

# Navigating Beyond Neuromas: A Riveting Case Report Unveiling Intriguing Differential Diagnoses

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## Abstract

The purpose of this research was to evaluate the pathophysiology, diagnosis, and differential diagnosis of metatarsalgia by describing a clinical case of a rare lower limb injury, fibrolipomatous hamartoma of the plantar nerve (HFL). Material and Method: We present the case of a 47-year-old patient with a one-year history of pain in the anterior region of the right foot, associated with paresthesia of the lesser toes and no history of trauma. Physical examination revealed hyperkeratosis on the third rocker over the 2nd metatarsal (MTT), local pain, and neuritic symptoms, along with a doubtful Mulder sign. X-rays showed a negative index with narrowing of the space in the second toe. Magnetic resonance imaging described findings suggestive of a possible Morton's neuroma in the second space. Initial conservative management was chosen. After the lack of response to conservative treatment, surgical intervention was decided, during which the digital nerve was visualized and excised. Subsequently, a Triple Weil osteotomy was performed on the second and third MTT to prevent transfer metatarsalgia. The postoperative period was uncomplicated, and the patient showed complete relief of symptoms without recurrence after surgical excision. HFL presents as a rare benign tumor with an atypical location in the foot, generating symptoms similar to Morton's neuroma. The imperative need for a differential diagnosis with this entity is emphasized.

**Keywords:** Morton's Neuroma; Forefoot; Pathological Anatomy

## Introduction

Metatarsalgia, a frequently encountered issue prompting consultations within Foot and Ankle Services, encompasses a spectrum of etiological factors categorized into three primary groups: mechanical, inflammatory or rheumatic in origin [1], and a miscellaneous third group. Within this diverse third category, we present an intriguing and rare case—a fibrolipomatous hamartoma classified among peripheral neurogenic tumors affecting the foot. This expansive narrative seeks to delve deeply into the clinical and surgical dimensions of this unusual tumor, which elicits neuritic pain and paresthesias along the distribution of the common digital nerve [1,2]. Furthermore, we aim to underscore the diagnostic challenges associated with this condition, often leading to confusion with Morton's neuroma, a more common place forefoot affliction [1,2].

Metatarsalgia, characterized by pain and discomfort in the forefoot region, is a symptom rather than a specific diagnosis, making it imperative to discern its underlying cause for effective management [1,3]. The diverse etiological factors contributing to metatarsalgia are broadly classified into three categories.

The first category encompasses mechanical causes, including abnormal foot biomechanics, excessive pressure on the metatarsal heads, and structural deformities such as hammertoes or claw toes.

The second category involves inflammatory or rheumatic conditions [1], such as arthritis or synovitis, which can contribute to pain and discomfort in the forefoot. Finally, the third category encompasses a heterogeneous group of miscellaneous causes, which may include neoplastic growths, nerve entrapments, or, as in the case we present, fibrolipomatous hamartomas [3,4].

Fibrolipomatous hamartomas are rare benign tumors characterized by the infiltration of fibrous and adipose tissue into peripheral nerves. [1,3] While these hamartomas can occur in various anatomical locations, their manifestation within the foot is particularly uncommon. The rarity of such cases necessitates a comprehensive exploration of clinical presentations, diagnostic challenges, and optimal management strategies. In this context, our contribution aims to elucidate the intricacies associated with fibrolipomatous hamartomas, focusing on a specific case involving the plantar nerve scenario that, to our knowledge, has been documented in the literature only once before.

The case under discussion involves a fibrolipomatous hamartoma situated within the plantar nerve, a peripheral neurogenic tumor that, despite its infrequency, poses substantial diagnostic and therapeutic implications. The patient presented with symptoms of neuritic pain and paresthesias along the distribution of the common digital nerve, a clinical manifestation that often mimics Morton's neuroma. Morton's neuroma is a common condition involving a benign growth of nerve tissue, typically between the third and fourth metatarsal heads. The similarity in symptomatology between fibrolipomatous hamartomas and Morton's neuroma underscores the importance of a meticulous differential diagnosis to ensure accurate and timely intervention.

