



Successful surgical treatment with tracheal resection for a symptomatic vascular ring in an adult

Naoko Imanishi¹ · Masaru Takenaka¹ · Yusuke Nabe¹ · Yusuke Nishimura² · Fumihiko Tanaka¹

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Abstract

Vascular rings are congenital anomalies of the aortic arch, which may cause compression of the trachea and esophagus. Compression symptoms usually present in infants and children. Adult presentations are quite rare, and compression symptoms may not be relieved by surgical correction due to tracheal remodeling and malacia caused by a long-standing compression. Here, we present an adult case of symptomatic vascular ring formed by the right aortic arch and persistent left ligamentum arteriosus. Respiratory symptoms were predominantly due to severe tracheal stenosis caused by tracheal wall thickening through remodeling during long-standing compression, and were relieved by surgery with transection of the persistent followed by resection and reconstruction of the trachea.

Keywords Vascular ring · Right aortic arch · Adult · Tracheal compression · Tracheal resection

Introduction

Vascular rings are congenital anomalies of the aortic arch such as double aortic arch and right aortic arch that form encirclement of the trachea and esophagus, which may cause compression symptoms [1]. Symptoms usually appear in infancy, and can be resolved soon after surgical correction of the anomalies [2]. Compression symptoms rarely appear in adulthood, and adult patients may be misdiagnosed for a long time. In such cases, symptoms may not be relieved by surgical correction, because long-term compression may lead to tracheomalacia and remodeling of the trachea. Accordingly, in adult cases, optimal treatment decision-making is sometimes difficult [3–5]. Here, we report a case of an adult right aortic arch that is successfully treated with transection of the persistent ligamentum arteriosus followed by resection and reconstruction of the trachea.

Case report

A 47-year-old female complaining of wheezes, dyspnea and dysphagia was referred. She had been treated as bronchial asthma for more than 40 years. Chest computed tomography (CT) revealed compression of the trachea and esophagus by a “ring” formed by the right aortic arch and a bundle structure suggesting the presence of persistent left ligamentum arteriosus (Fig. 1). Three-dimensional (3D) CT clearly revealed the right aortic arch without aberrant left subclavian artery, and showed that the trachea was narrowed for long segments above and below the compressed site (Fig. 2). Virtual bronchoscopy showed a severe tracheal stenosis caused by thickness of the tracheal wall at the compression site (Fig. 1).

Left posterolateral thoracotomy through fifth intercostal space was performed. The esophagus was compressed by a thick fibrous band indicating the persistent left ligamentum arteriosus (Fig. 3a). Esophageal compression was resolved soon after transection of the band. However, severe tracheal stenosis due to thickening of the compressed segment of tracheal wall was not relieved (Fig. 3b), and the tracheal segment including one cartilaginous ring was circumferentially resected. Tracheal reconstruction was performed with end-to-end anastomosis using interrupted 4-0 PDS sutures (Fig. 3c). Postoperative course was uneventful, and compression symptoms have been gradually relieved after surgery.

✉ Fumihiko Tanaka
ftanaka@med.uoeh-u.ac.jp

¹ Second Department of Surgery (Chest Surgery), University of Occupational and Environmental Health, Iseigaoka 1-1, Yahata-nishi-ku, Kitakyushu 807-8555, Japan

² Department of Cardiovascular Surgery, University of Occupational and Environmental Health, Kitakyushu, Japan

