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Case report

Presentation of the hypothenar hammer syndrome as a low incidence aneurysmal disorder of the ulnar artery

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ABSTRACT

Introduction and importance: The hypothenar hammer syndrome (HHS) is either rare or underdiagnosed condition that arises from vascular insufficiency of the ulnar artery in the hand. The most common accused mechanism is repetitive blunt trauma to hypothenar region while activating the latter as a hammer to do the job.
Case presentation: A 48 year old woman worked as a master of university claimed from pain and numbness in her left hand concurrent with a pulsatile mass. The second case was a 48 year old man who was a mechanic engineer in automotive industry that claimed of a pain and pulsatile mass in his non-dominant left hand.
Clinical discussion: Ulnar artery enters the hand from Guyon's canal where fixes to adjacent structure and is susceptible for injury. Aneurysmal formation is an outcome of arterial trauma which could result in distal necrosis if remains untreated.
Conclusion: This study has revealed that even whether a single blunt trauma or writing with pen could lead to aneurysmal formation. It has also showed primary aneurysmal resection with no further surgical procedure is both safe and reliable if digital ischemia is not concurrently present.

1. Introduction

Firstly in 1934 digital ischemia of the medial hand among factory workers due to arterial emboli formation in association with the ulnar artery disorder was described which 36 years later was named as hypothenar hammer syndrome (HHS) [1–4]. This disorder commonly develops when repetitive compression and/or vibration on susceptible segment of ulnar artery in its anatomical passage through the Guyon's canal leads to the artery compromise [3–9]. The basic histopathological reason for the latter is due to arterial intimal layer injury followed by platelets aggregation, luminal occlusion, arterial fragmentation, aneurysmal change, and thromboembolic events [4,10,11]. Although prevalence of the HHS was estimated <1% in general population studies considered that the disorder is underdiagnosed [1,5,8,9]. Signs and symptoms could be varies from asymptomic disease to even digital ischemia and necrosis of the hand which needs amputation [12]. The HHS regards in association with occupational and/or recreational repetitive activities in which hypothenar prominence of the hand compresses almost frequently to hold, grab, push, squeeze, screw, and work with objects. Therefore many field of industrial, constructive, and sport related jobs are susceptible for the HHS [1,3,6]. Diagnosis of the disorder is performed using Doppler sonography, physical examination, and definitively by arteriography [1,3–8]. Treatment is accomplished mostly conservatively, reduce traumatic activity, smoke cessation, medication prescription, and if adequate result does not achieved finally surgery will be planned [3–6]. In this report two HHS cases with aneurysmal change in ulnar artery were introduced according to the SCARE 2020 criteria [13]. Informed consent has been obtained from the patients for publication of the case details and accompanying images. The study also was approved by Institutional Review Board of Ebnesina Applied Science and Technology Educational Center with the registration code IR.TUMS.MED.REC.1399.022.

2. Cases presentation

First case was a 48 year old woman worked as a master of university who had a history of blunt trauma (ground-level falling) to her left palmar region presented with a pulsatile mass in her left hand hypothenar segment since some months prior to refer to our vascular clinic. She felt pain, numbness, and occasionally paresthesia in burden of the

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sensory branches of the ulnar nerve in left hand. She was otherwise healthy. Physical examination revealed a 2 × 2 cm pulsatile mass in hypothenar region that magnified natural hypothenar eminent obviously. Fig. 1 shows this finding. Either ulnar or radial artery pulses were palpable 2+ considering strength scale. The Allen's test was unremarkable and no sign of digital ischemia was present.

