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# Three-dimensional reconstruction simulation assisted pancreaticoduodenectomy forpancreatic head carcinoma in a patient with situs inversus totalis: Report of a rare case

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ABSTRACT

Situs inversus totalis (SIT) is a rare congenital malformation that refers to a complete reversal of the thoracic and abdominal viscera. An extremely small number of SIT patients with pancreatic head cancer have been reported. Surgical procedures in patients with SIT are more complicated and technically difficult due to the rearrangement of anatomical structures. Adequate and meticulous preoperative assessment of tumor regional anatomy, potential cardiovascular anomalies, and aberrant blood vessels by imaging examinations is critical for facilitating the safe and effective performance of the surgery. The current study reports the case of a 69-year-old female with pancreatic head cancer and SIT, and first focuses on the application of 3D reconstruction simulation assisted pancreaticoduodenectomy (PD) in patients with SIT. Based on the data of CT images, we reconstructed a real-time 3D simulation model with Myrian XP to assist with our surgical planning. Then, we performed smoothly PD with complete resection of the tumor. The patient was discharged without any serious complications. Follow-up has been 2 years and the patient is doing well with no recurrences.

**Keywords**: Situs inversus totalis, pancreatic head cancer, pancreaticoduodenectomy, three-dimensional reconstruction



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## Introduction

Situs inversus totalis (SIT) is a rare congenital malformation, which manifested as a complete left-to-right reversal of the thoracic and abdominal viscera compared to their usual positions. The reported incidence rate ranges from 1:8000 to 1:25000 [1]. Although no evidence shows this condition does increase the risk of developing cancer, cases of SIT combined with tumors have been reported [2]. The coexistence of SIT and pancreatic head cancer is extremely rare. Reviewing the literature, the first case of pancreatic head carcinomacoexistent with SIT whounderwent pancreatoduodenectomy (PD) was reported by Sakaguchi in 1985 [3]. Up to now, only 7 cases of successful PD in SIT patients with pancreatic head carcinoma have been documented [3-9]. The performance of PD in patients with SIT is more complicated and technically difficultdue to the rearrangement of anatomical structures.Adequate and meticulous preoperative assessment of tumor regional anatomy, potential cardiovascular anomalies, and aberrant blood vessels by imaging examinations is critical for facilitating the safe and effective performance of the surgery.

To the best of our knowledge,diagnostic imaging techniques such as computed tomography (CT), magnetic resonance imaging (MRI), and related three-dimensional (3D) reconstruction were applied to preoperative evaluation in previously documented cases of PD in SIT patients [7-9], but there is no report of 3D reconstruction simulation assisted PD for the pancreatic head carcinoma concurrent with SIT. Regarding the relative positions of tumor,invasion of superior mesenteric vasculature and other adjacent organs, 3D reconstruction has advantages to provide more detailedinformation and intuitive sense compared with two-dimensional (2D) CT, increasing the safety of the operation and the resection rate of tumor [10, 11].

The present study reports a case of SIT patient with pancreatic head carcinoma, and first focuses on the application of the 3D reconstruction simulation assisted PD in patientswith SIT.

**Casereport**

A 69-year-old femalewith known SITadmitted to our hospital withepigastricpain and jaundice for 10days.There were no past medical history and no familyhistory ofSIT and tumors.Investigations revealed cancer of pancreatichead with SIT. Laboratory examination showed alanine transaminase (ALT), aspartate transaminase (AST) and total bilirubin (TBIL) were 106 U/L, 102 U/L and 142.2 μmol/L, respectively, which were all within a normal range. Serum CA19-9 was 191.6 U/mL, which was more than 7 times higher than the normal upper limit. Chest radiography showed mirror-image dextrocardia. Endoscopic retrograde cholangiopancreatogram (ERCP) showed stenosis at the junction of duodenal bulb and descending part of the duodenum, and the duodenoscope failed to pass through this junction.

