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Solitary enchondroma in a metatarsal bone, an incidental discovery

Salomon Jasqui Remba[a,b,*,1], Raul Álvarez San Martín[a], Isaac Baley Amiga[a], Daniel Portman Santos[a]

[a] Office 073, ABC Medical Center, Mexico City, Mexico
[b] Office 165, Hospital Ángeles Lomas, Mexico

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ABSTRACT

INTRODUCTION: Enchondromas are rare benign bone tumours and their discovery is usually incidental, since in most cases it presents with no symptoms.
PRESENTATION OF CASE: A 30 year old male who comes to the office after suffering a forced inversion of the left foot, he has edema of the left lateral malleolus, pain and is unable to walk; no deformities or other alterations are observed and palpable pulses of adequate intensity and frequency are recorded. The radiography shows soft tissue edema and no bone lesions.

A tumour is found in the first metatarsal, which was afterwards corroborated by an MRI; once the diagnosis was confirmed, curettage of the lesion was performed with good outcome.
DISCUSSION/CONCLUSION: Multiple enchondromas and solitary enchondromas, left untreated, can transform into malignant bone tumours like chondrosarcomas, so early detection and treatment, in most cases with lesion curettage and bone grafting, become fundamental for a good prognosis.

1. Introduction

Benign bone tumours on the lower extremities have an incidence of less than 37% of the cases and more specifically between 1–5% when on the foot [1,2]. They are usually found incidentally or when there is already a malignant secondary tumour. Tumours can be divided into numerous types and subtypes; the most important classification is the one that separates them between benign and malignant tumours. This paper will specifically address the benign ones, which can be subdivided into osteomas, osteochondromas, osteoblastomas, osteoid osteomas, aneurysmal bone cysts, fibrous dysplasia, giant cell tumours and enchondromas. Each one of them has specific characteristics and will have different treatments and prognosis [3].

The relevance of this case relies on it being of great importance for physicians to know about this pathology and on the fact that it can cause severe complications when it is not found and treated on time.

We hope this case report helps to contribute to further investigation on the subject and to encourage medical staff to perform follow up studies, such as x-rays, to any patient who could present any remote indication of this clinical entity.

The elaboration of this case report has been reported in line with the SCARE 2020 guidelines, making sure it is compliant with all of the criteria of the SCARE 2020 Checklist [4].

2. Case presentation

A 30-year-old male patient comes to the office, by himself, for a consultation, after suffering a forced inversion of the left foot. The Patient doesn't have any relevant medical history, there is no apparent family history of relevance and he is not on any current medicactions. On physical examination, he presents edema of the left lateral malleolus, pain on mobility and walking, no deformities or other alterations and palpable pulses of adequate intensity and frequency. Imaging studies are ordered for exploration; the x-ray shows soft tissue edema and no bone lesions; a tumour is found in the first metatarsal (Fig. 1). Magnetic resonance imaging presents images of the tumour in addition to bone cortical injury due to the tumour's growth (Figs. 2 and 3).

2.1. Investigation

Foot and ankle tumours are rare, they are found only in 1–5% of the tumours of the extremities, the majority of these are usually benign and behave differently from other bone tumours [2,5]. These tumours are commonly misdiagnosed since foot tumours present symptoms that simulate other diseases [6].

Different types of benign bone tumours are described in the literature, finding the following incidence: osteochondroma 30%,

* Corresponding author at: Office 073, ABC Medical Center, Mexico City, Mexico. Office 165, Hospital Ángeles Lomas, Mexico.
E-mail address: salojasquiortopedia@gmail.com (S. Jasqui Remba).
[1] http://ortopediaespecializada.com/.

