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CASE REPORT

Giant enchondroma of the thumb distal phalanx. Presentation of a case and review of the literature☆

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Abstract We report on a case of giant enchondroma of the distal phalanx of the thumb, with a significant degree of clinical and radiographic deformity. It is extremely rare in this location and its treatment should be surgical, with curettage of the lesional cavity and filling with autologous bone graft. In this case, we performed a differential diagnosis of a giant cell tumour and aneurysmatic bone cyst (X-ray and nuclear magnetic resonance [NMR]).

PALABRAS CLAVE
Encondroma;
Tumor óseo de la mano;
Falange distal

Encondroma gigante falange distal del pulgar. A propósito de un caso y revisión de la bibliografía

Resumen Presentamos un caso de encondroma gigante de la falange distal del pulgar, con importante deformidad clínica y radiológica, extremadamente infrecuente en dicha localización y cuyo tratamiento es quirúrgico mediante curetaje de la cavidad lesional y relleno con autoinjerto óseo. En este caso realizamos diagnóstico diferencial con tumor de células gigantes y quiste óseo aneurismático (radiológico y resonancia magnética nuclear).

Introduction

Enchondroma is the most common benign, primary tumour of the hand,[1] especially in the phalanges and metacarpals, and more rarely in the carpal bones. They are generally single tumours (solitary enchondroma), but they occasionally simultaneously affect 2 phalanges or 1 phalanx and the metatarsal of the same digital area. The most frequent location is at the level of the proximal phalanx, followed by the middle phalanx and the metacarpals. The position in

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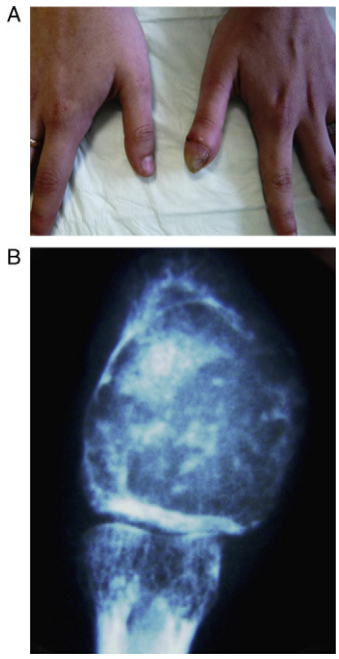

Figure 1 (A) Clinical aspect of the thumb. (B) Radiological image of the tumorous lesion.

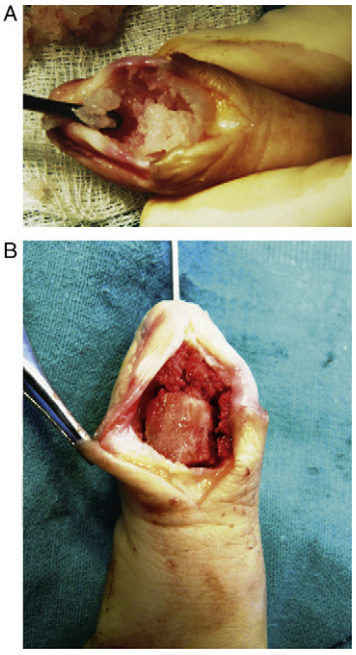

Figure 2 Surgical reconstructive technique. (A) Tumour cavity curettage. (B) Cavity filling with cancellous chips and a structural cortico-cancellous graft. Stabilisation with an axial Kirschner wire.

the distal phalanx is very rare and location in the thumb, exceptional.[2]

The objective of this article is to present a new case of rapidly growing enchondroma of the distal phalanx of the thumb, with clinical and radiological characteristics aggravating its prognosis. We also present the surgical technique performed and tumour development, as well as a review of the literature.

Clinical case

This was a 21-year-old woman, a monitor on transport adapted for the disabled, who came to the consultation due to pain and localised deforming swelling located in the left thumb, of a progressive nature. Clinical examination showed severe deformity of the distal end of the finger, of both the fleshy part and the nail, with global thickening, pain upon local finger palpation but with no limitations in interphalangeal joint function (Fig. 1A). The radiological examination revealed a central lytic image with small calcifications, occupying the entire distal phalanx, and which penetrated the cortical along its entire surface, thinning it, but without structural continuity (Fig. 1B). The patient provided a gammagraphy, which evidenced intense hyperuptake in F2 without affiliated swelling. Nuclear magnetic resonance (NMR) revealed an image compatible with giant cell tumour, making a differential diagnosis with enchondroma. An operation for excision and reconstruction was planned, without being able to eliminate the possibility of amputation given the severe bone deformity. The patient was operated on with regional anaesthesia (axillary block) and preventative ischaemia, preparing the contralateral iliac crest. Using a longitudinal, dorsal approach, after ungual removal, we performed curettage of the entire bone cavity, sending the content for anatomopathological study.

Next, given the cortical weakness after the curettage, we reconstructed the distal phalanx of the thumb using a structural cortico-cancellous graft and cancellous chips. The assembly was stabilised with a Kirschner wire as a guide (Fig. 2A and B). The biopsy confirmed that the tumour was an enchondroma without atypical cells. At 4 weeks, the Kirschner wire was removed and rehabilitation was initiated. We performed clinical and radiological controls periodically, with the last one at 8 months after the surgery. At that time, the patient was asymptomatic, with complete flexion-extension function of the distal phalanx, and the radiological study showed progressive resolution of the tumorous lesion with clear bone remodelling (Fig. 3A–C).

