Dupuytren’s Disease Involving the Wrist

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There is a well-established association with ectopic fibrosis in Dupuytren’s disease, but involvement of the wrist is rare. To our knowledge there have been only 5 cases reported in the literature. The authors present a patient with Dupuytren’s disease involving only the proximal palm and wrist who presented with functional limitation and a rapid clinical progression. (J Hand Surg 2007;32A:352–354. Copyright © 2007 by the American Society for Surgery of the Hand.)

Key words: Dupuytren’s disease, ulnar neuropathy, wrist, fibromatosis.

Dupuytren’s disease is a fibroproliferative disorder that is usually isolated to the palm and digits. There is a well-described association with ectopic fibrotic change, which may be observed in the soles of the feet (Ledderhose’s disease), over the dorsum of the proximal interphalangeal joints (Garrod’s knuckle pads), or involving the penis (Peyronie’s disease).1 Involvement of these sites can occur before the disease process is evident within the palm, and the disease is usually associated with a rapid progression and poor prognosis.1

Dupuytren’s disease involving the wrist is exceedingly rare: to our knowledge there have been only 5 cases reported in the literature.2–5 We present a case of Dupuytren’s disease isolated to the proximal palm and volar wrist, presenting with a rapid clinical progression and symptoms of ulnar neuropathy.

Case Report

A 36-year-old right-hand–dominant man was referred to the Combined Hand Service outpatient clinic with a 2-year history of a steadily enlarging mass of the right volar forearm that limited wrist extension. The patient was of mixed Northern and Central European ethnicity and had no known family history of Dupuytren’s disease. For the preceding 2 weeks the patient had experienced worsening pain and paresthesias in the right ring and small fingers. The symptoms were intermittent and more severe at night, and the patient reported occasionally being awakened from sleep.

On examination there was a firm mass of the ulnar aspect of the right volar forearm, wrist, and proximal palm involving the subcutaneous soft tissues and skin (Fig. 1). The mass was ill-defined, nontender, and fixed to the flexor carpi ulnaris tendon. The active range of wrist motion was from 15° of extension to 85° of flexion. There was no measurable sensory deficit in the fingertips by 2-point discrimination testing, but the patient reported paresthesias in the ring and small fingers, and a Tinel’s sign was elicited at the wrist. Dorsal hand sensibility was normal, and there was no tenderness or Tinel’s sign at the cubital tunnel.

A magnetic resonance image was obtained that showed an infiltrative soft-tissue mass that measured 4 cm in maximum transverse extent and surrounded and invaded the flexor carpi ulnaris (FCU) and palmaris longus tendons. The mass abutted the ulnar nerve on several sections but did not appear to invade it. The interpreting radiologist believed the appearance of the mass was suggestive of a fibroplastic sarcoma.

An incisional biopsy was performed under local anesthesia. Histologic examination of the specimen showed only fibrosis, but the diagnosis of sarcoma could not be definitively excluded. The mass was excised surgically en bloc with the FCU tendon and the surrounding skin (Fig. 1). The deep fascia of the forearm was involved with the disease process and was resected. The ulnar nerve was mildly hyperemic but morphologically normal in appearance.

Histologic evaluation was performed of the central portion of the mass, which was in continuity with the superficial palmar fascia, as well as involved portions of the deep fascia. This evaluation showed nodular proliferation of spindle-shaped fibroblasts surround-
ing dense collagenous tissue (Fig. 2). Immunohistochemical stains were performed on samples from the central mass, and results were positive for alpha–smooth muscle actin (Fig. 3) and negative for MAC1, cluster of differentiation 24, and S100. These findings supported the diagnosis of Dupuytren’s fibromatosis. The alpha–smooth muscle actin stain was used to confirm the presence of myofibroblasts. MAC1 is a macrophage surface antigen, and this stain was used to distinguish fibroblasts from macrophages. Cluster of differentiation 24 is a cell-surface glycoprotein associated with tumor growth, and S100 is a calcium binding protein expressed by melanocytic and neural tumors; these 2 stains were used to rule out neoplastic tumor growth.6

The postoperative course was uncomplicated, and the patient’s surgical site healed without incident. At 3 months after surgery, he was able to fully flex and extend the wrist. There was no evidence of recurrence, and the ulnar nerve symptoms had completely resolved.

Discussion

The first mention in the literature of Dupuytren’s disease involving the wrist appeared in 1965, in a review article by Hueston7, who reported that the palmaris longus or FCU tendons are occasionally

Figure 1. (A) The patient, a 36-year-old man with a firm mass involving the ulnar aspect of the distal forearm, wrist, and proximal palm, marked for dermatofasciectomy. (B) After en bloc resection of the mass, the overlying skin and FCU tendon, with preservation of the ulnar artery and nerve. (C) Appearance immediately after skin closure.

Figure 2. Histologic section of the mass showing a nodular proliferation of spindle-shaped fibroblasts surrounded by dense collagenous tissue (hematoxylin and eosin stain; magnification, ×80).

Figure 3. Histologic section with immunohistochemical staining for alpha–smooth muscle actin showing an abundance of myofibroblasts (magnification, ×80).
involved with the disease process. No illustrative cases, however, were presented. In 1968, Boyes and Jones presented 2 cases of recurrent Dupuytren’s contracture with primary involvement of the palm with extension into the palm in continuity. Since that time there have been only 3 other cases reported. In 2001, Dalton et al presented the case of a 47-year-old woman with a classic Dupuytren’s contracture of the right palm and isolated fibromatosis of the left wrist. The patient presented later with a classic Dupuytren’s contracture of the left hand.

The present case differs from earlier cases in that the wrist and proximal palm were the only anatomic sites involved and the patient presented with symptoms suggestive of ulnar neuropathy. Neuropathy is not a recognized feature of Dupuytren’s disease, but these symptoms may have arisen because of the unique anatomic vulnerability of the ulnar nerve at the distal forearm and wrist level. In the distal forearm, the ulnar nerve courses dorsal to the deep layer of the deep fascia of the forearm. In the wrist this fascial layer is in continuity with the volar carpal ligament. As it passes through Guyon’s canal, the nerve lies dorsal to the volar carpal ligament and volar to the transverse carpal ligament.8 If these fascial layers of the wrist and forearm are involved with Dupuytren’s disease and become thickened and fibrotic, it follows that the volume of Guyon’s canal may decrease, resulting in nerve compression. In addition, the patient in the present case had limitation of wrist extension, and it is well established that prolonged flexed posture is a predisposing factor for ulnar nerve compression at the wrist.10–12

The surgical treatment of Dupuytren’s disease involving the wrist should, like classic Dupuytren’s disease of the palm, be predicated on the severity of symptoms and physical limitations. In the present case, however, the patient presented with a clinical history and physical examination results suspicious for neoplasm. Because the result of the incisional biopsy was equivocal, an en bloc surgical resection (dermatofasciectomy) was performed. We believe that, in general, surgical treatment should be reserved for patients with sufficient functional limitations and should consist of local fasciectomy. Dupuytren’s disease involving the wrist is rare but must be included in the differential diagnosis of patients presenting with a mass of the volar aspect of the wrist, with or without neurologic symptoms.

Received for publication August 26, 2006; accepted in revised form October 18, 2006.

No benefits in any form have been received or will be received from a commercial party related directly or indirectly to the subject of this article.

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doi:10.1016/j.jhsa.2006.10.008

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