Journal of Medical and Pharmaceutical Innovation
ISSN : 2347 - 8136

Review Article

General Abdominal Surgery in Patients with Situs Inversustotalis

Takehiro Okabayashi MD[1], YasuoShima MD[1], Tatsuaki Sumiyoshi MD[1], Akihito Kozuki MD[1], Jun Iwata MD[2], Manabu Matsumoto MD[2], Yasuhiro Hata MD[3], Yoshihiro Noda MD[3], Akihito Nishioka MD[3]and Sojiro Morita MD[3]

[1]Department of Gastroenterological Surgery, Kochi Health Sciences Center
[2]Department of Diagnostic Pathology, Kochi Health Sciences Center
[3]Department of Radiology, Kochi Health Sciences Center

ABSTRACT:
In this article, we revisit the basic concept of SIT and review recently published articles on both changing trends in diagnostic and treatment modalities for SIT and notes for treating patients with SIT. Surgery in patients with SIT remains a technical challenge for the surgeon; however, it also remains extremely rare and is usually not encountered more than once in a surgeon's lifetime. Accurate modification to the standard protocol for general abdominal surgery is generally required to treat patients with SIT. Also, given the low, but real, incidence of associated gastrointestinal conditions, surgeons strongly favor preoperative evaluations to clearly delineate the anatomical variation because, generally, the contralateral disposition of the important organs demand an accurate dissection and exposure of the individual structures to avoid iatrogenic injures. These considerations, when paid appropriate heed, make a surgical approach not only safe, but also expedient, and can ensure the optimal treatment in patients who have these special instances of common disease processes.

Keywords: situs inversustotalis, Abdominal surgery, contralateral disposition

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Address for Correspondence:
Takehiro Okabayashi
Department of Gastroenterological Surgery, Kochi Health Sciences Center
2125-1 Ike, Kochi-City, Kochi 781-8555, Japan
Telephone: +81- 88-837-3000, Fax: +81- 88-837-6766
E-mail; takehiro_okabayashi@khsc.or.jp
Conflict of Interest: None declared

(Received 28 Septembert 2015; Accepted 30 October 2015; 1 November 2015) ISSN: 2347-8136 ©2014 JMPI

INTRODUCTION

Situs inversus is a rare, congenital condition in which the major visceral organs are reversed or mirrored from their normal arrangement, known as situs solitus. Aristotle was the first to reportsitus inversus in animals and considered it to signal a visitation from the gods. Dextrocardia (the heart being located on the right side of the thorax) was first seen and drawn in humans by Leonardo da Vinci(1452-1519), and then again noted by Marco Aurelio Severino in 1643, while situs inversus in humans was first described more than a century later by Matthew Baillie. Recent reports indicate that situs inversus occurs in 0.01% of the population, i.e., a 1 in 10,000 chance [1].

To the best of our knowledge, only 151 reported cases of successful general abdominal surgery for patients with situs inversus totalis(SIT) are accompanied bycomprehensive and publically available data, as revealed by a computer MEDLINE search of published literature [2-140]. In this article, we revisit the basic concept of SIT and review recently published articles on both changing trends in diagnostic and treatment modalities for SIT andnotes fortreating patientswith SIT.

DATA COLLECTION

Our survey of the literature from 1971 to 2014 revealed 151 patients with SIT who had undergone general abdominal surgery [2-140], with each case diagnosed as truly indicated for

surgical management in the individual department. For these cases, we reviewed country of the patient and clinicopathological data including types ofdisease and surgery involved, as well as the changing trends in the incidence of malignant neoplasms in patients with SIT and differences in surgical management for patients with SITacross the geographical locations. The changing trends were evaluated by Cochran-Mantel Haenszel statistics and analyzed using SPSS® (SPSS; Chicago, IL).*P* values less than 0.05 were considered to be statistically significant.

