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Case Report

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# Coil Embolization for a Ruptured Intercostal Artery Aneurysm Complicated with Neurofibromatosis Type 1

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## Abstract

A 52-yearsold man with Neurofibromatosis type-1 (NF1) presented with sudden dyspnea and chest pain due to spontaneous massive hemothorax. Three-Dimensional Computed Tomographic angiography (3DCT) clearly revealed an intercostal artery aneurysm. We performed endovascular treatment using coils for aneurysmal embolization. The patient was discharged from the hospital without any complications, and remains free of symptoms at 19 months.

**Keywords:** Coil embolization; Intercostal artery aneurysm; Neurofibromatosis type 1(NF1)

## Introduction

Neurofibromatosis type 1 (NF1), also known as Von Recklinghausen's disease, is a hereditary autosomal dominant disorder characterized mainly by café-au-lait spots and cutaneous neurofibromas. It may also involve vascular, bone, or ocular lesions. The incidence of concomitant vascular disease is only 3.6%, and arterial aneurysm, rupture, stenosis or arteriovenous fistula has been reported [1]. These arterial lesions are considered to be caused by arterial fragility [2]. Spontaneous rupture of a major artery is extremely uncommon. Here we report a NF1 patient who had spontaneous rupture of an intercostal artery with massive hemothorax.

## Presentation

Patient consent was obtained. A 52-year-old man was transported by ambulance for reasons of general malaise. He had many café-au-lait spots on his skin. He was diagnosed as having NF1 by skin biopsy of a café-au-lait spot. Circulation and breathing were stable. The hemoglobin level was 16.9 g/dl. The hematocrit level was 46.4%. On the second day of hospitalization, he suddenly complained of chest pain and dyspnea. Three-Dimensional Computed Tomographic angiography (3DCT) revealed a saccular aneurysm at the left fifth intercostal artery (Figures 1, 2) and massive pleural effusion in the left thorax (Figure 1).

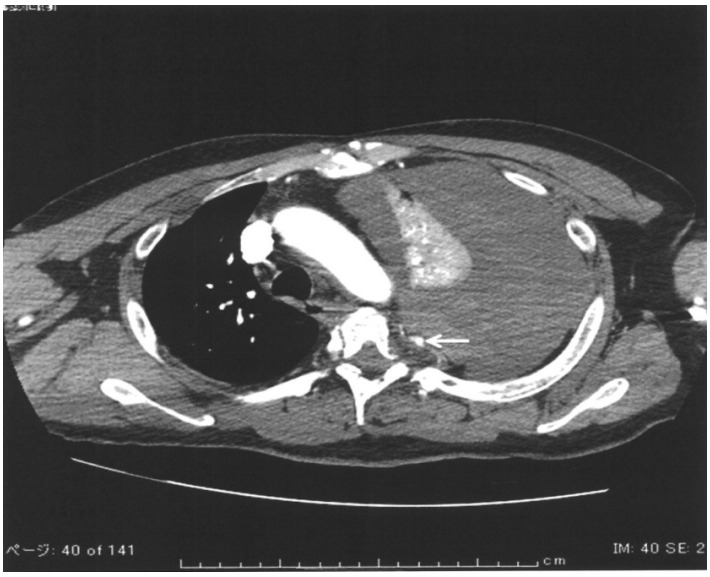


**Figure 1:** Contrast-Enhanced Computed Tomography (CECT) at the presentation of sudden dyspnea and chest pain. This shows massive pleural effusion and the left fifth intercostal artery aneurysm. White arrow

indicates the left fifth intercostal artery aneurysm.