Patient had a hand and wrist magnetic resonance arteriography (MRA) exam with herself. The report of the latter contained a 23 × 26 mm ulnar artery aneurysm just beneath the hypothenar muscles with compression features of the underlying ulnar nerve. There was no sign of bones or adjacent connective tissue pathologic involvement. Fig. 2 shows a crossed section of patient's MRA exam with intravenous contrast. Previous conservative therapy including of hand rest, acetylsalicylic acid, and amlodipine was unsatisfactory. Finally surgical resection of the aneurysm was projected for her with diagnosis of the HHS. Preoperative Doppler ultrasound exam was performed to evaluate palmar arch patency and contained normal findings. The procedure was initiated by a vertical incision on hypothenar region, continued with ulnar aneurysmal sac releasing from adjacent elements, and ended with complete resection of the sac with no additional vein graft or end to end arterial anastomosis. Surgical procedure was accomplished with no complications or injury to the nearby organs including the ulnar nerve. Fig. 3 illustrates intraoperative status prior to aneurysmal resection. Permanent pathologic study revealed sections of dilated arterial wall with degenerated media, proliferated intima plus clots formation consisting with arterial aneurysm. Patient's symptoms were completely resolved after surgery and no further complaint was remained after one year follow up.

The second case was a mechanic engineer in automotive industry with 48 years of age. He had positive history for repetitive blunt palmar trauma since several years prior. He claimed of pain and paresthesia in fourth and fifth digits of his left hand in combination with a pulsatile mass on the left palmar hypothenar region. He was neither smoker nor drinker. Also no history of other disease or surgery was remarked. Patient was under conservative treatment including of life style modification, aspirin and atorvastatin consumption for about two years as he remembered, however, there was no recovery occurred. Physical examination revealed a 1.5 × 1.5 cm pulsatile mass in hypothenar region of the patient's left hand. The Allen's test was negative, digital ischemia was absent, and both radial and ulnar artery pulses had a 2+ strength in palpation. Therefore the HHS was highly suspected and upper extremity

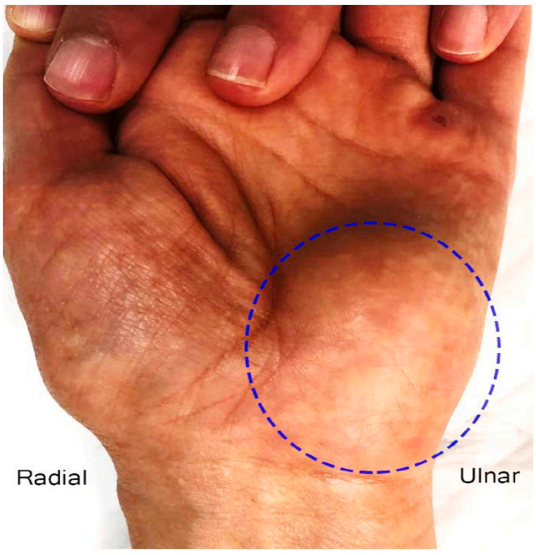

Fig. 1. Magnified hypothenar eminent of the palmar side of the left hand (blue dashes). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

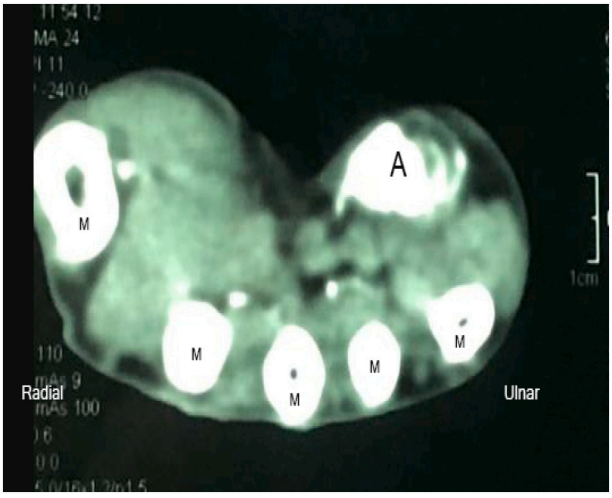

Fig. 2. Crossed section of left hand MRA; A: aneurysm with intimal injury; M: metacarpal bones.

computed tomography angiographic (CTA) study with intravenous contrast was ordered. Fig. 4 shows CTA findings. The HHS diagnosis was confirmed by CTA result that was in lined with the ulnar artery aneurysmal formation at the level of the fourth carpometacarpal joint in coronal view.