The diagnostic journey for our patient was characterized by challenges stemming from the overlapping clinical features of fibrolipomatous hamartomas and Morton's neuroma. Neuritic pain, characterized by sharp, shooting sensations along the nerve distribution, and paresthesias, abnormal sensations like tingling or numbness [3,4], are hallmark symptoms shared by both conditions. Imaging studies, including magnetic resonance imaging (MRI), played a crucial role in delineating the extent and nature of the lesion. The MRI revealed a fusiform enlargement of the plantar nerve with infiltration of fibrous and adipose tissue, confirming the diagnosis of a fibrolipomatous hamartoma.

The rarity of fibrolipomatous hamartomas, especially those affecting the plantar nerve, necessitates a nuanced approach to their management. Surgical intervention is often warranted to alleviate symptoms and prevent further progression of the tumor. In the case we present, the surgical excision of the fibrolipomatous hamartoma proved to be a viable and effective solution, providing relief from neuritic pain and paresthesias [4,5,6]

Importantly, the surgical approach required meticulous dissection to preserve the integrity of the affected nerve while excising the abnormal tissue, emphasizing the importance of tailored surgical strategies [5,7] in managing rare peripheral neurogenic tumors.

This case contributes significantly to the limited body of literature [5,8] on fibrolipomatous hamartomas of the plantar nerve. The scarcity of reported cases underscores the challenges associated with recognizing and diagnosing these tumors, especially when presenting with symptoms akin to more prevalent conditions like Morton's neuroma [1,3,4].

Our aim in presenting this case is twofold: first, to enhance the understanding of clinicians, particularly those in the realm of foot and ankle specialties, regarding the clinical nuances associated with fibrolipomatous hamartomas; and second, to underscore the importance of a meticulous and comprehensive diagnostic approach in cases of forefoot pain that extend beyond conventional pathologies [2,5,6].

In conclusion, metatarsalgia, as a symptom complex, demands a thorough investigation to identify its underlying cause for appropriate management. Fibrolipomatous hamartomas, though rare, represent a distinct entity within the spectrum of peripheral neurogenic tumors affecting the foot. The presented case of a fibrolipomatous hamartoma of the plantar nerve highlights the clinical challenges associated with this condition and emphasizes the need for a precise and comprehensive diagnostic approach [5,8]. By sharing our clinical and surgical experience, we hope to contribute to the evolving understanding of rare foot pathologies and encourage a heightened awareness of fibrolipomatous hamartomas in the differential diagnosis of forefoot pain.

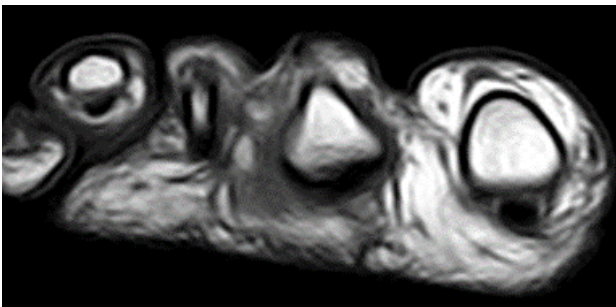
## Methods

The intricate landscape of forefoot pain demands a thorough and nuanced approach to differential diagnosis. While discussions often pivot around Morton's neuroma concerning metatarsalgia [2,5,8], our recent encounter with a distinctive case emphasizes the need to explore less common etiologies. Notably, the fibrolipomatous hamartoma of the plantar nerve (HFL) emerges as a rare and distinctive entity, urging healthcare practitioners to navigate unconventional diagnostic pathways.

