

Carpal tunnel syndrome caused by synovial osteochondromatosis of the finger flexor tendon

A case report

Dong Rak Kwon, MD, PhD^a, SeungBum Chae, MD, PhD^b, Yong Suk Moon, PhD^c, In Ho Woo, MD^{a,*}

Abstract

Rationale: Carpal tunnel syndrome (CTS) is the most common peripheral nerve neuropathy resulting from compression of the median nerve as it traverses the carpal tunnel. The pathophysiology of this condition is multifactorial, and majority of cases of CTS are idiopathic. We report cases of CTS caused by synovial osteochondromatosis (SOC), which has rarely been reported.

Patient concerns: A 45-year-old female was admitted to the clinic due to right hand tingling sensation for 4 months. On physical examination, the patient's symptoms and signs corresponded to the median nerve entrapment at wrist. However, there is mild swelling and tenderness around the second metacarpal bone. Pain was aggravated during wrist and finger flexion.

Diagnoses: An electrodiagnostic study revealed CTS. She was advised to begin splinting the hand using a wrist brace and to undergo physiotherapy. After 2 weeks, the tingling sensation decreased slightly. However mild swelling and tenderness around the second metacarpal bone did not improve. Ultrasonography showed multiple echogenic foci. Magnetic resonance imaging (MRI) revealed a nodule at the proximal metacarpal level with synovial thickening, enhancement, and a calcified shadow close to the flexor tendon. After confirming the presence of an osseous nodule with synovial thickening, the patient underwent surgery.

Interventions: Carpal tunnel release and mass excision with synovectomy of the adjacent structures were performed. Histologically, the lesion was compatible with a diagnosis of SOC.

Outcome: The symptoms have improved.

Lessons: CTS due to SOC on finger flexor tendon is rare but should be considered for possible etiology. Appropriate clinical examination, plain radiography, ultrasonography, and MRI will help physicians to diagnose this condition. In this paper, we report the successful diagnosis and treatment of CTS caused by SOC within the finger flexor tendon.

Abbreviations: CTS = carpal tunnel syndrome, SOC = synovial osteochondromatosis.

Keywords: carpal tunnel syndrome, etiology, synovial osteochondromatosis

1. Introduction

Carpal tunnel syndrome (CTS) is the most common peripheral nerve neuropathy resulting from compression of the median nerve as it traverses the carpal tunnel. The pathophysiology of this condition is multifactorial. Chammas et al classified etiology of CTS as idiopathic and secondary CTS.^[1] Majority of cases are classified as idiopathic cases.^[2] Less frequently, benign tumors

that affect nerve itself have been reported. Lipofibroma of median nerve was common among tumors, and neurilemmomas and hemangioma have been reported.^[3] Here, we present a case involving synovial osteochondromatosis (SOC) of the hand flexor tendon sheath. SOC may result in symptoms of pain, swelling, and limited motion of the affected joint. We also report the diagnostic and treatment process for CTS due to SOC within the finger flexor tendon.

Editor: N/A.

This research was supported by Basic Science Research Program through the National Research Foundation of Korea (NRF) funded by the Ministry of Education (NRF-2016R1D1A1B01014260).

The authors have no conflicts of interest to disclose.

^a Department of Rehabilitation Medicine, ^b Department of Orthopaedic Surgery, ^c Department of Anatomy, Catholic University of Daegu School of Medicine, Daegu, South Korea.

* Correspondence: In Ho Woo, Department of Rehabilitation Medicine, Catholic University of Daegu School of Medicine, 33, Duryugongwon-ro 17-gil, Nam-gu, Daegu 42472, Korea (e-mail: winho606@naver.com).

Copyright © 2018 the Author(s). Published by Wolters Kluwer Health, Inc. This is an open access article distributed under the terms of the Creative Commons Attribution-Non Commercial-No Derivatives License 4.0 (CCBY-NC-ND), where it is permissible to download and share the work provided it is properly cited. The work cannot be changed in any way or used commercially without permission from the journal.

Medicine (2018) 97:52(e13943)

Received: 21 July 2018 / Received in final form: 4 December 2018 / Accepted: 10 December 2018

<http://dx.doi.org/10.1097/MD.0000000000013943>

2. Case presentation

A 45-year-old woman presented at our institution with a 4-month history of tingling sensation on her right hand. There was no history of trauma. Sensory nerve conduction studies revealed prolonged distal latency (4.38 ms) with reduction in the amplitude (11.5 uV) of the right median sensory nerve action potential. Motor nerve conduction studies revealed a prolonged distal motor latency (5.73 ms) across the carpal tunnel with reduced amplitude (9 mV). The patient was diagnosed with CTS. She was advised to begin splinting the hand using a wrist brace to prevent prolonged flexion or extension and to undergo physiotherapy. After 2 weeks, the tingling decreased slightly. Despite treatment, the mild swelling and tenderness around the second metacarpal bone did not improve. Pain was aggravated during wrist and finger flexion. Ultrasonography showed multiple echogenic foci with shadowing at the proximal metacarpal level of the flexor tendon of the index finger measuring 11.9 × 9.6 mm in diameter (Fig. 1). A bone scan

