

Ulnar artery aneurysm: case report and review of the literature

D. MAZZACCARO, G. MALACRIDA, S. STEGHER, M.T. OCCHIUTO,
M. CALDANA, D.G. TEALDI, G. NANO

SUMMARY: Ulnar artery aneurysm: case report and review of the literature.

D. MAZZACCARO, G. MALACRIDA, S. STEGHER, M.T. OCCHIUTO,
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Aim. We report a case of ulnar and palmar arch artery aneurysm in a 77 years old man without history of any occupational or recreational trauma, vasculitis, infections or congenital anatomic abnormalities. We also performed a computed search of literature in PUBMED using the keywords "ulnar artery aneurysm" and "palmar arch aneurysm".

Case report. A 77 years old male patient was admitted to hospital with a pulsing mass at distal right ulnar artery and deep palmar arch; at ultrasound and CT examination a saccular aneurysm of 35 millimeters at right ulnar artery and a 15 millimeters dilatation at deep palmar arch were detected. He was asymptomatic for distal embolization and pain. In local anesthesia ulnar artery and deep palmar arch dilatations were resected. Reconstruction of vessels was performed through an end-to-end microvascular repair. Histological examination confirmed the absence of vasculitis and collagenopathies. In postoperative period there were no clinical signs of peripheral ischemia, Allen's test and ultrasound examination were normal. At follow-up of six months, the patient was still asymptomatic with a normal Allen test, no signs of distal digital ischemia and patency of treated vessel with normal flow at duplex ultrasound.

Conclusion. True spontaneous aneurysms of ulnar artery and palmar arch are rare and can be successfully treated with resection and microvascular reconstruction.

RIASSUNTO: Aneurisma dell'arteria ulnare: report di un caso e revisione della letteratura.

D. MAZZACCARO, G. MALACRIDA, S. STEGHER, M.T. OCCHIUTO,
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Obiettivo. Riportiamo un caso clinico di aneurisma dell'arteria ulnare e dell'arcata palmare in un uomo di 77 anni con anamnesi negativa per traumi ripetuti, vasculite, infezioni congenite o anomalie anatomiche. Abbiamo anche eseguito una ricerca computerizzata della letteratura in PubMed utilizzando le parole chiave "aneurisma dell'arteria ulnare" e "aneurisma dell'arcata palmare".

Caso clinico. Un paziente maschio di 77 anni è giunto in ospedale con massa pulsante a livello dell'arteria ulnare distale destra e dell'arcata palmare profonda omolaterale. Ecografia e TC con mezzo di contrasto dimostrarono la presenza di aneurisma sacciforme dell'arteria ulnare destra di 35 millimetri e dilatazione di 15 millimetri dell'arcata palmare profonda. Il paziente era asintomatico per segni di embolizzazione distale e non aveva dolore. In anestesia locale è stato sottoposto a intervento chirurgico di resezione delle due lesioni aneurismatiche, con ricostruzione della continuità vasale mediante anastomosi microvascolare termino-terminale. L'esame istologico dei pezzi operatori ha confermato l'assenza di vasculite e malattie del collagene. Dopo l'intervento non vi erano segni clinici di ischemia periferica dell'arto superiore destro; il test di Allen e l'esame ecografico erano normali. Ad un follow-up a sei mesi, il paziente era ancora asintomatico con normale prova di Allen, senza segni di ischemia distale digitale; all'eco-color-Doppler di controllo i vasi trattati erano pervi con flussi normali.

Conclusioni. Gli aneurismi idiopatici dell'arteria ulnare e dell'arcata palmare sono un evento raro e possono essere trattati con successo mediante resezione e ricostruzione microvascolare.

KEY WORDS: Ulnar artery aneurysms - Palmar arch aneurysms - Spontaneous true aneurysms.
Aneurismi dell'arteria ulnare - Aneurismi dell'arcata palmare - Aneurismi idiopatici.

Introduction

Aneurysms of ulnar artery and palmar arch are rare events (1); they are usually related to repetitive trauma to the involved upper extremity (2) or clinical findings in vasculitis such as Behçet's disease (3). They could be found more frequently in young males, and sometimes are related to anatomical abnormalities of the origin of



Fig. 1 - Ulnar artery aneurysm at Duplex ultrasound.

the vessels (4) or infections (5,6). We report a case of true aneurysm of the ulnar and palmar arch artery in a 77 years old man without history of any occupational or recreational trauma, vasculitis, infections or congenital anatomic abnormalities.