According to unfavorable recovery following conservative therapy patient was underwent the surgery following normal findings for palmar arch patency in preoperative Doppler ultrasound exam. Intraoperative photography of the ulnar artery aneurysm was illustrated by Fig. 5. Surgical procedure was accomplished by complete removal of the aneurysmal sac without adjacent elements iatrogenic injury. Additionally no extra procedure including of vein graft or anastomosis was considered.

Pathologic study demonstrated features of arterial aneurysm involving sections of arterial fragments with mural fibrins, hemosiderin macrophages, in addition to focally thin muscular layer. Patient was discharged from the hospital without previous complaints. His physical examination after one year in follow up visit was also unremarkable for the syndrome recurrence.

3. Discussion

Susceptible jobs for the HHS are engine mechanics, machinists, miners, butchers, carpenters, bakers, brick layers, metal workers, sawmill workers, vibration exposed workers, lathe operators, and athletes in volleyball, badminton, karate, hockey, handball, tennis, bicycle riding, golf, and weight lifting [1,3,9]. In this article, we presented a university master with history of one time blunt trauma to her left palmar side one year prior to our visit. She had no history of repetitive hand work except for occasional writing with pen. This type of mechanism whether considering one time trauma or occasional writing with pen have not introduced previously for the HHS formation. Interestingly, acute hyperextension injury to the left ring finger in an engine mechanic and also in a police officer because of using firearm led to the HHS formation [14,15]. Additionally, fracture of the hamate bone and using of walking sticks both could result in the HHS [5,6].

The ulnar artery enters the hand anterior to the flexor retinaculum just between the pisiform and hook of the hamate bone which called the Guyon's canal. In this anatomical zone the artery stabilized by adhesion to adjacent elements. The point where the ulnar artery is susceptible to injury is the segment between deep palmar branch and origin of the superficial palmar arch where the artery is covered by soft tissue layers including the superficial aponeurosis, palmaris brevis muscle, subcutaneous tissue, and skin subsequently [1,5]. Arterial vasospasm and

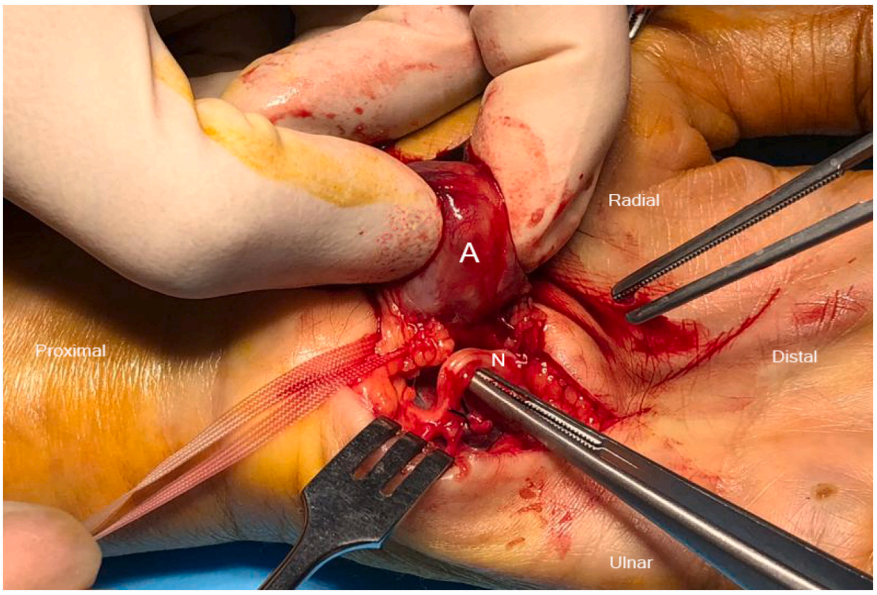

Fig. 3. Intraoperative condition of the aneurysm(A); N:ulnar nerve.

