CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 31 (2017) 43–46

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Thoracoscopic surgery in the prone position for esophageal cancer in patients with situs inversus totalis: A report of two cases

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ARTICLE INFO

Article history:
Received 7 September 2016
Received in revised form 4 January 2017
Accepted 4 January 2017
Available online 6 January 2017

Keywords:
Thoracoscopy
Video
Intestinal malrotation
Polysplenia
Laparoscopy

ABSTRACT

INTRODUCTION: Situs inversus totalis (SIT) is a rare congenital condition characterized by a complete transposition of thoracic and abdominal organs. Here, we present two successful cases of left thoracoscopic esophagectomy in the prone position for SIT-associated esophageal cancer.
PRESENTATION OF CASE: Our first case was of an 82-year-old man who underwent a left thoracoscopic esophagectomy in the prone position, followed by hand-assisted laparoscopic gastric mobilization. Surgical duration and blood loss were 661 min and 165 g, respectively. His postoperative course was uneventful. The second case was of a 66-year-old man who underwent a left thoracoscopic esophagectomy in the prone position, followed by gastric mobilization via laparotomy owing to a concomitant intestinal malrotation and polysplenia. Surgical duration and blood loss were 637 min and 220 g, respectively. We trained for the surgical procedures preoperatively using left-inverted and right-inverted thoracoscopic surgical videos of patients with normal anatomy.
DISCUSSION: Surgical procedures in SIT patients are challenging owing to their mirrored anatomy. Recognition of their variations is thus important to avoid intraoperative accidental injuries. Left-inverted and right-inverted thoracoscopic surgical videos of patients with normal anatomy were found to be useful for image training prior to the actual surgery.
CONCLUSION: Thoracoscopic surgical treatment for esophageal cancer associated with SIT in the prone position can be performed safely, similar to the manner performed for thoracoscopic surgery in the right decubitus position, or surgery via an open thoracotomy. Gastric mobilization via laparotomy should be considered in patients associated other anatomic variations.

1. Introduction

Situs inversus totalis (SIT) is a rare disorder characterized by the transposition of the thoracic and abdominal organs to the opposite side of the body, in a mirror image of the normal anatomy. The incidence of SIT is approximately 0.005%–0.01% of all live births [1]. Its occurrence makes surgical procedures more challenging owing to the mirror image positioning of organs [1]. There have been few reports of esophagectomy for esophageal cancer associated with SIT in the left decubitus position [1–4]. However, there has only been a single case report of this disease treated with thoracoscopic esophagectomy, with the patient in the prone position [5]. Here, we present two successful cases of left thoracoscopic esophagectomy in the prone position for esophageal cancer associated with SIT. We also discuss the ideal surgical positioning for thoracoscopic esophagectomy and image training utilizing left-inverted and right-inverted videos of thoracic surgery performed on patients with normal organ positions.

2. Case presentation

2.1. Case 1

An 82-year-old male patient presented to our hospital following a diagnosis of esophageal cancer during a cancer screening. An upper gastrointestinal endoscopy revealed a type 2 tumor, and an esophagogram showed a tumor measuring 1.5 cm in diameter in the upper thoracic esophagus. A chest X-ray revealed dextrocardia and a right aortic arch (Fig. 1A). Computed tomography revealed a thick wall around the upper thoracic esophagus. In addition, the thoracic and abdominal visceral positions were mirror images of the normal (Fig. 1B, C). The patient was clinically diagnosed with T2N1M0 stage IIB (Union for International Cancer Control [UICC], 7th edition) squamous cell carcinoma of the esophagus. For the administration of general anesthesia, patient was intubated with

Abbreviations: UICC, Union for International Cancer Control; SIT, situs inversus totalis.