OVERVIEW

SIT: Mapping the reported regions in the world

A surgically treated SIT patient also suffering from colon cancer was first reported in 1971 in the USA. Since then, reports of similar cases have gradually increased globally since [125], with some authors commenting on the increasing concentrations of SIT patientswhorequireabdominal general surgery, i.e., that a small number of countries have spent the largest sums (Fig. 1). The 29 countries of origin reported in the reviewed literature are as follows in order of frequency (Fig. 1), with the 11 countries with the highest reporting account for over 84% of the total case number:Japanaccounts for 19.9% of the world total, followed by India (12.6%), USA (11.9%), Turkey (10.6%), Korea (7.3%), Italy (5.3%), UK (5.3%), Greece (4.0%), China (2.6%), France (2.0%), and Saudi Arabia (2.0%).

SIT: Backgrounddiseases

The background diseasesinvolved with the 151 SIT patients could bedivided into nine categories (Fig. 2). Of all cases, 40.4%involvedcholecystitis, including acute cholecystitis, chronic cholecystitis, and cholecystlithiasis [2-60], while approximately 43%involved diseases of the alimentary tract, such as colorectal diseases, gastric neoplasms, appendicitis, super-obesity, gastro-esophageal reflux disease (GERD), and duodenal disorders [61-89,113-141], and only 15.9% ofpatients with SITshowedhepato-biliary-pancreatic neoplasms [90-112].

SIT: Kinds of abdominal surgical treatment

The 151 identified surgical procedures were categorized into nine different management categories, as represented here by a pie chart representing the number of identified surgical management protocols under each category (Fig. 3).Results for all general abdominal surgeries were analyzed and showed significant similarity to the known background diseases in patients with SIT. Cholecystectomy was reported as surgical treatment for 61 of the 151 (40.4%) cases in patients with SIT with significant similarity as clinical diseases wasperformed. Appendectomy, gastrectomy, and colorectal surgeryare also frequent procedures for patients with SIT, with 29.8% of thetotalcase reports reviewed citing one of these as a cause for surgery.In contrast, hepatobiliarypancreatic surgery (24 cases, 15.9%) remains achallenging procedure, although recent years have seen a gradual increase in surgical treatments for patients with SIT suffering from both obesity and GERD (15 cases, 9.9%) [75-89].

SIT: Changing trends

Among the 151reviewed general abdominal surgeries in patients with SIT, the background diseases ratio was 1.8:1, comprising benign diseases-64.9% (n= 98), andmalignant neoplasms- 35.1% (n= 53) (Fig.4). From 1970 to 2010, the absolute number ofreported SIT caseswho underwent general abdominal surgeryincreased three fold. Interestingly, the absolute value and prevalence of surgical management for malignant neoplasms in patients with SIT have also been increasing significantly. Indeed, approximately half (46.4%) of all reported cases from 2011 to 2014required surgical treatment for malignant neoplasms, although up to 2010, the rate of such cases was only 28.4%.

NOTES FOR TREATING PATIENTS WITH SIT

Cholecystectomy

Cholecystectomy is the most popular surgical procedureacross the reported patients with SIT [2-60].

