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Schwannoma of Foot, Case Report and Literature Review

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Abstract **Case Report**

Schwannomas are benign and rare nerve tumors, slow growing neoplasms originating from the peripheral nerve sheath. Its location in region of the head of the first metatarsal is a very rare presentation. A 43-year old patient reported a history of multiple pains over a year, with no sensorimotor deficit. MRI revealed presence of an isolated Schwannoma in the 1st intermetatarsal area, growing on the first metatarsal space with no associated fracture complication. The diagnosis was confirmed through preoperative biopsy. The patient benefited from surgical excision, bone curettage and adjuvant treatment with liquid nitrogen without bone filling. The final analysis of the surgical specimen confirmed the diagnosis of schwannoma. At the last follow-up, no recurrence was detected either clinically or imaging. This case is only the fourth schwannoma of the first metatarsal space reported in the literature.

Keywords: Intraosseous Schwannoma · Bone tumor· Foot tumor.

INTRODUCTION

Schwannoma, also known as a neurilemmoma, are benign and rare nerve tumors [1], it's slow growing neoplasms originating from the peripheral nerve sheath. That can occur anywhere in the body [2]. Few cases of this nerve sheath tumor have been reported in the long bones [3, 4]. Its location in region of the head of the first metatarsal is a very rare presentation [5-7]. The purpose of this work is to report the fourth case of intraosseous schwannoma of a metatarsal [5-7]; and to integrate this rare form of nerve tumor into the literature.

PATIENT AND OBSERVATION

It is a 43-year-old patient, military in operation, with no particular pathological history; especially family ones, and without vestibular or auditory symptomatology; describing a painful history having evolved over a year without sensitivo motor deficit. A difficult and painful walk and trigger pains were found during the interrogation. Clinical examination of the patient showed no lesion of the schwannoma type, no skin lesion of the neurofibroma type, no skin café au lait spots; it was possible to note a painful swelling in the first intermetatarsal space with a positive Tinel sign. Various paraclinic examinations were carried out following this consultation: simple radiography of the foot, ultrasound of the foot, MRI of the foot (Figure 1), and radio-guided diagnostic biopsy. The X-ray of the left foot was normal, the ultrasound found a lesion 3.2 cm wide, 2 cm high and 3 cm long, a cortical effraction without intraosseous invasion. MRI confirmed these dimensions as well as lateral cortical effraction of the first metatarsal. The MRI also showed that this lesion was in T1-weighted hypersignal and took the contrast after injection of gadolinium.

At this level a diagnosis of a benign lesion has been established. And after this assessment, a surgical intervention was decided. A dorsal approach was used. After locating and protecting the extensor tendons, the tumor was localized. The dorsal intermetatarsal nerve was located upstream and downstream of the tumor. The tumor could be removed in monobloc, and the nerve could be preserved. Once the extraosseous part was removed, cautious bone curettage was performed followed by adjuvant treatment with intraosseous liquid nitrogen (Figure 2). No bone filling was performed. Analysis of the surgical specimen concluded that benign schwannoma was diagnosed (Figure 3). The surgical follow-up was simple, partial support protected. The patient was doing well after 9 months of follow-up, with no clinical or radiological signs of lesion recurrence.

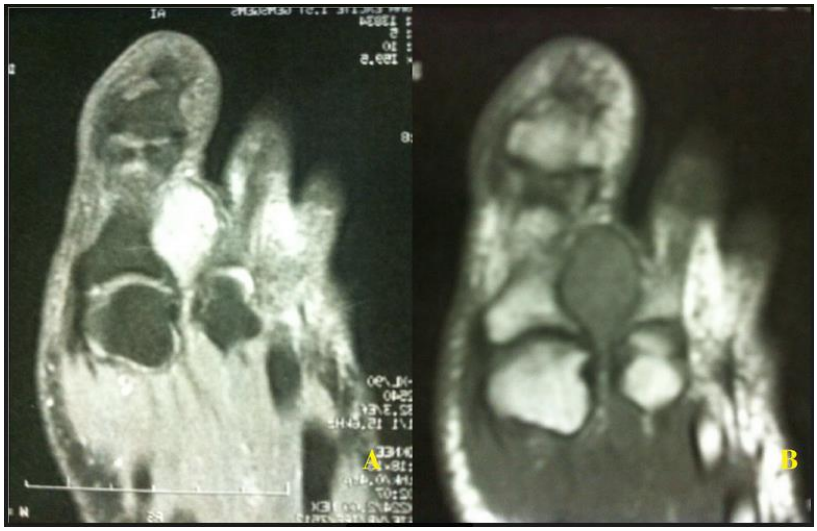

Figure 1: Preoperative T1 and T2 Axial section.

