

Case study

Carpal tunnel syndrome caused by anatomic anomalies muscles; a three cases report

Abstract

Carpal tunnel syndrome (CTS) is the most frequent peripheral compression neuropathy. Anatomic variations may be encountered during carpal tunnel surgery.1–3 Compression of the median nerve at the wrist is frequently encountered. Carpal tunnel syndrome usually occurs without any obvious extrinsic causes; several cases have however been reported caused by anomalous or hypertrophic muscles. A survey of the literature shows that compression neuropathy of the median nerve has been reported in relation with anomalies affecting three muscles: the first (or second) lumbrical, the palmaris longus and its anatomic variants and the superficial flexor of long fingers. We can suspect the presence of such an anomalous muscle when the compression syndrome concerns a patient who is not within the “usual” age group with symptoms initiated or aggravated by physical exercise.

This report presents three cases of carpal tunnel syndrome caused by anatomic anomalous muscles diagnosed peroperatively. (Epidemiologic, pathogenesis and diagnosis are discussed and literature was reviewed.)~~delet~~

Formatted: Highlight

Keywords

median nerve, nerve compression, carpal tunnel syndrome ,anomaly, muscle, hand surgery.

Introduction

Carpal tunnel syndrome (CTS) is a frequently encountered condition in middle-aged women; it is in most cases idiopathic. In all other “atypical” carpal tunnel syndromes, a more extensive search for external causes of the compression is necessary. An extrinsic structure is usually responsible for the compression and a systematic search for compressing structures is required (1). Compression of the median nerve by muscle anomaly is exceptional and performs a specific symptomatology: affects a young adult, manual worker, aggravated by manual activity. Clinical examination searches paresthesia during bending of MP. The ultrasound and MRI are used to make the diagnosis. The lifting of the median nerve compression by opening the carpal tunnel can relieve the patient (2). We present in this work 3 cases of secondary carpal tunnel

39 syndrome caused by muscle abnormality. The aim of this work is to study the different
40 arepidemiological, etiopathogenic, clinical and treatment of this rare disease. A
41 literature review was made. (delete and replace with present 3 rare cases of carpal
42 tunnel syndrome causes)

Formatted: Highlight

Formatted: Highlight

43 **Methods**

44 Our study is retrospective, collected in orthopaedic surgery department in Nabeul
45 hospital. Over the last ten years; we have collected three cases of carpal tunnel
46 syndrome caused by a muscle abnormality in 200 patients operated. Epidemiological
47 data, clinical features, investigations, treatment and follow up of the patients with this
48 type of injury were analysed.

Formatted: Highlight

49 **Results** delete

50 **Add cases :**

51

- 52 • Case 1

53 A 25-year-old female who suffered from paraesthesia, and numbness in the median
54 nerve territory of the left hand for several years. Her symptoms were aggravated by
55 exercise. She had a positive Phalen test and Tinel's sign at the carpal tunnel without
56 thenar atrophy. The clinical diagnosis of CTS was confirmed by electrophysiological
57 examination. Under loco regional anaesthesia, carpal tunnel release was performed
58 through the classic incision. After dividing the transverse carpal ligament, an aberrant
59 tendon was discovered on the anterior surface of the median nerve within its investing
60 tissue (Figure 1). The tendon was inserted deeply into the palmar aponeurosis and
61 evoke abnormal palmaris longus tendon: palmaris profundus. The median nerve was
62 congested. The palmaris longus tendon was palpable above the wrist crease. Further
63 exploration above the wrist was not indicated clinically and the origin of the palmaris
64 profundus was not established. After a few weeks the patient's symptoms resolved
65 completely.



67
68
69 Figure 1: Peroperating view showing palmaris profundus tendon with compression of
70 median nerve.

71 **Highlight in the figure with arrows**

72

73 • Case 2

74 A 35-year-old left-handed woman, without significant pathological history suffering from
75 numbness, tingling, pain, and weakness in the left hand and affecting the thumb, the
76 index and the middle finger, developed 6 months ago without any traumatic or
77 microtraumatic antecedent. Interrogation of the patient found a progressive evolution
78 over several months with gradual acroparaesthesia in the median nerve territory
79 associated with palmar pain. Her symptoms were aggravated by exercise. Physical
80 examination discovered a decrease in epicritic sensitivity with the Weber test, without
81 evidence for motricity impairment, using preserved prehensile strength and digital
82 winding as readouts. No abnormalities were seen on laboratory studies.