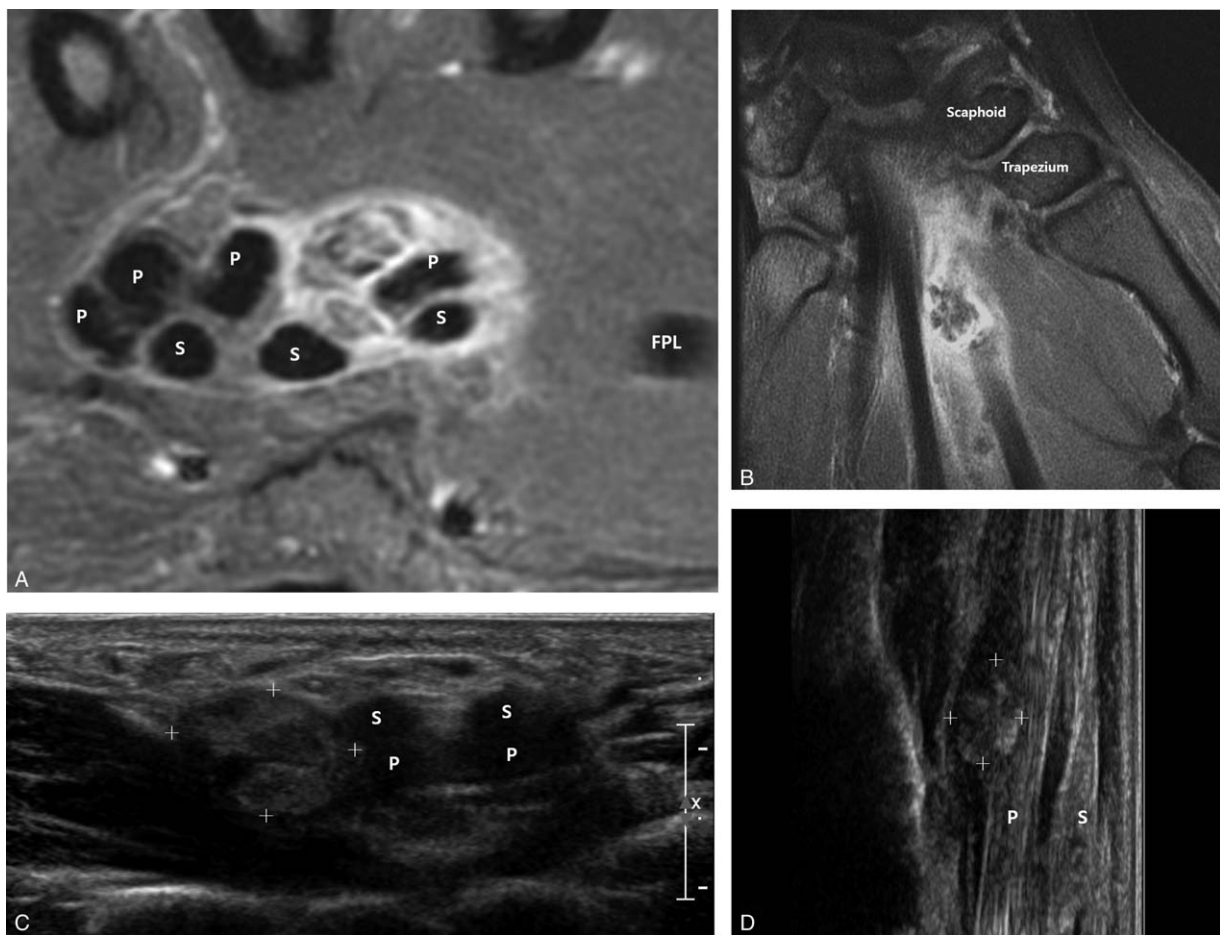


Figure 1. Synovial osteochondromatosis of the right hand in a 45-year-old woman presenting with a tingling sensation. (A, B) T1-weighted contrast enhanced axial (A) and coronal (B) radiographs showing synovial thickening and an osteochondral fragment surrounding the second flexor tendon. (B) Image of an osseous nodule at the proximal metacarpal level. (C) Transverse ultrasound of the right side of second flexor tendon showing multiple echogenic foci with shadowing (cross marks). (D) Longitudinal ultrasound showing multiple echogenic foci with shadowing (cross marks) beneath the flexor tendon. S = flexor digitorum superficialis tendon, P = flexor digitorum profundus tendon, FPL = flexor pollicis longus tendon.

demonstrated increased uptake of the right second metacarpal bone that was ascribed to a recent fracture or osteomyelitis as a differential diagnosis. Magnetic resonance imaging (MRI) revealed a nodule at the proximal metacarpal level with synovial thickening, enhancement, and a calcified shadow close to the flexor tendon.

