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CASE REPORT

Posterior interosseous nerve palsy secondary to pigmented villonodular synovitis of the elbow: Case report and review of literature

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KEYWORDS

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Summary Local tumor compression is the main mechanical cause of posterior interosseous nerve (PIN) palsy. The reported cases of these tumors do not include that of pigmented villonodular synovitis (PVNS). Here, we report a case of a 53-year-old male with a 9-year history of painless swelling in his left elbow and a few months of progressive weakness in his left hand. Imaging identified the mass, and histological examination of the biopsy specimens revealed PVNS. The mass was compressing the nerve at the arcade of Frohse, and we performed a complete resection of the mass. Following removal of the mass, the patient regained complete function in his left upper extremity, and no local recurrence has been detected after 2 postoperative years. The possibility of PVNS should be considered in the differential diagnosis of PIN palsy.

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Introduction

Posterior interosseous nerve (PIN) palsy can occur secondary to both mechanical and nonmechanical events. The former include local tumor compression, trauma, and iatrogenic injuries, while the latter include rheumatoid vasculitis and drug toxicity [1]. In particular, quite a few reports exist on local tumor compression. While the type of lesion varies

widely, no previous studies have described a case of PIN palsy caused by pigmented villonodular synovitis (PVNS), a rare, benign, proliferative disorder that affects synovial tissue, bursae, and tendons [2]. PVNS is rarely found in the elbow [3]. The present study is the first reported case of PIN palsy caused by PVNS of the elbow.

Case report

A 53-year-old man presented with a 9-year history of painless swelling in his left elbow. During the 4 months prior to presentation, he noted pain in the medial aspect of his

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Figure 1 Plain lateral radiograph of the elbow showing a soft tissue swelling extending from the antecubital fossa (fossa cubiti) to the proximal forearm (black arrowheads). There are no changes noted in the adjacent bones or joint space.

elbow, along with a 2-month history of progressive weakness of his left hand. There was no history of rheumatoid arthritis or trauma to the neck or left upper extremity. Physical examination revealed a non-tender mass in his left elbow. He was unable to extend his fingers at the metacarpophalangeal joint (MMT 1/5), but extension of his interphalangeal joints and wrist was normal. Full passive ranges of motion in each digit were preserved and no sensory disturbance was observed.

Plain radiographs of the elbow showed a soft tissue mass with no calcification, extending from the antecubital fossa to the proximal forearm (Fig. 1). Computed tomography (CT) of the left elbow showed a mass measuring 10cm compressed by the proximal end of the supinator muscle (Fig. 2). T1- and T2-weighted magnetic resonance images (MRI) revealed hyperintense signal in the muscle adjacent to the mass as well as several foci of low intensity. After administration of gadolinium-diethylenetriaminepentaacetic acid (Gd-DTPA) contrast, the



Figure 2 Sagittal CT scan of the elbow showing a mass compressed by the proximal portion of the supinator muscle (black arrowheads).

mass was observed to be heterogeneously enhancing (Fig. 3). Nerve conduction studies/Electromyography (EMG) confirmed PIN palsy with denervation of the muscles supplied by the PIN.

Microscopic examination of biopsy specimens obtained by open biopsy revealed numerous mononuclear cells, multinuclear giant cells, foam cells, and hemosiderin accumulation (Fig. 4). The histological diagnosis was PVNS and the mass appeared to compress the PIN.

Marginal excision of the mass was performed through a longitudinal medial approach. The mass was inferior to the brachioradialis muscle and superior to the deep head of the supinator muscle, causing compression of the PIN against the fibrous edge of the proximal portion of the supinator muscle (arcade of Frohse) (Fig. 5A). The mass was also compressed by the arcade of Frohse (Fig. 5B).

We performed no postoperative complementary therapy. The patient's PIN palsy improved without any rehabilitation following removal of the mass, and he had complete restoration of normal function in his left upper extremity

