International Journal of Surgery Case Reports 122 (2024) 110073

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.elsevier.com/locate/ijscr

Case report

Biliary stone in patient with situs inversus totalis

Morteza Azadbakht [a], Saleh Azadbakht [b,*], Samira Daniali [c]

[a] Department of Surgery, Faculty of Medicine, Lorestan University of Medical Sciences, Khorramabad, Iran
[b] Department of Internal Medicine, School of Medicine, Lorestan University of Medical Sciences, Khorramabad, Iran
[c] Drug-store, Lorestan University of Medical Sciences, Khorramabad, Iran

ARTICLE INFO

Keywords:
Situs inversus totalis
Thoracic
Abdominal viscera
Choledocholithiasis

ABSTRACT

Introduction and significance: Situs inversus totalis is characterized by the complete transposition of thoracic and abdominal viscera. Individuals can live asymptomatically with this condition; however, it may be associated with certain abnormalities of the organs involved.
Case presentation: Herein, we present a case of a situs inversus totalis woman presented with choledocholithiasis. Elective laparoscopic cholecystectomy was performed on the patient with intraoperative modifications. The patient was discharged in a healthy condition.
Clinical discussion: Several case reports have documented the typical presentation of cholelithiasis, which is characterized by pain in the left upper quadrant and epigastric region. Our patient exhibited similar symptoms and was diagnosed with choledocholithiasis.
Conclusions: Diagnostic and therapeutic management of morbidities in SIT individuals can be performed with recommended modifications that can lead to favorable outcomes.

1. Introduction

Situs inversus is a condition characterized by mirror inversion of organs of chest and abdomen relative to the normal position [1]. It is reported in 1 in 10,000 cases. Situs inversus totalis (SIT) is a more common type of situs inversus that is seen as a mirror inversion of heart and viscera (asymmetric body parts) [2,3]. It is the result of 270° clockwise rotation of the organs during embryonic development instead of anticlockwise rotation [4]. The condition is associated with several anomalies such as Kartagener's syndrome, cardiac, renal, skeletal and neurological disorders, and spleen malformations [5]. Underlying anatomical differences can impose challenges in the diagnosis of these comorbidities [6]. Additionally, surgical procedures on these viscera can be difficult and might require modifications [7].

2. Case presentation

The patient was a 59-year-old woman who complained of abdominal pain in the LUQ area and was presented to our center. The patient's pain started ten days before admission and had intensified in the last two days. The pain was accompanied by nausea and vomiting bile. The pain was crampy and spread to the epigastrium and back and left side. The patient has had intermittent and mild epigastric pain for the past two years, which would be exacerbated by the consumption of fatty and bulky foods. During that time, she had been treated with a proton pump inhibitor.

Due to the aggravation and lack of improvement, the patient went to emergency. She was hospitalized and initial measures such as fluid therapy and painkillers were started. She underwent ultrasonography where dextrocardia and situs inversus were reported, with the liver on the left and the spleen on the right. There was evidence of the onset of gallbladder inflammation and cholecystitis. The gallbladder was full of biliary sludge, the wall thickness of the gallbladder was 3 mm and spleen, kidneys and pancreas were normal.

The patient had hypertension and was taking amlodipine. She did not have any history of surgery whereas, her family history was unknown to us. Her vital signs such as blood pressure, pulse rate and body temperature were normal. The patient's growth status was normal, and the patient's history was reliable. The patient's skin was a natural color, not eclectic.

On examination of the head and neck, conjunctiva was absent, the patient's visual, olfactory, and gustatory status was normal. She had no anomalies and physical changes and surgical scars, chest and was symmetrical with no scoliosis and deformity.

* Corresponding author at: Gastroenterology and Hepatology, Lorestan University of Medical Sciences, Khorramabad, Iran.
E-mail address: md.azadbakht.s@gmail.com (S. Azadbakht).

https://doi.org/10.1016/j.ijscr.2024.110073
Received 22 June 2024; Received in revised form 16 July 2024; Accepted 22 July 2024
Available online 24 July 2024

The heart sound was louder on the right side. There was no murmur or abnormal sound. The lungs had a normal sound with no sound reduction or rumble. Bowel sounds were pronounced in the LLQ, the abdomen had a thunderous touch in the LUQ with no tenderness, rebounding, and no localized or generalized guarding. The pelvic and genital examination had no anomaly.

