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Spontaneous isolated true aneurysms of the brachial artery in children

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ABSTRACT

Upper extremity peripheral artery aneurysms are rare, and have been previously reported in children to be associated with congenital malformations and infectious or inflammatory processes. In this case series, we present two unique cases of spontaneous isolated true aneurysms of the brachial artery in two children. Both cases were incidentally found on examination, diagnosed by ultrasonography, and successfully managed by surgical excision and micro-vascular repair with vein grafting.

1. Introduction

True aneurysms of the brachial artery in children are extremely rare, and often are attributed to congenital malformation, iatrogenic, inflammatory or infectious processes, with reported incidences of less than 0.5% [1–3]. Likewise, *Spontaneous* occurrences of isolated aneurysms in otherwise healthy children, not clearly attributed to any of the processes above, are exceedingly rare, limited to only a few case reports [4]. Since the 1970s, only about 12 cases of truly idiopathic brachial artery aneurysms have been reported in the literature in children with ages ranging from 7 months to 18 years, with varying outcomes. We present two unique and more recent cases of young children (an infant and a toddler) who presented with incidental, spontaneous isolated true aneurysms of the brachial artery, diagnosed by multi-imaging modalities, and successfully managed surgically with excision and grafting without residual functional impairment.

2. Case report

2.1. Patient 1

An otherwise healthy 9 month-old female presented with a two-week history of an incidentally found left arm mass, unchanged in size since discovery. The patient had an uneventful birth history with no history of gestational infection or bacteremia as a neonate. She also had normal growth and development, and no prior history of trauma or placement of umbilical or central lines. She was clinically asymptomatic despite her examination notable for a painless, pulsatile, palpable, firm subcutaneous mass measuring approximately 1 × 1 cm just proximal to the medial aspect of the elbow (Fig. 1). She had strong Doppler signals distally in the radial and ulnar distributions, without an audible thrill or bruit. Ultrasonography of the mass demonstrated a 1.7 cm × 1 cm true fusiform shaped aneurysm of the brachial artery, although the presence of a vascular anomaly or alternate vascular beds for arterial dysplasia could not be excluded. Additional imaging including abdominal ultrasonography, echocardiography and chest radiography were normal with no gross evidence of aneurysms elsewhere. Magnetic resonance angiography was not performed on this patient. Rheumatologic work up ruled out vasculitis and Kawasaki disease.

Given the risk of aneurysmal rupture or thromboembolic complications [2,5,6], the decision was made to resect and reconstruct the lesion. In the operating room, under tourniquet control for ease of dissection, especially of the nerves in this area, the patient underwent exploration of the distal arm through a longitudinal incision up to the elbow crease with a transverse extension. The brachial artery was controlled proximally and distally prior to dissection of the aneurysm. A large aneurysm was

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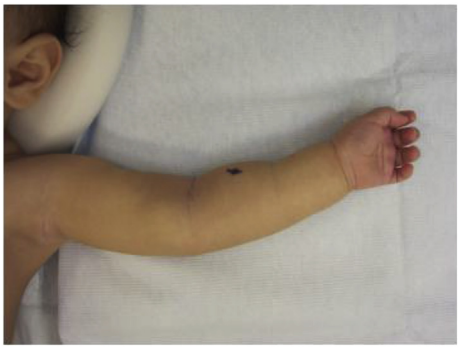

Fig. 1. Patient 1 with notable aneurysm surgical site marked out prior to surgical intervention.

identified (Fig. 2A) and isolated. The area of disease was resected along with a few millimeters of extra vessel on either side as margin. The resultant defect was then reconstructed with a reversed basilic vein graft (1.5 cm segment), repaired with interrupted 9-0 nylon sutures in an end-to-end fashion without spatulation using an operating microscope (Fig. 2B). Brisk perfusion was restored and pulses remained strong with the elbow in full flexion and extension in the operating room. Postoperatively, the patient was immobilized with the elbow in 50° of flexion and observed overnight. Aspirin was started and continued for 30 days. Immobilization was continued for several weeks and the patient returned to normal activities. Histopathology showed medial thinning, diffuse intimal myxoid thickening and focal calcifications but no evidence of inflammation. The patient had a remarkably stable exam on follow-up visit. At 6 months postoperatively with normal motor and sensory function of the hand and normal range of motion of the elbow. Repeat ultrasonography at that time showed a patent graft with no evidence of aneurysmal recurrence.