The patient received a 128-slice spiral CT scan (GE Healthcare Life Sciences, Chicago, IL, USA), including plain scan and triple-phase enhanced scan. Briefly, gastrointestinal preparation was done prior to the scan, which included routine fasting for 4 hours, iodine allergy test and drinking 500-800 ml water to fill the gastrointestinal tract. The intravenous injection of Ultravist (iodine concentration, 370 mg/mL; Bayer Healthcare Pharmaceutical, Germany; approval code, J20130157) was used for enhanced scan with a dose of about 80-100 ml. The arterial phase scanning was performed at 8 seconds after injection, and venous phase scanning was performed with an interval of 20-30 seconds after arterial phase scanning. The delayed phase scanning was performed with the same interval time to venous phase scanning, and subtle time adjustment might be done according to the patient's heart function in delayed phase scanning, which ensured the portal venous system got the best filling with contrast medium. Results of CT showed organs were relocated opposite to their normal anatomic position (a right-sided stomach, right-sided spleen, and inferior vena cava to the left of the aorta), and a 6.6 cm cystic-solid tumor in the head of the pancreas with a forward expansive growth pattern, in which the enhancement of solid part was lower than that of normal pancreatic parenchyma (Fig. 1).The CT images were collected in DICOM (Digital Imagingand Communications in Medicine) formatand then input to the Myrian XP 3D reconstruction software system (Intrasense, Montpellier, France) to make preoperative 3D visualizationreconstruction. The 3D model of the organs was reconstructed with automatic combination by extracting neighboring voxels with similar CT density, color matching, smoothing, and modification. The region of interest included liver, pancreas, pancreatic tumor, superior mesenteric vessels, splenic vessels, and other peripancreatic vessels. The 3D model was observed by rotating in different angles. Preoperative assessment of the anatomical relationship between tumor and surrounding vessels was performed to determine whether the tumor could be completely removed. In the present study, it clearly showed that the tumor was located in the pancreatic head and had a forward expansive growth pattern in the reconstruction model. No obvious malformationsother than the mirror image of the organs was observed. We rotated the 3D reconstruction model and observed the tumor from four angles (anterior, left, right and horizontal angle), which confirmed that there was no tumor invasion to the superior mesenteric vessels, splenic artery and vein, celiac trunk, abdominal aorta and inferior vena cava, and so on (Fig. 2). After we assessed that the tumor in the pancreatic head can be completely resected, we used the surgical simulation technique of the Myrian XP system to determine the pre tangent plane of the tumor. The superior mesenteric vein (SMV) was chosen as the reference to set the pancreatic tumor resection line and measured the length between pancreatic tumor resection line and SMV and we set the length to be 20.6mm, where the remnant pancreas could be preserved as much as possible under the premise of complete resection of the tumor (Fig. 3). The patient underwent PD performed with an end-to-side pancreaticojejunostomy, an end-to-side choledochojejunostomy, and an end-to-side gastrojejunos to my with a modified Child's procedure on May 23rd, 2018. The intraoperative findings were consistent with the results of the preoperative 3D reconstruction simulation and the tumor was completely resected (Fig. 4). The surgical duration was 380 minutes, with the estimated blood loss of 600 mL and infusion of 2 U red blood cell suspension. Postoperative pathology confirmed ductal adenocarcinoma of pancreatic head, with an invasion of the duodenal papilla and no lymph node metastasis. Immunohisto chemical detection showed CEA and p53 are weakly positive (Fig 4). The tumor staging was IIA with T3N0M0 based onthe 8th editions of the American Joint Committee on Cancer (AJCC) TNM staging system. The patient was discharged on postoperative day 16 without any serious complications. Follow-up has been 2 years and the patient is doing well with no recurrences. The present study was approved by the Ethics Review Committee of The First Affiliated Hospital of

Nanchang University (Nanchang, China). Informed written consent was obtained from the patient.

