Dual Aneurysm Formation in an Occupationally Predisposed Male: Atypical Hypothenar Hammer Syndrome

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Abstract
Hypothenar hammer syndrome is typically described as a chronic repetitive injury to the ulnar artery at the level of hamulus of hamate and shows certain occupational predilection. Our case is an occupationally predisposed patient who had two aneurysm formations from the superficial palmar branch of ulnar artery and the digital branch, respectively, diagnosed and treated surgically. We call it an atypical hypothenar hammer syndrome as the arterial involvement was not typically at the level of hamate bone and only corresponded to the site of recurrent friction. This article also discusses the role of imaging and treatment options.

Keywords
► aneurysm
► occupation
► ulnar artery

Key Messages
• Ulnar artery is prone to injury at the wrist level due to a relatively superficial course.
• Spectrum of changes includes “cork screw” configuration of the artery, aneurysm formation, thromboembolism, and ulnar nerve compression.
• Ultrasonography and angiography (conventional/computed tomography/magnetic resonance imaging) are used for diagnosis, whereas the latter is the gold standard.
• Treatment options include conservative to surgical intervention.

Introduction
Hypothenar hammer syndrome refers to changes in the ulnar artery at the level of hook of hamate secondary to repetitive injury as in occupational or sports-related injuries. Changes range from “cork screw” appearance of the artery, aneurysm formation, thrombosis, and embolism to digital arteries resulting in ischemia. We encountered a patient who had two aneurysms involving the distal ulnar artery and a digital branch from superficial palmar arch, respectively, secondary to chronic occupational injury diagnosed with multimodality imaging and managed surgically. To the best of our knowledge, this is the first reported case of dual aneurysm formation involving ulnar artery and branches, showcasing the role of multimodality imaging along with surgical treatment.

Case History
A 62-year-old man presented with swelling in his left central palm and vague episodic pain focally for the last 2 years. The patient is a laborer by occupation and used to carry milk in large utensils having sharp edges using both hands. On examination, the swelling was firm, pulsatile with normal overlying skin. Another focal area of redness was noted at the base of second metacarpal, which was mildly pulsatile. No obvious signs of digital ischemia were
noted. Radial and ulnar artery pulses were normally recordable at the wrist level.

Vascular origin lesion was suspected clinically and contrast-enhanced magnetic resonance imaging (MRI) of the hand was requested, which revealed a well-defined cystic lesion measuring 22 × 19 mm in the mid palm. The lesion was iso- to hyperintense signal on T1-weighted and hyperintense on T2-weighted sequences (►Fig. 1). The lesion was splaying the flexor tendons and was distal to the distal carpal row at the level of base of metacarpals. Another similar lesion was seen measuring 16 mm × 8 mm overlying the base of second proximal phalanx corresponding to the focal skin redness clinically. On dynamic contrast study, homogenous opacification of the lesions was seen. The larger lesion was continuous with the superficial palmar branch of ulnar artery (►Fig. 2) and the smaller lesion was related to a digital branch suggestive of aneurysms. Other distal digital branches appeared normal to the extent seen. MRI was followed by ultrasonography (USG) and Doppler, which showed the aneurysm arising from the superficial branch of ulnar artery. No thrombosis or hematoma was seen. The smaller lesion was partially thrombosed and showed feeder from a digital branch (►Fig. 3, ►Video 1). Radial artery and other digital arteries were unremarkable. On the basis of clinical scenario and imaging, diagnosis of dual aneurysms arising from superficial palmar branch of ulnar artery and digital branch, respectively, was made. The occurrence in this patient is likely the result of chronic occupational injury (continuous friction to the arteries by the sharp edge of the utensil). Digital subtraction angiography (DSA) confirmed the finding, and also revealed few digital arteries arising from the aneurysm (►Fig. 4). The patient underwent surgical excision of the aneurysms with reposition of the digital arteries and ensuring adequate distal flow (►Fig. 5).

**Fig. 1** T1-weighted images of hand in (A) axial at metacarpal base level, (B) axial at proximal phalangeal base, and (C) coronal planes showing two iso- to hyperintense lesions (arrows) on the palmar aspect, the former splaying the flexor tendons.

**Fig. 2** Postcontrast T1-weighted images of hand in (A) axial at metacarpal base level, (B) axial at proximal phalangeal base, and (C) coronal planes showing two homogenously enhancing lesions (arrows) on the palmar aspect, the former splaying the flexor tendons. (D) Postcontrast maximum intensity projection images showing palmar branch of ulnar artery and digital artery (arrows) leading to the aneurysms, respectively.

