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Metaphyso-Diaphyseal Chondroblastoma of the Humerus: About Two Cases

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Abstract **Case Report**

Chondroblastoma accounts for less than 2% of all bone tumors, it is a tumor of benign cartilage origin, it is typically located at the level of the epiphysis or process of a long bone. Most often located in the proximal humerus, the distal femur, the proximal tibia, it more rarely affects the tarsal bones or the pelvis. We report 2 cases of young patients (aged 14 years and 15 years) with metaphysodiaphyseal chondroblastoma of the right humerus which was revealed by painful shoulder swelling and joint limitation. Our goal is to discuss the diagnostic difficulties and evolutionary aspects of this rare tumor as well as its treatment.

Keywords: Chondroblastoma, Diagnosis, Evolution, Treatment.

1. INTRODUCTION

Chondroblastoma is a rare benign tumor that accounts for less than 2% of all primary bone tumors [1]. It usually occurs in the epiphysis of the long bones, its diagnosis is pathological after radiological assessment dominated by cross-sectional imaging, and is treated by curettage and filling, or even bone resection with or without replacement. The postoperative recurrence rate has been described as between 10% and 32% [1]. Although controversial, there are rare reports of malignant chondroblastoma in the literature. Many authors explain cases of malignant chondroblastoma as an entity of chondroblastic osteosarcoma that will need to be ruled out by a thorough histopathological study [2]. We present two observations of chondroblastoma of the right proximal humerus, both without recurrence after curettage and bone grafting.

Observation 1:

A 14-year-old girl consults for nocturnal pain, aggravated by the mobilization of the shoulder, evolving for 1 year, the clinical examination finds limited range of motion with abduction 40° passive internal rotation and active external rotation 30° very painful mobilization with swelling of the shoulder, loss of external bony reliefs. Vital signs were normal, laboratory profile normal.

Radiologically: a standard face and profile X-ray show a mixed osteolytic and osteocondensing image with a central and peripheral flaky appearance located at the level of the proximal end of the humerus blowing the cortical which is thinned, without periosteal reaction (fig. 1).

Magnetic resonance imaging (MRI) is performed which shows a tissue mass centered on the inner part of the right humeral head, of the conjugation cartilage and extended to the metaphyso-diaphyseal region, eccentric of polylobed contours, intermediate signal T1 and T2, enhanced in an intense and heterogeneous manner after injection of gadolinium, measuring 96*66*68mm (H*AP*T) (fig.2).

Bone scintigraphy revealed very intense pathological hyperfixation at the level of the tumor process of the proximal end of the right humerus (head and humeral neck), bilateral and symmetrical physiological hyperfixations at the level of the conjugation cartilages (Fig.3).

