#### 🍃 Case Report

# Fistula between the Thoracic Duct and an Unusual Vessel Aneurysm Branching Off the Abdominal Aorta Revealed by Aneurysm Rupture: A Case Report

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Fistulas between an aneurysm branching off the abdominal aorta and the thoracic duct are rare. We report a case of aneurysmal-thoracic duct fistula diagnosed by angiography when aneurysm ruptured, and we successfully treated by catheter embolization. A 42-year-old man was referred to our hospital with a chief complaint of sudden back and chest pain. Computed tomography showed both postmediastinal and retroperitoneal hematomas, with the aneurysm from the aorta being connected to the thoracic duct. After confirming the aneurysmal-thoracic duct fistula by angiography, we performed embolization of the aneurysm. The patient has remained well for 3 postoperative months, to date.

Keywords: aneurysm rupture, fistula, endovascular surgery

#### Introduction

Management of peripheral aneurysm rupture often requires surgery or embolization. However, some of the aneurysms identified by contrast-enhanced computed

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**(C) BY-NC-SA** ©2018 The Editorial Committee of Annals of Vascular Diseases. This article is distributed under the terms of the Creative Commons Attribution License, which permits use, distribution, and reproduction in any medium, provided the credit of the original work, a link to the license, and indication of any change are properly given, and the original work is not used for commercial purposes. Remixed or transformed contributions must be distributed under the same license as the original. tomography (CT) shows anatomical complications, which are difficult to diagnose or treat. A 42-year-old man was admitted to our hospital with sudden back pain. Angiography revealed an aneurysmal-thoracic duct fistula, with the aneurysm directly branching off the abdominal aorta. To the best of our knowledge, no other cases similar to ours have been reported previously.

### **Case Report**

A 42-year-old man was referred to our hospital for sudden back and chest pain. He had a history of dyslipidemia. He had no history of traumatic injury, but had received acupuncture treatment on his back. At hospitalization, his blood pressure was 125/69 mmHg and heart rate was 91 bpm. Laboratory examinations revealed white blood cell count of 15,400 cells/µL, hemoglobin level of 13.3 mg/dL, and C-reactive protein level of 0.05 mg/dL. Contrastenhanced CT showed a post-mediastinal hematoma and unusual vessels, which branched off at the same level as the abdominal aorta (superior mesenteric artery [SMA]) and formed an aneurysm. From the aneurysm, unidentifiable vessels branched to both cranial and caudal sides of the aorta (Fig. 1). The vessel branching off on the cranial side of the aneurysm ran along the aorta, and seemed to be connected to the left subclavian artery. The patient was diagnosed with aneurysm rupture attributed to an unusual vessel leading to the aneurysm. After obtaining informed consent from the patient, we performed emergent angiography to embolize the ruptured aneurysm. The procedure was performed under local anesthesia as follows: A 5-Fr sheath was introduced via the left radial artery, and a 6-Fr sheath via the right femoral artery. At first, to identify the unusual vessels branching off the aneurysm and the point at which the rupture had occurred, an angiogram of the left internal thoracic artery (LITA) was obtained to prevent the sheath from passing through the aneurysm.



Fig. 1 (a) An unusual vessel is seen to branch off at the same level as the abdominal aorta (superior mesenteric artery [SMA]). (b) Distal side of the unusual vessel can be seen to have formed an aneurysm. (c) From the aneurysm, the thoracic duct ran in parallel with the aorta on both the cranial and caudal sides. (d) Post-mediastinal hematoma and unusual vessels can be seen. (e) A schema of anatomic relationship between aorta, aneurysm, and thoracic duct.



Fig. 2 DSA images show the aneurysm in detail. The distal side of the aneurysm can be seen as being connected to the thoracic duct (aneurysmal-thoracic duct fistula [arrow]).

However, the angiogram obtained from the LITA and left common carotid artery (LCCA) showed neither an unusual vessel nor the aneurysm. However, in the venous phase, the vessel, which was considered to be unusual on a CT image, could be confirmed. Thus, a 6-Fr sheath was introduced from the right femoral artery, and a 4-Fr CXI support catheter (Cook Medical in Japan, Tokyo, Japan) was advanced to the aneurysm branching off the abdominal aorta. Digital subtraction angiography (DSA) images were obtained. With the assistance of a road map, the upper side of the unusual vessel branching off the aneurysm was catheterized using a 0.014-inch guide wire (Cruise, ASAHI INTECC J-sales, INC., Tokyo, Japan). Then, DSA images were obtained again (Fig. 2).

This procedure revealed that the unusual vessel branching off the aneurysm was a thoracic duct, which ran into the left subclavian vein. A diagnosis of aneurysmalthoracic duct fistula associated with aneurysm rupture was made. Therefore, to isolate the aneurysm, both the aneurysmal ostium from the aorta and the thoracic duct had to be embolized. Using a 0.018-inch embolization coil (Tornado<sup>®</sup>, Cook Medical in Japan, Tokyo, Japan), we embolized both the cranial and caudal sides of the thoracic duct. DSA images then showed that all flow from the distal side of the aneurysm had disappeared. Similarly, using an 8-mm Amplatzer<sup>TM</sup> vascular plug (AVP, St. Jude Medical, St. Paul, MN, USA), we embolized the proximal side of the aneurysm. Angiography, performed before and after AVP2 placement, confirmed total occlusion of the aneurysm and successful embolization (Fig. 3). A vascular closure device was used to close the right femoral puncture site, and the embolization procedure was completed.



