CASE REPORT

True brachial artery aneurysm in a child aged 2 years

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SUMMARY

Congenital brachial artery true aneurysms are exceedingly rare. Most are pseudoaneurysms secondary to trauma or infection. We report a boy aged 2 years who presented with painless, pulsatile swelling on the medial aspect of the right arm, 4 cm above the elbow joint that had been present since birth. Spiral CT angiography showed a fusiform aneurysm of the distal right brachial artery with a peripheral crescent-shaped thrombus. Distal arteries were normally opacified. There was no evidence of abnormal dilation or stenosis in any other artery. The aneurysm was surgically resected, with vascularisation re-established using a reversed great saphenous vein graft. His postoperative course was uneventful. Early surgery should be performed for moderately sized to large aneurysms that recently increased in size, exhibited luminal thrombus formation or caused neurovascular distal limb compromise. Early surgery could prevent complications such as a ruptured aneurysm, thromboembolism or limb ischaemia or loss.

BACKGROUND

An aneurysm is defined as an abnormal, localised, widened/dilated, ballooning of the wall of an artery, vein or heart, leading to its weakness.¹

Among peripheral artery aneurysms, those of the lower extremity (eg, femoral, popliteal) are common. Upper extremity aneurysms are rare.^{2–3}

Most of the brachial artery aneurysms are pseudoaneurysms that are commonly caused by arterial damage due to a diagnostic or therapeutic intervention. It may also be due, however, to direct trauma leading to wall disruption and damage.⁴

Congenital brachial artery aneurysms are rare. Only 5% of peripheral artery aneurysms are located in the upper extremity. Most congenital brachial artery aneurysms are associated with other systemic diseases.⁵

Only 14 cases of brachial artery aneurysms in patients <12 years of age have been reported since 1950. In nine of these cases, a brachial artery aneurysm was associated with other aneurysms. Only five cases had a true brachial artery aneurysm not associated with other central or peripheral artery aneurysms or any systemic disease.⁵

CASE PRESENTATION

A boy aged 2 years presented with a painless, pulsatile swelling on the medial aspect of his right arm 4 cm above the elbow joint that had been there since birth. The swelling recently increased, almost doubling, during the last 2 months. There were no symptoms of pain, numbness, bruising, skin

discolouration or wasting of the right forearm or hand. There was no past history of birth trauma, fracture, therapeutic or diagnostic venipuncture, or intra-arterial cannulation. He had not undergone any surgery or hospitalisation since birth. There was no history of fever, dyspnoea, loss of consciousness, blackouts, headache, fits, joint dislocation, joint or skin laxity, or other swelling on the body.

Most congenital brachial artery aneurysms are associated with central or peripheral arterial aneurysms due to a systemic disease (eg, Ehlers-Danlos syndrome, Marfan syndrome, Kawasaki disease, giant cell arteritis, polyarteritis nodosa).^{5–6} The whole course of his gestation was uneventful. There was no family history of aneurysm, congenital vascular disease or connective tissue disorder.

On examination, his height and weight were appropriate for his age. There was no pallor or lymphadenopathy. Physical examination of the swelling showed a 40×30 mm fusiform, pulsatile, expansile swelling on the medial aspect of the right arm 4 cm above the medial epicondyle of the right humerus. The swelling was non-tender, non-reducible and transversely mobile. There was no scarring, skin discolouration or prominent veins visible over the swelling. Right radial, ulnar and all other pulses were palpable with normal volume. Radial, ulnar and median nerve sensory and motor functions were intact. No bruit was audible on auscultation. Heart rate was 100 bpm with no added heart sounds or murmurs. The remainder of the systemic examination was unremarkable.

INVESTIGATIONS

The complete blood picture showed a haemoglobin concentration of 9.8 g/dL with normal platelet and white cell counts. Erythrocyte sedimentation rate, coagulation profile, C reactive protein, renal function tests, liver function tests, lipid profile and proteins C and S were within the normal range. The anti-dsDNA immunoglobulin G test was negative.

