



Case Report

Intraneural ganglion of the ulnar nerve

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ABSTRACT

We report the case of a 76-year-old right-hand dominant man with hypertension and type 2 diabetes mellitus who presented with numbness of the right ring and little fingers for 8 months and a palpable mass lesion in the right forearm for 3 months before his hospital visit. The patient exhibited grip power weakness and atrophy of intrinsic muscles. Magnetic resonance imaging confirmed a well-defined oval mass of $1.5 \times 0.6 \times 0.6 \text{ cm}^3$ between the ulnar bone and nerve. This lesion was hyperintense on T2-weighted images and isointense on T1-weighted images. A zigzag incision was made for ulnar nerve exposure, and the tumor lesion was identified and carefully excised with minimal damage of ulnar nerve. A cystic mass invaded ulnar nerve extending approximately 1.5 cm was resected and reconstructed by sural nerve grafting. The patient was examined 6 months after surgery. His grip power and muscle strength increased, and no numbness or pain was detected. The outcome of this case suggests a potential surgical benefit exists even in cases of apparent chronic or severe denervation atrophy and that ganglion excision and nerve grafting should be considered.

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1. Introduction

Ganglia represent 60% of all hand and wrist tumors; however, intraneural ganglion cysts are rare, benign, mucinous cysts that occur within the epineurium. These cysts are also referred to as intraneural mucoid cysts or intraneural mucoid pseudocysts. This condition usually affects middle-aged men and presents with symptoms of compression neuropathy. Intraneural ganglion cysts can result in pain, paresthesia, and neurological deficit of the involved nerve. This tumor predominantly occurs in the lower extremity, with the common peroneal nerve and its branches involved in almost all cases.¹ Very few cases of ulnar nerve involvement have been reported.^{2–7} Here, we present the case of a patient with a histopathologic diagnosis of intraneural mucoid ganglion involving the ulnar nerve at the level of the forearm. Despite chronic motor weakness and muscle atrophy, the patient achieved significant recovery of muscle function following excision of the ganglion and nerve grafting.

2. Case report

A 76-year-old right-hand dominant male with hypertension and type 2 diabetes mellitus presented to our hospital with complaints

of numbness of the right ring and little fingers for 8 months. A palpable mass lesion was discovered in his right forearm 3 months before the hospital visit. The patient had been taking medication for hypertension and type 2 diabetes for more than 10 years. He had been previously evaluated by a nerve conduction study, which suggested carpal tunnel syndrome as well as “chronic” ulnar neuropathy. He denied having any nighttime paresthesia or numbness of any kind.

Physical examination of the patient's right upper extremity revealed no evidence of traumatic injury and no significant bone or joint deformity. Normal light touch sensation in cutaneous nerve territories of all the upper extremities was found. Motor strength of both the upper extremities of the patient was strong, except the ulnar intrinsic function of his right hand. Grip power weakness in the patient's right hand was noted. Handgrip strength of both hands was measured by a dynamometer and was found to be 42 and 47 kg in the right and left hands, respectively. In addition, there was obvious evidence of interosseous muscle atrophy in the patient's right hand (Fig. 1). Froment and Wartenberg signs were positive for the right hand. There was no intrinsic muscle weakness, atrophy, or pathologic signs in the left hand. Thumb opposition and abduction were strong, and bilateral thenar muscles were well preserved. Tinel sign was negative at the hand, wrist, elbow, and brachial plexus. Further, the subject showed no provocative signs of nerve compression at the elbow or wrist.

An electromyogram revealed ulnar neuropathy at the forearm. Focal abnormalities in the dorsal interosseous and abductor digiti

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Fig. 1. Preoperative image of the patient's hands. The right hand shows significant deepening of the dorsal web space with suggestion of dorsal interosseous muscle atrophy. The left hand shows no similar condition.