Fig. 3 Both cranial and caudal sides of the thoracic duct were embolized with a 0.018-inch embolization coil (Tornado<sup>®</sup>, Cook Medical, Tokyo, Japan). The proximal side of the aneurysm was embolized with an 8-mm Amplatzer<sup>™</sup> vascular plug. The DSA image illustrates successful embolization.

The postoperative contrast-enhanced CT showed no blood flow into the aneurysm, revealing disappearance of the post-mediastinal hematoma. The patient made good progress after the operation, and discharged on postoperative day 7. The follow-up CT examination after 3 months revealed total embolization of the aneurysm, with no post-mediastinal or retroperitoneal hematoma.

#### Discussion

Aneurysms are usually asymptomatic until they enlarge or rupture. In the present case, the aneurysm became obvious when it ruptured and caused sudden back pain. The aneurysm was connected to the abdominal aorta at the proximal side and with the thoracic duct at the distal side. To the best of our knowledge, there are no prior reports of congenital fistula between an aneurysm branching off the aorta and the thoracic duct at the abdominal level. However, a few cases with a fistula between the abdominal aorta and the thoracic duct, resulting from surgical or traumatic injury, have been reported.<sup>1)</sup> In the present case, the anatomy of the aorta and the thoracic duct through the aneurysm was complex. We consider the proximal side of the aneurysm (aneurysm connected to the aorta) and the distal side of the aorta (aneurysmal-thoracic duct fistula) to be pathologically different.

Several mechanisms such as trauma, infection, and genetic susceptibility have been suggested to explain the formation of aneurysms. Although aneurysms occur in older age groups, our patient was relatively young with a peculiarly shaped aneurysm. Thus, we suppose that the aneurysm in the presenting patient was neither pseudoaneurysm secondary to trauma nor atherosclerotic aneurysm. We consider the mechanism of forming aneurysm in the present case is congenital.

With regard to the distal side of the aneurysm, the fistula was either congenital or had been acquired after birth. The present patient had no history of traumatic injury, but he had received acupuncture treatment on his back. In East Asia, acupuncture is a relatively safe and widely used therapy for chronic pain.<sup>2)</sup> Generally, during acupuncture, a needle is not inserted deeper than the muscle layers. In the present case, the abdominal aorta was located at a depth of 7 cm from the skin. If the aorta had been punctured by accident, it might have caused pain and bleeding from the posterior mediastinum. Therefore, acupuncture was considered not to be responsible for the fistula. Although it cannot be ruled out that the fistula was congenital, we believe that this fistula between an aneurysm and the thoracic duct had resulted from aneurysmal perforation. We drew this conclusion because, on the DSA images, the thoracic duct could gradually be imaged from the aneurysm, indicating the fistula to be acute. Had the fistula been congenital and connected to the aorta for a long time, the thoracic duct should already have been developed and would have been quickly imaged.

With regard to the proximal side of the aneurysm, diagnostic considerations can be applied. Arteriovenous malformation (AVM) is defined as abnormal connections of an artery and a vein without capillary vessels, which can be seen at other sites in the body. Prior case reports have described congenital AVM in the posterior mediastinum or retroperitoneum.<sup>3,4)</sup> However, few cases with AVM originating from the aorta, and none with AVM connected to both the aorta and the thoracic duct, have been reported. Another differential diagnosis we considered was a tumor, such as hemangioma, lymphangioma, angiolipoma, or angiosarcoma. Previous case reports have described tumors in the retroperitoneum, and several of them have attempted to differentiate vascular tumors from AVM based on clinical features or imaging studies.<sup>5-7)</sup> However, it may be difficult to distinguish hemangioma from AVM.<sup>4)</sup> Biopsy with needle aspiration or a surgical procedure is essential for obtaining a definitive diagnosis. Furthermore, for asymptomatic AVM, surgical treatment should be performed. However, in the present case, we avoided needle biopsy because we suspected that it might promote bleeding from the aneurysm. Instead, catheter embolization was prioritized over surgical resection because approaching to the aneurysm directly was anatomically difficult and saving the patient's life was the top priority. However, although the life-saving surgery was successful, a definitive diagnosis based on tissue specimens could not be made. Therefore, follow-up CT examinations at regular intervals are essential in order to check neovascularity or a tendency for enlargement.

## Conclusion

We experienced a case of an aneurysmal-thoracic duct fistula, with aneurysm rupture, that was successfully managed by percutaneous direct embolization. This case is very rare in that there have been no other reports describing an aneurysm connected to both the abdominal aorta and the thoracic duct. Because a definitive diagnosis was not obtained with our catheter embolization approach, regular follow-up examinations are essential.

## **Disclosure Statement**

All authors have no conflict of interest.

## **Author Contributions**

#### Writing: TY

Critical review and revision: all authors Final approval of the article: all authors Accountability for all aspects of the work: all authors

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