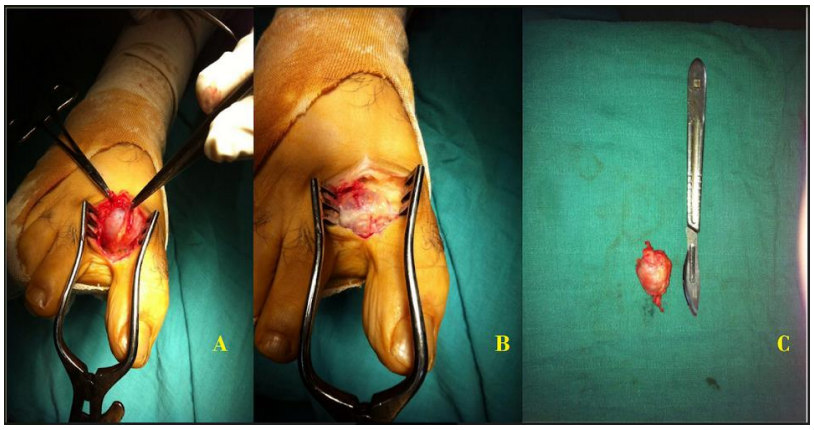

Figure 2: Intraoperative photos.

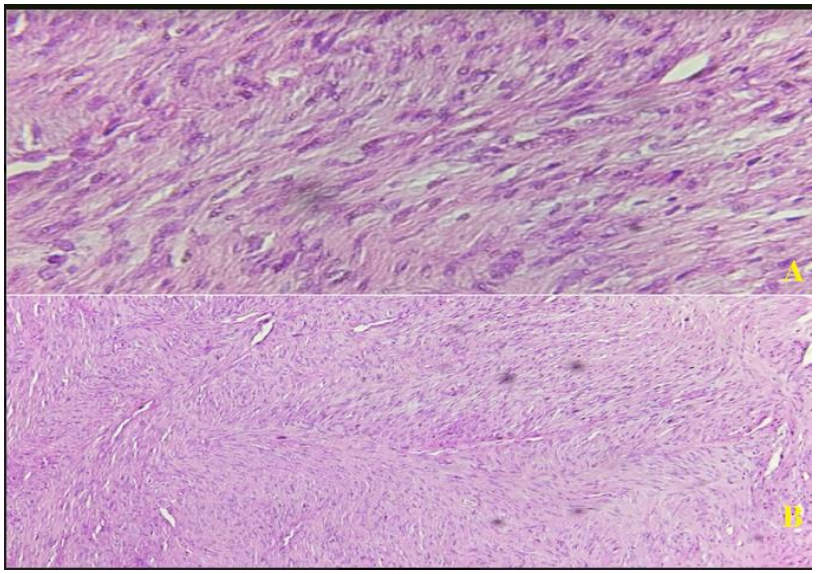

Figure 3: Hypercellular areas indicating an Antoni A area, hypocellular areas indicating an Antoni B area, and Verocay bodies are seen under the microscope.

Patient Perspective

The patient was satisfied with the treatment he received.

Informed Consent

The patient has been informed of the publication of this article; he has given his consent.

DISCUSSION

Peripheral nervous tumors are rare and usually benign [1]. They account for 1 to 10% of all soft tissue tumors [8]. Schwannoma, also known as a neurilemmoma, is a benign tumor arising from the sheath of myelinated nerve fibers that can occur anywhere in the body [2]. Schwannomas have been commonly seen associated with neurofibromatosis 1 and neurofibromatosis 2, but mostly NF 2. When multiple schwannomas are present in the absence of other stigmata of NF, then it can be defined as schwannomatosis [9].

