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UP-TO DATE REVIEW AND CASE REPORT • ELBOW - HAND

Ulnar intraneural cysts as a cause of cubital tunnel syndrome: presentation of a case and review of the literature

Dimitrios V. Papadopoulos[1,2] · Ioannis Kostas-Agnantis[1] · Dimitrios Kosmas[1] · Evridiki Tsiomita[1] · Maria A. Korompilia[1] · Ioannis D. Gelalis[1] · Anastasios V. Korompilias[1]

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Abstract

Introduction Intraneural cysts usually involve the common peroneal nerve, and in many cases, they are causing symptoms due to neural compression. It is hypothesized that these cysts originate from the adjacent joints while articular pathology is a major contributing factor for the formation of these lesions. Although ulnar nerve is the second most commonly affected nerve, these lesions usually develop distally at the Guyon tunnel, so cubital tunnel syndrome due to epineural cysts is very rare. In such cases, elaborate preoperative work-up is mandatory and surgical treatment should follow certain well-defined principles.

Case description A 60-year-old female patient presented with complaints of pain along the medial side of her elbow, forearm and hand and a tingling sensation in the same distribution for the past 2 months. The patient had sustained an injury 15 years ago, and a distal humerus fracture was diagnosed at that time. Radiological signs of posttraumatic elbow arthritis were evident at the initial evaluation. The patient was diagnosed with cubital tunnel syndrome which was further confirmed by nerve conduction studies, and she underwent surgical decompression of the nerve. During surgery, intraneural cysts were identified and addressed by excision, while dissection of the articular branch of the nerve was also performed. Pain and numbness subsided shortly after surgery, while the patient remained free of symptoms until the last follow-up.

Keywords Intraneural cysts · Cubital tunnel syndrome · Diagnosis · Surgical management

Introduction

Neural cysts are relatively uncommon lesions usually developing around the peroneal nerve, with ulnar nerve being the second most common location [1]. Several other terms have been also used to describe these lesions including intraneural ganglion cysts, intraneural mucoid cysts or simple intraneural ganglions [2]. Even though many different theories have been proposed regarding the pathogenesis of these lesions, the exact cause has not been fully elucidated. According to the most widespread theory, neural cysts originate from adjacent joints and contain synovial fluid [3, 4]. The core principle involved is that in cases of arthritis high intra-articular pressure due to joint effusions drives synovial fluid extra-articularly through vulnerable capsular sites with the least resistance. One such site is the entry point of articular nerve branches allowing synovial accumulations to propagate under the neural sheath distally and proximally forming cysts along the parent nerve.

Although neural cysts are considered to be a rare cause of cubital tunnel syndrome, there are studies estimating the prevalence rate of these cysts to be 3–8% in these patients [5]. In most cases, these lesions are diagnosed intraoperatively, though in suspected cases of cubital tunnel syndrome due to neural cysts, as in patients with elbow arthritis or palpable soft tissue masses additional imaging studies including magnetic resonance imaging (MRI) or ultrasound may be needed for preoperative diagnosis [6–8]. Despite the fact that the management of these lesions in cases of associated neurological symptoms is quite straightforward and includes surgical decompression of the nerve, there is some controversy regarding the optimal surgical technique for addressing the cysts and reduction in the recurrence rate. The most

✉ Dimitrios V. Papadopoulos
di_papadopoulos@yahoo.gr

[1] Department of Orthopedics, University Hospital of Ioannina, Stavros Niarchos Avenue, 45500 Ioannina, Greece

[2] Karaoli kai Dimitriou 5, 45332 Ioannina, Greece

common treatment strategy entails drainage of the contents through simple incision of the cysts or excision of the cysts and disconnection of the parent nerve with the joint by dissecting the articular branch [9].

This report aims to describe a case of cubital tunnel syndrome caused by ulnar intraneural cysts in a patient with posttraumatic elbow joint arthritis. The overt signs of arthritis in the adjacent joint further support the theory that arthropathy is a major precipitating factor for the development of these lesions.