**Fig. 3** (A and B) Gray scale and Doppler of the palmar lesion show classical “to-and-fro” motion consistent with aneurysm (white arrow). Black arrow shows origin from the ulnar artery. (C and D) Gray scale and Doppler of the lesion at base of index finger appear hypoechoic with partial arterial flow suggestive of partly thrombosed aneurysm.
Occupational change was advised. The patient is currently on follow-up and has no significant symptoms.

Video 1


Discussion

Hypothenar hammer syndrome\(^1\) refers to the spectrum of distal ulnar artery abnormalities including thrombosis, aneurysm, or distal digital emboli and ischemia secondary to repetitive irritation to the artery by the hamulus (hook of hamate) and is typically located at the level of the distal carpal row. It is commonly seen in male patients involving the dominant hand with certain occupational predisposition such as in carpenters, metal workers, and in certain sports such as baseball and golf.\(^2\) The overall incidence of aneurysm formation in hypothenar hammer syndrome is unknown\(^3\) however, in a study by Vays-sairat et al of 17 patients with hypothenar hammer syndrome, 10 patients had aneurysm formation.\(^4\)

Ulnar artery is the terminal branch of brachial artery and at the wrist after passing through the Guyon’s canal; it enters the palm and divides into the superficial and deep palmar branches contributing to superficial and deep palmar arches, respectively. The artery is quite superficial in its course in the palm directly lying above the carpal prominence, hence amenable for continuous friction and trauma. The symptoms include palpable swelling in the palm, pain, digital ischemia, and neuropathic symptoms like paresthesia due to involvement of sensory branches of ulnar nerve that course nearby.\(^5,6\) USG is generally the initial modality and shows thrombosis or aneurysm of the artery. Aneurysm typically shows to-and-fro pattern (yin yang phenomenon) on Doppler and shows continuity with the feeding artery. Distal flow can also be assessed on Doppler studies.\(^7\) Angiography—noninvasive (computed tomography/MRI) or invasive (DSA)—aids in confirming the vascular changes, in addition to therapeutic interventions by the latter.\(^8\) Due to repetitive trauma, the ulnar artery undergoes intimal hyperplasia, fragmentation of internal elastic lamina resulting in characteristic “cork screw” appearance as in alternate ectasia, and stenosis, which can be appreciated on imaging.\(^9\) Angiographic features also include aneurysm formation, occlusion of ulnar artery, and digital arteries.\(^8\) In addition, DSA aids in mapping the palmar arch as some variations require changes in the management.

Our case was atypical in many ways. Even though the cause is occupational, the changes in the ulnar artery (aneurysm) are not typically at the level of hamate but more distal to it (corresponding to the friction by the edge of the utensil) supporting the evidence that the repetitive trauma is the major and sole cause of the changes rather than an underlying arteritis (commonly fibromuscular dysplasia) as proposed by some authors.\(^2,10\) Our case had two aneurysms, again corresponding to the sites of friction. We did not encounter “cork screw” changes in the artery or thrombosis.

Management options depend on the clinical symptoms and range from conservative to resection of the thrombosed

Fig. 4 (A) Selective digital subtraction angiography of radial and ulnar arteries shows large aneurysm (thick white arrow) arising from the superficial palmar branch of ulnar artery (thin arrow). (B) Second aneurysm filling from a digital branch (white arrow). DB, digital branch; DPA, deep palmar arch; RA, radial artery; SPA, superficial palmar arch; SPB, superficial palmar branch of ulnar artery; UA, ulnar artery.

Fig. 5 (A and B) Intraoperative images showing the aneurysm (black arrow) arising from the superficial palmar branch of ulnar artery (arrowhead). White arrow is a digital branch arising from the aneurysm that was reposed. (C) Second aneurysm (black arrow) arising from digital branch of index finger.
segment/aneurysm, including occupational change to prevent further progression, thromboembolism if present managed by use of antiplatelets, heparin infusions, and vasodilators.\textsuperscript{11,12} Surgery is favored in the presence of aneurysms as it can remove the source of emboli and any compressive effect on the nerves and thus obliterate pain.\textsuperscript{1} Our case underwent surgery in view of two aneurysms, which caused pain relief by removing the source of emboli and hence future digital ischemia.

Financial Support and Sponsorship
Nil.

Conflicts of Interest
There are no conflicts of interest.

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