

Case Report

Open Cholecystectomy in Situs Inversus Totalis: A Case Report

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ABSTRACT

Situs Inversus Totalis (SIT) is a rare congenital positional anomaly, which is characterized by transposition of abdominal viscera associated by Dextrocardia. Its incidence is reported variously between 1 in 6,000 to 35,000 live births. This condition can pose difficulties in diagnosis and management. Herein we present a case of SIT. A 60 years old patient presented with pain at left Hypochondrium. Who was found to have SIT with cholelithiasis and managed by open cholecystectomy.

Key words: Situs inversus, Diagnosis, Left Hypochondrium, Cholelithiasis.

INTRODUCTION

Situs inversus totalis (SIT) is a congenital positional anomaly, which is characterized by transposition of abdominal viscera associated with right sided heart (Dextrocardia).^[1,2] First historical description of situs inversus (SI) was described in animals by Aristotle^[3-5] and by Fabricius in humans.^[6] Marco Severino first recognized dextrocardia in 1643. More than a century later, Mathew Baillic described the complete mirror image, reversal of the thoracic and abdominal organs in situs inversus.^[7]

SIT is a rare condition; its incidence has been reported variously between 1 in 6,000 to 35,000 live births,^[8] with male: female ratio of 3:2.^[9] Most of the cases of SI are thought to be due to a sporadic genetic mutation, but other pattern of inheritance have been described (autosomal dominant, autosomal recessive and x-linked recessive), and more than 20 genes are responsible for this phenomenon.^[10-12] There are two major types: Situs inversus

and situs ambiguous: Inversus indicates a position that is the mirror image of normal, whereas ambiguous refers to abnormal position of an organ, but in an unordered way.^[10] The normal development requires a 270 degree counter clockwise rotation to normal anatomy but in SIT the 270 degree rotation is in clockwise direction.^[13] The heart is located on right side of thorax, the stomach and spleen on right side of abdomen and the liver and gall bladder on the left side. The left lung is trilobed and right lung is bilobed. The transposition of organs may be associated with other congenital anomalies, such as renal dysplasia, biliary atresia, congenital heart disease or pancreatic fibrosis.^[14,15] SIT may be associated with bronchiectasis, chronic sinusitis and deficient tracheobronchial cilia and it is known as Kartagener's Syndrome.^[14,16]

Many people with SIT are unaware of this unusual anatomy until they seek medical attention for an unrelated condition. Cholelithiasis is one of the most common

diagnoses requiring surgical management, it can be difficult to correctly diagnose in a patient with SI. We present a case of Situs inversus totalis, which was managed by Open Cholecystectomy.

CASE REPORT

60 yrs old patient, who was a carpenter by profession, presented with pain at left hypochondrium on and off from last 6 months. Pain was dull in nature, radiated to left shoulder and left scapula. There was a history of single episode of biliary colic. Abdominal examination was unremarkable except a supraumbilical midline laparotomy healed scar (Fig. no. 1) of 5 cm.



Figure.1: Supra umbilical midline scar



Figure 2: Cholelithiasis in Left sided gall bladder

Laparotomy was done 3 yrs back for intestinal injury, which he received by a sharp wooden piece while working at the workshop. He was afebrile and not jaundiced. He denied any past medical history. His apex beat was found in right hemithorax at 5th intercostals space on

physical examination. Imaging by an ultrasound scan showed abdominal situs inversus with cholelithiasis (Single stone of size 14 mm) in a left sided gall bladder without any intra or extra biliary duct dilatation (Fig. no. 2).

He was found hypertensive in the out patient department and his Blood pressure was controlled by the physician. A preoperative chest X-Ray showed Dextrocardia (Fig no. 3) consistent with situs inversus and there was no evidence of bronchiectasis. Electrocardiography showed extreme rt. Axis deviation, non progression of R wave and finding were suggestive of Dextrocardia. Laboratory investigations revealed normal liver functions, normal coagulation profile along with normal other preoperative investigations. After consent the patient underwent open cholecystectomy. The operative technique performed was identical as for a routine open cholecystectomy. However the positions of team members were completely reversed. The surgeon stood on the left side of the patient and the left subcostal incision was utilized (Fig no. 4). There were no adhesions encountered upon entering the peritoneal cavity. The callot's triangle was identified. The relative positions of cystic duct, common bile duct and cystic artery were normal, with no exception of being 180 degree reversed. Both cystic artery and cystic duct were ligated separately and divided. Gall bladder removed from gall bladder fossa (Fig no. 5).

