Radiology Case Reports 19 (2024) 901–905

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

Direct sac puncture embolization for a left subclavian aneurysm with Marfan syndrome: A case report ☆

Akemi Ohtani, MD[a], *Masato Saito, MD, PhD*[a,*], *Naoki Hirokawa, MD, PhD*[b], *Hiroki Okuda, MD, PhD*[a], *Hiroki Sato, MD*[a], *Koh-ichi Sakata, MD, PhD*[a]

[a] *Department of Radiology, Sapporo Medical University Hospital, 291 South 1 West 16, Chuo-Ku, Sapporo City, Hokkaido 060-8543, Japan*

[b] *Department of Diagnostic Radiology, KKR Sapporo Medical Center, 3-40 Hiragishi 1-6, Toyohira-Ku, Sapporo City, Hokkaido 062-0931, Japan*

ARTICLE INFO

Article history:
Received 11 November 2023
Accepted 22 November 2023

Keywords:
Direct sac puncture
Subclavian artery aneurysm
Type II endoleak
Marfan syndrome

ABSTRACT

Subclavian artery aneurysms, occasionally related to connective tissue diseases, including Marfan syndrome, are rare and conventionally managed with surgery or endovascular treatment. However, in some cases, both interventions are challenging because of the inability to reach an aneurysm through a safe route or postoperative adhesion. This report describes the case of a 43-year-old patient with a left subclavian artery aneurysm and Marfan syndrome. In this case, the patient's 5 previous surgeries related to Marfan syndrome made surgery and endovascular treatment difficult. Therefore, an alternative was researched, and we decided to perform a method of percutaneous embolization with coils and N-butyl cyanoacrylate using the direct puncture technique, which succeeded in eliminating the blood flow in the left subclavian artery aneurysm. No severe complications were associated with the procedure. The patient was free from the risk of an aneurysm rupture post-treatment, and the left back pain improved. Follow-up computed tomography 2 years postsurgery revealed the aneurysm being under control without re-enlarging. Our method is considered an effective and safe therapeutic option for cases in which surgical approach and transarterial access routes are limited.

☆ Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

* Corresponding author.
E-mail address: saitomasa@sapmed.ac.jp (M. Saito).

https://doi.org/10.1016/j.radcr.2023.11.059

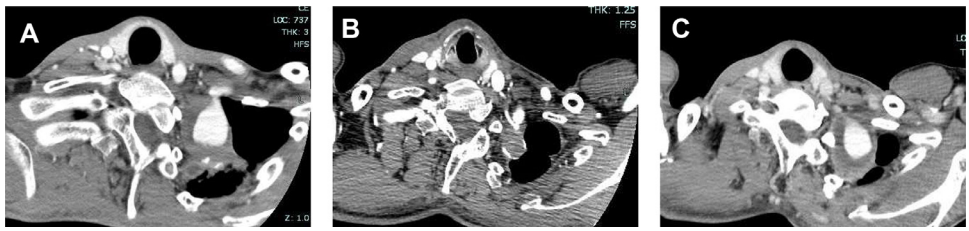

Fig. 1 – Because of the enlargement of the false lumen remnant of the left subclavian artery (A, 15 years ago), thoracic endovascular aortic repair and internal carotid artery revascularization were performed. Once, it looked in good condition (B, 12 years ago), but gradually re-enlarged (C, just before our treatment).