The appearance of the limbs was not normal or abnormal. The pulses were detected to be full and symmetrical.

Lab results:

WBC = 6.8, HB = 13.2, PLT = 254, AST = 118, ALT = 173, ALP = 727, BIL-T = 0.8, BIL-D = 0.3.

According to the results of tests and ultrasound for the patient, MRCP (Magnetic resonance cholangiopancreatography) was requested (Fig. 1).

The result showed a dilated gallbladder containing sludge with a normal wall. Dilated intrahepatic bile ducts and common biliary duct (CBD) were reported. CBD diameter was 9 mm. A stone with a diameter of 4.5 mm was seen in the distal CBD at the site of the ampulla. Some sludge and biliary sand were seen as well. The pancreatic ducts were normal in diameter. According to the ultrasound report and test results, gastrointestinal consultation was requested for the patient.

The work has been reported in line with the SCARE criteria [8].

2.1. Gastroenterology

Due to abnormal LFT and ultrasound and presence of calculi seen in MRCP, based on calculi, ERCP (endoscopic retrograde cholangiopancreatography) was performed. (Fig. 2): After conscious sedation, endoscope was passed through the mouth, stomach and second part of the duodenum and was placed in front of the papilla. The following was identified: 1–10 mm dilatation in CBD, filling defect in the distal part of CBD, normal right and left hepatic duct and intrahepatic duct.

Guidewire was then placed in CBD and balloon sweeping was performed several times, and a small stone and some sludges were removed. At least four fluoroscopic shots were obtained, biliary drainage was performed, and a video scope was removed. The patient was monitored for several hours and no complications of the procedure were seen. The patient went to the hospital a few days later for a cholecystectomy. At the last visit, the patient's symptoms were milder than the previous visit. Vital signs were normal. The abdomen was soft and biochemical analysis showed that liver enzymes were normalized, and amylase was 13. ECG was performed before the surgery.

2.2. Description of the surgery

First, the laparoscopic equipment was placed on the left side of the operating table due to the location of the patient's liver and gallbladder on the left side, and after general anesthesia, the gas was inserted into the abdominal cavity with a needle and the abdominal pressure was set at 13 mmHg. A cut was made at 10 mm above the umbilicus and the optics port was inserted, the camera was inserted at 10 mm and 30°. Directly, one port number 10 was inserted in the subglyphoid region, and two ports number 5 were inserted in the LUQ and left side of the patient at the umbilical cord (Video 2).

In the initial examination, the gallbladder was thickened, the anatomical elements of the Calot triangle were not well defined, and it was not possible to identify the lymph node and cystic duct in the early stages. Infundibulum was attached to the common duct (shown in Video 3), slowly performed with laparoscopic suction. It was looped up and attached to the infundibulum, and the cystic duct came down parallel to the common hepatic duct. First, the common duct was carefully separated from the infundibulum, and after detachment of the common duct, cystic duct appeared, which was slowly dissected by taking the cystic duct. After identifying the cystic duct and cystic artery and determining the critical view, the cystic duct was ligated and cut with a purple clip (which at this stage also marked the nipple sign). The cystic artery was cut and ligated, the gallbladder was removed from the liver bed and the necessary lavage was performed. Homeostasis was ensured, the ports were removed under direct vision, the gas was removed from the abdominal cavity, the fascia was sewn at the entrance of port 10 with vicryl 1 thread and the skin was sewn with 3-0 nylon sutures. The skin was sewn with nylon 3 zero thread. The patient was transferred to the ward in good general condition.

The patient was monitored for two days after the operation. During this period, there were no abnormal problems, presented in terms of