Abdomen was closed in layers with drain placed. Open cholecystectomy was carried out uneventfully. The post operative recovery of the patient was uncomplicated. Drain was removed on second postoperative day. He tolerated the regular diet on second post operative day. His Stitches were removed and he was discharged on 7th postoperative day. He was called on 14th post operative day in follow up and his prognosis of the case was good.



Figure No. 3: X-Ray showing Dextrocardia Figure No. 4: Left subcostal incision Figure No. 5: Gall bladder being removed

DISCUSSION

SIT is a rare condition, affecting approximately 1 in 10,000 individuals. [8] In 1600, Fabricius reported the first known case of SI in humans. [6] Though SI in its own is not pathological, it may be associated with Cardiorespiratory, Hepatopancreaticobiliary, Gastrointestinal, Neurological, Orthopaedic and Urological anomalies, some of which may be life threatening. [17,18] About 25% patients (with right sided heart) may have Kartagener's syndrome as well (also known as primary ciliary dyskinesia or immotile syndrome), characterized by SI, paranasal sinusitis and bronchiectasis. [19] Thus the patients with SI, who are scheduled for surgery, should be assessed preoperatively for any potentially serious cardiac or respiratory abnormalities as the mirror-imaged orientation while operating on a left sided gall bladder requires mental adaptability and mental dexterity to cope up with any evolving difficulties or potentially dangerous intra-operative situations. [20] Our patient was not having any life threatening condition including Kartagener's syndrome, reported so far.

There have been isolated reports of SI associated with peptic ulcer, [21] ulcer perforation, [22] amoebic liver abscess, [23] acute cholecystitis, [24] cholelithiasis, [3,25] acute appendicitis [26] and intestinal obstruction, [27] but there is no evidence to suggest that cholelithiasis occurs in greater frequency in SI population. [28]

In surgery department most common presentation of patients is abdominal pain. [26,29] Though SI does not predispose one to gall bladder disease, but it leads to diagnostic confusion. [30] Most of the patients present with left sided upper abdominal pain, however, about 10% of patients with left sided cholelithiasis present with right sided abdominal pain. [30] This phenomenon has been observed for both visceral biliary pain and somatic pain in cases of cholecystitis and suggests that the central nervous system may not share in general transposition. [31] Apart from the confusion related to the side of pain, the spectrum of clinical presentation related to complication of left sided cholelithiasis is similar to that occurring right sided gall bladder. [30] In our study the case of cholelithiasis was having pain at left hypochondrium radiating to left shoulder and left scapula.

The recognition of SI is important for preventing surgical mishap that result from the failure to recognize a reversed anatomy or an atypical history. If a surgery is planned on the basis of radiographic finding in a patient with SI, The surgeon should play careful attention to image labeling to avoid errors such as intervention on the incorrect side of the patient. An apical beat in the right fifth intercostal space, reversed side of the liver dullness and the right testicles hanging lower than the left, occasionally suggest SIT. [30] Echocardiogram, Ultrasonography, abdominal CT, Chest scan and MRI will

confirm the presence of gall stones and the left sided gall bladder. Some authors suggest that it would be very useful to perform a Magnetic Resonance Cholangio-Pancreatography (MRCP) procedure in order to reveal the exact anatomy of biliary tract, [32] thus decreasing intra-operative complications and enabling better planning of surgical procedures.

There have been several published case reports of performing surgery on patients with SIT such as Cholecystectomy, Common bile duct exploration, Appendectomy, Nissan Fundoplication, Hemicolectomy, Roux-en-Y gastric bypass, Sleeve gastrectomy, and Gastric banding. [5,20,33-38] In the published literature, there have been only 40 reports of open cholecystectomy in the prelaparoscopic era and 20 reports of laparoscopic cholecystectomy in patients with SI. [16,30,39] While both laparoscopic and open cholecystectomy procedures are possible in SIT. We planned the open cholecystectomy in our case, as the patient had undergone laparotomy 3 years back and the scar was situated at the midline, just above umbilicus (Fig. 1), thus due to expected adhesions and to avoid injury, open cholecystectomy was planned. In our case, the surgical team changed sides with the primary surgeon on the patient's left and the first assistant on the right. Cholecystectomy was performed through left subcostal incision. The mirror image anatomy poses difficulty in orientation during cholecystectomy. There are several important aspects of management of gall stones in the patients with situs inversus that are worth highlighting. In our case both the surgeon and first assistant were right handed and cholecystectomy in these patients is technically more demanding and needs reorientation of visual-motor skills to the left upper quadrant. [33]

The anatomical variation and the contralateral disposition of the biliary tree demand an accurate dissection and exposure to biliary structures to avoid iatrogenic injuries. In our case, it was important that

we identified the critical view of safety and dissected triangle of callot (Common Hepatic duct medially, cystic duct inferiorly and the liver edge superiorly), while there is no evidence to suggest that there is increased risk of bile duct injuries in patients with SIT, the orientation and ergonomic challenges may result in an increased operation time. [34] Apart from mirror image transposition, patients with SIT usually do not have associated extrahepatic biliary, venous and arterial anomalies. [35-38] Chandraraj S in 1976 [40] reported that the common hepatic artery originating from the superior mesenteric artery variant occurs in 17% of persons with normal anatomy. In our case we did not encounter any adhesions, biliary, venous and arterial anomalies.

