Cubital Tunnel Syndrome caused by an Ulnar Nerve Tuberculoma: A rare case report and Literature Review

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Abstract

Background: Tuberculosis has a very weak affinity of peripheral nerves, and the presence of a tuberculoma within a peripheral nerve is only detected sporadically.

Case Report: A 64-year-old male patient presented to Qena General Hospital Orthopaedic Department in February 2022 with symptoms and signs suggestive of cubital tunnel syndrome. During the operation, a fusiform swelling containing a caseous material was noticed within the ulnar nerve. The lesion was found to be an ulnar nerve tuberculoma. The patient showed partial neurologic recovery after surgery followed by an incomplete course of anti-tuberculous medication.

Conclusion: Although extremely rare, tuberculous affection of the ulnar nerve can occur. The previously reported cases involved the nerve either proximal or distal to the cubital tunnel, unlike our case which presented with a tuberculoma within the tunnel. Thus, in addition to the usual clinical and electrophysiological assessment, it is advisable to perform ultrasound evaluation in all cases of cubital tunnel syndrome; first to assess the nerve for subluxation, and second to exclude any intra- or extra-neural lesion that may be the cause of entrapment.

Keywords: Tuberculosis; Tuberculoma; Cubital tunnel syndrome; Ulnar nerve.

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Introduction

Tuberculosis is a dreaded cause of morbidity in the developing countries, with both pulmonary and extrapulmonary sequelae. Among the rare sequelae of tuberculosis that may be encountered in orthopaedic practice is its impact on the peripheral nervous system **(Omar et al., 2020)**. This can be induced either by tuberculous synovitis arising within a nerve tunnel or, much less frequently, by a tuberculoma within the nerve itself. Peripheral nerve tuberculomas are, in general, a very rare occurrence owing to the very low affinity of tuberculosis to peripheral nerves as opposed to its relatively higher affinity to the central nervous system **(Omar et al., 2020; Sinha, 1975)**.

We report the case of a patient who presented with symptoms mimicking cubital tunnel syndrome that was found to be secondary to an ulnar nerve tuberculoma. To our knowledge, tuberculomatous affection of the ulnar nerve within the cubital tunnel has not been previously reported.

Case report

A 64-year-old right handed male farmer presented in February 2022 with a history of numbness with gradual deterioration of sensation in his right ring and little fingers and the ulnar side of the palm over the preceding 6 months. This was associated with gradual weakening of the hand grip and medial elbow pain. The patient's medical history was unremarkable, apart from hypertension controlled on medication. The patient had no previous history of trauma or surgery at the affected upper extremity. Physical examination the right elbow showed no cubitus varus or valgus deformity. The ulnar nerve was tender to palpation within the cubital tunnel, and the slight bulge felt at the time of examination was not explained. Clawing of ulnar two digits of the right hand was noted, with atrophy of palmar and dorsal interossei. The tender ulnar nerve could be palpated and rolled within the cubital tunnel. The range of motion was full for both elbow flexion-extension and forearm pronation-supination, with no instability at both joints. There was weakness of abduction and adduction of the fingers. Compared to the left hand, grip and key pinch were weak at the right hand, with positive Froment's test. Tinel test was positive at the cubital tunnel but negative at Guyon's canal. Plain radiographs of the right elbow were unremarkable. Nerve conduction velocity study was consistent with a picture of axonal injury of the right ulnar nerve at the level of the elbow, which was mainly motor. Routine blood tests were normal, apart from elevated erythrocyte sedimentation rate (ESR) (1^{st} hour: 40; 2^{nd} hour 85) and elevated serum creatinine level (2.9 mg/dL).

