Cardiology in the Young

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Transcutaneous embolisation of an axillary artery aneurysm in a child with Takayasu arteritis

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Brief Report

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Abstract

We report the case of a 10-year-old girl with Takayasu arteritis who developed acute onset wrist drop diagnosed with a large right axillary artery aneurysm compressing the surrounding structures. Our case is unique because it describes a rare presentation of Takayasu arteritis (axillary aneurysm) in a child that was treated successfully in an unconventional manner by transcutaneous embolisation following failure of trans-arterial approach.

Takayasu arteritis is a type of granulomatous vasculitis involving main circulatory vessels.[1] This disease most commonly affects women of child-bearing age, but has also been reported in young children.[2] It presents among all ethnicities but appears to be more prevalent in the Asian population and northwestern Turkish population.[3] Treatment options in children have been restricted to limited reports focusing on immunosuppressive therapies.[4] The aim of this report is to describe an unconventional management of an axillary artery aneurysm in a young girl with Takayasu arteritis with an emphasis on a successful direct transcutaneous embolisation of the aneurysm.

Case

Our patient is a 10-year-old female, who presented with diffuse, extremely painful skin ulcerations. She was admitted to the American University of Beirut-Medical Center and was diagnosed with pyoderma gangrenosum. She received intravenous Solumedrol with limited response. During her hospital stay, she developed right wrist drop. Computed tomography angiography showed a large ascending aortic aneurysm and an axillary aneurysm 5 cm × 5 cm in dimension compressing the nerve plexus. The ascending aorta was surgically repaired and the histopathological examination confirmed Takayasu arteritis diagnosis, with chronic and focal granulomatous inflammation in the arterial media with prominent loss of elastic fibre and fibrosis and replacement of the intima by granulation tissue. The patient was started on tumour necrosis factor alpha inhibitors, and after few cycles her skin lesions markedly improved and she regained full motor power of her right hand. Given her marked improvement with full hand motor function recovery, the family declined further intervention on the axillary aneurysm.

Eight months later, she presented with acute right axillary pain. She had a severely tender pulsating axillary mass with stretching of the overlying skin. Duplex confirmed the presence of a large axillary pseudo-aneurysm (Supplementary Fig S1). Patient was taken to angiography for possible embolisation. Unfortunately, the procedure failed to secure the aneurysm due to the tortuosity of the collateral vasculature (Fig 1). After establishing a coaxial system consisting of a 60-cm 6 French sheath, a 5 French Bern catheter, and a variety of micro-catheter and wire combinations (including Mirage, X-Pedion, and Echelon micro systems from ev3, Plymouth, MN, USA and Renegade HI-Flo micro system from Boston Scientific, Natick, USA), multiple unsuccessful attempts were made to reach the aneurysm. At this point, direct puncture of the aneurysm under fluoroscopy guidance was done, using an 18-gauge needle (Cook, Bloomington, Indiana) (Fig 2). Following needle insertion, multiple coils (Axium, ev3) were placed in the aneurysm and the cavity was plastered with Onyx (ev3). Angiographic control done through the collateral catheter showed complete exclusion of the aneurysm (Fig 1). Patient improved after the procedure; pain subsided, and she went back to school performing normal activities. Two months later, she presented to the Emergency Department with haemodynamic and respiratory collapse and was found to have free rupture of her aortic root at the level of the first surgical repair (known long-term anastomotic complication in Takayasu arteritis); she succumbed in the paediatric ICU shortly after presentation.

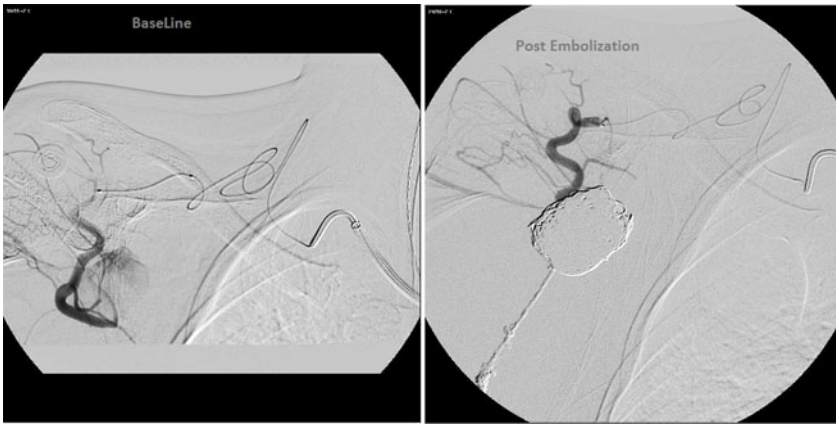

Figure 1. Side-by-side baseline selective angiography and completion post direct embolization.

