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CASE REPORT

Bizarre parosteal osteochondromatous proliferation (Nora's lesion) of the forefoot

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Nora's tumour is an uncommon benign tumour, involving mostly tubular bones of hands and feet, and characterized by a proliferation of chondroid, bony and fibrous tissues. Main differential diagnoses are osteochondroma, chondrosarcoma, and osteosarcoma. The authors report a case involving the left foot. The diagnosis was suspected by imaging characteristic features and was confirmed by histological examination. Treatment was surgical, with complete excision. There are no clinical or radiological signs of recurrence on further review one year postoperatively.

Keywords : Nora's lesion ; osteochondroma ; tumour.

INTRODUCTION

Bizarre parosteal osteochondromatous proliferation is a rare tumour, first described In 1983 by Nora *et al* (5) in the hand and foot. This lesion can be confused with other benign and malignant conditions ; therefore, radiological and gross histological appearances must be considered. Despite the high risk of local recurrence, local excision seems to be the treatment modality of choice. Since the original description, this condition has been reported sporadically and all publications are limited to isolated cases or very small series. We report an additional case of Nora's lesion of the lateral aspect of the fifth metatarsal and proximal phalanx of the little toe.

CASE REPORT

A 42-year-old male presented with a 2 years history of a progressive growing tumour at the plantar aspect of his left foot, with footwear discomfort ; there was no history of trauma.

Clinical examination revealed a tender non-mobile and remarkably hard mass measuring approximately 5 cm × 3 cm, located between the fourth and fifth metatarsals plantarly, and extending laterally to the little toe (fig 1). The overlying skin was intact and not adhering ; the mobility, sensibility and vascularity of the lateral toes were normal.

Radiographs (fig 2) and computed tomography scan of the left foot (fig 3) showed soft-tissue swelling with calcifications in the plantar region of the fourth and fifth metatarsals web space, and on the lateral aspect of the fifth metatarsal and

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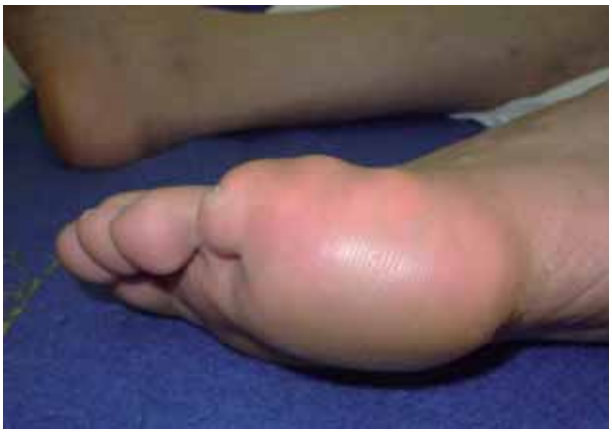


Fig. 1. — Aspect on clinical examination

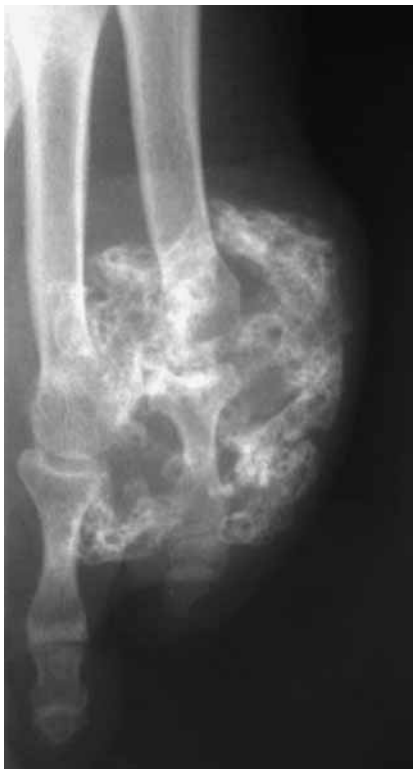


Fig. 2. — Dorsoplantar plain radiograph of the left foot demonstrating the tumour between the fourth and fifth metatarsals, with no evidence of bony erosion of the cortices.

proximal phalanx of the little toe, without cortical irregularity. A diagnosis of ectopic calcification or tumorous process was made.

