📕 Case Report 🗐

# Endovascular Treatment of a Ruptured Pseudoaneurysm of the Intercostal Patch after Descending Aortic Aneurysm Repair

Yoshikatsu Nomura, MD, PhD,<sup>1</sup> Ryota Kawasaki, MD, PhD,<sup>2</sup> Motoharu Kawashima, MD,<sup>1</sup> Hiroshi Tanaka, MD, PhD,<sup>1</sup> and Hirohisa Murakami, MD, PhD<sup>1</sup>

Anastomotic pseudoaneurysm and patch aneurysm are lifethreatening complications following thoracoabdominal and descending thoracic aortic aneurysm (DTAA) repair. The aortic wall tissue is fragile in patients with Marfan syndrome, who are at high risk of anastomotic pseudoaneurysm and patch aneurysms. We experienced a rare case of ruptured pseudoaneurysm of the intercostal patch after DTAA repair in a patient with Marfan syndrome. A hematoma was separated from the pseudoaneurysm caused by adhesion of the left lung after DTAA repair, which made diagnosis difficult. To prevent type II endoleak and achieve thoracic endovascular aortic repair, we treated the patent intercostal arteries by embolization.

*Keywords:* pseudoaneurysm of intercostal patch, Marfan syndrome, TEVAR

#### Introduction

Anatomic pseudoaneurysm and patch aneurysm are lifethreatening complications following thoracoabdominal aortic aneurysm (TAAA) and descending thoracic aortic aneurysm (DTAA) repair. Various methods exist for reconstruction of the intercostal arteries during TAAA and

<sup>1</sup>Department of Cardiovascular Surgery, Hyogo Brain and Heart Center at Himeji, Himeji, Hyogo, Japan <sup>2</sup>Department of Radiology, Hyogo Brain and Heart Center at Himeji, Himeji, Hyogo, Japan

Received: August 5, 2020; Accepted: October 3, 2020 Corresponding author: Yoshikatsu Nomura, MD, PhD. Department of Cardiovascular Surgery, Hyogo Brain and Heart Center at Himeji, 520 Saisho-ko, Himeji, Hyogo 670-0981, Japan Tel: +81-79-293-3131, Fax: +81-79-295-8199 E-mail: y\_katsu1027@yahoo.co.jp

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## **Case Report**

A 49-year-old woman with Marfan syndrome was admitted to our hospital complaining of hemoptysis. She presented with a history of valve-sparing aortic root reimplantation for annuloaortic ectasia and aortic valve regurgitation, descending aortic replacement with intercostal arteries patch reconstruction for Stanford type B aortic dissection, and permanent pacemaker implantation in another hospital. A chest computed tomography (CT) scan showed a hematoma in the apex of the left lung and a 32-mm intercostal patch aneurysm (Fig. 1). Two pairs of Th8 and 9 intercostal arteries were reconstructed in a patch, and all four arteries were patent. Because the patch aneurysm and hematoma were separated, we initially ruled out a rupture of the patch aneurysm. However, the hematoma gradually expanded, and the hemoptysis did not stop (Fig. 2). As a result, we reconsidered and diagnosed the case as a ruptured pseudoaneurysm of the intercostal patch aneurysm. Thoracic endovascular aortic repair (TEVAR) was the first-line treatment. However, due to retrograde flow from the intercostal arteries, the use of TEVAR alone was considered to carry a risk of a type II endoleak. Therefore, embolization of the patent intercostal arteries and subsequent TEVAR were planned. The patient was operated under general anesthesia. A motor evoked potential (MEP) monitor was also used to identify spinal cord injury. An 8Fr sheath was inserted into the right femoral artery through the right inguinal incision,



Fig. 1 (A) Contrast enhanced computed tomography image showing the intercostal patch aneurysm 32-mm in diameter (arrow). (B) The hematoma was separated from the aneurysm and accumulated in the apex of the lung.



Fig. 2 The hematoma was seen gradually expanding.



Fig. 3 (A) Angiography after embolization of the intercostal artery. A faint extravasation was observed at the head of the patch aneurysm (arrow). (B) No endoleak was recognized after embolization of the patent intercostal artery and endovascular stent graft of the ruptured pseudoaneurysm of the intercostal patch.

and angiography was performed at the level of the patch aneurysm. A very faint extravasation was observed at the head of the patch aneurysm. After selectively identifying the bilateral Th8 and 9 intercostal arteries using the micro catheter and micro guide wire, transarterial embolization (TAE) was performed using platinum coils. The MEP did not change during embolization, and to protect the spinal cord, the mean blood pressure was maintained above 80 mmHg. After TAE, the sheath was replaced with a 20 Fr DrySeal sheath<sup>®</sup> (W. L. Gore & Associates, Flagstaff, AZ, USA), and TEVAR was performed using the Conformable TAG<sup>®</sup> (TGU282815J, W. L. Gore & Associates, Flagstaff, AZ, USA) (Fig. 3). Hemoptysis disappeared immediately after TEVAR; however, bloody sputum persisted until postoperative day 4. The hematoma in the left thoracic cavity gradually disappeared, and the endoleak was not detected on postoperative CT findings. The patient was discharged on postoperative day 20. No adverse events, including infection, occurred up to 2 years postoperatively.

