DOI: https://doi.org/10.30795/jfootankle.2020.v14.1144

Case Report

Interdigital neuroma in a patient with macrodactyly of the hallux: case report

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Abstract

A patient with macrodactyly of the hallux, returned 2 years after amputation of the distal phalanx, complaining of pain and swelling in the plantar foot. The Tinel, Moulder, and Gauthier signs were all present. Diagnostic hypotheses were: neuroma of the amputation stump, compressive neuroma, neurofibroma, or schwannoma. Histopathological diagnosis demonstrated that the tumor was a neuroma. This is a rare and unique case associated with macrodactyly, in which nerves tend to be hypertrophic. The location in the first intermetatarsal space is uncommon. The treatment proposed was resection of the entire involved nerve; symptoms improved and there was no relapse.

Level of Evidence V, Therapeutic Studies; Expert Opinion.

Keywords: Neuroma; Foot deformities, congenital; Orthopedic surgery; Neurofibrosarcoma.

Introduction

Macrodactyly is a rare congenital condition in which one or more of the fingers or toes are disproportionately larger than the others. It is caused by hypertrophy of all of the mesenchymal tissues and simultaneously involves soft tissues and bony components of the digits[1]. It is present at birth or detected in early childhood and tends to be progressive throughout the normal period of skeletal maturation[2].

The etiology of idiopathic forms is still unclear[2], although it is consensus that the origins are multifactorial[2]. Unilateral manifestation is more common and few bilateral cases are described.

Macroscopically, all of the structures of the affected digits are enlarged. The flexor tendons appear normal, although larger. The digital nerves are thickened and tortuous[2].

Treatment of macrodactyly is decided on a case-by-case basis. Several aspects should be taken into consideration: type of macrodactyly, velocity of disease progression, the digits involved, and the age of the patient. The primary objective of treatment of macrodactyly of the toes is to obtain feet that can fit into footwear, enable the patient to walk, and achieve a good clinical appearance of the toes. Surgical treatment is needed to achieve this and can encompass resection of redundant tissues, tenodesis, epiphysiodesis, and shortening of the bone, and in some cases may require amputation of the digit or ray[1].

We present a rare case of macrodactyly of the great toe, in which, 2 years after the first surgical procedure, a large, painful, and palpable neuroma developed in the plantar region, located in the first intermetatarsal space of the foot.

Case report

This study was approved by the Institutional Review Board and registered on the Plataforma Brasil database under CAAE (Ethics Evaluation Submission Certificate) number: 29530019.1.0000.5501.

The patient, S.R., was a 46-year-old, white, male laborer who was referred to the foot and ankle surgery clinic at the University Hospital in Taubaté, SP, Brazil, complaining of an excessively large left hallux that made it difficult to wear shoes. He was diagnosed with macrodactyly of the hallux and we recommended amputation of the distal phalanx of the great toe, with resection of excessive soft tissues, thereby reducing both width and length of the toe.

Study performed at the Hospital Municipal Universitário de Taubaté, Taubaté, SP, Brazil.

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E-mail: ortopediahenrique@gmail.com. **Conflicts of interest:** none. **Source of funding:** own. **Date received:** March 30, 2020. **Date accepted:** June 27, 2020. **Online:** August 30, 2020.

How to cite this article: Silva HSB, Lara LCR, Grajales JÁ, Torres Filho LCA, Furtado Lancia LF. Interdigital neuroma in a patient with macrodactyly of the hallux: case report. J Foot Ankle. 2020;14(2):187-90.

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The patient did not have a family or genetic history of macrodactyly and had no associated comorbidities.

Two years after the first operation, the patient returned to the clinic, complaining of progressive pain in the plantar region of the forefoot, exacerbated by walking. Physical examination revealed a palpable and painful nodule. This tumor was mobile, of a fibroelastic consistency, with a largest dimension of approximately 6cm, and was located in the first intermetatarsal space. The Tinel, Moulder, and Gauthier signs were all positive. Palpation of the hallux amputation stump was painless and there were no signs of paresthesia, or shock in response to percussion.

We ordered radiological (X-rays) and magnetic resonance (MRI) examinations of the foot. The X-rays did not show any changes, beyond the amputation of the distal phalanx of the hallux. The MRI images showed a nodular mass located in the first intermetatarsal space, measuring 6cm long by 2cm wide, with no signs of malignancy or invasion of soft tissues (Figure 1).

The patient was scheduled for surgery. Through a plantar incision and careful dissection, the nerve was identified and found to be greatly thickened, enlarged, and yellowed. The perineurium was soft and the interior was hard, without signs of malignancy when examined macroscopically. It did not resemble a stump neuroma, since the nerve was uninterrupted both before and after the neuroma (Figure 2).