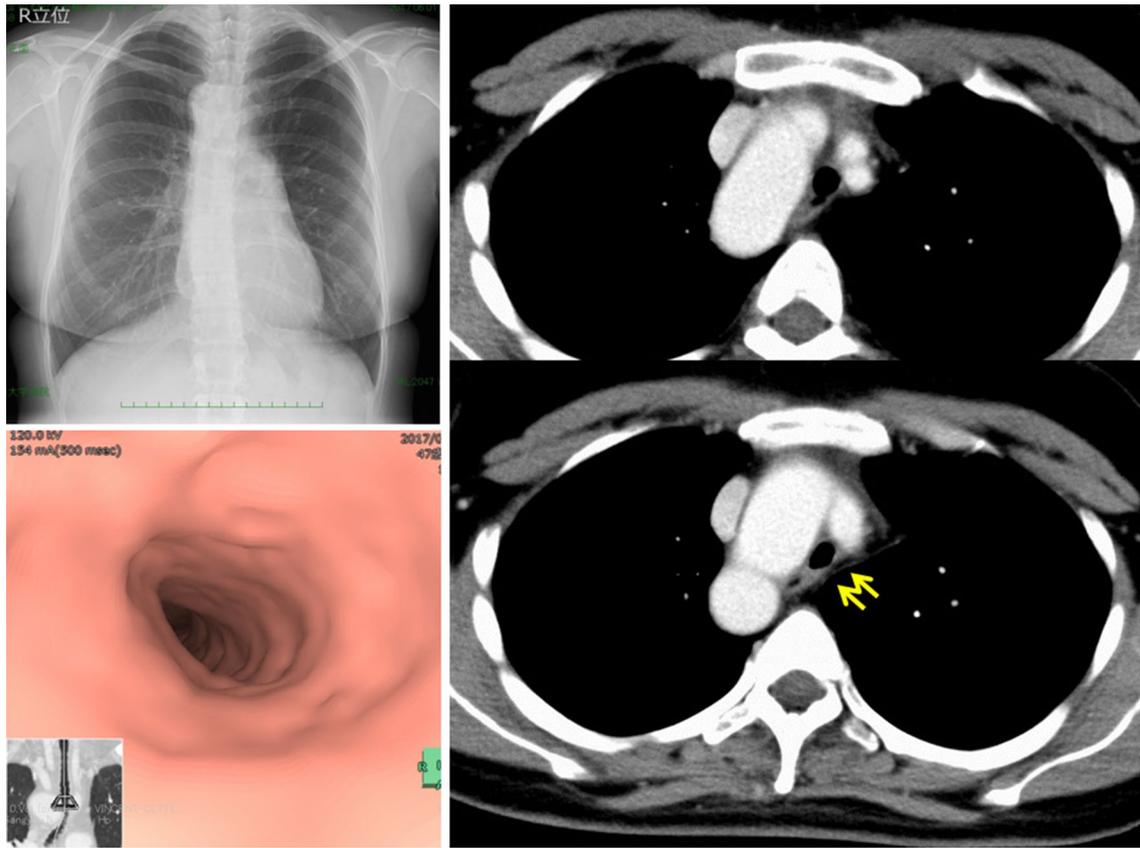


Fig. 1 Chest (left upper) roentgenogram showed right aortic arch. Chest computed tomography (right) revealed severe compression of trachea and esophagus by right aortic arch and a bundle structure

(yellow arrows). Virtual bronchoscopy (left lower) indicated a severe tracheal stenosis caused by thickness of the tracheal wall at the site of tracheal compression

Discussion

Respiratory symptoms in vascular rings may result not only from direct mechanical compression of the trachea, but also from tracheal remodeling and malacia which develop secondary to long-standing compression [3–5]. The latter mechanisms, often seen in adult cases, may not be resolved by surgical correction of vascular rings. In such cases, symptom relief may not be easily achieved, and a variety of treatments including tracheoplasty, tracheal reinforcement, and placement of tracheal stent have been tried [3]. In the present case, preoperative CT with 3D-reconstruction and virtual bronchoscopy suggested

that respiratory symptoms was partly due to tracheal compression and mainly due to tracheal stenosis caused by tracheal wall thickening through remodeling during long-standing compression. Accordingly, careful preoperative evaluation is the key to make optimal treatment decision-making for adult cases with symptomatic vascular rings.

Conclusion

We present a rare adult case of vascular ring that is successfully treated with transection of the persistent ligamentum arteriosus followed by resection and reconstruction of the trachea. Careful preoperative evaluation is the key for successful treatment.

