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CASE REPORT

Acute Leg Ischaemia in a Child due to a Thrombosed Popliteal Aneurysm

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Introduction: The case of an idiopathic thrombosed popliteal aneurysm is described in an otherwise healthy 6 year old child. This is the fourth reported case and the second youngest patient to present with an idiopathic isolated popliteal aneurysm.

Report: A 6 year old boy presented with an acutely ischaemic right foot. Computed tomography angiography confirmed a thrombosed popliteal aneurysm. A femoropopliteal bypass was performed with reversed long saphenous vein and ligation of the aneurysm. Yearly follow up is ongoing with ultrasound surveillance; the child's growth and development is unaffected, and the graft is patent. There was a readmission over six years later with claudication on the right side. There was evidence of thrombus in the graft with associated distal embolisation, which was managed conservatively with anticoagulation.

Discussion: Given the rarity of such presentations in the paediatric population, there is minimal good quality data to guide treatment. There have been three previous cases of idiopathic popliteal aneurysms all managed with a reversed long saphenous vein femoropopliteal bypass with resection of the aneurysm. Management should be guided based on the clinical picture and should be undertaken in specialised tertiary centres if possible. Surgical intervention is the treatment of choice in patients with an ischaemic limb.

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CASE REPORT

A 6 year old boy presented in March 2012 to a tertiary vascular centre with a 2 day history of an acutely ischaemic right foot, with restricted walking distance. On examination, the foot was pale and cold; there was a palpable right femoral pulse with absent popliteal and pedal pulses. At the time of presentation, the patient was otherwise well, with no known past medical history. He was started on a heparin infusion after review by the vascular team.

Duplex ultrasound of the right lower limb arteries suggested that there was an occlusion of the superficial femoral artery at the level of the adductor hiatus. There was evidence of collateralisation; dampened arterial flow reappeared in the distal popliteal artery and the crural vessels to the ankle.

Subsequent computed tomography angiography showed that there was a 1.6 cm thrombosed right popliteal artery aneurysm, which was partially calcified; there was triple vessel runoff to the ankle (Fig. 1).

Surgical procedure

Surgery was performed as an emergency on the day of admission; a right femoropopliteal bypass with reversed great saphenous vein was performed using a medial approach, along with a ligation of the popliteal aneurysm. The long saphenous vein was harvested from mid-thigh to below the knee. The superficial femoral artery was identified and controlled at the level of the adductor hiatus. Below the knee, the popliteal artery was dissected and controlled below the aneurysm. In total, 1,000 units of heparin was administered, and the clamps were put in place.

The upper anastomosis of the reversed saphenous vein was end to side. Tunnelled via the popliteal fossa, the graft was deliberately left long in order to accommodate growth. The popliteal artery was dissected and ligated below the aneurysm and the distal anastomosis was end to side.

There were good pulses in the graft, and distal Doppler signals were present throughout.

Post-operative period

In the post-operative period there were no complications. The heparin infusion was stopped the day after surgery. An ultrasound scan confirmed a patent graft. The patient complained of headaches, but these were chronic and had not worsened during his admission.

Following discharge there were no issues regarding his right leg for almost three years; however, in 2015 he re-