83 Functional exploration with electromyography identified a sensitive injury downstream of
84 the carpal tunnel. Standard X-rays did not find abnormalities. Surgery was performed
85 under loco-regional anaesthesia with an axillary block. Carpal tunnel release was
86 performed through the classic incision. Abnormal lumbrical tendon was easily exposed
87 after skin incision and opening of the mid-palmer fascia and the flexor retinaculum. The
88 median nerve was flattened (Figure 2). Postoperatively, the patient went home with scar
89 care every other day and a prophylactic treatment for algoneurodystrophy with vitamin C
90 for one month. No immobilization was used. Complete healing occurred after 21 days.
91 The last clinical review at the two-month postoperative, showed entire disappearance of
92 the acroparaesthesia and total functional recovery including full mobility of the fingers,
93 absence of pain, and restoration of the grip strength. No specific rehabilitation was
94 necessary.



95
96
97 Figure 2: Peroperating view showing hypertrophy of lumbrical tendon and compression
98 of median nerve.

99 [Highlight in the figure with arrows](#)

100 • Case 3:

101
102 A 65-year-old female who has suffered from paraesthesia, and numbness in the median
103 nerve territory of the right hand for five years. Her symptoms were aggravated by
104 exercise. Tinel's sign and Phalen test were positive. There was no evidence of thenar
105 atrophy. The clinical diagnosis of CTS was confirmed by electrophysiological
106 examination. Under loco regional anaesthesia, carpal tunnel release was performed
107 through the classic incision. After dividing the transverse carpal ligament, we identified a

108 muscle in the carpal tunnel. Pull on the muscle led to proximal interphalangeal joint
109 flexion of middle finger (Figure 3). The median nerve was congested. After a few weeks
110 the patient's symptoms resolved completely.



111
112 | [Highlight in the figure with arrows](#)

113
114 Figure 3: Peroperative view showing hypertrophic and engaged the flexor digitorum
115 superficialis of the middle finger into the carpal canal.

116

117 Discussion

118 Carpal tunnel syndrome caused by a space occupying lesion is rare and more
119 complicated than idiopathic carpal tunnel syndrome. Compression of the median nerve
120 with anatomic anomalies muscles is very rare and produces an atypical clinical
121 presentation. Facing anatomic anomalies during carpal tunnel surgery is not uncommon.
122 In a study of 382 patients, Tountas et al. reported that 38 hands had an anomalous
123 anatomy. Still and Kleinert [3] reported a series of 9 cases of which 8 presented with a
124 neuro-compression: a carpal tunnel syndrome in 6 cases, the anomalies involved the

125 palmaris longus in 4 cases, the flexor digitorum superficialis in two and abnormal
126 lumbricals in another two cases.

127 The pathogenesis of carpal tunnel syndrome is varied. In most cases no specific cause
128 can be disclosed; these cases are usually termed “idiopathic” or “primary”. Secondary
129 carpal tunnel syndrome can be caused by various conditions .Abnormal or aberrant
130 muscles have been well described, particularly in manual workers. These anomalous
131 muscles may be hypertrophic or abnormal lumbricals, a hypertrophic flexor digitorum
132 superficialis or an abnormal palmaris longus (profundus or reversed). All these muscles
133 compromise the available space in the carpal canal, resulting in compression of the
134 median nerve [4].

- Lumbrical muscle

136 The first or second lumbrical muscle is usually held responsible for the compression.
137 The relationship of the lumbrical muscles and the carpal tunnel was investigated by
138 Cobb et al. [5]. They found on cadavers that incursion of the lumbricals into the carpal
139 canal during flexion of the fingers is a normal occurrence. It may be a possible cause of
140 occupation-related carpal tunnel syndrome [6, 7].