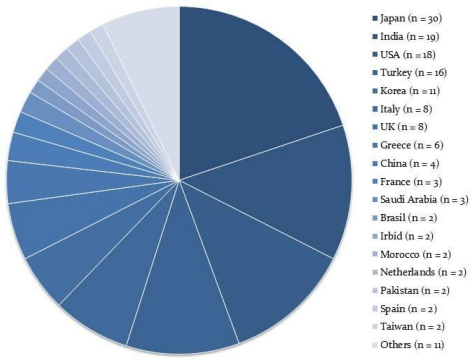

Figure 1

Figure 1: Mapping the reported regions in the world

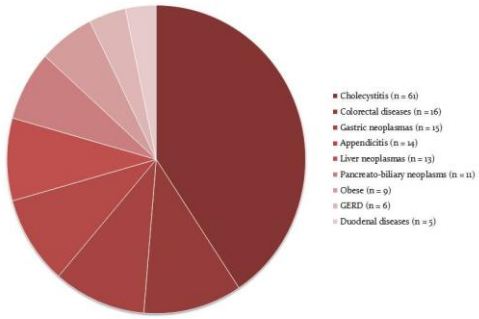

Figure 2

Figure 2: Background diseases in patients with situs inversustotalis

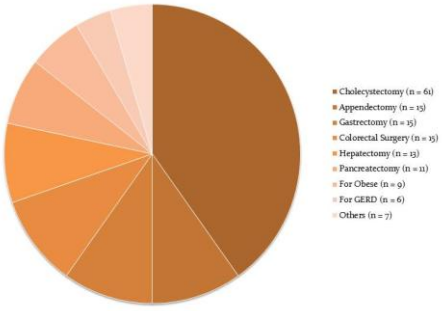

Figure 3

Figure 3: Kinds of abdominal surgical treatment

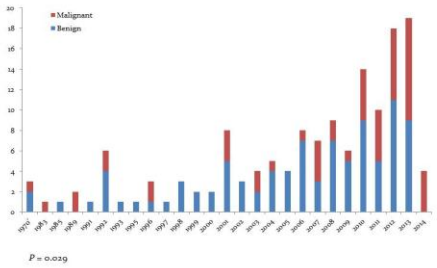

Figure 4

Figure 4: Changing trends

Laparoscopiccholecystectomyisatechnicallyestablishedprocedureinpatients suffering from cholecystitis;however, SITisstill an extremely rare condition and preoperative knowledge of the presence of malrotation is of prime importance for thesurgeon in terms of planning the operation strategy and minimizing the possibility of intra-operative complications. The mirror image anatomy in patients with SITposes difficulty in orientation during surgery, and isthe most challenging factor for performing cholecystectomy, especially laparoscopically because the surgeon needs to reorient visual images and surgical steps in an anatomical field that has undergone clockwise rotation. Thus, such surgical techniques should always be customized and arranged for the SIT condition.

Gastric Surgery

Gastric cancer is the second most common malignancy worldwide, and surgical treatment remains the only curative management option [141].With SIT [61-74], abnormal vascularization of the arteries and veins are common; therefore,preoperativeconfirmation of any abnormal vascularization is very important, due to the possible risk of misidentifying anatomy and unanticipated injury of important vessels during the lymph node dissection. The laparoscopic approach is being increasingly used (40.0%) for patients with SIT suffering from gastric neoplasms. Particularlywith laparoscopic surgery, it is essential to determine thepresence of vascular anomalies by preoperative enhanced computed tomography(CT) orangiography, because surgeons using a laparoscope cannot directly confirm directly the location of vessels by palpation.

In recent years, several authors have reported surgical procedures for obesity or GERD in patients with SIT, including Nissen fundoplication, gastric banding, sleeve gastrectomy, and gastro-jejunal bypass operation [75-89]. Of 12 such patients reviewed here, 11 patients (91.7%) underwent laparoscopic surgery. Surgical procedures, especially laparoscopic procedures, areconsidered more difficult in patients with SIT than in otherpatients because of the mirror-image anatomy. Thus, because laparoscopic gastricsurgery on patients with SIT presentstechnical challenges for the surgeon, it remains relativelyrare.

Liver surgery

Recent advances in surgical technology and perioperative management mean that hepatic surgical procedures, including liver resection and radiofrequency ablation, have become safer and are now the mainstay of curative treatment for liver tumors, such as primary liver cancer and secondary metastatic liver tumors. The diagnosis of liver neoplasms in patients with SIT is not difficult with the aid of radiological diagnostic modalities, but attention should be paid to detectingconcurrent abnormalities, especially in the hepatobiliary and cardiovascular system [90-100]. There are 13 reported cases of hepatectomy for liver malignancy in patients with SIT in the English literature, including 12 with hepatocellular carcinoma and 1 with secondary metastatic liver tumor from colon cancer. Considering that variation of the hepatic artery occurs in 20% of all such cases, it could