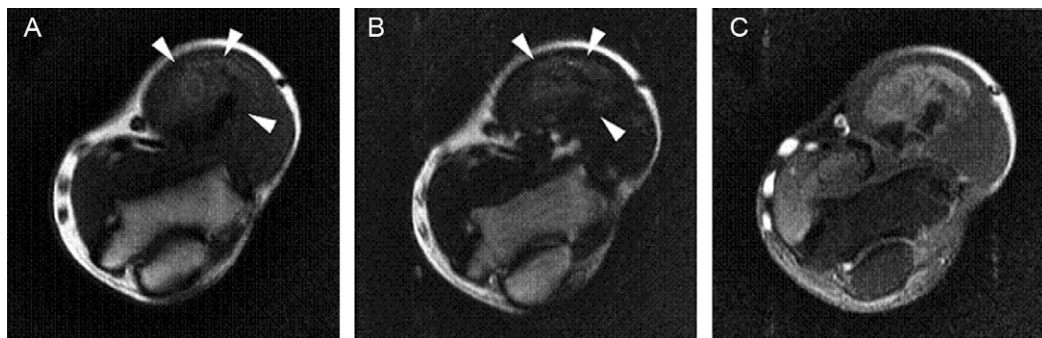


Figure 3 T1- (A) and T2-weighted axial MRI (B) showing a hyperintense mass (white arrowheads). The mass was heterogeneously enhancing after administration of gadolinium-diethylenetriaminepentaacetic acid (Gd-DTPA) contrast and fat suppression (C).

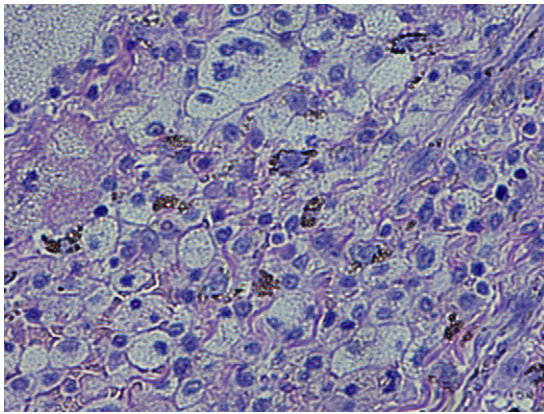


Figure 4 Photomicrograph showing mononuclear cells, polynuclear giant cells, foam cells, and hemosiderin accumulation.

3 months postoperatively. Electromyography taken 4 months postoperatively showed recovery of normal nerve conduction velocity. The Enneking functional score for the upper arm was 30/30 (100%). No local recurrence was detected 2 years postoperatively.

Discussion

The PIN may be affected by various lesions in the forearm including traumatic lesions, compressive lesions in the radial tunnel, tumors, vasculitis, and miscellaneous lesions [4]. Quignon et al. reported tumor compression as the most frequently reported etiology of PIN palsy, at a rate of 39.4%, followed by muscle aponeurosis at 27.3%, and trauma at 13.6% [5].

The radial nerve divides into the PIN and the superficial radial nerve. The PIN supplies the extensor carpi radialis brevis and supinator muscles and passes between the superficial and deep layers of the supinator muscle along with the radial recurrent artery. The most superior portion of the superficial layer of the supinator muscle forms an arch known as the arcade of Frohse (Fig. 6). The PIN is particularly vulnerable in this region [6]. In the present case, compression was caused by a tumor in this region. This tumor was identified by CT imaging (Fig. 2) and histological examination of biopsy specimens (Fig. 4).

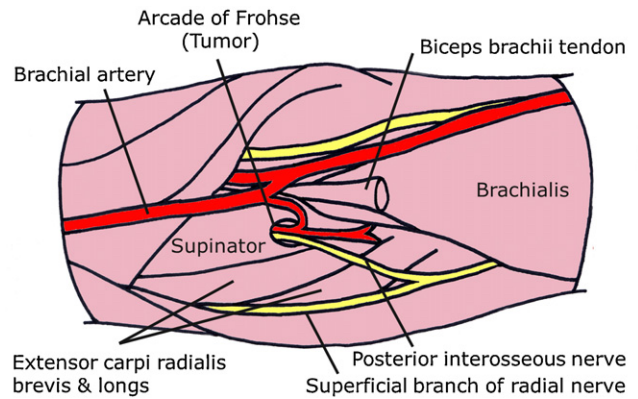


Figure 6 Anatomical diagram shows the relationship between the posterior interosseous nerve (PIN) and other structures. The PIN and the radial recurrent artery pass through the arcade of Frohse.

The PIN is primarily a motor nerve, and the superficial radial nerve is anatomically distinct from the PIN. As such, sensory disturbance usually does not accompany PIN palsy. However, the PIN gives off terminal sensory branches to the ligaments and articulations of the carpal joints as well as sensory innervation to the periosteum of the radius and interosseous membrane of the forearm. Entrapment of a motor nerve may cause a diffusely localized, dull, aching pain [7]. Therefore, patients with PIN compression by a soft tissue mass typically present with gradual onset of symptoms including deep forearm and elbow pain that appears prior to the development of weakness of the extensor muscles of the fingers without sensory loss. These findings are consistent with those in this case. In cases of nerve compression combined with other localized pain, EMG studies are considered valuable [8]. In addition, as the superficial radial nerve passes above the supinator muscle, it can be compressed by the mass itself and paraesthesia can occur [9]. Notably, we did not observe this in the present case.