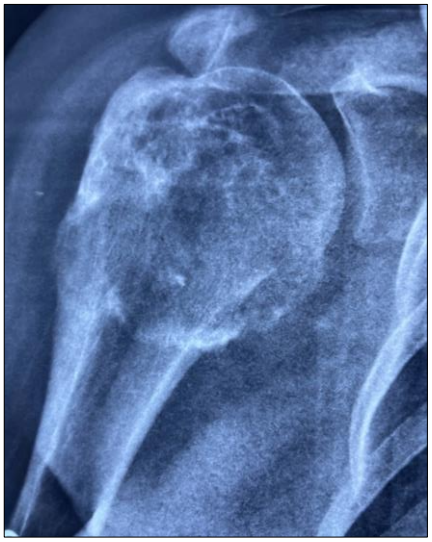

Fig.1: Lytic lesion of the proximal end of the right humerus

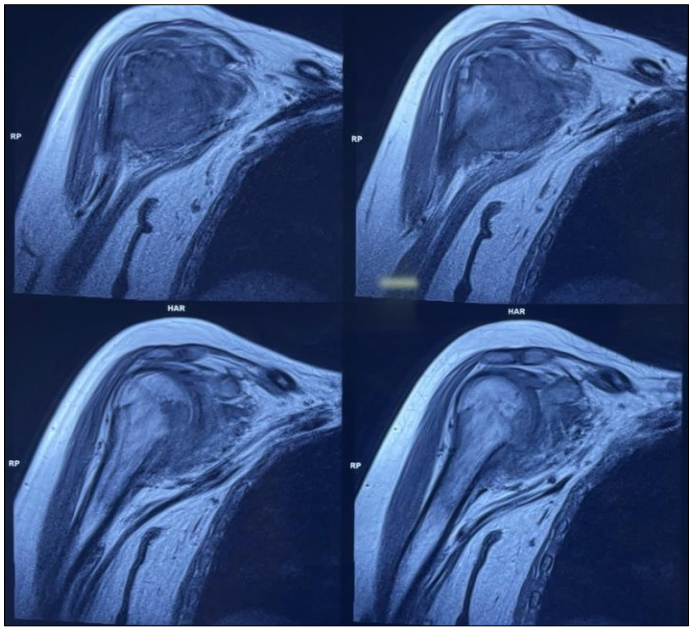

Fig. 2: Tumor process of the proximal end of the right humerus

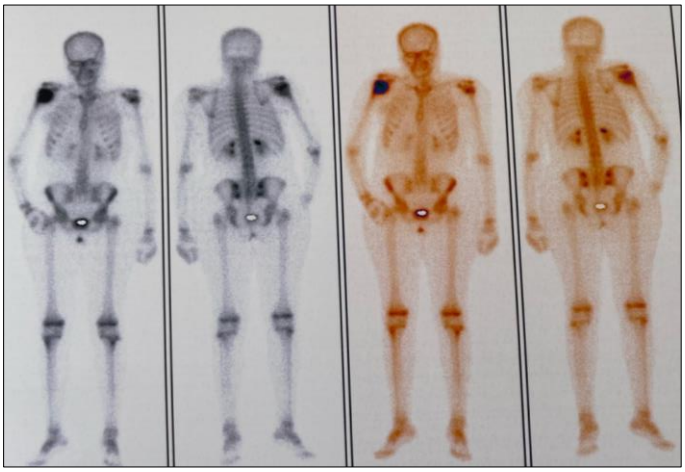

Fig. 3: Hyperfixation at the proximal end of the right humerus

A biopsy is performed by an anterolateral approach at the level of the mass, it has carried the sample of several brownish fragments, the largest of which measures 1.2*0.5 cm. Histopathological examination shows a range proliferation of ovoid cells, with abundant eosinophilic cytoplasm and fine-chromatin, finely nucleolate nucleus, accompanied by numerous osteoclastic multinucleated giant cells. Presence of chondroid tissue foci and hemorrhagic sufficiency. Figures of mitosis are present but not atypical. Morphological appearance is in favor of chondroblastoma.

The extension assessment including scintigraphy, CT and TAP came back in favor of a single proximal humeral lesion without secondary localizations the final diagnosis of chondroblastoma of the proximal end of the humerus is retained based on histological data.

The patient underwent extensive curettage of the entire tumor and a massive bone graft on the removal of an iliac crest graft without operative incident, including joint break-in. The evolution is favorable with improvement in joint amplitudes, the abduction being greater than 100° with a follow-up of 1 year without recurrence.

Observation 2:

A 15-year-old boy, trauma to the right shoulder one year before his first consultation, treated with analgesics, the course is marked by the appearance of a large and painful swelling as well as a joint limitation of the right shoulder, the range of motion being zero. Normal vital signs, normal laboratory workup.

Radiologically: A standard X-ray (Fig.4) shows an osteolytic image of the humeral head. Resonance imaging (MRI) shows a strongly aggressive tumor lesion (Fig.5). Bone scintigraphy shows significant right scapulohumeral hyperfixation extended to the glenoid, acromion and right upper humeral extremity (fig.6).

