Journal of Hand Surgery Global Online 5 (2023) 841–842

Contents lists available at ScienceDirect

Journal of Hand Surgery Global Online

journal homepage: www.JHSGO.org

Case Report

Tremor Induced by Focal Peripheral Nerve Entrapment: A Case Series

Bianief Tchiloemba, MD, [*,†] Min Cheol Chang, MD, [‡] Benjamin Ferembach, MD, [§] Elisabet Hagert, MD, [‖,¶] Jean Paul Brutus, MD [*]

[*] *Exception MD Clinic, Montreal, Canada*
[†] *Department of Plastic Surgery, University of Calgary, Calgary, Canada*
[‡] *Department of Rehabilitation Medicine, College of Medicine, Yeungnam University, Daegu, Republic of Korea*
[‖] *Centre Médico-chirurgical de Touraine, Saint Cyr sur Loire, France*
[§] *Aspetar Orthopaedic and Sports Medicine Hospital, Doha, Qatar*
[¶] *Department of Clinical Science and Education, Sodersjukhuset, Karolinska Institutet, Stockholm, Sweden*

ARTICLE INFO

Article history:
Received for publication July 1, 2023
Accepted in revised form July 15, 2023
Available online August 25, 2023

Key words:
Entrapment
Guyon canal syndrome
Lacertus syndrome
Peripheral nerve
Tremor

Little is known about tremors caused by peripheral nerve entrapment. We report two cases of tremors caused by peripheral nerve compressions. Two patients presented with intentional tremors combined with peripheral nerve compression symptoms on their affected hand. Based on the clinical findings and evaluations, the first patient was diagnosed with double-crush compression of the ulnar nerve at the cubital tunnel and Guyon canal, and the second patient was diagnosed with lacertus syndrome. The first patient underwent surgical release of the cubital tunnel and Guyon canal in two stages. The second patient underwent release of the lacertus fibrosus. At the 1-month follow-up after surgery, the tremors had completely resolved, and neurological symptoms improved. Peripheral nerve entrapment should be considered a potential cause of tremors in patients with tremors combined with symptoms of peripheral neuropathy. Surgical release can be curative.

Peripheral nerve entrapment (PNE) in the upper extremities, such as carpal tunnel syndrome, cubital tunnel syndrome, or lacertus syndrome, is common and can cause a range of symptoms, including neuropathic pain, tingling, numbness, clumsiness, and weakness.[1,2] Tremors are rarely reported as caused by PNEs. In 1986, Little et al[3] reported a case of tremor caused by ulnar nerve compression in the cubital tunnel. Because tremor is more commonly associated with central nervous system disorders, clinicians do not usually consider PNE as a cause of tremor, delaying accurate diagnosis and appropriate treatment.[4]

In this study, we report two causes of tremors—a double-crush compression of the ulnar nerve at the cubital tunnel and Guyon canal syndrome in the first and lacertus syndrome in the second. Written informed consent was obtained from these two patients for the publication of this case report and accompanying images.

Case Reports

Case 1

A 19-year-old right-handed woman with no specific medical history presented to our hand clinic complaining of intentional tremor associated with weakness, loss of dexterity, and claw deformity in her left hand. She also experienced shooting and electric shock-like pain in the left volar forearm, fifth digit, and ulnar aspect of the fourth digit. Her symptoms had developed 7 months previously and had worsened during that time, now interfering with her daily activities, sports, and school. She denied any history of trauma but recalled the onset of symptoms after playing golf in the summer. Three electrodiagnostic studies of the left upper limb were negative. Magnetic resonance imaging scans of the brain and neck were performed. No abnormal findings were observed. The patient was examined by seven different neurologists.

On physical examination, the tremor was rhythmic with a frequency of approximately 5 Hz. It manifested during purposeful motor movements but was not observed at rest. A claw deformity was present. Paresthesias were reported in the left little finger and

Declaration of interests: No benefits in any form have been received or will be received related directly to this article.

Corresponding author: Bianief Tchiloemba, MD, Department of Plastic Surgery, University of Calgary, 2204 7 Ave NW, Calgary, AB T2N 0Z6, Canada.
E-mail address: bianief.tchiloemba@ucalgary.ca (B. Tchiloemba).

https://doi.org/10.1016/j.jhsg.2023.07.008

the ulnar aspect of the fourth digit. Reduced grip strength in the left hand was observed, with initial grip strength of 29 pounds versus 59 pounds on the contralateral side. Specifically, weakness was noted in the flexor digitorum profundus to the little finger, the flexor carpi ulnaris, and the intrinsics.

Froment sign was positive. The Tinel sign was positive over the cubital tunnel and Guyon canal in her left upper limb. Atrophy of the interossei and hypothenar left intrinsic muscles was observed on inspection. The scratch collapse test was positive over the left cubital tunnel and Guyon canal. The cold spray test was used for confirmation. We decided to proceed with a cubital tunnel release under Wide Awake Local Anesthesia No Tourniquet (WALANT).[5] At follow-up 1 month after the cubital tunnel release, her pain in the volar aspect of the left forearm had disappeared, but the tremor, sensory, and motor symptoms persisted. A few weeks later, we also performed a Guyon canal release under WALANT. At follow-up 1 month after Guyon canal release, her tremor had completely resolved and the claw deformity was corrected (see Video 1, available online on the Journal's website at https://www.jhsgo.org). In addition, the neuropathic pain in the left finger had disappeared. Her grip strength increased considerably after her second surgery, with her left grip increasing from 29 to 41 pounds, and her 3 finger pinch going from 21 before surgery to 30 after surgery.

