Gastric cancer with situs inversus totalis: does it really create difficulties for surgeons?

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Abstract

Introduction: Situs inversus totalis (SIT) is a very rare condition that is seen at a rate of one in about 6000–8000 births. [1]. In this situation, the organs or organ systems are transposed to the opposite side of the body from their normal locations (mirror image of the normal), and it is most often detected during a radiological assessment [1–3]. The coexistence of SIT and gastric cancer in the literature is limited to a few case reports [4, 5]. In these cases, the research has shown that laparoscopic and open gastrectomies have been performed successfully. Moreover, the surgical procedure to be performed does not need to change, even though the anatomy of these patients is different.

Aim

Here we present the case of a patient diagnosed with gastric cancer and SIT, accompanied by a review of English studies on this subject.

Material and methods

Our study presents a case of gastric cancer with SIT and offers a discussion in light of the relevant literature. We searched for published studies of gastric cancer with SIT using different keyword combinations, including “situs inversus totalis and gastric cancer”, “situs inversus totalis and gastric malignant”, and “situs inversus totalis and gastric resection” in the PubMed, MEDLINE, and Google Scholar databases. Those studies published between January 1, 2000 and December 1, 2016 were reviewed, and any with full-text versions available and sufficient details on the patients were included in our study. Literature reviews and repeated reports were excluded from the study. The data recorded from the previous studies included the date of publication, age, sex, tumour location, existence of a vascular anomaly, surgical procedure performed, lymph node dissection, tumour/node/metastasis (TNM) staging, and postoperative complications.
Results

Case report

A 72-year-old female patient presented to our clinic with complaints of epigastric pain and burning. Her medical history showed no known comorbidities, and the abdominal and lymph node examinations did not reveal any problems. An oesophagogastroscopy revealed a lesion of about 1 × 1 cm in size with an ulcerated surface, located in the antrum close to the pylorus, with a slight tumorous appearance. Her biopsy results showed that it was an adenocarcinoma. A pulmonary radiograph showed dextrocardia (Figure 1), while the thoracoabdominal computerised tomography (CT) revealed SIT (Figures 2 A, B). There was no evidence of intra-abdominal acid, distant organ metastasis, peritoneal carcinomatosis, pathological lymph nodes, or vascular anomalies.

Endoscopic ultrasonography (EUS) was applied to the patient for endoscopic mucosal resection compliance. The EUS was evaluated as T1N0. An endoscopic mucosal resection was recommended for this patient, but a surgical procedure was planned since she and her family did not accept the recommendation. Subsequently, a distal subtotal gastrectomy and D1 lymph node dissection were performed via laparotomy. Billroth 2 gastroenterostomy anastomosis and Braun’s enteroenterostomy was performed for reconstruction. The operating time was 150 min and blood loss was 100 ml. The patient was then discharged on the eighth postoperative day without any problems. The pathological analysis of this patient revealed a tumorous lesion of approximately 1.5 × 1 cm, compatible with well differentiated (G1) adenocarcinoma, which had involvement up to the submucosa. None of the excised 17 lymph nodes exhibited metastasis (T1bN0M0). In light of these results, the patient was taken into the follow-up program without any adjuvant therapy.

Literature review

In total, 21 cases and 20 articles published in English between 2000 and 2016 were found through a comprehensive search of the PubMed, Google Scholar, and MEDLINE databases. Twelve (57.1%) of these patients were male, 9 (42.9%) were female, and their
mean age was 61.8 ±10.97 years old. The tumour localisations were in the antrum in 13 (61.9%) patients, corpus in 2 (9.5%) patients, esophago-gastric junction in 2 (9.5%) patients, corpus antrum junction in 2 (9.5%) patients, and cardia in 1 (4.8%) patient. In 1 (4.8%) of the patients the tumour localisation was not specified.