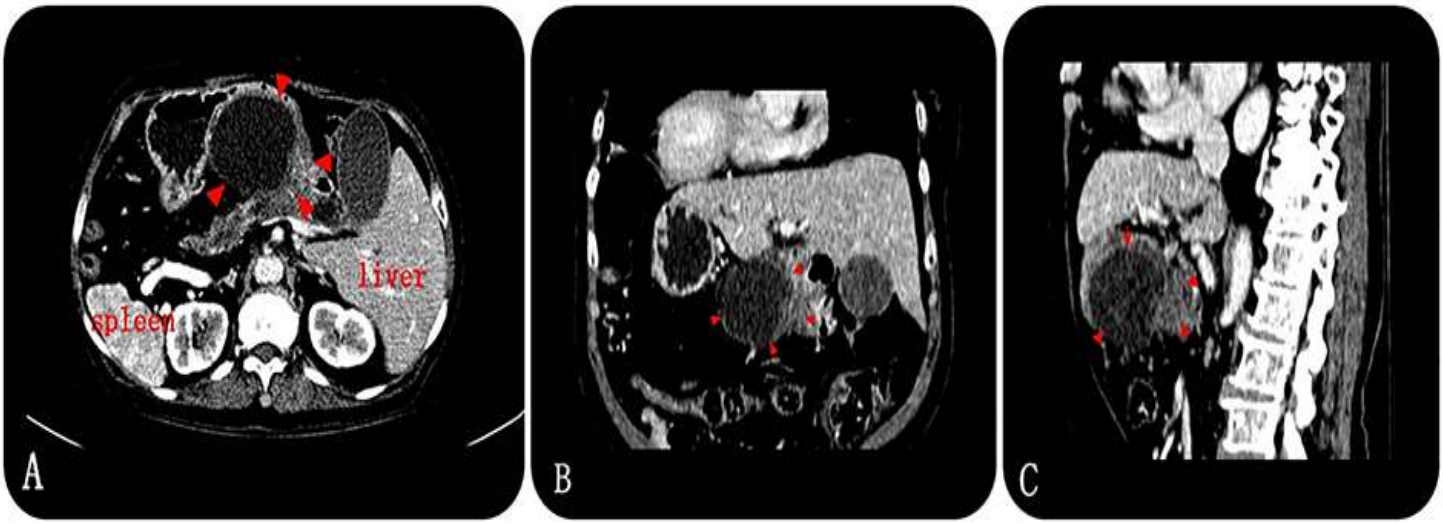


**Figure 1**. The contrast-enhanced CT showed a mass in the head of pancreas concurrent with situs inversus totalis.

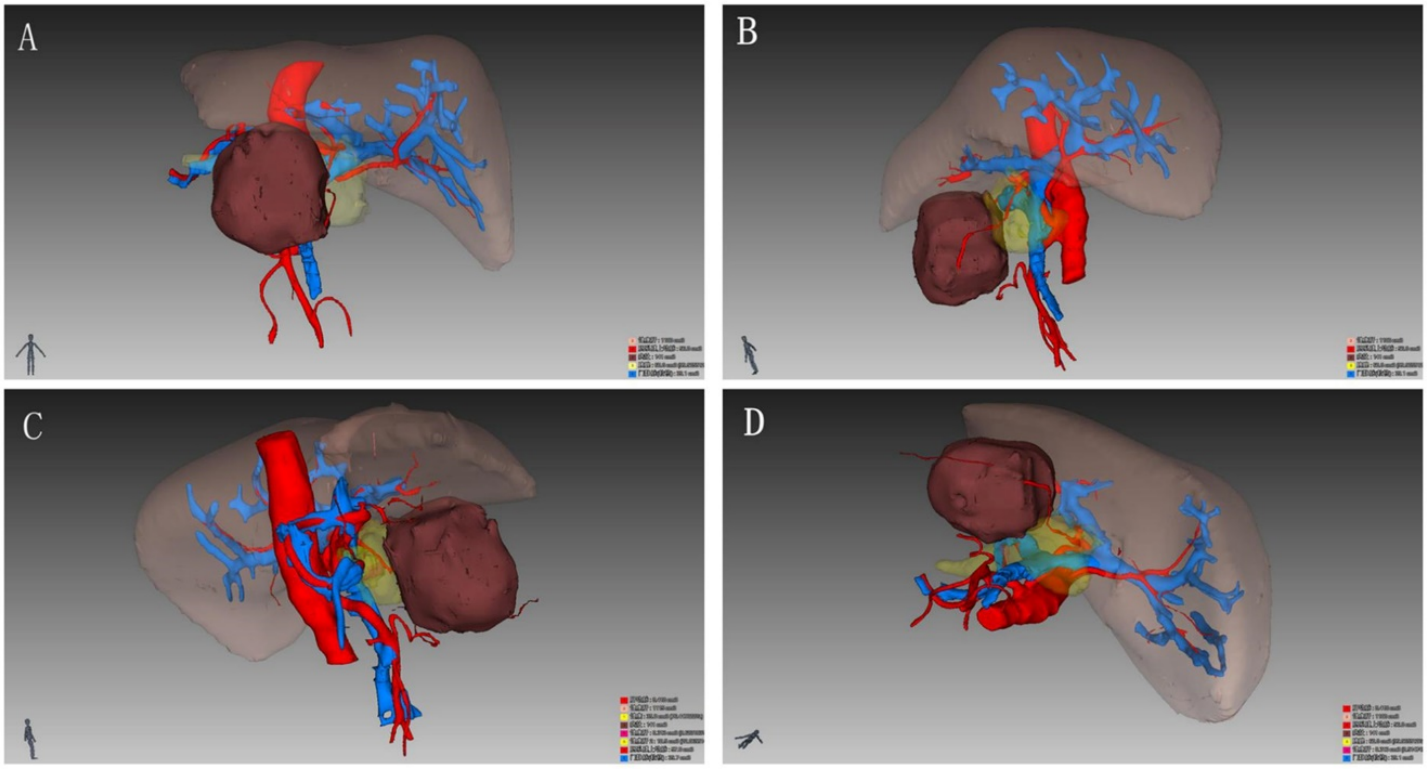


**Figure2**. The 3D reconstruction simulation system showed the relationship between tumor and peripheral vessels from different angles (A: anterior angle, B: left angle, C: right angle and D: horizontal angle).

