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Case Report

Pisiform–Hamate Coalition With Entrapment Neuropathy of the Deep Palmar Branch of the Ulnar Nerve

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Pisiform and hamate coalition, a rare congenital anomaly, is most often identified as an asymptomatic incidental finding on radiographs. Some patients have symptomatic coalition with ulnar-sided wrist pain or ulnar nerve neuropathy from ulnar nerve compression. Sports activities such as cycling and weight lifting can cause compression of the ulnar nerve in the hand. This report describes a case of a pisiform and hamate coalition in a 36-year-old man who reported decreased right-hand dexterity and right ulnar-sided wrist pain. The patient, an amateur weight lifter, developed right claw hand through repeated bench press training. Intraoperative findings revealed compression of the deep palmar branch of the ulnar nerve between the tendinous arch of the hypothenar muscles and pisiform and hamate coalition. Surgical resection of the tendinous arch and the enlarged hook of hamate relieved the claw hand deformity.

Carpal coalition is a rare congenital anomaly that is mostly identified as an asymptomatic incidental finding on radiographs. Carpal coalition can be of 2 forms: an osseous coalition in which the carpals are united by an osseous block, or a nonosseous coalition by which the carpals are united by cartilage or fibrous tissue.[1] The most common coalition is the lunate–triquetrum coalition.[2,3] Pisiform–hamate coalition is a rare coalition that was first reported by Cockshott.[4] Few reported cases have included symptomatic coalition with ulnar-sided wrist pain.[5,6] Only one article in the literature described ulnar nerve neuropathy with ulnar nerve compression by a pisiform–hamate coalition.[7] Sports activities such as cycling and weight lifting reportedly cause compression of the ulnar nerve in the hand.[8] This report describes a case of pisiform–hamate coalition in a 36-year-old man with ulnar nerve neuropathy triggered by repetitive bench press training. To our knowledge, this report is the first to describe a pure motor deficit of ulnar nerve with pisiform–hamate coalition.

Case Report

A 36-year-old man was referred to our hospital because of decreased dexterity of the right hand and right ulnar-sided wrist pain. The patient had a history of hyperuricemia and hyperlipidemia, particularly fatty liver. He had no upper-extremity trauma or surgery. He had started a yearlong weight loss program of weight training with bench presses of 100 kg (220 lb). Physical examination revealed no cubitus varus or valgus deformity in the left elbow. Tinel test results were negative at the cubital tunnel and Guyon tunnel. He showed no disturbance of sensation on either the left forearm or left hand. Semmes–Weinstein monofilament test revealed no diminished sensation. The right hand showed mild clawing of the ring and little fingers, and atrophy of the dorsal and palmar interosseous muscles was found (Fig. 1). Grip and key pinch strength of the affected hand was 30 and 6.0 kg, respectively, whereas that of the unaffected hand was 40 and 9.0 kg, respectively. Froment sign was positive on the right hand. Anteroposterior radiographs of the right hand (Fig. 2) and computed tomography (CT) revealed an unusual hamate shape compared with the unaffected side on the sagittal reconstruction view: a large hook of hamate and pisiform (Fig. 3). We observed a type 1 coalition according to Minaar's classification,[2] which was characterized by an incomplete fusion resembling pseudarthrosis. Ultrasonographic examination (Fig. 4) indicated no space-occupying lesion. A nerve

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Figure 1. Right hand showing mild clawing deformity of the fourth and fifth fingers with atrophy of the dorsal interosseous muscles.

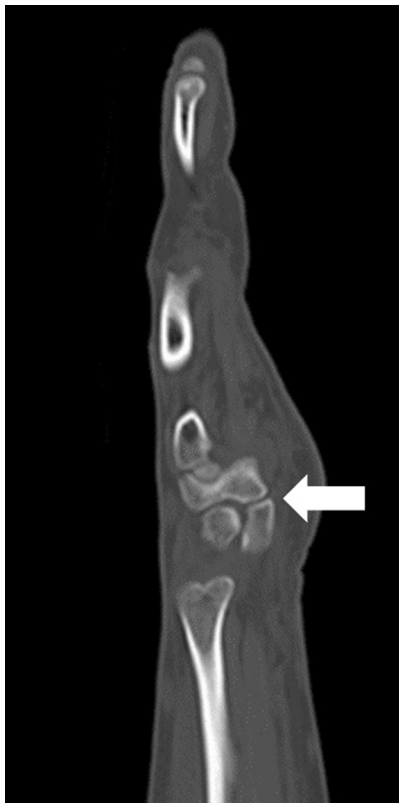

Figure 3. Sagittal reconstruction CT of the right wrist showing a type 1 pisiform–hamate coalition.

