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Case Report

Median Nerve Intraneural Lipoma as a Cause of Carpal Tunnel Syndrome: A Case Report and Review of Current Management Principles

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With few cases reported on the combination of intraneural lipoma resulting in carpal tunnel syndrome, our aim is to present further case evidence of such a condition and review current literature regarding workup and management of this condition.

CASE REPORT

A 53-year-old female presented with symptoms of numbness, pain, and tingling isolated to the radial three digits of the right hand for 4-5 years. Her symptoms progressively worsened over the preceding 2-3 months causing sleep disturbance and fatigue with activities requiring prolonged use of the hand. As her symptoms worsened, she noticed a lump forming on the volar surface of the wrist causing exacerbation of her symptoms with applied pressure. Of note, she also reported a history of two benign tumors that were removed from the ipsilateral palm roughly 20 years prior.

On physical exam she had a soft, mobile mass measuring roughly 2.5cm at the level of the distal wrist crease. She exhibited no thenar muscle atrophy, she had a positive Tinnel's sign to the median nerve, 5/5 muscle strength to flexion/extension to all digits, as well as intrinsic musculature. She had intact 2-point discrimination without deficit to the entirety of the hand, and brisk capillary refill to all digits.

Given her presentation, carpal tunnel syndrome (CTS) due to median nerve compression by an adjacent soft tissue mass was the principal concern. Patient underwent nerve conductions studies, EMG, and MRI with contrast of the wrist for further evaluation. Electrophysiologic studies were consistent with median nerve neuropathy at the carpal tunnel without evidence of neuronal loss. MRI revealed a homogeneous, non-enhancing mass measuring 8 x 8 x 28mm within the median nerve.

Following these studies, the patient underwent marginal excision of the mass with associated open carpal tunnel release. Intraoperatively, following release of the carpal tunnel an intradural mass was encountered at the level of the wrist crease. The majority of the mass was located on the ulnar aspect of the median nerve with few nerve fascicles running ulnar to the mass. The mass was then atraumatically dissected free from the native nerve tissue. Final pathology was consistent with mature adipose tissue and lipoma without atypia. At the patient's first follow up appointment she noted complete relief of her neurological symptoms. 6 months post operatively the patient continues to do well without recurrence of her previous symptoms or recurrence of the lipoma.

DISCUSSION

Idiopathic carpal tunnel syndrome represents one of the most common median nerve pathologies. An estimated 4% of the population will experience symptoms of carpal tunnel syndrome worldwide (Chammas et al. 2014). Additionally, lesions of adipose tissue are some of the most commonly encountered soft tissue masses in the body. These masses rarely occur in close relation to peripheral nerves

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Conflicts of Interest Statement for Dr. Amato

b Dr. Palumbo is an orthopaedic surgeon, specializing in surgery of the hand.

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Conflicts of Interest Statement for Dr. Palumbo

(Flores and Carneiro 2007; Okubo et al. 2012) but can create an atypical etiology of CTS with involvement of the median nerve (Okubo et al. 2012). Given the anatomy of the wrist and palmar tissue these lesions may not be readily identified on clinical exam making this particular diagnosis challenging (Bagatur and Yalcinkaya 2009). Many different names and classifications of these lipomatous lesions have been used interchangeably for many years throughout the literature. Broadly these lesions can be classified as extraneural and intraneural depending on the lesions' relationship with native neural tissue (Walter et al. 2011). Recently, intraneural lesions were further be broken down into forms depending on their relationship to the fascicle structures: encapsulated intraneural lipoma, (those involving no component of the nerve fascicles and surrounded by its own capsular tissue), and hybrid intraneural lipomas (the components of the mass are found intermixed within the fascicles and contain sctructureal aspects of each of the other nerve lesions). Lastly, lipomatosis represents a form of diffuse fatty deposition within the nerve without any distinctive capsular component (Marek et al. 2018). A summary table of these lesions is listed in Figure 1. Hybrid intraneural lipomas represent an even rarer form of peripheral nerve compression and neuropathy with reports of 24 cases in the literature (Marek et al. 2018). The median nerve is affected to the greatest extent based on published case reports (Gosk et al. 2015; Jalan et al. 2011; Flores and Carneiro 2007).

Though a rare cause of CTS, certain characteristics of this patients' presentation suggested an irregular cause of her symptoms. The unilaterality of her symptoms vs the more frequent bilateral presentation, the relatively benign results of the nerve conduction studies without evidence of gross neuronal compromise, and the slowly progressive mass (Bagatur and Yalcinkaya 2009; Nakamichi and Tachibana 1993). Interesting is the patient's history of previously excised benign palmar masses which may have represented asymptomatic palmar lipomas, of which have also been implicated in compressive neuropathy (Bagatur and Yalcinkaya 2009).