Complete resection of the nerve was performed until apparently healthy neural tissue was reached. The surgical wound was sutured closed by planes up to the level of the skin. The foot was bound and the patient instructed to walk immediately. During postoperative recovery the patient remained free from pain and infection and his compressive symptoms improved (Figure 3). Anatomopathological examination revealed fragments of twisted nerve trunks surrounded by fibrosis arranged concentrically, with no signs of malignancy, confirming the preoperative hypothesis that the mass was an interdigital neuroma.

We did not assess the results of surgery by applying the AOFAS scale before and after the operation, since this was a single case, but we did assess pain with an analog scale. The preoperative pain score was 8, which reduced to 3 during the postoperative period, demonstrating good recovery.

Discussion

Macrodactyly is a rare congenital anomaly, which can be associated with other syndromes, although in this study it was an isolated case, with no association with other pathologies. Generally, all structures of the affected digits are enlarged: dermis, subcutaneous tissues, tendons, bones, and nerves.[1-3]

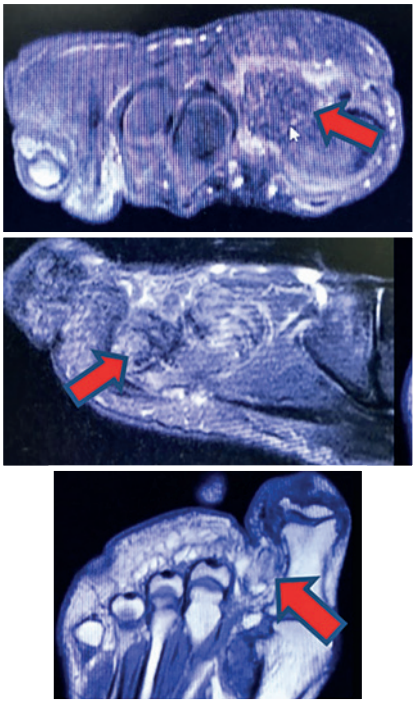

Figure 1. Magnetic resonance images showing details of the nodular lesion.

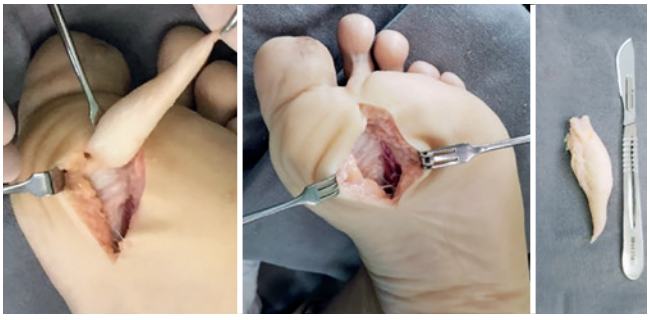

Figure 2. Images showing resection of the neuroma, and illustrating its size

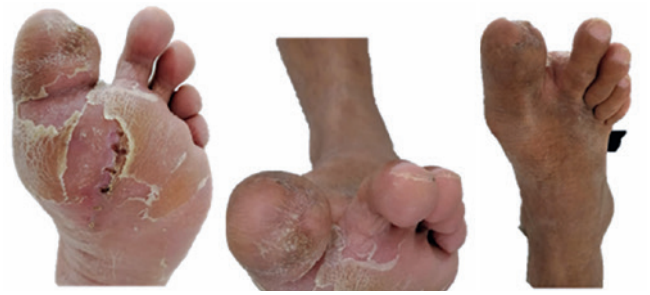

Figure 3. Postoperative appearance of the foot and the surgical scar.

We reported a rare case of an enlarged interdigital nerve, located in the first intermetatarsal space. The patient had previously presented with macrodactyly, which had been treated with surgery. However, 2 years after that first operation, he returned with pain and swelling in the plantar region.

The anatomopathological examination revealed fragments of twisted nerve trunks surrounded by fibrosis, arranged concentrically, and with no signs of malignancy. The patient was diagnosed with an isolated interdigital neuroma.

These symptoms are uncommon in the first space and even rarer in conjunction with macrodactyly. Taken together, the size of the nodule and the clinical status were strongly suggestive of neuroma and were reinforced by the clinical signs present[3].

In the literature, interdigital neuroma, known as Morton's neuroma, is frequently seen in the second and third intermetatarsal spaces, in 29% and 60% of cases respectively. In contrast, it is very rare in the first space, occurring in just 1 to 2.5%[4].

There are very few cases in this region in the literature. Another possibility in the differential diagnosis is schwannoma, a rare lesion that is uncommon in the foot, observed in just six cases in the literature[5-8]. A neurofibroma is easily differentiated with a histopathological study, which will show a myxoid stroma with collagen fibers in the interior, plexiform arrangement, and deformed neuronal axons[5].

We decided to conduct open surgery to treat the nodule because of its size, facilitating safe resection of the lesion.

New treatment methods exist that are reported to improve the painful symptoms, including use of high intensity laser treatment, which has shown promising results. Pain is reduced in up to 80% of cases and the size of the nerve thickening is reduced in up to 51%[9].

