



Attrition rupture of ulnar nerve in a patient with rheumatoid elbow arthritis

A case report

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Abstract

Rationale: Cubital tunnel syndrome has been recognized as a common pathology in rheumatoid arthritis (RA) of the elbow. We encountered a patient with RA of the elbow showing attrition rupture of the ulnar nerve. This pathology is extremely rare, and we discussed preventive measures for similar cases in the future based on our case.

Patient concerns: A 53-year-old woman, received drug treatment for RA since 30 years earlier, had numbness in the left ulnar nerve territory, dorsal interossei muscle atrophy, and resulting claw hand.

Diagnoses: Plain x-ray examination showed bone destruction of the left elbow joint and marked osteophyte formation in the medial joint space. In nerve conduction velocity (NCV) tests, the Motor NCV was immeasurable in the ulnar nerve territory. Based on these findings, a diagnosis of left cubital tunnel syndrome was made, and anterior transposition of the ulnar nerve was planned.

Interventions: When the ulnar nerve dissection was advanced, about 80% portion of the ulnar nerve was ruptured. After the ends of the divided nerve were freshened, end-to-end anastomosis was possible by anterior transposition of the ulnar nerve.

Outcomes: Two years after the operation, numbness and muscle atrophy also remained. There were no changes in the level of daily activities after the operation. However, motor NCV, showed improvement (22.8 m/s) after the operation.

Lessons: In patients with RA showing ulnar neuropathy symptoms, marked osteophyte formation in the medial joint space or valgus deformity may indicate attrition nerve rupture. In the future, when such patients with RA are examined, active nerve exposure and dissection should be considered in terms of ulnar nerve protection.

Abbreviations: NCV = nerve conduction velocity, RA = rheumatoid arthritis.

Keywords: attrition nerve rupture, motor nerve conduction, rheumatoid arthritis, ulnar nerve

1. Introduction

Cubital tunnel syndrome is the second most common peripheral neuropathy of the upper limb.^[1] Its cause is trauma or ulnar nerve entrapment at the elbow, and resulting sensory disturbance and motor paralysis in the ulnar nerve territory are observed.^[2] In

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Received: 23 January 2018 / Accepted: 30 March 2018 http://dx.doi.org/10.1097/MD.000000000010535 1965, Deandrade and Casagrande^[3] were the first to report cubital tunnel syndrome in rheumatoid arthritis (RA) of the elbow. Thereafter, cubital tunnel syndrome has been recognized as a common pathology in RA of the elbow.^[4]

We encountered a patient with RA of the elbow showing attrition rupture of the ulnar nerve. This pathology is extremely rare, and we discussed preventive measures for similar cases in the future based on our case.

2. Case report

A 53-year-old woman had received drug treatment for RA since 30 years earlier. She had also undergone operations such as right ankle arthrodesis and posterior cervical spinal fusion for multiple arthritic disorders. She visited our hospital due to numbness in the left ulnar nerve territory persisting for 1 year. At the first consultation, numbness in left ulnar nerve territory peripheral to the forearm, dorsal interossei muscle atrophy, and resulting claw hand were observed (Fig. 1A). Froment sign was positive, and Tinel like sign was positive at the cubital tunnel. The range of motion of the left elbow was 140° for flexion and -40° for extension. The Visual Analog Scale (VAS) score was 2/10, and the Quick Disabilities of the Arm, Shoulder, and Hand (Q-DASH) score was 75.00/100. Plain x-ray examination showed bone destruction of the left elbow joint and marked osteophyte formation in the medial joint space (Fig. 1B). In nerve conduction

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Figure 1. Preoperative physical findings of plain x-ray findings. A: Dorsal interossei muscle atrophy and a claw hand of the affected limb were observed. B: Plain x-ray examination showed marked osteophyte formation in the medial joint space and valgus deformity.

velocity (NCV) tests, the Motor NCV was immeasurable in the ulnar nerve territory. Based on these findings, a diagnosis of left cubital tunnel syndrome was made, and anterior transposition of the ulnar nerve was planned.

When the ulnar nerve dissection was advanced from a proximal site, adhesion was marked at the elbow joint. The joint capsule on the medial side of the elbow showed rupture, and the continuity of the ulnar nerve was interrupted at this site (Fig. 2A). About 80% portion of the ulnar nerve was ruptured (Fig. 2B). After the ends of the divided nerve were freshened, end-to-end anastomosis was possible by anterior transposition of the ulnar nerve (Fig. 2C). The ruptured joint capsule was covered with the flexor aponeurosis (Fig. 2D). After the operation, external immobilization with the elbow in flexion for 2 weeks was necessary.

Two years after the operation, the range of motion of the left elbow joint was 135° for flexion and -30° for extension. The grip strength was 0 kg (0% compared with the unaffected side), and the pinch strength was 0.25 kg (14.3% compared with the unaffected side). The VAS score was 2/10, and the Q-DASH score was 65.91/100. Tinel sign was observed at the site 13 cm peripheral to the medial condyle of the humerus. There was persistent numbness in the ulnar nerve territory. The dorsal interossei muscle atrophy and claw hand also remained. There were no changes in the level of daily activities after the operation. However, Motor NCV, which was immeasurable before the operation, showed improvement (22.8 m/s) after the operation.