minimi muscles suggested axonal injury, and absent motor unit action potentials with no recruitment suggested high-grade nerve injury. There were no other significant motor or sensory conduction abnormalities. Electromyogram suggested a focal lesion at the ulnar nerve, and a magnetic resonance imaging (MRI) scan of the right forearm (Fig. 2) revealed a well-defined oval mass with dimensions of $1.5 \times 0.6 \times 0.6 \text{ cm}^3$ between the ulnar bone and nerve. This lesion was hyperintense and isointense on T2- and T1-weighted images, respectively. The patient underwent surgical exploration with a preoperative diagnosis of tumor with severe mononeuropathy of the right ulnar nerve. A zigzag incision was performed for ulnar nerve exposure, and the tumor lesion was identified (Fig. 3) and carefully excised with minimal damage of ulnar nerve. A 1.5-cm cystic mass invading the ulnar nerve was resected (Fig. 4) more than 8 months after the patient first presented with the symptoms, and the nerve was reconstructed by sural nerve grafting (Fig. 5). Neurolysis was performed carefully under $\times 3.5$ loupe magnification, and a ganglion with dimensions of $1.5 \times 0.6 \times 0.6 \text{ cm}^3$ was excised. The nerve fascicles were found to be flattened and separated from each other. There was no clear ganglion stalk or origin. The pathologic specimen revealed a benign cyst with a fibrous wall lined by cuboidal synovial cells.

The patient was evaluated 2 weeks postoperatively, with no significant complaints except weakness of his right hand. At 6 months after the surgery, the patient's grip power and muscle creases increased and no numbness or pain was detected. The handgrip strength of his right hand was found to be 49 kg by a dynamometer. Eleven months after the surgery, the patient regained all hand functions and the interosseous muscle strength in the right hand was only minimally weaker than that in the contralateral hand.

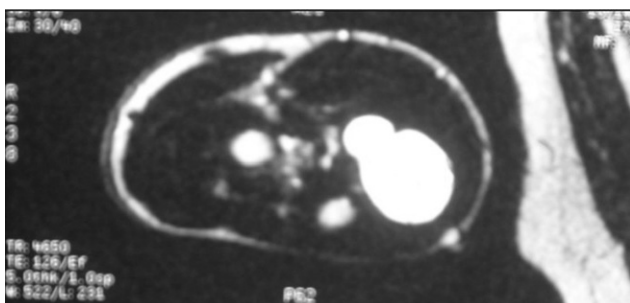


Fig. 2. T2-weighted magnetic resonance imaging showed a mass lesion in the forearm.

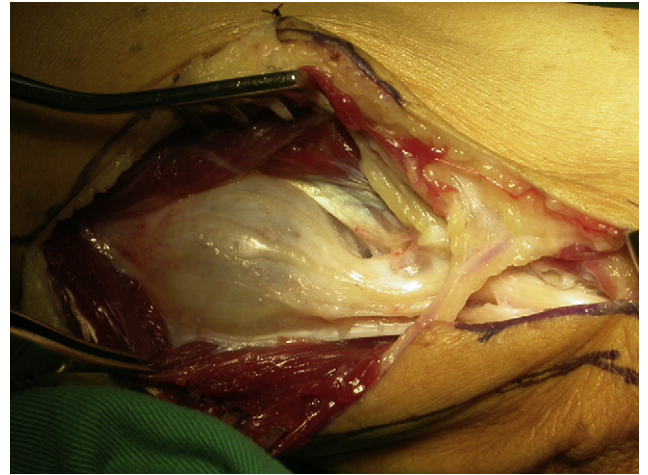


Fig. 3. Intraoperative image shows the multicystic lesion invasion of the ulnar nerve.

3. Discussion

Intraneural ganglion cyst is a rare, benign nerve sheath tumor.⁸ Since the first description of ganglia involving nerves,⁹ most reported cases concern the common peroneal nerve at the head of the fibula.^{3,10–14} It usually affects middle-aged men and presents with symptoms of compression neuropathy. This cystic lesion was first described by Zum-Bush according to Chick and colleagues⁸ in 1895 and subsequently confirmed histologically by Hartwell⁹ in 1901. The most common nerve involved in upper-extremity is the ulnar nerve.^{2–4,6,7,15,16} In addition, other sites have been described including the median nerve,¹⁷ radial nerve,¹⁸ superficial radial nerve,¹⁹ posterior interosseous nerve,²⁰ digital nerve,¹⁸ and brachial plexus.^{21,22}

Here, we presented a rare case of an intraneural ganglion compressing the ulnar nerve in the right forearm. A similar case of ulnar nerve compressed by a 1-cm diameter intraneural ganglion at the retrotrochlear groove was previously reported by Ming Chan and colleagues.⁵ In their case, the patient had symptoms for 3 months without muscle atrophies, and the patient achieved almost complete electrophysiological and clinical recovery after the excision of the ganglion. In our case, the patient developed grip power weakness and atrophy of intrinsic muscles because he experienced



Fig. 4. The resected cystic mass measured approximately $1.5 \times 0.6 \times 0.6 \text{ cm}^3$.