Discussion

Enchondroma is the most common primary tumour of the tubular bones of the hand, developing between the first and fourth decade.[1] In the majority of the cases (75%), they are monostotic or singular (solitary enchondroma).

Their most common position is at the level of the proximal phalanx, followed by the middle and the metacarpals, and they are found more rarely in the carpal bones. At the level of the distal phalanx, such tumours are extremely infrequent, representing 11.4% of the cases,[3] and if we speak of affecting the thumb, they are exceptional (1% of total hand enchondroma).

They present no specific clinical signs and symptoms in the initial stages, and very slight ones in advanced stages. Many cases are discovered in radiological examinations performed for other reasons. In other cases, the patient seeks a consultation after suffering a trauma and the pathological fracture is diagnosed after radiological study. On some

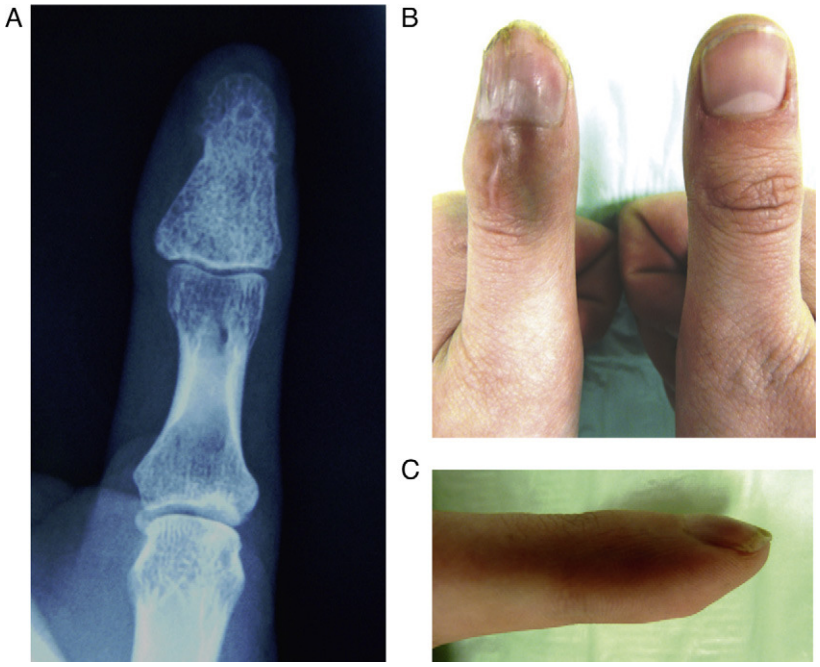

Figure 3 (A) Radiological image at 8 months. (B) and (C) Clinical aspect at 8 months.

occasions, the fracture is accompanied by a bone avulsion of the deep flexor tendon insertion, so active flexion of the distal phalanx is limited, apart from having pain.[4–9] In this case, the strongest clinical manifestation was the progressive, deforming swelling of the distal end of the finger, along with pain, without any prior traumatic antecedents.

The radiological image of the enchondroma is characteristic: a well-defined radiolucent or lytic lesion with small calcifications, the bone appearing penetrated and with the cortical thinned in cases of prolonged development, with a minimal sclerotic halo around it. Takigawa[3] classified the enchondroma according to their position and extension, from the central enchondroma to the eccentric, with cases of very extensive affectation and others of a polycentric nature. Our case is considered gigantic (occupying the entire distal phalanx).

Performing complementary studies, such as bone gammagraphy, computerized axial tomography or NMR, can help to confirm the diagnosis, tumour demarcation and its extension. In our case, the NMR led to the differential diagnosis with giant-cell tumour.

Enchondroma treatment must be considered individually in each case. In initial cases of small extension, a simple curettage with a teaspoon through a cortical window is normally sufficient.[9,10] In intermediate cases of greater extension, the defect created after tumour curettage should be filled with a bone graft[11] extracted from the iliac crest or from the distal end. Exceptionally, in the case of an associated pathological fracture with or without tendon avulsion, fixation with Kirschner wires or a miniplate with screws is needed. However, it must not be forgotten that when an enchondroma in the hand is fractured, its initial treatment should be conservative, because such a fracture makes the enchondroma become ossified; once the fracture consolidates, curettage and graft filling of the remaining enchondroma can be performed to prevent refracture. In the last few years, bone substitutes are being used more and more often to fill the bone defect created. They are very expensive but they simplify the surgery and make it possible to use regional anaesthesia, avoiding hospital admission.[12] In our literature review, we have seen that Wulle[13] utilised plaster in 3 cases. Yamamoto et al.[14] and Bauer et al.[15] use hydroxyapatite and Yasuda et al.[16] injects calcium phosphate cement into the bone defect. In cases of massive affectation of the phalanx or metatarsal, Bikels et al.[17] complements tumour curettage by placing 1 or 2 Kirschner wires inside the centre, filling the cavity with bone cement (PMMA). Finally, the approach used will always be conditioned by the size of the lesion and its position. Either the lateral or palmar approach can be used.[18] However, in our case, given the ungual deformity involved and the greater dorsal affectation, we chose a dorsal approach facilitated by the ungual excision.

Level of evidence

Level IV evidence.

Ethical Disclosures

Protection of human and animal subjects. The authors declare that no experiments were performed on humans or animals for this investigation.

Confidentiality of Data. The authors will declare that they have followed the protocols of their work centre on the publication of patient data and that all the patients included in the study have received sufficient information and have given their informed consent in writing to participate in that study.

Right to privacy and informed consent. The authors must have obtained the informed consent of the patients and /or subjects mentioned in the article. The author for correspondence must be in possession of this document.

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