Case Report

Determination of the best surgical strategy for thoracic esophageal resection concurrent with double aortic arch using a three-dimensional model: a case report

Kenji Hamada, Yusuke Taniyama, Yoshiyuki Yashima, Yohei Ozawa, Toshiaki Fukutomi, Hiroshi Okamoto, Chiaki Sato, Michiaki Unno, Takashi Kamei

Department of surgery, Tohoku University Hospital, Sendai, Japan

Correspondence to: Yusuke Taniyama. Department of surgery, Tohoku University Hospital, Sendai, Japan. Email: hama.k-base-0531@surg.med.tohoku.ac.jp.

Background: A double aortic arch (DAA) is a relatively rare vascular malformation, which rarely causes problems once the patients reach adulthood. However, a DAA makes an esophageal cancer surgery difficult to perform, especially during upper mediastinal dissection. Herein, we report a strategy for surgery in esophageal cancer patients concurrent with DAA.

Case Description: A 73-year-old man was diagnosed with middle thoracic esophageal cancer of cT3N4M0 stage III (UICC-TNM 7th) concurrent with DAA. After two courses of neoadjuvant chemotherapy, surgical intervention was planned. To develop a surgical strategy for an esophagectomy with this complicated malformation, we created a three-dimensional printer model for this case. According to this simulation, the bilateral thoracoscopic approach with prone position seemed to be an ideal method for upper mediastinal dissection. As we expected, the dissection of upper mediastinum was difficult only with the right-side approach; especially, the oral side of esophagus posterior to the right aortic arch (RAA) was impossible to dissect from the right side. By switching the approach from left side, oral esophagus was easily dissected by retracting the oral esophagus from the cranial side of the left aortic arch (LAA). Surgery was successfully performed, and the patient was discharged 26 days after surgery without major complications.

Conclusions: To the best of our knowledge, this is the first surgical report using a three-dimensional printer for esophageal cancer. The bilateral approach is appropriate for esophageal cancer surgery concurrent with a DAA. A three-dimensional printer is useful for simulating esophageal surgery with major vascular malformations.

Keywords: Case report; three-dimensional printing; double aortic arch (DAA); vascular malformation; esophageal cancer

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Introduction

A double aortic arch (DAA) is classified as a Stewart type I vascular malformation (1), which is a very rare congenital vascular malformation that occurs in approximately 0.1% of the adult population (2). DAA forms a complete vascular ring around the trachea and esophagus, which are then compressed. Major symptoms include noisy breathing, recurrent upper respiratory infection, dyspnea, and dysphagia during infancy and childhood (3). However, some cases are asymptomatic throughout their lives, and are detected incidentally in adulthood, as seen in the present case.

Cancer is the most common cause of death globally, with esophageal cancer being the seventh most diagnosed malignancy (4). Surgical resection plays a major role in treating esophageal cancer. In thoracic cancer, mediastinal lymph node dissection plays an essential role in curative surgery. However, in cases where thoracic cancer is concurrent with DAA, dissection of the upper mediastinum is difficult because aortic arches create a ring that surrounds

