Case Report

Congenital aneurysm of the palmar digital artery: A case report and literature review

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A B S T R A C T
Spontaneous arterial aneurysms of the hand are uncommon but are well-described in the adult population. In the pediatric population, however, congenital or true aneurysms of the hand are exceptionally rare. A case report and a literature review were performed for published cases of arterial aneurysms of the hand in the pediatric population. A 13-month-old child presented with an aneurysm of the common digital artery and underwent surgical excision without need for reconstruction. Literature review found 13 documented cases. Patient characteristics and management strategies were summarized. There are very few documented cases of hand arterial aneurysms in the pediatric population, with our patient being the third youngest ever reported. No cases were associated with hereditary disease, and aneurysm excision was performed in all cases. Our report highlights the need to include arterial aneurysm in a differential diagnosis when evaluating a pediatric patient with a palpable hand mass.

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I n t r o d u c t i o n
Arterial aneurysms of the hand are uncommon but well-described phenomenon in the adult population, usually in the setting of chronic, repetitive blunt trauma to the ulnar artery (hypothenar hammer syndrome) [1,2]. Nontraumatic causes of aneurysms in adults are associated with atherosclerosis, infection, and other less common causes [3]. Congenital aneurysms or true arterial aneurysms of the hand are

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exceptionally rare in the pediatric population [4–16]. Very little is known regarding their causation and treatment algorithms have not been developed in the pediatric population. We present a report of an isolated aneurysm of the common digital artery in a 13-month-old infant and reviewed the literature on congenital aneurysm to determine what treatment protocols exist for the diagnosis and treatment of these rare conditions.

**Case report**

A 13-month-old healthy boy without a history of trauma presented with an enlarging, pulsatile mass of the right palm (Fig. 1A). Workup included Doppler ultrasonography, which revealed an arterial lesion with a proximal and distal arterial connection, small thrombi within and a characteristic “yin-yang” sign (Fig. 2). Right upper extremity angiogram demonstrated an aneurysm of the common digital artery of the third web space just distal to its takeoff from the superficial palmar arch (Fig. 3). The patient underwent operative exploration utilizing the operating microscope. The common digital nerve (middle and/or ring) was adherent to the outer wall of the aneurysm and required meticulous dissection (Fig. 1B). The 1.5 cm aneurysm of the common digital artery of the third web space was excised. Following resection and tourniquet deflation, there was prompt restoration of perfusion to all of fingers. The distal portion of the common digital artery demonstrated pulsatile back-bleeding after release of the microvascular clamp, and it was subsequently clipped without need for microvascular reconstruction. Pathologic characterization of resected specimen was consistent with a true congenital arterial aneurysm. The patient recovered well from the procedure with full hand function.

**Literature review**

With exclusion of hypothenar hammer syndrome cases from our search, 193 reported cases of hand arterial aneurysms were found in the literature, and only 13 such cases in the pediatric population (Table 1) [4–16]. Average patient age at presentation ranged from 5 months to 18 years (median = 4 years), and none had a prior history of trauma. None had a history of hereditary collagen disease (eg, Ehlers Danlos, Marfan’s Syndrome) or Kawasaki disease. Locations of the aneurysms included: ulnar artery (n = 9), superficial palmar arch (n = 1), and palmar digital artery (n = 2). One pediatric pseudoaneurysm is described. In all cases, management consisted of operative exploration and aneurysm excision with none undergoing interventional procedure to stent or sclerose the aneurysm. Arterial reconstruction was performed in 5 cases, with interposition vein grafts utilized in 3 cases to span longer defects.

*Fig. 1 – (A) Patient presented with an enlarging pulsatile palmar mass. (B) Intraoperative photograph demonstrating 1.5 cm aneurysm of the common digital artery.*
Discussion

Aneurysms can be of the true, involving the entire vessel wall, or false type, or pseudoaneurysm which represented blood collecting between the layers of the vessel wall [3]. Both types of aneurysms rarely occur in arteries of the hand. These small-caliber arteries require high pressures to distend, and are much more likely to be fully transected by penetrating trauma. The most common cause of palmar or digital artery aneurysms is either penetrating trauma for false aneurysms or blunt trauma for true aneurysms [3]. Other less commonly reported etiologies of true arterial aneurysms of the distal upper extremity include infection, atherosclerosis, neurofibromatosis, hemophilia, and congenital deformity [17,3]. The vast majority of unexplained hand aneurysms occur in older patients, and as a result, their cause is often assumed to be chronic blunt microtrauma. Aneurysms of the hand presenting in younger patients, however, are more likely to be described as congenital when a true etiology is unclear.

Of the 13 reported pediatric arterial aneurysm cases found during our literature search, 4 were accompanied by either agenesis or hypoplasia of one or more arteries within the hand [4,6,12,16]. This may have contributed to aneurysm formation in each patient via elevated pressures within the existing arterial system. None of the reported cases were associated with other signs of congenital disease. Common conditions associated with aneurysm formation in larger
arteries include Marfan’s syndrome, Loeys-Dietz syndrome, osteogenesis imperfecta, Ehlers-Danlos syndrome, and other collagenopathies [18]. These conditions predispose to arterial dilatation through abiotrophy, or premature tissue deterioration from usual stresses or strains. For this reason, characteristic aneurysm formation in these patients tends to occur during adulthood in large arteries like the ascending aorta, where pressures and changes in diameter are maximal [19]. Physiologic arterial wall pressures in the hand are substantially below those required to dilate healthy vessels, and it is unlikely that these collagen vascular diseases alone could weaken vessels of the hand to their aneurysmal threshold. Our patient demonstrated no other evidence of collagen vascular disease, nor did imaging show hypoplasia of other arteries within the affected hand.