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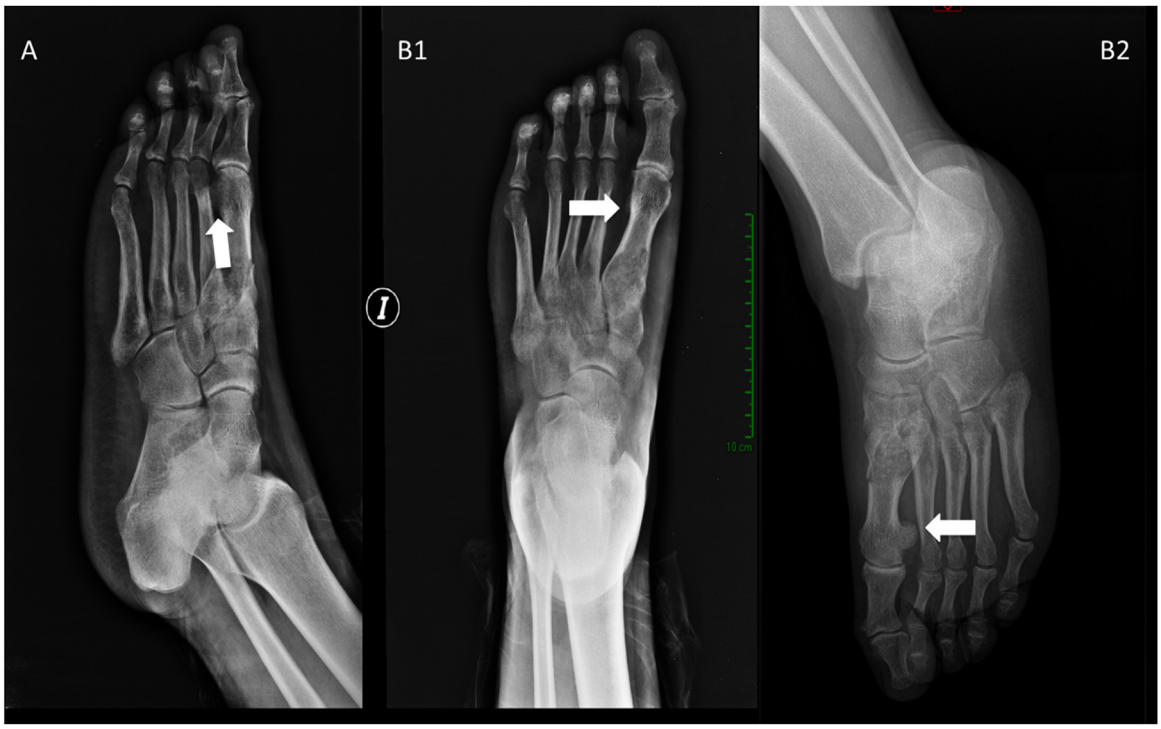

Fig. 1. Oblique (A) and anteroposterior (B1–B2) radiographs of the left foot shows a tumor in the first metatarsal (arrow).

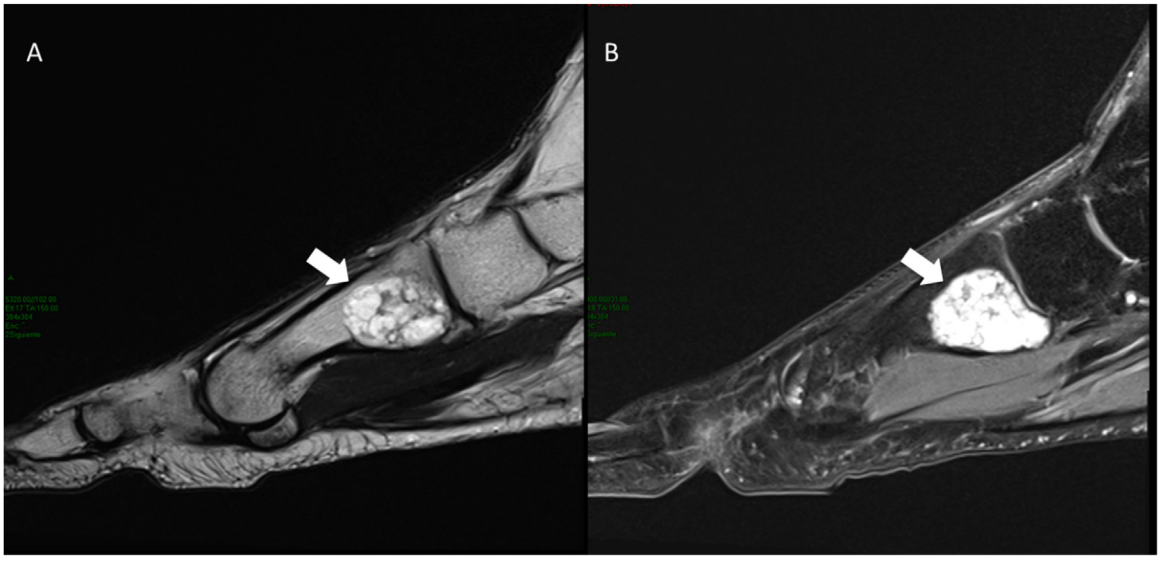

Fig. 2. MRI with gadolinium of the left foot shows in sagittal section in the first metatarsal at the level of the base and the proximal diaphysis heterogeneous intramedullary lesion, hypointense in T1 (A) and hyperintense in T2 (B) (arrow).

osteoblastoma 14%, osteoid osteoma 12%, aneurysmal bone cyst 9.1%, fibrous dysplasia 5–7%, giant cell tumour 20% and enchondroma 2.6% [3].