Based on the data above, the diagnosis of cubital tunnel syndrome was made, with no apparent explanation for the elevated ESR and no previous medical history to explain the elevated creatinine level. As there was a neurological deficit, surgical treatment was decided and the patient's consent was obtained. Surgical exploration was done under general anesthesia. The ulnar nerve was explored through a curved 10-cm incision on the medial aspect of the elbow. A fusiform swelling that occupied about 4.5 cm of the length of the ulnar nerve, with a slight bulge, was found **(Fig. 1)**. A scanty amount of a cheesy white material appeared though this sweeling **(Fig. 2)**. After fully decompressing the nerve, the intraneural lesion was addressed. Tangential excision of the caseating granuloma was done, taking care to excise the lesion in a direction as parallel as possible to the nerve fibers and to spare most of the nerve tissue **(Fig. 3)**.

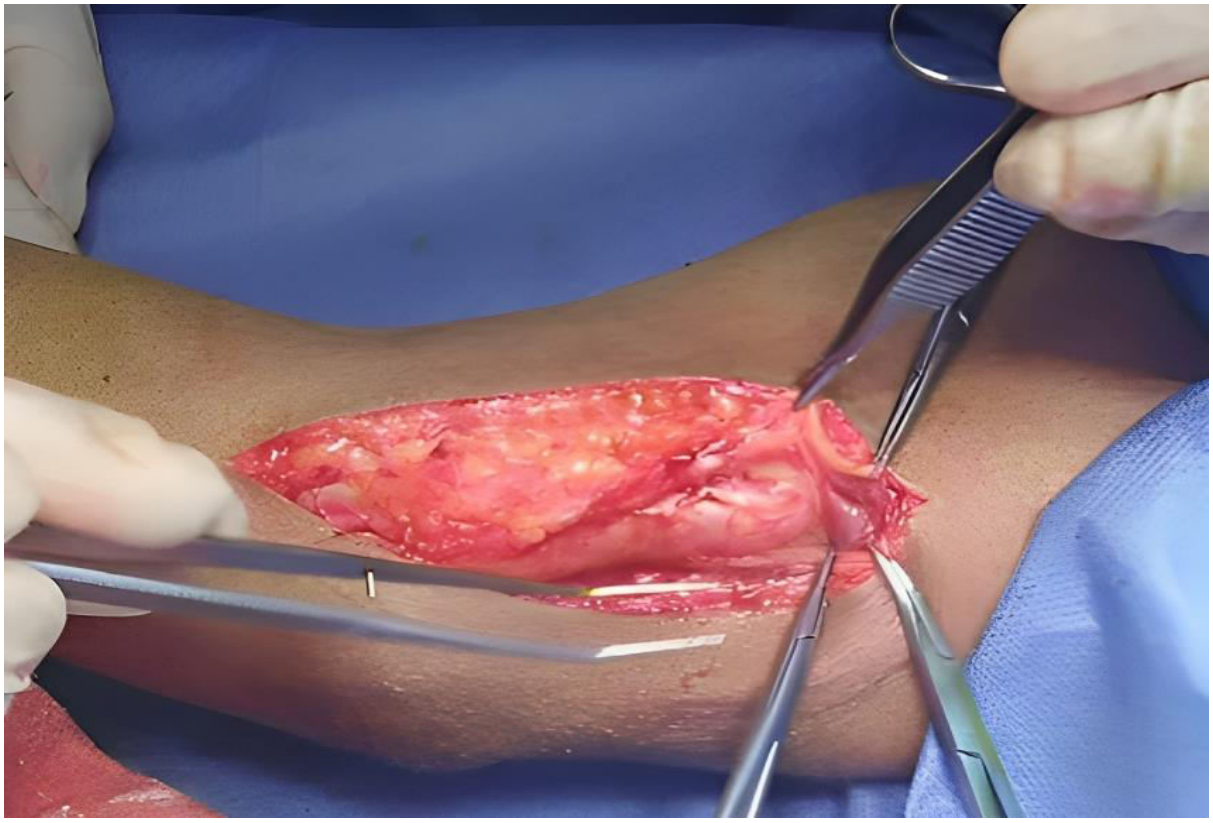

Fig. 1. The fusiform swelling found in the ulnar nerve

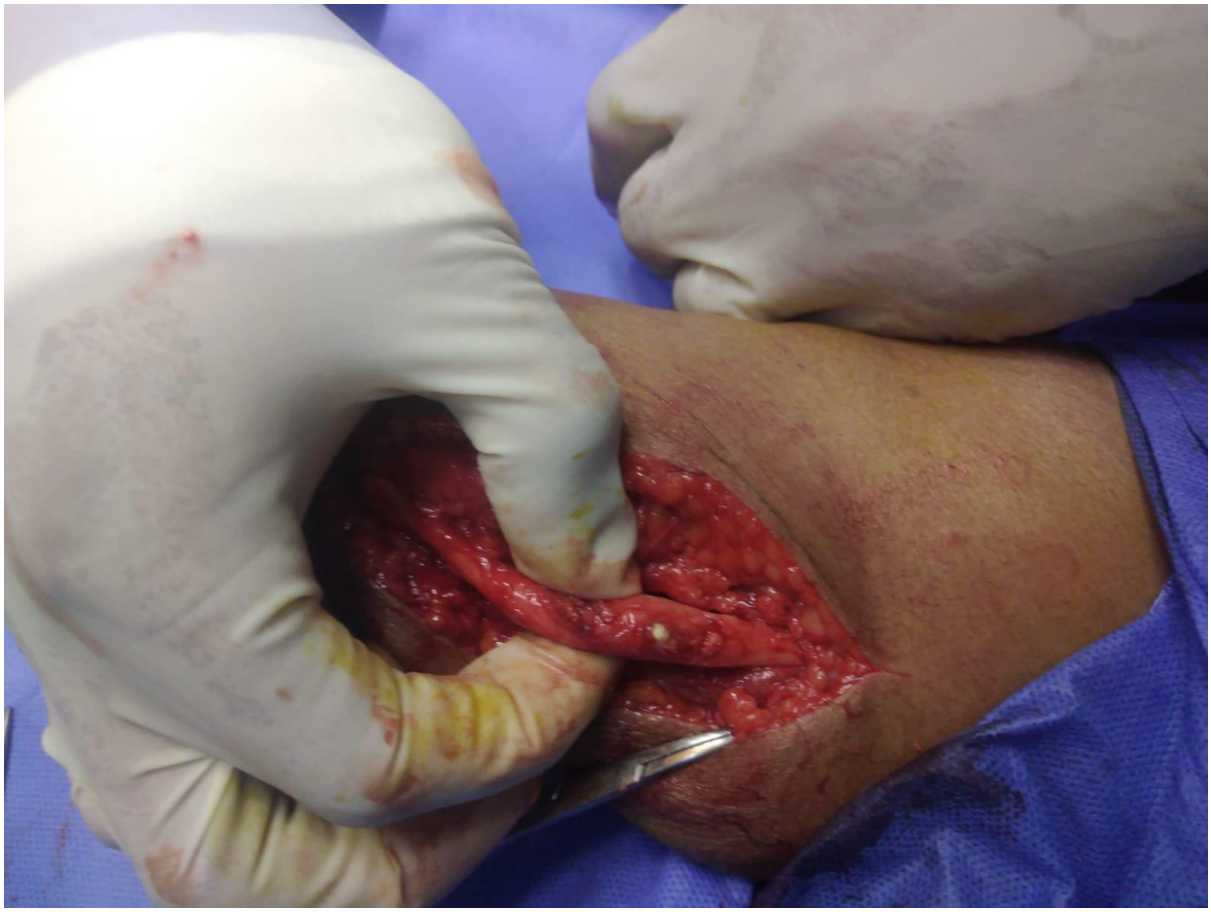

Fig. 2. Cheesy white material leaking from the nerve

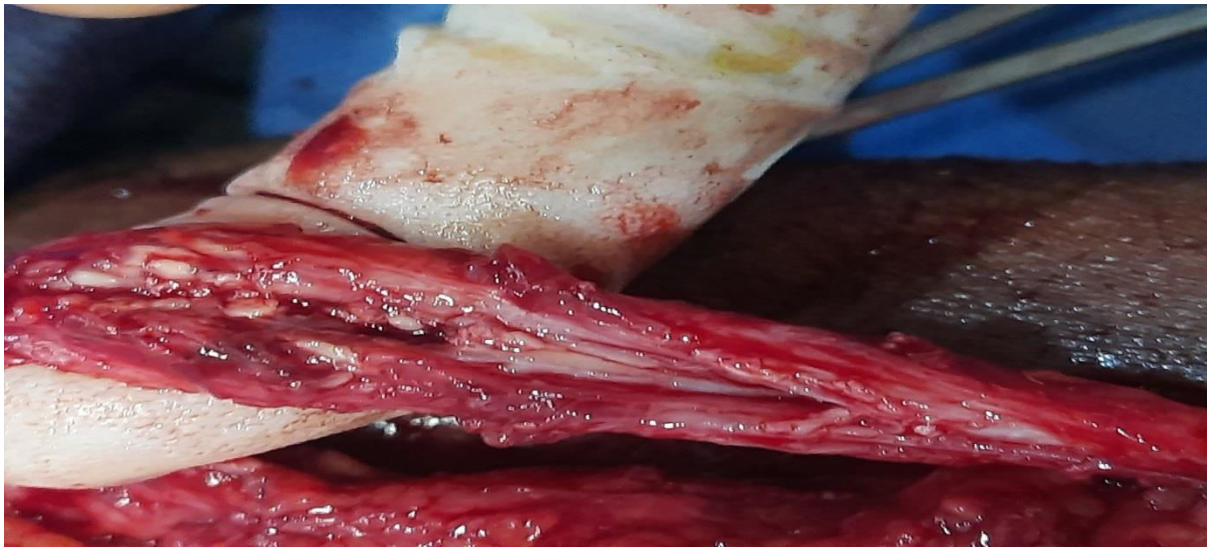

Fig. 3. The nerve after tangential excision of the lesion

Postoperatively, free elbow range of motion was allowed. And the obtained material was sent for microbiological and cytological studies, which proved it to be tuberculous caseous material **(Fig. 4)**.

