



Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Successful hybrid TEVAR for distal anastomotic pseudoaneurysm and coarctation following previous palliative left subclavian artery to descending aorta bypass: A case report

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ARTICLE INFO

Article history:

Received 29 June 2020

Received in revised form 24 July 2020

Accepted 3 August 2020

Available online 15 August 2020

Keywords:

Coarctation of the aorta

Anastomotic pseudoaneurysm

Thoracic endovascular aortic repair

ABSTRACT

INTRODUCTION: Anastomotic pseudoaneurysm is one of the most common but catastrophic complications in coarctation of the aorta (CoA); this is equally true even if the initial surgery is not directly related to the coarctation. Redo open heart surgery is usually required for the pseudoaneurysm; however, redo surgery remains challenging with high morbidity and mortality rates.

PRESENTATION OF CASE: A 38-year-old woman with CoA, who had undergone left subclavian artery (LSCA) to descending aorta bypass 21 years prior, was referred to us for the treatment of distal anastomotic pseudoaneurysm. Zone 2 thoracic endovascular aortic repair (TEVAR) with LSCA debranching was performed to exclude the distal anastomotic pseudoaneurysm and expand the CoA using a stent graft. The patient completely recovered and resumed work without delay.

DISCUSSION: In patients who require surgical treatment for both pseudoaneurysm and CoA, hybrid TEVAR can be an alternative surgical option instead of conventional open repair.

CONCLUSION: TEVAR for concomitant pseudoaneurysm and native CoA is feasible and less invasive, especially for young patients who have to resume work early after surgery.

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1. Introduction

Coarctation of the aorta (CoA) is a birth defect in which a part of the aorta is narrower than usual. Anastomotic pseudoaneurysm is one of the common late complications in patients who undergo surgical treatment for CoA itself [1–4], with a reported incidence rate ranging between 11%–24% [1,2]. Similarly, anastomotic pseudoaneurysm is a complication in those who have undergone palliative surgery for CoA [5]. Due to spontaneous rupture risk, redo open surgery is usually required for the pseudoaneurysm [1–3]; however, it remains challenging and is associated with high morbidity and mortality rates [1,2].

In patients with post CoA pseudoaneurysm after palliative surgery, such as extra-anatomical bypass from ascending aorta to the descending aorta, additional surgical repairs are essential for both the anastomotic pseudoaneurysm and native CoA. In these patients, redo open surgery may be more difficult compared to those who had initially only undergone radical reconstruction.

Abbreviations: CoA, coarctation of the aorta; LSCA, left subclavian artery; TEVAR, thoracic endovascular aortic repair; ABI, ankle-brachial index; CT, computed tomography; PG, pressure gradient.

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<https://doi.org/10.1016/j.ijscr.2020.08.007>

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Here, we describe a successful concomitant endovascular repair of a distal anastomotic pseudoaneurysm after bypass surgery from the left subclavian artery (LSCA) to the descending aorta and native CoA. We report this case in line with the SCARE criteria [6].

2. Presentation of case

A 38-year-old woman with Turner syndrome complicated by CoA was referred to our department for the surgical treatment of an anastomotic pseudoaneurysm at the descending aorta. While investigating the cause of the delay in the manifestation of secondary sexual characteristics, the patient was diagnosed with Turner syndrome 21 years prior. Subsequently, computed tomography (CT) imaging revealed the CoA, and she had undergone bypass surgery from the LSCA to the descending aorta (using an 8-mm prosthetic graft).

On admission, she developed slight leg fatigue. The right and left preoperative ankle-brachial index (ABI) was 0.90 and 0.89, respectively. Enhanced CT showed a pseudoaneurysm in the anastomotic site of the descending aorta measuring 58 mm in diameter. The diameter of the CoA lesion was 7.5 mm (Fig. 1). The arterial pressure was 98/56 (73) mmHg and 76/54 (64) mmHg in the aorta proximal and distal to the CoA, respectively (Fig. 2).