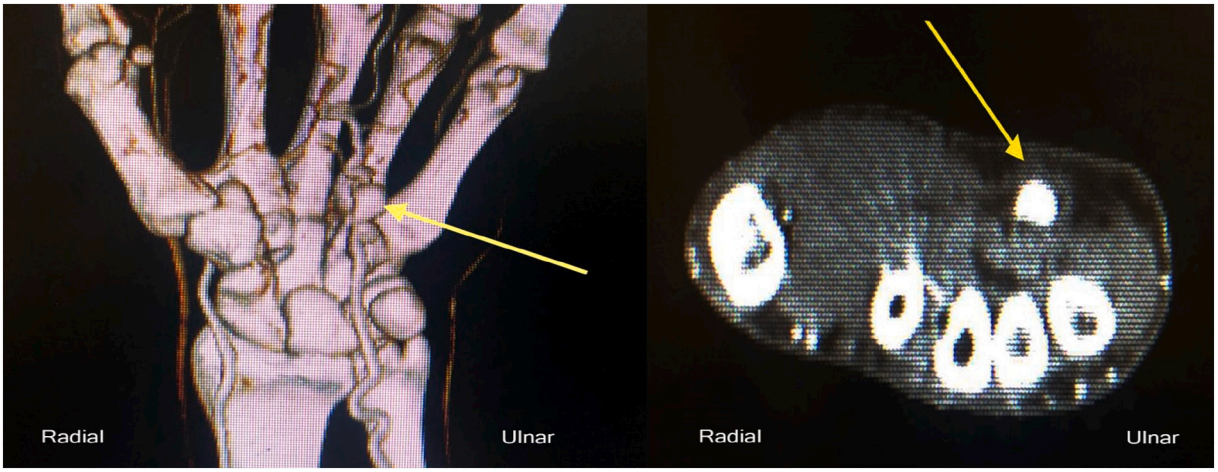

Fig. 4. CT angiography of the ulnar aneurysm; yellow arrow pointed to the ulnar artery aneurysm. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

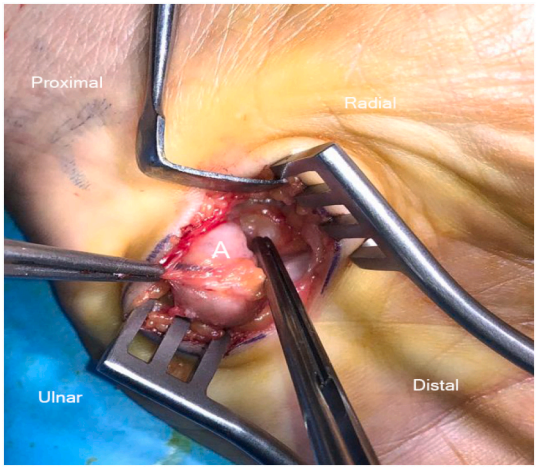

Fig. 5. Ulnar artery aneurysm(A); intraoperative view.