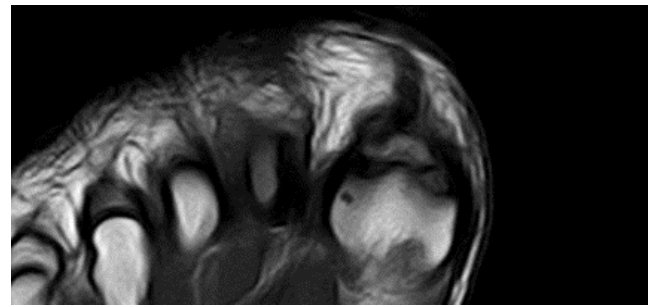
A 47-year-old individual sought our hospital's expertise with a persistent one-year history of discomfort in the right forefoot, accompanied by unsettling paresthesia in the lesser toes. The absence of notable medical history or traumatic incidents introduced an additional layer of complexity to the diagnostic challenge. The patient's vivid depiction of acute and enduring pain, intensified during weight-bearing activities on the 2nd and 3rd metatarsals (MTT), triggered an in-depth exploration of the underlying pathology.

During the physical examination, subtle indicators such as a mild diversion between the 2nd and 3rd toes and hyperkeratosis on the third rocker in 2MTT hinted at the intricate interplay of factors contributing to the patient's discomfort. Localized pain, neuritic symptoms radiating to the corresponding toes, and a Mulder sign that allowed room for doubt further heightened the diagnostic complexity. Remarkably, the neurovascular examination yielded normal results, injecting an element of intrigue into the clinical presentation.

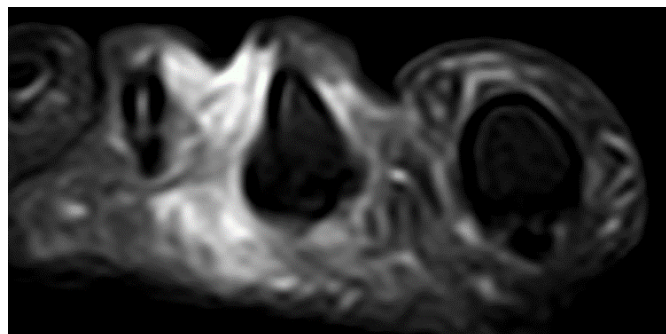
The diagnostic odyssey unfolded through a series of imaging studies, with X-ray and magnetic resonance imaging (MRI) providing glimpses into the labyrinthine nature of the forefoot pathology. The X-ray unveiled a negative index with a narrowed 2nd space, casting shadows of potential abnormalities. The MRI findings in T1 and T2 SAT in coronal and axial planes of the forefoot (Figure 1, 2 and 3), suggestive of Morton's neuroma in the 2nd space, with no other lesions described in the rest of the anatomy, propelled the investigative process further, guiding us into a realm where commonplace diagnoses may inadequately encapsulate the clinical narrative.



**Figure 1.** Coronal plane MRI in T1 Hospital Infanta Elena de Madrid).



**Figure 2.** Axial plane MRI in T1 Hospital Infanta Elena de Madrid)



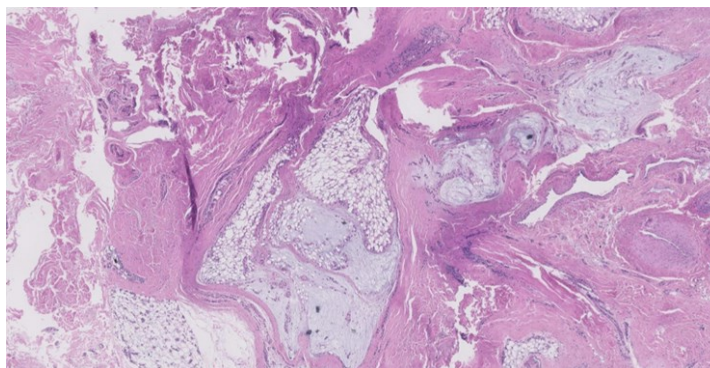
**Figure 3.** Coronal plane MRI in T2 fat SAT Hospital Infanta Elena de Madrid)

