Successful Resection of Esophageal Carcinoma with Aberrant Right Subclavian Artery Using Video-assisted Thoracoscopic Surgery: Report of Two Cases

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Abstract. *The right non-recurrent inferior laryngeal nerve (NRILN) is a rare nerve anomaly that communicates the laryngeal nerve to the right vagal nerve trunk directly in the neck, which is usually accompanied by aberrant right subclavian artery (ARSA). We report on two cases of thoracic esophageal carcinoma undertaken in patients with these abnormalities: a 73-year-old woman with progressive dysphagia and a 63-year-old asymptomatic man. Although there have been 10 cases of thoracic esophageal carcinomas associated with ARSA and NRILN in literature, as far as we are aware of, this is the first report to describe successful resection using video-assisted thoracoscopic surgery (VATS). We found that the combination of preoperative recognition of the ARSA using three-dimensional computed tomography (3D-CT) and VATS in the prone position allowed for visual magnification with an excellent thoracoscopic view and facilitated successful tumor resection and preservation of NRILN.*

Non-recurrent inferior laryngeal nerve (NRILN) is a rare nerve anomaly that heads directly into the larynx *via* the right vagal nerve trunk in the neck, and has an incidence of 0.52–0.7% (1, 2). In comparison, the normal right inferior laryngeal nerve typically branches from the right vagal nerve trunk in the upper mediastinum before turning around the right subclavian artery and turning superiorly to the larynx. Furthermore, NRILN is often associated with an aberrant right subclavian artery (ARSA).

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Key Words: Esophageal cancer, video-assisted thoracoscopic surgery, nonrecurrent inferior laryngeal nerve, aberrant right subclavian artery.

In esophagectomy, cervical and mediastinal lymphadenectomy along the bilateral inferior laryngeal nerves is reported to be important for curative resection because these lymph nodes are frequently involved with cancer (3, 4). However, nerve injury can cause serious complications, such as vocal cord paralysis and aspiration pneumonia. In addition, intraoperative failure to identify vessel anomalies in the mediastinum may cause critical vessel injury. Therefore, surgeons should be aware of these anomalies in esophagectomy for esophageal cancer. The preoperative recognition of ARSA and the intraoperative identification of NRILN are crucial in preventing injury to vessels and nerves. In this aspect, video-assisted thoracoscopic surgery (VATS) has paid much attention in esophageal surgery because of reduced surgical trauma and visual enhancement.

We report on two cases of thoracic esophageal carcinomas associated with ARSA and NRILN. Although there have been 10 cases of thoracic esophageal carcinomas associated with ARSA and NRILN in the literature, as far as we are aware of, this is the first report to describe successful resection using VATS.

Case 1. A 73-year-old woman presenting a complaint of progressive dysphagia for over seven months was introduced to our hospital. Upper gastrointestinal endoscopy revealed a protruding tumor located approximately 25-29 cm distal from the incisors, which was histopathologically-diagnosed as squamous cell carcinoma (Figure 1A). Barium esophagography also revealed a 50 mm-long stricture in the upper third of the intrathoracic esophagus (Figure 1B).

Three-dimensional computed tomographic (3D-CT) images revealed ARSA arising from the descending aorta and passing behind the oral side of the esophageal tumor (Figure 2A). Enhanced CT imaging revealed that the upper thoracic esophagus was surrounded by the trachea, descending aorta, and ARSA in the upper mediastinum (Figure 2B). The preoperative diagnosis was Ut, type 2, cT3,