3. Discussion

Attrition rupture of the ulnar nerve in RA of the elbow is extremely rare. To our knowledge, only 3 cases have been reported. Moore and Weiland reported a 56-year-old woman with a 37-year history of RA showing attrition rupture of the ulnar nerve. She had developed ulnar neuropathy symptoms 2 years earlier, and the preoperative motor NCV of the ulnar nerve was immeasurable. Kalaci et al^[6] reported a 68-year-old man with a 30-year history of RA showing attrition rupture of the ulnar nerve. He had had ulnar neuropathy symptoms persisting

for about 10 years such as intrinsic muscle atrophy and sensory impairment. The preoperative ulnar nerve motor NCV was immeasurable. On the other hand, Ochi et al^[7] reported a 68-year-old woman with a 47-year history of RA showing attrition rupture of the ulnar nerve. She had had ulnar neuropathy symptoms for 2 years, and the preoperative motor NCV of the ulnar nerve was 10.8 m/s. One of the 3 previously reported patients underwent no suturing of the ulnar nerve, and another underwent nerve suturing. In the other patient, the surgical technique was unknown. After the operation, improvement was observed in sensory impairment but not in motor paralysis. In none of these studies, changes in the NCV of the ulnar nerve after the operation were evaluated.

Our patient showed improvement in sensory impairment in the ulnar nerve territory but no recovery of motor paralysis 2 years after the operation. Gaul^[8] performed nerve suturing in 41 patients with ulnar nerve injuries, and observed improvement in motor paralysis. Kim et al^[9] surveyed treatment results in 654 patients with ulnar neuropathy, and observed improvement in motor paralysis in 42 (72%) of 58 patients who underwent nerve suturing. Woo et al^[10] described that functional recovery after ulnar nerve injuries at the elbow requires about 5 years. In our patient, no improvement was observed in motor function, but the NCV of the ulnar nerve was recovered 2 years after the operation, which may be a pathology that can lead to functional improvement after long-term follow up.

4. Conclusions

Based on the findings in our patient as well as the previously reported 3 patients, leaving ulnar neuropathy symptoms untreated in rheumatoid elbows has a risk of developing attrition nerve rupture. [5–7] In patients with RA showing ulnar neuropathy symptoms, marked osteophyte formation in the medial joint space or valgus deformity may indicate attrition nerve rupture (Fig. 1B). [7] In the future, when such patients with RA are examined, active nerve exposure and dissection should be considered in terms of ulnar nerve protection. In addition, when

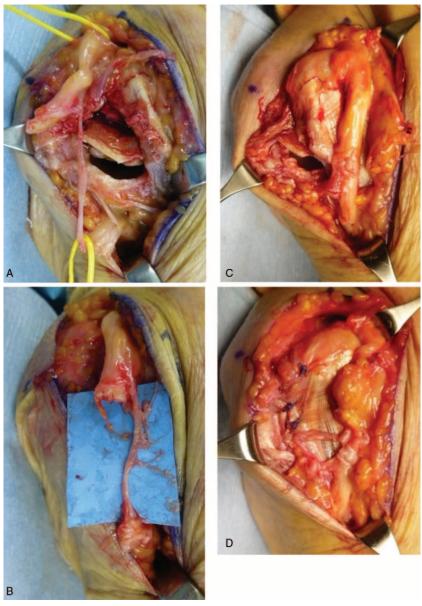


Figure 2. Surgical findings. A: Rupture of the joint capsule on the medial side of the elbow joint was observed, and there was no continuity of the ulnar nerve. B: About 80% of the ulnar nerve was damaged. C: Anterior transposition of the ulnar nerve and end-to-end anastomosis were performed. D: The ruptured joint capsule was covered with flexor aponeurosis.

preoperative informed consent is obtained, an adequate explanation of the possible presence of attrition rupture is also necessary.

Author contributions

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References

[1] Fernandez E, Pallini R, Lauretti L, et al. Neurosurgery of the peripheral nervous system: cubital tunnel syndrome. Surg Neurol 1998;50:83–5.

- [2] Palmer A, Hughes T. Cubital tunnel syndrome. J Hand Surg Am 2010;35:153-63.
- [3] Deandrade JR, Casagrande PA. Ulnar nerve entrapment in rheumatoid arthritis: a case report. Arthritis Rheum 1965;8:294–301.
- [4] Balagtas-Balmaseda OM, Grabois M, Balmaseda PF, et al. Cubital tunnel syndrome in rheumatoid arthritis. Arch Phys Med Rehabil 1983;64: 163–6.
- [5] Moore JR, Weiland AJ. Bilateral attritional rupture of the ulnar nerve at the elbow. J Hand Surg Am 1980;5:358–60.
- 6] Kalaci A, Aslan B, Yanat AN. Spontaneous rupture of ulna nerve due to neglected cubital tunnel syndrome associated with rheumatoid arthritis. J Clin Rheumatol 2007;13:217–8.
- Ochi K, Ikari K, Momohara S. Attritation rupture of ulnar nerve in an elbow of a patient with rheumatoid arthritis. J Rheumatol 2014;41:2085.
- [8] Gaul JS. Intrinsic motor recovery-a long-term study of ulnar nerve repair. J Hand Surg Am 1982;7:502–8.
- [9] Kim DH, Han K, Tiel RL, et al. Surgial outcomes of 654 ulnar nerve lesions. J Neurosurg 2003;98:993–1004.
- [10] Woo A, Bakri K, Moran S. Management of ulnar injuries. J Hand Surg Am 2015;40:173–81.