Clinical note

Scintigraphic depiction of non-ossifying fibromas and the role of SPECT/CT

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A B S T R A C T

Non-ossifying fibromas (NOF) are a benign entity of the developing bone, relatively common in children and young adults. Their location is most frequently metaphyseal. They are usually asymptomatic (unless associated to a fracture) and have a self-limited behavior, with spontaneous regression through a sclerotic consolidation. Plain X-ray is the main imaging tool for its diagnosis. However, an unclear X-ray may lead to further imaging studies.

We present the case of a 17-year-old male with back pain and lower limb dysmetria referred for a bone scintigraphy to complete the diagnostic and assess disease extension and the subsequent MRI evaluation.

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Representación gammagráfica de fibromas no osificantes y el rol de SPECT/CT

R E S U M E N

Los fibromas no osificantes (NOF) son entidades benignas del hueso en desarrollo, relativamente frecuentes en niños y adultos jóvenes. Su localización más habitual es la metáfisis de los huesos largos, suelen ser asintomáticos (excepto si se asocian a una fractura) y normalmente se autolimitan, regresando espontáneamente mediante una consolidación esclerosante. La radiografía simple es la principal herramienta para su diagnóstico. Sin embargo, una radiografía dudosa puede llevar a la realización de otras pruebas de imagen.

Presentamos el caso de un chico de 17 años con dolor de espalda y dismetría de miembros inferiores, remitido para la realización de una gammagrafía ósea con la finalidad de completar el diagnóstico y evaluar la extensión de la afectación, y la posterior valoración mediante RM.

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Introduction

Non-ossifying fibromas (NOF) are benign fibrous cortical defects of the developing skeleton occurring in 30–40% of all children and young adults. They are usually well-defined asymptomatic lesions involving the metaphysis of long bones, especially in the lower limbs, and frequently become a happenstance diagnostic.1,2 Plain X-ray findings are characteristic and may establish the definitive diagnosis.3,4 However, when associated to a fatigue-type stress fracture they can cause pain, and its radiographic diagnostic may be more difficult, leading to further imaging investigations.5 As they present typical imaging features, a histologic diagnosis is not necessary, being part of the so-called “leave me alone” lesions.4,5 During their natural evolution they tend to ossify and disappear, making therapy unnecessary, and only large lesions at risk of bone fracture are considered for surgery.3

In the current note the case of a young male who underwent different imaging tests during the diagnostic process of a NOF is presented. One has to bear in mind that probably a proper interpretation of the plain X-ray would have been enough to reach the diagnosis, with no need of a scintigraphic evaluation; however, as it is an uncommon entity in Nuclear Medicine departments, we would like to present this situation to ring a bell to nuclear medicine physicians when facing such findings.

Clinical case

We present the case of a seventeen-year-old male who consulted his general practitioner because of periodic mild mechanic back pain of a few months of evolution that ceased spontaneously without need of drug intake. Physical examination did not show findings at the spine level, but evidenced lower limbs dysmetria and mild pain at palpation of the left tibia. An X-ray of both knees was performed as primary imaging procedure.

Conventional X-ray showed bilateral, cortically based, metaphyso-diaphyseal, lucent lesions (Fig. 1, images A and
B). They had well-defined, sclerotic borders and narrow zone of transition. One could see thick sclerotic septa and no evidence of matrix within them. There were no evidence of cortical discontinuity, periosteal reaction or other signs of aggressivity. Even if these findings are quite indicative that we may be in front of a NOF, the patient was referred to his reference orthopedic surgeon, who requested a bone scintigraphy and an MRI for a better characterization of the lesions, and evaluation of disease extension, as occasionally multiple NOF can coexist in the same patient.

Scintigraphic blood-pool images of the knees 5 min post-injection of 1110 MBq (30 mCi) of 99mTc-hydroxymethylene diphosphonate (HMDP) showed mild hyperemia proximally in both tibias, more evident in the left one (Fig. 2, image A). Static late images 2 h post-injection (Fig. 2, image B) showed a moderate uptake of radiotracer at the medial margin of the left tibia, and much lighter laterally at the right one. Whole body scan 2.5 h post-injection did not demonstrate other significant pathologic uptake. For a better characterization of tibial lesions, a SPECT/CT was performed 3 h post-injection, proving intraosseous asymmetrical lytic lesions in the posterior margin of both tibias, apparently depending on the cortical bone and extending into the medullary cavity, with a sclerotic rim, internal septa and focal cortex involvement. Left lesion was of bigger entity (Fig. 3, images A and B).

Scintigraphic findings, together with the X-ray, patient’s age and the clinical behavior of the lesions, were compatible with a bilateral NOF. However, an MRI of both knees was performed, proving well-demarcated bilateral lesions with global low T1 and intermediate-high T2 signal (Fig. 4, images A and B). A heterogeneous center was

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Fig. 1. Conventional X-ray. Anterior view of the right leg (image A) and the left leg (image B). One can see a manifest metaphyso-diaphyseal lesion in image B, showing a lucent center with sclerotic borders. The lesion is less obvious in image A, evidencing only a small area of radiopacity at the lateral metaphyso-diaphyseal margin of the right tibia.