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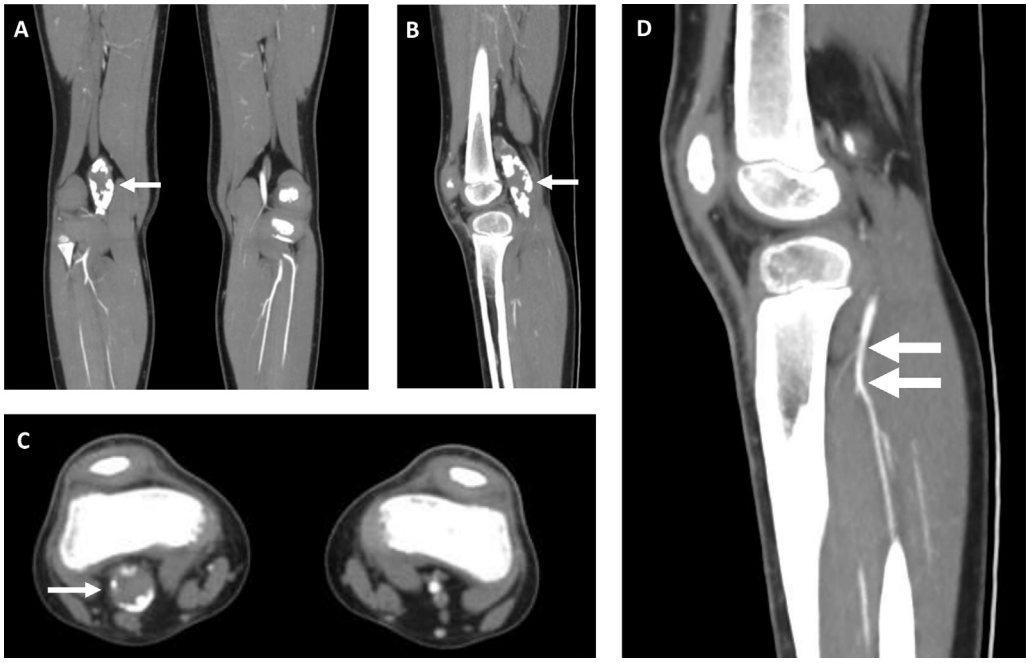

Figure 1. Computed tomography peripheral angiography. (A) Coronal view showing thrombosed, partially calcified right popliteal artery aneurysm (arrow). (B) Sagittal view. (C) Transverse view. (D) Sagittal section showing triple vessel runoff; upper arrow shows branching of anterior tibial artery and tibio-peroneal trunk, lower arrow shows branching of peroneal and posterior tibial arteries.

presented to the vascular clinic with right foot numbness, which was triggered by cold temperatures. It was not positional, and there were no concerning clinical features or on duplex ultrasound of the graft; this was managed conservatively as vasospasm.

It was chosen to follow up yearly with duplex ultrasound to screen for stenosis of the graft; all scans have shown a patent graft, with good flow. The thrombosed popliteal aneurysm is stable in size (in 2017 the aneurysm was measured at 1.4 cm).

Readmission

Over six years later, in September 2018, the patient presented with a two week history of pain in his right calf and foot. He was unable to walk long distances or play football for more than a few minutes; however, there was no rest pain. The right foot was seen to be paler that the left, particularly after a warm bath.

On examination there was a pulse present in the graft; however, there was no dorsalis pedis or posterior tibial pulse. Right foot capillary refill was also slower. A duplex ultrasound scan done on the same day suggested that there was a small amount of thrombus in the graft with some associated dilation; the graft remained patent. There was, however, evidence of an obstruction of the anterior tibial and mid-posterior tibial artery, with good collateral flow. The most likely explanation for these findings is that the thrombus in the graft had embolised into his native tibial vessels. After discussion with the haematology team, the plan was to anticoagulate with low molecular weight heparin for at least three months.

Follow up one week later showed a better perfused right foot with a biphasic Doppler signal present in the anterior and posterior tibial arteries. The hope is that collateralisation will resolve the symptoms, while the anticoagulation plan should reduce the risk of further embolic events.

DISCUSSION

True aneurysms are most commonly caused by atherosclerosis and hypertension; however, this is only true for the adult population.[1] In 1991, Sarkar et al. proposed a nine category clinic-pathological classification of arterial aneurysms in children: (i) arterial infection; (ii) giant cell aortoarteritis; (iii) autoimmune vasculitis; (iv) Kawasaki disease; (v) medial degeneration (Ehlers-Danlos or Marfan syndrome); (vi) other forms of degeneration of the media; (vii) arterial dysplasias; (viii) congenital idiopathic factors; (ix) extravascular events leading to pseudoaneurysm.[2] Full work up for these conditions yielded nothing, implying that this case fitted firmly into the congenital idiopathic variant and there was no evidence clinically or radiologically of aneurysm disease at other locations.