Patients with arterial aneurysms of the hand most commonly present with a pulsatile mass accompanied by varying degrees of tenderness, numbness and/or tingling and cold intolerance depending on the level of nerve involvement. Ultrasound (US) is the mainstay of initial diagnosis, because it is readily available, cheap, has excellent spatial resolution, and displays imaging in real-time without use of radiation. When hand arterial aneurysms are viewed with color Doppler ultrasonography, a characteristic imaging appearance described as the “yin-yang” sign can be seen—this is caused by the back-and-forth flow typically seen in pseudoaneurysms or saccular aneurysms [20]. Information about the aneurysm and/or pseudoaneurysm neck maybe obtained with US imaging, which is necessary if minimally invasive, percutaneous treatment options such as thrombin injection are being considered.

Magnetic resonance angiography (MRA) can also be utilized for initial diagnosis. Although MRA also does not emit radiation and can potentially characterize vascular anatomic relationships, it has a number of relative disadvantages. Often in young pediatric patients, it requires sedation or general anesthesia, image acquisition can be time-consuming, and the spatial resolution obtained is typically inferior to alternative imaging modalities, especially with regard to hand arterial anatomy and/or aneurysm relationship. Computed tomography angiography with postprocessing techniques provides a high level of detail with satisfactory spatial resolution if the contrast bolus is correctly timed. The challenges of computed tomography angiography however, include the high radiation dose associated with computed tomography imaging—a major issue in pediatric patients and getting the contrast bolus timing right to optimize imaging of the hand vessels. Invasive digital subtraction angiography provides real-time imaging with great spatial resolution and anatomic detail, but is invasive and like MRA, often requires sedation or general anesthesia [21].

Surgical management consists of aneurysm resection with or without microvascular repair or reconstruction. Arterial aneurysm resection alone is sufficient in the presence of intact collateral circulation. Arterial repair and/or reconstruction is indicated if there is any concern about the adequacy of distal perfusion. Although preoperative arteriography can help predict the possible need for arterial repair or reconstruction, the decision is best made intraoperatively following aneurysm resection and tourniquet deflation. The color of the finger(s) can be evaluated, distal flow can be assessed with a hand-held Doppler, and pulsatile back-bleeding can be confirmed from the distal arterial stump. When distal circulation is compromised following aneurysm resection, primary microvascular repair can be performed if the proximal and distal arterial stumps can be mobilized and reapproximated in a tension-free manner. Vein grafting would be required in cases where primary repair is not possible.

Minimally-invasive treatment by using percutaneous US-guided thrombin injection is a recognized technique in appropriately selected cases with a narrow simple communication defect with the normal vessel [22]. The technique is preferable and safer in larger vessels but likely carries a higher risk in smaller vessels with small margins for error, as in our case.

**Conclusions**

Arterial aneurysms of the hand in the pediatric population are very rarely encountered. Our case represents the third youngest patient ever reported, and only the third

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**Table 1 – Patient characteristics of all reported cases of congenital palmar aneurysms and pediatric cases of true palmar aneurysms.**

| Age (yr) | Sex | True/False | Artery | Cause   | References |
|----------|-----|------------|--------|---------|------------|
| 5 mo     | F   | True       | Ulnar  | Congenital | Iyer et al.¹ |
| 8 mo     | M   | True       | Common digital | Congenital | Itoh et al.⁵ |
| 18 mo    | M   | True       | Ulnar  | Congenital | Al-Omran² |
| 1 yr     | M   | True       | Ulnar  | Congenital | Offer and Sully⁶ |
| 2 yr     | M   | True       | Ulnar  | -         | Amjad³ |
| 4 yr     | F   | True       | Superficial palmar arch | Congenital | Deune and McCarthy¹⁸ |
| 4 yr     | F   | False      | -      | Congenital | Lourie¹⁰ |
| 8 yr     | M   | True       | Ulnar  | -         | Witt et al.¹⁰ |
| 12 yr    | M   | True       | Ulnar  | Congenital | Parsa¹⁷ |
| 12 yr    | M   | True       | -      | -         | Rikukawa et al.¹¹ |
| 16 yr    | M   | True       | Common digital | Blunt trauma | Shütze et al.¹⁹ |
| 18 yr    | M   | True       | Ulnar  | Blunt trauma | Green¹² |
|          |     |            | Ulnar  | -         | Martin¹³ |
pediatric patient with an aneurysm involving the common digital artery. No cases found in the literature review were associated with hereditary disease, and aneurysm excision was performed in all cases. Microvascular reconstruction becomes mandatory in cases where there is inadequate collateral circulation following aneurysm excision. Our report adds to the existing body of literature, and highlights the need to include arterial aneurysm in a differential diagnosis when evaluating a pediatric patient with a palpable hand mass.

Ethical approval

This study was exempt from Institutional Review Board from our institution as it is a case report and literature review.

Statement of human and animal rights

All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2000 and 2008.

Statement of informed consent

Informed consent was obtained from the parental guardians in this case report.

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