Magnetic resonance imaging of the foot showed a soft-tissue mass, arising from the lateral aspect

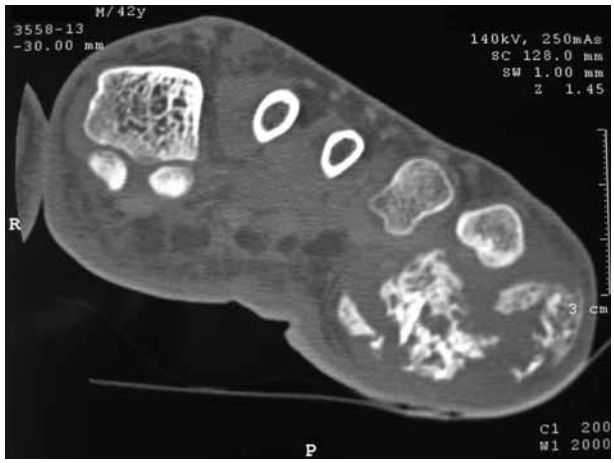


Fig. 3. — Computed tomography scan showing the lesion between the fourth and fifth metatarsals (coronal view).

of the fifth metatarsal and proximal phalanx measuring $50 \times 40 \times 30$ mm, with low signal intensity on T1 weighted imaging ; the adjacent cortices were not violated and tumour margins were well defined (figs 4A,4B).

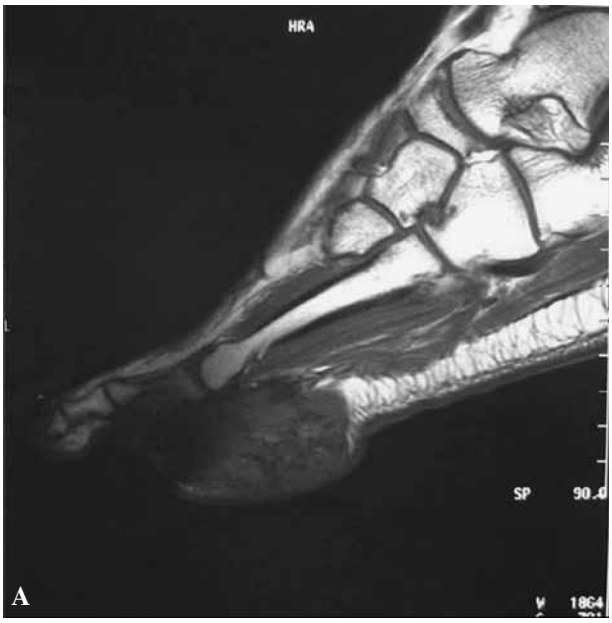
Routine laboratory determinations were normal.

A wide open excision biopsy was performed, through a longitudinal incision on the lateral side of the left foot over the fifth metatarsal bone ; the mass was found to be multilobulated with additional portions extending laterally and plantarly to the proximal phalanx of the fifth toe ; it was easily removed with the pseudocapsule, as there was no adherence to any of the surrounding bony or soft tissue structures (fig 5). Histopathological analysis confirmed a disorganized proliferation of fibrous tissue, cartilage and bone, with the cartilaginous component showing at places irregular groups of enlarged binucleated and 'bizarre' chondrocytes (fig 6). The clinical and histopathological features of the tumour were consistent with a bizarre parosteal osteochondromatous proliferation.

There are no clinical or radiological signs of recurrence two years post-operatively.

DISCUSSION

Bizarre parosteal osteochondromatous proliferation (BPOP) was first described in 1983 by Nora *et al* in a report of 35 cases (*8*) ; since the study of



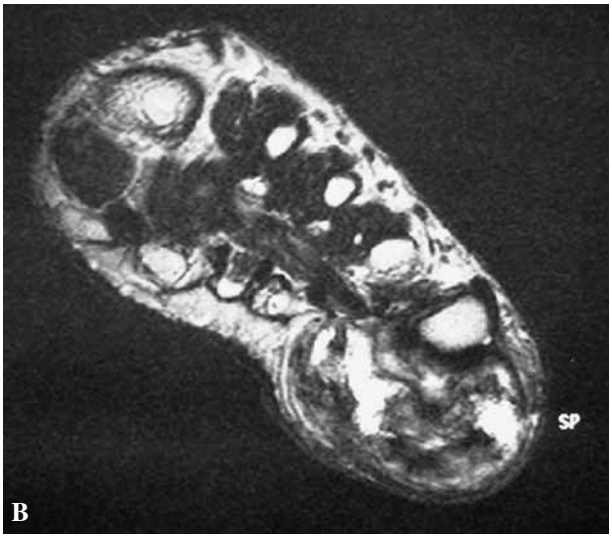


Fig. 4. — MRI showing a heterogeneously low signal intensity mass on T1 weighted imaging, with no evidence of cortical flaring, or adjacent soft tissue swelling. A : axial view ; B : sagittal view.