## Results

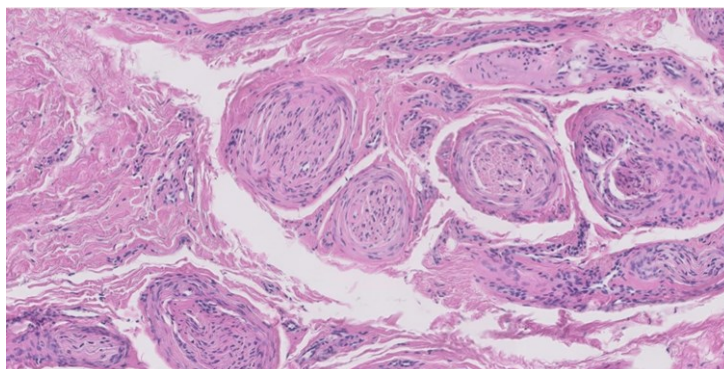
The patient was initially treated conservatively with infiltration of local anesthetic and corticosteroids (3 ml of 2% mepivacaine + 2 ml of betamethasone), oral analgesia, and metatarsal offloading insoles. Following the failure of conservative treatment and persistence of symptoms one year later, a decision was made for surgical exploration of the 2nd metatarsal space. Preoperative prophylactic antibiotic therapy (2g cefazolin) was administered intravenously. A popliteal block was performed, and a pneumatic tourniquet was applied at the ankle level. A longitudinal dorsal approach was made along the midline of the second intermetatarsal space to avoid damage to the dorsal cutaneous nerves. Precise dissection was carried out until the identification of the deep transverse metatarsal ligament, followed by its longitudinal section. The digital nerve was visualized, surrounded by a fibro-fatty tissue mass, and excised. All samples were sent to the laboratory for histopathological examination. Subsequently, a Triple Weil osteotomy of the 2nd and 3rd metatarsals was performed to prevent transfer metatarsalgia. All osteotomies were secured with screws. After thorough irrigation with physiological saline, layered closure was performed, and a compressive dressing was applied.

The postoperative period was uneventful. The patient commenced ambulation immediately postoperatively wearing an inverted heel shoe without loading on the forefoot for 1 month. After the first postoperative month, the patient was evaluated on an outpatient basis with a weight-bearing foot X-ray, examining the results of the pathological anatomy. From a clinical standpoint, there were no complications at the wound site, with slight swelling on the dorsum of the foot. The postoperative X-ray revealed a metatarsal formula plus-minus without apparent forefoot overload, allowing the patient to remove the post-surgical footwear at that time. Histology (Figure 4) revealed the presence of an exceptional entity in the foot: a fibrolipomatous hamartoma of the plantar nerve with infiltration of the perineurium and epineurium by fibroadipose tissue that separates and compresses nerve fascicles, causing atrophy of neural elements.

It is accompanied by perineural fibrosis and focal areas of myxoid material deposition, devoid of stromal cells (Figure 5). No evidence of Morton's neuroma was observed. The subsequent patient evaluation was conducted at 3 months, demonstrating complete relief of symptoms without recurrence following surgical excision.



**Figure 4.** Pathological anatomy section Hospital Infanta Elena de Madrid.



**Figure 5.** Pathological anatomy section Hospital Infanta Elena de Madrid.



## Discussion and Conclusions

Pain in the forefoot is a common reason for consultation, and the differential diagnosis can be challenging. Excluding mechanical or inflammatory metatarsalgia, Morton's neuroma is the most common cause. Morton's neuroma (MN) is a highly prevalent cause of neuropathy in this location [1,2,5,6].