#### Discussion

The reconstruction of the intercostal artery is an important surgical technique for protecting the spinal cord during TAAA and DTAA repair. Although various techniques were reported for reconstruction of the intercostal arteries, the island technique allows reimplantation of as many intercostal arteries as possible at once.<sup>1,2)</sup> However, this technique carries an increased risk of aneurysmal change, especially among patients with connective tissue disorders. Kulik et al. reported that the incidence of the intercostal patch aneurysm was 7.1%.<sup>3)</sup> In Marfan syndrome, the incidence rate was reported to be 22.7%.<sup>3)</sup> Another study reported an incidence rate of 25% among patients with Marfan syndrome.<sup>4)</sup> Hence, we avoid using the island patch technique in patients with Marfan syndrome.

Until a few years ago, open thoracic surgical repair

was the only treatment for this aneurysm, and it required cardiopulmonary bypass and hypothermic circulatory arrest.<sup>3)</sup> Although the result of conventional open thoracic surgical repair is good, endovascular treatment is minimally invasive, and it can be applied to cases where left thoracotomy is not possible. Several reports of endovascular treatment were published.3-7) However, this treatment exhibits various demerits, such as development of a type II endoleak from the patent intercostal artery. In addition, landing the aorta with connective tissue disease is necessary. No report exists on the embolization of the patent intercostal arteries during TEVAR, but the occurrence of a postoperative type II endoleak due to the absence of embolization was observed.<sup>3,7</sup>) In the present case, a postoperative type II endoleak was not observed by embolization of the patent intercostal artery prior to TEVAR. Treatment by endovascular repair using a physician-modified endograft with an intercostal artery directional branch was reported for intercostal patch aneurysm in a patient with Loyes-Dietz syndrome.<sup>8)</sup> This technique is possible in cases with a large diameter of the intercostal artery and the presence of a landing zone. The rupture of pseudoaneurysm and patch aneurysm is rare but fatal condition. Other than the present case, a rupture has been reported in only one case.<sup>7)</sup> In this patient, the hematoma was separated from the aneurysm owing to adhesion after left thoracotomy, which made diagnosis difficult. Due to the rupture of the pseudoaneurysm in this case, embolization of the intercostal artery was performed to prevent endoleak, which was time-consuming. If the type II endoleak remains, reaching the intercostal artery using the transcatheter approach is difficult after DTAA or TAAA repair. Therefore, if possible, the intercostal artery should be embolized.

## Conclusion

We reported a successful treated case of a ruptured pseudoaneurysm of the intercostal patch after TAAA repair. Although the occurrence of type II endoleak in the future is unclear, embolization of the intercostal artery prior to TEVAR is effective for endoleak prevention.

## **Disclosure Statement**

The authors report no conflicts of interest.

## Additional Note

This article was written based on enough informed consent and patient's agreement.

The study was reviewed and approved by the institutional review bords (IRB number: 2-21).

# **Author Contributions**

Conception and Design: YN, RK Data collection: YN, RK, MK Writing: YN Critical review and revision: all authors Final approval of the article: all authors Accountability for all aspects of the work: all authors

#### References

- 1) Okita Y, Omura A, Yamanaka K, et al. Open reconstruction of thoracoabdominal aortic aneurysms. Ann Cardiothorac Surg 2012; 1: 373-80.
- Safi HJ, Miller CC 3rd, Carr C, et al. Importance of intercostal artery reattachment during thoracoabdominal aortic aneurysm repair. J Vasc Surg 1998; 27: 58-66; discussion, 66-8.
- Kulik A, Allen BT, Kouchoukos NT. Incidence and management of intercostal patch aneurysms after repair of thoracoabdomial aortic aneurysms. J Thorac Cardiovasc Surg 2009; 138: 352-8.
- 4) Juthier F, Rousse N, Banfi C, et al. Endovascular exclusion of patch aneurysms of intercostal arteries after thoracoabdominal aortic aneurysm repair. Ann Thorac Surg 2013; **95**: 720-2.
- 5) Kitahara H, Yoshitake A, Hachiya T, et al. Hybrid thoracic endovascular aortic repair for intercostal patch aneurysm after thoracoabdominal aortic replacement. Ann Vasc Dis 2015; 8: 340-2.
- 6) Glebova NO, Hicks CW, Alam R, et al. Technical aspects of branched graft aortic reconstruction in patients with connective tissue disorders. J Vasc Surg 2016; 64: 520-5.
- 7) Schwill S, LeMaire S, Green SY, et al. Endovascular repair of thoracic aortic pseudoaneurysm and patch aneurysms. J Vasc Surg 2010; **52**: 1034-7.
- 8) Tenorio ER, Tallarita T, Mirza AK, et al. Endovascular repair of large intercostal artery patch aneurysm using branch stent-graft in a patient with Loeys–Dietz syndrome. J Thorac Cardiovasc Surg 2020; **159**: e95-9.