

Carpal Tunnel Syndrome Due to Median Nerve Cavernous Hemangioma; A Case Report

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ABSTRACT

Intraepineural hemangiomas are extremely rare. Only 13 cases of median nerve hemangiomas have been reported in the literature. We report a case that presented with bilateral paresthesia, hypoesthesia and pain at median nerve sensory area. During the operation an intra-neural hemangioma was noticed on the left side. After total excision of the mass and release of the median nerve, there were no recurrence of symptoms after 5-months post-operative follow-up period. Histological examination of the mass confirmed the diagnosis of cavernous hemangioma. Although rare, any mass in the vicinity of the median nerve should be carefully examined and peripheral nerve hemangiomas should be considered in the differential diagnosis of carpal tunnel syndrome.

INTRODUCTION

Carpal tunnel syndrome is a common disease seen at a rate of 1% to 5% in general population with common morbidities like diabetes.^[1] Compared to other etiologies, median nerve cavernous hemangioma is one of the rarest causes of carpal tunnel syndrome. There are only 13 patients reported in the literature (2 male, 11 female).^[2-12] Case reports are important in understanding the pathophysiology, the presentation, course and clinical findings of the disease in such extremely rare cases. As a rare cause of this common syndrome, cavernous hemangioma needs to be considered in the differential diagnosis requiring further examination.

CASE REPORT

In September 2024, a 73-year-old woman presented to outpatient clinic with hypoesthesia and paresthesia on

bilateral hands for 18 months. The patient described hypoesthesia on bilateral first, second and third fingers and lateral – radial side of fourth finger, and had a history of pain with daily activities on bilateral hands for the last six months. The patient did not describe any weakness during last 18-months period. Bilateral grip and muscle strengths of fingers was 5/5. Tinel's and Phalen's signs were positive both of left and right side. Additionally, the patient did not have any palpable mass both of forearms, hands and wrists on examination. There was no trauma and relevant medical disease in anamnesis. According to the EMG tests and physical examination the patient was diagnosed with bilateral carpal tunnel syndrome, severe on the left side, moderate on the right side. No imaging test was used due to cost effective reasons.

Under general anesthesia and pneumatic tourniquet, cardinal line of left hand was determined and marked for in-



Figure 1. Intraoperative image (Arrow 1: The median nerve, arrow 2: The mass).

cisions in operating room. After suitable incisions, subcutaneous soft tissue was exposed and dissected till palmar fascia. Palmar fascia was released, and transverse carpal ligament was fibrotic and released by suitable incision with preserving the median nerve. After the dissection of the transverse carpal ligament, an intraepineural extra-fascicular 5x5x4mm tumoral mass with vascular component was inspected on the median nerve (Fig. 1). Using the microscopic magnification, the mass was removed and epineural sheet was repaired delicately in order to prevent any neurological deficit. Post operative histological examination confirmed that the tumor was a cavernous hemangioma.

After the surgical procedure, symptoms were relieved immediately, with no neurological deficit. The patient was followed-up with an interval of two weeks for 5-months and no recurrence was observed both on left and right hands.

DISCUSSION

The epineurium, the perineurium and the endoneurium are the three connective tissue compartments of the peripheral nerve covering from outside to inside. There is a wide spectrum of benign to highly malignant tumors that affects these connective tissue compartments.^[13] Cavernous hemangiomas are one of the benign and rare lesions of peripheral nerves.

All reported cases of cavernous hemangiomas of the median nerve are presented with carpal tunnel syndrome symptoms.^[2-12] Even though our patient did not present with a mass on the volar side of the wrist, ten of the reported cases were presented with mass on the relevant area at the physical examination. Preoperatively ultrasound and MRI imaging is helpful for evaluating the tumoral pathologies of peripheral nerve and its correlation with the surrounding soft tissue and musculotendinous structures.^[14] Even though, ultrasound and EMG are used to diagnose the carpal tunnel syndrome, American Academy of Orthopaedic Surgeons additionally recommends the use of Six-Item Carpal Tunnel Syndrome Symptoms Scale which contains numbness in the median nerve territory, nocturnal numbness, thenar atrophy or weakness, positive Phalen's test, loss of 2-point discrimination and positive Tinel sign.^[15] In our clinical practice, we primarily use physical examination and EMG tests for diagnosing patients with carpal tunnel syndrome. USG and MRI imaging tests are used for limited number of patients due to cost effective reasons.

As in our experience the cavernous hemangiomas of the median nerve may cause carpal tunnel syndrome, but the cavernous hemangiomas mas also present with Reynald syndrome as reported in one case.^[6]

In the literature, there are only 13 cases reported for median nerve cavernous hemangioma between 1913 and 2017.^[2-12] Due to reported cases, 85% of the patients were female and only two of the patients were male. The age range of the patients was variable. The eldest patient was 64-year-old male, followed by 43 and 35-years. The rest of patients were younger than 22 years. The presented patient is 73-year-old female. Due to the literature the tumor is mostly located in the carpal tunnel, and only one of the reported cases was located at the proximal 1/3 of the forearm.