Considering her relatively young age, we initially planned a radical redo procedure, such as a resection of the CoA and anastomotic

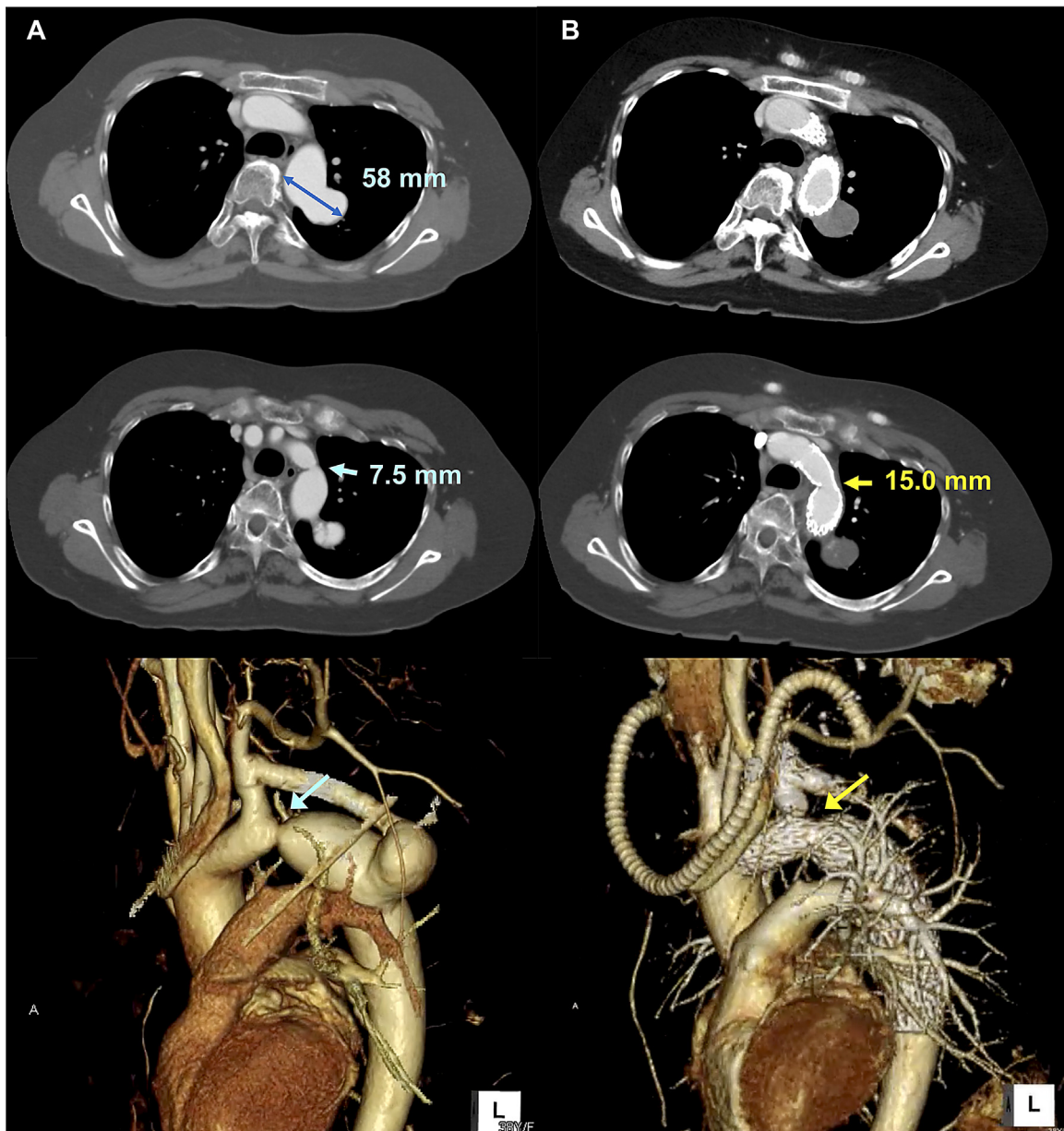


Fig. 1. Enhanced computed tomography (CT) imaging. A: Preoperative CT findings show that the anastomotic pseudoaneurysm was 58.0 mm in size. The diameter of the coarctation of the aorta (CoA) is 7.5 mm (blue arrows). B: On postoperative CT, CoA expanded to 15.0 mm in size (yellow arrows). No significant endoleakages were observed in the pseudoaneurysm.

pseudoaneurysm, followed by descending graft replacement with LSCA reconstruction. To resect the CoA and remove the pseudoaneurysm, graft replacement of the distal arch and descending aorta with redo left wide thoracotomy was required. Removal of the previous bypass graft was equally required. However, as the patient wished to return to work early, we decided to perform zone 2 debranching thoracic endovascular aortic repair (TEVAR). Regarding the reconstruction of LSCA before zone 2 TEVAR, we selected axillo-axillary bypass instead of the left common carotid artery to LSCA bypass, since the patient had a short and webbed neck, which is one of the characteristic physical features in Turner syndrome.