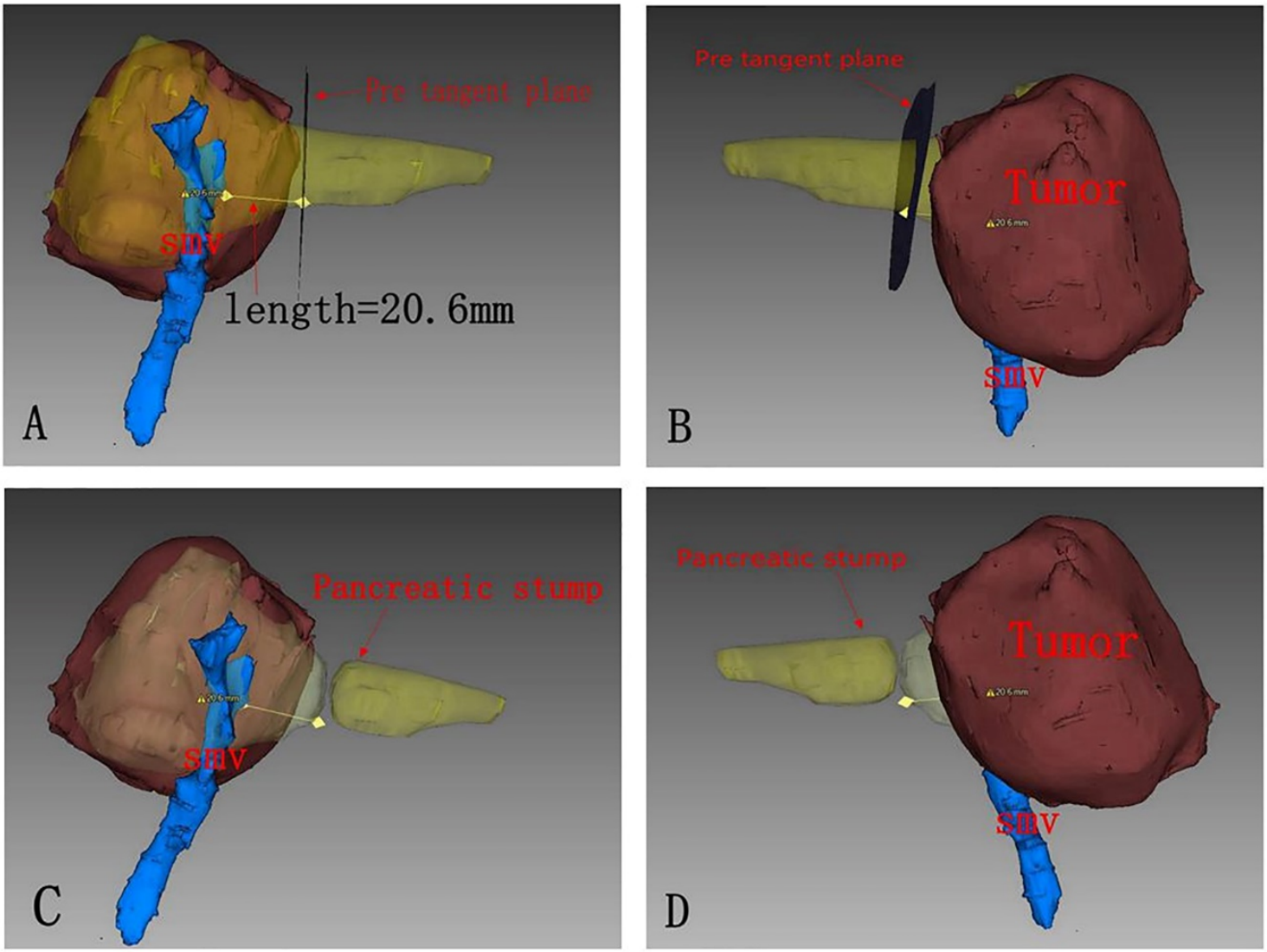


**Figure3**. The 3D reconstruction simulation system showed the pre tangent plane and the length between pancreatic tumor resection line and SMV (super mesenteric vein) from posterior and anterior angles (A and C are posterior angle, B and D are anterior angle).