Case report

A 77 years old male patient was admitted to hospital with pulsing mass at distal right ulnar artery and deep palmar arch; at ultrasound and CT examination a saccular aneurysm of 35 millimeters at right ulnar artery and a 15 millimeters dilatation at deep palmar arch were detected (Figs. 1, 2 and 3). He was asymptomatic for distal embolization and pain. History was negative for Behcet's disease, vasculitis, collagenopathies, peripheral catheterization, smoking and drug use. He hadn't any previous history of infections, except for hepatitis C virus; however, he hadn't cryoglobulinemia. He hadn't Marfan disease, even if he had a dilatation of ascending aorta. He denied traumatic event involving upper extremity and he was an engineer so we excluded pseudoaneurysm due to occupational micro-trauma. By the clinical history it was a spontaneous true aneurysm of ulnar artery and deep palmar arch. Blood laboratory tests were normal, except for glucose level (113 mg/dL) and a little transaminase elevation (aspartate aminotransferase 63 U/L); he had no eosinophilia. He hadn't neither a history of artery disease nor peripheral obstructions (all arterial pulses were present) and a duplex of carotid artery was normal. At clinical examination of upper extremity radial and ulnar pulses were present bilaterally but there was a pulsing mass synchronous with heart note at the middle third of the right forearm. Auscultation revealed no heart murmure, and transthoracic echocardiography showed no valvular heart disease and a normal ejection fraction. Due to the risk of rupture and distal embolization we decided to treat those aneurysms with a surgical approach; endovascular therapy was not indicated for tortuosity of the vessels, while thrombin injection couldn't be performed because there wasn't a suitable neck. In local anesthesia we performed a resection of ulnar artery and deep palmar arch dilatations (Fig. 4); reconstruction of the vessels was perfor-

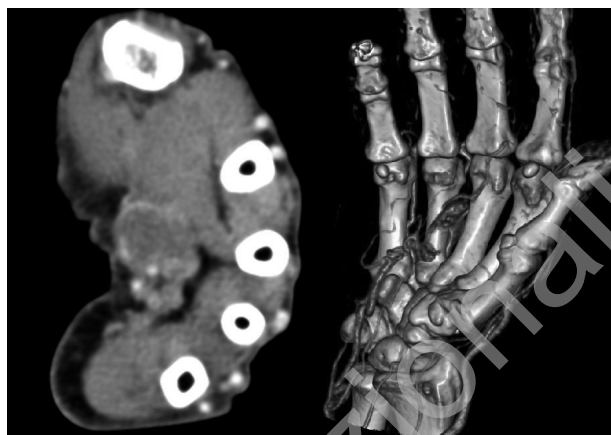


Fig. 2 - Palmar arch aneurysm at CT scan.



Fig. 3 - Ulnar artery aneurysm at CT scan.

med using an end-to-end microvascular repair (Fig. 5). Histological examination of ulnar aneurysm confirmed the absence of vasculitis and collagenopathies. The pathologist described just fibrotic tissue with scleroalinosis and multiple haemorrhagic foci. After the operation there were no clinical signs of peripheral ischemia, Allen's test and ultrasound examination were normal. At a follow-up of six months, the patient was still asymptomatic with a normal Allen test, no signs of distal digital ischemia and patency of treated vessel with normal flow at duplex ultrasound.

Discussion

Distal ulnar artery and palmar arch aneurysms are uncommon clinical findings. They are usually associated with hypotenar hammer syndrome, which is typically an occupationally acquired disease (7); rarely, they could be found as a clinical sign in vasculitis (3) or in case of abnormal vessel conformation (4).

A computed literature search was conducted in PUBMED using the keywords "ulnar artery aneurysm"



Fig. 4 - Intraoperative findings of ulnar (on the left) and palmar arch aneurysms (on the right).

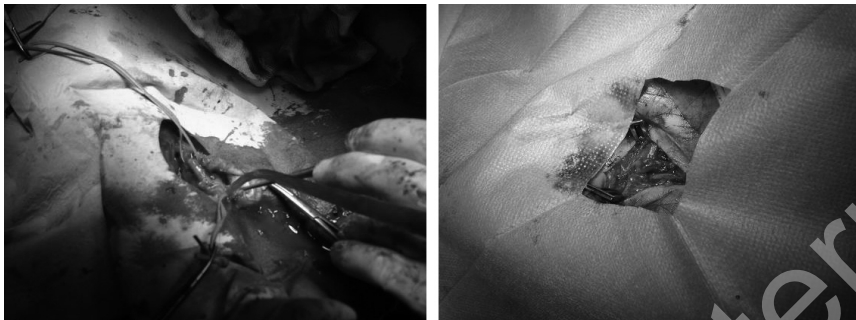


Fig. 5 - Vessel end-to-end reconstruction of ulnar (on the left) and palmar arch arteries (on the right).

and “palmar arch aneurysm”. Most of cases have been reported in young people, usually after violent trauma or repetitive micro-trauma to the involved upper extremity, and they are pseudoaneurysms more frequently, as reported since 1949 in *Journal of The Royal Army Medical Corps*: the literature of the war, so rich in vascular injuries, has however few examples of these lesions (8).