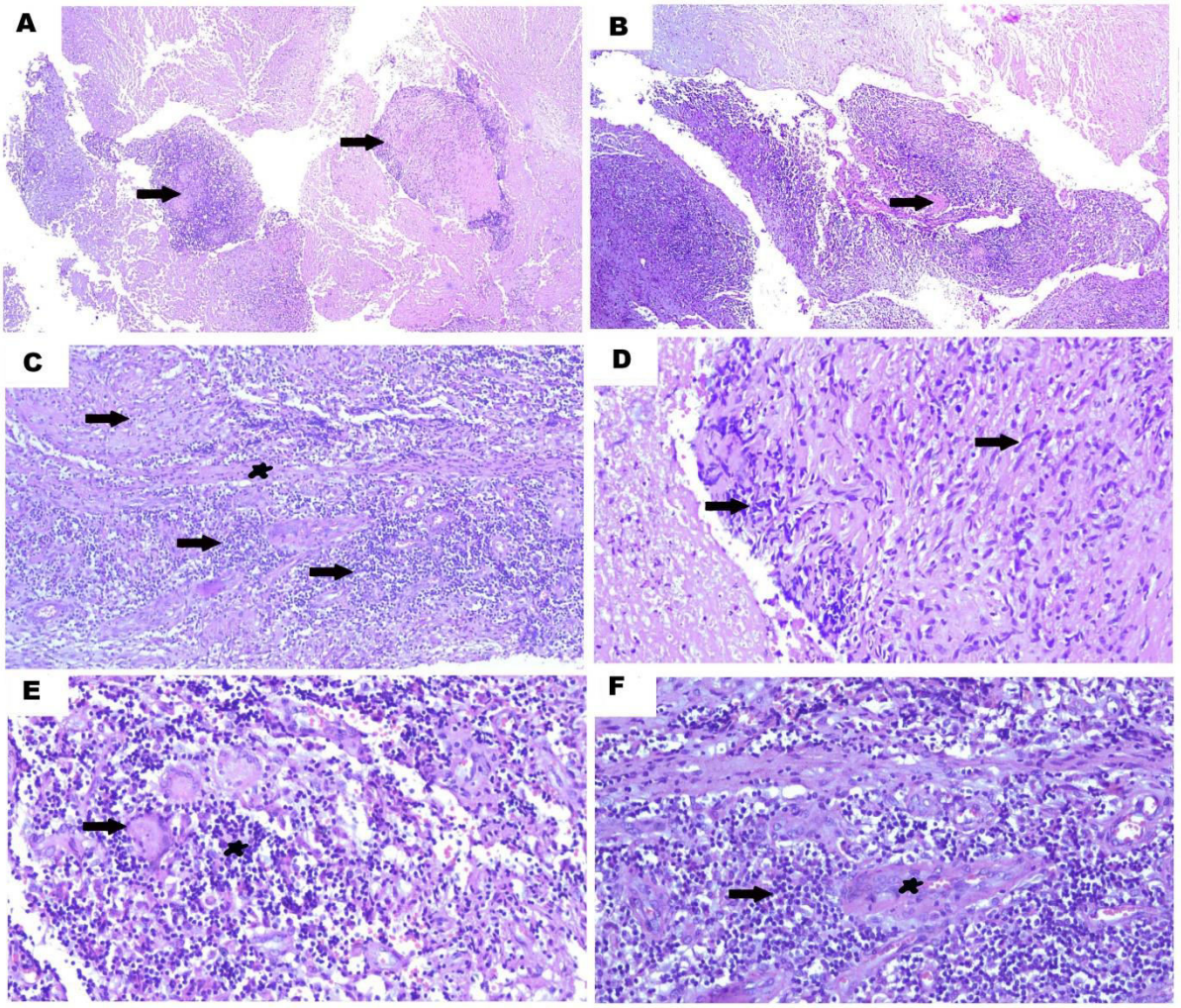

Fig. 4. Ulnar nerve tuberculoma sectioned with Hematoxylene and eosin stain (H&E). A, B) Epithelioid tuberculous granulomas. C) Segregated granulomas (arrows) separated by

complete fibrous capsule (asterisk). D) Tuberculous granuloma mainly composed of fibrous tissues infiltration. E) Tuberculous granuloma consisting of huge aggregation of epithelioid macrophages, multinucleated giant cells (arrow), and lymphocytes (asterisk). F) Tuberculous granuloma with thickened blood vessel wall (asterisk) and lymphocytes accumulation (arrow).

The patient was referred to the local chest hospital where the investigations confirmed that the condition was tuberculous in aetiology, but found no evidence of any other extra-pulmonary or pulmonary foci of tuberculosis. The standard anti-tuberculous medication was prescribed. And the patient was lost to follow up for twelve months thereafter.

After we managed to regain contact with the patient in March 2023, he was re-evaluated at our outpatient clinic. The patient reported that he suffered serious gastrointestinal trouble about 4 months into his anti-tuberculous medication, so he discontinued DOTS regimen and never re-instituted it again. On examination there was partial neurologic improvement, more sensory than motor, based on clinical evaluation by the same pre-operative examiner (**Fig. 5**). The mean DASH score improved from 63.3 to 50.8 over this 13-month period. This partial recovery did not meet the patient's expectations, thus he was reluctant to do any further investigations, including nerve conduction studies. We, therefore, relied exclusively on clinical evaluation for assessing the patient's outcome.

