                                 | AI Jumaily and Hoche et al. | 2001 | Chronic Cholecystitis | Female | 46 |
| 22                                 | Singh and Dhi et al. | 2002 | Chronic Cholecystitis | Female | 28 |
| 23                                 | Trongue et al. | 2002 | Chronic Cholecystitis | Male | 68 |
| 24                                 | Polychronidis et al. | 2002 | Chronic Cholecystitis | Male | 35 |
| 25/26                              | Oms and Badia et al. | 2003 | Acute Cholecystitis | Female | 20/65 |
| 27                                 | Jesudason et al. | 2004 | Chronic Cholecystitis | Female | 32 |
| 28                                 | Kang and Han et al. | 2004 | Chronic Cholecystitis / CBD calculi | Female | 43 |
| 29                                 | Dociro et al. | 2004 | Chronic Cholecystitis | Female | 41 |
| 30                                 | Pitiakoudis et al. | 2005 | Chronic Cholecystitis | Female | 47 |
| 31                                 | McKayand Blake et al. | 2005 | Acute Cholecystitis | Female | 32 |
| 32                                 | Kamitani et al. | 2005 | Chronic Cholecystitis | Male | 76 |
| 33                                 | Puglisi et al. | 2006 | Chronic Cholecystitis | Female | 43 |
| 34                                 | Bedioui et al. | 2006 | Chronic Cholecystitis | Female | 58 |
| 35                                 | Aydin et al. | 2006 | Chronic Cholecystitis | Male | 35 |
| 36                                 | Machado and Chopra et al. | 2006 | Chronic Cholecystitis | Female | 65 |
| 37                                 | Kumar and Fasui wt al. | 2007 | Chronic Cholecystitis | Female | 57 |
| 38                                 | Fernandes et al. | 2008 | Chronic Cholecystitis | Female | 43 |
| 39                                 | Hamdi and Abu hamdan wt al. | 2008 | Acute Cholecystitis | Male | 41 |
| 40                                 | Pavlidis et al. | 2008 | Acute Cholecystitis | Female | 34 |
| 41                                 | Taskin et al. | 2009 | Chronic Cholecystitis / Gastric banding | Female | 20 |
| 42                                 | Masood et al. | 2009 | Chronic Cholecystitis | Female | 42 |
| 43                                 | Pereira-Gradouro et al. | 2009 | Chronic Cholecystitis | Female | 70 |
| 44                                 | Romano et al. | 2009 | Cholecystitis | Female | 67 |
| 45                                 | Eisenberg D et al | 2009 | Cholecystitis | Male | 61 |
| 46                                 | Patali et al. | 2010 | Chronic Cholecystitis | Male | 68 |
| 47                                 | Hall et al. | 2010 | Chronic Cholecystitis | Male | 53 |
| 48                                 | Gonzalez Valverde et al. | 2010 | Chronic Cholecystitis | Female | 46 |
| 49                                 | Sanduc and Toma et al. | 2010 | Chronic Cholecystitis | Female | 64 |
| 50/51/52/53/54/55                   | Palle NM et al. | 2010 | 5 Cholecystitis / 1 Acute Cholecystitis | Female / Male | 45/36/43/27/48/59/33 |
| 56                                 | Han et al. | 2011 | Chronic Cholecystitis | Male | 45 |
| 57                                 | Ozsoy et al. | 2011 | Chronic Cholecystitis | Female | 65 |
| 58                                 | Uldag et al. | 2011 | Cholecystitis | Male | 49 |
| 59                                 | Borgaonkar et al. | 2011 | Cholecystitis/ Appendicitis | Female | 47 |
| 60                                 | Seo KW et al. | 2011 | Cholecystitis / Gastric cancer | Male | 60 |
| 61                                 | Evoli LP et al. | 2011 | Cholecystitis | Female | 48 |
| 62                                 | Isuco et al. | 2012 | Cholecystitis | Female | 52 |
| 63                                 | Elbeshy et al. | 2012 | Cholecystitis | Female | 24 |
| 64                                 | Lochman et al. | 2012 | Acute Cholecystitis | Female | 75 |
| 65/66                              | Demirgulmaz et al. | 2012 | Cholecystitis / Cholecholecholeithiasis | Female / Male | 55/51 |
| 67                                 | de Campos Martins, Marcus Vinicius Dantas et al. | 2012 | Cholelithiasis | Female | 59 |
| 68                                 | Pahwa, Harvinder Singh et al. | 2012 | Cholelithiasis | Female | 46 |
| 69                                 | Bozkurt et al. | 2012 | Cholelithiasis | Male | 49 |
| 70                                 | Salama et al. | 2013 | Cholelithiasis | Male | 10 |
| 71                                 | Aya et al. | 2013 | Cholelithiasis | Female | 35 |
| 72                                 | Ali MS et al. | 2013 | Cholelithiasis | Female | 43 |
| 73                                 | Khangte et al. | 2013 | Cholelithiasis | Male | 65 |
| 74                                 | Raghveer et al. | 2014 | Cholelithiasis | Male | 55 |
| 75                                 | Reddy et al. | 2014 | Acute Cholecystitis / Cholecholecholeithiasis | Female | 45 |
| 76                                 | Fang et al. | 2015 | Gallbladder polyp / Rectal cancer | Female | 39 |
| 77                                 | Deguchi et al. | 2015 | Acute Cholecystitis | Male | 66 |
| 78                                 | Rosen H et al. | 2015 | Acute Cholecystitis | Male | 36 |
| 79                                 | Phothong et al. | 2015 | Cholelithiasis | Female | 39 |
| 80                                 | Alsabek et al. | 2016 | Cholelithiasis | Female | 50 |
| 81/82/83                           | Zeeshan et al. | 2016 | Acute Cholecystitis / Cholelithiasis / Cholecholeithiasis | Female | 46/44/33 |
| 84                                 | Jain-jun et al. | 2017 | Chronic Cholecystitis | Female | 36 |
artery were then doubly clipped and divided, through the subcostal port using the right hand. The gallbladder was dissected from its peritoneal attachments using electrocautery and was retrieved using Endoscopic bag through the infraumbilical port. The total operative duration was 80 min, which was longer than the conventional laparoscopic cholecystectomy performed in patient without underlying anatomical variation. It can be attributed to the modification in the technique required to adjust to the mirror image anatomy.

