Late open conversion after endovascular treatment for the coarctation of aorta in adult due to restenosis with thrombus

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ABSTRACT
A 22-year-old man was referred to our hospital for rib notching found on a radiograph and hypertension. Computed tomography revealed coarctation of the descending aortic isthmus. Because he refused open surgery, endovascular treatment was performed. The 2-year follow-up computed tomography scan showed infolding of the stent graft and thrombus formation. He had presented with intermittent claudication; therefore, graft interposition was performed. Endovascular surgery plays an important role in the treatment of coarctation of the aorta. However, insufficient dilatation can lead to restenosis accompanied by thrombus formation, and excess ballooning can cause aortic wall injury. Careful performance of the procedure and close postoperative follow-up are essential. (J Vasc Surg Cases Innov Tech 2022;8:338-44.)

Keywords: Coarctation of the aorta; Endovascular treatment; Late open conversion

Coarctation of the aorta (CoA) is a congenital disease that causes narrowing and thickening of the intima in the distal aortic arch around the ductus arteriosus, which can lead to hypertension in the upper body.1,2 If it is left untreated, it can cause atherosclerotic disease in the upper body, including cerebral vascular disease and myocardial infarction at a young age.3-5

Along with surgical repair, endovascular therapy for CoA has been performed for a long time.6-8 However, a number of studies have reported cases of restenosis due to intimal regrowth through the stent mesh and aortic wall injury caused by the device itself when balloon angioplasty and bare metal stenting were used.9 Although early- and long-term outcomes have improved dramatically with the development of covered stents in recent years,10,11 restenosis requiring reintervention can still occur in rare cases with extensive endothelial regrowth.12 Reintervention can be usually performed with repeated endovascular treatment, including additional stenting or balloon redilation, and rarely requires open conversion. We have reported a case of restenosis after endovascular treatment that had required late open conversion. The patient and his parents had provided written informed consent for the report of his case details and imaging studies.

CASE REPORT
A 22-year-old man, with no pertinent medical history, was referred to our hospital because of abnormal radiograph and hypertension. Computed tomography revealed coarctation of the descending aortic isthmus. Because he refused open surgery, endovascular treatment was performed. The 2-year follow-up computed tomography scan showed infolding of the stent graft and thrombus formation. He had presented with intermittent claudication; therefore, graft interposition was performed. Endovascular surgery plays an important role in the treatment of coarctation of the aorta. However, insufficient dilatation can lead to restenosis accompanied by thrombus formation, and excess ballooning can cause aortic wall injury. Careful performance of the procedure and close postoperative follow-up are essential. (J Vasc Surg Cases Innov Tech 2022;8:338-44.)

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Author conflict of interest: none.

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The editors and reviewers of this article have no relevant financial relationships to disclose per the Journal policy that requires reviewers to decline review of any manuscript for which they may have a conflict of interest.

https://doi.org/10.1016/j.jvscit.2022.04.008

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because he had no symptoms. At 2 years postoperatively, his ABI had decreased further to 0.6, and refractory hypertension at the upper extremity had developed. The patient complained of fatigue on exertion and agreed to undergo reoperation. Open graft interposition was performed under the partial cardiopulmonary bypass through the left fourth intercostal space thoracotomy (Fig 4). When the stenotic lesion was incised, the lumen was severely collapsed and was almost completely filled with organized thrombus. The stent graft was transected and removed, leaving the distal two stents in the native arterial wall because the stents were highly adherent to the arterial wall and no thrombus was inside. The stenosed lesion was replaced with a J Graft Shield Neo 18-mm straight vascular graft (Japan Lifeline Co, Ltd, Tokyo, Japan). For spinal cord protection, the mean blood pressure was maintained at >100 mm Hg until the patient was discharged from the intensive care unit. The continuous infusion of low-dose naloxone was continued (1 μg/kg/h) until postoperative day 3. Cerebrospinal fluid drainage was not used. The postoperative course was uneventful, and he was discharged from the hospital on postoperative day 5. One year later, his ABI had recovered to 0.99/1.06, and he had not presented with signs of recurrent stenosis.

**DISCUSSION**

**Restenosis after stenting.** Restenosis after endovascular treatment of CoA occurs in 0% to 25% of cases, and its onset varies from 6 months to nearly 10 years postoperatively. The pathology of CoA is characterized by the loss of elasticity of the arterial wall, intimal thickening, and secondary fibrotic changes in the intima. The loss of elasticity results from tears in the inner elastic plate and the reduction of elastic fibers at the level of the tunica media. Because these degenerative processes can develop even after endovascular treatment, intimal regrowth through the stent mesh can occur when treated with bare metal stents, leading to restenosis. Covered stents, especially balloon-expandable stents, are recommended considering the stronger radial force compared with self-expandable covered stents. Balloon-expandable stents were not available owing to insurance restraints in Japan. In the present patient, CT had shown thrombus formation in the stenosed lesion. Therefore, we chose open surgery with graft interposition. The unique aspect of our findings was that thrombus had formed in the stent graft, which did not allow for endovascular reintervention. Disturbed rheology of post-stenotic turbulent blood flow has a significant effects on the adhesion and activation mechanisms used by platelets with high wall shear stress and playing an important role in accelerating platelet activation and thrombus growth. Given this mechanism, thrombus formation is a possible consequence for restenosis after stenting; therefore, reintervention should be well discussed before thrombus formation occurs because it will limit the available treatment options.