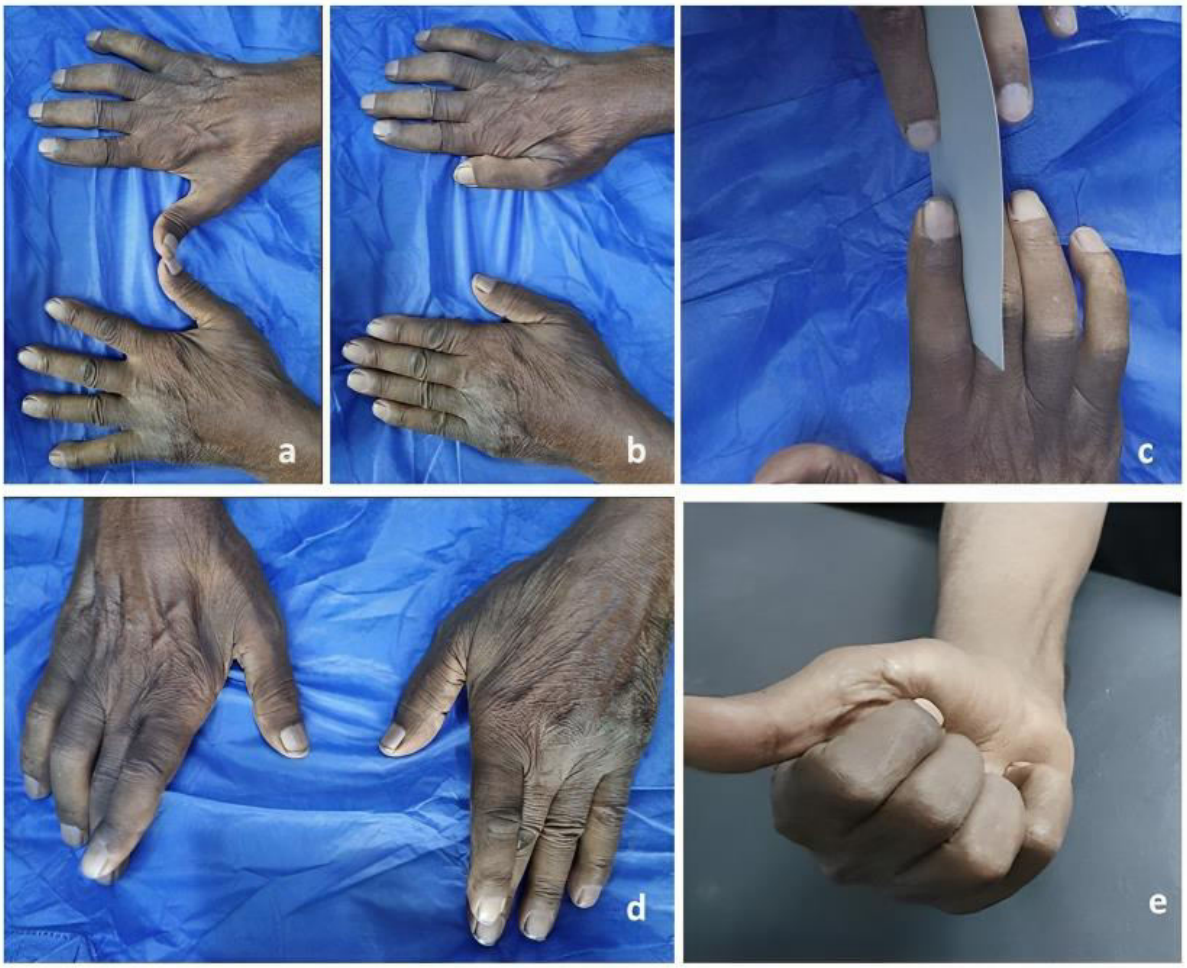

Fig. 5. Clinical assessment 12 months postoperatively, showing partial neurologic recovery. a) Testing finger abduction. Notice the wasting of the first dorsal interosseous muscle bulk on the affected right side. b) Testing finger adduction. c) Testing the palmar

interossei using the card test. d) Performing the crossed finger test. e) Testing the hand grip strength.

Discussion

Unlike leprosy, which has a well-known affinity to peripheral nerves, peripheral nerve affection is very rare in the case of tuberculosis and only sporadic reports have recorded peripheral nerve tuberculomas. Although central nervous system tuberculosis is in itself an uncommon form of human mycobacterial infection, peripheral nerve affection is yet far more uncommon.