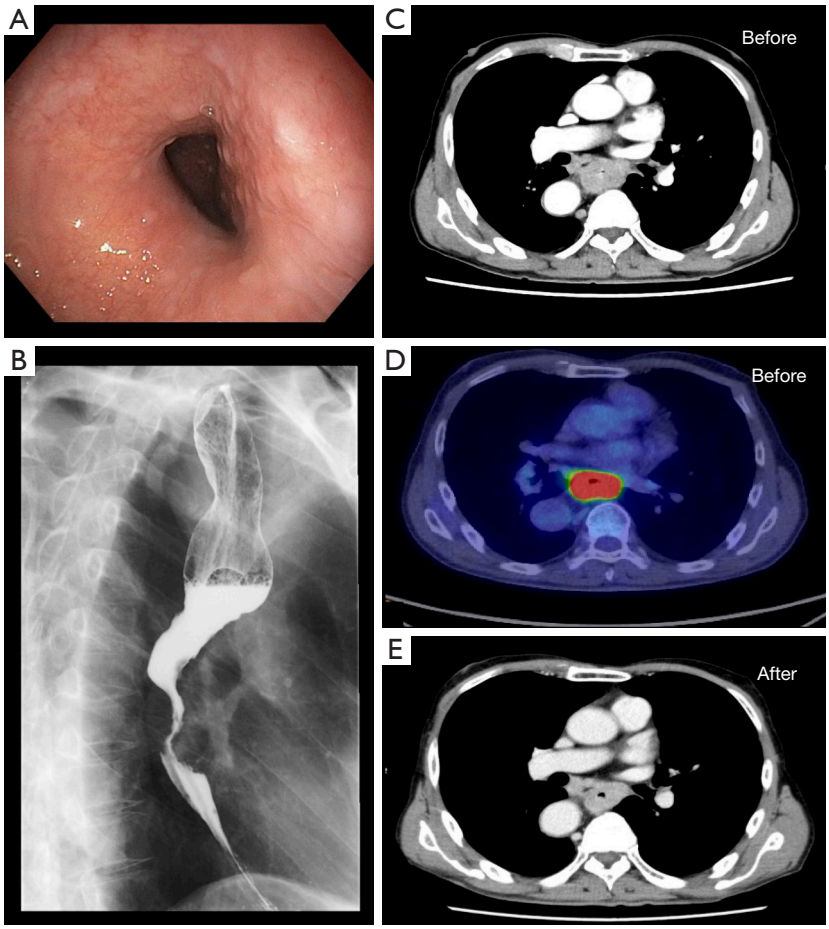

Figure 1 Preoperative Image. (A) Endoscopic findings show a type II tumor surrounding the wall in the middle thoracic esophagus; (B) barium esophagography show the narrowing of the middle and lower mediastinum; (C-E) enhanced computed tomography (CT) and positron emission tomography CT-showing esophageal cancer before neoadjuvant chemotherapy and then shrinking of the esophageal cancer lesion after chemotherapy.

the esophagus and regional lymph nodes.

Three-dimensional (3D) printing has rapidly evolved over the last few years. Although it has been successfully used in the field of maxillofacial surgery (5), limited cases have been reported in gastroenterology (6). Herein, we report our experience with simulation using a 3D printed model, that was useful in a case of esophageal cancer surgery concurrent with DAA. We present the following case in accordance with the CARE reporting checklist (available at https://tcr.amegroups.com/article/view/10.21037/tcr-22-468/rc).

Case presentation

A 73-year-old man presented with chief complaint of dysphagia. Endoscopic examination revealed a type II tumor located 25 cm from the incisors. Squamous cell carcinoma was confirmed by histopathologic examination of a biopsy specimen. Laboratory examination showed no abnormalities excepting mild elevation of tumor markers. Chest radiography demonstrated widening of the upper mediastinal silhouette. Enhanced computed tomography (CT) showed thickening of the walls in the middle thoracic esophagus and some swollen lymph nodes in the upper thoracic paraesophageal lymph nodes, recurrent laryngeal lymph nodes, and dorsal lymph nodes around the thoracic aorta (*Figure 1*). DAA was also detected on the enhanced CT images (*Figure 2*), which showed encirclement of both the trachea and thoracic esophagus by the right and left aortic arches (LAAs). The right aortic arch (RAA) was

