

**Case Report**

# Rare Presentation of an Ulnar Artery Aneurysm in a Six-Month-Old Baby: Case Discussion

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Ulnar artery aneurysm cases have been rarely reported in the literature previously. A number of these cases occur in the adult population and are mostly occupational associated. In children, however, this condition is much less. Only 10 cases in children have been reported in the literature to the best of our knowledge, and the minimum age was 1 year. The etiology is mainly post-traumatic. We present a case for discussion of an ulnar artery aneurysm in a 6-month-old baby with the habit of hitting his hand against a table and the floor.

**Keywords:** infants, hand trauma, aneurysm, ulnar artery

## Introduction

Of all the peripheral artery aneurysms, ulnar arteries have been rarely involved. Few cases have been reported in the literature, and a number of them were in the adult population and mostly occupational associated. They have been found mainly in mechanics and carpenters because of traumatic disruption of the ulnar artery at the palmar aspect of the hand when they use their hands as a hammer.<sup>1)</sup> This condition, which was initially described by Rosen and named by Conn in 1970,<sup>1)</sup> has been defined in the literature in detail as the hypothenar hammer syndrome. The symptoms of this condition can vary from painful pulsatile swelling or hard thrombosed swelling and sometimes gangrene of the fingers from distal embolization. However, in children and mainly in infants, the incidence


of this condition is much less. Only 10 cases have been reported in the literature for ulnar artery aneurysm in children, and to the best of our knowledge, the minimum age was 1 year.<sup>2)</sup> The etiology explained in all the case reports on this condition was post-traumatic and not hereditary. This is because of the close nature of the ulnar artery with the hamate bone at the palmar aspect. This article revisited the latest trends in the presentation and treatment of ulnar artery aneurysms as we encountered an ulnar artery aneurysm in a 6-month-old baby.

## Case Report

A 6-month-old baby boy presented at the Vascular Surgery clinic with a 2-month history of swelling noticed by his parents on the hypothenar eminence of the left hand (Fig. 1). The swelling was gradually increasing in size. The parents gave no history of any unusual crying of the baby, which concludes that it was painless. On our examination, the swelling was around 4×2 cm in size. It was pulsatile on palpation, and there were no motor deficits in the left hand. This swelling was localized in the left hand hypothenar eminence, and both the radial and ulnar pulses were palpable with a good capillary refill. On taking a detailed history, the mother explained that the baby had the habit of hitting his left hand mainly on the table while having food and on other objects. With the clinical history and examination, an aneurysmal swelling was confirmed, most likely arising from the ulnar artery or the palmar arch because of its location. He came in with an arterial duplex, which confirmed only an aneurysmal sac. There was no detailed anatomical description because of the compromised examination due to the age of the child. No other investigations were carried out to confirm the diagnosis because of financial limitations and the very young age of the child, and we opted for surgical excision directly. Intraoperatively, it was an aneurysmal swelling involving the ulnar artery from below the wrist and at the distal forearm involving the superficial palmar arch distally and taking the digital blood supply of the little finger and the ring finger with it (Fig. 2). This aneurysm was excised and repaired with an interposition reverse cephalic vein graft from the forearm of the same side (Fig. 3). A segment of

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**Fig. 1** Pre-op picture of the left hand swelling showing the ulnar artery aneurysm.



**Fig. 2** Aneurysm involving digital arteries.



**Fig. 3** Arterial reconstruction hand vessels with an ipsilateral reverse cephalic vein.

the Y-shaped cephalic vein with a tributary was taken from the distal forearm of the same side. The proximal end of the reverse cephalic vein was anastomosed to the distal end of the ulnar artery, and the two distal ends of the reverse cephalic vein in a Y-shape fashion were anastomosed, one at the superficial palmar arch and one at the common digital artery of the fourth and fifth fingers at the fourth web space. The histopathology of the aneurysmal sac confirmed a dilated vascular channel with fibrin and an attenuated intima, compatible with an aneurysm with

no signs of vasculitis. Postoperatively, he had palpable pulses in both the ulnar and radial arteries, and his motor movement was intact. He was not kept on any anticoagulation, and no further follow-up studies were conducted.