After confirming the presence of an osseous nodule with synovial thickening, the patient underwent surgery. Intraoperative exploration showed that the nodule was attached to the flexor tendon of the index finger. The flexor tendon sheath appeared to be swollen with adhesions between adjacent structures. There was migration of the mass into the carpal tunnel during finger flexion. We excised the mass with synovectomy of the adjacent structures and released the carpal tunnel. Histologically, the lesion was composed of calcified nodules. A mineralized chondroid nodule, covered by fibrous tissue, was also visible. These features were compatible with a diagnosis of SOC (Fig. 2).

After the operation, the patient experienced complete resolution of her symptoms. At the follow-up 2 months after surgery, the patient remained symptom-free with no recurrence of the mass.

3. Discussion

SOC is a rare condition caused by metaplasia of the subsynovial connective tissue and involves the development of cartilage in the

synovial membrane of bursae, joints or tendon sheaths.^[4] This condition typically involves the monoarticular joint, with large joints being frequently affected. 60% to 70% of cases involve knee joint and the shoulder, elbow, and hip follow with respect to frequency of involvement.^[4,5] However, SOC rarely affects the temporomandibular joint, spinal facet joint, acromioclavicular joint, metatarsophalangeal/interphalangeal joint, wrist joint, ankle joint, or biceps tendon and so on.^[6–8]

In this case, SOC affected the flexor tendon sheath with the patient complaining of a tingling sensation in the thumb and index finger. It appeared that a swelling associated with the lining of the flexor tendon resulted in changes to the carpal tunnel. Intraoperative findings revealed a mass that was entrapped during finger flexion and thickening of the transverse ligament. The clinical features of SOC are known to include pain, swelling, mechanical symptoms, and reduced joint function caused by compression of the mass and the development of loose bodies. In our current case, the patient complained about tenderness and mild swelling.^[8] Diagnosis of SOC is confirmed by histopathology, hyaline cartilage nodule under the synovial tissue of the tendon sheath. Pathological changes can be identified in the radiograph, ultrasound, and MRI. In general, X-ray examination shows multiple calcified nodules.^[9] However, our X-ray examination did not reveal any alteration. An ultrasound

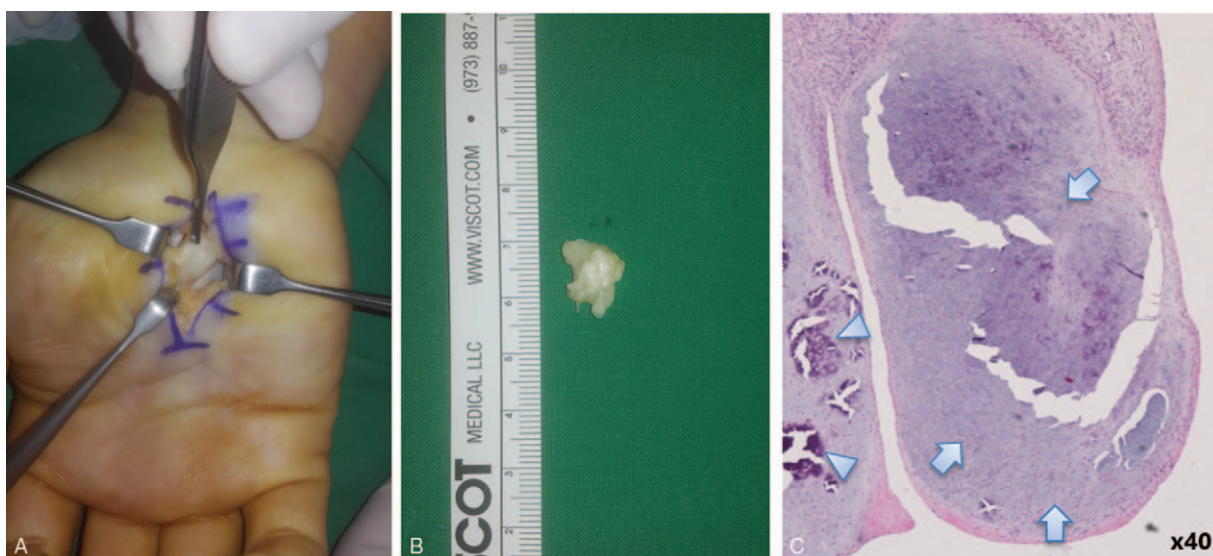


Figure 2. Images of extra-articular synovial osteochondromatosis. (A) Intraoperative view presenting an osteochondral fragment surrounding the second flexor tendon. (B) Debrided specimen measuring $1.5 \times 0.7 \times 0.6$ cm. (C) Microphotograph showing a calcified nodule (arrow head) and mineralized chondroid nodule (arrow) covered by fibrous tissue (hematoxylin and eosin staining, $\times 40$).

