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Case Report

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Lipomatosis of dorsal cutaneous branch of ulnar nerve – a rare cause of sensory neuropathy of dorsum of the hand

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ABSTRACT

Wrist pain along the ulnar aspect is a diagnostic dilemma for a long time, this is because of the complex anatomy and numerous pathologies related to these structures. The most common cause is the tear of the triangular fibrocartilage complex. The remaining etiologies are lunotriquetral ligament tears, extensor carpi ulnaris tendon disorders, distal radioulnar joint disorders, impingement and impaction syndromes, pisotriquetral joint disorders, and ulnar wrist masses. Imaging is an important cornerstone in the evaluation of ulnar-sided wrist pain which aids the clinical history and physical examination. X-rays, USG, CT, magnetic resonance imaging (MRI), and MR arthrography are the currently available modalities that are often used to guide diagnosis and treatment. However, while requesting MRI, the need for inclusion of distal forearm is not well understood. In this case report, we describe a rare cause for ulnar-sided wrist pain which was diagnosed accurately with MRI. The inherent properties of MRI and the inclusion of the distal forearm helped us in reaching the final diagnosis. With this case report, we describe the ability of MRI to detect the nature of the lesion which helped to preclude the need for biopsy, hence entitling it as a "do-not touch lesion."

Keywords: Lipomatosis, Fibrolipoma, Ulnar nerve, Dorsal cutaneous branch of ulnar nerve, Magnetic resonance imaging

INTRODUCTION

Lipomatosis of the nerve or fibrolipomatous hamartoma is a benign pathology of peripheral nerves. The pathology is the excessive fat and mature fibroblasts within the neural sheath.

The ulnar nerve gives off a dorsal cutaneous branch at about 5 cm proximal to the wrist joint. It courses posteriorly deep to the flexor carpi ulnaris, passes through the deep fascia, and runs along the ulnar side of the dorsum of the wrist and hand. Here, it bifurcates into two dorsal digital branches. The first branch carries the sensory information from the ulnar side of the little finger, and the second branch is from the adjoining aspects of the little and ring fingers.

USG can be used for an initial evaluation, where it can be used to see thickening and altered echoes of the nerve fibers. However, due to the lack of soft-tissue resolution/when the lesion is relatively small, USG is unable to characterize the lesion. Magnetic resonance imaging (MRI) is considered imaging of choice for neural lipomatosis due to its high soft-tissue resolution and its ability to detect fat.

In this report, MRI findings of a case of lipomatosis of the dorsal cutaneous branch of the ulnar nerve are presented. As for the date, there is no effective treatment for lipomatosis of a nerve

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is available; removal of the whole mass can result in severe sensory and motor deficits that result.

Unnecessary biopsies and surgical interventions may be avoided by knowledge of the unique MR features of this rare pathology.

CASE REPORT

A 44-year-old female computer operator presented with a palpable soft swelling on the posterior aspect of the left hand and wrist for a few years. She also felt pain and burning sensations over posteromedial aspects of the 4th and 5th fingers of the left hand for a few months. There was no prior trauma, and there were no other comorbidities or systemic illnesses. On physical examination, she had an elongated, soft, and mildly tender swelling on the dorsum of the ulnar aspect of the wrist and hand. She had undergone USG which showed a fusiform iso-hypoechoic swelling without increased vascularity. MRI wrist joint was requested for further evaluation.

Interpretation

Non-contrast MRI scan of the left wrist and hand was performed using 1.5 Tesla systems. Axial T1, T2, fat-suppressed T2 (T2FS), coronal T1, T2FS, and sagittal T2FS images were obtained.

An elongated tubular structure was seen along the course of the dorsal cutaneous branch of the ulnar nerve in the distal forearm which further divided into the two separate structures in the dorsum of wrist and hand [Figure 1a and b]. The lesion showed hyperintense signal intensity on T1, T2 sequences with suppression on T2FS (T2 Fat Saturated), suggesting the presence of fat [Figure 2a and b]. Fine fibrillar longitudinally oriented structures were seen within the lesion consistent with thickened nerve fibers. Adjacent osseous structures were normal without any evidence of osseous deformity or cortical destruction. The main ulnar nerve, radial and median nerves were normal. No mass was seen in Guyon's canal or carpal tunnel. No atrophy/denervation edema of hypothenar muscles was noted. The rest of the visualized structures of hand and wrist were unremarkable. There was no evidence of macrodactyly.

Diagnosis of lipomatosis of dorsal cutaneous branches of the ulnar nerve was made based on imaging findings. Biopsy and surgical interventions were not performed as they were deemed to be of no significant benefit; hence, the patient was managed conservatively.