Although vascular anomalies were seen in 10 (47.6%) cases, they were not specified in 8 (38.1%) of them. In 1 out of the 3 (14.3%) patients with vascular anomalies the left hepatic artery exited from the superior mesenteric artery, in one the left gastric artery exited from the aorta, and in the final one the left gastric artery was double branching.

Nineteen of the patients received curative surgeries, with 11 of these having had laparoscopic resections. Two of the patients did not receive curative procedures; one of them received a palliative surgery, and no surgical procedure was performed on the other. Only 1 of these cases developed a mechanical obstruction. The demographic and clinical characteristics of the patients (20 studies, 21 patients) have been summarised in Table I [6–25].

Discussion

Situs inversus totalis is a congenital anomaly with an asymptomatic course, and it develops via the clockwise rotation of the embryonic midgut at 270°, instead of a counter-clockwise rotation at 270°. Thus, all of the thoracic and abdominal visceral organs are located symmetrically according to the midline in reverse. In other words, SIT is the mirror image of the normal [1]. Situs inversus can include both the thoracic and abdominal cavities (totalis), or only one cavity (partial). The term situs solitus, on the other hand, refers to the normal localisation of the bodily organs [1, 2].

The aetiology of situs inversus remains a controversial issue. It has been suggested that it is related to a genetic defect occurring during the second week of the embryonal period [2]. It can be accompanied by pathologies like bronchiectases (Kartagener syndrome), polysplenia, and genitourinary anomalies [3–5]. In addition, cancers of various organs have been seen in patients with situs inversus [26, 27]. Allen [28] first described a case of gastric carcinoma in a male patient with situs inversus in 1936; however, this condition has been limited to case reports in the literature [2, 17]. Generally, it has been suggested that there is no direct relationship between SIT and gastric cancer [15, 23].

In cases of SIT, the most significant preoperative stage, especially before performing laparoscopic procedures, is the careful and cautious assessment of anatomical variations using preoperative imaging methods [8, 16]. If possible, CT angiography is recommended for the detection of accompanying vascular anomalies [29]. In our case, the CT was helpful in unveiling the local and vascular structures.

For SIT patients, the treatment modality is no different than that for normal gastric cancer. The previous literature has shown that these patients most often received successful surgical treatments [6–24]. For example, open, assisted or total laparoscopic, and assisted robotic gastrectomy procedures have been performed successfully. Yamaguchi et al. [8] reported the first laparoscopy-assisted distal gastrectomy case in 2003. Following this case, 6 more cases of laparoscopy-assisted distal gastrectomies were reported. Min et al. [19] published the first case of a totally laparoscopic distal gastrectomy in 2013, followed by a 2015 study by Morimoto et al. [21] in which the authors presented the first totally laparoscopic total gastrectomy case. Moreover, Kim et al. [16] presented the first and only robot-assisted distal gastrectomy case in the literature in 2012.

Some of the cases in the literature also received additional organ resections alongside the gastric resections. Among these, it was reported that 2 patients had a splenectomy and cholecystectomy, 1 had a low anterior resection, and 1 had a cholecystectomy [6, 7, 14, 24].

Situs inversus totalis can pose a challenge during surgery because of the extraordinary anatomy of these patients. Some surgeons have recommended that the operator and assistant positions should be reversed, especially during laparoscopic surgeries [11, 13, 18]. However, there are other authors who have reported successful laparoscopic procedures in the normal positions [14]. In our case, the surgeon and assistant worked in their standard places, successfully completing the surgical procedure.

The structure of gastric cancer is complex. The lymph node status alone may not be sufficient to show the disease. Appropriate lymph node dissection should be performed in patients with all gastric cancers [30].

When the postoperative complications were investigated, it was seen that the rate of complications was very low, with a mechanical obstruction seen in only one case [15, 18].