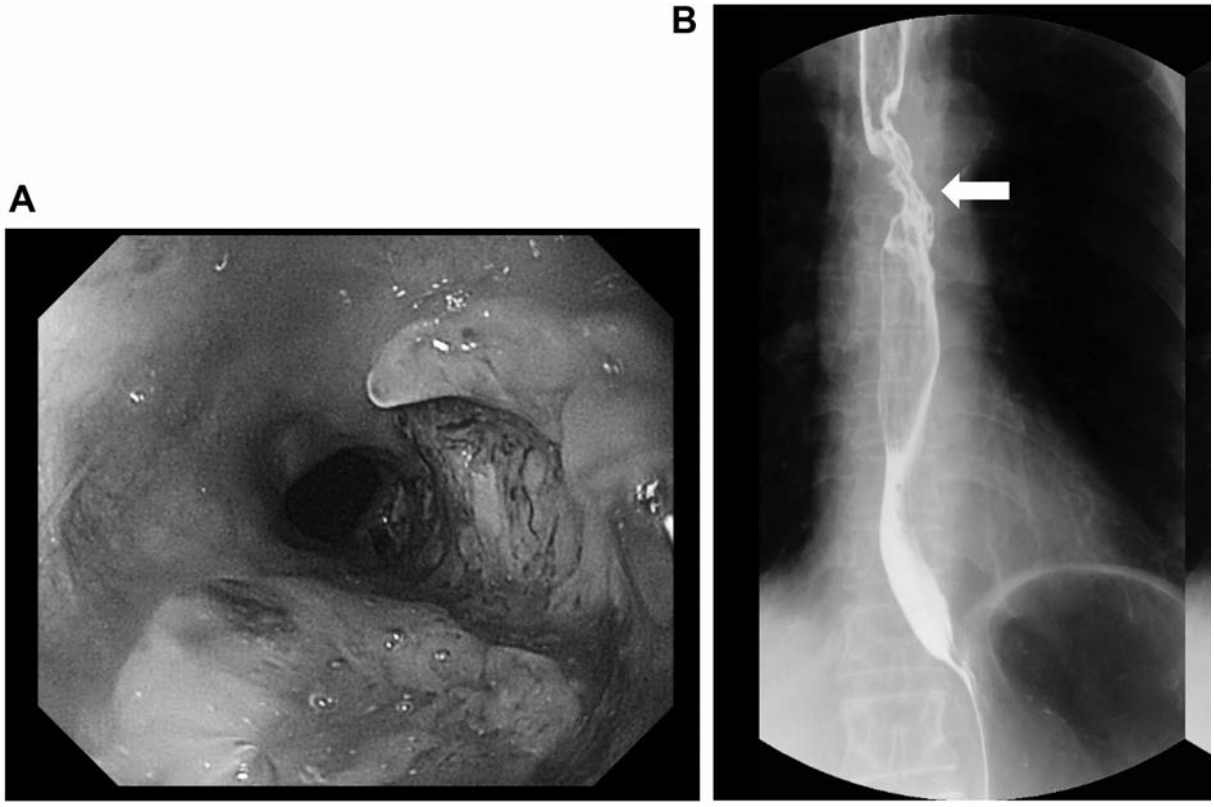

Figure 1. *Upper gastrointestinal endoscopy showed a type 3 esophageal cancer located approximately 25-29 cm distally from the incisors. Barium esophagography showed a malignant stricture (arrow) in the intrathoracic upper third of the esophagus.*

cN1, cM0, cStageIIIA (5), and the patient was treated with two courses of neoadjuvant chemotherapy using fluorouracil (5-FU) and nedaplatin (cis-diammine-glycolatoplatinum), followed by sub-total esophagectomy with three-field lymphadenectomy using VATS. The diagnosis was esophageal carcinoma accompanied with ARSA.