Hybrid TEVAR was performed in a hybrid operative suite. Under general anaesthesia, an axillo-axillary bypass using a ringed 6 mm expanded polytetrafluoroethylene graft (PROPATEN 6 mm, WL GORE & Associates, Inc., USA) was established prior to TEVAR. An 8 Fr sheath was inserted through the exposed LSCA. Through the 8 Fr sheath, the orifice of LSCA and the anastomotic site of the

bypass graft were occluded using an occlusion balloon. The pressure gradient (PG) across CoA was approximately 30 mmHg after occlusion of the previous graft. A 20 Fr sheath was inserted through the exposed right common femoral artery. A stent graft measuring 21 mm in diameter and 100 mm in length (Conformable GORE TAG) was delivered through the CoA, and the proximal endograft was deployed. Subsequently, a stent graft measuring 26 mm in diameter and 100 mm in length (Conformable GORE TAG) was deployed to the distal aortic arch, which was distal to the CoA lesion. After touching up at the distal and proximal edges of the stent graft using a balloon catheter (GORE® Tri-Lobe Balloon catheter), the CoA was carefully expanded using the same balloon catheter. Postoperative PG improved to 0 mmHg. Post ballooning, the LSCA and old bypass graft were occluded using 3 embolisation devices (2 Vascular-plugs, 14 mm; and a RUBY-coil, 35 cm, 8 mm) to prevent type 2 endoleakage (Fig. 3). The final aortography showed no endoleakage. The

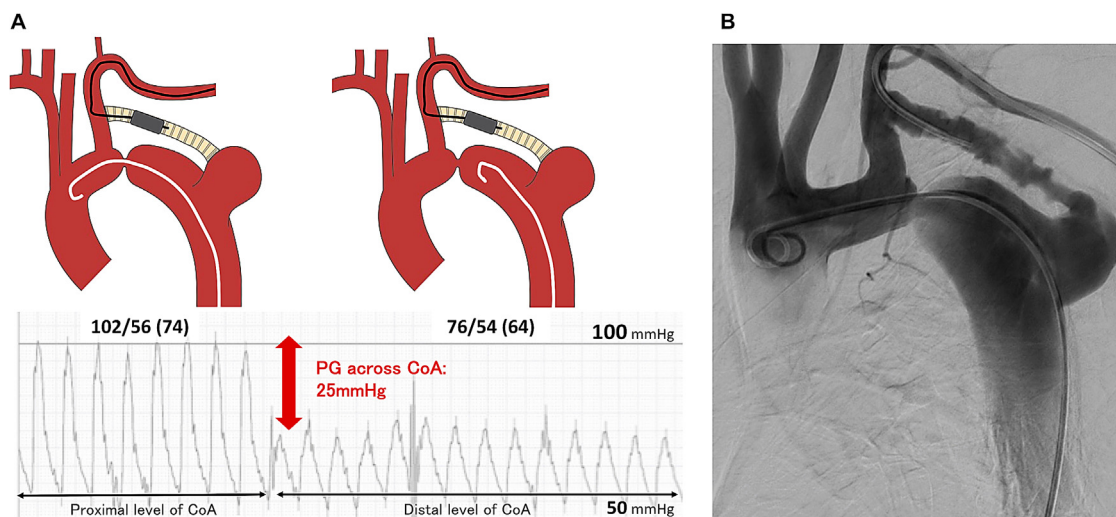


Fig. 2. Aortic pressure study and angiography. A: Under occlusion of the bypass graft, the pressure gradient across CoA was approximately 25 mmHg. B: Preoperative angiography showed blood flow from CoA and prosthetic graft to pseudoaneurysm.