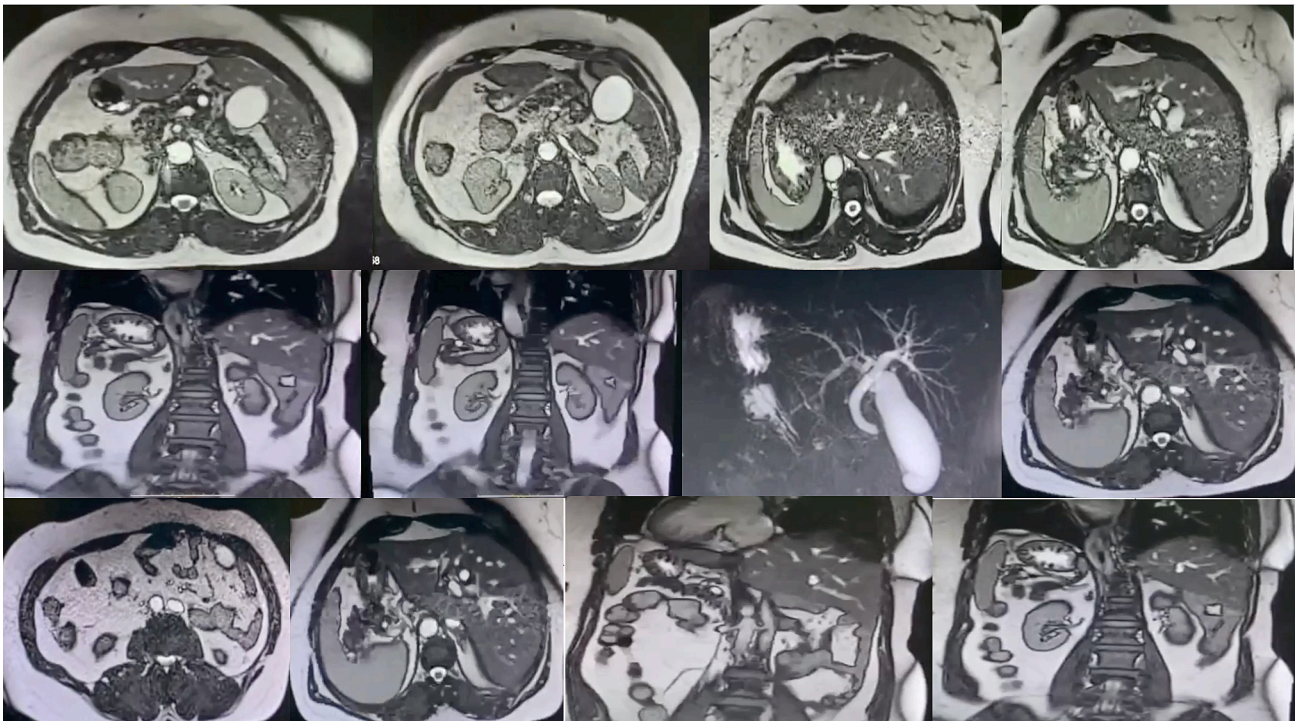

Fig. 1. Magnetic resonance cholangiopancreatography.