For surgery, the patient was placed in the lateral position and the surgical table was rotated to the prone position. Thoracoscopy was performed through five thoracic ports under CO_2 insufflation at a pressure of 7 mmHg. The combination of CO_2 insufflation and the prone position resulted in an excellent view of the posterior mediastinum because the lung was collapsed and shifted away. The thoracoscope revealed that the RSA branched from the descending aorta and continued superiorly between the esophagus and the vertebral column. The RILN, which is normally located around the RSA, could not be found and intrathoracic esophageal mobilization with radical dissection of the para-esophageal and mediastinal lymph nodes was performed. Although lymph node dissection was difficult along the left inferior laryngeal nerve because the space between the trachea and vertebra was very narrow due to the ARSA, we were able to complete the dissections safely under magnifying thoracoscopy (Figure 3). During cervical lymph node dissection through the cervical collar incision, we recognized the right NRILN branching from the right vagal nerve trunk and running directly to the larynx. The left recurrent laryngeal nerve was normally located in the tracheoesophageal groove. Simultaneously, an abdominal team mobilized the stomach and made a gastric conduit using hand-assisted laparoscopic surgery. The gastric conduit was manipulated *via* a retrosternal route, which facilitated cervical esophagogastric anastomosis using circular stapling instruments.

The postoperative course was uneventful. Microscopically, the tumor was diagnosed as moderately-differentiated squamous cell carcinoma invading the *muscularis propria* and no nodal involvement was found. The histopathological efficacy of the neoadjuvant chemotherapy was grade 2. According to the tumor, nodes, metastases (TNM) classification version 7 (Union for International Cancer Control) (5), this tumor was T2N0M0, stage IB.

Case 2. A 63-year-old man, managed for alcoholic liver disease at another hospital, was examined by gastrointestinal endoscopy and esophageal tumor was found incidentally. Further upper gastrointestinal endoscopy revealed a 0-IIc-type tumor located approximately 33–35 cm distal from the