**Fig 1.** Computed tomography (CT) scan showing stenosis and kinking of the descending aortic isthmus and screw-like development of the internal thoracic and intercostal arteries.

**Fig 2.** Final angiogram after stent deployment. Although the image still showed an hourglass shape, no delay was present in the contrast flow; therefore, it was considered acceptable.
Aortic wall injury. Aortic wall injury (AWI), including aortic rupture and dissection, is a rare, but important, complication of endovascular treatment, which can lead to a fatal outcome. When using a covered stent, balloon dilation to more than three times the diameter of the coarctation has been suggested as constituting a risk of AWI. Post-dilation to avoid migration can also increase the risk of rupture. Dilating the stent to smaller than the diameter of the distal landing zone has been effective in reducing aortic coarctation and decreasing the risk of rupture. In the present case, we were concerned about the occurrence of aortic rupture resulting from strong balloon dilation. Thus, we used the minimum post-dilation necessary to remove the stenosis. Endovascular treatment plays an important role in the treatment of CoA because of its minimal invasiveness. However, emergency treatment will be required once an AWI has occurred, and the mortality rate is high. Thus, surgeons should be familiar with safe and effective techniques to avoid complications.

Review of the literature. A semi-systematic review was conducted for a better understanding of stent selection for CoA. All randomized controlled trials and observational studies that had examined the outcomes of endovascular stenting for CoA were identified using a two-level strategy. First, the MEDLINE database was searched for studies reported from 2000 through March 9, 2022 using PubMed. Second, the relevant studies were identified through a manual search of secondary sources, including references from the initially identified studies, reviews, and commentaries. The search terms included “adult coarctation of aorta” or “adult aortic coarctation” or “adult CoA” and “endovascular” or “TEVAR” or “transcatheter.” Our search identified 27 eligible studies from 528 records, including 2 randomized controlled studies and 25 observational studies, with 2022 patients enrolled. The patient characteristics and outcomes are summarized in the Table. The mean age of the patients across all the studies was 23.1 years. The pressure gradient had improved from 41.6 to 4.1 mm Hg, and the stenosis had been dilated to 14.6 mm.

Because our review included both planned and unplanned procedures for redilation and/or restenting,
### Table. Characteristics of included studies

| Investigator   | Study design | Follow- up, months | Patients, Age, n | Pathophysiology | Devices                               | PC, mm Hg | Diameter, mm | Outcomes                                                                 |
|----------------|--------------|--------------------|------------------|-----------------|---------------------------------------|-----------|--------------|--------------------------------------------------------------------------|
| Macdonald et al, 2003 | R            | 36                 | 15               | 25 Native, n = 10, recurrent, n = 5 | Palmaz, n = 8; Memotherm, n = 7 | 42.5      | NA           | NA Redilation for restenosis, n = 1                                    |
| Tyagi et al, 2003     | R            | 41                 | 21               | 28.6 Native, n = 21 | Palmaz, n = 5; Memotherm, n = 16   | 68.4      | 8.3          | 4.3 13.9 Redilation for restenosis, n = 1                               |
| Johnston et al, 2004  | R            | 18                 | 32               | 15.2 Native, n = 9, recurrent, n = 23 | Palmaz, n = 31; Intratherapeutics, n = 1 | 31        | 1.7          | 8.1 13.5 Aortic dissection, n = 1; redilation for restenosis, n = 8     |
| Sulik-Gajda et al, 2017| R            | 104.4              | 26               | 23 Recurrent, n = 26 | Palmaz, n = 7; Andrasent, n = 7, bare CP, n = 2; Smart, n = 1; covered CP, n = 2 | 40.5      | 13.1         | 7.5 13.1 Redilation for restenosis, n = 10; aortic aneurysm, n = 1     |

(Continued on next page)
it was difficult to directly compare the vulnerability to restenosis with the use of self-expandable or balloon-expandable or covered or bare metal stents. Early studies in our review reported no differences in performance between the self- and balloon-expandable devices. However, Butera et al reported good procedural outcomes with balloon-expandable stents in patients with recurrent CoA.

