



## Case Report

## Surgical challenges during open pancreaticoduodenectomy in a patient with situs inversus totalis: A rare case report and literature review

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## ABSTRACT

**Background:** Situs inversus totalis is a rare congenital anomaly defined by a mirror-image of thoracic and abdominal viscera. Discrete cases of situs inversus totalis and its association with gastrointestinal tumors have been reported. Here we report the first case of pancreatic-head serous cystadenoma in patient with situs inversus totalis.

**Case presentation:** A 68-year-old woman presented with an abdominal mass that appeared four months ago. She was otherwise asymptomatic and her physical examination was unremarkable. Chest X-ray revealed dextrocardia. CT scan confirmed situs inversus totalis with an irregular, clear border, heterogenous pancreatic-head mass measuring 11 cm. Laboratory studies were within the reference range and pancreatic tumor markers were normal. We performed an elective open pancreaticoduodenectomy followed by an end-to-side pancreaticojejunostomy, an end-to-side choledochojunostomy, and a side-to-side gastrojejunostomy. The immediate postoperative course was uneventful, and she was discharged four days later without any complications. Four-month of follow-ups revealed no recurrent or relapsed disease.

**Discussion:** Although the steps of the Whipple procedure are almost the same in SIT patients. The main differences during the operation in SIT patients are the anatomical variations and how the surgeon will cope with them to avoid any mistakes.

**Conclusion:** The surgeons should improve their skills and gain control in both hands to easily adjust with the anatomic variations of situs inversus totalis and reduce the operation time and the associated risk of long operation time.

## 1. Introduction

Situs inversus totalis (SIT) is a rare congenital anomaly characterized by an entire reversal of both thoracic and abdominal organs [1]. Its incidence rate varies from 1:8000 to 1:25,000 [2]. SIT develops when the cardiac tubes rotate to the left, and the heart and other internal organs are positioned in a mirror image of the normal-organ anatomy (i.e., left-sided liver and gallbladder, lung with three lobes, and inferior vena cava and right-sided aorta, lung with two lobes, stomach, spleen, and cardiac apex) [1,3].

It can present either as an isolated, asymptomatic condition or as a component of other syndromes [4]. Cardiovascular, gastrointestinal,

and respiratory congenital anomalies, even anatomic ones (i.e., Kartagener syndrome, polysplenia, biliary atresia, and midgut malrotation), were observed to be more frequent in SIT [5]. The current data proposes no proven association between SIT and malignancies occurrence [6,7]. However, scattered reports have shown that SIT has a remarkably high incidence of pancreatic, bile duct, and ampullary tumors [8].

In this review, we aim to report the first case in the literature of pancreatic-head serous cystadenoma and to address these inquiries: 1) Does the presence of SIT suggest surgical management of pancreatic serous cystadenoma? 2) What are the technical differences between SIT and non-SIT patients when performing the Whipple procedure? 3) Does the presence of SIT affect the outcome and prognosis? 4) Address the

**Abbreviations:** SIT, Situs inversus totalis; PD, Pancreaticoduodenectomy.

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difficulties and challenges of performing the Whipple procedure in the presence of anatomical variations.

## 2. Case presentation

A 68-year-old woman presented to our department with an abdominal mass that appeared four months ago. Her vital signs were within normal limits. Her physical examination was unremarkable apart from the mentioned mass. She had a history of well-controlled hypertension for seven years. The remainder of her medical, surgical, and familial history was unremarkable. Laboratory tests were within reference ranges. Chest X-ray revealed dextrocardia. An electrocardiogram exposed right axis deviation and inverted P waves in leads I, aVR, and aVL (Fig. 1). CT scan exposed dextrocardia. The ascending aorta to the left of the midline, the aortic arch crosses the midline posteriorly to the right, and a right-sided descending aorta (Fig. 2). The inferior vena cava was found to the left of the descending aorta and its major branches. A right-sided stomach and spleen and a left-sided liver were observed (Fig. 3). An irregular, clear border, heterogenous pancreatic-head mass measuring 11 cm, pancreatic atrophy and calcifications with mild dilation of the pancreatic duct were noticed (Fig. 4). Pancreatic amylase, serum alpha-fetoprotein, carcinoembryonic antigen, CA-125, and CA19-9 were within normal limits.