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http://dx.doi.org/10.1016/j.ijscr.2017.01.009

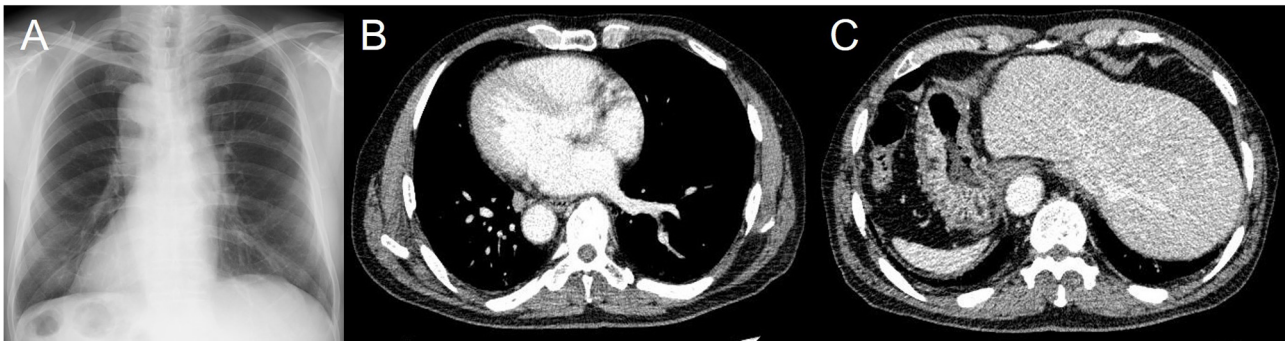

Fig. 1. (A) Chest X-ray showing dextrocardia and a right aortic arch. (B) Computed tomography images of the chest showing that the thoracic position of the organs is a mirror image of the normal position. (C) Abdominal computed tomography showing that the abdominal position of the organs is a mirror image of the normal position.

a single lumen endotracheal tube. The surgeons stood on the left side and a video monitor was placed to the right of the patient in the prone position. Placement of trocars was the same as our standard procedure because it is required to approach both the upper and the lower mediastinum easily. We performed a thoracoscopic subtotal esophagectomy with radical lymph node dissection using five ports via the left thoracic cavity, with the patient in the prone position under artificial pneumothorax (Fig. 2A), followed by hand-assisted laparoscopic gastric mobilization in the supine position. While processing around the gastrosplenic ligament, we performed the operation by inserting the right hand into the peritoneal cavity and using the main instruments with the left hand, unlike the standard procedure. The forceps of the assistant was inserted into the abdominal cavity from the left side of the abdomen. The assistant's forceps retracted the colon downwards, similar to the standard procedure. The surgical duration and intraoperative blood loss were 661 min and 165 g, respectively; in total, 49 lymph nodes were resected. The surgeons underwent image training prior to the surgery using left-inverted and right-inverted thoracoscopic surgical videos of patients with normal anatomy, to recognize the inverted anatomy of this condition (Fig. 2B). The definitive diagnosis was of a pT1bN0M0 p-stage IA (UICC, 7th edition) basaloid squamous cell carcinoma of the esophagus. Postoperative upper gastrointestinal endoscopy showed no leakage. Oral intake resumed on postoperative day 7, and the patient was discharged on postoperative day 34.

2.2. Case 2

A 66-year-old man presented to our hospital for the treatment of esophageal cancer. He had undergone endoscopic hemostasis for a bleeding gastric ulcer, and an esophageal type 2 tumor was detected in the middle thoracic esophagus during a follow-up endoscopy. A chest X-ray revealed dextrocardia and a right aortic arch. Computed tomography revealed a thick wall around the middle thoracic esophagus and multiple splenic nodules on the right side of his abdominal cavity (Fig. 3A). His duodenum was devoid of a horizontal portion. A larger part of the subsequent colon was not fixed in the retroperitoneum and was present on the right side of the abdominal cavity. These findings of computed tomography suggested an intestinal malrotation (Fig. 3B). He was clinically diagnosed with T2N1M0 stage IIB (UICC, 7th edition) squamous cell carcinoma of the esophagus. For general anesthesia, the patient was intubated with a single lumen endotracheal tube. We performed a thoracoscopic subtotal esophagectomy with radical lymph node dissection using five ports via the left thoracic cavity, with the patient in the prone position. A laparoscopic observation of the abdominal cavity identified the mirror image of his abdominal organs and intestinal malrotation. Gastric mobilization was performed using open laparotomy with the patient in the supine position. The surgical duration was 637 min, and intraoperative blood loss was 220 g. The definitive diagnosis was of a pT1bN0M0 p-stage IA (UICC, 7th edition) basaloid cell carcinoma of the esophagus. Postoperative upper gastrointestinal endoscopy showed no leakage. Oral intake resumed on postoperative day 7, and the patient was discharged on postoperative day 17.