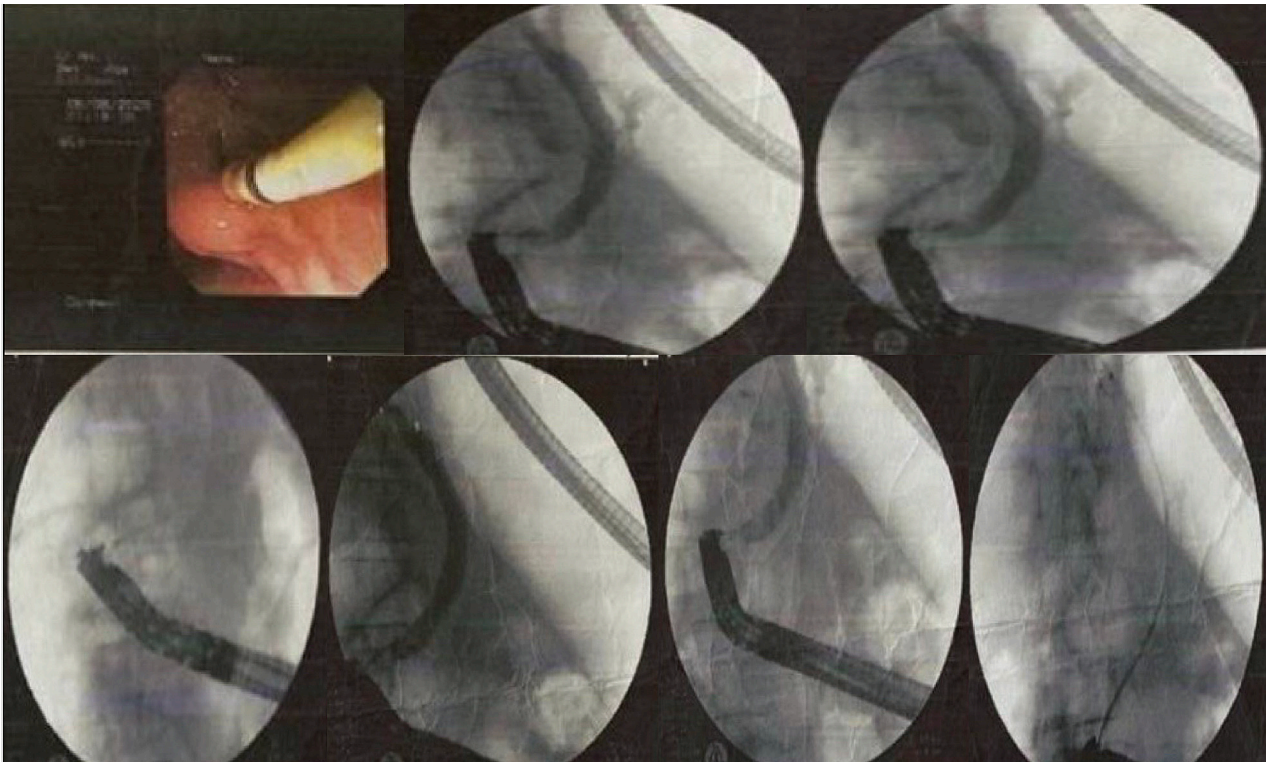

Fig. 2. Endoscopic retrograde cholangiopancreatography.

clinical symptoms and blood report tests. Finally, she was in good general condition and was discharged. At the outpatient visit after discharge, the patient had no evidence or symptoms in favor of possible complications. The patient is still under follow-up. Her LFT from last follow-up was normal.

3. Discussion

SIT is an autosomal recessive congenital disorder that is presented as anatomical mirroring of abdominal and thoracic viscera [9]. Several case reports have shown usual presentation of cholelithiasis characterized by left upper quadrant and epigastric pain [10]. Our patients had similar presentations and was diagnosed with choledocholithiasis.

Case reports have shown the use of laparoscopic cholecystectomy for this procedure with no major complications. The modifications in the technique have been proposed in case reports which included changing the positions of surgical equipment, surgeons and assisting staff, ports for the dissection of Calot's triangle, and the direction of the clip applicator aligning with cystic duct and artery [11]. A similar approach was adopted in our case report and the patient was discharged in healthy condition without any major complications [12]. A study has also suggested surgeons standing between the legs of the patient (Lloyd-Davis position) [7]. For right-handed surgeons, it is also recommended to switch right and left hands for dissection, crossing laparoscopic devices for dissection in Calot's triangle and the right hand in the left subcostal [13]. Furthermore, they can also dissect using the right hand from the epigastric port and the first assistant can hold the fundus and retract Hartmann's pouch [14–17].

4. Conclusion

With modification in laparoscopic cholecystectomy, the procedure can be performed without complications in SIT patients. Diagnosis of comorbidities can be challenging in SIT patients and special operative techniques and surgical skills are required to provide therapeutic efficacy.

Supplementary data to this article can be found online at https://doi.org/10.1016/j.ijscr.2024.110073.

Consent to participate

From the under 16 years old was given by a parent or legal guardian.

Consent for publication

Not applicable.

Patient consent

Informed consent was obtained from the patient for publication and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

All procedures performed in this study involving human participants were in accordance with the ethical standards of the institutional Lorestan University of Medical Sciences (IR.LUMS.REC.1397.001) committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Funding

No funding was secured for this study.

Author contribution

Dr. Morteza Azadbakht: conceptualized and designed the study, drafted the initial manuscript, and reviewed and revised the manuscript.

Dr. Saleh Azadbakht: Designed the data collection instruments,

collected data, carried out the initial analyses, and reviewed and revised the manuscript.

Dr. Samira Daniali: Coordinated and supervised data collection, and critically reviewed the manuscript for important intellectual content.

Guarantor

Dr. Morteza Azadbakht.

Research registration number

N/A.

Conflict of interest statement

The authors deny any conflict of interest in any terms or by any means during the study.

Availability of data and material

Data sharing is not applicable to this article as no datasets were generated or analyzed during the current study.