After taking the informed consent, she underwent an elective open pancreaticoduodenectomy (PD). The operator stood on the left side of the patient. A bilateral subcostal Kocher's incision was performed (Fig. 5). The hepatic artery was located to the right of the portal vein, and the common duct was to the left. We noticed that the transverse mesocolon was shorter than usual with no more than 3 cm midcolic vein length. The hepatic flexure of the colon was shifted to expose the duodenum and the head of the pancreas, which were then mobilized by Kocher's maneuver. A "reversed" Kocher maneuver was performed; the peritoneum was incised at the left edge of the duodenum; rather than the right, and the duodenum and the head of pancreas were retracted away from IVC (to the right; rather than the left) in a cephalad direction. The pancreatic head was located to the left of the midline, and the tail extended towards the right upper quadrant. The cecum and appendix were in the left lower quadrant, and the sigmoid colon was in the right lower quadrant. The superior mesenteric artery was identified posterior to the neck of the pancreas, and the confluence of the portal and superior mesenteric veins was positioned posterior to the body of the pancreas.

The splenic vein crossed the right upper quadrant and drained to the lateral wall of the superior mesenteric vein and portal vein confluence. We performed cholecystectomy and pancreaticoduodenectomy followed by an end-to-side pancreaticojejunostomy, an end-to-side choledochojenostomy, and a side-to-side gastrojejunostomy.

The operative time was 330 minutes and Intraoperative blood loss was approximately 500 ml. She did not need a blood transfusion. Gross examination showed a well-circumscribed encapsulated mass measures 12 × 8 cm in the pancreatic head (Fig. 6). The cut surface was a multi-located, sponge-like appearance with a central scar. No invasion towards the duodenum was noticed. The gallbladder was dilated and measured 11 × 5 cm with wall thickness less than 1 mm, and smooth, green-colored mucosa. No polyps or gallstones were noted. The histologic report confirmed the diagnosis of pancreatic serous cystadenoma (Fig. 7). The immediate postoperative course was uneventful, and she was discharged four days later without any complications. Four-month of follow-ups revealed no recurrent or relapsed disease.

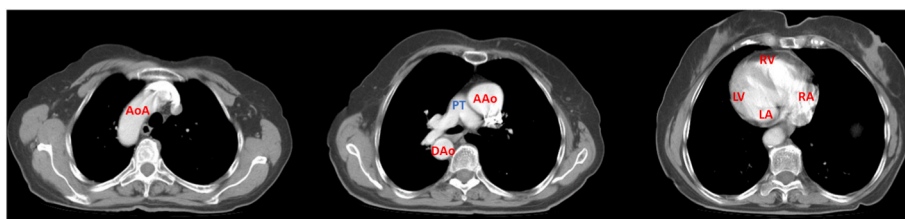
This case report has been reported in line with the SCARE Criteria [9].

## 3. Discussion

The current data proposes no proven association between SIT and malignancies occurrence, especially pancreatic adenocarcinoma since some evidence reinforces the genetic basis for their development [6,7]. However, pancreatic adenocarcinoma remains the furthest fatal solid malignancy [10]. Contrary, pancreatic serous cystadenoma is the common pancreatic cystic tumor and is almost always defined as a benign tumor [7]. The first case of pancreatic serous cystadenoma was reported in a 46 years old female patient by Paunipagar et al. [11]. They only focused on diagnostic imaging and have declared nothing regarding its management or prognosis. Our case is the second patient who had SIT and presented with a giant pancreatic serous cystadenoma. From the literature review (Table 1), the data poses an equal male-female distribution of SIT patients with pancreatic mass (male: 5; female: 3) with a mean age at presentation of 60 years (range 47–79 years). We noticed that most of the pancreatic neoplasms in the SIT patient were adenocarcinomas, and those patients presented with different symptoms. Jaundice and epigastric pain were predominant; less frequent symptoms were weight loss, clay-like stool, and nausea (Table 1) [3,6,12–15].