2.2. Patient 2

A 3-year-old female with no relevant past medical history presented with a several week history of an incidental small 1 cm mass on her right arm, reportedly to have significantly increased in size by the mother. Patient had no history of gestational infection or bacteremia as a neonate, no prior history of trauma and no placement of umbilical or central lines. She was asymptomatic despite her exam notable for a pulsatile mass along the medial aspect of the right upper extremity with intact distal pulses bilaterally in the upper and lower extremities. Ultrasonography demonstrated a saccular aneurysm of the right brachial artery measuring 3.0 by 0.9 cm. Additional imaging, including full body MRA, echocardiography and labs were unremarkable. She also underwent surgical excision via a longitudinal incision, without the use of a tourniquet, followed by reconstruction with reversed saphenous vein graft (4 cm segment) using a similar micro-vascular technique as patient 1. (Fig. 3). The patient recovered well with strong and intact distal pulses on exam. On follow-up visit at 7 months, she was recovering uneventfully without any functional or physical deficits of her arms. Repeat ultrasonography demonstrated a patent saphenous vein graft with no evidence of recurrent aneurysm or stenosis. Histopathology demonstrated transmural lympho-plasmacytic infiltrate associated with disruption of elastic lamellae consistent with arteritis. However, full rheumatological workup was negative.

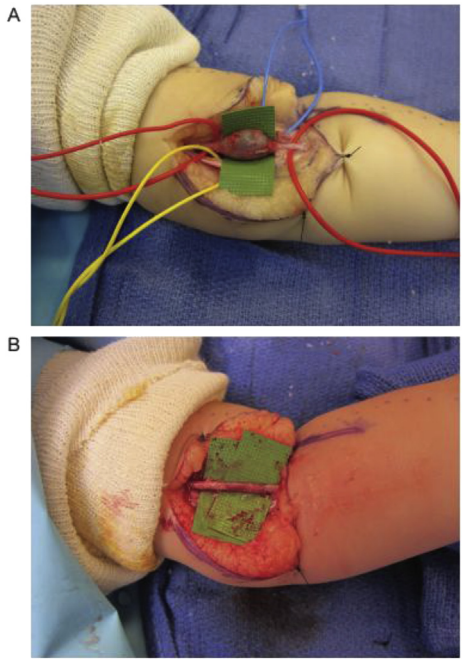

Fig. 2. A. Patient 1 brachial artery aneurysm after dissection: Venous drainage marked with blue vessel loop, median nerve marked with yellow vessel loop. B. Shows reverse basilic vein graft post anastomosis. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

2.3. Comment

Spontaneous true aneurysms of the brachial artery in children are exceeding rare and very few cases have been previously reported in the literature [4]. In most cases, the pathophysiology of these aneurysms is unclear, and unrelated to infectious, congenital

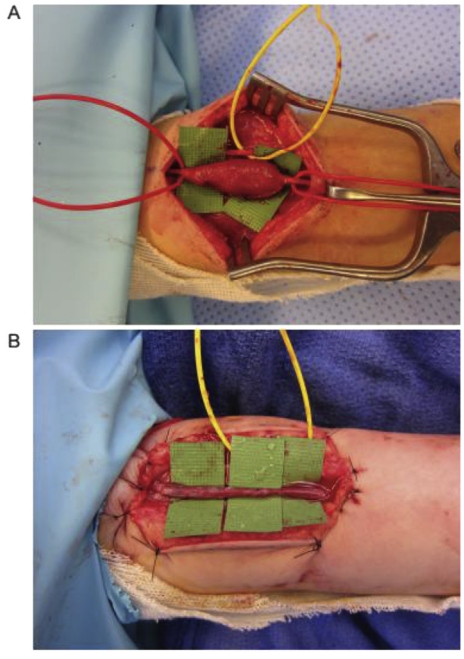

Fig. 3. A. Patient 2 aneurysm after dissection: Venous drainage marked with yellow vessel loop. B. Shows saphenous interposition graft post anastomosis. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)