Intraneural Nodular Fasciitis in a Child: A Case Report and Review of the Literature

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We present the case of an 11-year-old boy with intraneural nodular fasciitis of the median nerve at the distal part of the carpal tunnel. Complete excision of the involved median nerve and cable grafting using 4 fascicular segments of the sural nerve was performed for persistent pain and numbness after initial resection of the tumor in piecemeal fashion. Pain and numbness were completely resolved and there was no evidence of recurrence 24 months after the final surgery. *(J Hand Surg Am. 2016;41(9):e299–e302. Copyright © 2016 by the American Society for Surgery of the Hand. All rights reserved.)*

Key words Child, intraneural, nodular fasciitis.

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NODULAR FASCIITIS IS A BENIGN SOFT tissue tumor characterized by proliferation of fibroblasts, and often confused with sarcomas.[1,2] The most common locations are the upper extremities (48%), trunk (20%), head and neck (17%) and lower extremities (15%).[3] The patients range in age from infants to the elderly, with the majority between 20 and 50 years old.[4] Both sexes are equally affected.[1–3] The lesions appear as solitary, well-circumscribed, round to oval, subcutaneous nodules less than 2 cm in diameter.[2]

Nodular fasciitis was originally described in 1955 by Konwaler et al[5] and is synonymous with pseudosarcomatous and infiltrative fasciitis, terms that reflect the lesion's rapid growth, dense cellularity, alarming mitotic activity, and permeative nature. Although only 10% to 15% of patients report a history of preceding trauma, nodular fasciitis is considered to be a benign, nonneoplastic proliferative process.[4]

An intraneural lesion is rare and only 6 cases have been reported. All 6 cases occurred in patients ranging from 32 to 79 years of age.[6–11] We present the case of an 11-year-old boy with intraneural nodular fasciitis of the median nerve.

CASE REPORT

An 11-year-old boy had noticed numbness and pain in the thumb and index finger of the right hand 2 months prior to visiting our clinic. There was no history of trauma and no family history of nodular fasciitis. The initial examination revealed hypoesthesia in the right thumb, index, middle, and ring fingers. There was weakness of opposition, but no atrophy of the thenar muscle. The Tinel sign was present over the distal wrist crease. The Phalen and reverse Phalen tests were negative. Grip strength was 15 kg in the left hand but recorded as 0 kg for the right hand because the patient could not squeeze the dynamometer owing to numbness of the fingers. Key and tip pinch strength were 2.2 kg and 0.2 kg on the right side and 4.4 kg and 1.8 kg on the left side, respectively. The Semmes-Weinstein monofilament test revealed diminished light touch (3.22) in the right thumb and right index and middle fingers, but was normal (1.65) in the little finger. Although symptoms

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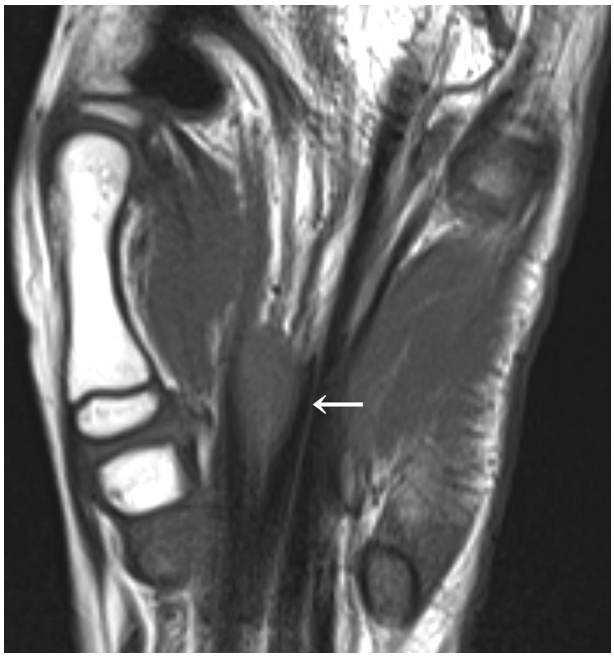

FIGURE 1: A T1-weighted coronal image shows an isointense mass at the center of the palm (arrow).

were suggestive of carpal tunnel syndrome, the patient was only 11 years old, an age at which carpal tunnel syndrome is not commonly observed. A magnetic resonance imaging examination was performed and demonstrated a 14 × 8 × 7-mm lesion at the distal end of the carpal tunnel in the terminal branches of the median nerve. The lesion was isointense on T1-weighted images and hyperintense on T2-weighted images (Fig. 1).