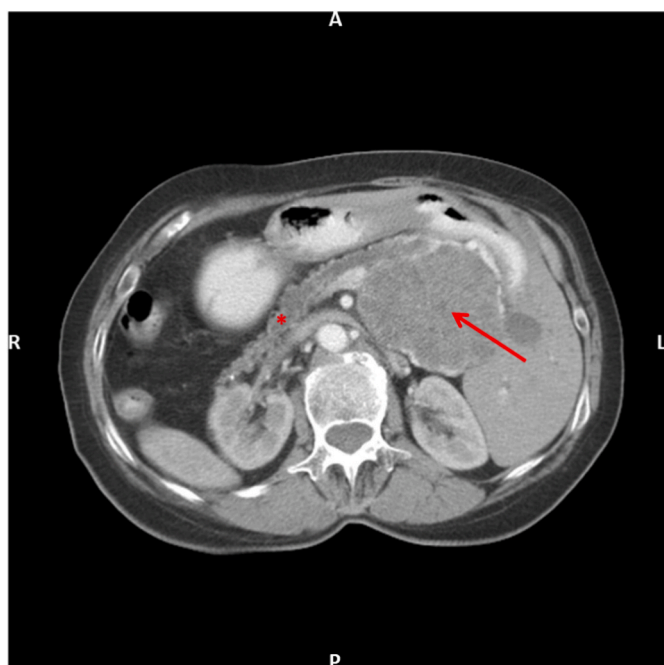
Fig. 1. An electrocardiogram of patient with situs inversus totalis exposes a right axis deviation and inverted P waves in leads I, aVR, and aVL.



**Fig. 2.** Shows the thoracic anatomic variations in patient with situs inversus totalis (from the left to right). the aortic arch (AoA) crosses the midline posteriorly to the right. The ascending aorta (AAo) to the left of the midline, a right-sided descending aorta (DAo), and the pulmonary trunk (PT) in between. Finally, dextrocardia.



**Fig. 3.** Reveals the abdominal anatomic variations in patient with situs inversus totalis. A right-sided stomach and spleen and a left-sided liver are noted. The inferior vena cava (IVC) is found to the left of the descending aorta (Ao) and its major branches (red arrows from the left to right: celiac trunk, superior mesenteric artery, and inferior mesenteric artery). (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)



**Fig. 4.** Shows an irregular, clear border, heterogenous pancreatic-head mass measuring 11 cm (red arrow), pancreatic atrophy and calcifications with mild dilation of the pancreatic duct (red asterisk) in patient with situs inversus totalis. (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)



**Fig. 5.** An open pancreaticoduodenectomy on patient with situs inversus totalis through left-subcostal Kocher's incision.

Surgery is the only curative treatment for tumors of the head of the pancreas and based on the published cases (Table 1), pancreaticoduodenectomy represents the golden standard procedure in managing pancreatic-head carcinoma in SIT patients [3,6,12–16]. The initial obstacle during surgery in SIT patients is the anatomical variations; therefore, using MRI, three-dimensional angiography with multidetector-row CT, and Doppler ultrasound can be practical to





**Fig. 6.** Shows a well-circumscribed encapsulated mass measures  $12 \times 8$  cm in the pancreatic head.

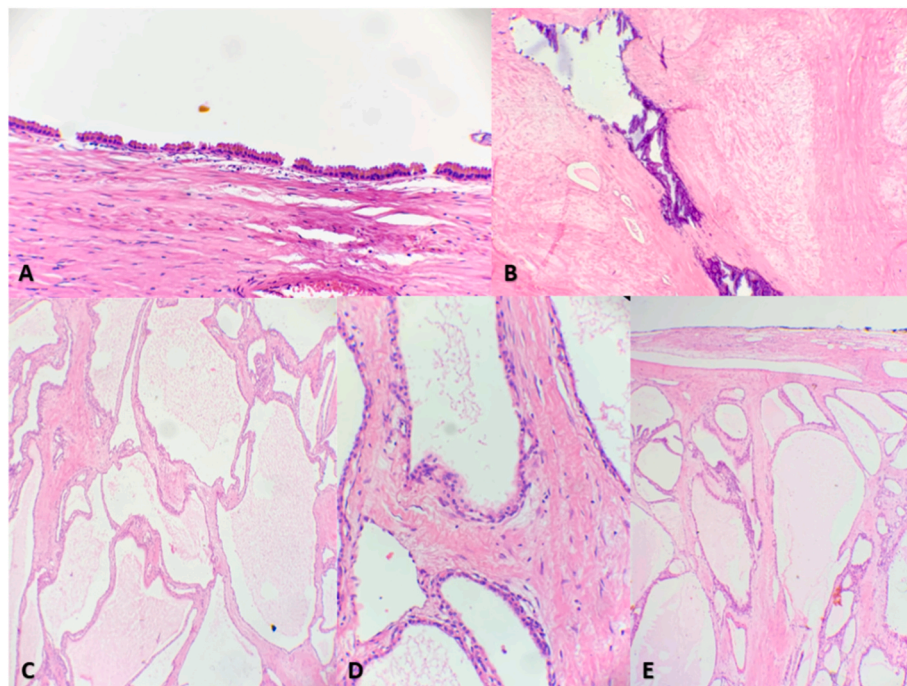
evaluate the tumor perfusion and to identify the anatomical variations before surgery rather than to confirm the diagnosis (Table 1) [6,12,14]. We were unable to order MRI, magnetic resonance cholangiopancreatography (MRCP), or CT angiogram as the patient could not afford their high cost. However, the CT scan along with the extreme caution and attention during the operation prevented any vascular injury and life-threatening hemorrhage.