Abdominal ultrasonography and echocardiography showed no abnormalities. Arterial Doppler images of the right upper limb showed a right brachial artery aneurysm with turbulent flow through the aneurysm and normal arterial flow of the right radial and ulnar arteries. CT angiography (CTA) showed fusiform dilation of the distal right brachial artery 4 cm proximal to the right elbow joint (figure 1). The aneurysm was thick walled with a luminal peripheral crescent-shaped thrombus. The right axillary, radial, ulnar and distal palmar arteries and arteries of the left upper limb, lower limbs,

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Figure 1 CT angiography showing balloon-like dilation of the distal end of the right brachial artery.

chest, abdomen, neck and brain were normally opacified with no evidence of abnormal dilation or stenosis. Preoperative lower limb venous mapping was performed to select a great saphenous vein graft.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis for brachial artery aneurysm includes pseudoaneurysm, haematoma and arteriovenous malformation.

TREATMENT

As brachial artery aneurysms are rare, with few cases reported to date, no definitive treatment or surgical procedure has been established.⁷ Although endovascular stenting to repair popliteal artery aneurysms has been reported, no such technique has been suggested for repairing a true brachial artery aneurysm.⁸ Hence, the best therapeutic option for a brachial artery aneurysm is surgical repair, thereby preventing upper extremity ischaemia.¹

We planned early surgery because of the recent increase in the size of the aneurysm and the presence of the thrombus seen on CTA. It was designed to prevent thromboembolic complications, rupture of the aneurysm and/or limb loss.

Figure 2 Operative view of fusiform aneurysm arising from the right brachial artery.

Figure 3 A reversed great saphenous vein graft interposed.

Under general anaesthesia, a 7 cm vertical skin incision was made over the right brachial artery aneurysm on the medial aspect of the right arm. A saccular aneurysm was identified arising from the distal end of the right brachial artery (figure 2). Vascular clamps were applied to the right brachial artery proximal and distal to the aneurysm. The aneurysm was resected, a 5 cm reversed great saphenous vein graft interposed and an end-to-end anastomosis created using the proximal and distal ends of the right brachial artery. It was held in place with polypropylene 6/0 sutures (figure 3). Histopathological examination confirmed that it was a true aneurysm. All three vessel wall layers were intact with an organised thrombus in the vessel lumen.

OUTCOME AND FOLLOW-UP

The patient remained hospitalised for 5 days and was discharged on postoperative day 3 after surgery. He was prescribed 75 mg aspirin orally, once per day, for 6 months. CTA performed 1 week after surgery showed a patent vascular graft. The patient was advised to appear for follow-up at 10 days postoperatively and then monthly for 3 months, quarterly for a year and yearly. CTA/MRI was advised at 6 months and then annually.

There were no thromboembolic complications, vascular graft stenosis, right forearm or hand ischaemia, or aneurysm recurrence or development at any other site during 6 months of follow-up. Distal neurovascular status of the right upper limb was intact throughout the postoperative period.

DISCUSSION

Aneurysms may be true or false. If all three layers of the vessel wall are involved, it is said to be a true aneurysm. In false aneurysms, only a single layer of fibrous tissue is involved. An aneurysm may also be congenital or acquired (mycotic, syphilitic, traumatic) and is classified as central or peripheral according to its site.¹

Among peripheral arterial aneurysms, those in the lower extremity (eg, popliteal, femoral) are commonly encountered. Upper extremity aneurysms are rare.^{2 3}

Most brachial artery aneurysms are pseudoaneurysms that are commonly caused by arterial damage due to a diagnostic or therapeutic intervention. They may also be caused by direct trauma leading to wall disruption and damage.⁴

Congenital brachial artery aneurysms are rare. Only 5% of peripheral artery aneurysms are in the upper extremity.⁵