Introduction

Marfan syndrome is one of the connective tissue disorders inherited in an autosomal dominant pattern. This disorder affects the skeletal, ocular, and cardiovascular systems. Subclavian artery aneurysms are rare, accounting for <1% of all peripheral aneurysms, and the underlying causes of these aneurysms are connective tissue diseases, including Marfan syndrome or Ehlers-Danlos syndrome, which are rare [1,2]. As a treatment for subclavian artery aneurysms, although surgery is historically the first choice, this treatment is associated with a risk of complications due to artery deterioration and previous operations in patients with connective tissue disorder [2]. Recently, the use of minimally invasive endovascular approaches, including revascularization, has been reported [2–4]. However, these interventions are sometimes unsuccessful due to their inability to reach an aneurysm through a safe route or postoperative adhesion. In such cases, another option is sought, and optimal treatment strategies are not yet well known. In this case report, we describe the case of a patient with a left subclavian artery aneurysm and Marfan syndrome, effectively treated with embolization with coils and N-butyl 2-cyanoacrylate (NBCA) using a direct sac puncture (DSP) technique percutaneously. To our knowledge, only a few reports have mentioned percutaneous embolization of a subclavian artery aneurysm using the direct puncture technique.

Case report

A 43-year-old patient with a left subclavian artery aneurysm and Marfan syndrome underwent 5 surgeries for Stanford type B aortic dissection and impending abdominal aortic aneurysm (AAA) rupture 23 years ago. The Bentall procedure, vascular graft replacement within the descending aorta, and left subclavian artery revascularization were adopted. Subsequently, the patient was diagnosed with an enlarged false lumen remnant of the left subclavian artery (Fig. 1A). Therefore, the patient underwent thoracic endovascular aortic repair and internal carotid artery revascularization. The aneurysm was isolated by blocking blood flow into the aneurysm, and the patient's condition appeared to be under control for a while. However, the aneurysm continued to enlarge, likely secondary to type II endoleaks from the vertebral artery, and an additional intervention was necessary (Figs. 1B and C). Unfortunately, because of the patient's previous surgeries, open surgery and transarterial endovascular treatment via the femoral or radial artery were considered difficult. Then, the patient was referred to our department for minimally invasive treatment. The patient had a history of left back pain and pregabalin intake (75 mg daily), and his pain was under control. However, we could not judge whether his pain was associated with an aneurysm.

First, the aneurysm and subclavian arterial blood flow were evaluated using various imaging modalities. Contrast-enhanced computed tomography showed a 26-mm aneurysm with mural thrombus and endoleak findings (Fig. 1C) and revealed left vertebral-occipital arteries and left costocervical trunk-occipital artery anastomoses (Fig. 2). Four-dimensional CT of the vertebral artery revealed retrograde blood flow into the aneurysm. The costocervical artery appeared to function as an outflow and inflow vessel. To-and-fro vertebral artery flow was recognized on Doppler ultrasonography (US). Accordingly, 3 access routes to the aneurysm were considered: (1) from the occipital artery to the aneurysm via vertebral artery anastomosis, (2) via vertebral artery puncture, and (3) via direct puncture to the aneurysm. The first option requires a skin incision and exposure of the occipital artery and features a long pathway through a small tortuous anastomotic vessel that may be unreachable; the second option requires a deep puncture and is associated with a high risk of bleeding, and the third option—DSP—is simple and feasible and has a lower risk of complications. Thus, embolization via DSP was finalized.

The following procedures were performed under local anesthesia induced by 1% xylocaine. We identified an anechoic region with a flow signal using the US, directly punctured with an 18-gauge Surflo cannula (Terumo Corporation, Tokyo, Japan). Subsequently, we placed a 4-Fr sheath introducer (introducer set; Medikit, Co., Tokyo, Japan). Digital subtraction angiography of the sheath revealed vertebral and costocervical arteries traffic with the aneurysm (Fig. 3A). The vertebral artery was the main inflow vessel; however, significant upstream contrast reflected to-and-fro blood flow. To prevent nontarget embolization, coil embolization of the vertebral artery was performed before filling the aneurysm with NBCA. We used a co-axial system comprising a 4-Fr catheter and a 2.4-Fr steerable microcatheter (LEONIS Mova; Sumitomo Bakelite, Tokyo, Japan) to access the vertebral artery. While adjusting the tip angle, a steerable microcatheter was advanced