crush injury are the outcome when the hypothenar eminent applies as a hammer and hook of the hamate makes its anvil effect [1,2,10,12]. Histopathologically hyperplasia and irregularity of the intimal tunica occurs followed by internal elastic lamina fragmentation, luminal occlusion with organized thrombi formation, with or without aneurysmal generation [3,11]. These latter changes could be asymptomatic or present with cold hand, severe pain, cold intolerance, ulnar neuropathy, paresthesia, cyanosis, pulsatile mass, and digital ischemia [1,4,5,9]. The HHS could firstly manifests as the Raynaud's phenomenon but in the former there is no hyperemia phase in comparison to the latter. Although the severity of the colour and temperature change in the HHS is higher than in Reynaud's phenomenon [6–9]. Other helpful features that were observed more in the HHS are being male, history of repetitive traumatic injury to wrist and/or hand, asymmetric distribution of symptoms, decreased ulnar and/or radial pulse, digital ulceration, and positive Allen's test [3,9]. Due to involvement of the superficial arterial palmar arch in the HHS, frequently the three medial digits of the hand would be influenced by the disease [9]. Other differential diagnosis of the HHS are including of hand-arm vibration syndrome, Raynaud's disease, systemic lupus erythematous, scleroderma, giant cell arteritis,

Buerger's disease, thromboangiitis obliterans, rheumatoid arthritis, vasculitis, arterial emboli with cardiac origin, and thoracic outlet syndrome [3,5,7]. Prevalence of the HHS among workers and general population was low and reported 1.1–1.3% and 1% respectively [9]. But it seems the syndrome is underdiagnosed due to anatomical variations, subclinical symptoms, and that the physician regarded the HHS as a musculoskeletal disorder [6]. Male smokers are vulnerable for the syndrome [1]. Although the radial artery is less susceptible for injury following blunt trauma it has revealed that vasospasm could be happen secondary to injury to the ulnar artery [1,2]. Overall involvement of right and left hand by the HHS was 53 and 25% respectively; while bilateral involvement was reported 22% [1]. Physical examination clue that is highly suggestive for diagnosis of the HHS is concurrent finding of positive Allen's test and signs of Reynaud's phenomenon except for hyperemic phase [5]. However the Allen's test is negatively false among 17% of subjects [9]. Although Doppler ultrasonography evaluation is helpful and functional the definitive diagnostic exam for the HHS is arteriography of the ulnar artery showing minimal changes of the affected segment of the artery [1]. In current report arteriography was conducted through MR and CT imaging approach which both were either reliable or diagnostic. Treatment of the syndrome is basically directed on conservative therapy including of cessation of offending activities, prevention of exacerbating factors, certain smoking discontinuation, lowering lipid of the diet, prescription of anti-platelet agents, heparin derivate, prostaglandin E1, and calcium canal blockers followed then by cervical sympathectomy if needed [9]. Surgical intervention is reserved for refractory symptoms, poor collateral arterial branching, and in whom conservative therapy is unsatisfactory or digital necrosis is happened [15,16]. Arterial aneurysm formation is another indication for surgery as like for this study cases. Surgical approaches include proximal ligation and resection of the diseased segment with/without further arterial reconstruction by whether vein graft or end to end arterial anastomosis [1,3–6]. We performed aneurysmal resection with no further procedure. Symptoms recurrence rate following conservative therapy is about 28% during 12 months of therapy initiation [1]. Both patients have no signs of disease recurrence after one year of operation. Conclusively the HHS is a complex syndrome involving vessels, nerves, muscles, joints, and digits which is either preventable or curable without needing organ resection if diagnosis occurs prior to its irreversibility phase [3].

4. Conclusion

The HHS is a complex syndrome which could be triggered by single blunt trauma and progressed to arterial aneurysmal formation through the time. In absence of digital ischemia primary resection of the aneurysmal segment of the artery with no further surgical procedure is either safe or reliable when the definitive therapy is considered.

Provenance and peer review

Not commissioned, externally peer-reviewed.

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Ethical approval

This study was performed under supervision of the Ebnesina Applied Science and Technology Educational Center, Tehran, Iran with reference code IR.TUMS.MED.REC.1399.022.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Research registration

N/A.

Guarantor

Abbas Hajian (abbashajian@ymail.com)

CRediT authorship contribution statement

AM: Study design, supervision, chief surgeon, data collection, interpret results.

AH: Study design, surgical first aid, interpret results, article draft.

Declaration of competing interest

The authors declared that they have no competing interests.

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