

Case report

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Intravascular treatment of left subclavian artery aneurysm coexisting with aortic coarctation in an adult patient

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Abstract: Coexistence of aortic coarctation with aneurysm of subclavian artery is a uncommon situation and may require unusual treatment in patients.

A 40-year-old patient diagnosed incidentally with left subclavian artery aneurysm coexisting with aortic coarctation. Patient was initially referred for hybrid treatment. Initially ostium of the left subclavian artery was covered with a stent-graft. Over a 30-month follow-up period aneurysm became thrombosed all the way up to the ostium of internal mammary artery. The patient did not present with neurological symptoms or signs of upper limb ischemia. Taking into consideration good blood supply to the axillary artery via reversed blood flow in the thyrocervical trunk, hence we decided not to proceed with cervicoaxillary bypass grafting.

Implantation stent-graft into aorta coarctation with covering axillary artery is proper way of treatment and may need no other surgical procedures.

Keywords: Aortic coarctation; Coarctation-associated aneurysms; Coarctation in adults; Coarctation complication

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1 Introduction

Incidence of aortic coarctation is estimated at 4/100 000 of live births. In 5% to 8% of patients coarctation appears as a component of various congenital heart conditions [1]. When aortic coarctation is significant and causes a perfusion deficit in the lower part of the body, surgical treatment is required. Currently, beside classical surgery, treatment methods include intravascular and hybrid procedures, as well as extra-anatomical grafts.

2 Methods

Intravascular intervention was performed on a 56-year-old man, who had undergone suspected for arterial hypertension and suspected coronary artery disease 4 years before. A left subclavian artery aneurysm was diagnosed incidentally at that time. CT attenuated angiography revealed dilatation of the left subclavian artery orifice to 25 mm and mild aortic coarctation. Patient was hospitalized due to exacerbation of coronary disease symptoms. He underwent coronarography and coronaroplasty with implantation of a stent into the left coronary artery. Existence of aortic arch pathology was confirmed during the procedure and patient was referred to a vascular surgeon. He had a chest angio-CT performed at the vascular surgery outpatient clinic. Imaging revealed mild aortic coarctation and an aneurysm of the ostium of the left subclavian artery, about 37 mm in diameter (Fig. 1). Since the size of the aneurysm increased by 12 mm over a period of 4 years, patient was referred for intravascular treatment. A short stent-graft (TAG GORE) was implanted into the descending aorta, entirely covering the ostium of the left subclavian artery (Fig. 2 and 3). A small, late retrograde flow into the aneurysmal sac was left to thrombose. No ischemic neurological events or significant disruption of left upper limb perfusion were noted during the postoper-

ative period. Considering these circumstances, left-sided cervicoaxillary bypass grafting was not performed.

Follow-up angio-CT examination performed 18 months from implantation of the stent-graft demonstrated complete thrombosis of aneurysmal sac, all the way to the origin of the left internal mammary artery and left verte-

bral artery (Fig. 4). Blood supply to the subclavian artery observed in a dynamic study mainly came from the thyrocervical trunk. Until now, patient has not complained of any symptoms that might result from occlusion of the left subclavian artery at the place where it arises from the aortic arch.

Ethical approval: The research related to human use has been complied with all the relevant national regulations, institutional policies and in accordance the tenets



Figure 1: Left subclavian artery aneurysm. Mild aortic coarctation is visible below its origin.



Figure 2: Intraoperative aortic arch angiography visualizing the origin of the left subclavian artery. Aortic coarctation is also demonstrated



Figure 3: Control angiography following stent-graft implantation into the aortic arch. A complete occlusion of the left subclavian artery ostium.



Figure 4: Follow-up angio-CT performed 30 months after stent-graft implantation.

of the Helsinki Declaration, and has been approved by the authors' institutional review board or equivalent committee.

Informed consent: Informed consent has been obtained from all individuals included in this study.

3 Results

After covering the ostium of the aneurysmal left subclavian artery the absence of neurological symptoms and lack of perfusion disturbances in the left upper limb were observed. Cervicoaxillary bypass grafting was not performed, as the axillary artery received proper blood supply from reversed flow in the thyrocervical trunk. We concluded, that in similar cases, our way of treatment is proper and safe for patient and can be recommended for other surgeons.

4 Discussion

The coarctation does not require treatment if it fails to demonstrate perfusion deficit in the lower body or if it has never been diagnosed (asymptomatic). Per experiences of previous studies, vascular pathologies other than congenital heart defects are associated with coarctation, including aneurysms of the aortic arch, subclavian artery and intercostal arteries [2,3]. Aneurysms of these structures are diagnosed over a range of ages and may require surgical intervention due to continuous growth.