actually be related to the SIT. A long straight"left" portal vein branched to the "left liver" and "right" medial area was another notable finding. The round ligament terminated in this portion and was diagnosed as the "left" umbilical portion. Normally, the right umbilical vein disappears in the early fetal life, whereas the left umbilical vein partly remains resulting in the umbilical portion of the portal system, round ligament of liver, and Arantius ligament. If the left umbilical vein disappears early and the right umbilical vein remains, the latter develops into the umbilical portion and the round ligament deviates to the right. In addition, three main hepatic veins drain the liver into the inferior vena cava below the diaphragm. In the notes reviewed here the single hepatic vein sometimes drained directly to the right atrium. Thus, despite the rarity of SIT, its preoperative detection in patient undergoing liver surgery is crucial to preventingpotential injury to the hepatic veins during their dissection. These anomalies were the cause of surgical difficulty, whereas the surgical course was uneventful and the surgical technique for this patient did not differ from that for usual patients. Needless to say, cautious preoperative evaluation with imaging modalities, especially multi-detector CT, was a key factor for successful surgical treatment.

Pancreatic surgery

Improvements in surgical technique, increased surgical experience, and advances in anesthesia, intensive care, and parenteral nutrition have made pancreatic surgery a substantially safer procedure and the mainstay of curative treatment for periampullary and pancreatic head malignancy [142]. For pancreatic surgery in patients with SIT, advanced diagnostic imaging techniques, such as three-dimensionalCT, are highly recommended whenever available [104-112]. Especially in pancreaticoduodenactomy, advanced diagnostic imaging techniquesare very important for careful preoperative planning andto prevent misunderstanding of the arrangement of theabdominal viscera.Depending on the situation, the surgeon should change their position to ensure anadequate surgical procedure, if necessary. Furthermore, the assistantsshould support the operation appropriately given that thesurgeon has to 'reverse' the handling technique.

Appendectomy

The condition of appendicitis in patients with SIT may present difficulties in terms of the differential diagnosis for cholecystitis. Approximatelyone third of patients with appendicitis have pain localizedoutside the right lower quadrant, because the position of theappendix can vary considerably in any appendicitis patient.However, in cases with SIT, rare types of appendicitis can be encountered including subhepatic, lateral pouch, mesocoliac, left-sided (associated with SIT), intra-herniary, and lumbarappendicitis, becauseit is the only organ in the body that has no constant position.After establishing the diagnosis of appendicitis in patients with SIT, whether acute or chronic, the surgicaloptions are the same as for normal patients [128-140].A laparoscopic approach for appendectomy is superior in a patient with SIT, compared with the access that a McBurney incision provides, although the laparoscopic approach has to be tailored in each individual SIT case, because there are no standard port positions in these situations and the surgeon has to modify the port placements, adheringto the basic principles of laparoscopy.

Colorectal surgery

Colorectal surgery for patients with SIT is not as technically difficult compared to stomach, liver, and pancreatic surgery. However, attention to the fundamentals of the operative procedures, such as careful handling and maintaining a dry operating field, is very important. Of course, accurate preoperative anatomic assessment and careful preoperative planning of surgical procedures are also imperative to ensure a safe and smooth procedure, as mentioned in previous reports of colorectal surgery in patients with SIT [115-126]. Although advanced surgical skill is required for radical lymphadenectomy in a patient with SIT, careful recognition of the mirror image anatomy is more important.The reported prevalence of a laparoscopic approach for patients with SIT and suffering from colorectal diseases is 40.0%, and such procedures can be performed safely by a skilled surgeon and surgical team after thorough preoperative planning including assessment of the anomaly.

PERSPECTIVE AND CONCLUSIONS

Surgery in patients with SIT remains a technical challenge for the surgeon; however, it also remains extremely rare and is usually not encountered more than once in a surgeon's lifetime. Accurate modification to the standard protocol for general abdominal surgery is generally required to treat patients with SIT. Also, given the low, but real, incidence of associated gastrointestinal conditions, surgeons strongly favor preoperative evaluations to clearly delineate the anatomical variation because,

generally, the contralateral disposition of the important organs demand an accurate dissection and exposure of the individual structures to avoid iatrogenic injures. These considerations, when paid appropriate heed, make a surgical approach not only safe, but also expedient, and can ensure the optimal treatment in patients who have these special instances of common disease processes.

ACKNOWLEDGMENT: This work was supported by the Kochi organization for medical reformation and renewal grants.

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