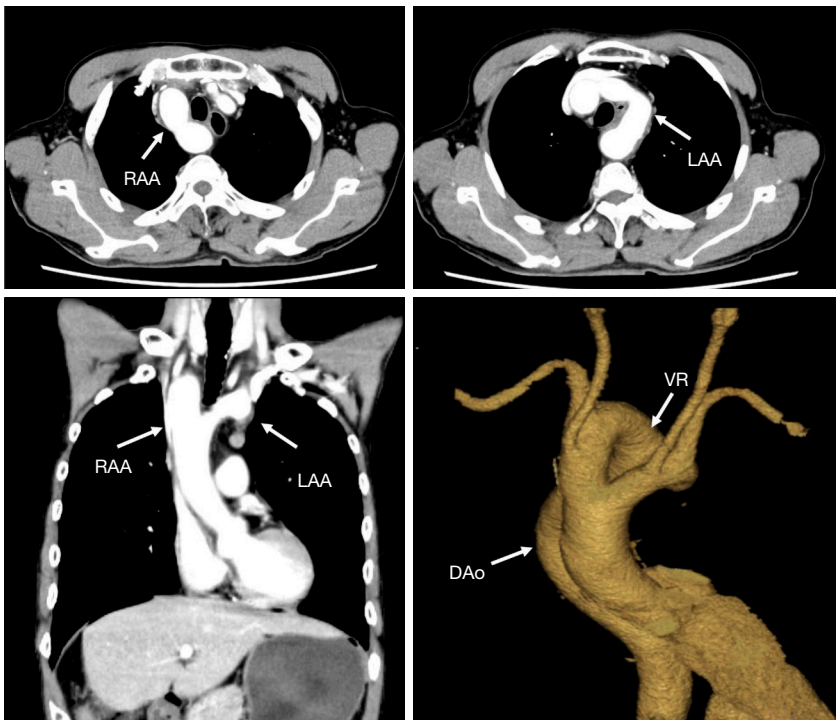

Figure 2 Preoperative CT shows the RAA and the LAA. Three-dimensional CT shows the double arch of aorta, VR, and DAo. CT, computed tomography; RAA, right aortic arch; LAA, left aortic arch; VR, vascular ring; DAo, descending aorta.

more dominant than the LAA, and the descending aorta was located on the right side of the mediastinum (*Figure 2*). Positron emission tomography CT (PET-CT) revealed accumulation of lymph nodes, suggestive of metastatic lymph nodes. No distant metastases were detected on PET-CT. Hence, the patient was diagnosed with middle thoracic esophageal cancer cT3N4M0 Stage III (UICC-TNM 7th) and DAA.

The patient underwent two courses of neoadjuvant chemotherapy using fluorouracil (5FU) and cisplatin; however, no obvious treatment effect was observed in either the tumor or metastatic lymph nodes.

3D printed model for intraoperative simulations

To aid in the development of a surgical strategy for an esophagectomy with this complicated malformation, a 3D printer model (Rais 3D N2 Plus) was created for this case (*Figure 3A*,*3B*). An AB resin was used to create the model. The creation time for the blood vessels, trachea, and heart was approximately 37 h, and that for the ribs and vertebrae was approximately 46 h. To enable recognition of each individual organ, the originally white model was colored manually. The esophagus was excluded from this 3D printed model. Tape was added after the model fabrication was completed, which made it easier to understand the anatomical location and relation of each organ—both before and after the dissection of the esophagus. Ribs and vertebrae were added to the model to simulate the thoracoscopic approach. This simulation led us to conclude that the bilateral approach was the best method for such a case (*Figure 3C*,*3D*). Additionally, the ideal port placement was determined in this simulation.

Intraoperative findings

The patient was placed in a prone position with both hands raised, and the operation was initiated via right thoracoscopy. The descending aorta could be detected at the right side of the vertebrae, with the RAA found at the cranial side compared to the level of the usual aortic arch. Esophageal dissection was started from the middle and inferior mediastinum (*Figure 4A*). The arch of the azygos vein was cut and divided for a better surgical view. In the upper mediastinum, the right recurrent laryngeal nerve could be detected from the RAA to the right side of the