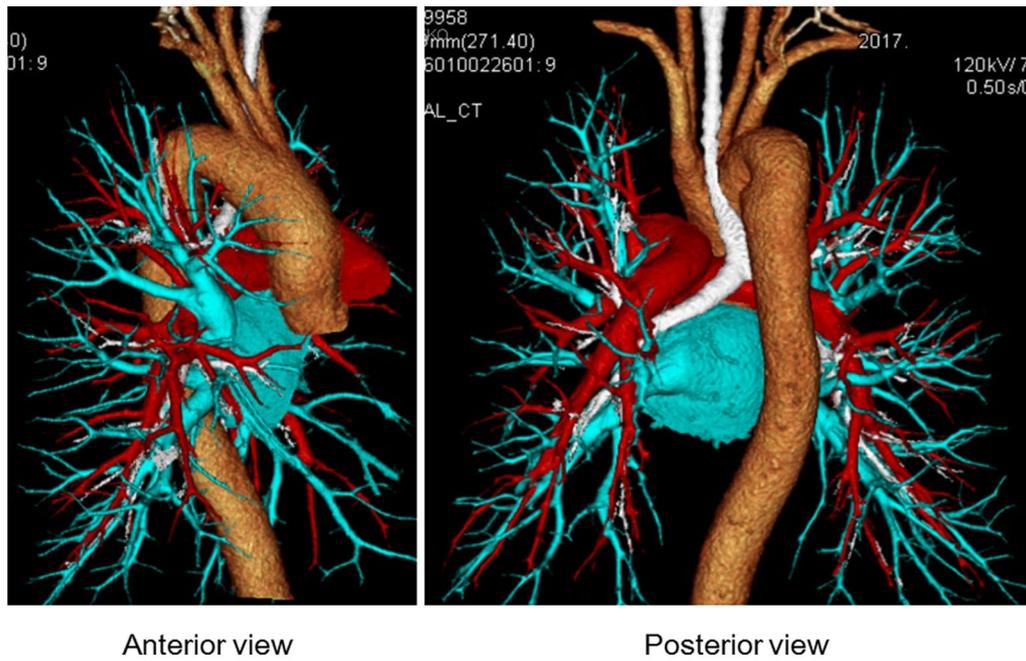


Fig. 2 Three-dimensional computed tomography showed the right aortic arch (left, anterior view), and showed that the trachea was narrowed for long segments above and below the compressed site (right, posterior view)

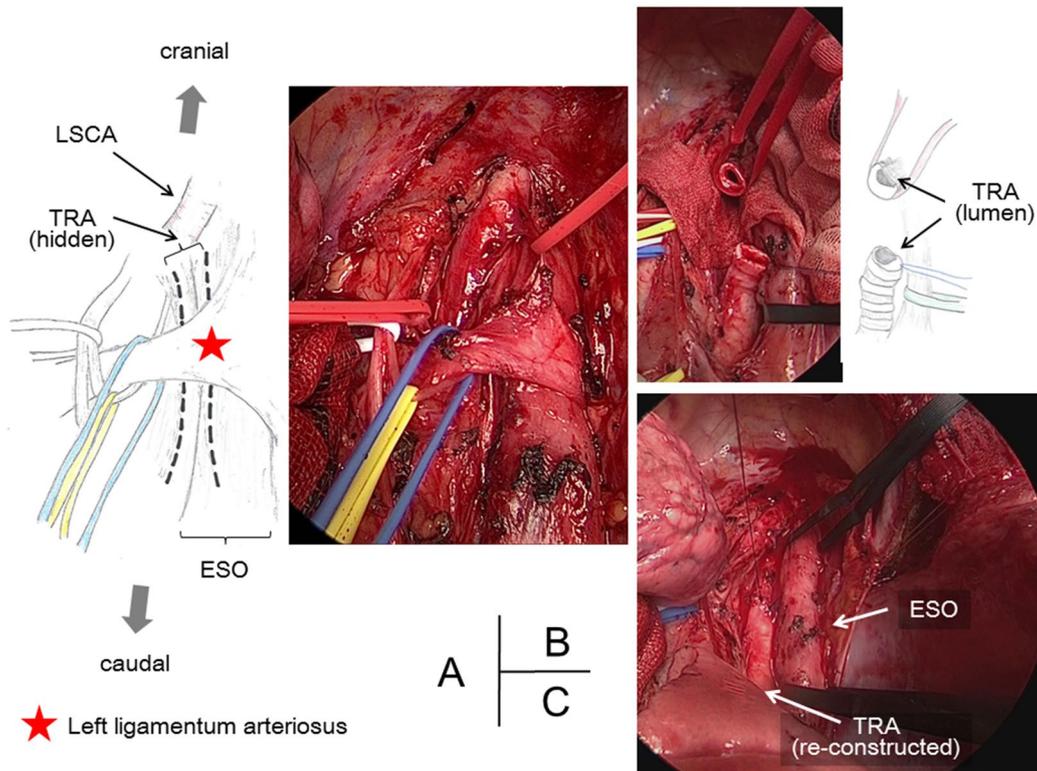


Fig. 3 a (left) Through left thoracotomy, a thick fibrous band (left ligamentum arteriosus) was exposed and taped (blue). Esophagus (ESO) was compressed by the band. Vagus nerve (white) and recurrent nerve (yellow) were also taped. The hidden trachea (TRA) was also indicated by dotted lines. *LSCA* left subclavian artery. **b** (right

upper) The trachea was exposed by transection of left ligamentum arteriosus. The cut surface revealed severe stenosis of the lumen due to thickening of the compressed segment of the tracheal wall. **c** (right lower) The thickened tracheal segment was circumferentially resected and reconstructed

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Compliance with ethical standards

Conflict of interest The authors declare that they have no competing interests.

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