The previously recorded cases of peripheral nerve tuberculomas involved the ulnar nerve **(Hasan and Prakash, 1964; Sinha, 1975; Nucci et al., 1988; Chandra et al., 2013; Chatterjee et al., 2015; Song et al., 2016; Djaharuddin et al., 2017; Omar et al., 2020; Maurya et al., 2022)**, the optic nerve and optic chiasma **(Schlernitzauer et al., 1971; Benchekroun et al., 2021)**, the lumbosacral plexus **(Stoeckli et al., 2000)**, the sural nerve **(Lee et al., 2019)**, and the recurrent laryngeal nerve **(Varghese et al., 2012)**. The pathogenesis of tuberculoma spreading to peripheral nerves is unclear **(Maurya et al., 2022)**.

Although tuberculous affection of the peripheral nervous system was known much earlier **(De Massary 1931)**, the earliest case of ulnar nerve tuberculoma was reported, as a "new clinical entity" in 1964 **(Hasan and Prakash, 1964)**. Since then, only 9 cases of ulnar nerve tuberculoma were reported in the English-language literature.

The presence of a tuberculoma within a nerve in a tunnel is an extremely rare occurrence. The association between tuberculosis and carpal tunnel syndrome was recorded fairly well in the literature. Since the one-century-old case series by Kanavel **(Kanavel, 1923)**, several reports of carpal tunnel syndrome secondary to tuberculous infection have been published. However, all of these were due to tuberculous *synovitis* rather than intraneural tuberculoma **(Lee, 1985; Nsegue et al., 2022; El-Rosasy et al., 2023)**. Atypical mycobacteria can also cause synovitis that may lead to carpal tunnel syndrome **(Yao et al., 2017)**.

Our patient's case was initially diagnosed as cubital tunnel syndrome. Ulnar nerve compression within the cubital tunnel syndrome is second in frequency only to median nerve compression within the carpal tunnel. It can be caused by extraneural compression as well as intraneural lesions. The reported causative intraneural lesions in patients with cubital tunnel syndrome included intraneural ganglion cysts **(Li et al., 2018)**, intraneural haematomas **(Xu et al., 2011)**, intraneural haemangiomas **(Kline and Moore, 1992)** and intraneural lipomas **(Balakrishnan et al., 2012)**.

Most of the previously reported cases (six out of nine) involved ulnar nerve at the distal part of the upper arm **(Sinha, 1975; Chatterjee et al., 2015**; **Song et al., 2016; Djaharuddin et al., 2017; Omar et al., 2020; Maurya et al., 2022)**. The case reported by Chandra et al in 2013 involved the medial aspect of right forearm, just distal to elbow joint **(Chandra et al., 2013)**, and the most distal location reported for an ulnar nerve tuberculoma was that by Nucci et al, being in the palm of the hand **(Nucci et al., 1988)**. To our knowledge, no case of ulnar nerve tuberculoma found within the cubital tunnel was previously reported.

Only one of the previously reported patients was treated conservatively with anti-tuberculous drugs followed by physiotherapy, with reported partial recovery **(Djaharuddin, 2017)**. In all the other cases, including ours, the systemic anti-tuberculous treatment was preceded by surgical excision of the tuberculoma. The ulnar nerve tuberculoma was associated with pulmonary affection in only two of the 9 previously reported

patients **(Song et al., 2016; Omar et al., 2020)**.

In our patient, we depended only on clinical and electrophysiological findings to establish the diagnosis of cubital tunnel syndrome. We did not perform a preoperative ultrasound examination, which we now do in every case of cubital tunnel syndrome. Dynamic assessment of the ulnar nerve for nerve subluxation, which is a somewhat frequent finding in cubital tunnel syndrome, and preoperative detection of pathologies within the tunnel or within the nerve itself are two important benefits of using ultrasonography in cubital tunnel syndrome.

Conclusion

Although very rare, tuberculomatous involvement of the ulnar nerve can occur. In this very rare case, it occurred within the cubital tunnel. Besides clinical and electrophysiological assessment, ultrasound evaluation is advisable in all cases of cubital tunnel syndrome; first to assess the nerve dynamically for subluxation, and second to exclude the presence of an intra- or extra-neural lesion that may have caused the entrapment.

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