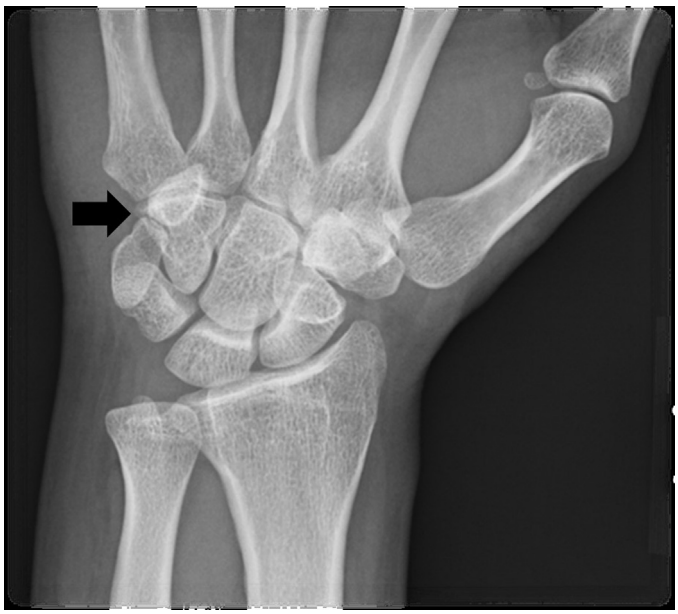

Figure 2. Anteroposterior plain radiograph of the right wrist showing unusual shape as a large hook of hamate and pisiform.

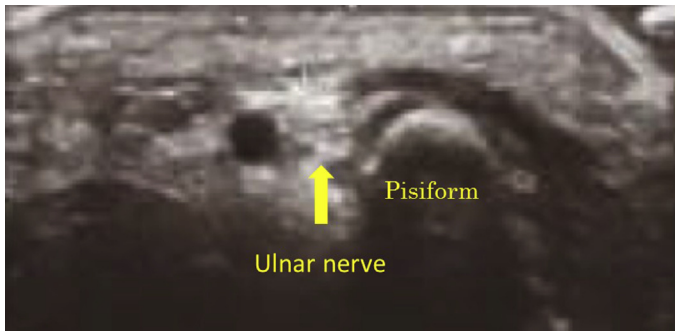

Figure 4. Axial view of ultrasonography showing no space-occupying lesions.

conduction examination performed at the initial visit indicated normal amplitude of the right first dorsal interosseous muscle motor nerve conduction velocity of the ulnar nerve and normal sensory nerve conduction velocity on the affected side. However, 4 months later, the amplitude of the right first dorsal interosseous muscle motor nerve conduction velocity of the ulnar nerve had declined. We speculated that repeated compression of the palm during weight training might have caused neuropathy of the deep palmar branch of the ulnar nerve. We advised the patient not to perform bench press exercises. We observed the motor deficit for 10 months with no improvement. For this reason, surgical exploration was performed. Under general anesthesia, Guyon canal was opened and the deep palmar branch of the ulnar nerve was released. The deep branch of the ulnar nerve was compressed between the tendinous arch of the hypothenar muscles from the volar side and the hypertrophied hook of hamate from the dorsal side (Fig. 5A, 5B). We resected the tendinous arch of the hypothenar muscles and the hypertrophied hook of hamate (Fig. 5A). The deep branch was released completely. One week after surgery, the mild claw hand had recovered (Fig. 6). At 10 months after the operation, grip strength and key pinch strength in the affected hand was 39 and 8.0 kg, respectively, compared with 41 and 10 kg, respectively,