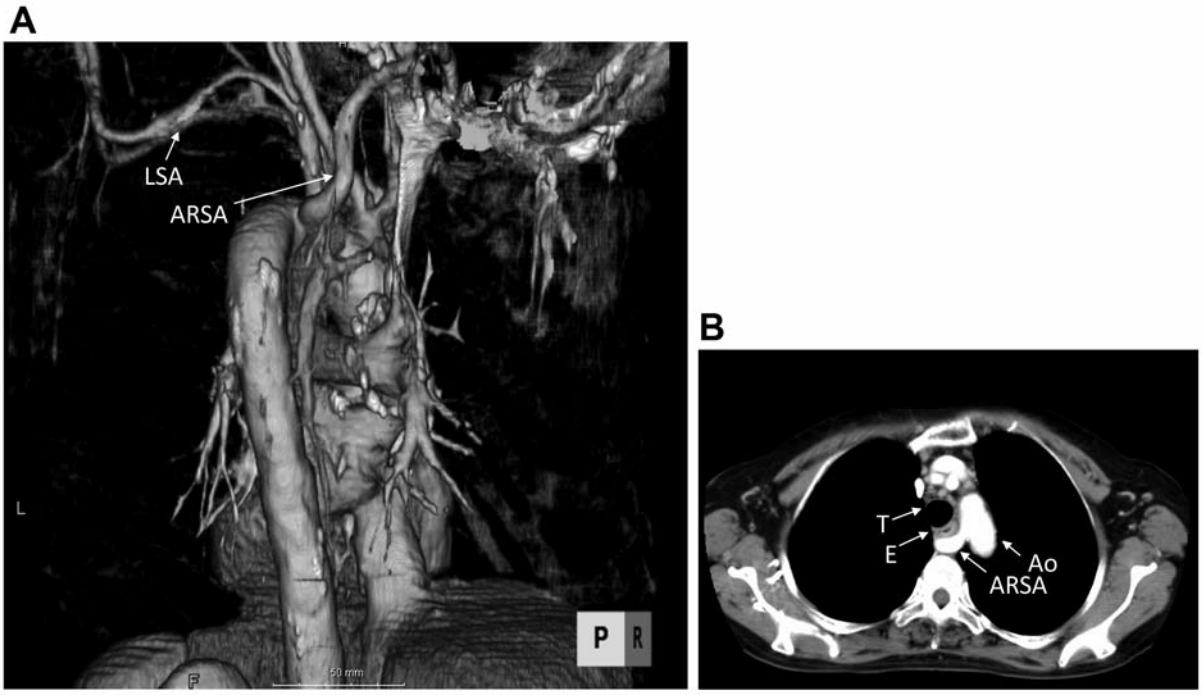

Figure 2. *Three-dimensional computed tomography (3D-CT) (right-posterior view). 3D-CT revealed the aberrant right subclavian artery (ARSA), which arose from the descending aorta (Ao). Enhanced CT showed the upper thoracic esophagus encircled by the trachea, descending Ao, and ARSA. LSA: Left subclavian artery; T: trachea; E: esophagus.*

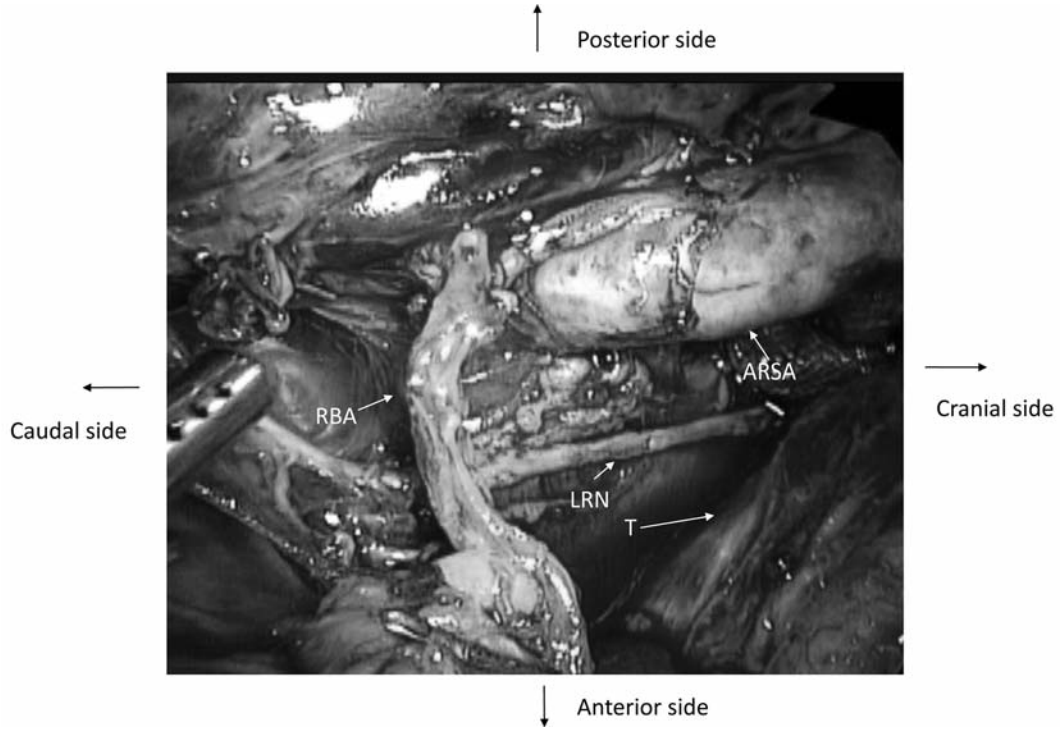

Figure 3. *A magnified view through the thoracoscope showing the upper mediastinum after esophagectomy. It was revealed that the right subclavian artery ran superiorly between the esophagus and the vertebral column to the upper thoracic cavity. RBA: Right bronchial artery; LRN: left recurrent nerve; T: trachea.*

Figure 4. *Upper gastrointestinal endoscopy showed a 0-IIc-type tumor located approximately 33–35 cm distal from the incisors.*

incisors that was histopathologically-diagnosed as squamous cell carcinoma (Figure 4). Enhanced CT imaging revealed no esophageal tumor or metastatic lymph nodes but revealed ARSA running posteriorly from the esophagus (Figure 5). Preoperative diagnosis was Mt, 0-IIc, cT1b, cN0, cM0, cStageIA (5). A subtotal esophagectomy with three-field lymphadenectomy using VATS was undertaken under a diagnosis of esophageal carcinoma with ARSA. We also suspected the presence of NRILN preoperatively.