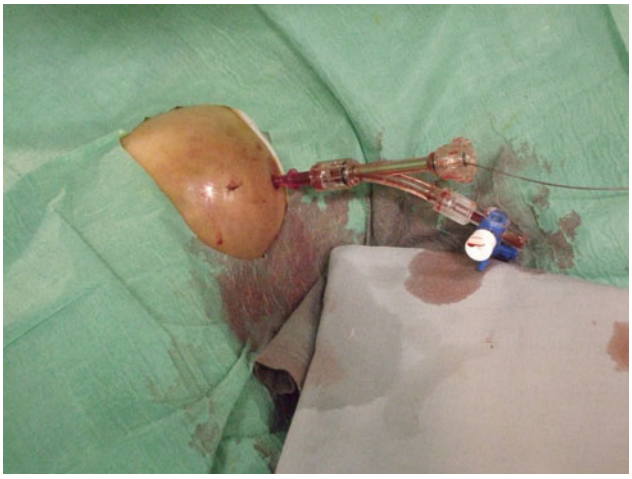

Figure 2. Direct aneurysm puncture.

Discussion

Takayasu arteritis is a vasculitis entity divided into two stages, the generalised inflammatory state and the pulseless stage. During the first stage, patients will complain of systematic symptoms, such as low-grade fever and weakness. Arterial stenosis and aneurysms can be observed during the late stage also called the pulseless or vascular inflammatory stage.[5] Most patients with Takayasu arteritis have stenotic or occlusive changes; aneurysmal degeneration is relatively rare and is related to the destruction of the arterial media during the acute progression phase. Although Takayasu arteritis has been extensively researched among the adult population,[6] small number of reports has been published regarding its manifestation in children.[7]

Takayasu arteritis is related to other autoimmune disorders among which are juvenile rheumatoid arthritis, systemic lupus erythematosus, and Crohn's disease.[8] The association of pyoderma gangrenosum and Takayasu arteritis has rarely been reported in the Western literature, but is well known in the Japanese population.[9] Our patient presented initially with pyoderma gangrenosum and further investigations revealed the diagnosis of Takayasu arteritis. Upper extremity aneurysms are rare and often detected at the time of onset of symptoms. To our knowledge, axillary aneurysm secondary to Takayasu arteritis has been reported only once in the literature.[10] Management of Takayasu arteritis varies between steroids and tumour necrosis factor alpha inhibitors depending on the disease's stage. It is quite possible that our patient improved at first on steroids and infliximab from reduction of the inflammatory process surrounding the axillary aneurysm; nevertheless, this did not prevent later expansion and further brachial plexus compression. Duplex ultrasound was instrumental in confirming the pathology and documenting expansion compared to the previous studies. It showed an irregular shape with thickening of the wall that was severely tender under the probe raising the possibility of a contained rupture (Supplementary Fig S1). Since duplex findings correlated perfectly with the clinical diagnosis, no further imaging was done. There was no need at that point to control the old aortic repair. Although the conventional treatment approach would have been exploration and repair of the axillary artery, in this particular situation, with the subclavian artery being occluded and well collateralised all what was needed was to exclude the pressurised aneurysm sac and stop the leak. Thus, an embolisation procedure without revascularisation was contemplated. After several unsuccessful attempts at percutaneous embolisation, we decided to try the transcutaneous approach. The use of thrombin injections that are commonly done in peripheral pseudo-aneurysms was debated, but decided against since the behaviour of the thrombin in this setting and possible collaterals occlusion could not be predicted.

Conclusion

Our case is unique because it describes a rare presentation of Takayasu arteritis (axillary aneurysm) in a child that was treated successfully in an unconventional manner by transcutaneous embolisation.

This direct access technique could be used more often in applicable cases where trans-arterial embolisation is more challenging or inaccessible.

Supplementary material. To view supplementary material for this article, please visit https://doi.org/10.1017/S1047951119001914

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Conflicts of Interest. None.

Ethical Standards. This report is in compliance with our institution's Human Research Protection Program (HRPP) and with our Institutional Review Board (IRB) policies.

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