Ulnar Neuropathy due to Cubital Tunnel Syndrome Caused by Anconeus Epitrochlearis: A Case Report

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INTRODUCTION

Ulnar neuropathy involving the cubital tunnel is the second most common entrapment neuropathy, following carpal tunnel syndrome\textsuperscript{1}. A number of risk factors for the development of cubital tunnel syndrome have been suggested, including diabetes mellitus, varus or valgus deformity of the elbow, repetitive tasks that require protracted elbow flexion, and the repetitive use of vibrating tools\textsuperscript{2}. Anconeus epitrochlearis (AE) muscle is a congenital accessory muscle, which originates in the medial epicondylye (ME) of the humerus and inserts onto the olecranon (OL) process the ulna. It can be found in normal elbows with an incidence ranging between 4\% to 34\%\textsuperscript{3,4}. AE is a common variation in the cubital tunnel retinaculum of the elbow; however, its association with ulnar neuropathy is rare.

CASE REPORT

A 24-year-old, right-handed male patient presented with a 1-month history of continuous tingling paresthesia involving left medial hand and fingers accompanied by clumsiness of left hand and fingers. The pain onset was sudden and marked by a severe aching pain in the medial elbow and medial scapular areas, without any prior cause. Two weeks of excruciating pain led to clumsiness of the left hand and fingers gradually occurred. Magnetic resonance imaging (MRI) of the left elbow revealed a swollen ulnar nerve with perineural enhancement involving anconeus epitrochlearis (AE) muscle within the cubital tunnel. The operation revealed adhesion of the AE muscle to the left ulnar nerve. Open in-situ decompression of the ulnar nerve without transposition eventually alleviated the pain and weakness associated with ulnar neuropathy. The patient slowly recovered 6 months after surgery. The AE muscle is a common anatomic variation, with a prevalence of up to 34\%. However, the prevalence of ulnar neuropathy associated with the AE muscle is unknown. It is a congenital accessory muscle between the medial humeral epicondyle and the olecranon covering the posterior aspect of the cubital tunnel. It is usually diagnosed intraoperatively and not preoperatively in the absence of no imaging studies. The clinical presentation of ulnar neuropathy caused by the AE usually differs from idiopathic disease, including younger age at onset, rapid progression with a short duration of symptoms, and edema of the AE muscle on the MRI.

Key Words: Anconeus epitrochlearis, Cubital tunnel syndrome, Peripheral nerve, Ulnar nerve

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detected in the cubital tunnel. The patient complained of persistent tingling paresthesia involving the medial wrist, hand, and little finger.

The electrophysiological study performed at the hospital where he was previously treated revealed acute ulnar neuropathy around the left elbow without mention of denervation. Because clinical and electrophysiological findings were consistent with acute ulnar neuropathy at the elbow with advanced motor signs, an MRI of the elbow was requested. MRI of the left elbow revealed diffuse swelling of the left ulnar nerve at and distal to the cubital canal (Figure 1B-D). The left ulnar nerve was compressed against ME by the AE muscle at the level of the cubital tunnel. Mild perineural enhancement of the left ulnar nerve just distal to the cubital tunnel was observed (Figure 1C, D). Laboratory parameters including erythrocyte sedimentation rate, C-reactive protein, rheumatoid factor, antinuclear antibody, and creatinine kinase were normal. Considering the course of ulnar nerve palsy and abnormal MRI findings, an exploration of the ulnar nerve was performed.

During the surgery, the fibrous facial layer (Osborne's ligaments) over the cubital tunnel was not thickened or hypertrophied. The ulnar nerve was swollen with a slightly rubbery consistency (Figure 2A). The adhesion between the ulnar nerve and AE muscle was found and dissected (Figure 2B). The AE was excised and the ulnar nerve

**Figure 1.** Pain distribution and magnetic resonance imaging (MRI) findings of cubital tunnel syndrome associated with anconeus epitrochlearis (AE).

- **Ⓐ:** Pain distribution. Black area denotes the painful area with paresthesia and allodynia.
- **Ⓑ:** Sagittal (left) and coronal (right) T2-weighted images of the left elbow showing the ulnar nerve (arrowheads) compression by AE (arrow), with increased signal intensity of the nerve, suggesting intraneural edema.
- **Ⓒ:** Axial fat-suppression T2-weighted MRI (left) reveals compression of the swollen ulnar nerve (arrowhead) by the AE (arrow).
- **Ⓓ:** Enhanced, axial T1-weighted images of the lesion immediately distal to the cubital tunnel. Mild perineural enhancement was observed in the surrounding flexor carpi ulnaris muscle. ME: medial epicondyle; OL: olecranon.

