

Cubital Tunnel Syndrome Caused By Uric Acid Tophi: An Unusual Case



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Abstract

We present a rare case of ulnar nerve compression by deposits of uric acid crystals at the cubital tunnel. Diagnosis of ulnar neuritis was largely based on the symptoms, patterns of paresthesia, and also specific patterns of muscle weakness. Nerve compression by tophi was documented during surgery. The tophi were completely resected and the nerve fully decompressed. Surgical treatment was followed by prompt recovery from ulnar neuropathy and also clinical symptoms. At twenty two months follow-up there was no evidence of recurrence. In the clinical setting of gout, tophi might be recognized as another cause of cubital tunnel syndrome. Given the high index of suspicion, early diagnosis and careful excision is associated with a satisfactory outcome

Keywords: Ulnar nerve compression; Tophaceous gout; Cubital tunnel syndrome

Introduction

Ulnar nerve compression at the elbow is quite common and second in incidence only to carpal tunnel syndrome [1,2]. Compression can occur at the epicondylar groove and also at the cubital tunnel which is indeed the most common site of entrapment. Compression at this site can be caused by a wide variety of anatomic and physiologic factors [3-5]. Consequently the differential diagnosis of ulnar neuropathies at the elbow region should include all possible anatomical sites of compression, systemic and metabolic disorders, such as diabetes mellitus or hypothyroidism, and also lesions that cause additional proximal or distal nerve compression, such as ganglia and soft-tissue tumors [2]. Entrapment neuropathy associated with tophaceous gout is however a rare occurrence. We present an unusual case of tophi compressing the ulnar nerve in the cubital tunnel which was successfully managed with surgical excision.

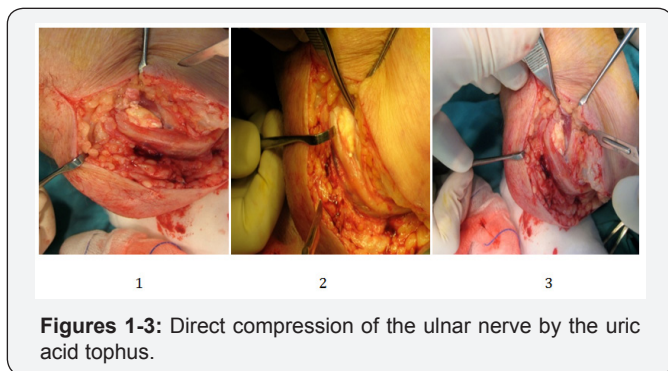
Case Report

The sixty seven-year-old man recently presented to our department with symptoms and signs of right cubital tunnel syndrome including persistent numbness and tingling in the ring and small finger of approximately sixteen months duration. He also complained of hand weakness and clumsiness as well as vague medial elbow pain. There was no history of trauma, previous surgical procedure in the same upper limb or recent illness; however, he mentioned a 20-year medical history significant for gout. Allopurinol was recommended but he has not been completely

compliant with his medication. In addition, he occasionally needed non-steroidal anti-inflammatory drugs to treat acute attacks. Regarding the course of the disease, the patient has experienced only some sporadic attacks of acute inflammatory arthritis of the metatarsal-phalangeal joint at the base of the big toe. He has had no other signs of gouty arthritis in any other joint or history of swelling of his right elbow. He was moderately obese and also suffered from hypertension.

On examination, atrophy of the lumbrical and interosseous muscles of the hand most evident in the dorsal thumb web space was revealed. Weakness of the intrinsic muscles was documented with a Wartenberg's sign, a Froment's sign, and also side-to-side confrontational testing [6]. The scratch collapse test that has recently been described for the assessment of cubital tunnel syndrome was also taken into consideration [7]. The test was found to be positive since a temporary loss of shoulder external rotation resistance when the examiner scratches the skin over the area of nerve compression. A positive Tinel test was evident along the course of the nerve as well as a positive elbow flexion test. Ulnar neuropathy was classified according to the McGowan grading system as III [8]. Static two-point discrimination was eight mm in the small finger. There were no signs of arthritis, redness or swelling of the involved elbow and the elbow range of motion was almost full and painless except for flexion which produced radiating pain in the medial side of the elbow down to the hand.

Laboratory data for this patient including the serum uric acid level at the time of presentation were within normal values but marked variability in uric acid measurements has been documented in his medical records. At operation a tophus 10x10x12 mm arose at the bed of the cubital tunnel compressing the nerve. Complete cubital tunnel release with resection of the uric acid tofi and anterior subcutaneous transposition of the nerve was performed. No other soft tissue of structure looked inflamed (Figures 1-3). Ulnar nerve compression by tophus was documented by examination of tophaceous material with compensated polarized light microscopy that identified the characteristic monosodium urate crystals. Operation was followed by prompt relief of pain and sensory disturbances. At the latest follow-up, 22 months postoperative. Sensory deficit was improved almost too normal but his muscle strength was 3/5 according to the Medical Research Council Scale for Muscle Strength.



Discussion

Gout affects approximately 1-2% of the population in the developed countries, being the most common inflammatory arthritis in men. Indeed, epidemiological data show that in the past twenty years its prevalence has doubled [9]. Nonetheless, and despite its early recognition and the availability of effective therapy, the proper diagnosis and therefore treatment are still problematic [10]. Yet, in order to prevent complications accurate diagnosis and appropriate therapy are mandatory. Gout is characterized by chronic hyperuricemia and manifests as deposits of monosodium urate crystals known as tophi in the joints, tendons, and surrounding soft tissues. It may affect the musculoskeletal system at any site. However, it classically presents with recurrent and intense pain in the first metatarsophalangeal joint [11]. Other joints frequently involved in the early stage of gout include the midfoot, ankle, heel, and knee. The wrist, fingers, and elbows are more typical sites of attack later in the course of the disease. Extra-articular manifestations include tophi and renal stones [12]. Untreated or severe gout leads to visible and palpable tophaceous deposits and a destructive arthropathy. However, with the variety of medications available today for the treatment of acute attacks, the incidence of tophaceous gout is progressively diminishing.