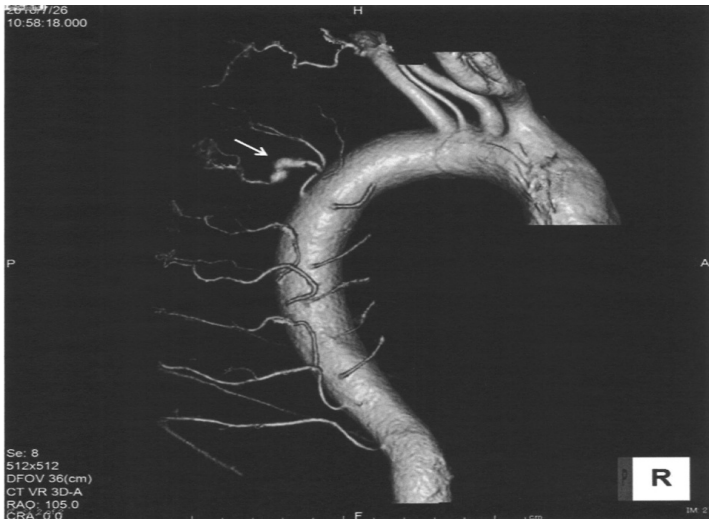


**Figure 2:** Three-dimensional computed tomographic angiography at the presentation of sudden dyspnea and chest pain. White arrow indicates the fifth intercostal artery aneurysm.

The size of the aneurysm was 8 mm × 4 mm × 3 mm. The pleural effusion did not have a contrast effect on imaging. The hemoglobin level was 9.1 g/dl, and hematocrit was 25.8%. He was in clinical shock. Left chest drainage was performed. Approximately 2800 ml of blood was removed from the left thorax. A blood transfusion was provided. The discharge liquid was blood, but sustained bleeding did not occur after the drainage. His circulation and respiration improved after these procedures. We concluded that the bleeding was controlled. We decided to perform scheduled embolization of the aneurysm. Using the Seldinger technique and the right femoral approach, a 6Fr introducer sheath (Radifocus Introducer II H, Terumo Corporation, Tokyo, Japan) and a 4.2Fr guiding catheter (Goodtec catheter GCB4-APR2, Goodman, Nagoya, Aichi, Japan) were placed. A 1.7Fr micro catheter (Excelsior SL-10, Stryker, Tokyo, Japan) over a microguidewire (Chikai AIN-CKI-200-RC, Asahi Intecc, Seto, Aichi, Japan) was advanced through the guiding catheter into the left fifth intercostal artery. We performed embolization of the aneurysm and the proximal artery with coils. We used seven coils (ED coil14 standard, Kaneka Medix, Osaka, Japan) as follows: two coils (8 mm-20 cm), two coils (5 mm-15 cm), two coils (4 mm-12 cm), and one coil (3 mm-12 cm). Selective arteriography to the left fifth intercostal artery was performed after the embolization. The aneurysm al cavity and the proximal portion of the aneurysm were filled with coils. There was no extravasation around the aneurysm and no blood flow to the distal artery (Figure 3).

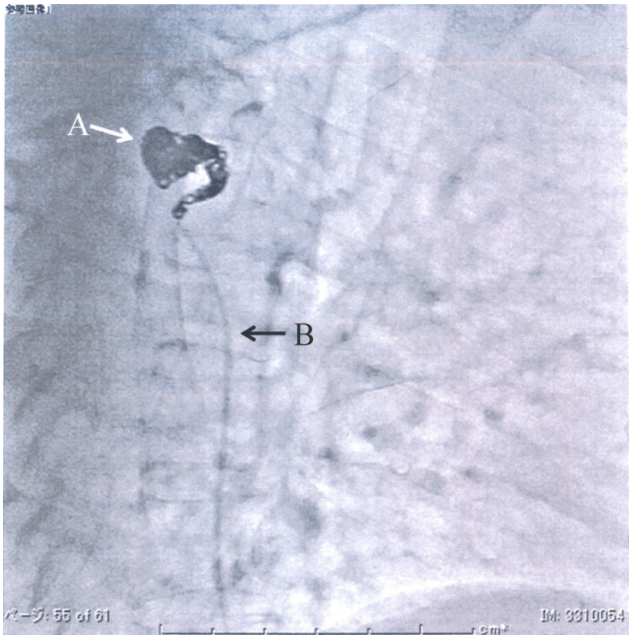


**Figure 3:** Selective arteriography to the left fifth intercostal artery after the embolization. The intra-aneurismal cavity and the proximal portion of the aneurysm are filled with coils. White arrow A indicates the fifth intercostal artery aneurysm. Black arrow B indicates the catheter for arteriography.