The superiority of covered stents compared with bare metal stents has not yet been determined. Even in our review, the AWI rate reported by the studies that had used only covered stents ranged from 1.8% to 11.8%. However, Butera et al compared the results of 71 bare metal stents and 72 covered stents and reported no AWI with the covered stents. In contrast, AWI had occurred in eight cases with bare metal stents. Covered stents prevented postprocedural pseudoaneurysm formation and might enable safer bailout procedures because the bleeding point is covered even if aortic

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**Table.** Continued.

| Investigator | Study design | Follow-up months | Patients, n | Age, years | Pathophysiology | Devices | PG, mm Hg | Diameter, mm | Outcomes |
|--------------|--------------|------------------|-------------|------------|----------------|---------|-----------|-------------|----------|
| Fitozzi et al, 2018 | R | 35 | 169 | 26 | Native, n = 169 | Bare CP, n = 98; Genesis, n = 2; Sinus XL, n = 69 | 57.9 | 1.9 | 3.7 | 13.5 | Aortic dissection, n = 4; restenting for restenosis, n = 4; redilation for restenosis, n = 12; surgery for restenosis, n = 2 |
| Erben et al, 2019 | R | 38 | 82 | 44 | Native, n = 32; recurrent, n = 50 | Palmaz, n = 9; covered CP, n = 47; stent graft; n = 25 | 25.7 | 4.6 | 97 | 18.8 | Aortic rupture, n = 2; type I endoleak, n = 1; aortic dissection, n = 3; restenting for restenosis, n = 3; redilation for restenosis, n = 5; stent graft for pseudoaneurysm, n = 1 |
| Hatoum et al, 2020 | R | 12 | 20 | 18.4 | Native, n = 15; recurrent, n = 5 | Bare CP, n = 10; Genesis, n = 3; Formula, n = 1; covered CP, n = 3; Advanta VI2, n = 3 | 50 | 0.30 | N/A | N/A | Redilation for restenosis, n = 4 |
| Promphavan et al, 2020 | R | 10 | 2 | 7.7 | Native, n = 8; recurrent, n = 4 | BeGraft, n = 12 | 25 | 2 | 6 | 11.6 | Redilation for restenosis, n = 2; pseudoaneurysm, n = 1 |
| Sasikumar et al, 2020 | R | 57 | 45 | 25.8 | Native, n = 35; recurrent, n = 10 | Bare CP, n = 17; andra, n = 6; Palmaz, n = 2; covered CP, n = 7; Advanta VI2, n = 13 | 45.8 | 4.3 | N/A | N/A | Redilation for restenosis, n = 6 |
| Egbe et al, 2021 | R | 46 | 44 | 26 | Native, n = 9; recurrent, n = 35 | Palmaz, n = 2; Genesis, n = 4; IntraStent Max LD, n = 25; bare CP, n = 4; covered CP, n = 9 | 22 | 3 | 9 | 17 | Redilation for restenosis, n = 2; restenting for restenosis, n = 1 |
| Homsi et al, 2021 | R | 48 | 18 | 21.2 | Native, n = 16; recurrent, n = 2 | Palmaz, n = 5; bare CP, n = 6; covered CP, n = 4; Advanta VI2, n = 2; BeGraft, n = 1 | 49.7 | 6.3 | 51 | 12.6 | Redilation for restenosis, n = 4; surgery for pseudoaneurysm, n = 1 |
| Nagendra et al, 2021 | R | 64 | 56 | 33.6 | Native, n = 47; recurrent, n = 9 | Genesis, n = 10; Intrastent Max LD, n = 2; covered CP, n = 37 | 28 | 5.3 | 8.2 | NA | Aortic dissection, n = 1; redilation for restenosis, n = 7 |
| Yilmazer et al, 2021 | R | 14 | 11 | 14 | Native, n = 5; recurrent, n = 6 | BeGraft, n = 11 | 30 | 5 | 71 | 15 | None |
| Sadeghipour et al, 2022 | RCT | 12 | 92 | 30 | Native, n = 92 | Bare CP, n = 46; Sinus-XL, n = 46 | 63.9 | 1.5 | NA | NA | Aortic dissection, n = 1; stent graft for pseudoaneurysm, n = 1; restenting for restenosis, n = 1 |

CP, Cheatham-Platinum; NA, not applicable; P, prospective; PG, pressure gradient; R, retrospective; RCT, randomized controlled trial.
rupture occurs.26 Butera et al29 also demonstrated that no difference was found in the incidence of restenosis due to neointimal hyperplasia (n = 7 with bare metal stents [10%] vs n = 5 with covered stents [7%]). No neointimal hyperplasia was observed in adult patients treated with covered stents.

In the present case, we did not consider the use of a bare metal stent because the coarctation was severely kinked and a risk of AWI was present. If available, balloon-expandable covered stents were a potentially favorable option owing to resisting buckling due to external pressure of restenosis. The only available covered stent within the health insurance coverage in Japan was the self-expandable thoracic endovascular aortic repair device. Therefore, it might have been a possible option to deploy an additional balloon-expandable bare stent, including the Palmaz extra-large stent (Cordis Europa NV, Roden, Netherlands) inside to reinforce the radial strength. Because the VBX stent (W. L. Gore & Associates, Inc, Newark, DE), a balloon-expandable covered stent, including the Palmaz Cheatham-platinum stents for endovascular treatment of patients with native post-ductal aortic coarctation: immediate and intermediate-term results. JACC Cardiovasc Interv 2014;7:416-23.

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Submitted Jan 26, 2022; accepted Apr 4, 2022.