Case 2

A right-handed 36-year-old man presented for the treatment of an intentional tremor in his right hand combined with the weakness of the right-hand grip, which persisted for 3 years. He also experienced pain and numbness in the elbow and forearm associated with clumsiness and decreased endurance. He had undergone a radial nerve release to treat right lateral elbow pain previously. He had no specific medical or trauma history. An electrodiagnostic study and magnetic resonance imaging of the right upper limb revealed no specific abnormalities.

On physical examination, the tremors were intentional and rhythmic. Weakness was observed in the right flexor carpi radialis, flexor pollicis longus, and flexor digitorum profundus of the index finger. Tenderness exists on palpation of the median nerve under the lacertus fibrosus. The scratch collapse test was positive at the level of the lacertus fibrosus.

The patient was diagnosed with lacertus syndrome.[6] The patient underwent lacertus release on the right side using WALANT. At follow-up, 1 month after surgery, her tremor and neuropathic pain had completely disappeared and her motor weakness had greatly improved.

Discussion

Our two patients had peripheral focal entrapment within the cubital tunnel and Guyon canal and under the lacertus fibrosus, respectively. The tremors in both patients completely disappeared after surgical release, and we believe that these were induced by PNE (Guyon canal syndrome and lacertus syndrome).

The neurophysiological mechanism underlying the occurrence of tremors because of PNE is unknown. Tremors caused by peripheral neuropathy may result from a hyperactivated stretch reflex induced by minimal weakness or impairment of the stretch reflex.[7] In animal studies, after partial injury to the peripheral nerve, which can reduce sensory input, there can be an increase in gains within the reflex pathway, magnifies the likelihood of tremor oscillations.[8,9] A progressive PNE involves some large-diameter sensory axons and reduces sensory input to stretch reflex pathways while preserving some reflex connections.[3,8,9] Decreased sensory input then causes increased synaptic excitability of the still-functioning afferent neural fibers. Repetitive discharge by mechanical compression of the ulnar and median nerves within the cubital tunnel, Guyon canal, and underneath the lacertus fibrosus appeared to induce intentional tremors in our patients.

Guyon canal syndrome is a relatively rare peripheral neuropathy that involves compression of the distal portion of the ulnar nerve as it passes through a narrow anatomical canal in the wrist. The most common causes of ulnar nerve entrapment in Guyon canal are ganglion and repeated trauma. In some cases, it is idiopathic. Cubital tunnel syndrome is the second most common PNE in the upper extremity after carpal tunnel syndrome. The prevalence of lacertus syndrome has not been documented, but it has been reported that it occurs frequently, alone or in association with carpal tunnel syndrome.[10] Lacertus syndrome is often misdiagnosed as carpal tunnel syndrome because the symptoms are similar. Clinicians should consider the possibility of lacertus syndrome in patients with neuropathic pain and/or neurological symptoms of the hand or persistent median nerve paresthesia after treatment of carpal tunnel syndrome.

Several cases of tremors caused by peripheral nerve disorders have been reported. However, most of the cases were peripheral polyneuropathies, such as diabetic, uremic, alcoholic, immune-mediated polyneuropathy, vasculitis neuropathy, and motor neuron disease.[7] Regarding focal PNE, to the best of our knowledge, only one case of tremor occurring after cubital tunnel syndrome was reported in 1986.[3]

In conclusion, we have reported that tremors can occur because of focal PNE. As many clinicians are unaware of tremors after PNE, its diagnosis and appropriate management may often be missed or delayed. Clinicians should consider the possibility of peripheral nerve disorder-related tremors when patients with peripheral entrapment syndrome(s) present with intentional tremors. Nerve decompression under local anesthesia can be curative.

References

1. Kwak S, Jeong D, Choo YJ, Chang MC. Management of neuropathic pain induced by cubital tunnel syndrome using pulsed radiofrequency: two case reports. *Medicine (Baltimore)*. 2019;98(21):e15599.
2. Lee SH, Choi HH, Chang MC. Effect of ultrasound-guided partial release of the transverse carpal ligament with a needle in patients with refractory carpal tunnel syndrome. *Pain Physician*. 2022;25(1):E141–E145.
3. Little JW, Burchiel K, Nutter P. Tremor and peripheral nerve entrapment. Case report. *J Neurosurg*. 1986;64(1):145–147.
4. Lenka A, Jankovic J. Tremor syndromes: an updated review. *Front Neurol*. 2021;12:684835.
5. Hagert E. Clinical diagnosis and wide-awake surgical treatment of proximal median nerve entrapment at the elbow: a prospective study. *Hand (N Y)*. 2013;8:41–46.
6. Hagert E, Jedeskog U, Hagert CG, Fermin TM. Lacertus syndrome: a ten year analysis of two hundred and seventy five minimally invasive surgical decompressions of median nerve entrapment at the elbow. *Int Orthop*. 2023;47(4):1005–1011.
7. Said G, Bathien N, Cesaro P. Peripheral neuropathies and tremor. *Neurology*. 1982;32(5):480–485.
8. Eccles RM, Kozak W, Westerman RA. Enhancement of spinal monosynaptic reflex responses after denervation of synergic hind-limb muscles. *Exp Neurol*. 1962;6:451–464.
9. Gallego R, Kuno M, Núñez R, Snider WD. Disuse enhances synaptic efficacy in spinal motoneurons. *J Physiol*. 1979;291:191–205.
10. Tang JB. Median nerve compression: lacertus syndrome versus superficialis-pronator syndrome. *J Hand Surg Eur Vol*. 2021;46:1017–1022.