Ortner Syndrome Associated With Aortic Pseudoaneurysm After Repair of Aortic Coarctation 30 Years Previously

Naoto Fukunaga, MD; Tadaaki Koyama, MD; Yasunobu Konishi, MD; Takashi Murashita, MD; Mitsuru Yuzaki, MD; Yu Shomura, MD; Hiroshi Fujiwara, MD; Yukikatsu Okada, MD

A healthy 48-year-old woman with hypertension was referred to our hospital because of an aortic pseudoaneurysm. She had undergone a replacement of the descending aorta by a 16-mm Dacron graft for aortic coarctation via a left thoracotomy 30 years before. Sudden onset of hoarseness had been presented 1 month previously, and exploration by flexible fiberoptic laryngoscopy for the causes of hoarseness revealed paralytic left vocal cord (Figure 1 and online-only Data Supplement Movie I). Three-dimensional computed tomography showed aortic pseudoaneurysm with a maximum diameter of 50 mm distal to the left subclavian artery, indicating that the pseudoaneurysm had possibly developed at the proximal anastomotic site of a previous operation (Figure 2), and the diameter of the previous Dacron prosthetic graft had dilated by $\approx 1.3$ times.

On admission, laboratory examinations were within normal limits. Transthoracic echocardiography demonstrated moderate aortic valve regurgitation with a left ventricular ejection fraction of 62%.

We assumed that hoarseness occurred because of compression of the left recurrence laryngeal nerve by aortic pseudoaneurysm. A diagnosis of Ortner syndrome associated with aortic pseudoaneurysm was made.

Figure 1. A flexible fiberoptic laryngoscopy reveals complete left vocal cord paralysis.

Figure 2. Three-dimensional computed tomography shows aortic pseudoaneurysm with a maximum diameter of 50 mm distal to the left subclavian artery.
Total arch replacement with a 4-branched knitted prosthetic graft was performed via a median sternotomy considering lung injury by pleural dissection via a left rethoracotomy and no dehiscence of the previous distal anastomosis. At the previous proximal anastomotic site, prosthetic graft was dehisced from native aorta and the pseudoaneurysm had derived from there. The new knitted prosthetic graft was anastomosed to the previous one. Cardiopulmonary bypass, circulatory arrest, and total operative time were 279, 52, and 456 minutes, respectively. Histopathological analysis of resected aortic wall showed no evidence of inflammation or infection. The postoperative course was uneventful. She has been followed up in the outpatient clinic, and her voice quality has improved gradually.

Ortner syndrome is a clinical entity defined as hoarseness caused by impairment of the left recurrent laryngeal nerve because of cardiovascular diseases. It was first described in 1897 by Norbert Ortner in a patient with left atrial enlargement secondary to mitral valve stenosis. Subsequently, the entity has been described in other cardiovascular diseases including mitral valve prolapse, atrial and ventricular septal defect, patent ductus arteriosus, pulmonary hypertension, and thoracic aortic aneurysm. In the present case, an aortic pseudoaneurysm after repair of aortic coarctation compressed the left recurrent laryngeal nerve, resulting in Ortner syndrome. To our knowledge, this is the first description of Ortner syndrome caused by pseudoaneurysm after repair of aortic coarctation.

With regard to improvement of hoarseness, early detection and treatment of underlying cardiovascular diseases generally result in its resolution. We expected her voice quality to improve because of relatively early detection of the aortic pseudoaneurysm after the onset of hoarseness. The late complications after repair of aortic coarctation are primarily recurrent stenosis and pseudoaneurysm formation. The incidence of pseudoaneurysm formation at the site of repair was 5.4%. As the interval after repair of aortic coarctation lengthened, the number of formed aneurysms increased.

As the interval after repair lengthened, we must address another problem: the durability of the implanted prosthetic graft. Reports of nonanastomotic rupture of a Dacron graft in the late stage have been published. As in our case, the diameter of the previously implanted Dacron graft dilated by 1.3 times. The optimal surgical treatment in our case was to replace the Dacron prosthetic graft that had been implanted 30 years before. However, we took into consideration the risk of lung injury during pleural dissection via rethoracotomy, and we ascertained that the distal anastomosis was intact using preoperative computed tomography. This was why we left the previous Dacron prosthetic graft in place and performed total arch replacement alone. Recently, thoracic endovascular aortic repair has undergone a number of technical advancements. If a previous Dacron prosthetic graft tends to dilate, thoracic endovascular aortic repair will be feasible in the future. It is important to address the long-term follow-up carefully using imaging modalities.

**Disclosures**

None.

**References**

Ortner Syndrome Associated With Aortic Pseudoaneurysm After Repair of Aortic Coarctation 30 Years Previously
Naoto Fukunaga, Tadaaki Koyama, Yasunobu Konishi, Takashi Murashita, Mitsuru Yuzaki, Yu Shomura, Hiroshi Fujiwara and Yukikatsu Okada

*Circulation.* 2012;125:e937-e938
doi: 10.1161/CIRCULATIONAHA.111.086421
*Circulation* is published by the American Heart Association, 7272 Greenville Avenue, Dallas, TX 75231
Copyright © 2012 American Heart Association, Inc. All rights reserved.
Print ISSN: 0009-7322. Online ISSN: 1524-4539

The online version of this article, along with updated information and services, is located on the World Wide Web at:
http://circ.ahajournals.org/content/125/20/e937

Data Supplement (unedited) at:
http://circ.ahajournals.org/content/suppl/2012/05/18/125.20.e937.DC1

**Permissions:** Requests for permissions to reproduce figures, tables, or portions of articles originally published in *Circulation* can be obtained via RightsLink, a service of the Copyright Clearance Center, not the Editorial Office. Once the online version of the published article for which permission is being requested is located, click Request Permissions in the middle column of the Web page under Services. Further information about this process is available in the Permissions and Rights Question and Answer document.

**Reprints:** Information about reprints can be found online at:
http://www.lww.com/reprints

**Subscriptions:** Information about subscribing to *Circulation* is online at:
http://circ.ahajournals.org//subscriptions/