Ulnar nerve palsy caused by synovial protrusion in synovial chondromatosis of the elbow: a case report and literature review

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Abstract

We describe an unusual case of ulnar nerve compression (cubital tunnel syndrome) caused by synovial protrusion in primary synovial chondromatosis of the elbow in a 59-year-old man. Magnetic resonance imaging is a useful tool for diagnosing this rare condition. Surgical excision of the intra-articular multiple loose bodies and ulnar nerve decompression were performed. The clinician should be aware of primary synovial chondromatosis as one of the causative factors of cubital tunnel syndrome.

Keywords

cubital tunnel syndrome, elbow, synovial chondromatosis, ulnar nerve

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Introduction

We report a case of ulnar nerve compression syndrome by a protruded capsule in synovial chondromatosis (SC) of the elbow joint and also conducted a literature review.

Case report

A 59-year-old man presented with progressive numbness in the right little finger and ulnar half of the right ring finger for 3 months. He had a 6-year history of right elbow pain. Physical examination revealed that active and passive ranges of motion (ROMs) of the right elbow were limited between 20° and 120° of flexion with terminal pain. Muscle weakness appeared during finger abduction and adduction for the right hand, although there was no apparent muscle atrophy. The right hand grasping power was weaker than that of the left hand. There was hypoesthesia over the right little finger and ulnar half of the right ring finger. Froment's sign was negative, and claw finger deformity was not seen. There was a positive Tinel's sign over the right cubital tunnel. Motor nerve conduction study revealed a reduced conduction velocity, delayed latency, and decreased amplitude and area, which were consistent with ulnar nerve compression at the elbow joint.

Plain X-ray and computed tomography (CT) of the right elbow showed a few small loose bodies at the anterior and posterior aspects of the elbow joint and some small osteophytes at the radial neck, radial fossa, coronoid fossa and olecranon fossa (Fig. 1). Magnetic resonance imaging (MRI) revealed a T1-low and short-tau inversion recovery (STIR)-high intensity cystic lesion image, including T1-low and STIR-iso intensity multiple small nodules at the cubital tunnel and intra-articular numerous small tumours in the right elbow joint.

Surgical treatment was attempted because the symptoms were progressively worsened. Arthroscopic findings under general anaesthesia revealed many small white loose bodies inside the elbow joint, although the synovium was whitish and appeared to be inactive (Fig. 2a). Surgical exploration of the ulnar nerve

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Figure 1. Anteroposterior (a) and lateral (b) plain radiographs of the right elbow. A small loose body was seen at the anterior part of the elbow joint. Three-dimensional computed tomography views (c and d) demonstrated some bone spur formations (c and d, arrowheads) and a few loose bodies (c and d, arrows). Short-tau inversion recovery (STIR)-weighted magnetic resonance imaging coronal (e) and sagittal (f and g) views of the right elbow. STIR-low-iso weighted nodular mass lesions were found inside the joint (e and f). STIR-high weighted cystic mass with STIR-low-iso weighted small tumours were localized to the distal part of the medial epicondyle, coincident with the cubital tunnel (g, arrowheads).

showed a protrusion of the joint capsule compressing the nerve at the cubital tunnel (Fig. 2b). This protruded capsule was filled with multiple small loose bodies and was excised. After the medial joint capsule proximal to the medial epicondyle was incised, many loose bodies were removed from the anterior and posterior joint cavities (Fig. 2c) and partial synovectomy was performed. In addition, subcutaneous anterior transposition of the ulnar nerve was performed. Histopathology revealed that these loose bodies were benign chondromas or osteochondromas with no evidence of malignancy (Fig. 2d and 2e). Postoperatively, his symptoms gradually improved with no specific rehabilitation program. At the 1-year postoperative follow-up, his complaints of elbow pain and numbness of the fingers had completely resolved. Elbow ROM recovered from 0° to 140° of flexion with no difference in the grasping power between both hands. He still was asymptomatic with no neurological deficits, and had no clinical and radiologic evidence of recurrence 2 years after surgery.

Discussion

SC is a benign disorder characterized by synovial metaplasia, with formation of numerous cartilaginous nodules (with or without ossification) in the joint, tendon sheath or bursa.¹ Although the most frequent location is the knee and hip joints, SC could occur in upper extremity joints, such as the shoulder, elbow and wrist.²⁻⁴ The common symptoms of SC are pain, swelling, stiffness and locking because of loose bodies impinging the affected joint, resulting in secondary osteoarthritis for a long duration.^{2,3} The involvement of the ulnar nerve at SC of the elbow joint is extremely rare.