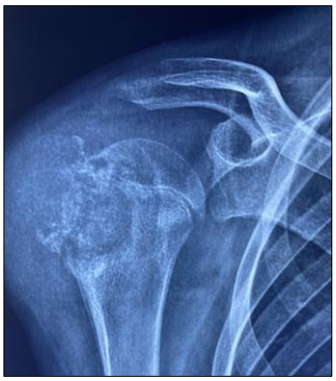

Fig. 4: Osteolytic of the proximal end of the humerus

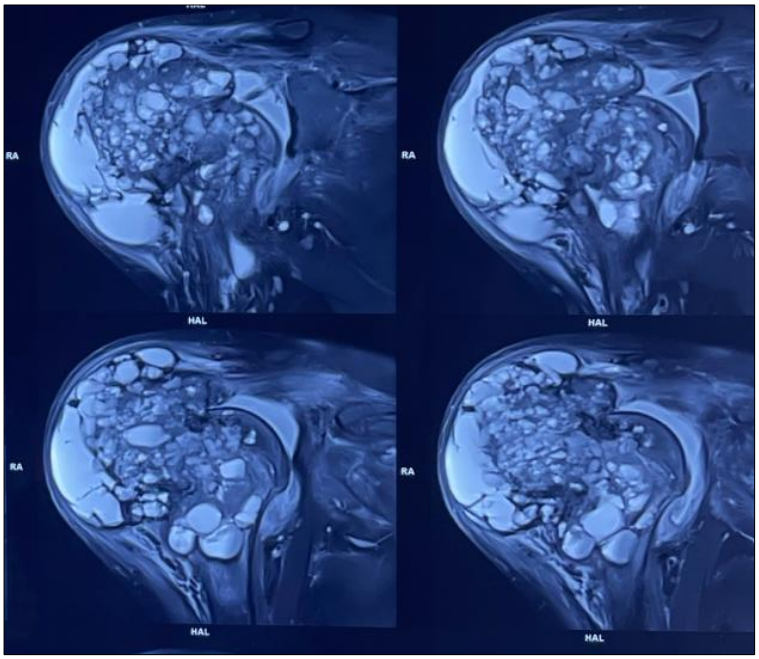

Fig. 5: Aggressive tumor lesion

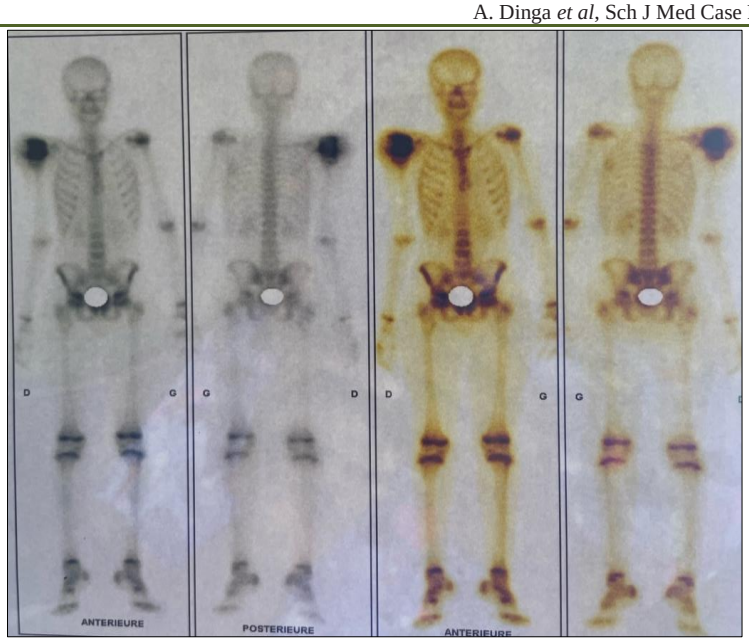

Fig. 6: Right scapulohumeral hyperfixation.

A biopsy was done, histopathological examination found numerous osteoclastic multinucleated giant cells, the morphological appearance was in favor of chondroblastoma; In view of the clinical, radiological, and histological picture, the diagnosis of chondroblastoma is retained. The patient undergoes a curettage of the entire tumor, the residual cavity is filled with surgical cement. A follow-up CT scan (Fig. 7) with a follow-up of 1 year was done objectifying a right humeral metaphyso-diaphyseal lesion with cement-plasty material.

