

Fistulized Pseudoaneurysm Associated with Hypothenar Hammer Syndrome to the Skin in a 12-Year-Old Patient: A Case Report

Metin Onur Beyaz¹, Sefer Kaya¹, and İbrahim Demir²

¹Department of Cardiovascular Surgery, Tayfur Ata Sökmen Medical Faculty, Hatay Mustafa Kemal University, Hatay,

²Department of Cardiovascular Surgery, Kirsehir Education and Research Hospital, Kirsehir, Turkey

In this case, we present a condition where the extension of the hamate hook in the Guyon canal can damage the ulnar artery or its branches, leading to the development of an aneurysm or pseudoaneurysm. The patient, a 12-year-old female, presented to our clinic with a complaint of an uncontrolled palm lump that has been growing for several months and began to bleed in a pulsatile manner after trauma. She was an amateur volleyball player who trained twice weekly for two hours. Color Doppler ultrasound examination revealed a 1.1×0.8 cm pseudoaneurysm in a branch of the ulnar artery. Aneurysmectomy and primary repair were performed. Timely diagnosis and treatment planning are crucial for ulnar artery pseudoaneurysms or aneurysms to prevent ischemic events in a later period.

Key Words: Hypothenar hammer syndrome, Ulnar artery, Skin fistula

Received July 17, 2023

Revised September 30, 2023

Accepted October 10, 2023

Published on November 20, 2023

Corresponding author: İbrahim Demir

Department of Cardiovascular Surgery,
Kirsehir Education and Research Hospital,
Kervansaray Mah., Kirsehir 40200, Turkey
Tel: 90-5444083812

E-mail: ibrahimd128@gmail.com

<https://orcid.org/0000-0003-3813-922X>

Copyright © 2023 The Korean Society for Vascular Surgery

This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/4.0>) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

Cite this article; Vasc Specialist Int 2023. <https://doi.org/10.5758/vsi.230070>

INTRODUCTION

First described in 1934, Hypothenar Hammer Syndrome (HHS), which involves aneurysmal dilatation of the superficial palmar branch of the ulnar artery in the hypothenar region, is exceptionally rare in the pediatric age group [1,2]. The available literature provides limited documentation regarding the frequency of occurrence of HHS. The prevailing occurrence of this medical condition is approximately 14% within the subset of the populace considered susceptible. However, in contradistinction, the frequency of new cases arising among patients exhibiting manual vascular issues is notably confined to a range of 1.1% to 1.6% [3]. The superficial branch of the ulnar artery is susceptible to compression by the prominence of the hamate bone in the Guyon canal, which can lead to symptoms of carpal tunnel nerve entrapment in HHS [4]. In our case, a pediatric patient presented with a skin fistula of the pseudoaneurysm related

to HHS, without signs of nerve entrapment, necessitating urgent surgical intervention.

Due to the retrospective nature of the case report, IRB approval was waived.

CASE

A 12-year-old female presented with persistent palm swelling that had been occurring for several months. She did not report any other specific complaints, and there was no history of prior healthcare facility visits. The patient was referred to our clinic due to sudden-onset bleeding. She had no known systemic illnesses. The patient was an amateur volleyball player, training twice weekly for two hours. On physical examination, both radial and ulnar pulses were palpable. A pulsatile mass, approximately 1 cm in diameter, was observed in the hypothenar region at the time of presentation, at which time active bleeding had ceased (Fig.

1). There were no digital ulcers, cyanosis, or discoloration on the affected side. The Allen test was normal in both hands. Duplex ultrasonography confirmed the presence of a saccular-shaped aneurysm of the ulnar artery. Computed tomography was performed as relatives of the patient declined conventional angiography, and it confirmed the presence of a 1.1×0.8-cm saccular-shaped aneurysm (Fig. 2). Since the patient only reported a pulsatile mass for several months and did not exhibit hypothenar atrophy, hypoesthesia, or adduction deformities at the time of presentation, electromyography (EMG) was not performed; in addition, it was not performed given that the primary concern was bleeding.

After obtaining consent from the patient and her family and following a detailed explanation of the associated risks, a surgical plan was implemented with the patient under general anesthesia, employing a lazy S incision. Due to the risk of bleeding during the procedure, both proximal

and distal areas of the aneurysmal dilatation were explored. The pseudoaneurysm sac, which looked similar to a saccular aneurysm, was excised (Fig. 3), and primary repair was achieved using 8-0 Prolene sutures. Following the operation, the patient was moved to the intensive care unit, where monitoring included assessing oxygen saturation and palmar arch circulation at the levels of the first and fifth distal phalanges, with a focus on the arterial flow continuity (Fig. 4). During the first three months of postoperative follow-up, Doppler ultrasonography examinations of the surgical area did not reveal any new flow abnormalities or aneurysmal dilatations.

DISCUSSION

HHS is a condition characterized by a combination of symptoms that occur after segment occlusion or aneurysmal change of the superficial branch of the ulnar artery or ulnar artery itself in the hypothenar region [5]. This condition commonly affects males in their 5th decade, particularly those in certain occupational groups and athletes, due to chronic repetitive trauma to the superficial branch of the ulnar artery [6]. Aneurysmal dilatation is extremely rare in the pediatric age group [7]. Our patient was a pediatric case with no history of severe trauma but occasionally played volleyball. HHS can manifest as ischemic findings due to distal embolization following aneurysmal dilatation or as peripheral neurological symptoms due to compression [8]. Our case has different clinical characteristics and trauma history than other cases reported in the literature. In our case, although there was aneurysmal dilatation, there were



Fig. 1. Aneurysm sac fistulized to the skin.