For this review, we searched the key words 'synovial chondromatosis, elbow, ulnar nerve' using the Medline database to obtain the articles published from 1950 to until 2015. We retrieved six articles from the key words. We also scrutinized the reference sections of the selected articles to identify additional literatures, and, as a result, 10 articles were collected. Then, we excluded

Figure 2. (a) Arthroscopic view from the anterolateral portal showed multiple white loose bodies (arrows) and whitish synovium (arrowheads). h, humerus; r, radial head. (b) Intra-operative findings showed a protrusion of the capsule at the cubital tunnel. Arrows indicate the protruded capsule. m, medial epicondyle; t, triceps muscle; u, ulnar nerve. (c) Over 100 multiple loose bodies. Haematoxylin and eosin (d) and alcian blue (e) staining showed chondroid metaplasia with no evidence of malignancy.

three articles because those were not published in English and, finally, seven cases about ulnar nerve palsy associated with SC were reviewed (Table 1).^{5–11} In these cases, the nerve compression was not confirmed until the time of surgical exploration. Our patient had protrusion of the capsule to the cubital tunnel as an obvious space-occupying lesion on the pre-operative MRI. Also, MRI was useful to diagnose SC in our patient with intra-articular small multiple loose bodies in the elbow, although these were not detected on the preoperative plain X-ray and CT.

SC can be primary or secondarily associated with osteoarthritis, traumatic osteochondral fracture or osteochondritis dissecans. SC is most commonly seen in patients aged 30 years to 40 years old, and the prevalence in men is twice than that in women.^{2–4,9,12} The reported cases, including our patient, with compression ulnar neuropathy were older (mean of 54.6 years) than those without ulnar neuropathy. We diagnosed the present case as having primary SC because there was no trauma or obvious osteoarthritic change. However, it is hard to clinically and radiologically differentiate between primary and secondary SC because primary chondromatosis could result in osteoarthritis.⁴

Recently, clonal chromosomal alterations have been reported in some cases of SC and, especially, chromosome 6 anomalies have been observed in five of eight reported cases.^{13–15} Thus, cytogenetic analysis might help in diagnosing this entity more accurately.

Milgram et al.⁴ divided SC into three separate phases; the early phase consists of intrasynovial changes only, with no loose bodies (phase I); the transition phase is characterized by intrasynovial nodules and loose bodies (phase II); and, in the final phase, the synovium becomes quiescent and osteochondral loose bodies remain (phase III). If the synovium appears to be active in phases I and II, extensive synovectomy might be necessary to minimize its recurrence, whereas, in phase III, removal of loose bodies without extensive synovectomy might suffice. All reported SC cases with ulnar neuropathy only had removal of loose bodies and/or partial synovectomy. Our case was in stage III and was successfully treated by the removal of loose bodies and partial synovectomy, with no recurrence during 2 years of follow-up. However, long-term follow-up is necessary because SC has a potential risk of local recurrence and/or malignant transformation to chondrosarcoma.^{16–18}

Table 1. Synovial chondromatosis complicated by ulnar nerve compression syndrome in the elbow.

Reference	Age (years)	Sex	Dominant side	Affected side	History of elbow pain	Surgical treatment
Fahmy and Noble (1981) ⁵	62	Male	Not described	Right	5 years	Removal of loose bodies Partial synovectomy Nerve decompression
Lister et al. (1981) ⁶	55	Male	Not described	Left	Not described	Removal of loose bodies Nerve decompression
Ruth and Gloves (1996) ⁷	27	Male	Right	Right	2 years	Removal of loose bodies Anterior subcutaneous nerve transposition
Nogueira et al. (1999) ⁸	53	Female	Right	Right	1 year	Removal of loose bodies Nerve decompression
Kamineni et al. (2002) ⁹	51	Male	Right	Left	20 years	Removal of loose bodies Humeroulnar arthroplasty Anterior subcutaneous nerve transposition
Kim et al. (2008) ¹⁰	62	Female	Not described	Right	Not described	Removal of loose bodies Mini-medial epicondylectomy Anterior subcutaneous nerve transposition
Weerasuriya and Swaminathan (2011) ¹¹	68	Male	Right	Right	Not described	Removal of loose bodies Nerve decompression
Present study	59	Male	Right	Right	6 years	Removal of loose bodies Partial synovectomy Anterior subcutaneous nerve transposition

Conclusions

Cubital tunnel syndrome as a result of primary SC is uncommon. MRI is an effective diagnostic modality. The clinician should be aware of primary SC as one of the causative factors of cubital tunnel syndrome.

Author Contributions

All authors read and approved the final manuscript. KY, MM and YS were involved in the diagnosis and treatment of the case. TH helped to draft the manuscript. TT and JH helped in the design, interpretation and writing of the manuscript.

Declaration of Conflicting Interests

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Ethical Review and Patient Consent

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