Examples in the literature of idiopathic true popliteal aneurysms in the paediatric population are vanishingly rare. In 1994, Hurley described such a case in a 14 year old girl,[3] and, in Chile, Olguín et al. reported a similar case in a three year old,[4] both of whom were treated by a reversed venous graft with resection of the aneurysm. Notrica et al. reported a similar case in 2016, which was managed in almost the same way; they found that distal endarterectomies were required to re-establish flow.[5]

One case report in 2012 described a nine year old girl who sustained traumatic injury to her right femoral artery in 1964, managed with an ipsilateral saphenous vein bypass graft. In this case, there was no evidence of arterial or

venous disease on follow up for 44 years, until she presented with acute limb ischaemia of the right leg in 2008. Angiography confirmed occlusion of the graft, which was replaced surgically with another segment of saphenous vein. This is the longest reported patency of a saphenous vein graft. [6] Poor growth of vein grafts in the paediatric population has not been a documented issue, as evidenced by the abovementioned cases. However, given that a graft thrombus was observed in this case, there is a chance that the need for a revision of the graft may arise in the near future, particularly if there is evidence of further thrombotic events.

As a technical point, the decision was made to use interrupted rather than continuous sutures, to allow for growth. Remarkably, there appears to be no comparative data in the literature between the two techniques in this population; however, this has been the practice in the paediatric population for some time. [7]

Davis et al. undertook a retrospective analysis at the University of Michigan on 41 children who underwent surgical treatment for non-aortic arterial aneurysms. [8] Of three popliteal aneurysms, one was repaired with endovascular embolisation, one was resected with a subsequent primary anastomosis and one was managed with a reversed saphenous vein graft (it is unclear if this aneurysm itself was ligated or resected). They concluded that surgical treatment should be planned and undertaken on a case by case basis in this population, and their experience suggested that there was minimal peri-operative risk and sustained durability of the surgical interventions.

In contrast, a study from a Canadian tertiary paediatric centre described a different protocol; if presentation was with acute limb ischaemia, but without motor or sensory dysfunction, patients were trialled on 24 h of anticoagulation. If there was no response, thrombolysis with tissue plasminogen activator was undertaken, and, failing this, progression to surgical intervention was considered. Of the eight patients in this centre who had idiopathic causes of acute limb ischaemia, six resolved with anticoagulation alone. Therefore, they recommended a generally conservative approach in the paediatric population. [9]

These differences in the recommendations above are probably because the studies looked at different populations; Davis et al. only included patients who had a surgical intervention; [8] they did not compare patients with a control group managed conservatively, as Kayssi et al. did. [9] However, the paper by Kayssi et al. also included causes of ischaemic limb other than aneurysms, so it is unclear if the data apply to the case described here. It is worth noting that in these two papers, almost all the cases described were secondary to trauma (iatrogenic or otherwise); this may not directly be extrapolatable to the patient described here. It would be ideal to study the subset of patients who present with idiopathic true aneurysms and investigate the interventions used, post-operative complications, mortality, and the need for re-intervention; however, it appears that this occurrence is so rare in the paediatric population that there are currently not enough data to reach a conclusion.

In conclusion, the case of a six year old boy who presented with what was most likely an idiopathic true aneurysm has been reported. This is the second youngest presentation of a patient with a popliteal aneurysm, and was managed with a reversed long saphenous vein graft. Uniquely in this case, no resection of the aneurysm was undertaken. Review of the literature suggests that this is rare in the paediatric population and, as such, there is very little evidence to guide treatment of future cases. Management should therefore be guided based on the clinical picture and should be undertaken in specialised tertiary centres if possible.

CONFLICTS OF INTEREST

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