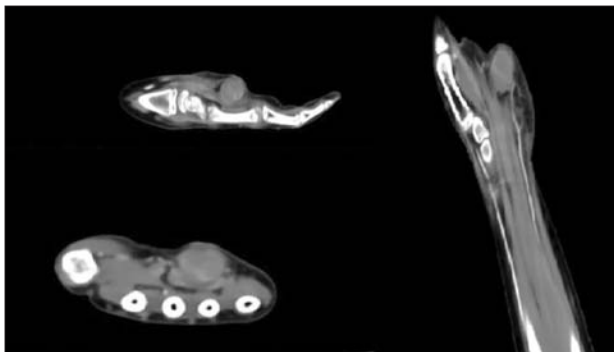


Fig. 2. Computed tomography images showing a 1.1×0.8-cm aneurysm.



Fig. 3. Aneurysmectomy and primary repair with lazy S incision.



Fig. 4. Monitoring of palmar arch circulation at the levels of the first and fifth distal phalanges.

no signs of nerve compression or distal ischemia in classic HHS, and the initial reason for the presentation was a rupture with fistulization to the skin. Additionally, our patient had a history of repetitive trauma that was not as severe as that associated with the use of agricultural tools or hammers.

In cases where the ischemic symptoms are not highly progressive in the classic HHS, conservative methods such as smoking cessation, calcium channel blockers, and hand care in warm environments similar to that used in the management of Raynaud phenomenon can be employed. However, when symptoms are progressive or not tolerable, surgical intervention becomes a priority, and an urgent surgical approach becomes the only option in cases like ours where bleeding occurs [9]. Although there is a lack of randomized controlled trials regarding the optimal surgical approach for the aneurysm change associated with the HHS, it generally involves resection of the aneurysmal segment and revascularization [7]. Regarding the revascularization methods, an interposition graft with an available vein is the most common method; however, end-to-end anastomosis can be used when there is no tension in the vascular structure. Primary repair can also be performed in small but symptomatic aneurysms or pseudoaneurysms. Prior to surgical planning, pulse assessment, Doppler ultrasonography, Allen test, and EMG should be performed. However, in our case, since the patient had no history of neurological symptoms and presented with bleeding, EMG was not performed, and the decision was made to proceed with aneurysm excision and primary repair for vascular continuity.

In summary, this case highlights the risk of ulnar artery

pseudoaneurysm and the possibility of fistulization to the skin, as observed in a 12-year-old female volleyball player and HHS. Early diagnosis, facilitated by color Doppler ultrasound, and prompt aneurysmectomy with primary repair are crucial to prevent ischemic events, emphasizing the significance of timely intervention in such cases.

FUNDING

None.

CONFLICTS OF INTEREST

The authors have nothing to disclose.

ORCID

Metin Onur Beyaz

<https://orcid.org/0000-0001-9338-8152>

Sefer Kaya

<https://orcid.org/0000-0003-2814-5327>

İbrahim Demir

<https://orcid.org/0000-0003-3813-922X>

AUTHOR CONTRIBUTIONS

Concept and design: ID. Analysis and interpretation: SK, MOB. Data collection: SK. Writing the article: ID. Critical revision of the article: all authors. Final approval of the article: all authors. Statistical analysis: none. Obtained funding: none. Overall responsibility: all authors.

REFERENCES

- 1) McCready RA, Bryant MA, Divilbiss JL. Combined thenar and hypothenar hammer syndromes: case report

- and review of the literature. *J Vasc Surg* 2008;48:741-744. <https://doi.org/10.1016/j.jvs.2008.03.055>
- 2) Conn J Jr, Bergan JJ, Bell JL. Hypothenar hammer syndrome: post-traumatic digital ischemia. *Surgery* 1970;68:1122-1128.
- 3) Marie I, Hervé F, Primard E, Cailleux N, Levesque H. Long-term follow-up of hypothenar hammer syndrome: a series of 47 patients. *Medicine (Baltimore)* 2007;86:334-343. <https://doi.org/10.1097/md.0b013e31815c95d3>
- 4) Pagliari M, Menna CR, Christodoulidis A, Soldà M, Molinari M. Unusual case of hypotenar hammer syndrome and carpal tunnel syndrome association. *Acta Biomed* 2018;90:158-161. <https://doi.org/10.23750/abm.v90i1-s.8016>
- 5) Swofford BP, Swofford DP. Management of hypothenar hammer syndrome a case report. *Am J Case Rep* 2018;19:150-152. <https://doi.org/10.12659/ajcr.906849>
- 6) Behar JM, Winston JS, Knowles J, Myint F. Radial artery aneurysm resulting from repetitive occupational injury: tailor's thumb. *Eur J Vasc Endovasc Surg* 2007;34:299-301. <https://doi.org/10.1016/j.ejvs.2007.04.009>
- 7) Amjad I, Murphy T, Zahn E. Diagnosis and excision of an ulnar artery aneurysm in a two-year-old boy. *Can J Plast Surg* 2010;18:e15-e16.
- 8) Çeçen S, Ersen B, Akin S, Tunali O, Aksu İ. Hypothenar hammer syndrome leading to carpal tunnel syndrome in the same hand in a pediatric patient: a rare case. *Türkiye Klinikleri J Case Rep* 2016;24:62-65. <https://doi.org/10.5336/caserep.2015-44981>
- 9) Yuen JC, Wright E, Johnson LA, Culp WC. Hypothenar hammer syndrome: an update with algorithms for diagnosis and treatment. *Ann Plast Surg* 2011;67:429-438. <https://doi.org/10.1097/sap.0b013e31820859e1>