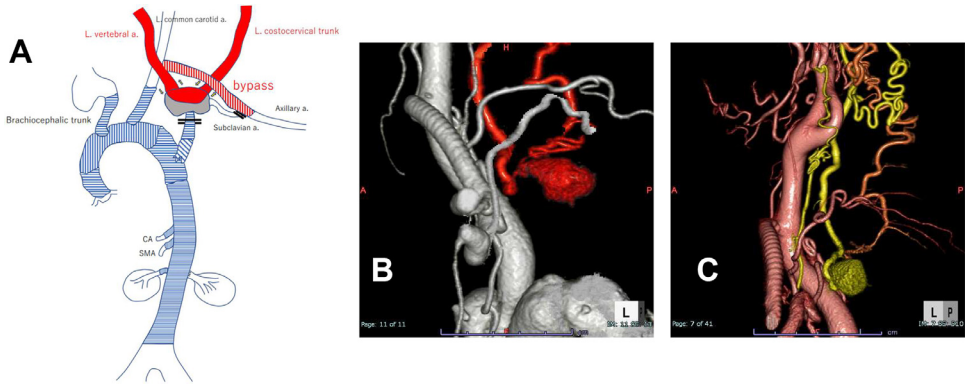

Fig. 2 – The schema expressing his vessel state before our treatment (A), left vertebral artery-left occipital artery, and left costocervical trunk-left occipital artery anastomosis revealed by CECT (B and C).

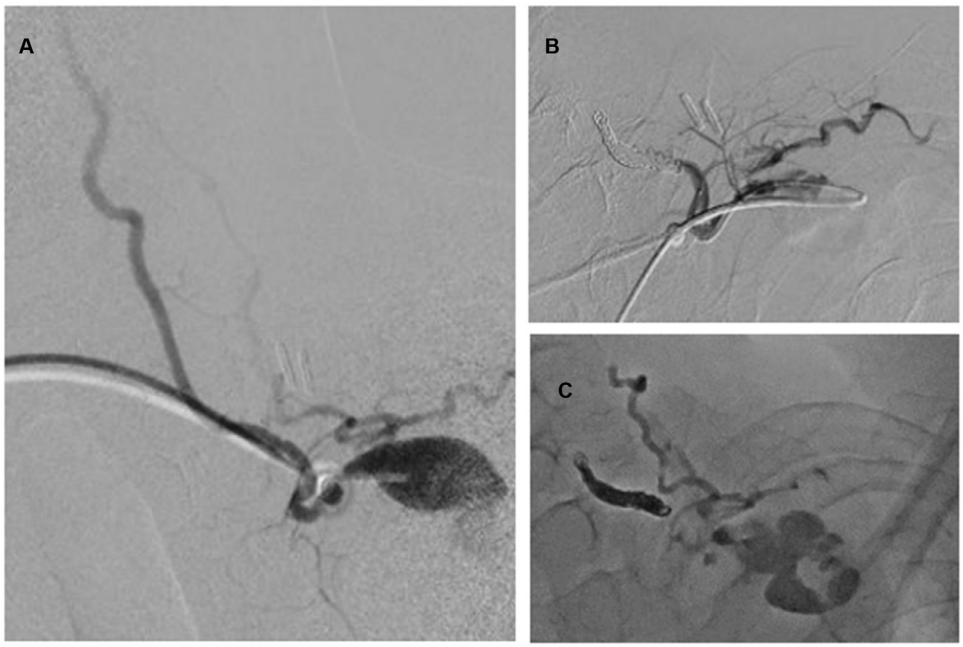

Fig. 3 – Images during treatment (DSA (A and B) and DA (C)).