Fig. 2. Planar scintigraphic selective acquisition of both knees, anterior view. Asterisk is always indicating right side. Blood-pool images (A) prove a proximal area of mild hyperemia in the left tibia, while late images (B) evidence radiotracer uptake of moderate intensity in this location and also a much slighter uptake laterally in the proximal right tibia. These findings indicate that the left lesion is moderately active, probably already in an ossifying phase as shown by the uptake in late images. The lesion in the right leg does not prove clear hyperemia, which could be explained because of lower ossifying activity.
evident in the biggest lesion, and all sequences showed a peripheral low signal rim corresponding to the sclerotic border.

All imaging procedures performed depicted characteristic features of NOF with apparently no risk of fracture. As previously introduced, no treatment was necessary and only recommendations such as avoiding certain sports that could favor a fatigue-type stress fracture were given to the patient. One year after diagnosis, the patient remains asymptomatic. A follow-up X-ray and a new bone scintigraphy have been performed, showing no changes with respect to the diagnostic ones.

Discussion

In few occasions NOF will reach a Nuclear Medicine department, as most frequently they are diagnosed in the basis of characteristic X-ray findings. Nevertheless, in the context of a young patient referred for scintigraphic characterization of long bone lesions without a clinical suspicion of malignancy, one has to consider NOF among the differential diagnosis.

As previously mentioned, NOF are benign developmental cortical defects usually involving the metaphysis of long bones. In our patient they were placed in the proximal tibia, one of the most frequent locations. They are often asymptomatic, even if they can become painful when associated to a pathologic fracture, commonly related to bone insufficiency, especially in larger lesions.

In the series by Shimal et al., 6% of all stress fractures had an underlying NOF, and Blaž et al. described a 3.5% of stress fractures in a group of patients with NOF and fibrous cortical defects (FCD).

It is important to distinguish NOF from FCD, as their target population and involved bones may be the same. In fact, histologic appearance of both entities is mainly identical. However, NOF tend to be of bigger size, even mimicking a fibrous dysplasia in X-ray and bone scintigraphy, show expanded cortices, may protrude into the medullar cavity, and more frequently can be symptomatic.

Bone scintigraphy is a sensitive technique to detect such bone lesions, especially if there is an associated stress fracture in early stages, but it is much less specific being more frequently used to assess disease extension, as occasionally multiple NOF can coexist in the same patient. Multiple NOF have been described in the Jaffe-Campanacci syndrome, even if they tend to be of larger size and number, and associated to extraskelatal manifestations (as neurofibromas and café-au-lait skin spots), which is not the case of our patient.

Radiotracer accumulation may reflect the biological activity of NOF, depending on its developmental stage. In our patient, mild hyperemia and moderate bone uptake suggest a healing phase, in opposition to higher hyperemia and uptake (which may indicate a more active process or a fracture), and to normal scintigraphic

Fig. 3. Axial (A) and coronal (B) slices of the SPECT/CT acquisition. Well-defined lytic lesions with sclerotic margins are evident posteriorly in both tibias, smaller and lateral in the right one, and more significant and medially based in the left side. Both present internal septa and focal cortical involvement.

Fig. 4. MRI study, saggital view of right (A) and left (B) knees, T2-weighted with fat saturation sequence. Both images reflect intermediate-high signal lesions involving the posterior border of the tibias, with a well-demarcated low signal periphery corresponding to the sclerotic rim.
appearance (frequent in quiescent and healed lesions, both without clinical significance).

SPECT/CT is a useful complementary tool for bone scans, increasing its specificity and allowing a more accurate diagnosis.9,10 As reported by Gayed et al.,9 diagnostic certainty of bone scans in the study of lower extremities’ abnormalities improved significantly with the addition of SPECT/CT, leading to a better localization and diagnosis in 30% of the cases. Its lytic appearance at the CT sequence, sometimes with punctual cortical involvement, may suggest a malignant diagnosis at first sight, even if the well-defined sclerotic borders together with the radiotracer uptake and its typical location in a young patient may raise the diagnosis of a possible NOF.

MRI is not as specific as conventional X-ray or CT and it can bring up the differential diagnosis with other benign entities (frequently with enchondroma, but also with the rarer chondromixoid fibroma). As we can deduce from its frequent regression, when NOF mature they begin to ossify and the signal becomes low in all sequences.

Summarizing, NOF is a benign bony defect relatively common in children and young adults, which diagnosis can be reached with a plain X-ray. However, occasionally such entities may arrive at a Nuclear Medicine department, so one must know that its radiotracer uptake degree depends on the developmental stage of the lesion and that they can present in multiple locations. Despite bone scintigraphy has low specificity in this situation, the addition of a SPECT/CT may improve its diagnostic performance.

Conflicts of interest

There are no conflicts of interest.

References