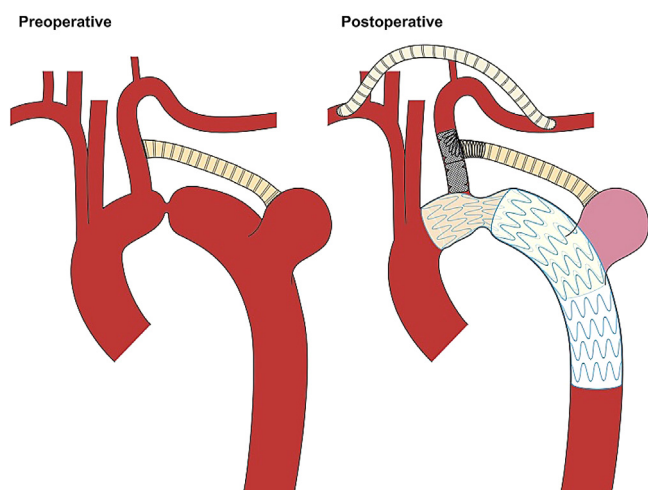


Fig. 3. Pre- and postoperative schema in the present case.

total operation time was 182 min, and no blood transfusion was required.

The patient was extubated in the operating room and showed an uneventful recovery without any neurologic deterioration. Postoperative ABIs of the right and left sides were 1.02 and 1.05, respectively. Postoperative enhanced CT imaging showed no endoleakages in the pseudoaneurysm, and the diameter of CoA had expanded from 7.5 mm preoperatively to 15.0 mm postoperatively (Fig. 1B). The patient was discharged on postoperative day 8 and returned to work on postoperative day 18.

3. Discussion

After various open surgeries for CoA, redo open surgery is usually the first-line treatment for anastomotic pseudoaneurysms; however, it carries an increased risk of morbidity and mortality due to the densely adherent pulmonary tissue, which increases the risk of bleeding, phrenic, or recurrent nerve palsy, and aortobronchial fistula during reoperation [2,5]. Several reports have shown that endovascular repair for anastomotic pseudoaneurysm can contribute to the improvement in early outcomes compared to conventional surgery [1,2]. In this case, owing to previous palliative treatment by LSCA-descending aorta bypass, surgical repair was

necessary for both the anastomotic pseudoaneurysm and native CoA. Therefore, the operative risk was greater in this case than in isolated pseudoaneurysms or CoA. Although the reported numbers of cases of TEVAR for anastomotic pseudoaneurysm in CoA are limited and long-term outcomes remain uncertain [1,2], hybrid TEVAR should be fully considered a treatment option, especially for patients requiring concomitant surgery for pseudoaneurysm and native CoA. To the best of our knowledge, this is the first case to perform hybrid TEVAR for concomitant anastomotic pseudoaneurysm and CoA.

In addition, the patient had Turner syndrome, which is characterised by the lack of an X chromosome. Turner syndrome is a representative genetic disease associated with aortic pathologies, including CoA, bicuspid aortic valve, and dissection or aortic aneurysmal formation of the thoracic aorta [4,7]. In a CoA patient with this genetic background, the potential risk of anastomotic pseudoaneurysm recurrence may be higher than that in CoA patients without Turner syndrome [7]. Given these facts, TEVAR might be more suitable for pseudoaneurysm in patients with Turner syndrome than conventional open repair because aortic anastomosis can be avoided using TEVAR. However, the potential risk of stent graft implantation to the aortic wall in these patients remains unknown. Therefore, further assessment of the durability of stent grafts with periodic CT follow-up should be performed over a long period.

4. Conclusion

In conclusion, this case suggests that TEVAR for native CoA is a useful and feasible surgical option, especially for young patients who have to resume work early after surgery.

Declaration of Competing Interest

None.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Ethical approval

This study was approved by the ethical committee of our hospital.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Takasumi Goto wrote the manuscript, performed the procedures, and collected all clinical data. Hiruyuki Nishi supervised this study, proofread this manuscript, and performed all the procedures.

Mutsunori Kitahara, Satoshi Sakakibara, and Yumi Kakizawa assisted in the clinical management of this patient.

Registration of research studies

This study is a case report and not a research study.

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Guarantor

Hiruyuki Nishi is fully responsible for this case report.

Availability of data and materials

All data are available from the corresponding author just on reasonable request.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Acknowledgment

We appreciate Dr Yuji Tominaga, who is a surgeon of congenital heart disease in Osaka University, for advising surgical therapy of CoA.

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