Case presentation

A 60-year-old right-handed female patient presented to the outpatient clinic of our department complaining of chronic intermittent pain in her right arm for the past 4 months, rated 4–7/10. Pain was aggravated by heavy work during certain periods of strenuous agricultural activities since she was a farmer, mainly located on the medial side of her elbow and radiating down to the ulnar side of her forearm and hand. She also reported that during flare-ups, which usually lasted roughly a week, she had to restrict work and use NSAIDS for alleviation of pain. For the past 2 months, flare-ups were more frequent and she additionally developed numbness and decreased sensation in the ulnar aspect of her hand, mainly felt along the ring and small fingers. She denied any motor neurological deficits such as muscle weakness. Her previous medical history revealed that she had suffered from a distal humerus fracture due to a fall 30 years ago, but not any other medical co-morbidities.

On inspection, there was not any obvious deformity of the right arm, while elbow's flexion–extension as well as pronation–supination were painless and within normal range. Tinel's sign was positive over the course of the ulnar nerve in the cubital tunnel. Froment's and Wartenberg's signs were negative. Motor examination did not reveal any signs of weakness, while sensation to light touch was affected along the small finger and the ulnar aspect of her ring finger. Posttraumatic arthritis was evident in plain X-rays as indicated by the presence of osteophytes around the trochlea and joint space reduction. Electrodiagnostic studies including nerve conduction studies and electromyography were also performed, demonstrating marked delay in motor and sensory nerve conduction velocities consistent with cubital tunnel syndrome.

Diagnosis of cubital tunnel syndrome was made, and surgical treatment was advised. Patient consented for surgery, and she underwent surgical exploration under general anesthesia. A curved 10-cm incision was made along the trajectory of the ulnar nerve on the medial aspect of the elbow, and dissection was carried out until identification of the ulnar nerve. The ulnar nerve was found to be swollen with multiple cysts along its course (Fig. 1). Cautious decompression of the nerve was performed, releasing the nerve in all possible sites of compression one by one, as described by Possner (Fig. 2) [10]. Decompression of the nerve was performed with great care due to dense adhesions between the nerve and the surrounding soft tissue. After decompression of the nerve at all common sites of compression along the cubital tunnel, the intraneural cysts were addressed. The first step during management of these lesions was incision and drainage of the cysts. Afterward, the wall of each of the cysts was cautiously resected under loupe magnification with great care not to damage the nerve fascicles (Fig. 3).

Postoperatively, free elbow range of motion was allowed. The patient reported subsidence of pain and numbness shortly after the surgery, and she remained free of symptoms until the last follow-up 3 months postoperatively.

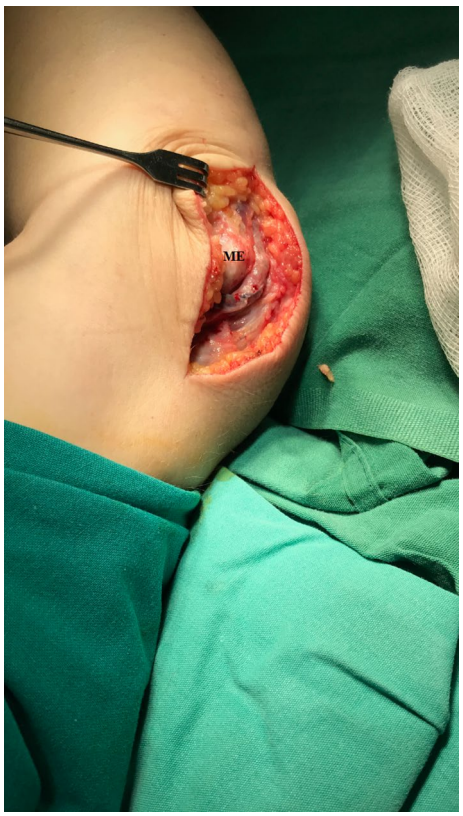

Fig. 1 An intraoperative image demonstrating the ulnar nerve as it passes behind the medial epicondyle (ME) after decompression of the nerve at all possible sites of compression at the cubital tunnel. The intraneural cysts are clearly visible along the course of the nerve