following the upper wall of the aneurysm, and the left vertebral artery was cannulated. We embolized the vertebral artery using appropriately sized detachable coils (Target XL, Stryker: 2, IDC, Boston Scientific: 2) (Fig. 3B). Subsequently, we slowly injected 33% of NBCA via a 4-Fr hook catheter. Within a short time, the cast stagnated proximal to the left deep costocervical artery. We continued injecting NBCA to fill the aneurysm. After injecting 6 mL of NBCA, we removed the 4-Fr catheter using a sheath introducer (Fig. 3C). After completing all procedures, we confirmed the lipiodol deposit area and the absence of bleeding on interventional radiology-CT. Furthermore, we confirmed the disappearance of blood flow in the sac using color Doppler US (Fig. 4). On the following day of the treatment, on X-ray, we confirmed that the coils and glue were at a suitable location without migration (Fig. 5). Furthermore, contrast-enhanced magnetic resonance imaging 1 week after treatment confirmed no blood flow in the aneurysm. Post-treatment, the patient experienced mild back and left arm pain and numbness for several days, which resolved spontaneously. No other complications were associated with the procedure. The patient has continued taking pregabalin at the same dose as before treatment for left back pain; however, it gradually diminished.

Follow-up CT 2 years after treatment showed that the aneurysm remained under control without re-enlarging.

Discussion

The incidence of subclavian artery aneurysms is rare, and subclavian artery aneurysms in patients with Marfan syn-

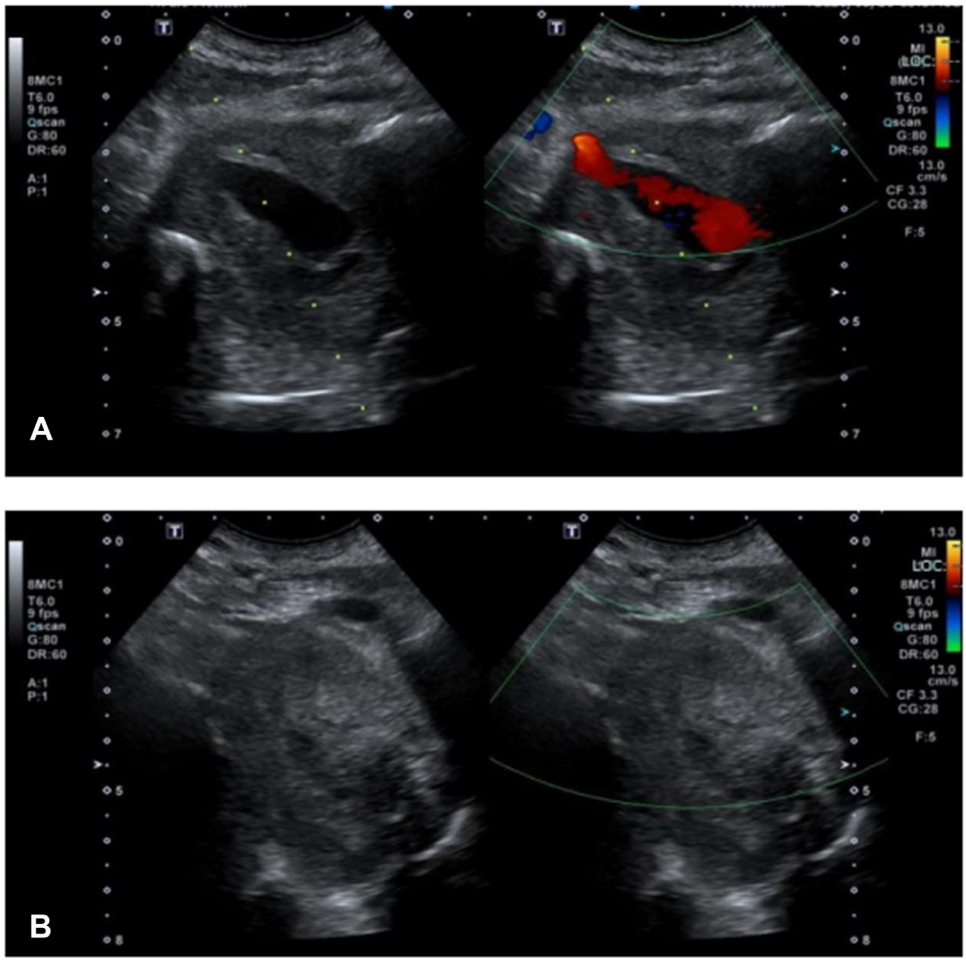

Fig. 4 – Ultrasound images before and after treatment (before (A) and after (B)).