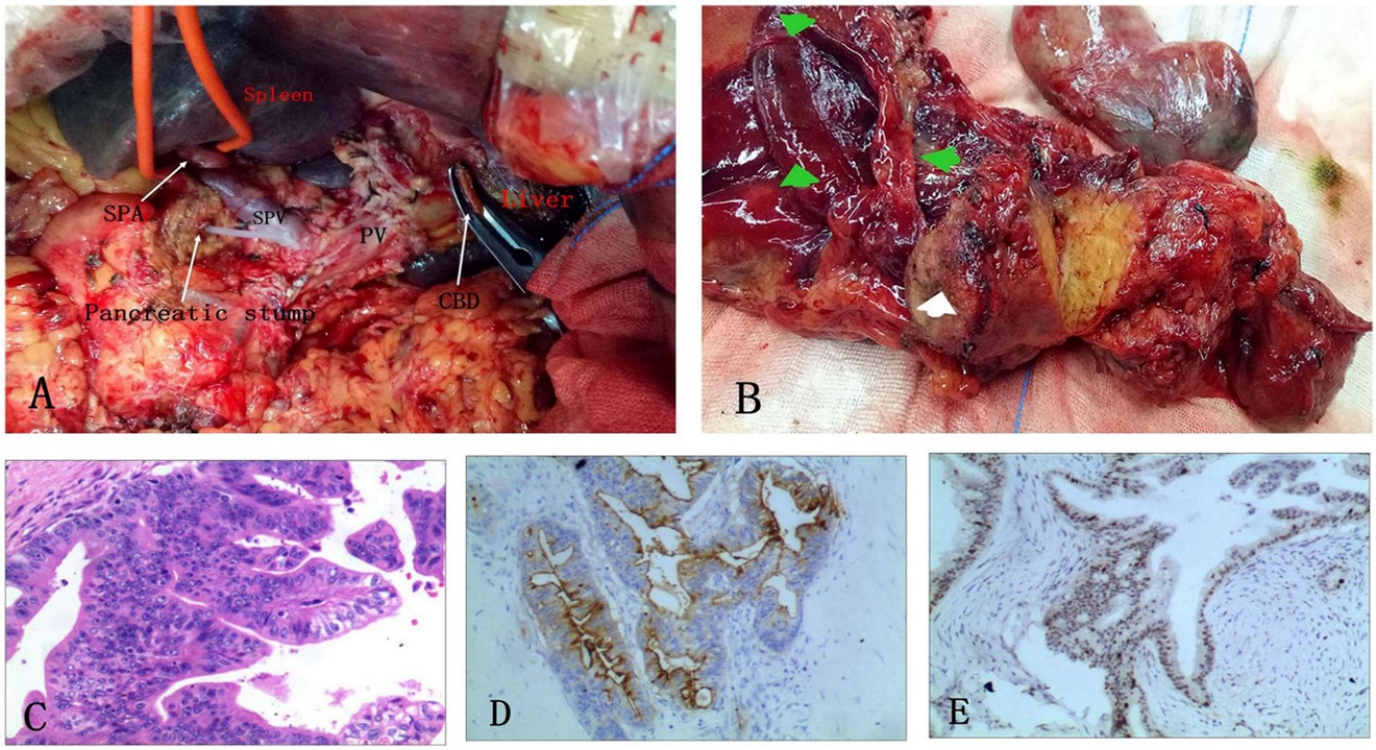


**Figure 4**. Intraoperative findings and postoperative pathological results. (A) Anatomy after tumor resection (PV, portal vein; CBD, common bile duct; SPV, splenic vein; SPA, splenic artery).(B) Tumor specimen: green arrows mark the cystic part of the tumor and white arrow marks the solid part of the tumor.(C) The pathology of tumor(H&E staining; magnification, x400). (Dand E)Immunohistochemical examination of tumor：specimens stained weakly positive for CEA (D) and P53 (E) (magnification, x100).

**Discussion**

The first case of SIT was reported by Fabricus in 1600 [2]. Although this anomaly does not affect normal health or longevity and often diagnosed incidentally during check-up examinations, SIT can be accompanied by cardiopulmonary malformations, digestive system abnormalities, and a variety of respiratory and urologic anomalies [12]. Typically, the risk of cardiovascular anomalies of patients with SIT is 10 times greater than normal individuals [13]. Moreover, approximately20-25% of individuals with SIT have concurrent chronicsinusitis and bronchiectasis, termed as Kartagener syndrome [6]. There was neither Kartagener syndrome nor obvious anatomic abnormalities except forSIT in our case. The exact etiology of SIT during embryonic development remains unclear. Some studies suggested that chromosomal abnormalities might lead to a reversal of right-left polarity during early embryonic development [14, 15]. However, other researches showed that the role of mechanical nodal fluid flow may be associated with left-right patterning [16, 17]. Furthermore, some studies suggested that the KIF3 complex, cell-adhesion factor N-cadherin and beta-catenin may be high-risk factors for tumors in subgroup of SIT, such as lung cancer or gastric cancer [18, 19], but pancreatic cancer concomitant with SIT was not reported before. The coexistence of SIT and pancreatic head cancer is extremely rare. To date, only seven cases of pancreatic head cancer with SIT have been reported, and the present report emphasizes the eighth case, as shown in Table 1.

| Author | Reference | Year | Age/sex | Malignancy | Operation | Journal |
|---|---|---|---|---|---|---|
| Sakaguchi | (3) | 1985 | 77/male | Pancreas head | PD | Nihon Geka Gakkai Zasshi |
| Quintini | (4) | 2003 | 56/male | Pancreas head | PD | Minerva Chir |
| Macafee | (5) | 2007 | 67/male | Pancreas head | PD | Eur J Surg Oncol |
| Sceusi | (6) | 2009 | 48/female | Pancreas head | PD | World J Surg Oncol |
| Maruyama | (7) | 2010 | 63/male | Pancreas head | PD | Surg Today |
| Zheng | (8) | 2013 | 47/female | Pancreas head | PD | Dig Liver Dis |
| Chen | (9) | 2015 | 56/male | Pancreas head | PD | ANZ J Surg |
| This case | - | 2020 | 69/female | Pancreas head | PD | - |

PD, pancreaticoduodenectomy.