CONCLUSION

Undiagnosed SIT with symptomatic gall stones provides a diagnostic dilemma for clinician as the symptoms are predominantly on left side. The principles of surgery are the same except for the change in position of operating team. Surgery in patients with SIT is technically more demanding in right handed surgeon and needs reorientation of visual-motor skills.

REFERENCES

1. Yokoyama T, Copeland, Jenkis N A, Montgomery C A, Elder F F and Overbeek PA. Reversal of left -right asymmetry: a situs inversus mutation. *Science*, 1993; 260 (5108), 679-682.
2. Gedda Sciacca A, Brenci G et al. Situs Viscrum Specularis in a monozygotic twin. *Acta Genet Med Gemellol (Roma)*, 1984; 83 (1), 81-5 PMID6540028.
3. Pathak K A, Khama R and Khanna N N. Situs inversus with cholelithiasis. *J Postgrad Med.*, 1995; 41(2), 45-6 [PUB Med].
4. Jobanputra S, Safar B and Wexner D D. Laparoscopic diverticular resection with situs inversus totalis (SIT): Report case. *Surg Innov.*, 2007; 14(4), 284-6 [Pub Med]

5. Kang S B and Han H S. Laparoscopic exploration of the common bile duct in a patient with situs inversus totalis. *J. Laparoendosc Adv Surg Tech*, 2004; 14(2): 103-6 [Pub Med].
6. Blegen H M. Surgery in situs inversus. *Ann Surg.*, 1949; 129: 244-251.
7. Harler D J and O'Leary P W. Cardiac malpositions and abnormalities of atrial and visceral sites In: Emmanouilides GC, Riemenschneider TA, Aleen HD, Gustessell HP (eds). *Heart disease in infants, Children, adolescents.. Williams & Wilkins, Baltimore, 5th ed. Vol 2. 1995; 1307-1336.*
8. Adeyekun A A, Onunu A N and Mazeli F O. Dextrocardia with situs inversus: A case report. *West African Journal of Medicine*, 2004; 22 (4): 358-360.
9. Yamaguchi S, Orita H, Yamaoka T, Mii S, Sakata H and Hashizuma M. Laparoscopic assisted distal gastrectomy for early gastric cancer in a 76 year old man with situs inversus totalis. *Surg Endosc.*, 2003; 17(2): 352-353.
10. Morelli S H, Young L, Reid B, Ruttenberg H and Bamshad M J. Clinical analysis of families with heart, midline and laterality defects. *Am J Med Genet.*, 2001; 101 (4): 388-92. [Pub Med].
11. Kumar S. and Fusai, G. Laparoscopic cholecystectomy in situs inversus totalis with left sided gall bladder. *Ann R Coll Surg Engl.*, 2007; 89 (2): 16-8 [Pub Med].
12. Casey B. Genetics of human situs abnormalities. *Am J med Genet.*, 2001; 101 (4): 356-8 [Pub Med].
13. Song J Y, Rana N and Rottman C A. Laparoscopic appendectomy in a female patient with situs inversus: Case Report and literature review. *JLSLS*, 2004; 8: 175-7.
14. Nursal T Z, Baykal A, Iret D and Aran O. Laparoscopic cholecystectomy in a patient with situs inversus viscerum: A case Report. *Can J. Surg.*, 1992; 35(1): 65-6.
15. Campos L and Sipes E. Laparoscopic cholecystectomy in a 39-year old female with situs inversus: Case report and literature review. *J Laparoendosc Surg.*, 1991; 1:123-6.
16. McKay D and Blake G. Laparoscopic cholecystectomy in situs inversus totalis: A case report. *BMC Surg*. 2005; 5:5.
17. Varano N R and Merkin R J. Situs Inversus : review of literature, report of four cases and analysis of the clinical implication. *J int Coll Surg.*, 1991;33:131-5.
18. Johnson J R. Situs Inversus with associated abnormalities. *Situs Inversus with associated abnormalities. Review of literature and report of three cases. Arch Surg.*, 1949; 58:149-54.
19. Maldjian P D and Saric M. Approach to dextrocardia in adults: Review, *AJR Am J Roentgenol.* 2007; 188(6 Suppl): S 39, 49; quiz S 35-8 [Pub Med]
20. Kumar S and Fusai G. Laparoscopic Cholecystectomy in situs inversus totalis with left sided gall bladder. *Ann R Coll Surg Engl.* 2007;89(2) :W 16-8.
21. Zaporozhets V K, Chupryna V V, Vasilenko, NJ and Mal'Ko. Lazvennaia bolezni zheludka pri polnoi inversiivnufrennikh organor [in Russian]. *Klin Med (Mosk)*, 1980; 58: 95-96.
22. Ghandhi DM, Warty PP, Pinto A C and Shetty SV. Perforated DU with dextrocardia and situs inversus. *J Postgrad Med.*1986; 32: 45-46.
23. Ansari Z A, Skaria J, Gopai M S, Vaish S K and Rai A N. Situs inversus with amoebic liver abscess. *J Trop Med Hyg*, 1973; 76: 169-170.
24. Heinmann T and Sailer A. Acute cholecystitis with situs inversus. *NY State J Med.*, 1979; 79: 253-254.
25. Mcfarland S B. Situs inversus with cholelithiasis. A case report. *J. Tenn Med Assoc*, 1989 82, 69-70.
26. Ucar A E, Ergul E, Aydin R, Ozgun Y M and Korukluoglu B. Left-sided acute appendicitis with situs inversus totalis. *Internet journal of Surgery*; 2007;12:9.
27. Ruben G D, Templeton, J M and Ziegler M M. Situs inversus. The complex inducing neonatal intestinal obstruction. *J Ped Surg.*, 1983; 18: 751-756. 10.1016/S0022-3468(83)80018-9.
28. Wood G O and Blalock A. Situs inversus totalis and disease of biliary track. *Arch Surg*, 1940; 40: 885-90.