A number of reports exist regarding PIN compression by tumors such as lipomas, ganglion cysts, fibromas, schwannoma, and synovial haemangiomas [10], but to our knowledge, no one has reported PIN palsy caused by PVNS. As the first to do so, we recommend that PVNS is considered in the differential diagnosis as a potential etiology of PIN palsy.

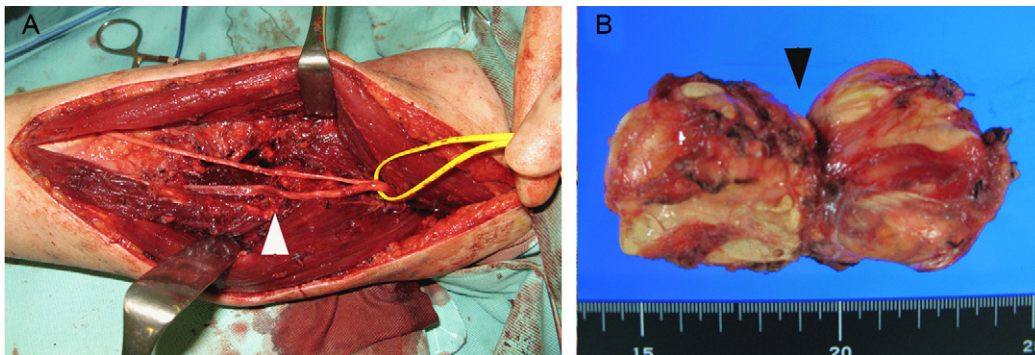


Figure 5 A. The tumor, which was located deep to the posterior interosseous nerve (white arrowhead), was resected and the nerve was released. B. The resected specimen, showing a narrowed region (black arrowhead) in the middle of the tumor.

Jaffe et al. identified PVNS of differing morphologies, noting that these represented variants of the same proliferative disorder of the synovium [2]. PVNS can be locally aggressive, but does not metastasize. It most often occurs in patients between the ages of 30 and 50 years and there is no gender bias [11]. The most commonly affected joint is the knee, followed by the ankle and hip. Involvement of the elbow is rare [3,12,13]. Ottaviani et al. reported that the incidence of PVNS of the elbow is 0.82% (1/122 cases) [13]. Neural involvement of PVNS is also rare and only three cases have been reported in the literature [11,14,15]. In these three cases, the nerves involved were the radial and ulnar nerves, peroneal nerve, and ulnar and median nerves. Thus, the present study is the first to describe PVNS involving the PIN.

The most common finding on plain radiographs of PVNS is soft-tissue swelling or swelling of the involved joint. Only 10% of cases show bone lesions similar to osteoporosis or cortical erosion of adjacent bones [13,14]. In addition, PVNS usually show high attenuation on CT imaging because of their high hemosiderin content. On T1- and T2-weighted MRI imaging sequences, foci of low signal intensity, related to high hemosiderin content, are observed [7,16]. In the present case, these characteristic findings were observed in the CT and MRI, and no changes in the plain radiographs were noted for the bones or joints.

Treatment of PVNS involves complete resection of all gross disease because of its high rate of recurrence. The recurrence rate after surgical resection is reportedly 8 to 56%, and differs by treatment, follow-up duration, definition of relapse, and outcome evaluation [13]. When PVNS occurs on the tendon sheath, it is relatively easy to resect, but because of the complexity of knee and elbow joint anatomy, complete resection in these joints is often difficult [2,17]. To minimize the risk of recurrence, complementary therapies such as isotopic synoviorthesis, adjuvant cryotherapy or external radiotherapy, have been suggested [12]. However, as their effects have not been proven, complementary therapies were not provided in this case.

In terms of PIN palsy treatment, treatments are divided according to causes. If definite compressive lesions such as PVNS are evident, then surgical decompression of the PIN is recommended as treatment. A review of the literature would reveal that rapid and complete recovery of function is likely to follow decompression, but delay in diagnosis and treatment may lead to poor outcomes [18]. In cases of PIN palsy caused by vasculitis, pharmaceutical approaches are more effective than surgery [1,4,19]. Moreover, no correlation seems to exist between the length of preoperative symptoms and the recovery time [1]. This case involved a definite compression lesion, and there was a long delay before the patient presented to our hospital. However, because tumor growth was slow and symptom onset was gradual, complete resolution of symptoms was achieved by tumor resection and nerve decompression.

Conclusion

As PVNS occurring in the elbow can compress the PIN, it should be considered in the differential diagnosis as a potential etiology of PIN palsy.

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

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