While this patient along with other previously reported cases have had physical exam findings that led to suspicion of her symptoms being the result of median nerve compression due to a nearby soft tissue mass, not all cases may present in a clear manner and may have little in the way of physical exam findings to lead practitioners to suspect and underlying mass (Gennaro, Merciadri, and Secci 2012). To our knowledge the currently reported cases of this condition have been identified through preoperative workup vs during carpal tunnel release. Given the increasing prevalence of carpal tunnel release being performed in the office setting as well as with endoscopic techniques this also presents a challenge should a similar lesion be identified at the time of surgery. To date we are not aware of reported cases of median nerve lipomas being identified in these particular operative settings, however should this be encountered in the office or via an endoscopic technique it would be advisable to proceed with the workup described below and ultimately address the lesion utilizing a more traditional open approach.

Workup will typically proceed in the traditional fashion for diagnosing CTS and consist of physical exam and likely nerve conduction studies. In the setting of a soft tissue mass present on physical exam MRI represents the imaging modality of choice in these lesions. Lipomas traditionally appear as a homogeneous mass, with sharp borders, high signal intensity on T1 and low signal intensity T2 fat suppression sequences, with no enhancement with gadolinium contrast (Ergun et al. 2010). High-grade liposarcomas, alternatively have a heterogeneous appearance with low intensity on T1-weighted images. In general, a larger size, deeper localization, or enhancement with contrast medium on MRI is suggestive of malignant soft tissue tumors (Nagano et al. 2015). Had this patient had more concerning findings during her diagnostic workup such as heterogenous appearance on MRI, lesions size greater than 5cm, and deep location of the mass, additional workup would have been warranted. Concerning MRI characteristics would indicate the patient for a preoperative image guided biopsy to better classify the mass and guide surgical planning. At this stage a coordinated effort between an orthopedic oncologist, musculoskeletal radiologist, and hand surgeon would also be appropriate in guiding final surgical plans (Cribb et al. 2005). Should biopsy results confirm the presence of a malignant soft tissue mass, surgical strategies will need to change drastically. This location offers a very challenging treatment dilemma in terms of the desire to eradicate the malignant tissue while attempting to retain limb function for the patient. Ultimately, eradication of the malignant tissue is the primary goal and should take precedent over preservation of limb function (Thomas et al. 2011).

Marginal excision of the mass is the ultimate treatment of extraneural and intraneural lesions of the median nerve. However, instances of diffuse nerve involvement such as with lipomatosis without clear surgical borders, conservative measures such as nerve decompression may need to be considered (Kim et al. 2005). The subset of hybrid intraneural lipomas represent an added surgical challenge as they form between nerve fascicles requiring careful dissection of the mass. Intraoperative ultrasound use has been reported to aid in surgical planning of tumor resection especially in instances without clear demarcation of the pathologic tissue (Haldeman, Baggott, and Hanna 2015). Another recent device, the Checkpoint Surgical nerve stimulator and nerve locator, allows for identification of nerve structures including individual fascicle groups. The primary benefit of using this device is the ability to discern nerve structures via stimulation even within areas of distorted anatomy (Checkpoint Surgical 2020). Especially in instances where the lipoma is intermixed within the fascicle structure of the nerve, the ability to accurately identify and preserve these structures during dissection is of utmost importance. While this may not be a necessary tool in all cases such as purely extraneural lipoma where there is no identifiable involvement of deeper neural structures, hybrid intraneural lipomas represent a subset of these lesions where this device can offer substantial aid to the surgeon.

Figure 1.

Extraeural Lipoma	Lipoma is separate from the native nerve tissue; this structure causes compression on the periphery of the nerve without invading the nerve proper.
Intraneural Lipomas	**Encapsulated Intraneural Lipoma:** Lipoma is found within the epineurium however; this structure resides on the periphery of the nerve and does not disrupt the organization of the nerve fascicles. **Hybrid Intraneural Lipoma:** This represents a lipoma with characteristics of each of the other forms. This lesion will consist of components within the nerve that disrupt the normal organization of the fascicles that may or may not be surrounded by its own capsule, as well as have a component extending beyond the epineurium exerting external compression.
Lipomatosis	Diffuse deposition of fatty tissue within the nerve proper that disrupts the fascicular structure and does not contain a clear capsule separating the lesion from the native nerve tissue.

Ultimately, for this patient observation for recurrence of the lesion will be paramount and would need to be suspected should her symptoms return. While data regarding the rates of recurrence of lipomas in this anatomic area is limited owing to the small number of reported cases. One study reporting on deep atypical lipomas of the extremities found a local recurrence rate of 8% (Sommerville et al. 2005). Additionally, there has been evidence to show increased rates of recurrence with increasing heterogeneity of the mass on preoperative imaging (Cribb et al. 2005). These imaging characteristics can help in the postoperative counseling of patients.

CONCLUSION

An intraneural lipoma resulting in CTS marks a rare cause of this condition with few reported cases. This case report provides further insight into the patient presentation, workup, and treatment of this condition. Atypical presentations of carpal tunnel warrant added suspicion and workup during evaluation. Ultimately, surgical resection provided significant and ongoing symptom relief to our patient with no associated morbidity, though she was counseled that continued monitoring for potential recurrence will be required.

DISCLOSURES

None of the authors has a financial interest in any of the products, devices, or drugs mentioned in this manuscript.

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SUPPLEMENTARY MATERIALS

Images

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