3. Discussion

SIT is a rare disorder characterized by a complete right–left transposition of the thoracic and abdominal organs compared with their normal positions [6,7]. There are only a few reports discussing esophagectomy in patients with SIT, owing to the low incidence of esophageal cancer in patients with this disorder; SIT itself has no pathophysiological significance [1]. However, approximately 20–25% of cases are associated with Kartagener's syndrome caused by ciliary dyskinesia [8]. Successful thoracoscopic esophagectomy has been reported in patients with SIT accompa-

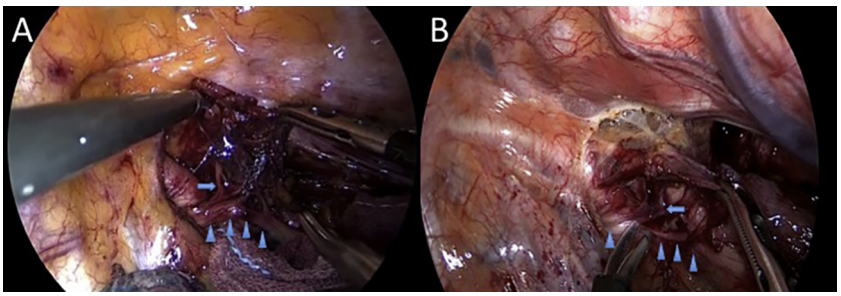

Fig. 2. (A) Thoracoscopic operative view of the patient showing that the thoracic organs are in mirror image positioning. The arrow shows the left-sided right recurrent laryngeal nerve; the arrowheads show the left-sided right vagus nerve. (B) Right-inverted and left-inverted view of a previous surgery in an anonymous patient with normal organ positioning for preoperative image training. The arrow shows the right recurrent laryngeal nerve, and the arrowhead shows the right vagus nerve.

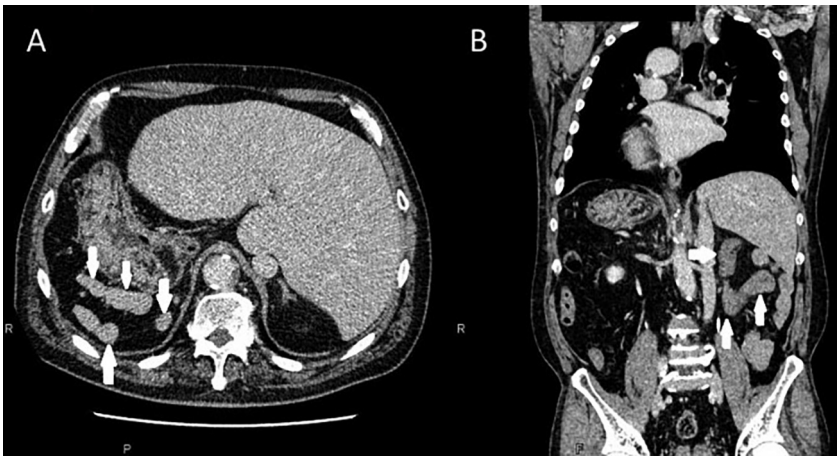

Fig. 3. (A) Abdominal computed tomography reveals that the abdominal position of the organs is a mirror image of the normal, with presence of multiple splenic nodules on the right side of the abdominal cavity. Arrows show multiple splenic nodules. (B) Findings of a computed tomography image suggest the occurrence of intestinal malrotation. The duodenum lacks a horizontal portion (arrows). A larger part of the subsequent colon was not fixed in the retroperitoneum and was present on the right side of the abdominal cavity.

nied with Kartagener's syndrome [2]. It is very rare for SIT and polysplenia syndrome to occur in the same patient [9].