The operation was commenced in the supine position. Through a cervical collar incision on the right side of the neck, we identified the NRILN branching from the right vagal nerve running to the larynx directly as expected preoperatively. Cervical lymph node dissection was performed preserving the NRILN. The left inferior laryngeal nerve was located normally. As with case 1, an abdominal team mobilized the stomach and manipulated a gastric conduit superiorly *via* a retrosternal route to facilitate cervical esophagogastric anastomosis. The patient was then moved to the prone position for subtotal esophagectomy and mediastinal lymphadenectomy under VATS. In this case, the thoracoscope clearly revealed that the RSA ran superiorly between the esophagus and the vertebral column. As in case 1, due to a narrow working space between the trachea and vertebra caused by the ARSA, lymphadenectomy along the left inferior laryngeal nerve was difficult.

Postoperatively, a tracheotomy was necessary due to left lower lobe pneumonia, which was successfully treated medically.

Microscopical examination revealed that the tumor was a moderately-differentiated squamous cell carcinoma invading the mucosal layer with no lymph node metastasis, and was classified as T1N0M0 stage IA (5).

Discussion

The NRILN anomaly originates during the development of the RSA. The right fourth arch will ultimately become the RSA, and the left sixth arch the arterial duct. In addition, the laryngeal nerves are pulled into the thoracic cavity with the descent of the heart, causing them to travel beneath the fourth and sixth arches before returning to the larynx. However, the distal portions from the fourth to the sixth arch on the right side occasionally disappear in an early embryonic stage. The ARSA then branches dorsally from the distal descending aorta, leaving the seventh intercostal artery attached to the descending aorta, and passes through the retroesophageal space and beneath the clavicle. Under such circumstances, the right laryngeal nerve heads directly into the larynx from the vagal nerve trunk without coursing beneath the RSA, and is termed the NRILN (12).

Although rare, NRILN is frequently reported in head and neck surgery, such as a thyroidectomy (6, 7). Toniato *et al.* classified the branched form of NRILN from the vagal nerve into three types: type 1 enters the larynx with the superior thyroid artery; type 2A accompanies the inferior thyroid artery cranially; and type 2B accompanies the inferior thyroid artery caudally (6). Our two cases were classified as type 2A.

During mediastinal and cervical lymphadenectomy performed in thoracic esophageal cancer surgery, the identification and preservation of the inferior laryngeal nerve is important. Without preoperative prediction of NRILN, surgeons must identify it intraoperatively. Although preoperative identification of the NRILN is difficult, it often closely accompanies the more readily evident ARSA. Thus, preoperative CT identification of ARSA can indicate the presence of NRILN. Moreover, identifying NRILN during the neck procedure, but before the thoracic procedure, offers improved safety with the upper mediastinal thoracic lymphadenectomy. In the second case, we initially identified the cervical NRILN, which allowed us to completely dissect the lymph nodes around the ARSA in the mediastinum.

VATS has traditionally been performed in the left lateral decubitus position; however, VATS in the prone position has received great attention in recent years because of its providing an excellent view of the surgical field (8, 9). We found that this technique was particularly useful in delineating the unusual vessel anomaly, facilitating the safe completion of the surgery. VATS in the prone position allowed for visual enlargement and an excellent thoracoscopic view, in combination with preoperative recognition using 3D-CT. During surgery of the second case, we first identified the cervical NRILN before the thoracic part, which we considered to be particularly useful.