Occupational true aneurysms of the ulnar artery in the palm have been reported as a result of hypothenar hammer syndrome: this nosological entity is an uncommon vascular overuse syndrome that is caused by trauma to the palmar portion of the ulnar artery, usually as a result of occupational or sports activities which involve repetitive micro-trauma on the heel of the hand. Typically, hypothenar hammer syndrome occurs in men with a mean age of 40 years, who are more frequently vibration-exposed workers, such as metal workers, auto mechanics, lathe operators, machinists, miners, sawmill workers, butchers, bakers, brick layers and carpenters (7). In our case, however, the patient hadn't either history of occupational nor recreative micro-trauma, as he worked as an engineer and didn't practice any sports.

True aneurysms have been reported as a result of anomalous vessels' anatomy, such as congenitally hypoplastic ipsilateral radial artery (9) or ulnar artery arising from the axillary artery (4); sometimes ulnar artery aneurysms are thought to be congenital in origin, as reported in the pediatric population (10). Angio-CT scan of our patient indeed showed a normal conformation of the vessels' anatomy and origin and the patient reported the oc-

currence of a pulsating palpable mass of his wrist and palm two years before, and he had been completely asymptomatic.

True aneurysms of ulnar artery and palmar arch can be also clinical findings of vasculitis or collagenopathies. Spontaneous aneurysms of the ulnar artery as a complication of rheumatic vasculitis have been reported since 1958 (11); Seishiro and coll. reported a case of a 54-year-old-man with a non-traumatic pseudoaneurysm of the proximal ulnar artery with eosinophilia (12), while Kisacik and coll. reported a case of a 21-year-old male with a ulnar artery aneurysm and a history of with Behçet's disease (3). Those rheumatic vasculitis have multisystemic involvement and specific clinical findings, such as oral and genital aphthous lesions for Behçet's disease. Moreover, histological samples have typical lymphocyte infiltration of aneurysmatic vessels' walls, with irregular necrotic zones and diffuse interstitial fibrosis. In our case, the patient hadn't any clinical sign of those diseases, and histological samples were not suggestive for vasculitis.

Nguyen and coll. reported a case of ulnar artery aneurysm in a patient with Marfan syndrome (13). This is a multisystem connective tissue disorder, which usually presents with musculoskeletal abnormalities; as for this syndrome, the major cardiovascular manifestation is a progressive dilatation of the ascending aorta, leading to aortic aneurysm formation and eventually to fatal aortic rupture or dissection. As for our case, the patient really had a dilatation of the ascending aorta, but he didn't fit the Gent criteria for the clinical diagnosis (14).

Vasculitides have also been described as secondary to infections (5,6,15), many viruses can be responsible for systemic vasculitis, the most frequent being hepatitis B virus-related polyarteritis nodosa or hepatitis C virus-related mixed cryoglobulinemia (16). Moreover, some bacteria, fungi or parasites can cause vasculitis, mainly by direct invasion of blood vessels or septic embolization: Inoue and coll. reported a case of mycotic aneurysm of the palmar artery associated with infective endocarditis (5), while Bacourt and coll. reported in 1987 the first case of aneurysm of the ulnar artery in *Streptococcus bovis* septicemia (6). Our patient had a history of previous hepatitis C virus infection; however, he hadn't cryoglobulinemia, and microbiological analysis of the vessel's sample was negative for bacteria, fungi and parasites.

The patient probably had a true spontaneous aneurysm of ulnar artery and palmar arch, which indeed is a rare event, as reported in literature.

As for treatment, surgery is the gold standard for large aneurysms which have high risk of rupture or distal embolization (17). Technically, the aneurysm is resected; vascular reconstruction could be performed or not (18), using an end-to-end ulnar artery microvascular repair or a bypass with inverted great saphenous vein. A single case of medical therapy has been reported in 1990

by Rothkopf and coll., using long-term anticoagulants (19), while Komorowska and coll. reported two cases of percutaneous thrombin injection for radial and ulnar artery pseudoaneurysms (20).