drome are uncommon. However, subclavian artery aneurysms are occasionally related to connective tissue diseases, including Marfan syndrome and Ehlers–Danlos syndrome [1,2]. Subclavian artery aneurysms are located primarily in the proximal segment of the subclavian artery, as in this case. These aneurysms are often managed by surgical treatment with median sternotomy or thoracotomy [3]. Recently, few reports have focused on less invasive treatments, such as endovascular procedures, including the use of endografts, to avoid such invasive surgery [3,4]. However, occasionally, we may encounter cases where both interventions are challenging because of various factors, including the inability to reach an aneurysm through a safe route or postoperative adhesion.

In this case, we observed a left subclavian artery aneurysm secondary to Marfan syndrome and type II endoleaks from the vertebral artery due to changes in blood flow caused by previous surgeries. First, we considered embolization by endovascular treatment but could not establish access via the femoral or radial artery. Although some reports have described embolization of head and neck aneurysms after establishing access via the vertebral artery, most have warned of a high risk of hemorrhage, hematoma, or vasospasm [5–7]. Thus, we considered DSP to be the most appropriate option. Several reports have described direct percutaneous sac injection in type II endoleaks after endovascular aortic repair [8]. This case's clinical condition was similar to previously described cases in which researchers used NBCA to iodized oil ratio of 50% or 33% to embolize AAA [9,10]. We used 33% NBCA to prevent cast deviation at the area distal to the branch, and an excellent therapeutic effect was confirmed. This ratio appears adequate.

Because of multiple contraindications for open surgery and intravascular embolization, this case proceeded successfully using a strategy that involved DSP of the subclavian artery aneurysm. Complete blood flow cessation to the aneurysm was confirmed post-treatment using coils and glue. Remarkably, we could prevent reenlargement of the aneurysm by completely blocking blood flow. Particularly, although embolization with NBCA alone appeared to provide a reliable therapeutic effect on the aneurysm, a combination of coil embolization appeared to be the best choice for reducing the risk of spinal cord infarction and other complications.

In this case, we embolized vertebral and costocervical arteries to block the route into the aneurysm; the former diverges a cervical branch, and one branch of the latter also diverges another branch toward the cervical spinal cord. Therefore, completely embolizing only target vessels using coils or NBCA adjusted to an appropriate concentration was re-

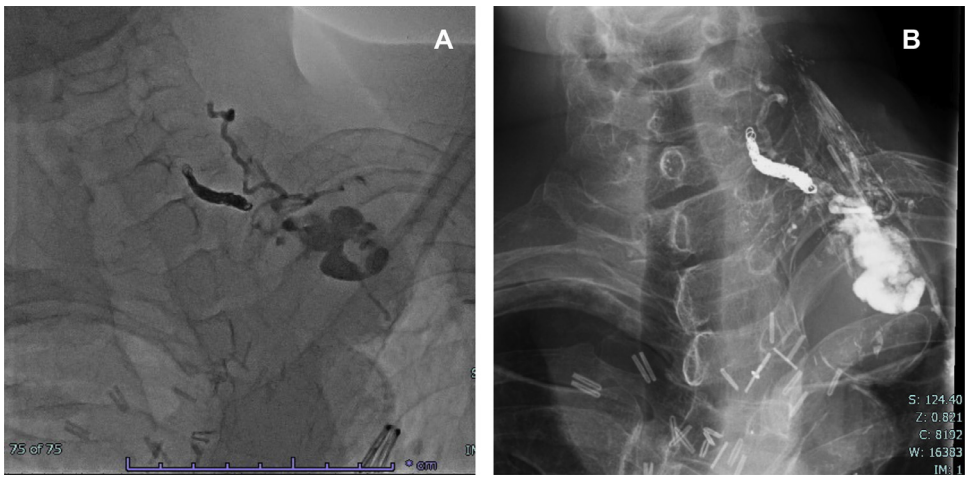

Fig. 5 – X-ray image just after the procedure (A) and X-ray of the next day (B).

quired. We completed the procedure with extreme caution to this point, that is, embolizing the vertebral artery with coils and costocervical artery with 33% NBCA, while observing the movement of the cast in realtime under X-ray. We are convinced that such a careful procedure yielded a highly positive result. In our case, we reduced the risk of aneurysm rupture. Aneurysm cases where the patient has a high risk of surgery and endovascular treatment are considered rare; however, the possibility that physicians encounter such cases is not very low.