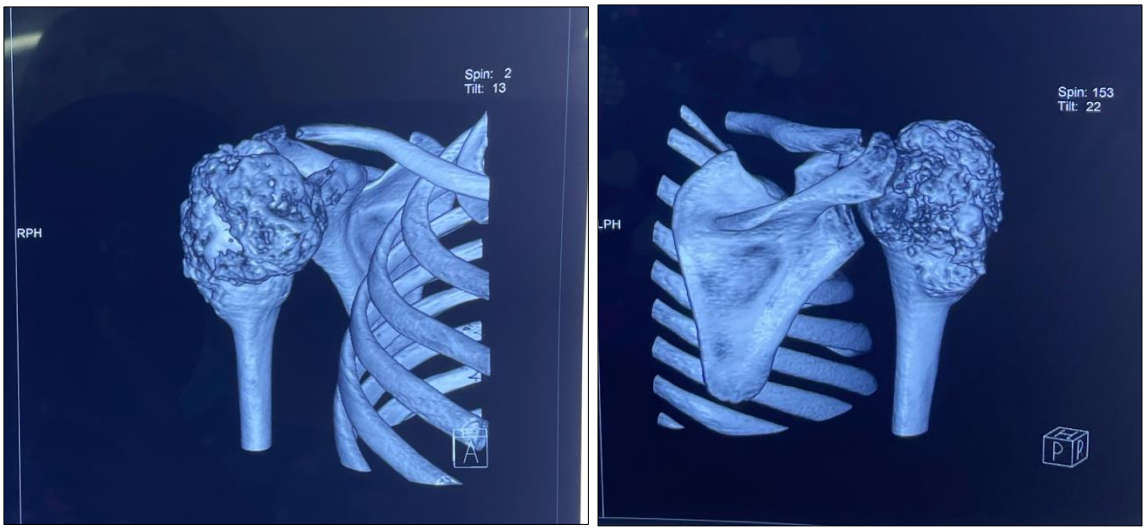

Fig. 7: Right humeral metaphyso-diaphyseal lesion with cementoplasty material.

2. DISCUSSION

Chondroblastomas are rare bone tumors first described by Ewing [4], in 1923. Clinically, the tumor is more common in men, with a wide age range between 10 and 25 years in 85% of cases[5] and may have an indolent course with lung metastases in a minority of reported cases [6].

Several reports of benign chondroblastoma with metastases to the lungs have been described, these metastatic lesions are microscopically indistinguishable from benign chondroblastoma, and to date, there are no studies that definitively identify the risk factors that would predict recurrence or more aggressive behavior of these tumors [3].

However, there are reports demonstrating some association between higher recurrence of these tumours and aneurystic bone cystic changes, tumour location, or duration of symptoms [1].

The tumor has a predilection for the lower limbs, the metatarsal, tibia and femur being the most

frequently involved bones and the epiphysis of long bones, Metaphyseal, diaphyseal or metaphyso-diaphyseal localizations, as in the case we present, are rare. Their incidence varies from 2 to 5% depending on the series (Table I) [Azorin *et al*., [7], Peh *et al*., [8], Fechner and Wilde [9], Aronsohn *et al*., [10], Nimbkar *et al*., [11], Brien *et al*., [12], Sotelo-Avila *et al*., [13], Dwaik and Devlin [14].

The radiological aspect of chondroblastoma usually appears as a well-defined lytic eccentric geographic spinal cord lesion, surrounded by a thin border of condensation and possibly containing areas of punctiform calcifications, thinning or cortical erosion in 75% of cases and a periosteal reaction in 15 to 30% of cases, as in our observations. On MRI, the tumor has an intermediate low intensity signal at T1, an intermediate heterogeneous signal at T2, and is surrounded by intense perilesional edema [15].

Macroscopically, the tumor has a variable appearance. It is most often soft, finely granular, and greyish in color with a few hemorrhagic foci that may correspond to hemorrhagic necrosis or cystic rearrangements such as aneurysmal cysts. It is small to medium in size, between 1.5 and 7 cm. The identification of chondroid foci is inconsistent [Kilpatrick *et al*., [16], Forest and de Pinieux [17].