References

[1] K. Eitler, A. Bibok, G. Telkes, Situs inversus totalis: a clinical review, Int. J. Gen. Med. (Mar 3, 2022) 2437–2449.
[2] P.J. Shogan, L. Folio, Situs inversus totalis, Mil. Med. 176 (2011) 840–843.
[3] S.J.S. Bajwa, A. Kulshrestha, J. Kaur, S. Gupta, A. Singh, S.S. Parmar, The challenging aspects and successful anaesthetic management in a case of situs inversus totalis, Indian J. Anaesth. 56 (2012) 295–297.
[4] W. Chen, Z. Guo, L. Qian, L. Wang, Comorbidities in situs inversus totalis: a hospital-based study, Birth Defects Res. 112 (2020) 418–426.
[5] N. Abu-Oddos, M. Abu-Jeyyab, M. Al Mse'adeen, A. Rawshdeh, M. Al-Jafari, S. I. Abu-Oddos, M. Shahin, B. Rawashdeh, Laparoscopic cholecystectomy in a patient with situs inversus totalis and a double superior vena cava, Am. J. Case Rep. 24 (2023) e938774-1.
[6] O. AlKhlaiwy, A.M. AlMuhsin, E. Zakarneh, M.Y. Taha, Laparoscopic cholecystectomy in situs inversus totalis: case report with review of techniques, Int. J. Surg. Case Rep. 59 (2019) 208–212.
[7] M. Azadbakht, S. Azadbakht, S. Daniali, M. Dehghani, Comparison of the prevalence of perforated appendicitis during and before COVID19 pandemic, Ann. Med. Surg. 82 (Oct 1, 2022) 104785.
[8] C. Sohrabi, G. Mathew, N. Maria, A. Kerwan, T. Franchi, R.A. Agha, The SCARE 2023 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, Int. J. Surg. Lond. Engl. 109 (5) (2023) 1136.
[9] D. Gunsahin, M. Ilie, O. Plotogea, D.N. Paduraru, A. Bolocan, O. Andronic, F. Musat, V. Baleanu, D. Davitoiu, M. Pahomeanu, B. Dumbrava, ERCP extraction of stones in situs inversus patients; state-of-the-art techniques, J. Mind Med. Sci. 11 (1) (2024) 256–260.
[10] C.O. Blacio Villa, J.C. Jeanneth Elizabeth, M.J. Villacreses Portero, C.V. Sogso Chano, Laparoscopic gallbladder surgery in a patient with situs inversus totalis and cholecystitis without stones, J. Adv. Zool. 44 (Jan 2, 2023).
[11] J.D. de Meira Júnior, J.R. Aranda, J.C. Vidales, G. Ochoa, Millan G. del Angel, E. V. Corria, M.A. Mercado, I.D. Rosado, Bile duct injury repair in a patient with situs inversus totalis, HPB 26 (Jan 1, 2024) (S461-2).
[12] Thakral AK, Gupta T, Farooq A, Zahoor M. Laparoscopic Cholecystectomy In Situs Inversus Totalis.
[13] N. Hirano, M. Iseki, K. Nakagawa, M. Mizuma, T. Kamei, R. Matsumoto, S. Miura, K. Kume, A. Masamune, M. Unno, A case report of perihilar cholangiocarcinoma in a patient with situs inversus totalis, Clin. J. Gastroenterol. (Apr 12, 2024) 1–8.
[14] Uluşahin M, Çekiç AB, Yıldırım R, Türkyılmaz S. Laparoscopic colecystectomy in a patient with situs inversus totalis. Farabi Tıp Dergisi; 3(1):24–7.
[15] S.V. Arya, A. Das, S. Singh, D.S. Kalwaniya, A. Sharma, B.B. Thukral, Technical difficulties and its remedies in laparoscopic cholecystectomy in situs inversus totalis: a rare case report, Int. J. Surg. Case Rep. 4 (2013) 727–730.
[16] S. Kumar, G. Fusai, Laparoscopic cholecystectomy in situs inversus totalis with left-sided gall bladder, Ann. R. Coll. Surg. Engl. 89 (2007) W16–W18.
[17] T.E. Pavlidis, K. Psarras, A. Triantafyllou, G.N. Marakis, A.K. Sakantamis, Laparoscopic cholecystectomy for severe acute cholecystitis in a patient with situs inversus totalis and posterior cystic artery, Diagn. Ther. Endosc. 2008 (2008) 465272.