Most congenital brachial artery aneurysms are associated with other systemic diseases (eg, Ehlers-Danlos syndrome, Marfan

syndrome, Kawasaki disease, giant cell arteritis, polyarteritis nodosa).^{5 6}

Since 1950, only 14 cases of brachial artery aneurysms in children <12 years of age have been reported. In nine of these cases, the brachial artery aneurysm was associated with other aneurysms. There were only five cases of an isolated true brachial artery aneurysm not associated with other central or peripheral artery aneurysms or any systemic disease.⁵

Patients with brachial artery aneurysms may be asymptomatic or present with a pulsatile mass. Other findings appear when thromboembolic complications occur.⁹ Complications include profuse bleeding, vascular collapse and/or thrombus formation leading to emboli and the risk of limb loss.¹⁰

Since brachial artery aneurysm is rare and few cases have been reported up to now, there is no established definitive treatment or surgical procedure.⁷

Although endovascular stenting has been reported for repair of popliteal artery aneurysms, no such technique to repair true brachial artery aneurysms has appeared in the literature.⁸

Therefore, to prevent upper extremity ischaemic sequelae, the best therapeutic option for brachial artery aneurysm is surgery.¹

Urgent surgery is not required if a brachial artery aneurysm is small and asymptomatic. Early surgical intervention is required, however, if the aneurysm is moderately sized or large, has recently increased in size, exhibits thrombus within the aneurysm or there is neurovascular compromise of the distal limb. If the aneurysm is small, surgical treatment involves aneurysm resection with end-to-end anastomosis. For large aneurysms, the definitive treatment is aneurysm resection and revascularisation of the brachial artery with a reversed saphenous vein graft interposed, anastomosed with the proximal and distal ends of the brachial artery.^{5 6}

In the present case, the brachial artery aneurysm size had recently increased and CTA showed thrombus formation. Early surgical reconstruction was planned to avoid aneurysm rupture and thromboembolic complications.

Children who undergo surgical resection of an aneurysm and arterial reconstruction should take oral aspirin for 6 months postoperatively to reduce the risk of occlusion at the sites of anastomosis.^{5 6}

Long-term follow-up and screening studies are recommended in children with a brachial artery aneurysm because of the risk of recurrence and the development of peripheral or central arterial aneurysms at other sites of the body, which can form even several years later. Duplex ultrasonography or magnetic resonance angiography is recommended at 1 and 2 years following arterial reconstruction.^{5 6}

Diligent workup is needed for accurate diagnosis of an aneurysm. Early surgical intervention is required for moderate to large aneurysms, aneurysms whose size recently increased, intra-aneurysm thrombus formation or if there is neurovascular compromise of the distal limb. In this case, meticulous workup confirmed the presence of a congenital, idiopathic, right brachial artery true aneurysm. It was not associated with central or peripheral arterial aneurysms at other sites of the body or any systemic disease.

Learning points

- ▶ Upper extremity peripheral arterial aneurysms are relatively rare. Congenital, idiopathic, brachial artery true aneurysms are exceedingly rare. Most brachial artery aneurysms are pseudoaneurysms secondary to trauma or infection. Thus, a diligent workup is needed for an accurate diagnosis.
- ▶ To avoid rupture and thromboembolic complications, early surgical intervention is required for moderately sized to large aneurysms, aneurysms that recently increased in size, intra-aneurysm thrombus formation or the presence of neurovascular compromise of the distal limb.
- ▶ The best therapeutic option is surgery, which results in an excellent outcome.
- ▶ Long-term follow-up and screening studies are recommended in children with congenital, true, brachial artery aneurysms because of the risk of recurrence and the development of peripheral or central arterial aneurysms at other sites of the body, which can form even several years later.

Contributors AG is the main surgeon who operated the case. Apart from the surgery that was he, who selected this case for publication. He also contributed greatly in the drafting and revision of this document. AA wrote the abstract, Case presentation, Investigations and Treatment sections of this article. NUK is a consultant epidemiologist and an experienced researcher. He critically reviewed the whole manuscript, edited and drafted again and gave a final touch to the report and made it ready for publication. SA did extensive literature search for this case report. She also wrote the Discussion section and provided the references for the article.

Competing interests None declared.

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