Conclusions

The coexistence of SIT and gastric cancer is a very rare condition. Although it has been reported that surgeons might experience challenges because of the extraordinary anatomy of these patients, the cases presented in the literature were reported to have been treated successfully. A careful radiological assessment should be conducted preoperatively because there might be accompanying anomalies, especially vascular
Table I. General characteristics of 21 gastric cancer cases with situs inversus totalis in English literature

| Author                  | Year | Age | Gender | Tumour localisation | Vascular variation | Operation | Lymph node dissection | TNM Stage | Post-operation complication |
|-------------------------|------|-----|--------|--------------------|-------------------|-----------|-----------------------|-----------|----------------------------|
| Iwamura [6]             | 2001 | 71  | F      | EGJ + rectal cancer | ND                | TG + LAR   | ND                    | T1bN0M0   | 1A                         |
| Murakami [7]            | 2003 | 71  | F      | Artrium           | No                | TG + splenectomy + cholecystectomy | D2          | D1 + β                    |
| Yamanouchi [8]          | 2003 | 76  | M      | Antrum            | No                | LAR       | ND                    | ND        | No                         |
| Ifergan et al [9]       | 2005 | 50  | F      | Antrum            | ND                | DG        | ND                    | ND        | No                         |
| Benjelloun et al [10]   | 2008 | 70  | M      | Antrum            | No                | DG        | ND                    | ND        | No                         |
| Ono et al [11]          | 2010 | 53  | M      | Antrum            | No                | LADG      | ND                    | ND        | No                         |
| Jin [12]                | 2011 | 60  | F      | Antrum            | No                | PD        | ND                    | No        | No                         |
| Benjelloun et al [13]   | 2008 | 70  | M      | Antrum            | No                | LAR       | ND                    | ND        | No                         |
| Futawatari [14]         | 2010 | 81  | M      | Antrum            | No                | DG        | D2                    | T1N0M0    | 1A                         |
| Haruki [15]             | 2010 | 81  | M      | Antrum            | No                | DG        | ND                    | ND        | No                         |
| Kang [16]               | 2010 | 60  | M      | Antrum            | No                | LADG      | D2                    | T3N2M0    | 3A                         |
| Seo [17]                | 2011 | 60  | M      | Antrum            | No                | LADG      | D2                    | T1N0M0    | 1A                         |
| Kim [18]                | 2011 | 71  | F      | Antrum            | No                | PD        | ND                    | No        | No                         |
| Kim [19]                | 2011 | 47  | M      | Artrium-corpus    | No                | LADG      | D2                    | T1aN0M0   | 1A                         |
| Fujikawa et al [20]     | 2013 | 52  | M      | Antrum-corpus     | No                | LADG      | PD                    | D1 + β    | 4A                         |
| Min et al [21]          | 2013 | 60  | M      | Artrium           | No                | LADG      | D2                    | T1N0M0    | 1A                         |
| Yamashita et al [22]    | 2014 | 42  | M      | Artrium           | No                | LADG      | D2                    | T1N0M0    | 1A                         |
| Morimoto et al [23]     | 2015 | 58  | M      | EGJ               | No                | LADG      | D2                    | T1N0M0    | No                         |
| Zhu [24]                | 2015 | 66  | F      | Antrum            | No                | LADG      | PD                    | D2        | No                         |

F – female, M – male, EGJ – esophago-gastric junction, ND – not data, LGA – left gastric artery, LHA – left hepatic artery, TG – total gastrectomy, LAR – low anterior resection, LADG – laparoscopic-assisted distal gastrectomy, DG – distal gastrectomy, PD – pylorus-preserving gastrectomy. Positive staging of lymph nodes: T1smN0M0, T1N0M0, T1N1M0, T1N2M0, T2N0M0, T2N1M0, T2N2M0, T3N0M0, T3N1M0, T3N2M0, T3N3M0, T4aN0M0, T4aN1M0, T4aN2M0, T4aN3M0, T4aN4M0.
ones. Overall, laparoscopies and robotic surgeries can be performed for suitable patients at experienced centres, consistent with oncological principles.

Conflict of interest
The authors declare no conflict of interest.

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