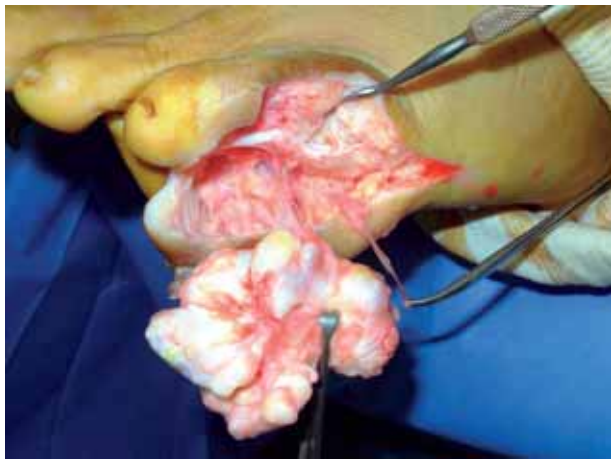


Fig. 5. — The excised lesion (5 cm / 4 cm / 3 cm), with a fibrocartilaginous outer capsule easily dissected from the surrounding bone and soft tissue structures.

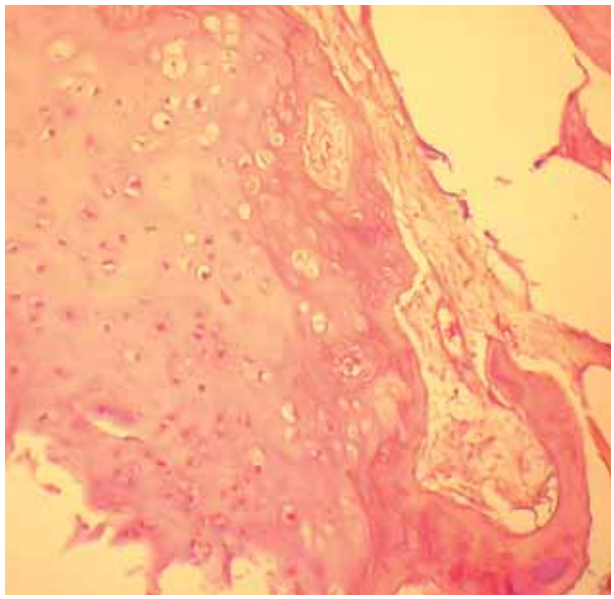


Fig. 6. — Histological findings : presence of disorganized endochondral ossification and irregular cartilaginous proliferation (Magnification × 200).

65 cases reported at the Mayo clinic by Meneses *et al* between 1956 and 1993, this condition has been reported sporadically and all publications are limited to isolated cases or small case series (*3*).

The lesion occurs in all age groups with even frequency in men and women (*4*), and is commonly found on the metacarpals and metatarsals and on the proximal and middle phalanges (*8*).

The cause of Nora's lesions is unknown. It may be related to a reparative process following trauma to the periosteum, as this was noted in 30% of cases in the series of Meneses *et al* (*3,10*).

Grossly the tumour may be confused with osteochondromas, but beside the localisation, some histological differences are apparent (*5,8*). The bony trabeculae are typically covered by a cartilaginous

cap with irregular endochondral ossification (*2,4*). The cartilaginous component shows irregular groups of numerous, bizarre, and enlarged chondrocytes which are commonly binucleated (*8,9*). This contrasts with the more regular alignment of chondrocytes in osteochondromas, and with malignant features such as hyperchromasia and/or cytological atypia noted in any aggressive process such as osteosarcoma or grade-I or II chondrosarcoma (*2,4,6*).

On standard radiographs and computerized tomographs, BPOP is typically well marginated without continuity with the cortex of the medullary canal, like in osteochondromas. The lesion is usually located in the metaphysis, and may exhibit a spiculated or irregular surface leading to a possible misdiagnosis of malignancy such as osteosarcoma. Magnetic resonance imaging (MRI) is required to provide a clearer view (*1,2,4*). The characteristics are clearly evident, the lesion displays heterogeneous low signal intensity on T1 sequences, with absence of marrow involvement of the adjacent bone as well as absence of adjacent soft tissue swelling (*4,7*).

The definitive treatment of this unusual lesion is complete surgical removal ; despite the high recurrence rate, which exceeded 50% within 2 years in the series of Meneses *et al* , and a 20% rate of re-recurrence within 13 years of the second operation, repeat excision seems to be preferrable to more aggressive surgery (*3,7*).

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