Some authors recommend using minimally invasive surgery to treat these neuromas, releasing the intermetatarsal ligament and performing osteotomies on metatarsal bones, creating more space between them and relieving the nerve compression. They report good results at 2-year follow-up.[10]

The results of surgical treatment for neuroma are excellent or good in 89% of cases, irrespective of the technique employed, compared to 85% for ablation with alcohol or radio frequency. The worst results are seen with conservative treatment, which is associated with 47% relapse or treatment failure[8].

Ultimately, surgical treatment is the best choice, improving symptoms in up to 89% of patients. Founded on this theory, a review study also considered that patients who are given good information will exhibit better results with surgical intervention[8].

In the case described here, the procedure performed was the best choice, in view of the size, characteristics, and location of the lesion. The tumor was very large and we needed a precise diagnosis.

We used an extended plantar surgical approach, affording an excellent view and allowing total resection. The patient recovered well after the operation, with relief from symptoms and no complications.

In current databases, it is uncommon to find cases diagnosed as neuromas in the first interdigital space associated with gigantism of the first toe, which is the main reason for this study.

Notwithstanding, other pathologies of the nerves can occur in the presence of macrodactyly, such as, for example: hamartomas, neurilemmoma (schwannoma), neurofibroma, or fibrolipomas, which are very rare in the lower limbs[9].

Interdigital neuromas located in the first space in the forefoot were described in 1% of cases by Bartolome and Wertheimer, in 1983, and in 2.5% of cases by Adante et al., in 1985, who studied 100 patients with Morton's neuroma, as reported by Thomas et al.[4].

All of these factors prompted us to report this uncommon case of a patient with both macrodactyly and a digital neuroma in the first space. We are not certain that the presence of this neuroma was because of the macrodactyly, but it is possible, as a result of compression of the nerve between the first and second metatarsals. The combination is possible, although they are two distinct pathologies.

Conclusions

This study is an alert that it is possible for a compressive neuropathy to develop in the first intermetatarsal patient in a patient with macrodactyly of the hallux.

Authors' contributions: Each author contributed individually and significantly to the development of this article: HSBS *(https://orcid.org/0000-0002-1549-0992) participated in the review process, approved the final version; LCRL *(https://orcid.org/0000-0003-1158-2643) conceived and planned the activities that led to the study, wrote the article, interpreted the results of the study; JAG *(https://orcid.org/0000-0003-4652-4400) wrote the article, participated in the review process; LCATF *(https://orcid.org/0000-0002-0778-2506) conceived and planned the activities that led to the study, approved the final version; LFL *(https://orcid.org/0000-0003-1048-7134) wrote the article, participated in the review process. *ORCID (Open Researcher and Contributor ID).

References

1. Pessôa FC, Costa EM, Martins IS. Macrodactilia: série de casos. Sci J Foot Ankle. 2018;12(3):255-9.
2. Monteiro AV, Chiconelli JR, Almeida SF. Macrodactilia: estudo retrospectivo de sete casos. Rev Bras Ortop. 1998;33(1):54-8.
3. Coughlin M, Saltzman CL, Mann RA (editors). Mann's surgery of the foot and ankle. 9ed. Philadelphia: Mosby; 2013.
4. Clinical Practice Guideline Forefoot Disorders Panel, Thomas JL, Blitch EL 4th, Chaney DM, Dinucci KA, Eickmeier K, Rubin LG, Stapp MD, Vanore JV. Diagnosis and treatment of forefoot disorders. Section 3. Morton's intermetatarsal neuroma. J Foot Ankle Surg. 2009;48(2):251-6.
5. Muratori F, De Gori M, Campo FR, Bettini L, D'Arienzo A, Scoccianti G, Capanna R. Giant schwannoma of the foot: a case report and literature review. Clin Cases Miner Bone Metab. 2017;14(2): 265-8.
6. Gimber LH, Melville DM, Bocian DA, Krupinski EA, Guidice MP, Taljanovic MS. Ultrasound evaluation of Morton neuroma before and after laser therapy. AJR Am J Roentgenol. 2017;208(2):380-5.
7. Park EH, Kim YS, Lee HJ, Koh YG. Metatarsal shortening osteotomy for decompression of Morton's neuroma. Foot Ankle Int. 2013;34(12):1654-60.
8. Valisena S, Petri GJ, Ferrero A. Treatment of Morton's neuroma: A systematic review. Foot Ankle Surg. 2018;24(4):271-81.
9. Murphey MD, Smith WS, Smith SE, Kransdorf MJ, Temple HT. From the archives of the AFIP. Imaging of musculoskeletal neurogenic tumors: radiologic-pathologic correlation. Radiographics. 1999; 19(5):1253-80.
10. Bauer T, Gaumetou E, Klouche S, Hardy P, Maffulli N. Metatarsalgia and Morton's disease: comparison of outcomes between open procedure and neurectomy versus percutaneous metatarsal osteotomies and ligament release with a minimum of 2 years of follow-up. J Foot Ankle Surg. 2015;54(3):373-7.