As mentioned, aortic coarctation usually coexists with other congenital heart defects, such as bicuspid aortic valve or interventricular septal defect. Coarctation may be accompanied by defects of the aortic arch, such as aneurysms located in various parts of the arch, or its hypoplasia, aneurysms of the arteries originating from the aortic arch (most commonly the left subclavian artery), intercostal artery aneurysms and aneurysms of the circle of Willis. Aortic coarctation is also frequently diagnosed in Turner syndrome [1]. High-grade coarctation producing severe arterial hypertension in the upper body usually requires repair in childhood. Lower-grade coarctation may exist as an isolated aortic arch defect in about 20% of cases and, if not diagnosed early, present with symptoms in adulthood. Arterial hypertension, vasculitis and atherosclerosis may, with age, lead to significant restriction of blood flow at the site of coarctation, prompting the diagnosis and implementation of appropriate treatment [4,5]. Similar problem

concerns aneurysms that accompany the coarctation. In case of our patient, size of the left subclavian artery aneurysm increased by 30% over a 4-year period, compelling the therapeutic team to perform surgery. It also provides the evidence for necessary monitoring of patients who had been treated surgically in childhood due to coarctation, as well as those with incidental finding of aortic coarctation. The first group of surgically treated patients often requires re-intervention; thus, periodic follow-up is necessary. The second group should be monitored due to possible enlargement of coexisting aneurysms and development of life-threatening conditions [6]. In the recent years, significant progress took place in the management of both primary and recurrent coarctations in pediatric as well as adult population due to development of intravascular techniques and stenting [7]. Introduction of stent-grafting in treatment of various aneurysms allowed for application of these techniques in a broad range of vascular pathologies associated with aortic coarctation [8]. Open surgery and hybrid techniques complement the scope of treatment of coarctation and related pathologies, particularly when open surgery removes both the coarctation itself as well as the coexisting aneurysm [9-11]. Our patient was initially qualified for hybrid treatment involving occlusion of the left subclavian artery ostium with a stent-graft and subsequent left-sided cervicoaxillary bypass grafting.

Conflict of interest statement: Authors state no conflict of interest.

References

- [1] Allen BD, Barker AJ, Gabbour M, Markl M, Rigsby C and Robinson JD. Aortic coarctation augments changes in thoracic aortic hemodynamics in pediatric and young adult patients with bicuspid aortic valve. *Journal of Cardiovascular Magnetic Resonance* 2013;15 (Suppl 1):P300
- [2] Cardoso G, Abecasis M, Anjos R, Marques M, Koukoulis G, Aguiar C, Neves JP. Aortic coarctation repair in the adult. *J Card Surg.*, 2014, 29, 512-518
- [3] Choudhary P, Canniffe C, Jackson DJ, Tanous D, Walsh K, Celermajer DS. Late outcomes in adults with coarctation of the aorta. *Heart*, 2015, 101(15), 1190-1195
- [4] Chariton-Ouw KM, Cordreanu ME, Leake SS, Sandhu HK, Calderon D, Azizzadeh A, Astrera AL, Safi HJ., Open repair of adult aortic coarctation mostly by a resection and graft replacement technique. *J Vasc Surg.*, 2015, 61(1), 66-72
- [5] Lemaire A, Cuttone F, Desgue J, Ivascau C, Caprio S, Saplacan V, Belin A, Babatasi G., Late complication after repair of aortic coarctation. *Asian Cardiovasc Thorac Ann*, 2015, 23(4), 423-429

- [6] Kudo M, Keneda K, Yokoyama S, Nagasaka S, Taki T, Yoshida Y, Marui A, Nishiwaki N., Adult congenital aortic coarctation; report of a case, *Kyobu Geka*, 2015, 68(2), 133-135
- [7] Rakhra SS, Lee M, Ivengar AJ, Wheaton GR, Grigg L, Konstantinov IE, Brizard CP, d'Udekem Y., Poor outcomes aftersurgery for coarctation repair with hypoplastic arch warrants more extensive initial surgery and close long-term, *Interact Cardiovasc Thorac Surg*, 2013, 16(1), 31-36
- [8] Meadows J, Minahan M, McElhinney DB, McEnaney K, Ringel R., Intermediate Outcomes in the prospective, multicenter coarctation of the aorta stent trial (COAST), *Circulation*, 2015, 131(19), 1656-1664
- [9] Kim SS, Jeong MH, Kim JE, Yim YR, Park HJ, LEE SH, Rhew SH, Jeong YW Kim JH, Cho JG, Park JC., Successful treatment of a ruptured subclavian artery aneurysm presenting as hemoptysis with a covered stent, *Chonnam Med J*, 2014, 50(2), 70-73
- [10] Taguchi E, Nishigami K, Miyamoto S, Sakamoto T, Nakao K., Impact of shear stress and atherosclerosis on entrance-tear formation in patients with acute aortic syndromes, *Heart Vessels*, 2014, 29(1), 78-82
- [11] Ananiadou OG, Koutsogiannidis Ch, Ampatzidou F, Drossos GE., Aortic root aneurysm in an adult patient with aortic coarctation: a single-stage approach, *Interactive Cardiovascular and Thoracic Surgery*, 2012, 15, 534-536
- [12] Prifti E, Kuci S, Krakulli K, Nuellari E., Pseudoaneurysm of the descending aorta complicating an untreated aortic coarctation, *Ann Thorac Surg* 2012, 99(1), 3-5