We identified 10 cases of esophageal cancer with NRILN in the literature (10-18). The 12 cases, including our two cases, are summarized in Table I. In nine out of the 12

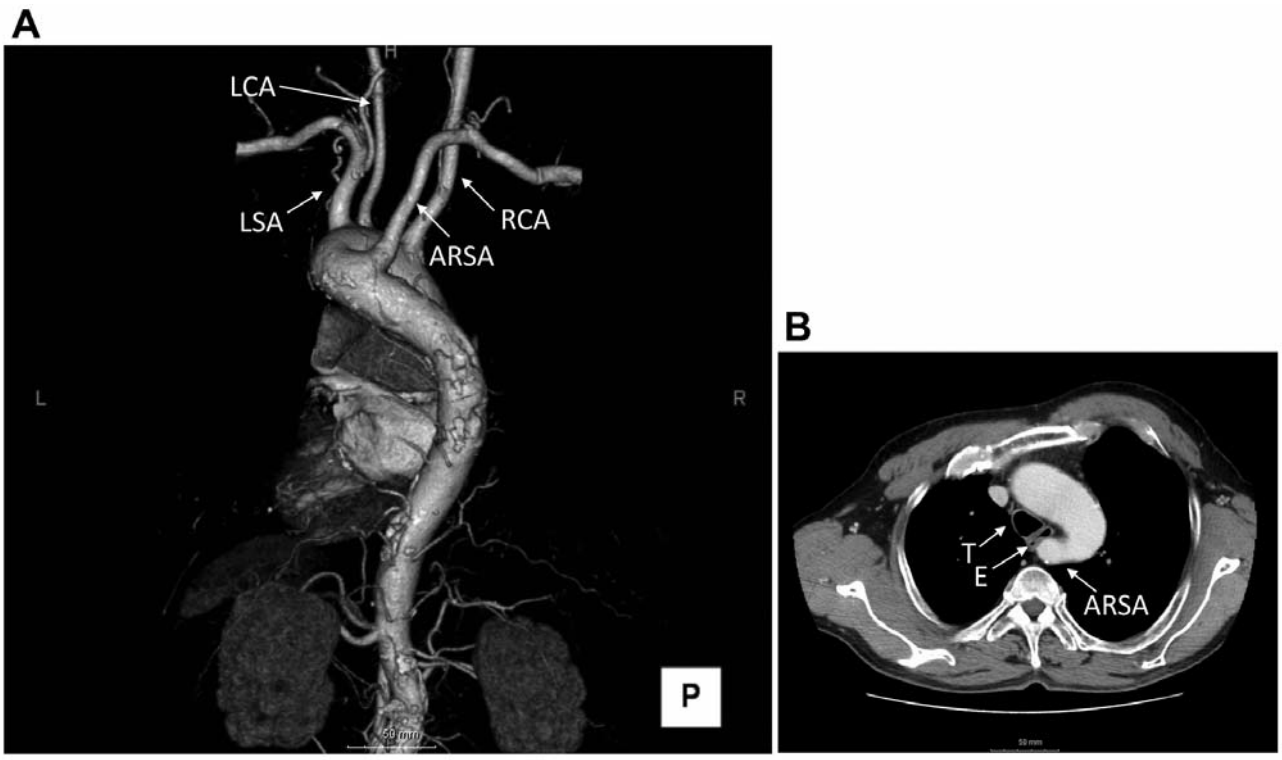

Figure 5. *Three-dimensional computed tomography (3D-CT) and enhanced CT revealed an abnormal vascular structure posterior to the esophagus: the aberrant right subclavian artery (ARSA). RCA: Right carotid artery; LCA: left carotid artery; LSA: left subclavian artery; T: trachea; E: esophagu.*

Table I. *Summary of reported cases of esophageal cancer with nonrecurrent inferior laryngeal nerve (NRILN).*