We chose to resect the aneurysm because of its risk of rupture and chose to restore vessels' continuity with an end-to-end ulnar artery microvascular repair as the lesion was short and had good proximal and distal diameters. At a follow-up of six months, the patient is still asymptomatic with a normal Allen test and no signs of distal digital ischemia.

Conclusion

True spontaneous aneurysms of ulnar artery and palmar arch are a rare event, which can be successfully treated with resection and microvascular reconstruction.

Competing interests

No financial competing interests.

Institutional review board approval

Our institution approved the report of this case.

References

1. Spartera C. *Chirurgia delle arterie*. 2005, Ed. Masson.
2. Latshaw RE, Weidner W. Ulnar artery aneurysms: angiographic considerations in two cases. *AJR Am J Roentgenol* 1978;131(6):1093-5.
3. Kısacık B, Kasıfoğlu T, Akay S, Yılmaz O, Yılmaz S, Simsek I, Erdem H, Pay S, Dinc A. Ulnar artery aneurysm in a patient with Behçet's disease. *Rheumatol Int* 2010; 30(3):383-5.
4. Pulakunta T, Potu BK, Vollala VR, Gorantla VR, Thomas H. Co-existence of superficial ulnar artery and aneurysm of the deep palmar arch in the hand. *Bratisl Lek Listy* 2009;110(11):738-9.
5. Inoue T, Otaki M, Wakaki N, Oku H. Mycotic aneurysm of the palmar artery associated with infective endocarditis. Case report and review of the literature. *Minerva Cardioangiolog* 2001;49(1):87-90.
6. Bacourt F, Borner P, Fourcade R, Davous N, Lacombe P. Aneurysm of the ulnar artery in *Streptococcus bovis* septicemia. *Presse Med* 1987;16(9):450.
7. Ablett CT, Hackett LA. Hypothenar hammer syndrome: case reports and brief review. *Clin Med Res* 2008;6(1):3-8.
8. Waterston RE. A case of aneurysm of the ulnar artery. *J R Army Med Corps* 1949;93(5):260.
9. Venkatanarasimha N, Manghat NE, Wells IP. Unusual presentation of ulnar artery aneurysm and dissection with associated anomalous radial artery: appearances on multi-detector row CT angiography. *Emerg Radiol* 2007;14(2):101-4.
10. Al-Omran M. True ulnar artery aneurysm of the hand in a 18-month-old boy: a case report. *J Vasc Surg* 2007;45(4):841-3.
11. Makarenko TP, Klimkovich IG. Spontaneous aneurysm of the ulnar artery as a complication of rheumatic vasculitis. *Khirurgiia (Mosk)* 1958;34(3):109-11.
12. Sekino S, Takagi H, Kato T, Matsuno Y, Sekido Y, Umamoto T. Nontraumatic pseudoaneurysm of the proximal ulnar artery with eosinophilia. *J Vasc Surg* 2005;42(6):1233-5.
13. Nguyen DQ, Murison M. Ulnar artery aneurysm in a patient with Marfan's syndrome. *J Plast Reconstr Aesthet Surg* 2006;59(10):1131-2.
14. Dean JC. Marfan syndrome: clinical diagnosis and management. *Eur J Hum Genet* 2007;15(7):724-33.
15. Pagnoux C, Cohen P, Guillevin L. Vasculitides secondary to infections. *Clin Exp Rheumatol* 2006;24(2 Suppl 41):S71-81.
16. Morgan AJ, Schwartz RA. Cutaneous polyarteritis nodosa: a comprehensive review. *Int J Dermatol* 2010;49(7):750-6.
17. Harris EJ Jr, Taylor LM Jr, Edwards JM, Mills JL, Porter JM. Surgical treatment of distal ulnar artery aneurysm. *Am J Surg* 1990;159(5):527-30.
18. Keymeulen K, Van Osselaer G. True aneurysm of the palmar arch. A resection without reconstruction. *Acta Chir Belg* 1995;95(3):144-6.
19. Rothkopf DM, Bryan DJ, Cuadros CL, May JW Jr. Surgical management of ulnar artery aneurysms. *J Hand Surg Am* 1990;15(6):891-7.
20. Komorowska-Timek E, Teruya TH, Abou-Zamzam AM Jr, Papa D, Ballard JL. Treatment of radial and ulnar artery pseudoaneurysms using percutaneous thrombin injection. *J Hand Surg Am* 2004;29(5):936-42.