The patient had an uneventful postoperative course and was discharged on postoperative day 1. Pathological examination of the gallbladder confirmed the presence of gallstones with chronic cholecystitis. No postoperative complications were noted during his follow up in the outpatient department.

3. Discussion

Situs inversus totalis (SIT) is a rare autosomal recessive congenital anomaly, with a global prevalence of 0.01% [1,5]. It is characterized by the transposition of both thoracic and abdominal viscera resulting in perfect mirror image of their normal anatomical position [5]. It can be associate with various congenital anomalies, such as Kartagener’s syndrome which comprises a triad of SIT, sinusitis and bronchiectasis, and Yoshikawa’s syndrome that is characterized by the presence of SIT, bilateral renal dysplasia, pancreatic fibrosis and meconium ileus [6].

Diagnosis of biliary colic in patient with SIT is challenging due to the underlying anatomical anomaly. They often have an unusual presentation in form of left upper quadrant or epigastric pain, leading to a delay in the diagnosis and management especially in those who are not known to have SIT, as in the reported case. However, there is no evidence suggest that patients with SIT are more susceptible to cholelithiasis [7].

Open Cholecystectomy was the mainstay of management of cholelithiasis in the prelaparoscopic era. Around 40 cases of open cholecystectomy in patients with SIT were reported in the literature [8]. With advancement in surgical techniques, minimal invasive surgery has been introduced. The first case of Laparoscopic cholecystectomy was successfully performed by mooret in 1987, and since then it has become the gold standard approach [9]. In 1991 Campos and Sipes performed the first successful laparoscopic cholecystectomy in patient with SIT [10]. Since then 91 cases have been reported in the literature (Table 1). None of these cases reported any complication or have been converted to open cholecystectomy. Thus, it is considered a safe procedure, and not contraindicated in SIT [11,9]. However, it is carries within technical challenges due to the underlying mirror image anatomy which demands meticulous dissection of the biliary tree to avoid iatrogenic injuries [12]. Various techniques have been advocated to overcome these difficulties.

In the current literature, the most frequently adopted technique is the four port technique with placement of the laparoscopic equipment, positioning of the surgical team, and ports sites are a mirror image of the standards used in the usual cases [13,14]. The surgeon stands on the right side of the patient along with the camera assistant, and the first assistant stands on the left side. Left-handed instruments are used to grasp Hartmann’s pouch through the subxiphoid port, and the right hand is used for dissection through the left mideclavicular subcostal port [14,15]. Modification of this technique have been reported in the literature, where the assistant retracts the gallbladder fundus while the surgeon perform the dissection through the epigastric port with the right hand [8,13]. Some authors adopted a complete mirror image approach by using the left hand for dissection through the subxiphoid port, which could be more suitable option for a left handed or ambidextrous surgeon [16]. Another alternative for the surgeon to be positioned between the patient’s leg while the patient is in Lloyd-Davis position [17]. Recently a laparoendoscopic single-site surgery technique have been reported, which had the advantages of easier dissection with the right hand and better cosmetic result [18–21].

No technique has been considered yet as a standard for such cases. Surgeons should choose any suitable approach taking in account meticulous dissection and critical view achievement before clipping the cystic duct and artery. Intraoperative cholangiogram can be performed in such cases to visualize the anatomy and avoid iatrogenic injury [22]. Rungsaulkij et al. used fluores- cent cholangiography by administration of indocyanine green to delineate the extrahepatic biliary tree anatomy [14].

4. Conclusion

SIT is a rare congenital anatomy with mirror image transposition of the viscera. This anatomical variation can influence the localiza-
This manuscript reports a case of symptoms in a patient with cholelithiasis leading to a delay in diagnosis and management. Laparoscopic cholecystectomy can be safely performed in these cases. However, it is considered technically challenging and often requires alteration in the technique compared to the conventional laparoscopic cholecystectomy.

Conflicts of interest
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Ethical approval
This case report is exempt from ethical approval by our institution.

Consent
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Author contribution
OA assisted in the surgery, drafted the manuscript and reviewed the literature. AM wrote the manuscript and reviewed the literature. EZ performed the surgery and reviewed the manuscript. MT supervised the management of the patient and reviewed the manuscript. All authors read and approved the final manuscript.

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