Case	Gender	Age, year	Complaint	Tumor location	Preoperative diagnosis of ARSA	Thoracic approach	Extent of LN dissection	Route of gastric conduit	Complication	Reference
1	Male	66	Dysphasia	NM	Yes	Transhiatal	NM	NM	NM	(10)
2	Male	70	Unclear	Ut	Yes	Open (transthoracic)	Three-field	Posterior mediastinal	None	(11)
3	Female	52	Dysphasia	Mt	No	Transhiatal →transthoracic	NM	NM	None	(12)
4	Female	55	Dysphasia	Mt	No	Thoracoscopy →open[a]	Three-field	NM	Injury of an ARSA during operation	(13)
5	Male	58	Dysphasia	Mt	Yes	Open (transthoracic)	Three-field	NM	Pneumonia	(14)
6	Female	70	Dysphasia	Mt	Yes	Open (transthoracic)	Three-field	NM	NM	(15)
7	Male	59	Swallowing pain	Mt	Yes	Open (transthoracic)	Three-field	NM	NM	(15)
8	Male	50	Dysphasia	Ut	Yes	Open (transthoracic)	NM	NM	None	(16)
9	Female	65	Dysphasia	Ce	Yes	Open (transthoracic)	Bilateral neck dissection	Posterior mediastinal	None	(17)
10	Male	67	None	Mt	NM	Mediastinoscopy	None	Posterior mediastinal	Vascular-digestive fistula	(18)
11	Female	73	Dysphasia	Mt	Yes	Thoracoscopy	Three-field	Retrosternal	None	Our case
12	Male	63	None	Lt	Yes	Thoracoscopy	Three-field	Retrosternal	Pneumonia	Our case

[a]Due to need for a median sternotomy due to injury of an aberrant subclavian artery. Ut: Upper thoracic esophagus, Mt: Middle thoracic esophagus, Ce: Cervical esophagus, Lt: Lower thoracic esophagus. NM: Not mentioned.

cases, ARSA was diagnosed preoperatively, and was not in two cases. The failure in recognition of the ARSA caused for intraoperative vessel injury (13) and the need to convert from a transhiatal to a transthoracic approach (12). In the present two cases, preoperative 3D-CT images clearly identified the ARSA, facilitating the safe completion of surgery. Almost all reported cases underwent open esophagectomy, apart from one, which first underwent thoracoscopic surgery and was converted to open esophagectomy because of intraoperative ARSA injury. Our two cases are the first to be successfully resected using VATS, without ARSA and NRILN damage.

Concerning the reconstruction route, Pop *et al*. reported an interesting case with a serious clinical course due to a fistula between the gastric tube (reconstructed by the posterior mediastinal route) and the ARSA (18). In this case, the narrow space between the ARSA and trachea may cause compression of the reconstructed gastric tube, especially in re-construction of the posterior mediastinal route. Therefore, we performed conduit reconstruction retrosternally, rather than through the posterior mediastinal route.

The anomaly caused significant difficulties when performing the lymphadenectomy around the left recurrent nerve because of inadequate working space due to the ARSA running dorsally to the esophagus. Sabljak *et al*. reported a case with partial resection of the ARSA during lymphadenectomy around the left recurrent nerve and reimplantation of the RSA to the right carotid artery (17). However, this combined esophageal and vascular procedure remains controversial because of its excessive invasiveness.

In conclusion, we successfully and safely performed esophagectomy under VATS in two cases of esophageal cancer accompanied by NRILN and ARSA. We found it beneficial to identify the ARSA preoperatively using 3D-CT, which may predict for the presence of NRILN. In such cases, we recommend esophagectomy under VATS in the prone position, which can provide an excellent surgical view and allow for safe lymph node dissection especially in the upper mediastinum. In addition, we recommend identifying the NRILN during the neck procedure before esophagectomy by thoracic procedure, and in reconstruction, the posterior mediastinal route should be avoided because of the narrow space.

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Received November 22, 2013
Revised December 11, 2013
Accepted December 13, 2013