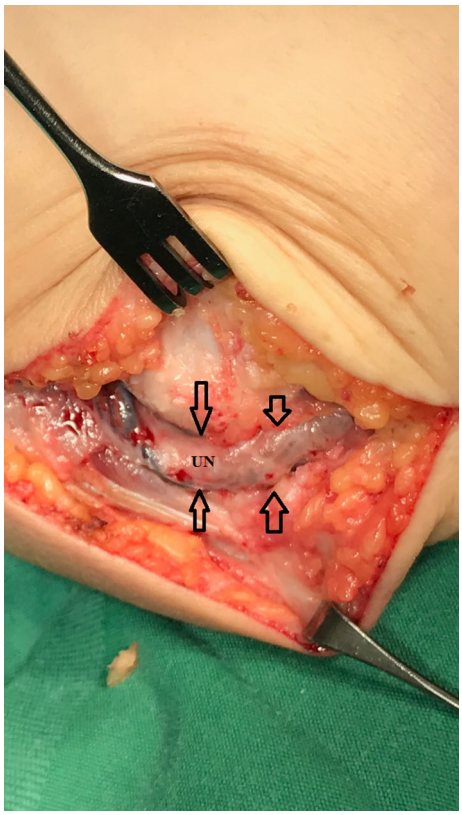

Fig. 2 Intraoperative image of the ulnar nerve (UN) at the cubital tunnel. The ulnar nerve is shown enlarged and swollen due to the numerous and closely located intraneural cysts (arrows)

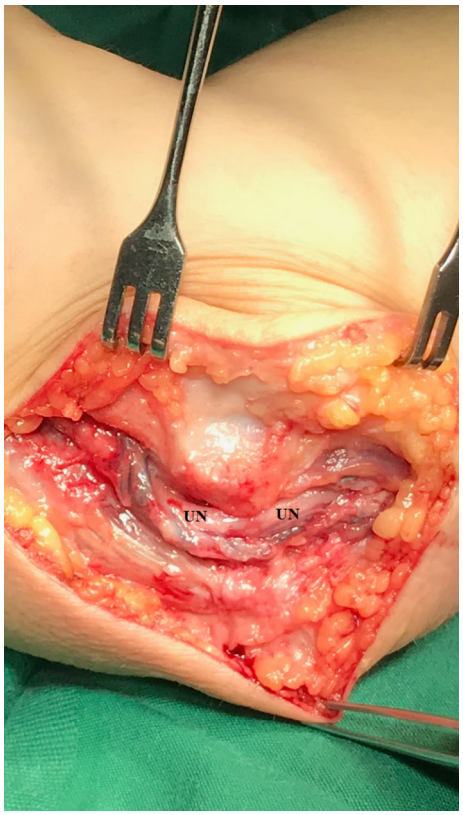

Fig. 3 Intraoperative image demonstrating the decompressed ulnar nerve (UN) after excision of the cysts

Discussion

The common peroneal nerve is reported to be the site of neural cysts's development in about 75% of the cases [11]. The first report of these lesions was back in 1901 by Hartwell who described an intraneural cyst within the median nerve [12]. Since then, several case reports and small case series have been published and shed light to various aspects of this disorder, such as the pathogenesis, clinical symptoms, imaging findings, electrophysiologic work-up and management of these lesions. According to a recent systematic review, neural cysts around the elbow comprise only 9% of all cases [13]. However, most of the studies regarding development of neural cysts around the ulnar nerve at the cubital tunnel have described cases of epineural cysts, while intraneural cysts are much less commonly reported with only few studies describing these lesions (Table 1) [5, 9, 14–20].

There is some controversy regarding the etiology of neural cysts. Three different theories have been proposed for the development of these lesions: the degenerative theory, the tumoral theory and the synovial articular theory. According to the degenerative theory, a certain degree of degeneration of the surrounding neural sheaths (i.e., epineurium, perineurium) results in mucus formation which is subsequently walled off by these sheaths and forms cysts along the nerve [21]. The tumoral theory, which is the least favored, speculates that these cysts are similar to other cystic lesions and neural tumors while the synovial theory described by Spinner is constantly gaining ground [4, 22]. According to Spinner et al., a connection of the joint with the parent nerve through an articular branch is responsible for the formation of the cysts. The fact that the vast majority of the existing

Table 1 Studies with similar reported cases of intraneural ulnar cysts

References	Number of cases	Management of the cysts
Son et al. [15]	1	Incision and drainage
Li et al. [19]	1	Epineurectomy and evacuation
Tong et al. [16]	6	Incision and drainage
Chang et al. [5]	3	Complete resection
Mobbs et al. [14]	1	Complete resection
Xu et al. [9]	1	Drainage and partial resection
Mingchan et al. [20]	1	Drainage and partial resection
Chick et al. [17]	1	Drainage and epineurectomy
Inhofe et al. [18]	1	Complete resection

studies describe cases of lesions that developed in the vicinity of joints reflects the major role of the intra-articular pathology.