Although the steps of the Whipple procedure are almost the same in SIT patients. The main differences during the operation in SIT patients are the anatomical variations and how the surgeon will cope with them to avoid any mistakes. Under normal circumstances, the surgeon stands on the patient's right side. This position provides more maneuverability to execute with the right hand. In our case, SIT imposes the surgeon to stand on the left side throughout the operation. Surgical incision of Whipple procedure can be either a vertical midline or bilateral subcostal

transverse; both could provide proper entrance [17]. Enormous attention is required to define the location of the major vessels and their branches during the operation. The Kocher maneuver was entirely reversed as the peritoneum was incised at the left edge of the duodenum rather than the right, and the duodenum with the head of the pancreas were retracted away from the IVC, to the right rather than the left. The surgeon should identify the arterial pulse within the porta hepatis to avoid possible vascular injury and life-threatening hemorrhage [18]. Portal vein dissection along the hepatic artery in the hepatoduodenal ligament was also reversed from the right to the left side of the patient. Stomach and jejunum transection, dissection of ligament of Treitz, rotating the duodenum under the mesenteric vessels, and pancreatic transection remain the same as in non-SIT patients.

Although Bilimoria et al. and Kyuno et al. [8,19] have established the safety and feasibility of performing open PD in patients with SIT. Chen et al. suggested that laparoscopic PD is more practical as it is minimally invasive and provides a broader field of vision. However, the difficulty in performing laparoscopic PD, the mandatory existence of accuracy, skills, and dexterity [20], and the diverse anatomical positions of the internal organs make the procedure further complicated and disadvantageous putting the final decision in the surgeon's hands based on the equipment and expertise [8,14]. The surgeon must have intense concentration while performing each step of the procedure and examine the abdominal viscera and the great vessels positions [5].

The median operative time for open PD in non-SIT patients is 354 minutes [21]. The operative time in our case was 330 minutes, while the operative times for PD in SIT patients were available only in two cases throughout the literature; 635 min [14] and 710 min [16]. We suggest that the surgical skills, dexterity, available equipment, and whether the tumor is benign or malignant could have a significant impact on operation time. Therefore, surgeons must have intense concentration while performing each step of the procedure and examine the abdominal viscera and the great vessels positions. In addition, they should improve their skills and gain control in both hands to easily adjust with the anatomic variations of SIT and reduced the operation time and the associated risk of long operation time (i.e., infections, prolonged duration of stay, 30-day mortality rate, and complications related to the



**Fig. 7.** Microscopic images of pancreatic serous cystadenoma revealing A) cuboidal epithelial lining B) calcified central scar C) cystic spaces ranged from 1 mm to 3 cm in diameter D) flattened epithelial lining E) the capsule mass.

**Table 1**

Exposes a summary of the relevant, published papers in patients with SIT.