- M.flexor digitorum superficialis

142 Anomalies of the muscle belly of the flexor digitorum superficialis of long fingers as a
143 cause for compression of the median nerve were reported and well [8, 9]. In two of them
144 the muscle belly was hypertrophic and engaged into the carpal canal, compressing the
145 median nerve [10]; in the others an abnormal muscle belly originated in the palm and
146 inserted onto the superficial flexor tendon. Resection of this muscle belly seems to be
147 necessary in such cases.

- Abnormal palmaris longus tendon

149 The palmaris longus muscle is without any doubt the most variable muscle in the
150 forearm. It can be absent (15% of all patients, more on the left side and in women),
151 hypertrophic, reversed (muscle belly distal rather than proximal), centrally placed,
152 digastric, duplicated or bifid. It can be located deep to the transverse ligament (palmaris
153 profundus) [11]. Anomalies in origin and insertion have been described and particular
154 accessory insertion slips are well recognized (palmaris accessorius). Lindly and Kleinert
155 [3] found (5.7%) anomalies in 526 CTS surgeries and there was one palmaris
156 profundus anomaly in their experience. Palmaris profundus was first described in 1908.
157 Reimann et al. [13] found one example in 530 cadaver arms.3. It arises as a separate
158 muscle in the middle third of the forearm, deep to the superficialis muscles. Its tendon
159 courses deep into the carpal tunnel as a tenth tendon that may stray from a dorsal to
160 palmar position within the canal, inserting into the palmar aponeurosis. The palmaris
161 profundus is not a variation of the palmaris longus. Several reports indicate that the
162 palmaris longus coexists with palmaris profundus [4, 14].

163
164 Ultra-sonogram, CT and MRI should be performed. Recently, arthroscope or minimal
165 invasive surgery has been preferred, however, in cases such as this involving space
166 occupying lesions, symptoms do not improve unless open transverse carpal ligament
167 release is performed in conjunction with removal of the SOL. Based on this study, it is
168 important to perform special tests such as CT or MRI to identify the SOL through
169 physical examination when unilateral CTS patients exhibit swelling or tenderness of
170 volar wrist crease area. The use of MRI scans is an excellent way to examine soft tissue,

171 and also has the advantage to make it possible diagnose exact location and border of
172 lesion and also involvement of surrounding tissue. However, CT scans are superior to
173 MRI with regard to detection of bony lesion. Ultra-sonogram has its advantages such as
174 wide availability, lower cost, and shorter examination time; however, accurate
175 characteristics of SOL cannot be identified [15].

176 However, imaging is not routinely done for carpal tunnel syndrome, and surgeon's first
177 encounter likely will be intraoperatively. The literature is not conclusive as to whether the
178 anomalous muscle can cause clinical nerve compression. In most cases, the authors
179 have advocated resection of the anomalous muscle [17]. There are reports where the
180 patients are managed with just release of the carpal ligament, without resection of the
181 muscle [18], or even with physical therapy alone. However, it must also be noted that,
182 the resection of these anomalous muscles did not lead to any residual deficits. Some
183 authors suggest excision of the muscle when there is a suspicion of it causing the
184 compression, only after identifying the existence of normal structures to preserve
185 function (19). In our cases, conventional open transvers carpal ligament without resection
186 of these anomalous muscle gave immediate painless.

187

188 **Conclusion**

189 All space-occupying lesions can compromise the free course of a nerve in an
190 anatomically limited tunnel, so do anomalous, aberrant or hypertrophied muscles. In
191 patients not belonging to the so-called typical population for carpal tunnel syndrome,
192 younger patients with a neurocompression syndrome related to physical activity, the
193 treating physician should be aware of the possibility of such pathology. A thorough
194 knowledge of every possible anatomic variation is indispensable to surgeons who
195 perform CTS surgery, as an appreciation of the possibilities increases the safety of the
196 procedure [16]. An anatomic anomaly may be the aetiology of the CTS and failure to
197 address it results in the persistence to the symptoms and ends in failure to the
198 treatment.

199 **Consent Disclaimer:**

200 As per international standard or university standard, patient's consent has been
201 collected and preserved by the authors.

