Case report: intraneural ganglion cyst of the ulnar nerve at the wrist

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Published online: 1 March 2011
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Abstract We report a case of a 69-year-old male who presented with pain, weakness, and clumsiness of his right hand. Initial evaluation suggested possible neoplastic process affecting his cervical spine, which was fortunately ruled out by bone biopsy. Subsequent electrodiagnostic studies and magnetic resonance imaging confirmed a lesion of the deep ulnar motor branch. Exploration of Guyon's canal was performed, and an intraneural ganglion involving the deep motor branch of the ulnar nerve was found and excised. Despite more than 14 months of symptomatic duration, the patient made a near-complete recovery with virtually no functional limitations. This provides supporting evidence for a functional benefit of intraneural ganglion excision and nerve decompression even in cases of chronic muscle atrophy.

Keywords Intraneural ganglion · Intraneural cyst · Ganglion cyst · Muroid cyst · Peripheral nerve cyst

Introduction

Intraneural ganglion cysts are benign, mucinous cysts occurring within the epineurium of nerves. They have also been called intraneural mucoid cyst or intraneural mucoid pseudocyst. They can result in pain, paresthesia, and neurological deficit of the involved nerve. Most commonly, these cysts occur within the common peroneal nerve, at the neck of the fibula [6], with only a few reported cases of ulnar nerve involvement in the English literature [2, 5, 9, 10,

13, 15]. We report a case of an intraneural ganglion cyst of the ulnar nerve at the level of the wrist with a significant delay in diagnosis. Despite chronic motor weakness and muscle atrophy, the patient achieved significant recovery of muscle function following excision of the ganglion and nerve decompression. This is contrary to our experience with weakness and atrophy following traumatic ulnar nerve injury and should raise awareness among hand surgeons regarding the differential diagnosis of intrinsic atrophy and timing of treatment.

Case Presentation

A healthy, physically active, 69-year-old, right-hand-dominant, white male presented to our clinic with complaints of chronic weakness of his right upper extremity. He described the onset of acute, deep, achy right hand and forearm pain and right hand weakness 8 months prior to presentation after an episode of heavy physical activity splitting wood. His pain had mostly resolved, but his hand clumsiness and weakness had persisted such that he continued to have trouble shaving. He had been previously evaluated by a nerve conduction study, which suggested carpal tunnel syndrome as well as “chronic” ulnar neuropathy. He denied any nighttime paresthesias or numbness of any kind. No significant medical problems were reported except for having broken both his legs after falling off a ladder a few years earlier.

On examination of the patient's right upper extremity, he had no evidence of traumatic injury and no significant bone or joint deformity. He had normal light touch sensation in all upper extremity cutaneous nerve territories. His motor strength was strong throughout both upper extremities, except the ulnar intrinsic function of his right hand. Finger

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abduction and adduction were minimal, and he had obvious evidence of interosseous muscle atrophy of his right hand (Fig. 1). His right hand tested positive for Froment's sign and Wartenberg's sign. He had no intrinsic muscle weakness, atrophy, or pathologic signs at his left hand. Thumb opposition and abduction were strong, and thenar muscles were well preserved bilaterally. He had no Tinel's sign at his hand, wrist, elbow, or brachial plexus. He had no provocative signs of nerve compression at his elbow or wrist. He did, however, have a positive Spurling's sign on the right and lower cervical spine tenderness.

Cervical spine plain films and MRI were obtained and revealed a possible neoplastic process of his cervical vertebral bodies concerning for multiple myeloma. He then underwent a bone scan that showed increased focal uptake in the medial left clavicle which necessitated a bone biopsy that showed no evidence of malignancy. An electromyogram was performed, with findings consistent with ulnar neuropathy at the wrist affecting the deep motor branch. Fibrillations and positive sharp waves in the first dorsal interosseous and abductor digiti minimi muscles suggested axonal injury, and absent motor unit action potentials with no recruitment suggested high-grade nerve injury. There was no other significant motor finding and no significant sensory conduction abnormality. This suggested a focal lesion at the deep motor branch of the ulnar nerve, and an MRI of the right hand (Fig. 2) revealed a very faint abnormality in zone 2 of Guyon's canal. Ulnar nerve exploration and decompression were recommended though significant recovery of motor function was not expected given the apparent severity of nerve injury and the elapsed time since onset of weakness and atrophy. The patient underwent right ulnar nerve exploration and decompression

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

Fig. 2 MRI image showing abnormality (arrow) in zone 2 of Guyon's canal. P pisiform

at Guyon's canal with excision of an intraneural ganglion from the deep motor branch more than 14 months after his initial presentation of symptoms. The ganglion was identified just deep and distal to the origin of the hypothenar muscles, which were released (Fig. 3). Careful internal neurolysis was performed under $\times 3.5$ loupe magnification, and a 1.2-cm ganglion was excised. The ulnar nerve grouped fascicular structure was in continuity without significant scar. There was no clear ganglion stalk or origin. Surgical pathology was consistent with a ganglion.

Fig. 3 Intraoperative image with the elevator identifying the ganglion within the deep motor branch of the ulnar nerve

The patient was evaluated 1 and 2 weeks postoperatively, with no significant subjective changes noted in regard to his weakness. At 4 months postsurgery, the patient had some clinical recovery of his ulnar nerve intrinsic muscle function and stated that his function was significantly improved from preoperative function. He continued to have a Froment's sign, but had virtually no functional limitations. Ten months after surgery, the patient had regained all previous hand functions, with a very faint hint of a Froment's sign, and interosseous muscle strength only minimally weaker than at his contralateral hand.

