Ulnar Artery Aneurysm in a 6-Month-Old: A Case Report

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Abstract

Background: Pediatric ulnar aneurysms are rare and, unlike their adult counterparts, cannot be explained by repetitive trauma to the palm. A small number of case reports describe diagnostic difficulty with these lesions and different treatments. **Methods:** We present the case of a 6-month-old with an ulnar artery aneurysm of unknown cause. The diagnosis was supported with magnetic resonance imaging, and the lesion was resected. **Results:** Because the hand remained well perfused, the ulnar artery was not reconstructed. **Conclusions:** Although the early result was good, the long-term outcome of this approach is unknown.

Keywords: ulnar artery, aneurysm, pediatric, excisional biopsy, idiopathic, hypothenar muscles

Introduction

Aneurysms occur as a weakening and expansion of all layers of a vessel wall. By contrast, false aneurysms or pseudoaneurysms typically arise from penetrating trauma; a hematoma forms, organizes, and recannulizes. This mimicking lesion lacks a true intimal layer.

In the adult upper limb, true aneurysms occur most commonly in the ulnar artery at the wrist. In this location, the ulnar artery is drawn over the rigid hook of the hamate and is protected only by thin layers of soft tissue. Repetitive microtrauma, such as that sustained with long-time use of the hypothenar eminence for pounding, weakens the vessel, and the so-called hypothenar hammer syndrome results. Patients complain of a palpable mass, sensory changes resulting from compression of the ulnar nerve in the Guyon canal, or embolic sequelae affecting the ulnar digits. Pain and pallor may eventually progress to ulceration and frank necrosis of the fingertips. Surgery may be used to interrupt the natural history of this disease, although less or noninvasive measures are often appropriate.^{5,7}

Ulnar artery aneurysms in young children are at least anatomically similar to their adult counterparts. Diagnosis takes place as it would in an adult; however, because the same repetitive trauma has likely not occurred, predisposing factors such as connective tissue disease may be sought. Because the natural history of these pediatric lesions is poorly understood, the adult disease process is used as a facsimile and surgery may be undertaken.

We present the case of 6-month-old boy with an idiopathic ulnar artery aneurysm. Surgical resection without reconstruction of the ulnar artery was successful at early follow-up.

Case Report

A previously healthy, 6-month-old white boy developed an enlarging, firm, mobile, 2 × 2.5-cm mass in the left hypothenar eminence over the previous day and refused to bear weight with the affected hand (Figure 1). No trauma was reported. A radiograph was unremarkable. Ultrasound and magnetic resonance studies (Figure 2) suggested an avascular cystic structure.

An excisional biopsy was undertaken for symptom relief and definitive diagnosis. The mass arose from the ulnar artery, protruded through the hypothenar muscles, and was rubbery, tan in color, and encapsulated. The mass bifurcated in continuity with the ulnar artery and included proximal sections of the common digital artery to the fourth web space and the superficial palmar arch (Figure 3). The mass was closely adherent to the ulnar nerve in the Guyon canal, and the nerve was dissected away from the mass from the wrist to the midpalm. When the tourniquet was let down, prompt reperfusion of the hand including the small finger was apparent. There was, however, no palpable pulsation of the mass or of the vessels in immediate continuity with it. The ulnar artery was ligated proximal and distal to the mass,

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Figure 1. Preoperative photographs of a mass in the hypothenar eminence.

Figure 2. Coronal (a) and axial (b) magnetic resonance images of a hypothenar mass.

and the mass was resected. The hand and small finger remained pink and demonstrated brisk capillary refill.

A pathologist evaluated the mass and found an arterial aneurysm containing thrombus material.

Three months after the operation, the patient's hand was warm and well perfused, and the patient had full active range of motion of his fingers.

Discussion

Older children could presumably experience repetitive blunt trauma during skateboarding, snowboarding, or other athletic activity that might lead to ulnar artery aneurysm. Pediatric ulnar artery aneurysm arising from a single blunt traumatic event has been reported.²

Pediatric ulnar artery aneurysms without trauma have been rarely reported.^{1,3,6,8,9} At approximately 1 year, children begin to walk, and we speculate that repeated falls on the palms may constitute repetitive blunt trauma in this relatively older cohort. In children younger than 1 year, it is

difficult to assume the adult mechanism of aneurysm formation is responsible.

Several authors reporting pediatric ulnar artery aneurysms have sought an underlying connective tissue or autoimmune disorder, but no such association has been discovered.^{1,3,6,9} A number of associated vascular abnormalities have been noted, however, including incomplete palmar arches,^{1,8,9} a persistent median artery,⁹ and absence of the radial artery.³ These associated differences suggest that ulnar artery aneurysm in pediatric patients may be congenital, ie, that the aneurysm is present from birth. Iyer et al. speculated that acute thrombosis might bring a congenital lesion to clinical attention.⁶

A presumptive diagnosis may be based on the presence of a pulsatile mass^{1,3,8,9}; however, as in our case, this is not always present.⁶ Ultrasound^{1,3,6} and advanced imaging^{1,3,6,8,9} have been helpful, but they do not always offer a definitive diagnosis.⁶ Angiography is the gold standard for evaluation of ulnar artery aneurysm in adults. This technique requires arterial access, however, which necessitates sedation in a child. Other drawbacks include the risks of thrombosis, allergy to contrast dye, nephrotoxicity, and increased exposure to radiation.⁵ In the reported case, a precise diagnosis would not have changed our decision to perform an excisional biopsy, and we determined our diagnostic strategy accordingly.

Observation is an appropriate treatment for adults with ulnar artery aneurysm in whom there is little risk for ischemia.⁵ Offer and Sully treated a 1-year-old boy with this lesion conservatively for 1 year until the mass became symptomatic and was excised.⁸ Oral medication may be used for adults, as may intra-arterial thrombolytics,⁵ although the risks of these treatments in children likely outweigh their uncertain benefit. Open surgical options include resection alone, resection and repair, and resection and reconstruction.⁵ Once the diseased segment is resected, blood flow to the hand and small finger must be assessed by observation of capillary refill, by palpation or Doppler examination of distal vessels, and/or by the status of reverse-flow pulsatile spurting from the superficial arch distal to the lesion. As in our case, simple resection may be undertaken if this does not compromise perfusion.^{1,6,8} Resection alone is expedient and avoids additional complexity and, potentially, autograft donor site morbidity. Much evidence suggests that the radial artery alone may support the hand. Repair or reconstruction of ulnar artery lesions may nonetheless minimize the risk of critical ischemia in the distant future.⁴ When resection of an ulnar artery aneurysm does jeopardize distal blood flow, short, simple gaps should absolutely be repaired. Larger gaps and gaps involving branches of the ulnar artery may likewise require reconstruction with vein or artery graft.^{3,9}

Figure 3. Intraoperative photographs of the mass in situ (a), reflected (b), and resected (c).
 Note. In (c), the ulnar nerve, previously underlying the mass, is indicated with surgical ink.

More and better data will inform the management of pediatric ulnar artery aneurysms. We add our experience to the early stages of this undertaking.

Ethical Approval

This study was approved by our institutional review board.

Statement of Human and Animal Rights

This article does not contain any studies with human or animal subjects.

Statement of informed consent

There is no identifiable information about this patient in the case report.

Declaration of Conflicting Interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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