In conclusion, although only a few reports have mentioned percutaneous embolization of subclavian artery aneurysms using the direct puncture technique, it is an effective and safe therapeutic option for cases in which surgical approach and transarterial access routes are limited. Furthermore, to our knowledge, this is the first report of embolizing subclavian artery aneurysms using a combination of coils and NBCA, possibly leading to novel treatment options.

Patient consent

We obtained written informed consent from the patient for this report.

REFERENCES

[1] Ito H, Kurazumi H, Sakata K, Kobayashi Y. Successful surgical treatment of right subclavian artery aneurysm with a hypoplastic left vertebral artery using temporary cerebral perfusion to prevent cerebral ischemia. Ann Vasc Dis 2011;4:60–3. doi:10.3400/avd.cr.10.00006.

[2] González JM, García BA, Lebrun JM, Docampo MM. Combined surgery for the treatment of bilateral subclavian artery aneurysm in Marfan syndrome. J Vasc Surg 2007;45:180–2. doi:10.1016/j.jvs.2006.08.045.

[3] Morimoto K, Matsuda H, Fukuda T, Iba H, Tanaka H, Sasaki H, et al. Hybrid repair of proximal subclavian artery aneurysm. Ann Vasc Dis 2015;8:87–92. doi:10.3400/avd.oa.15-00006.

[4] El Khoury R, Greenspahn BR, Jacobs CE, White JV, Schwartz LB. Minimally invasive repair of left subclavian artery aneurysm. Cardiovasc Revasc Med 2020;21:165–7. doi:10.1016/j.carrev.2019.05.013.

[5] Blanc R, Piotin M, Mounayer C, Spelle L, Moret J. Direct cervical arterial access for intracranial endovascular treatment. Neuroradiology 2006;48:925–9. doi:10.1007/s00234-006-0157-1.

[6] Desai JA, Almekhlafi MA, Hill MD, Goyal M, Eesa M. Ultrasound guided V3 segment vertebral artery direct percutaneous puncture for basilar artery mechanical thrombectomy in acute stroke: a technical report. BMJ Case Rep 2013;2013:bcr2012010601. doi:10.1136/bcr-2012-010601.

[7] Nakatsukasa M, Kitamura Y, Mayanagi K. Coil embolization of a ruptured basilar aneurysm with direct percutaneous puncture of the cervical vertebral artery: a case report. Surg Cerebral Stroke 2015;43:477–81. doi:10.2335/scs.43.477.

[8] Uthoff H, Katzen BT, Gandhi R, Peña CS, Benenati JF, Geisbüsch P. Direct percutaneous sac injection for postoperative endoleak treatment after endovascular aortic aneurysm repair. J Vasc Surg 2012;56:965–72. doi:10.1016/j.jvs.2012.03.269.

[9] Park YS, Do YS, Park HS, Park KB, Kim DI. Experience of direct percutaneous sac injection in type II endoleak using cone beam computed tomography. Ann Surg Treat Res 2015;88:232–5. doi:10.4174/astr.2015.88.4.232.

[10] Ugajin A, Fujii H, Nakamura H, Fujita A, Sasaki T, Mato N, et al. Transcatheter proximal coil blocking with n-Butyl-2-cyanoacrylate injection via the pulmonary artery alone for Rasmussen's aneurysm. Case Rep Radiol 2019;2019:1725238. doi:10.1155/2019/1725238.