29. Nelson M Jand Pesola G R. Left lower quadrant pain of unusual cause. *J Emerg Med.* 2001; 20 (3):241-5.
30. Takei H T, Maxwell J G, Clancy T Vand Tinsley E A. Laparoscopic Cholecystectomy in situs inversus totalis. *Journal of Laparoscopic Surgery*,1992; 2(4):171-6.
31. Sato M, Watanable Y, Iseki S, Akehi S, Tachibana M, Sato N, et al. Hepatalithiasis with situs inversus, first case report. *Surgery*, 1996; 119:598-605.
32. Kamitani S, Tsutamoto Y and Hanasawa K. Laparoscopic Cholecystectomy in situs inversus totalis with “Inferior cystic artery: a case report. *World J Gastroenterol.* 2005; 11: 5232-5234.
33. Aydin U, Unalp O, Yazici P, Gurcu B, Sozbilen M.and Coker A. Laparoscopic cholecystectomy in a patient with situs inversus totalis. *World J Gastroenterol.* 2006; 12(47): 7717-9. [Pub med]
34. Ersoy E, Koksall H and Ege B. Laparoscopic gastric banding for morbid obesity in a patient with situs inversus totalis. *Obes Surg.* 2005;15(9): 1344-6 [Pub Med]
35. Fujiwara Y, Fukunaga Y, Higashino M, Tanimura, S, Takemura, M, Tanaka, Y, et al. Laparoscopic Hemicolectomy in a patient with situs inversus totalis. *World J Gasteenterol.* Oct 2007;13 (37):5035-7. [Pub Med].
36. Catheline J M, Rosales C, Cohen R, Bihan H, Fournier J L, Roussel J,et al. Laparoscopic Sleeve gasterectomy for a super-super-obese patient with situs inversus totalis. *Obes Sug.* 2006; 16(8):1092-5
37. Tsung A, Feliz A and Kane TD. Laparoscopic nissen fundoplication in an infant with situs inversus. *JLap-aroendosc Adv Surg Tech.* 2007; 17(5): 698-700 [Pub Med] .
38. Koo KP, Laparsopic nissen fundoplication in a patient with situs inversus totalis: An ergonomic consideration. *J Laparoendosc Adv Surg Tech*, 2006; 16(3):271-3 [Pub Med].
39. Oms NM and Badia J M. Laparoscopic cholecystectomy in situs inversus totalis-the importance of being left handed. *Surg Endosc*, 2003;17:1859-61.
40. Chandraraj S. Observation on some additional abnormalities in situs inversus vicerum. *Journal of anatomy*, 1976;122: 377-88.

How to cite this article: Aziz I, Albina. Open cholecystectomy in situs inversus totalis: a case report. *Int J Health Sci Res.* 2018; 8(1):292-297.