MN is a neuropathy of the plantar nerve due to entrapment in the distal part of the transverse intermetatarsal ligament, where the nerve lies between the metatarsal heads. It is usually located between the 3rd and 4th metatarsals (75% of cases), and occasionally between the 2nd and 3rd (17% of cases). It very rarely occurs in other locations.

Medical history and physical examination can usually diagnose MN. The typical pain is triggered by palpation of the interdigital space and Mulder's maneuver [1,2]. The primary treatment is conservative, consisting of orthopedic treatment (wide shoe with soft sole and low heel, metatarsal support, splint, or strapping), medication (NSAIDs, antidepressants, anticonvulsant analgesics, or opioids), and corticosteroid infiltration [5,6,7].

Other options include non-surgical treatments (radiofrequency ablation, cryotherapy, or neurolytic procedures). In case of failure of conservative treatment, surgical excision of MN is indicated [5,7].

Sudden, lancinating, and periodic pain in the forefoot associated with paresthesias may be due to other rare etiologies, as found in our case:

The fibrolipomatous hamartoma (FLH) of the plantar nerve is an extremely rare benign lesion, also known as neural fibrolipoma or perineural lipoma or intraneural lipomas or lipomatous hamartomas. It is a benign, uncommon lesion first described by Mason in 1953 [9].

It is located in peripheral nerves of the upper limbs, with the median nerve being the most common location. Its manifestation in the lower extremities and proximal peripheral nerves is rare.

Although the etiology is still unknown, it is believed to result from hypertrophy of adipocytes and fibroblasts in the perineurium. There is no association with genetic syndromes [9,10], although there is a familial predisposition. It can be associated with neurofibromatosis and macrodactyly [6,8], characterized by bone deformity and increased adjacent soft tissues (lipomatous macrodystrophy). There is no gender predilection, but the presence of macrodactyly is more common in women. In our case, there was no association with macrodactyly or neurofibromatosis.

Clinically, it presents as a slowly growing, asymptomatic mass, later causing pain, weakness, and paresthesias similar to the picture produced by MN [9,10]. Therapeutic management is not clearly standardized; however, conservative treatments are initially favored, progressing to surgical treatment after the failure of the former. FLH and MN exhibit similar characteristics. Both conditions involve middle-aged adults, and in the physical examination, the neuritic pain produced by FLH and MN is very similar [9,10].

Currently, magnetic resonance imaging (MRI) is the gold standard for diagnosing foot and ankle pathology. MN in MRI appears hypo- or hyperintense on T1- and T2-weighted images, respectively, depending on the degree of fibrosis (Figure 1,2,3). This clinical entity can be observed as a fusiform or teardrop-shaped soft tissue mass located between the metatarsal heads of the affected space [7]. FLH in MRI presents a pathognomonic image with fusiform thickening of the affected nerve, with hypointense images on T1 and STIR corresponding to nerve fascicles, separated by hyperintense areas in T1 and hypointense areas in STIR, corresponding to adipose tissue (Figure 1,2,3). These features are pathognomonic and constitute the so-called cable sign [4,8,9].

The definitive and differential diagnosis [10] between FLH and MN, as demonstrated by the aforementioned elements, can be made using MRI, with histological examination taking a secondary role (Figure 4,5). It should be noted that sometimes these two pathologies may coexist [9,10].

In our case, the clinical presentation was not accompanied by macrodactyly, the MRI (Figure 1,2,3) described findings of a possible Morton's neuroma, and yet the histological examination resulted in the diagnosis of fibrolipomatous hamartoma of the plantar nerve.

FLH is a benign and uncommon tumor with an atypical location in the foot. It produces clinical neuropathy similar to the symptoms presented by MN. We emphasize the importance of always considering a differential diagnosis with this entity. Although it is rare and uncommon, in middle-aged patients presenting with neurological deficits or peripheral compressive syndromes, it is essential to consider this condition early on to ensure proper treatment.

## Conflict of Interest

The authors declare that they have no conflict of interest.

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