In this paper we will discuss specifically the enchondroma, since this was the histopathological diagnosis the patient received.

Enchondromas are very rare benign bone tumours; only 2.6% of all benign bone tumours are enchondromas and they are usually asymptomatic. The typical age of appearance is between 10 and 39 years of age, commonly found in hands and feet [3].

The average size is about 3 cm and they are in most cases benign; in the rare case that an enchondroma is found to be bigger than 5 cm, we should consider a low-grade malignancy [7].

Enchondromas are found in the central medullar canal of the metaphysis of the bone and are caused by an abnormal growth of benign rests of cartilage that will be displaced from the growth plate [7].

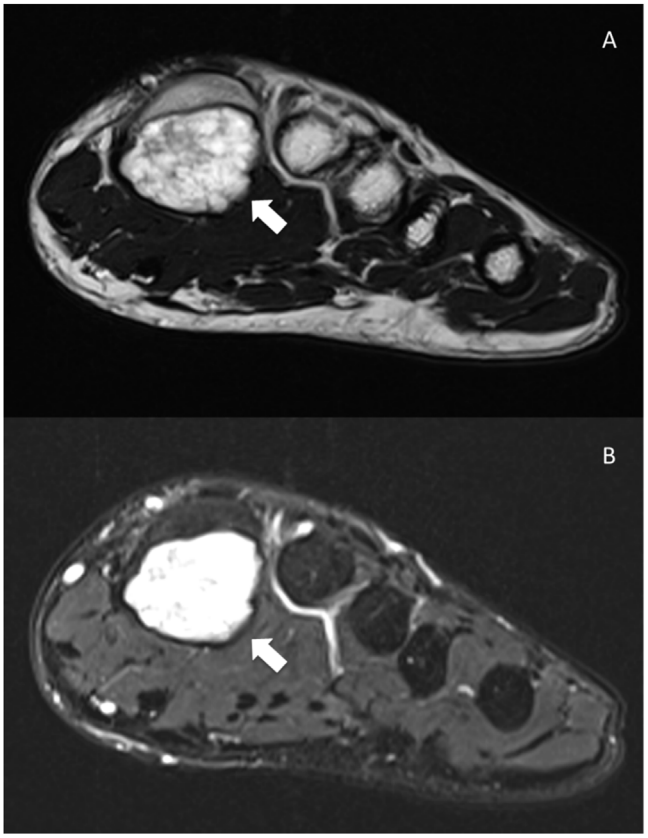

Fig. 3. MRI with gadolinium of the left foot shows in coronal section in the first metatarsal at the level of the base and the proximal diaphysis heterogeneous intramedullary lesion, hypointense in T1 (A) and hyperintense in T2 (B) (arrow).

These tumours are usually found as solitary lesions. However, there is a clinical entity called Ollier's Disease in which we can find multiple enchondromas, also called enchondromatosis [3].

Although enchondromas by themselves are usually asymptomatic, they can cause several complications, like pathological fractures and malignant degeneration [7].

Malignant transformation to chondrosarcoma statistics are not very accurate, since most of them are found as a secondary chondrosarcoma, thus finding the primary asymptomatic enchondroma only after the chondrosarcoma was symptomatic. Nevertheless, in most of the cases where the enchondroma had transformed into a malignant tumour, the patient had presented vague symptoms, such as night pain and resting pain, which motivated the patient or his physician to perform an x-ray, thus finding the tumour [8].

Solitary enchondromas are believed to be incidental tumours, with no genetic association; however, enchondromatosis or multiple enchondromas have been found in more than one member of the same family, suggesting some genetic association. There has been some research on PTHR1 mutations, but none of the studies have been conclusive. There are other studies that are relating mutations in the gene encoding isocitrate dehydrogenase 1 (IDH1) and IDH2 with solitary cartilaginous tumours, but these are still inconclusive and have a lot of research pending [8].

2.2. Differential diagnosis

Differential diagnosis must include all bone tumours; however, the most important differentiation has to be made between enchondroma and chondrosarcoma, given that the latter is malignant. According to Vanel et al., enchondromas are composed by "islands of intramedullary hyaline cartilage surrounded by marrow fat, and a chondrosarcoma, a diffuse cartilaginous replacement (invasion) of the marrow which leads to complete 'trapping' of host lamellar bone trabeculae"; he proposes that the marrow around the cartilage should be visible in an Magnetic Resonance Image (MRI). In his study we can find that in some cases this is true; however, not enough to make it a gold standard and not enough to take the risk of not performing surgery to obtain a histopathological diagnosis [9].