Discussion

A number of authors have cited Hartwell (1901) [7] as being the first to report an intraneural ganglion cyst [10, 12, 15], but Spinner et al. [11] sought and successfully retrieved a specimen that confirmed the first reported case dated back to 1810 with the first description, an ulnar ("cubital") intraneural cyst, being credited to Beauchêne of France. Upon close investigation of the specimen, Spinner found the cyst to be mucin filled, creating a fusiform cystic enlargement of the nerve with similar enlargement of an articular branch at the level of the elbow. They believed this to further confirm the applicability of the "unified articular (synovial) theory" in explaining the pathogenesis of intraneural ganglion cyst formation.

Three major theories have been proposed to explain intraneural ganglion cyst formation: (1) degenerative theory, (2) synovial (articular) theory, and (3) tumoral theory [12]. The degenerative theory speculates that mucoid degeneration of the epineurium or perineurium leads to de novo cyst formation within the nerve sheath. The synovial (articular) theory proposes that "pedicles" connect the synovial joint to the affected parent nerve by way of cystic fluid "infiltration" along the articular branch. The tumoral theory, least favored of the three, compares an intraneural ganglion to other cystic lesions or tumors and hypothesizes that even an organized hematoma can transform into an intraneural ganglion cyst. Recent advances in imaging, histological investigation, and mathematical analysis [3] point to the unifying synovial (articular) theory as being the most logical and applicable explanation of intraneural ganglion origin.

Zielinski [15] reported that the intraneural ganglion found through intraoperative exposure in their reported case originated from the carpus and grew into the muscular branch and then into the ulnar nerve through bifurcation. All of the cited reports of intraneural ganglion of the ulnar nerve at the level of the wrist [1, 5, 9, 10, 15] involved the deep branch of the ulnar nerve. Our presented case involved a similar ganglion, found in the deep motor branch of the ulnar nerve, just below the level of the hypothenar muscles. We

found no communication between the ganglion and any extraneural synovial space. Yet in this location, two synovial spaces exist in close proximity: wrist joints and flexor tendon sheaths. In our opinion, it is reasonable to assume either of these to be potential sources for intraneural ganglions.

In accordance with the above-mentioned unifying synovial (articular) theory, it has been suggested that decompression combined with disconnection of the articular nerve branch at or near the joint of origin can eliminate recurrence [14]. In the present case, we found no clear joint of origin, thus a formal exploration of surrounding joints was not carried out on this patient. However, further investigation into the benefits of arthroscopy or arthrotomy may be warranted.

Ming Chan et al. [10] concluded that the outcome of surgery to remove intraneural ganglions 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 [1] reported no correlation of surgical outcome in relation to age, signs or symptoms, or to diagnostic delay in their four cases of intraneural ganglion cyst involving the upper extremity. In our literature review of upper extremity intraneural ganglions, we have found that the time from symptomatic presentation to surgery for complete to near-complete recovery ranged from as little as 4 weeks to more than 1 year [1, 4, 8, 10, 13, 15]. Incomplete or moderate recovery was found in patients symptomatic for 8 months and 2 years [1, 5]. In the two cases of little to no recovery after complete excision, one patient was symptomatic for only 2 months [1] and the other for 6 months [2]. It was difficult to compare how extensive the cystic destruction was in each of these cases due to the fact that not all authors provided detailed descriptions of intraoperative findings.

In our present case, the patient was symptomatic for more than 14 months before surgical intervention, and he was able to make a nearly full recovery. Our experience with muscle recovery following traumatic ulnar nerve injury and repair has been much less impressive than in the present patient. Multiple factors may explain this apparent discrepancy. The present patient's outcome may be extraordinarily relative to other patients with similar lesions. Though his age is advanced, he is very fit and a physically active nonsmoker, and thus may have a youthful potential for nerve healing. The severity of his nerve injury may not have been significant until the few weeks or months prior to surgery. Though his early atrophy would argue against this, combined with the very distal level of nerve involvement and consequent close proximity to motor end plates, his actual time period of denervation may have been much briefer than suggested. Also, his degree of functional recovery and apparent return of muscle bulk may not reflect a similar quantity of nerve or muscle fiber recovery; he may perceive normal function with a

significantly reduced number of neuromuscular functional units. Finally, the ganglion may have fluctuated in size, providing time periods of little or no nerve injury that may have allowed the nerve to intermittently recover. Any of these factors may have had the effect of decreasing the degree of nerve injury and resultant period of muscle denervation, perhaps preserving motor end plates despite evident muscle fiber atrophy and allowing for improved recovery. Nevertheless, a strict reading of his symptoms, clinical exam findings, and electromyogram results would suggest chronic high-grade nerve injury with denervation atrophy. His degree of muscle recovery suggests to us that a delay in diagnosis and surgery does not always result in poor outcomes.

In summary, we report a rare case of an intraneural ganglion of the ulnar nerve at the level of the wrist, with only a handful of cases in the English literature previously reported. Though our patient had a prolonged symptomatic period prior to surgical intervention, and despite findings suggestive of severe nerve injury, he was able to regain near full muscle strength, with no signs of recurrent weakness or recurrent ganglion 10 months postoperatively. The literature on intraneural ganglions is limited to case reports, thus making it difficult for surgeons to predict outcome of surgical treatment. The outcome reported here suggests that a potential surgical benefit exists even in cases of apparent chronic or severe denervation atrophy, and that nerve decompression and ganglion excision should be considered.

Conflicts of Interest The authors declare that they have no conflicts of interest.

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