In our case, we followed-up the patient for 5 months with 2 weeks intervals in outpatient clinic. There was no recurrence of the carpal tunnel syndrome symptoms and no neural deficit due to surgical trauma, and no palpable mass on physical examination. Due to reported literature, there were recurrence in 4 of the 13 cases within the first six months in the post operative period. 9 of these patients had no recurrence in the post operative follow-up period ranging from three months to three years.^[11]

Conclusion

Cavernous hemangiomas should be considered on the differentials on the patients presented with carpal tunnel syndrome in pediatric, adolescents and adult patients. There is no clear recommendation regarding the management of the peripheral nerve tumors in the current guidelines. The patients with cavernous hemangiomas may present with palpable mass on physical examination. For patients with no palpable mass, preoperative ultrasound imaging is more useful than MRI to visualize the mass causing the carpal tunnel syndrome. For patients with no imaging tests, the presence of the mass will be determined

during the surgery and this may require more meticulous dissection of the mass. In every case, microscopic magnification should be used during the dissection to make sure that the entire mass is removed effectively.

Informed Consent

Retrospective study.

Peer-review

Externally peer-reviewed.

Authorship Contributions

Concept: B.M.; Design: A.B.I.; Supervision: G.F.; Fundings: B.M.; Materials: A.K.Y.; Data: K.C.; Analysis: B.M.; Literature search: B.M.; Writing: B.M.; Critical revision: G.F.

Conflict of Interest

None declared.

REFERENCES

1. Padua L, Cuccagna C, Giovannini S, Coraci D, Pelosi L, Loreti C, et al. Carpal tunnel syndrome: Updated evidence and new questions. *Lancet Neurol* 2023;22:255–67. [CrossRef]
2. Kojima T, Ide Y, Marumo E, Ishikawa E, Yamashita H. Haemangioma of median nerve causing carpal tunnel syndrome. *Hand* 1976;8:62–5. [CrossRef]
3. Peled I, Iosipovich Z, Rouso M, Wexler MR. Hemangioma of the median nerve. *J Hand Surg Am* 1980;5:363–5. [CrossRef]
4. Petrovici V. Cavernous hemangioma of the palm with symptoms resembling carpal tunnel syndrome. *Z Plast Chir* [Article in German] 1980;4:40–7. [CrossRef]
5. Patel CB, Tsai TM, Kleinert HE. Hemangioma of the median nerve: A report of two cases. *J Hand Surg Am* 1986;11:76–9. [CrossRef]
6. Prosser AJ, Burke FD. Haemangioma of the median nerve associated with Raynaud's phenomenon. *J Hand Surg Br* 1987;12:227–8. [CrossRef]
7. Coessens B, De Mey A, Lacotte B, Vandebroek D. Carpal tunnel syndrome due to an haemangioma of the median nerve in a 12-year-old child. *Ann Chir Main Memb Super* 1991;10:255–7. [CrossRef]
8. Louis DS, Fortin PT. Perineural hemangiomas of the upper extremity: Report of four cases. *J Hand Surg Am* 1992;17:308–11. [CrossRef]
9. Oztekin HH. Carpal tunnel syndrome due to a cavernous hemangioma of the median nerve. *Acta Orthop Traumatol Turc* [Article in Turkish] 2003;37:170–2.
10. Doğramaci Y, Kalaci A, Sevinç TT, Yanar AN. Intra-neural hemangioma of the median nerve: A case report. *J Brachial Plex Peripher Nerve Inj* 2008;3:5.
11. Vekris MD, Stafilas KS, Zacharis KX, Xenakis TA, Soucacos PN, Beris AE. Intrinsic haemangioma of the median nerve: Report of a case and review of the literature. *Microsurgery* 2008;28:89–90. [CrossRef]
12. Al-Garnawee M, Najjar M. Median nerve cavernous hemangioma. *Basic Clin Neurosci* 2017;8:255–9. [CrossRef]
13. Belakhova SM, Rodriguez FJ. Diagnostic pathology of tumors of peripheral nerve. *Neurosurgery* 2021;88:443–56. [CrossRef]
14. Chick G, Hollevoet N, Victor J, Bianchi S. The role of imaging in isolated benign peripheral nerve tumors: A practical review for surgeons. *Hand Surg Rehabil* 2016;35:320–9. [CrossRef]
15. American Academy of Orthopaedic Surgeons. Management of carpal tunnel syndrome. Evidence-based clinical practice guideline. Available at: <https://www.aaos.org/cts2cpag>. Accessed Feb 9, 2025.

Median Sinir Kavernöz Hemanjiyomuna Bağlı Median Sinir Hemanjiyomu; Olgu Sunumu

İntraepinöral hemanjiyomlar oldukça nadirdir. Literatürde median sinir kavernöz hemanjiyomlarına bağlı sadece 13 vaka bildirilmiştir. Bu çalışmada bilateral parestezi, hipoestezi ve median sinir duyuşal alanında ağrı ile başlayan bir vakayı bildirmekteyiz. Hastanın ameliyatı sırasında sol el median sinirde intranöral kitle tespit edildi. Kitlenin tamamen çıkarılması ve median sinirin serbestleştirilmesinden sonra, hastanın 5 aylık postoperatif takip süresinden sonra semptomlarda tekrarlama olmadığı görüldü. Çıkarılan kitlenin histolojik incelemesi sonrası kavernöz hemanjiyom tanısı doğrulandı. Literatürde nadir olmakla birlikte, median sinirin çevresinde tespit edilen herhangi bir kitle dikkatlice incelenmeli ve karpal tünel sendromunun ayırıcı tanısında periferik sinir hemanjiyomları göz ardı edilmemelidir. Çıkarılan kitle mutlaka histopatolojik olarak incelenmelidir.

Anahtar Sözcükler: Karpal tünel; kavernöz hemanjiom; median sinir.