2.3. Treatment

The physician, an orthopedic surgeon, with a sub-specialty in articular replacement, decided to perform surgery in which the following was done: A biopsy was taken with a fluoroscope on the proximal, distal, anterior and posterior borders of the tumour. Also, a percutaneous biopsy was done, obtaining a tissue cylinder, which in the trans-operative pathology exam was identified as a tumour with a chondroid aspect, without malignancy data.

During open surgery a bone window was performed -observing chondroid tissue in the region of the bone lesion- as well as curettage and surgical washing with a solution. Control of surgical edges was achieved with radiofrequency. A sample from the left iliac crest was taken for stem cell sampling.

The bone graft was bathed in chips; it was morselized and combined with demineralized bone matrix. A paste construct was made and placed at the site of bone loss of the tumour, that zone already having undergone curettage and edge control. A left suropodalic splint was placed along with Tegaderm in the left anterior superior iliac crest region. The patient was transferred to the recovery room without complications, after surgery, pain was managed with regular NSAID's, without need for any other medications.

3. Results

3.1. Outcome and follow-up

An adequate follow up is fundamental to achieve the best prognosis; in this case, the patient had the suropodalic splint for 6 weeks, there were several routine follow up appointments and x-ray studies were used to verify the absence of the tumour or any other tumour in this site. No pathological signs where found in these x-rays and after rehabilitation the patient was able to walk normally without assistance, so the surgeon discharged the patient with no further complications.

4. Discussion

In order to prevent bone lesions with a bad prognosis, it is fundamental that clinicians around the world focus on preventive medicine and on early detection and diagnosis, making sure we can find benign tumours, like most of the enchondromas, on time, and perform early treatment, in order to prevent malignization and further complications.

There are many tools healthcare professionals can use to detect and diagnose these lesions; the first and most important one will always be a complete and thorough clinical history. Sánchez-Torres LJ and Santos-Hernández M published a very complete article called "The art of bone tumour diagnosis" in which we can see the utility of each of the imaging techniques. They found that while the histopathological study is the gold standard for the specific diagnosis of the bone tumour, the simple radiography is still the gold standard for initial detection, since this is a cheap and simple non invasive method, that is widely available in most clinics and hospitals in the world. Computerized axial Tomography (CAT),

Magnetic Resonance Imaging (MRI), Gammagraphy, Angiography and Positron Emission Tomography (PET) are also valid and useful techniques; however, these are not so easily found and are a lot more expensive; in many cases they are used as a more specific study once we already detected a lesion on a simple radiography or as a follow up method [10].

In this case, the patient was previously asymptomatic, the lesion was detected incidentally by imaging studies after an ankle sprain, and the radiography and the MRI were fundamental tools for the diagnosis of this tumour, since without them the medical team would have never known of it. Afterwards, the histopathological studies were used to decide definitive diagnosis and prognosis, finding that this specific lesion was an enchondroma, allowing the physician to elect the best treatment for this patient.

Herget et al., has performed a literature review about enchondromas, enchondromatosis and the risk of secondary chondrosarcoma; in this review they mention that the enchondroma is usually found incidentally, since in many cases it is completely asymptomatic. They also found that most of these hyaline cartilage forming tumours are found in the diaphysis of long bones and, in many cases, they are found as a single lesion, also known as a solitary enchondroma [8].

As we said before, the most common place to find an enchondroma is on the diaphysis of long bones, but concerning the exact location of the tumour, a study made by Potter et al., found that the proximal part of the humerus is the most common place for single enchondromas to be found, appearing in 30% of the 33 patients studied, followed by the distal part of the femur 18%, the proximal part of the femur 15%, and the proximal part of the tibia 12% [11].

In the case under study here, the patient also presented an asymptomatic tumour, which was found incidentally, and it was found in a metatarsal bone, which is considered a long bone.

Patients with multiple enchondromas or enchondromatosis have usually a worse prognosis that those who present a solitary enchondroma; Verdegaal et al. analysed 161 patients with multiple enchondromas and found that the overall risk of chondrosarcomas development was 40% [12].

This paper's patient only had a single tumour, also called solitary enchondroma and was found and treated on time, which confers a better prognosis.

In the clinical study performed by Potter et al., 79% of the patients were successfully treated with intralesional curettage with bone-grafting of the lesion, finding tumour recurrence in only one patient, attributed to an incomplete curettage of the lesion; the rest of the patients had a more radical resection performed with the same results, the authors of the article conclude the radical interventions were unnecessary [12].