examination revealed osteochondral nodules with hyperechoic foci and acoustic shadowing.^[5]

Treatment options usually involve the removal of loose bodies with or without removal of the synovial membrane. Existing literature currently recommends synovectomy with removal of loose bodies in the presence of active synovitis.^[8] The recurrence rate after surgery ranges from 3.2% to 22.2%.^[10] In our current case, we performed excision of the mass and released the carpal tunnel. At the 2-month follow-up, the patient reported no recurrence of symptoms. However, previous studies have reported recurrence at 18 months after the initial mass excision and synovectomy, and recurrence at 5 years after removal of loose bodies with synovectomy.^[11,12] Therefore, our patient may require regular follow-up.

In summary, although SOC in the hand is an extremely rare condition, such cases should not be neglected as a potential cause of median nerve compression in the carpal tunnel. Appropriate clinical examination, plain radiography, ultrasonography, and MRI will help physicians to diagnose this condition. In this paper, we report the successful diagnosis and treatment of CTS caused by SOC within the finger flexor tendon.

4. Method

This was a case report. Ethics committee or institutional review board approval was not obtained. It was not necessary for the case report. The patient signed informed consent for the publication of this case report.

Author contributions

Conceptualization: Dong Rak Kwon.

Data curation: Yong Suk Moon, Dong Rak Kwon.

Formal analysis: Yong Suk Moon, Dong Rak Kwon.

Investigation: Dong Rak Kwon, SeungBum Chae.

Project administration: In Ho Woo, Dong Rak Kwon.

Writing – original draft: In Ho Woo, Dong Rak Kwon.

Writing – review & editing: In Ho Woo, Dong Rak Kwon.

References

- [1] Chammas M, Boretto J, Burmann LM, et al. Francisco carlos dos santos neto, jefferson braga silva. Carpal tunnel syndrome—Part I (anatomy, physiology, etiology and diagnosis). *Rev Bras Ortop* 2014;49:429–36.
- [2] Stevens JC, Beard CM, O Fallon WM, et al. Conditions associated with carpal tunnel syndrome. *Mayo Clin Proc* 1992;67:541–548.
- [3] Johnson J, Kilgore E, Newmeyer W. Tumorous lesions of the hand. *J Hand Surg* 1985;10A:284–6.
- [4] Sim FH, Dahlin DC, Ivins JC. Extra-articular synovial chondromatosis. *J Bone Joint Surg Am* 1977;59:492–5.
- [5] McKenzie G, Raby N, Ritchie D. A pictorial review of primary synovial osteochondromatosis. *Eur Radiol* 2008;18:2662–9.
- [6] Sakamoto A, Naka T, Shiba E, et al. Extra-articular tenosynovial chondromatosis of the finger: A case series study of three cases, one including excessive osseous invasion. *Open Orthop J* 2017;11:417–23.
- [7] Fetsch JF, Vinh TN, Remotti F, et al. Tenosynovial (extraarticular) chondromatosis: an analysis of 37 cases of an underrecognized clinicopathologic entity with a strong predilection for the hands and feet and a high local recurrence rate. *Am J Surg Pathol* 2003;27:1260–8.
- [8] Khadilakar MS, Patil AA, Shah NS, et al. Extra-osseous tenosynovial chondromatosis of the middle finger: a case report. *J Orthop Surg* 2012;20:406–8.
- [9] Murphey MD, Vidal JA, Fanburg-smith JC, et al. Imaging of synovial chondromatosis with radiologic-pathologic correlation. *Radiographics* 2007;27:1465–88.
- [10] Loonen MP, Schuurman AH. Recurrent synovial chondromatosis of the wrist: case report and literature review. *Acta Orthop Belg* 2007;71:230–5.
- [11] Rogachefsky RA, Zlatkin MB, Greene TL. Synovial chondromatosis of the distal radioulnar joint: a case report. *J Hand Surg* 1997;22-A:1093–7.
- [12] Roulot E, Le Viet D. Primary synovial osteochondromatosis of the hand and wrist. Report of a series of 21 cases and literature review. *Rev Rhum Engl Ed* 1999;66:256–66.