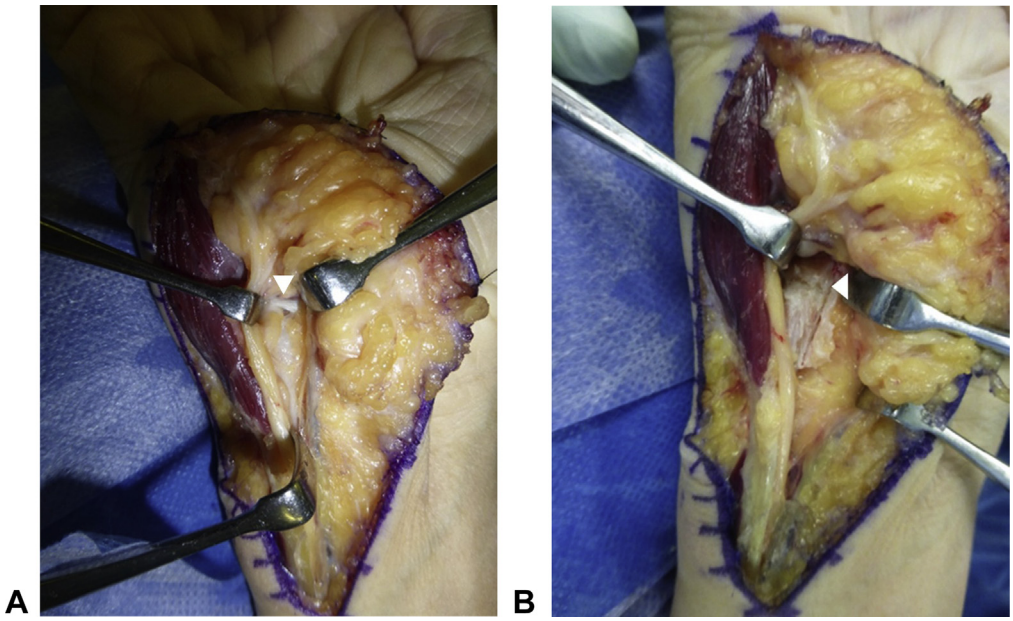

Figure 5. **A** Deep branch of the ulnar nerve compressed from the dorsal side by the tendinous arch of the hypothenar muscles (arrowhead). **B** Deep branch compressed from the dorsal side by the hypertrophied hook of hamate (arrowhead).

Figure 6. Right hand at 1 week after surgery showing recovery of the mild claw hand.

on the unaffected side. Electrophysiological examination revealed recovery of the amplitude of the motor nerve conduction velocity of the ulnar nerve of the right dorsal first interosseous muscles.

Discussion

The patient in this case report presented with ulnar-sided wrist pain and pure motor neuropathy attributable to a lesion of the deep branch of the ulnar nerve with mild clawing of the ring and little fingers. As far as we know, this description is the first in the literature for a pure motor deficit with pisiform–hamate coalition. Classification of the level of the lesion in ulnar neuropathies of the hand presented by Wu et al[9] demonstrated that the patient in this report had a pure motor disturbance, type 3 ulnar nerve neuropathy, as indicated by a lesion of the deep branch of the ulnar nerve just distal to the superficial branch but proximal to the branch to the hypothenars. For this patient, we speculate that the ulnar-side wrist pain derived from neuropathic pain. We identified the motor nerve disturbance as probably resulting from entrapment of the deep branch of the ulnar nerve. Krivickas and Wilbourn[8] reported that sports activities such as weight lifting, and specifically bench press lifting, cause entrapment neuropathy as carpal tunnel syndrome and ulnar nerve neuropathy. In this case, we suspected that repetitive compression to the hand involved in bench press lifting contributed to the development of compression of the deep palmar branch of the ulnar nerve. Preoperative CT provided information related to pisiform–hamate coalition that was not initially recognized by plain radiographs. In fact, pisiform–hamate coalition is a rare coalition. An incidence of 0.11% to 0.76%[1] is estimated for this rare coalition, which is inferred as resulting from metaplasia of the pisohamate ligament into bone.[1] The patient in this case report presented the coalition categorized as type 1 by Minaar's classification: incomplete fusion resembling pseudoarthrosis. The classification was developed by Minaar to classify lunotriquetral fusions based on radiographs.[2] Berkowitz et al[7] demonstrated 2 cases of pisiform–hamate coalition with mixed motor and sensory neuropathy of ulnar nerve. Two cases were type 1 coalition, which involved the floor of Guyon canal and compression of the ulnar nerve. Total excision of the pisiform and partial resection of the articulating portion of the hook of hamate decompressed

the ulnar nerve and relieved the symptoms. In the patient in the current case report, the deep branch of the ulnar nerve was compressed between the tendinous arch of the hypothenar muscles and the hypertrophied hook of hamate. We resected the tendinous arch of the hypothenar muscles and the hypertrophied hook of hamate, rather than the pisiform, and decompressed the deep branch of the ulnar nerve completely. We believe that surgical procedures used for this coalition with ulnar nerve neuropathy should be chosen depending on the characteristics of the patient's pathophysiology. Awareness of this pathophysiology can raise confidence in the proper treatment and surgical management of this rare condition, whereas delayed diagnosis and treatment may lead to functional damage.[10] Indeed, surgical resection of the hook of hamate and the tendinous arch resolved the pure motor neuropathy of the deep branch of the ulnar nerve. Surgeons should be aware that repeated compression motion to the palm such as that inherent in excessive weight training can cause ulnar nerve neuropathy in patients with pisiform–hamate coalition.

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