Surgery was performed with the patient under general anesthesia. The median nerve was deformed by a mass of solid white tissue at the ramification into the common digital nerves (Fig. 2). A biopsy was performed of the thickest part of the mass without iatrogenic injury to the median nerve.

Hematoxylin-eosin staining showed findings of nodular fasciitis, with a proliferation of spindle-shaped cells in a myxoid matrix with scattered inflammatory cells and dense collagen fibers. Immunohistochemical analysis revealed that the spindle-shaped cells were positive for smooth muscle actin but negative for desmin, S100, and epithelial membrane antigen (Fig. 3). With a pathological diagnosis of nodular fasciitis, the expected course was for the lesion to reduce spontaneously, and so no further treatment was planned.

Although there was temporary improvement, pain returned 3 months after surgery and was associated with a gradual increase in numbness. Atrophy of the thenar muscle also appeared at 3 months after surgery.

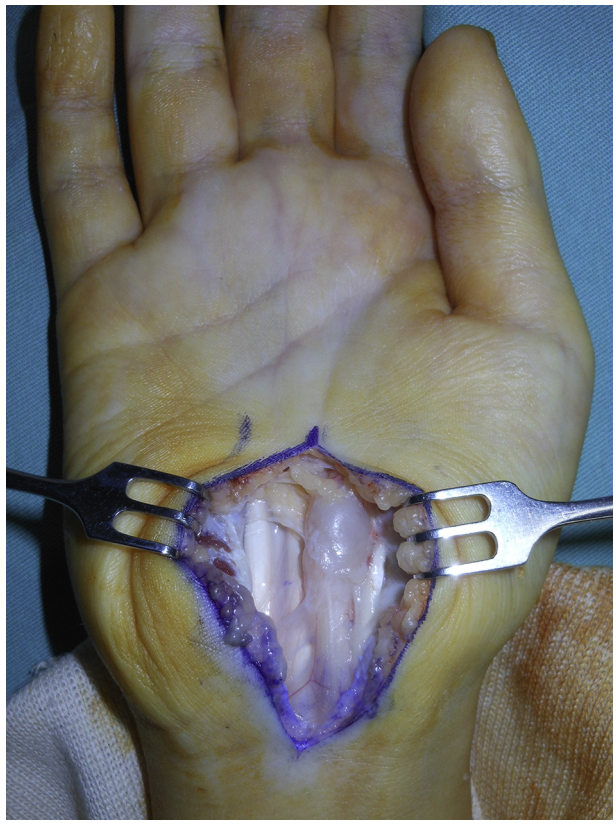

FIGURE 2: The carpal tunnel was released.

A secondary operative procedure was performed 9 months after the initial surgery. The tumor, which was adherent to the surrounding tissue, was resected in a piecemeal fashion under magnification. The median nerve was not resected, but a neurolysis was performed. Pain and numbness persisted after the second procedure and a follow-up magnetic resonance imaging examination 5 months later demonstrated a slight increase in tumor size. The patient and parents were advised to consider additional surgery to completely resect the lesion. Seven months after the second surgery, a third procedure was performed in which the median nerve and the tumor were completely excised. The terminal branches of the median nerve, including the motor branch, were reconstructed with nerve grafting using 4 fascicular segments of the sural nerve (Fig. 4).

Twenty-four months after the final surgery, pain and numbness were completely resolved. However, atrophy of the thenar muscle remained. Grip strength was 21.2 kg for the right hand and 20.6 kg for the left. Key and tip pinch strength were 2.7 kg and 0.9 kg on the right side and 3.8 kg and 2.8 kg on the left side, respectively. The Semmes-Weinstein monofilament test was normal in the right thumb, index, and middle fingers.