202

203 **Competing interests**

204 The authors declare that they have no competing interests

205

206

207 **References**

- 208 1. Chen CH, Wu T, Sun JS, Lin WH, Chen CY. Unusual causes of carpal tunnel
209 syndrome: space occupying lesions. *J Hand Surg Eur Vol.* 2012 Jan;37(1):14-9.
- 210 2. Sbai MA, Benzarti S, Msek H, Boussen M, Khorbi A. Carpal tunnel syndrome caused
211 by lipoma: a case report. *The Pan African Medical Journal.* 2015; 22: 51.
- 212 3. Lindley SG, Kleinert JM. Prevalence of anatomic variations encountered in elective
213 carpal tunnel release. *J Hand Surg Am.* 2003 Sep;28(5):849-55.
- 214 4. De Smet L. Median and ulnar nerve compression at the wrist caused by anomalous
215 muscles. *Acta Orthop Belg.* 2002 Dec;68(5):431-8.
- 216 5. Cobb TK, An KN, Cooney WP, Berger RA. Lumbrical muscle incursion into the carpal
217 tunnel during finger flexion. *J Hand Surg Br.* 1994 Aug;19(4):434-8
- 218 6. Carroll M., Montero C., Rare anomalous muscle cause of carpal tunnel syndrome.
219 *Orthop. Rev.*, 1980; 9: 83-85.
- 220 7. Butler B Jr, Bigley EC Jr. Aberrant index (first) lumbrical tendinous origin associated
221 with carpal-tunnel syndrome. A case report. *J Bone Joint Surg Am.* 1971 Jan;53(1):1602.
- 222 8. Hutton P, Kernohan J, Birch R. An anomalous flexor digitorum superficialis indicis
223 muscle presenting as carpal tunnel syndrome. *Hand.* 1981 Feb;13(1):85-6.
- 224 9. Neviasser RJ. Flexor digitorum superficialis indicis and carpal tunnel syndrome. *Hand.*
225 1974 Jun;6(2):155-6.
- 226
- 227 10. Smith RJ. Anomalous muscle belly of the flexor digitorum superficialis causing
228 carpal-tunnel syndrome. Report of a case. *J Bone Joint Surg Am.* 1971 Sep;53(6):1215-
229 6.
- 230 11. Brones, M. F., & Wilgus, E. F. S. (1978). Anatomical variations of the palmaris
231 longus, causing carpal tunnel syndrome: Case reports. *Plastic and Reconstructive*
232 *Surgery*, 62(5), 798-800.
- 233 12. Tountas CP, MacDonald CJ, Bihrl DM. Carpal tunnel syndrome. A review of 507
234 patients. *Minn Med.* 1983; 66: 479 – 482.
- 235 13. Reimann AF, Daseler ED, Anson BJ, Beaton LE. The palmaris longus muscle and
236 tendon. A study of 1600 extremities. *Anat Rec.* 1944; 89: 495 – 505.
- 237 14. Dyreby JR, Engber WD. Palmaris profundus rare anomalous muscle. *J Hand Surg*
238 *[Am].* 1982; 7: 513 – 514.
- 239 15. Kang HJ, Jung SH, Yoon HK, Hahn SB, Kim SJ. Carpal tunnel syndrome caused by
240 space occupying lesions. *Yonsei Med J.* 2009;50(2):257–261.
- 241 16. Suk JI, Walker FO, Cartwright MS. Ultrasonography of peripheral nerves. *Curr*
242 *Neurol Neurosci Rep.* 2013 Feb;13(2):328.
- 243 17. Bakriga B, Amouzou KS, Ayouba G, Kombate, N, Dellahn Y, Walla A, et al.
244 Anomalous muscles: a rare cause of carpal tunnel syndrome: a case report.
245 *International Journal of Orthopaedics.* 2017;4(1):705-07.
- 246 18. Javed S, Woodruff M. Carpal tunnel syndrome secondary to an accessory flexor
247 digitorum superficialis muscle belly: case report and review of the literature. *Hand (NY).*
248 2014;9(4):554-55.
- 249 19. Laxminarayan Bhandari, Michelle Palazzo, .ANOMALOUS MUSCLES
250 ENCOUNTERED IN THE CARPAL TUNNEL: A REPORT OF TWO CASES. *Journal of*
251 *Clinical and Diagnostic Research* 2017 Oct 10 PD03 - PD04.