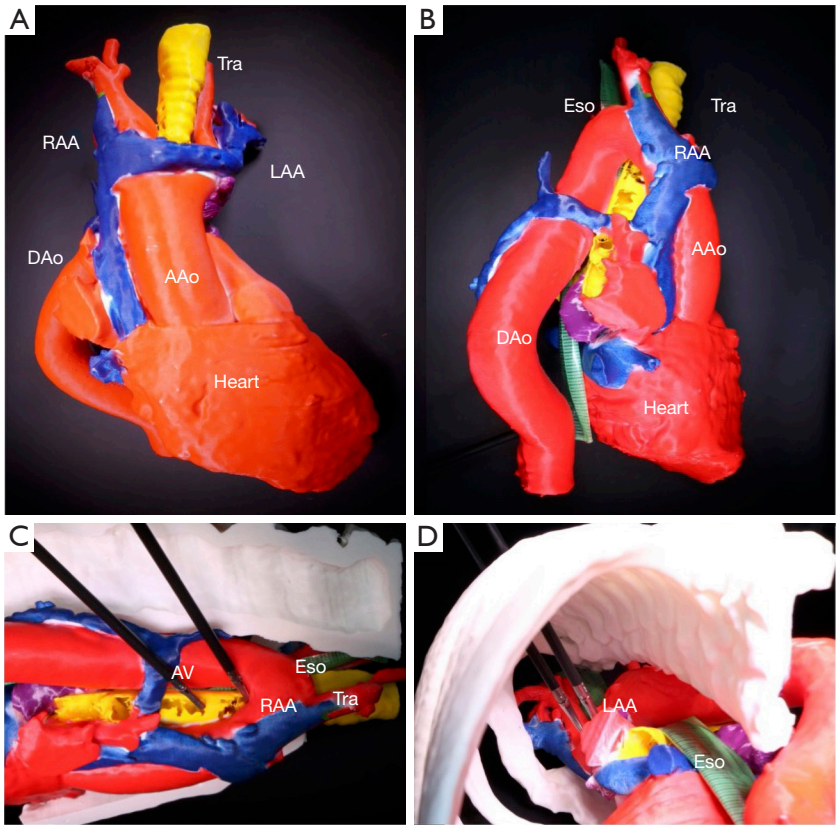

Figure 3 Three-dimensional (3D) printing of the model. (A,B) View of the 3D printer from the front and right sides; (C) simulation of a right thoracoscopic approach. This angle seemed to be appropriate for the approach inside the ring formed by the DAA; (D) simulation of the left thoracoscope. This angle seems to be appropriate for the dissection of the cephalad side of LAA. DAA, double aortic arch; RAA, right aortic arch; LAA, left aortic arch; AAo, ascending aorta; DAo, descending aorta; Tra, trachea; Eso, esophagus; AV, azygos vein.

trachea. After dividing the esophagus, dissection of the right recurrent laryngeal nerve lymph node was performed. The esophagus was detached as much as possible from the trachea and aortic ring (*Figure 4B*). The approach was switched to the left for dissection of the upper mediastinum, which was hidden by the RAA from the right thoracic view. After incision of the left mediastinal pleura, the LAA was exposed, and the oral side of the esophagus was detected at the caudal side of the aortic ring. The oral esophagus was retracted from the cranial side of the LAA. The esophagus was dissected towards the oral side till the levels of previous dissections. Additionally, since the A #106tbL lymph node invaded the left vagus nerve, the lymph nodes were dissected from the bifurcation of the left vagus and recurrent laryngeal nerve (*Figure 4C*). The left recurrent nerve was located at the LAA, as usual (*Figure 4D*).

The abdominal procedure was started after placing the patient in supine position. Dissection of the celiac and para-stomach lymph nodes was performed, followed by creation of a gastric conduit. The retrosternal route was selected for reconstruction and anastomosed using the hand-sewn procedure.

The patient experienced temporary left recurrent laryngeal nerve palsy and minor leakage in the anastomosis; however, the condition resolved with conservative treatment. The patient was discharged 26 days after surgery.

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). 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 editorial office of this journal.