In this revision, the surgeon's team performed intralesional curettage with bone grafting of the lesion, obtaining similar results to the ones reported in Potter et al.'s study.

Bone tumors are found incidentally relatively often, when these are found early, the prognosis tends to be good, the authors of this case report are concerned about the lack of screening tests and methods to detect such pathology on time and prevent it's progression to a malignant tumor or metastasis, the publication of case reports like this one are of great importance for the medical community and have the objective of creating awareness on the risks and incidence of this problem, hopefully inviting physicians to perform more screening tests when there is any kind of suspicion or indication.

Conflicts of interest

We have no conflicts of interest.

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Ethical approval

The study is exempt from ethnical approval.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

This manuscript has been read and approved by all the authors. SJR, RASM, IBA and DPS contributed in the acquisition of the clinic patient data for the case presented, as well as the research of the related literature and the preparation of the manuscripts, DPS and IBA made the design of the study images and finally SJR and RASM made the critical revision of the manuscript.

Registration of research studies

Not Applicable.

Guarantor

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Provenance and peer review

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References

[1] Nina C. Lindfors, Ilona Koski, Jouni T. Heikkilä, Kimmo Mattila, Allan J. Aho, A prospective randomized 14-year follow-up study of bioactive glass and autogenous bone as bone graft substitutes in benign bone tumors, J. Biomed. Mater. Res. 94B (1) (2010).
[2] L.R. Ramos Pascua, S. Sánchez Herráez, Ó. Fernández Hernández, A. Sladaña Díaz, Tumores óseos del pie, Rev. Del Pie Tobillo XXV (1) (2011) 6–18.
[3] David N. Hakim, Theo Pelly, Myutan Kulendran, Jochem A. Caris, Benign tumours of the bone: a review, J. Bone Oncol. 4 (2015) 37–41.
[4] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, for the SCARE Group, The SCARE 2020 guideline: updating consensus surgical CAse REport (SCARE) guidelines, Int. J. Surg. 84 (2020) (in press).
[5] S. Boriani, R. Biagni, P. Ruggieri, R. Casadei, N. Fabbri, Tumores Óseos del Pie: Experiencia del Istituto Ortopédico Rizzoli, Revista de Medicina y Cirugía del Pie (1) (1992) 71–81.
[6] Ernesto Andrés Delgado Cedillo, Genaro Rico Martínez, Luis Miguel Linares González, Eréndira Estrada Villaseñor, Saúl Renán León Hernández, Raul Ble Campos, Epidemiología de tumores óseos y partes blandas del pie y tobillo, Acta Ortopéd. Mex. 21 (3) (2007) 144–150.
[7] Kakarla Subbarao, Benign cartilaginous lesions, J. Med. Sci. Res. 1 (1) (2013) 29–36.
[8] G.W. Herget, P. Strohm, C. Rottenburger, U. Kontny, T. Krauss, J. Bohm, N. Sudkamp, M. Uhl, Insights into Enchondroma, Enchondromatosis and the risk of secondary Chondrosarcoma. Review of the literature with an emphasis on the clinical behaviour, radiology, malignant transformation and the follow up, Neoplasma 61 (4) (2014) 365–378.
[9] D. Vanel, J. Kreshak, F. Larousserie, M. Alberghini, J. Mirra, M. De Paolis, P. Picci, Enchondroma vs. chondrosarcoma: a simple, easy-to-use, new magnetic resonance sign, Eur. J. Radiol. 82 (12) (2013) 2154–2160.

[10] L.J. Sánchez-Torres, M. Santos-Hernández, El arte de diagnosticar tumores óseos, Acta Ortopéd. Mex. 26 (1) (2012) 57–65.
[11] Benjamin K. Potter, Brett A. Freedman, Ronald A. Lehman Jr., Scott B. Shawen, Timothy R. Kuklo, Mark D. Murphey, Solitary epiphyseal enchondromas, J. Bone Jt. Surg. 87 (A) (2005) 1551–1560.
[12] Suzan H.M. Verdegaal, Judith V.M.G. Bovée, Twinkal C. Pansuriya, Robert J. Grimer, Harzem Ozger, Paul C. Jutte, Mikel San Julian, David J. Biau, Ingrid C.Mvander Geest, Andreas Leithner, Arne Streitbürger, Frank M. Klenke, Francois G. Gouin, Domenico A. Campanacci, Perrine Marec-Berard, Pancras C.W. Hogendoorn, Ronald Brand, Antonie H.M. Taminiau, Incidence, predictive factors, and prognosis of chondrosarcoma in patients with ollier disease and maffucci syndrome: an international multicenter study of 161 patients, Oncologist 16 (12) (2011) 1771–1779.