**Table 1:** List of reported cases of pancreatic headcancer with situs inversus totalis

The PD is still the curative treatment for pancreatic head cancer since it was performed by Whipple in 1935[20]. With the development of surgical techniques and perioperative care, the mortality rate of PD has dropped to less than 5% in some centers [21, 22],however, the postoperative morbidity of PD remains as high as up to 50% [23] and thereby is considered to be the main factor for long postoperative hospital stay and high cost for PD. Due to its high complexity of the surgical operation and postoperative morbidity, PD for the pancreatic head cancer can be very challenging for surgeons, especially unusual surgical field of vision with mirror-image anatomy in patients with SIT. We summarized an application of 3D reconstruction simulation for preoperative assessment assisted PD for the pancreatic head cancer patient concurrent with SIT.

Several factors affect the complete resection of pancreatic head tumor during PD, including invasion of superior mesenteric vein or portal vein, invasion of abdominal aorta or inferior vena cava, or other important adjacent organs. Some studies suggested that 3D reconstruction is an optimal method for preoperative assessment to reveal the tumor, vascular variance and tumor-induced vascular changes, which can improve the surgical quality and reduce surgical risk [10, 11, 24]. Multi-slice spiral CT has also 3D reconstruction function, which can be used for showing the pancreatic head tumor and peripancreatic vessels. But 3D reconstruction of CT can not clearly show the relationship between tumor and its adjacent vessels, and not provide the 3D, intuitive, multi-angle observation when the vessels are squeezed, oppressed or infiltrated by tumor or partially overlapped by tumor and surrounding organs, or anatomical abnormalities. Therefore, the assessments by the reconstruction of CT on whether pancreatic tumor can be resected are often unsatisfactory. A meta-analysis showed that the sensitivity and specificity of multi-slice spiral CT in the evaluation of the pancreatic head tumor resectability were generally 68% and 89%, respectively, and the sensitivity for the evaluation of vessel involvement was 68% [25]. 3D reconstruction simulation system can reconstruct a model of tumor and its surrounding organs to reveal their exact relationship, which can improve the resection rate of tumor and reduce tissue injury.

As far as we know, there is still no literature reporting on the application of 3D reconstruction simulation in pancreatic head cancer concurrent with SIT. In the present study, we reprocessed the CT data of the pancreatic head cancer patient with SIT by using the 3D reconstruction simulation system (Myrian XP).The pancreatic head tumor, pancreatic tissue, and peripancreatic vessels are marked in different colors, and the relationship between tumor and adjacent vessels is observed by 3D graphics zooming, rotation and transparency, which overcomes the limits of CT image. After 3D reconstruction, it showed clearly the location of tumor and its relationship with the vessels and adjacent organs from different angles in the SIT patient, which facilitates the formulation and implementation of surgical treatment. In our study, we amputated the tumor according to the pancreatic tumor resection line and the length between pancreatic tumor resection line and SMV, which was preoperative set by 3D reconstruction simulation during operation. we successfully removed the tumor with no tumor residue in the pancreatic stump. Therefore, we suggested that the method for the pancreatic tumor resection line is convenient and helpful to improve tumor resection rate and worthy of promotion, but the optimal length through which we can

preserve normal pancreatic tissue as much as possible remains unknown.In conclusion, the assistance of preoperative assessment by 3D reconstruction of the imaging results is very important in facilitating the surgical treatment for SIT patient with pancreatic head cancer, which can improve the resection rate of tumor and reduce the risk of abdominal viscera damage, and should beroutinely performed when available.

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**Authors' contributions**

Peng Du, Xinjiang Zhang and Yong Liparticipated in the treatment of this patient and helped to draft the manuscript. Peng Du performed the data analyses and wrote the manuscript. Yong Li gave the final approval of the version to be submitted. All authors read and approved the final manuscript.

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**Competing interests**

There are no competing interests

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