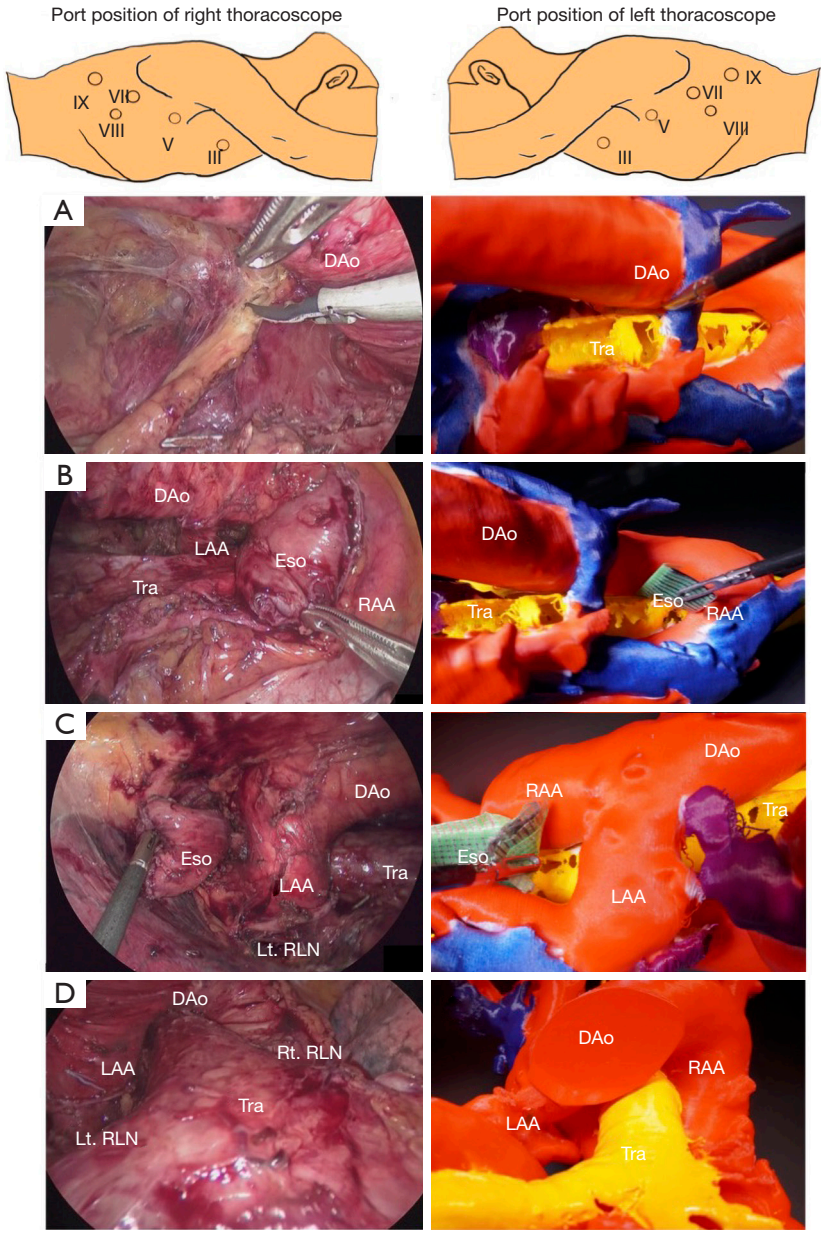

Figure 4 These photos are the intraoperative finding and the corresponding images of the 3D model. (A) The middle and lower mediastinum from the right thoracoscopic approach; (B) the upper mediastinum from the right thoracoscopic approach. The approach angle inside the ring formed by DAA is appropriate; (C) the upper mediastinum from the left thoracoscopic approach. The oral side of the esophagus is pulled out from the cephalad side of the LAA; (D) the ring formed by the DAA seen from dorsal of trachea in the left thoracoscopic approach. DAA, double aortic arch; RAA, right aortic arch; LAA, left aortic arch; DAo, descending aorta; Tra, trachea; Eso, esophagus; Lt. RLN, left recurrent laryngeal nerve; Rt. RLN, right recurrent laryngeal nerve.

Discussion

Although DAA is an anatomically complex vascular malformation, it does not affect the patient's life if the aortic ring does not cause stenosis of the trachea or esophagus. However, when a patient with this malformation develops esophageal cancer, most surgeons would be distressed concerning the best surgical strategy. To the best of our

knowledge, there are seven surgical case reports of thoracic esophageal cancer patients with DAA (7-13). Four case reports describe the use of left thoracic approach because of a right-sided descending aorta (7,8,10,11). Three case reports describe the use of right thoracic approach (9,12,13). Although 3D-CT was performed in most cases, no cases reported the use of bilateral thoracoscopic approach.

