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Aneurysm of Distal Ulnar Artery in 8 Year-Old Boy – A Case Report.

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ABSTRACT

The aneurysm of hand is an extremely rare condition, especially in children. We report case of 8 year-old boy with a 4-week history of a palmar soft tissue mass, and no record of trauma to this area. Angiography of the left brachial artery revealed a properly developed artery with a dilatation in the area of the hypothenar eminence, as well as four smaller aneurysms located on the digital branches of the superficial palmar arch from second to fifth finger. Following a consultation with a hand surgery specialist, a surgical treatment was proposed to the patient and his parents. The aneurysm was resected. The ulnar artery was reconstructed using an inverted vein graft harvested from the distal saphenous vein. At the 3-month follow-up, the patient had no complains, and both function and sensation of his left hand were assessed to be normal.

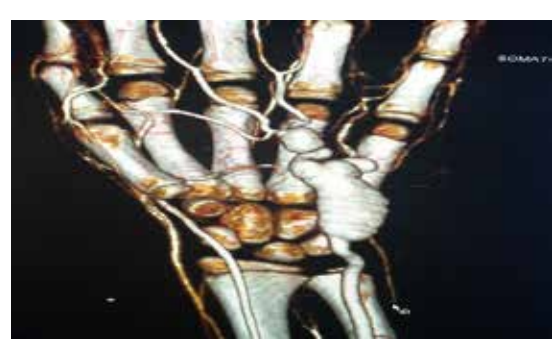
KEYWORDS aneurysm, children, ulnar artery

Introduction:

The aneurysm of the hand is an extremely rare condition, especially in children. A variety of variables, including vessel wall anatomy and potential causative factors, should be considered when clinically differentiating between a true and a false aneurysm (pseudoaneurysm). Majority of seudoaneurysms localized on the hand occur in adults, usually as a result of a penetrating trauma. A hematoma, which initially forms around the damaged vessel wall, is eventually replaced by a fibrous tissue leading to the formation of a false aneurysm.[1]By contrast, a true aneurysm involves the dilatation of all three layers of the vessel wall (intima, media and adventitia), and is most often caused by either blunt trauma or repetitive micro trauma. The anatomical location of the ulnar artery, which in its distant portion (Guyton's canal) is practically subcutaneous, makes this vessel particularly susceptible to trauma and thus aneurysmal formation. When an aneurysm of the distal portion of the ulnar artery coexists with thrombosis, hypothenar hammer syndrome, a rare clinical entity effecting primarily male manual laborers, can be recognized.[2,3] Clinically, aneurysms typically present as tense, usually painless pulsatile masses, located in the palmar region of the hand. High-resolution duplex color Doppler examination is useful in confirming the diagnosis of palmar aneurysm. Duplex color Doppler is a noninvasive test, which allows the examiner to evaluate the damage to the vessel wall and the presence of a hematoma. Angiography is currently the "gold standard" in aneurysmal diagnosis providing crucial information for preoperative planning such as location, size and shape of the arterial lesion in question, as well as the presence of any collateral circulation. [4] Several modalities of treatment for hand aneurysms exist and include both conservative and surgical options. In his or her decision making process the clinician should consider such variables as aneurysm location, etiology and symptoms. Conservative methods of treatment which include a low calorie diet, and intravenous administration of prostaglandin E1 and vasodilatation agents, have been suggested by some as the first line of therapy. On the other hand surgical treatment is preferred in cases of serious damage to the artery. Surgical techniques include excision of the aneurysm followed by either arterial ligature or arterial reconstruction, and depend, in part, on the size and the localization of the aneurysm and on the presence of ischemic symptoms distally from the vessel lesion. [5] Some experts are even more restrictive in their inclusion criteria for surgical intervention, supports surgery only for cases of ulnar artery embolization or fusiform deformity (with or without peripheral symptoms), in order to prevent digital artery embolism.

Case report:

A 8- year-old boy presented with a 4-week history of a palmar soft tissue mass, and no record of trauma to this area, was admitted to the Department of Pediatric Surgery. A clinical examination revealed a soft, pulsatile, non-tender mass approximately 4 cm in diameter in the palmar region of his left hand, and the second, and a second, 1 cm mass in the vicinity of the third metacarpal bone. Pulse on both, the radial and the ulnar artery was normal. Compression of the ulnar artery during Allen's test caused both tumors to collapse. Capillary refill time and sensation of all fingers was normal. Initial diagnosis was vascular malformation. Angiography of the left brachial artery (following the administration of 60 ml of iomeron via the right femoral artery) revealed a properly developed artery with a dilatation in the area of the hypothenar eminence, as well as four smaller aneurysms located on the digital branches of the superficial palmar arch from second to fifth finger.(Fig.1)



Diagnostics was furthered by a contrast enhanced computer tomography which showed an aneurysm of the distal part of the ulnar artery 33 x 13 mm in size and four smaller aneurysm of the superficial palmar arch. (Fig.2)