Schwannomatosis was first described in 1973 as neurofibromatosis type 3 [9]. Along the years, many papers have been published noting further cases of schwannomatosis without features of NF, there by signalling a new entity. As per Jacoby et al., the presence of two or more schwannomas in the absence of radiological evidence of vestibular lesions in patients older than 18 years is definitely indicative of schwannomatosis [10]. According to Baser et al., MRI should be used to rule out presence of vestibular schwannoma and also any presence of NF mutations should also be ruled out [9]. Moreover, some authors recommend in view of the presence of several lesions, we must investigate the genetic mutation in the same context of schwanomatosis [10].

Intraosseous schwannoma accounts for only 0.2% of intraosseous tumors [3]; most frequently benign and rarely measure more than 5 cm in diameter [7]. Few bone sites have been reported [3-7]. Peers was the first to describe a schwannosarcoma in 1934 and Gross et al. An intraosseous benign neurofibroma in 1939 [2,11]. In 1984, de la *Monte et al*. [10 report 64 cases documented in the literature. The mechanism of bone localization is of three types: extraosseous localization and progressive erosion, infiltration through a feeder canal and intraosseous primary localization. Intraosseous forms are most commonly found in the skull, mandible and sacrum, but forms have also been reported in the upper limb [3,8] and lower limb [4-7].

Our case is only the fourth schwannoma of the first metatarsal space reported on the literature [5-7]. The first case which proves the existence of the first mechanism already described. The age of the patients is between 20 and 50 years, without predisposition according to sex [1]. Clinically, schwannoma is a slow growing, benign tumor that may be present for years before becoming symptomatic. Intraosseous schwannoma most commonly presents with complaints of swelling [10]. However, pain may be present in about 50% of the cases [1], whereas no symptoms are present in about 25% [10]. In the case presented in this article, the patient was long asymptomatic, he became aware of swelling after the onset of disabling pain.

The possibility of schwannoma of the first metatarsal space was not envisaged first in our case, despite a radiological aspect with MRI very evocative; But also because of the extreme rarity of this site. Histopathological examination provided a definitive diagnosis for the case. Intraosseous schwannoma radiographically is unilocular or multilocular and is usually associated with bone resorption and may be rarely associated with pathological fracture [10]. Intraosseous schwannoma appears as a limited lytic tumor with dense banks, cortical rupture and some rare internal calcifications [4]. On the MRI, the schwannoma appears: in sequence T1 in isosignal with the muscle and in T2 in frank hypersignal. The signal is homogeneous for lesions less than 3 cm, with sharp contours.

The malignant forms of schwannomas are heterogeneous in both T1 and T2 in relation to haemorrhagic necrosis plaques or myxoid plaques. In addition to diagnostic guidance, MRI can be used to determine tumor volume, its relationship to vascular structures and its muscular, fatty and bone extension [9]. The diagnosis of certainty is based on the anatomopathological analysis with the presence of Schwann ovoid and fusiform cells divided into two zones: compact plant (Antoni A cell and Verocay nodule) and myxoid peripheral (Antoni B cell) palissadic with weak interstitial tissue.

The tumor is peripheral and respects the nerve fibers unlike the neurofibroma. Malignant transformation is exceptional, and treatment is based on simple excision with bone filling [1,8,10]. Taking into account our patient's history of schwannoma, cortical erosion and despite the results of the initial biopsy, we preferred not to bone filling. Tumor lesions of soft tissues at the forefoot and intermetatarsal spaces are relatively frequent [10]. Close to the nervous or nervous structures, they are clinically difficult to distinguish.The most frequent are Morton's neuromas (second space in 19% and third intermetatarsal space in 75% of cases), followed by inter- and sub-metatarsal bursitis, gouty tophoses, epidermoid cysts and giant cell tumors of tendon sheaths.

Given the difficulty of ensuring the benign character of a lesion, a diagnostic biopsy must be systematic, especially since there are signs of aggressiveness (cortical erosion, heterogeneous aspect, height greater than 5 cm, deep seat, Perilional edema, necrosis zone and invasion of neurovascular structures) [6,9,10].

CONCLUSION

This case is only the fourth intraosseous schwannoma reported at a first intermetatarsal space; which proves extraosseous localization and progressive erosion mechanism. Our goal was to recall as much as possible the possibility of a benign schwannomatous

lesion at this level; But also, the requirement of absolute caution in front of any lesion of the potentially malignant aggressive forefoot.

Competing interests

The authors declare no competing interest.

Authors' contributions

All authors contributed to the conduct of this work. All authors also claim to have read and approved the final manuscript.

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