The 3D model allowed a better understanding of the positioning of the blood vessels, trachea, and heart preoperatively, thus increasing the surgeons' confidence (*Figure 3A*,*3B*). The bilateral thoracoscopic approach was determined to be a safe way to operate in this case via preoperative simulation using a 3D printed model (*Figure 3C*,*3D*). Trans-hiatal approach might be one of the choices. However, LN dissection below the aortic arch was deemed necessary in the present case, and this procedure is difficult to perform by trans-hiatal approach. In addition, trans-cervical approach using mediastinal scope would also be one of the choices for the case with normal anatomy. But it was not suitable for this case because the space composed by DAA was too narrow to perform a surgery. Therefore, we could safely perform three-field lymph nodes dissection for an esophageal cancer patient with DAA. In the field of gastroenterology, 3D printing has been used in limited cases, generally concerning the liver (6). To the best of our knowledge, this is the first surgical report making use of a 3D printed model for esophageal cancer management. Thus, 3D printing might be useful in surgery of the gastrointestinal tract with vascular malformations, such as DAA.

Preoperative simulations suggested that performing a dissection through a left side approach was easier in the middle and lower mediastinum. However, the right thoracoscopic approach was selected for the present case because it is the standard technique followed by us, and was deemed necessary for the upper mediastinum.

In case the right-side approach was found to be difficult, it was planned to continue the dissections around the concerned area from the left.

However, the 3D simulation indicated that most of the procedure could be successfully performed by the right side approach. The space between the descending aorta and the trachea seemed to be narrow due to the right descending aorta at the right thoracoscopic approach site. However, this issue was not considered to be a major problem since the narrow mediastinum also occurs in normal esophagectomy, which can be addressed by opening the mediastinum space by pressing either the bronchus or heart. Although, exposure of the esophagus was slightly more difficult than was expected due to the protruding descending aorta, which made it arduous for the thoracoscope to obtain a good surgical view (*Figure 4A*). Most of the dissection was possible with the selected approach. The remaining dissections were completed using the left-side approach with ease.

In the upper mediastinum, the RAA was found to be located cephalad compared to the LAA location indicated by the 3D printed model. Therefore, the right thoracic approach angle was considered appropriate for the ring formed by DAA (*Figure 3C*). However, for dissection of more cephalad structures than DAA, the right thoracic approach becomes strenuous. This is because the RAA is positioned in front of the upper esophagus and regional lymph node. Thus, the left thoracic approach was selected for further neck dissection. Intraoperatively, the approach angle inside the ring formed by DAA was easy using the right thoracoscopic approach (*Figure 4B*). However, dissection of the tissue cephalad from the DAA was difficult from the right side, as expected from our simulation. By switching to a left thoracoscopic approach, the oral side of the esophagus was pulled out from the cephalad side of the LAA, and the remaining dissection toward the neck could be performed with ease (*Figure 4C*).

The bilateral thoracoscopic approach may also be useful for preserving the recurrent laryngeal nerve in advanced cancers with DAA, as seen in the present case. Intraoperatively, the right recurrent laryngeal nerve recurred at the RAA, which might be difficult to visualize using only the left-side approach because of the existence of the LAA. In fact, none of the previous cases that underwent surgery from the left side could identify the right recurrent laryngeal nerve (7,8,10,11). Therefore, the left recurrent laryngeal nerve is difficult to identify using the right-side approach because of the existence of RAA. The approach was simulated using a 3D printed model, preoperatively.

To conclude, the surgery for was safely performed using a bilateral thoracoscopic approach aided by adequate preoperative simulation using a 3D printed model. Thus, 3D printer is useful in gastrointestinal surgeries with vascular malformations.

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Footnote

Reporting Checklist: The authors have completed the CARE

reporting checklist. Available at https://tcr.amegroups.com/article/view/10.21037/tcr-22-468/rc

Peer Review File: Available at https://tcr.amegroups.com/article/view/10.21037/tcr-22-468/prf

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://tcr.amegroups.com/article/view/10.21037/tcr-22-468/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). 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 editorial office of this journal.

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