The deep palmar arch was visible but poorly developed. Following a consultation with a hand surgery specialist, a surgical treatment was proposed to the patient and his parents. The size and location of the aneurysm, its brief history and unclear etiology as well as an absence of detectable microaneurysms (common in the elderly) and no neurological symptoms were all indications for a surgical approach. Intaoperatively a longitudinal left palmar incision extending along the axis of the fourth metacarpal bone, was performed exposing an aneurysm which extended from the pisiform bone to the bifurcation of the third digital common artery. The aneurysm was resected, along with a 5 cm segment of the ulnar artery. Dissection of the aneurysmal wall revealed the presence of a small thrombus. Next, the ulnar artery was reconstructed using an inverted vein graft harvested from the distal saphenous vein. An end-to-end anastomosis was preformed between vein graft and ulnar artery as the same as vein graft and palmar superficial arch. The end-to- side anastomosis was preformed between vein graft and digital common artery. Microsurgical technique and a running 9-0 polyprolene suture were used for all the anastomoses. Suction drainage was placed in the wound and the incision was closed in a typical manner. Postoperatively, a low molecular weight heparin was administered subcutaneously at a dose of 2850 IU AXa for 14 days. Postoperatively, a small area of necrosis developed at the edge of the skin flap, but it did not require further treatment. The patient did not report any sensory or functional deficits. At the 3-month follow-up, the patient had no complains, and both function and sensation of his left hand were assessed to be normal. Furthermore Allen's test was negative, and the ultrasound Doppler scan showed good vessel patency in the area of the vascular reconstruction. A histological examination of the excised mass confirmed it to be an arteriovenous hemangioma.

Discussion: True aneurysms of ulnar artery are extremely rare. In fact, a quick search of the medical literature yields no positive matches. It is clear however that the pathophysiology of this condition is strongly influenced by the unique vascular anatomy of the hand, specifically the small caliber of vessels, which decrease absolute flow, and the thick protective palmar fascia. An ulnar artery aneurysm comorbid with digital ischemia is termed "hypothenar hammer syndrome". Thrombosis formed within the aneurysm can lead to chronic digital ischemia. The signs and symptoms of cold, paresthesia, numbness, cyanosis and palpable tumor in the region of the hypothenar eminence are often first to appear. It is recommended that patients presenting these symptoms undergo extensive diagnostic work-up. However angiography which is currently considered to be the "gold standard" in both, the aneurysmal diagnostic work-up and pre-operative planning, remains controversial due to potential complications, including the risk of distal embolization. Contrast enhanced computer tomography and high-resolution duplex color Doppler ultrasound are also useful for aneurysmal evaluation. Choosing the best treatment modality can be difficult, as many factors must be considered, including first and foremost, the location of the lesion, and, to a lesser extent patient's age and the presence of comorbidities. Most authors recommend a surgical excision of the aneurysm, followed by an end-to-end anastomosis or replacement with an interposition venous conduit. An alternative method involves ulnar artery ligation followed a bypass of the damaged vessel. For patients unfit for surgery, percutaneous embolization of the supply vessel may be a good treatment option but it requires caution as it can lead to aneurysmal enlargement due to the activation of the collateral circulation.[6]

A true aneurysm is formed due to the weakening or ballooning of the vessel wall. In the false aneurysm, the arterial wall is disrupted and a hemorrhage develops in the surrounding tissues. Although the etiology of our patient's aneurysm remains unknown, we assume that it is a result of congenital abnormalities of the arterial wall. This hypothesis was confirmed both histologically and clinically with the discovery of four additional smaller aneurysms in the digital arteries. Although uncommon in children, false aneurysms resulting from injury are extremely difficult to treat surgically. It is commonly believed that in child patients restoration of the normal blood flow is always preferable. Sakamoto and Arai reported on the case of a 4 year old boy with a false aneurysm of the superficial palmar arch which developed ten days following a suturing of a hand wound. The patient was treated surgically, with an en-block removal of the lesion and adjacent vessel, followed by an end-to-end anastomosis using microsurgical techniques. Our patient had neither neurological conditions nor symptoms of micro emboli, which is more typical for the elderly. His young age, coupled with the size and unclear etiology of the aneurysm, were all indications for choosing a surgical treatment option. During the three month follow-up an ultrasound Doppler scan showed good blood flow through the venous graft. Unfortunately regardless of its good patency at the time of the follow-up, we cannot accurately predict its patency in the future. Percentage of permeable vascular grafts in the adult population over a ten year period following the initial surgery is only 28%. Surprisingly this does not appear to affect the actual efficiency of the limb, only influencing the Patient's subjective self - assessment. Our conclusion is that surgery is the preferred method of treatment for aneurysms in children. Microsurgical techniques involving the removal of the lesion and the reconstruction of the artery should be used.

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