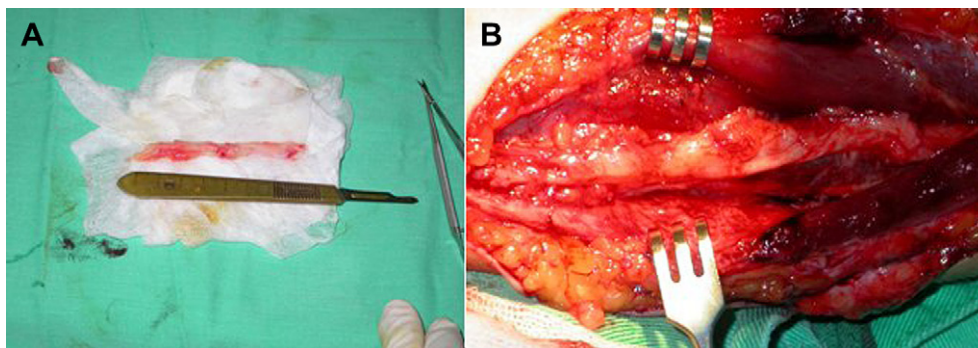


Fig. 5. (A) A sural nerve graft of approximately 7.0 cm was prepared. (B) Nerve grafting using the sural nerve.

the symptoms for a longer duration. Chan and others⁵ concluded that the outcome of surgery to remove intraneural ganglia largely depended on the duration and extent of cystic destruction, and that early recognition and prompt intervention is usually associated with good outcome. Allieu and Cenac encountered four cases of intraneural ganglion cyst involving the upper extremity and reported no correlation of surgical outcome in relation to age, signs or symptoms, or diagnostic delay in all the cases. In our literature review of intraneural ganglia in the upper extremity, we found that the time from symptomatic presentation to surgery for complete to near-complete recovery ranged from as short as 4 weeks to more than 1 year.^{5–7,16,18,20} Patients who were symptomatic for 8 months and 2 years have been reported to have incomplete or moderate recovery.^{3,16} In the two cases of little to no recovery after complete excision, one patient was symptomatic for only 2 months¹⁶ and the other for 6 months.² It was difficult to compare the extent of cystic destruction in each of these cases because not all the authors had provided detailed descriptions of the intraoperative findings.

In this case, the patient was symptomatic for more than 8 months before surgical intervention; despite this, he achieved nearly complete recovery of motor and sensory function after the operation. Since ganglia are not a common cause of ulnar nerve neuropathy, they are frequently misdiagnosed because imaging studies are not always performed preoperatively.²³ Although computed tomography (CT) and ultrasound can also be useful for imaging of the cyst, MRI provides the most detailed image of the cyst and its relation to adjacent structures.^{6,24–28} However, detailed history, clinical examination, and electrophysiologic studies are crucial to the early diagnosis and evaluation of this problem. The symptoms involve pain, numbness, sensory impairment in the medial border of the forearm and hand, reduced grip strength, and wasting of the intrinsic hand muscles.²³ Total excision of the epineural ganglion may not always be possible without damaging the nerve, especially if the ganglion is intraneural.²³

In summary, we presented the case of a patient with intraneural ganglion cyst in the ulnar nerve at the level of the forearm with a significant delay in the diagnosis. Despite chronic motor weakness and muscle atrophy, the patient achieved significant recovery of muscle function after excision of the ganglion and nerve grafting with sural nerve. This is contrary to our experience with weakness and atrophy following traumatic ulnar nerve injury, and this case report will raise awareness among surgeons regarding the differential diagnosis of intrinsic atrophy and timing of treatment.

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