Neural cysts may displace nerve fascicles and cause compressive neuropathy leading to neural entrapment syndromes such as cubital tunnel syndrome. In several cases, they may pose a differential diagnosis problem, since they can masquerade as nerve sheath tumors or other mass occupying lesions [14]. Preoperative identification of the exact cause of cubital tunnel syndrome is necessary when a more complex pathology such as neural tumors or neural cysts is suspected. Further work-up with additional imaging studies including MRI or ultrasound is recommended for better preoperative planning. Two imaging MRI findings have been described: the signet ring and the transverse limb signs which are considered to be highly specific of these lesions [15]. Although MRI is the most commonly performed study for diagnosis of these lesions, in most cases MRI fails to identify the communicating articular branch. Recently, high-resolution MRI with or without arthrography has been suggested as a reliable method for demonstration of this neural branch [7].

Surgical management of ulnar nerve neural cysts in cubital tunnel should follow the same principles applied in all cases of cubital tunnel syndrome and in any case of neural cysts. Exploration and decompression of the nerve at every possible site of compression along its course in cubital tunnel should be performed, while cysts must be addressed accordingly. Although the best method of addressing these cysts is controversial, many authors recommend dissection of the articular branch of the nerve since based on the popular synovial theory this branch is the culprit for the development of the cysts through which the parent nerve is connected with the joint [9, 13]. Similar to the 4-D technique that has been proposed for the treatment of neural cysts around the common peroneal nerve (dissection of the nerve, disarticulation of the tibiofibular joint, decompression of the cyst and disconnection of the articular branch), identification and division of the articular branch are believed to be a critical step of the procedure in order to decrease the risk of recurrence, which may be up to 11% [12, 23]. Simple incision and drainage of the cysts are believed to result in complete resorption of the cysts and by many are considered to be the treatment of choice [15]. Although some believe that more radical interventions such as complete cyst resection are contraindicated due to the high risk of iatrogenic nerve injury, we consider that cautious resection of the cysts is the optimal method for reduction in risk of recurrence. In the largest case series including six patients with cubital tunnel syndrome due to intraneural cysts, Tong et al. showed that additional subcutaneous transposition of the nerve following drainage of the cysts and ligation of the connecting articular branch yield excellent results, though this practice has not been evaluated by other authors [16].

There are strengths and limitations of this case report that must be addressed. This report describes a patient in whom development of intraneural ulnar cysts can be attributed to a profound risk factor such as the posttraumatic elbow arthritis. The proximity of the arthritic joint with the affected nerve underlines the causative role of the articular pathology and further supports the synovial theory. The main limitation of the study is the relatively short follow-up; the possibility of recurrence cannot be excluded. Additionally, even though this study includes intraoperative images that clearly demonstrate the intraneural cysts before and after decompression, the articular branch of the nerve is not shown, while there are no intraoperative images during the microsurgical decompression of the nerve and resection of the cysts.

Conclusively, although ulnar compression neuropathy at elbow due to intraneural cysts is a rare event, surgeons must always consider the possibility of this disorder in cases of cubital tunnel syndrome associated with elbow joint arthritis. In such cases, further work-up including MRI is recommended and surgical management should be based on three main principles: decompression of the ulnar nerve at all possible sites of entrapment in cubital tunnel, dissection of its articular branch and excision of the cysts.

Compliance with ethical standards

Conflict of interest The authors Dimitrios V. Papadopoulos, Ioannis Kostas-Agnantis, Dimitrios Kosmas, Evridiki Tsiomita, Maria A. Korompilia, Ioannis D. Gelalis, and Anastasios V. Korompilias declare that they have no conflict of interest.

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