**Figure 2.** Intraoperative findings during decompression of the left cubital tunnel.

- **Ⓐ:** The left ulnar nerve (arrowhead) is compressed by the anconeus epitrochlearis (AE) muscle (arrows) within the Osborne's ligament.
- **Ⓑ:** The left ulnar nerve (arrowhead) is decompressed following resection of the Osborne's ligament and the AE muscle (arrow). MABC: medial antebibrachial cutaneous nerve; ME: medial epicondyle.
was distally dissected along its course to the flexor carpi ulnaris muscle. Although postoperative course was uneventful, the rate of recovery from ulnar neuropathy was not as fast as expected. The pinch and grip strength of the left hand and fingers did not show any improvement despite improvement in paresthesia until two months postoperatively. Then, the pinch and grip strength was gradually recovered with minimal paresthesia in the fifth finger and hypothenar area at 6 months postoperatively. No evidence of ulnar neuropathy was detected at 12 months postoperatively.

**DISCUSSION**

1. AE Muscle

AE is a small triangular muscle originating in the ME of the humerus, inserting laterally on the OL and proximal posterior surface of the ulnar. Its anatomic course is similar to that of the cubital tunnel retinaculum. It was postulated that the cubital tunnel retinaculum is the remnant of the AE muscle. It expands and initiates the extension of the elbow and is regarded as a continuation of the triceps brachii muscle and enables extension of the forearm. Thus, AE protects the ulnar nerve and assists the triceps muscle in preventing subluxation of the ulnar nerve. In contrast to the triceps, this muscle is innervated by a branch of the ulnar nerve. The AE, when present, forms the roof of the cubital tunnel, replacing Osborne’s ligament. It was suggested that Osborne’s ligament is a fibrous remnant of the AE that remains after the muscle regresses. The anconeus muscle occupies more space than the typically present Osborne fascia. The prevalence of this anomalous muscle is estimated to be 3% to 34% based on cadaveric and imaging studies and is often found bilaterally.

2. AE and Cubital Tunnel Syndrome

The AE muscle exerts a compressive force on the ulnar nerve and its association with cubital tunnel syndrome has long been reported. Although the AE is a common anatomical variant of the elbow, the prevalence of cubital tunnel syndrome due to AE is unknown. Its association with cubital tunnel syndrome is generally detected during operation, not preoperatively. It was suggested that ulnar neuropathy associated with AE might be dynamic rather than static compression and exhibited different clinical characteristics compared with idiopathic cubital tunnel syndrome including more rapid progression, younger age at onset, and shorter duration of symptoms.

The current case also appeared as a sudden outbreak in young patients, as previously reported. The neurophysiological findings of ulnar neuropathy associated with AE also differ from those involving patients with idiopathic cubital tunnel syndrome. Velocity drop or conduction block of the ulnar nerve indicating the subacute onset of symptoms is common rather than chronic demyelination generally seen in idiopathic ulnar neuropathy.

Ultrasound and MRI are reliable imaging modalities used to show structural abnormalities of the cubital tunnel and can reveal the causes of secondary ulnar neuropathy and anatomical variants, as in the current patient. Edema of the AE in the MRI was associated with medical elbow pain. In the MRI of the present case, the AE size was not large enough to directly compress the ulnar nerve, but close adhesion with the nerve was suspected (Figure 1). The adhesions between the AE and the ulnar nerve were confirmed by surgical findings (Figure 2). The ulnar nerve was swollen. A weak perineural enhancement of the ulnar nerve and flexor carpi ulnaris muscle was observed in the distal cubital tunnel. However, it is not known exactly whether this perineural enhancement is pathognomonic for ulnar neuropathy triggered by AE.

3. Treatment of Ulnar Neuropathy Associated with AE

Complete excision of the AE is widely accepted as the definitive treatment for the ulnar neuropathy associated with AE, but whether to transpose the ulnar nerve remains controversial. Several treatment options have been suggested including muscular release or complete excision with in-situ decompression of the ulnar nerve and complete excision of the muscle with transposition of the ulnar nerve to prevent subluxation, depending on the surgeon’s decision about the extent of neurolysis to prevent dislocation of the ulnar nerve and the extent of muscle excision. More rapid and complete recovery with a low incidence of recurrence than idiopathic cubital tunnel syndrome is a common finding in most studies investigating the AE muscle. A more comprehensive study investigating the pathophysiology of cubital tunnel syndrome caused by AE is warranted in the near future.

**CONCLUSION**

The manifestations of ulnar neuropathy associated with AE differ
from those of idiopathic cubital tunnel syndrome. They include younger age at onset, more rapid progression with a shorter duration of symptoms, neurophysiologic findings of velocity drop or conduction block of the ulnar nerve, and perineuritis and edema of AE on MRI. Therefore, when ulnar neuropathy due to cubital tunnel syndrome is suspected in young patients with rapid progression, an imaging study is essential to identify the structural lesion around the cubital tunnel.

Conflicts of Interest:
No potential conflict of interest relevant to this article was reported.

REFERENCES