DISCUSSION

Mason *et al.* first described lipomatosis of the nerve in 1953.^[1] Neural fibrolipoma, intraneural lipoma, perineural lipoma, or fibrolipomatous hamartoma are prior synonyms for the same entity.^[2] The term "lipomatosis of the nerve" was accepted as the accepted term for this entity by The World Health Organization in 2002.^[3] Lipomatosis of the nerve is portrayed by an unusual invasion of the nerve by fat and fibrous tissue making the influenced nerve thicker and mimicking a space-occupying lesion. Case reports are referencing related macrodactyly, some of the time enough to cause localized gigantism.^[1,4,5] It could be found in any age, however, most normally happens in the young.^[2] The upper extremities are more frequently involved than the lower extremities with median nerve affection the commonest.^[2] Lower extremity nerves involvement is extremely rare.^[2] A few of the associated rare congenital disorders with lipofibromatous hamartoma are Klippel-Trenaunay-Weber syndrome or Proteus syndrome.^[2]

The dorsal cutaneous branch of the ulnar nerve arises about 5 cm proximal to the wrist. It courses posteriorly deep to the flexor carpi ulnaris, passes through the deep fascia, and runs along the ulnar side of the dorsum of the wrist and hand. Here, it bifurcates into two dorsal digital branches. The first



Figure 1: (a) Coronal T1 along distal radioulnar joint. T1 hyperintense lobulated well circumscribed lesion surrounding dorsal cutaneous branch of ulnar nerve, which appears mildly thickened. (b) Axial T2 at the level of hook of hamate. T2 hyperintense lobulated well circumscribed lesion surrounding dorsal cutaneous branch of ulnar nerve, which appears mildly thickened.



Figure 2: (a) Zoomed axial T1 at proximal wrist. T1 hyperintense lobulated well circumscribed lesion surrounding mildly thickened dorsal cutaneous branch of ulnar nerve. (b) Zoomed axial T2FS in proximal wrist suppression of T2 hyperintense signals on T2FS sequence, suggesting presence of fat.

branch carries the sensory information from the ulnar side of the little finger, and the second branch is from the adjoining aspects of the little and ring fingers.

In the absence of macrodactyly, radiographs are often unremarkable; they may show a radiolucent soft-tissue mass that corresponds to fat. Bone and soft-tissue overgrowth are seen in the case of macrodactyly. After puberty, no osseous overgrowth is usually expected. In ultrasonography, alternating hyperechoic (fat), and hypoechoic (nerve fascicles) bands in a diffusely enlarged nerve, giving a "cablelike" appearance are reported.

Conventional modalities like radiographs have a limited role. Both ultrasound and computerized tomography can help in the initial detection; however, MRI due to its unparalleled soft-tissue resolution is considered the gold standard. MRI gives the diagnosis of lipomatosis of the nerve with better certainty, blocking the requirement for histopathologic evaluation and hence restricting the requirement for superfluous biopsies.^[2,5]

The hypertrophied fat that encompasses the nerve filaments and fusiform expansion of the nerve brings about "spaghettilike" in longitudinal section and as "coaxial link like" in crosssections; they are generally pathognomonic for lipomatosis of the nerve on T1-weighted and T2-weighted sequences without edematous changes on T2.^[2,4,5] With fat suppression methods, the high signal of the fat tissue vanishes aside from the nerve filaments. The lesion is usually non-enhancing. However, mild enhancement of the surrounding structures may be seen.

A regular MRI sequence describes the lesion, yet it needs explicitness in the differential analysis of delicate softtissue growths because numerous lesions show fluctuating qualities with no particularity. A diffusion-weighted image (DWI) used in association with a conventional MRI adds to diagnostic confidence. A DWI permits both quantitative and subjective examinations of tissue cellularity and cell membrane uprightness; and hence, it has been generally utilized for lesion localization and characterization. Differentiation between benign from malignant soft-tissue tumors can be aided by DWI done in increasing "b values."^[6]

CONCLUSION

Even though the location is rare and can confound initial clinical diagnosis, the characteristic MRI findings are typical for lipomatosis of the nerve. Lipomas of the hypothenar region and Guyon's canal clinically interpreted as pressure neuropathy of the ulnar nerve have been accounted for in literature. However, to the best of our knowledge, there has not been a single case of lipofibromatous hamartoma of the dorsal cutaneous branch of the ulnar nerve which has been reported in the literature.

As a final statement about lipomatosis of the nerve, it is a "do not touch" lesion in which biopsy is contraindicated. Hence, imaging diagnosis is of paramount importance to prevent unnecessary intervention-related morbidity.

Declaration of patient consent

Patient consent is not required as the patient's identity is not disclosed or compromised.

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Conflicts of interest

There is no conflict of interest.

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