Author Year	Sex	Age	Diagnosis	Symptoms	Investigations	Procedure	Stage	Outcome and Follow-up
Macafee et al., 2007	Male	67	Papillary adenocarcinoma	Epigastric pain, nausea, weight loss, and jaundice	CT scan, ERCP, EUS	PD <sup>a</sup>	T3 N0 M0 Stage IIA	A peritoneovenous shunt was performed 9 months postoperatively for intractable ascites, but there was no evidence of recurrent disease
Scusi et al., 2009	Female	48	Ductal adenocarcinoma	Epigastric pain and jaundice	US, CXR, ERCP <sup>b,c</sup> , and CT scan	Percutaneous transhepatic cholangiogram. Then PD	T3 N0 M0 Stage IIA	Five-month follow-up: no evidence of recurrent disease
Maruyama et al., 2009	Male	63	Invasive ductal adenocarcinoma	Epigastric pain and jaundice	CXR, CT scan, ERCP, and three-dimensional angiography with multidetector-row computed tomography	PD	T3 N1 M0 Stage III	Postoperative course was complicated by pancreaticoenteric anastomotic leakage. Uneventful recovery with no clear follow-up period
Zheng et al., 2013	Female	47	Adenocarcinoma	Epigastric pain and jaundice	MRI	PD	N/A <sup>d</sup>	Uneventful recovery with no clear follow-up period
Chen et al., 2015	Male	56	Adenocarcinoma	Epigastric, pain, jaundice, and weight loss	MRI, CT scan, Electrocardiogram, CXR, and three-dimensional angiography with multidetector-row computed tomography	Open PD	T3 N1 M0 Stage III	One-year follow-up: uneventful recovery without tumor recurrence or metastasis
Hussan et al., 2021	Male	52	Invasive ductal adenocarcinoma	Epigastric pain, jaundice, weight loss, and clay-like stool	Gastroscopy, Doppler US, and CT scan	Laparoscopic PD	T3 N0 M0 grade IIA	15-day follow-up: uneventful recovery
Nagai et al., 2022	Male	79	N/A	asymptomatic	CT	Open PD	T2, N2, M0 Stage III	N/A
Current case	Female	68	Serous cystadenoma	Asymptomatic	CXR, CT scan, ECG, and US	Open PD	N/A	6-month follow-up: uneventful recovery with no recurrent or relapsed disease

<sup>a</sup> PD: Pancreaticoduodenectomy.<sup>b</sup> ERCP: Endoscopic retrograde cholangiopancreatography.<sup>c</sup> Unsuccessful ERCP due to difficulty with cannulation of the inverted ampulla of Vater.<sup>d</sup> N/A: not applicable.

pancreas itself).

The follow-up period differs tremendously across patients with a minimum of 15 days and a maximum of up to one year (Table 1). However, postoperative recovery of all patients was rapid and uneventful with no evidence of recurrent disease and no fatalities except for one patient [14] who required a peritoneovenous shunt after nine months for intractable ascites, yet, there was no evidence of recurrent disease [3,6,12–15].

#### 4. Conclusion

In conclusion, the lack of informative data regarding SIT association with other diseases indorses more efforts to be achieved. However, the presence of pancreatic serous cystadenoma in a patient with SIT seemed would not have changed the prognosis nor the outcome of its surgical management. Although pancreaticoduodenectomy is more demanding and challenging to perform in situs inversus totalis, it usually results with good outcomes. Therefore, the surgeons should have a complete state of focus and take extreme care at each step during the operation. In addition, they should improve their skills and gain control in both hands to easily adjust with the anatomic variations of SIT and reduced the operation time and the associated risk of long operation time.

#### Ethic approval

Ethical approval from the IRB of Al Assad University Hospital was obtained.

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No funding was required.

#### Contributions

FA is the supervisor of this work. FA and SJ collected the patient's data and perform the literature review. SJ and HD drafted the manuscript. DA prepared the microscopic images. FA performed the procedure and revised the final manuscript. All authors discussed and contributed to the final manuscript.

#### Registration of research studies

Name of the registry:

Unique Identifying number or registration ID:

Hyperlink to your specific registration (must be publicly accessible and will be checked):

#### Guarantor

Sami Jomaa.

#### Consent

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

## Provenance and peer review

Not commissioned, externally peer reviewed.

## Declaration of competing interest

The authors have declared that they have no conflict of interests.

## Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.amsu.2022.104610>.

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