Likewise, ulnar nerve compression at the elbow is quite common and second in incidence only to carpal tunnel syndrome [1,2]. Potentially, entrapment can occur at the arcade of Struthers,

the medial intermuscular septum, the medial epicondyle, the cubital tunnel, and the deep flexor pronator aponeurosis. The most common site of entrapment is the cubital tunnel. Regional anatomical studies have revealed variability in the level of previous unidentified fibrous bands and suggest that proximal and distal ends of the cubital tunnel be explored [3,13]. Furthermore, a wide variety of lesions that cause nerve compression in the groove has been described in the literature. These lesions include tumors, ganglia, synovitis, osteochondromas, fracture fragments, arthritic spurs from the epicondyle or the olecranon [14-16].

Tophi are not a common cause of ulnar nerve compression and have not been highlighted in the literature. The present report identifies the case of a man with ulnar nerve compression caused by tophus. Cubital tunnel syndrome due to gouty tophi is a rare condition, and to our knowledge, only four such cases have previously been reported [17-19]. Nevertheless, in three of the aforementioned cases tophi were found at the epitrochlear groove and in only one case at the distal cubital tunnel arising from the joint capsule. In the present case report the tophus was identified at the bed of cubital tunnel at the level of the joint capsule. On the other hand, however, tophi have been recognized as a potential cause of compressive neuropathies and specifically for carpal tunnel syndrome. In these references, mostly case reports, carpal tunnel syndrome is associated with flexor tendinitis in the carpal tunnel. Other structures where deposits of tophi can be found include carpal tunnel floor, transverse carpal ligament, flexor tendons, tendons sheath or even the median nerve [20-22].

In addition, quite a few cases are associated with bilateral involvement [21-24] and some fewer cases with acute compression [25]. Furthermore, there are references for patients who suffered an acute attack of gout in the wrist after surgical decompression of carpal tunnel syndrome [26-28]. The acute inflammatory reaction was resolved only when treated postoperatively with a combination of anti-inflammatory and anti-gout medication. The differential diagnosis of compressive neuropathies in the arms includes syndromes involving the nerve roots and brachial plexus, as well as the peripheral nerves. Often these conditions coexist. In the present case physical examination began at the neck and proceeded distally. There was no evidence of cervical disease. Electromyography and nerve conduction studies were not performed in this case. It is well known that electro diagnostic testing is a tool used as an adjunct to careful clinical evaluation in order to clarify both the nature and the location of the nerve dysfunction. Taken alone without clinical correlation, it does not establish a diagnosis neither can predict the surgical outcomes. Even in patients with clinical evidence of cubital tunnel syndrome, electromyography and nerve conduction velocities may have a false-negative rate in excess of 10%. Therefore, electro diagnostic studies should be reserved for when the clinical picture is not typical or obscured by other co morbidities [2,29,30].

In the present case the clinical presentation of ulnar nerve compression was indisputable and the diagnosis straightforward and thus no electro diagnostic testing were performed. Also, no

radiographs of the elbow were taken in this case. Plain radiographs show typical changes only in advanced chronic gout when bony abnormalities have developed. In general bone abnormalities indicative of deposition of urate crystals are a late feature of gout typically found 15 years or more after the initial attack and are always present in patients with subcutaneous tophi. These abnormalities are most frequently asymmetric and confined to previously symptomatic joints [31].

In this case since there were no symptoms or clinical signs of elbow joint pathology and for that reason no radiographs were obtained. Furthermore, in a similar case of cubital tunnel syndrome caused by tophi [19], elbow radiographs did not reveal either degenerative changes or calcific deposition. Optimum treatment of ulnar nerve compression consists of early diagnosis and proper surgical treatment [2]. Various surgical techniques for decompression of the ulnar nerve have been described in the literature but a definitive gold standard does not exist. However, the chance for complete recovery is inversely related to the initial neuropathy grade but most patients exhibit an improvement of at least one McGowan grade. Therefore, treatment can be expected to be most successful in cases of mild neuropathy where it is estimated that more than 90% treated operatively achieve excellent results regardless of the procedure [32]. The advocated procedures include simple decompression, subcutaneous or sub muscular anterior transposition, medial epicondylectomy, and also partial medial epicondylectomy and yield reasonable long-term results even for patients with severe nerve lesions [33-36].

On the other hand there are some authors that do not recommend in situ decompression for severe cases of compressive neuropathy or favor anterior transposition of the nerve also in recurrent cases [2,37,38].

In the present case the simplest surgical option that would address the pathology was pursued. Furthermore, since there were no signs of the disease at the medial epicondyle no epicondylectomy was performed [1]. Twenty two months after the operation the patient remains symptom free. However, given that gout is a chronic disease a recurrence of tophi compression may be anticipated. Overall, cubital tunnel syndrome is common and can result in severe disability if diagnosis and treatment are delayed. A prompt diagnosis of nerve compression allows for earlier treatment that may result in a more favorable outcome. Complications are preventable with accurate diagnosis and appropriate therapy. In the differential diagnosis of cubital tunnel syndrome tophaceous compression over the ulnar nerve should be taken into consideration. A thorough history focused on determining associated risk factors is the keystone for the correct diagnosis. Once compression occurs the management should be surgical.

Conflict of Interest Statement

The authors of this article certify that they have NO affiliations with or involvement in any organization or entity with any

financial or non-financial interest in the subject matter or materials discussed in the manuscript.

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