Our first patient had an unremarkable medical history and possessed an acceptable performance status to undergo esophageal surgery, even at his age. Our second patient had multiple splenic nodules on the right side of his abdominal cavity and intestinal malrotation. Polysplenia, a recognized situs ambiguous, should be considered in cases with multiple splenic nodules on the right side of the abdominal cavity. This syndrome is associated with heterotaxia syndrome, which is defined as the occurrence of multiple splenic nodules and described as a left isomerism, or referring to the duplication of left sided structures [10–13]. It has been reported that 50–90% of polysplenia syndrome cases are associated with cardiac complications [11]. Some degree of intestinal malrotation is also seen in approximately 70% of the patients with this syndrome, although most cases are asymptomatic [14]. Midgut volvulus associated with intestinal malrotation was previously reported in an adult patient with SIT [15]. Since the computed tomography image in our second case displayed no cardiac malformation or bilateral bilobed lungs, we diagnosed the patient with intestinal malrotation and polysplenia associated with SIT. Recognition of SIT and its variation is important in order to avoid accidental injuries intraoperatively. Gastric mobilization via laparotomy should be considered in SIT patients associated with other anatomical variations.

A PubMed search using the terms "esophagus" and "situs inversus totalis" from 1976 to June 2016 revealed only 4 cases of thoracoscopic esophagectomy via the left thoracic cavity in the right decubitus position for SIT patients [1–4]. Average blood loss and surgical duration of these cases were 517 min and 260 g, respectively [4]. Esophagectomy in the prone position may improve patient outcome as a less invasive surgical technique, with fewer complications (e.g., pneumonia) and less intraoperative blood loss [16,17]. Compared with the lateral position, placement of the patient in the prone position improves visualization and ergonomics during surgeries; may improve the quality of mobilization and lymphadenectomy; and appears to contribute to an enhanced learning curve [18]. These potential advantages of thoracoscopic surgery in the prone position may be maintained in patients with SIT. In our cases, the surgical duration was almost similar and the amount of bleeding was minimal compared to thoracoscopic surgery in the right decubitus position.

Owing to the mirror-image positioning of thoracic and abdominal organs, surgical procedures in patients with SIT are often considered more challenging [1,3]. While operating on patients with SIT, it is characteristic for a surgeon's right hand to be positioned in the caudal side of the patients in the prone position, which is the opposite of patients with normal anatomy. Most surgeons lack the surgical experience required for SIT. It is important to recognize the complex anatomy in these cases, so that surgical planning can be improved [12]. The surgeons in this study performed esophagectomy in patients with SIT for the first time. Although we have encountered several similar cases before, the surgeons in this study have not been able to perform in the previous cases. In rare cases, such as those of patients with SIT, the selection of easier and more practical surgical methods and utilization of prior image training with right-inverted and left-inverted videos will be helpful to ensure surgical safety. This work has been reported in line with the SCARE criteria [19].

4. Conclusion

Thoracoscopic surgical treatment for esophageal cancer associated with SIT in the prone position can be performed safely, similar to the manner performed for thoracoscopic surgery in the right decubitus position, or surgery via an open thoracothomy. Left-inverted and right-inverted thoracoscopic surgical videos of patients with normal anatomy were useful as preoperative image training.

Conflicts of interest

None.

Consent for publication

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

Funding

The authors declare no supportive funding.

Ethical approval

This work does not require a deliberation by the ethics committee.

Authors' contributions

Nakano T is the first author of this article; Nakano T, Kamei T, Sakurai T, Taniyama Y, Sato C, and Ohuchi N were attending doctors and performed clinical treatment including surgical operation; Onodera Y and Ujiie N constructed the imaging and prepared the figures; all authors have read and approved the final manuscript.

Guarantor

Toru Nakano and Noriaki Ohuchi are accept full responsibility for the work and had controlled the decision to publish.

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