Histologically, the tumor is highly cellular with sparse intercellular substance. The cells are medium-sized, rounded, or polyhedral, with well-defined cytoplasmic boundaries. Their nuclei are rounded or oval-shaped or kidney-shaped often marked by indentations, cleavages or incisures. Chromatin is finely dispersed with fine nucleoli. The cytoplasm is acidophilic, rich in glycogen. Reactive giant cells with the osteoclast phenotype are present with a highly variable abundance and distribution from one tumor to another. They sometimes predominate in contact with chondroid patches, or in the vicinity of hemorrhagic rearrangements, as in our observation [Kilpatrick *et al*., [16], Forest and de Pinieux [17]. Necrotic rearrangements associated with intercellular calcium deposits form a basophilic mesh very suggestive of chondroblastoma (chicken-wire of the Anglo-Saxons).

The main differential diagnoses are with GCTs (giant cell tumors), clear cell chondrosarcoma and chondroblastic osteosarcoma, the distinction is made on the anatomopathological, immunohistological and genetic aspects.

Table I: Chondroblastomas of metaphyseal, metaphyso-diaphyseal and diaphyseal localization.

Authors (years)	Age	Sexe	Localisation
Dahlin (1972)	13	F	Distal metaphysis of the femur
Fechner (1974)	-	-	Proximal metaphysis of the femur
Schwinn (1976)	14	M	Metaphyseal of the radius
Aronsohn (1976)	18	M	Proximal metaphysis of the femur
Bloem (1985)	25	F	Metaphysis of the humerus
Nimbkar (1980)	-	-	Proximal metaphysis of the femur
Sotelo – Avila (1986)	11	F	Proximal metaphysis and diaphysis of the femur
Ippolito (1986)	15	M	Diaphysis of the first metacarpal
Dwaik (1992)	-	-	Meta diaphysis of the femur
Peh (2000)	13	M	Metaphysis of the thumb
Azorin (2005)	13	F	Distal diaphysis of the femur
Cas clinique K. Znati (2007)	15	F	Distal metadiaphysis of the femur
Notre observation	14 – 15	F – M	Metaphyso-diaphyseal of the humerus

The main treatment modality for the management of chondroblastomas is intralesional curettage and bone grafting, as in the cases we report. In a series reported by Suneja *et al.*, only 7 (13.2%) out of 52 patients had a local recurrence after treatment with intralesional curettage alone [18]. Dahlin and Ivins, in 1972, reported 38 cases of chondroblastomas with 3 recurrences after curettage, which were treated with repeated curettage and bone grafting with or without radiation [19]. The cases presented demonstrate successful treatment using curettage and the use of chemical cauterization (phenol) with grafting In the 1st case, high-speed deburring, phenol, bone cement, and cryosure were recommended to reduce the local recurrence of these tumors [20]. As in our 2nd case where cement was used. (Surgery is the basic treatment in case of doubt, diagnosis with a malignant tumor, cancer resection with replacement is required).

Rybak *et al.*, reported percutaneous radiofrequency ablation as an alternative method to surgery in selected cases [21]. Despite many advances and alternates in the treatment of these lesions, sometimes with multiple recurrences, amputation has also been recommended [22]. Although it is a benign tumor, a few cases of metastases have been reported in the literature. In fact, the prognosis of chondroblastoma is mainly dominated by the risk of local recurrence after curettage-graft. The recidivism rate varies from 5% to 38% depending on the series. These recurrences are often amenable to new curettage-graft therapy, but they are sometimes associated with tumor extension in the joint

or adjacent soft tissue requiring a heavier and more extensive surgical procedure that can compromise the functional prognosis [Accadbled *et al*., [23].

3. CONCLUSION

Chondroblastoma is a benign cartilage tumor most often found in the epiphysis of long bones. Metaphysodiaphyseal localization of the humerus is rare. It can be confusing with malignant tumors in the management, which is more cumbersome. Despite successful initial treatment, patients must be followed postoperatively in the long term due to the risk of local recurrence and joint invasion, which will be responsible for total destruction of the joint.

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