Re-rupture did not happen during the embolization procedure. The patient left the hospital 16 days after embolization. At 19 months after discharge, there were no symptoms or signs of recurrence.

## Discussion

Chest vascular lesions of NF1 patients may occur in the intercostal artery, subclavian artery, internal thoracic artery, vertebral artery, brachial artery, and thyrocervical artery. Aneurysm rupture, blood vessel tearing, or breakdowns of the tumor vessel are the main causes of hemothorax in such patients.

Because the blood vessel tissue is fragile in NF1, thoracotomy (open surgery or video-assisted thoracic surgery [3]) and a direct approach to diseased vessels are often difficult. Similarly, ablation of the bleeding part may not be tolerated. Endovascular Treatment (EVT) techniques have improved recently [4-6]. Although EVT carries a risk of rupture, the risk of EVT is considered to be less than those of thoracotomy and ligation of fragile vessels. It is important to carefully operate the guide wire and avoid over-packing the aneurysm with coils. One operator has contended that liquid embolic agents such as N-butyl-cyanoacrylate or gelatin

are better than solid coils [7]. However, there are many reports of successful coil embolization of intercostal artery aneurysms in NF1 patients, including our report, and we concluded that our strategy was technically feasible and effective. It should be noted that complications such as paraplegia due to embolization of the Adamkiewicz artery are possible [6]. In our case, preoperative diagnosis of the intercostal artery aneurysm was possible due to the clear imaging by 3DCT angiography [8]. As NF1 can be diagnosed by a simple physical examination and interview, we should consider the possibility of aneurysm rupture when we encounter a spontaneous hemothorax in NF1.

## References

1. Brasfield RD, Gupta TKD (1972) Von Recklinghausen's disease: a clinicopathological study. Ann Surg 175: 86-104.

2. Leier CV, DeWan CJ, Anatasia LF (1972) Fatal hemorrhage as a complication of neurofibromatosis. Vasc Surg 6: 98-101.

3. Miyazaki T, Tsuchiya T, Tagawa T, Yamasaki N, Nagayasu T (2011) Spontaneous hemothorax associated with von Recklinghausen's disease: report of a case. Annals of Thoracic and Cardiovascular Surgery 17: 301-303.

4. Dominguez J, Sancho C, Escalante E, Morera JR, Moya JA, et al. (2002) Percutaneous treatment of a ruptured intercostal aneurysm presenting as massive hemothorax in a patient with type I neurofibromatosis. J Thorac Cardiovasc Surg 124: 1230-1232.

5. Arai K, Sanada J, Kurozumi A, Watanabe T, Matsui O (2007) Spontaneous hemothorax in neurofibromatosis treated with percutaneous embolization. Cardiovasc Intervent Radiol 30: 477-479.

6. Aizawa K, Iwashita C, Saito T, Misawa Y (2010) Spontaneous rupture of an intercostal artery in a patient with neurofibromatosis type 1. Interact Cardiovasc Thorac Surg 10: 128-130.

7. Keeling AN, Costello R, Lee MJ (2008) Rasmussen's aneurysm: a forgotten entity? Cardiovasc Intervent Radiol 31: 196-200.

8. Nakamoto K, Hashiyada H, Fujii T, Takeshige M, Yoshida K, et al. (2015) Visualization of Vascular Anastomosis as Responsible Sites of Hemoptysis by Three-Dimensional Angiography for the Guidance of Endovascular Treatment. Int J Radiol Imaging Technol 1.