## Discussion

The exact incidence of the ulnar artery aneurysm is not stated in the literature because of its rarity. However, in adults, the incidence is sufficient to conclude the causal mechanism, and in almost all the cases, the mechanism was post-traumatic.<sup>1)</sup> Blunt trauma is associated with a true arterial aneurysm; penetrating trauma is associated with a pseudoaneurysm of the artery.<sup>3)</sup> In a few of the cases reported in the literature, an ulnar artery aneurysm in adults was associated with connective tissue disorders such as Marfan and Kawasaki.<sup>4)</sup> In children, there were only 10 cases reported in the literature with a minimal age of 1 year, and all of them were post-traumatic. It was concluded in one of the studies that it was possibly congenital just because of no history of trauma by the parents; however, unnoticed trauma should always be kept in mind.<sup>5)</sup>

The mechanism of injury to the ulnar artery at the palmar aspect is quite well understood. The division of the ulnar artery into superficial and deep branches occurs in Guyon's canal. Here, it is bounded laterally by the hook of the hamate bone, and around 1–2 cm of the superficial branch remains uncovered at this point before it turns deep into the palmar aponeurosis and forms the superficial palmar arch.<sup>6)</sup> The clinical symptoms of ulnar artery aneurysms in children are pulsatile swelling and, in some cases, painful swelling, cyanosis, pallor, and a positive Allen's test.<sup>3)</sup> However, the symptoms in adults vary in trends from thrombosed swelling to distal embolization, numbness, paresthesia, and pain. The differential diagnosis of soft tissue swelling at the ulnar aspect of the hand in children includes lipoma, neurofibroma, soft tissue sarcoma, synovial tumors, and ganglion cysts.<sup>6)</sup>

There are several diagnostic modalities available for arterial aneurysms, of which arterial duplex ultrasound is reliable, easily available, and of low-cost modality in children. It also helps in identifying the mural thrombus in the aneurysmal sac with flow present or not and the size of the actual aneurysmal sac.<sup>7)</sup> Most importantly, it can be easily carried out in children without general anesthesia. Like some other noninvasive studies, it has a few drawbacks, which include dependence on the operator and issues such as the noncooperative nature of children. Another modality reported to be more accurate in this condition is conventional arteriography. Arteriography also excludes other anatomical abnormalities in the course of a vessel leading to disruption of the vascular lining.<sup>8)</sup> Magnetic resonance angiography (MRA) is another option, which

will help in detailing the vascular anatomy and differentiating the vascular swelling from other similar swellings, mainly soft tissue sarcomas, synovial tumors, and tendinous and ligamentous structures.<sup>9)</sup> Both conventional arteriography and MRA are difficult to perform in infants because they are only carried out under general anesthesia.

The treatment options for ulnar artery aneurysms include excision of the aneurysm alone, excision and reconstruction with reverse autologous vein grafts, and ulnar artery ligation.<sup>3)</sup> Few reports of thrombin injection have been reported, but ultimately the aneurysm was excised.<sup>10)</sup> The role of thrombin injection is better explained in a pseudoaneurysm of the peripheral arteries. Reconstruction or no reconstruction is also a topic of debate. No reconstruction is opted for if there is a good blood supply from the radial artery or collaterals. However in infants, if expertise of a microvascular surgeon is present, it is ideal to reconstruct, so we have a patent vessel if there is any vascular injury of the same limb further in life. Reconstruction also depends on whether the aneurysm is thrombosed or not and how good the proximal and distal ends of native vessels are at the time of resection.

## Conclusion

This was a rare presentation of an ulnar artery aneurysm that we encountered in an infant. It was post-traumatic, and we opted for its excision because of its progressive increase in size. The decision of reconstruction of the hand vessels was made because of (i) the availability of a hand surgeon in our setup and (ii) the very young age of the patient.

## Disclosure Statement

The authors declare no conflict of interest.

## Author Contributions

Study conception: all authors

Data collection: ZS

Analysis: MAM, SAS

Investigation: MAM

Writing: MAM

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