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Case Report

Calcitonin Protocol Therapy for a Treatment of a Bone Giant Cell Tumor in Childhood: A Case Report

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Abstract

Giant cell tumors (GCTs) are rare tumors and represent 5 to 10% of all primary bone tumors. Those tumors are rare before skeletal maturity (1.7-2.5%). They appear as an expansile lytic lesions involving the metaphyso-epiphyseal area of long bones. We report a metaphyso-epiphyseal upper tibia location in a 13-year-old boy treated by curettage followed by calcitonin protocol injections with 13 years follow-up.

Skeleton GCTs are highly uncommon in pediatric age group. Calcitonin therapy for GCTs in children has not been reported in the literature. This protocol can be an interesting alternative treatment for these tumor locations.

Keywords: Giant Cell Tumors; Pediatrics; Calcitonin; Tumor

Introduction

Giant cell tumors (GCTs) are almost benign and usually occur at the epiphyso-metaphyseal level of long bones in young adults. Its location in a child remains exceptional. We report the case of 13-year-old child complained of pain and swelling in the proximal tibia. MRI followed by core surgical biopsy confirmed a giant cell tumor diagnosis which is quite rare in this age group. Many therapeutic procedures were already reported and curettage was the most procedure performed in adult and leads often to tumor recurrence. We report a calcitonin treatment protocol case treating such bone tumor location.

Case Report

We report a case of a 13-year-old boy treated conservatively during two months for a pathological fracture of the upper extremity of the right tibia on a well-defined lytic image, taken for unicameral bone cyst (Figure 1). The evolution was marked by the extension of the lytic image questioning about the initial diagnosis (Figure 2).

The patient was admitted for the evaluation and further management of his complains. There was no soft tissue involvement, although the overlying skin was warm. No appreciable lymphadenopathy was noted.

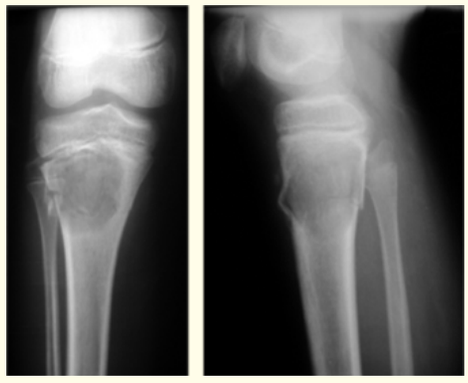

Figure 1: Pathological fracture on expansile lytic lesion.

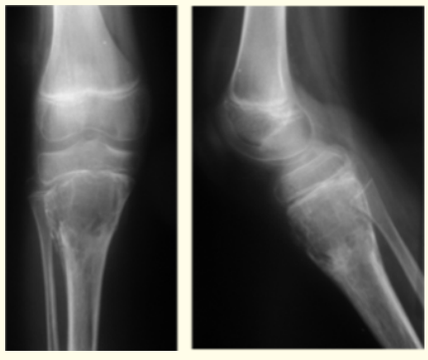

Figure 2: Radiographic aspect after 45 days of cast immobilization.

The CT and MRI confirmed the well-defined aspect and vascularized expansile lesions crossing the physis without surrounding sclerosis (Figure 3). Considering the age of the patient and the radiological appearance, there were multiple differential diagnoses such as osteosarcoma, chondroblastoma, brown tumor, aneurysmal bone cyst and giant cell tumor were kept. The inflammatory assessment was normal.

A GCT diagnosis was confirmed by a surgical biopsy (Figure4). The patient had the calcitonin treatment protocol combining a lesion curettage, intrasite calcitonin injection and external fixation for better stability and skin control (Figure 5).

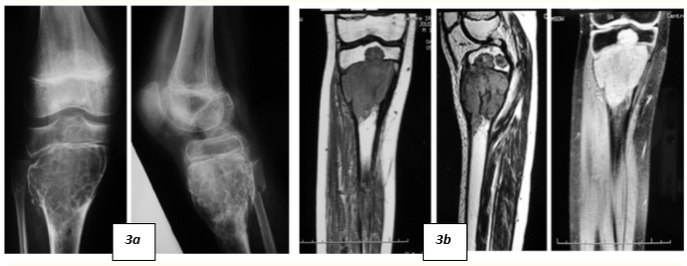

Figure 3: a: X-ray shows an extension of the lytic lesion with thinned cortex. b: MRI (coronal view) shows a sharp well defined lytic lesions extending to epiphysis. The lesion was iso-intense to the surrounding muscle on T1-weighted imaging, heterogeneous high intense on T2-weighted fat-suppression imaging.

Figure 4: Bone destruction by a tumor composed of giant cells admixed with round mononuclear cells in histological and cytological examination (HE stain x 200,x 400).

This therapeutic protocol is compound of four successive steps

- First step include bone lesion curettage made through a cortical fenestration by an osteotome. The tumor was removed with curettes of different sizes. Then the cavity was firstly irrigated with sterile saline then injected with 100 to 200 IU of calcitonin.
- Second step include intramuscular injection of 100 IU of calcitonin each day for 15 days.
- Third step start after healing of the operative scar and consist of a daily intra-lesional calcitonin (100 IU) injection for one month.
- Fourth step include a daily intramuscular calcitonin (100 IU) injection for 2 months.

Figure 5: A: external fixation stabilizing patient knee; B: Radiological aspect during the protocol with no signs of recurrence; C: AP and lateral radiographs obtained at the 1-year follow-up show recurrence

The radiological aspect at 1 year follow-up showed a tumor recurrence with expansile lytic image (Figure 5C) confirmed by a second surgical biopsy. After discussion with a multidisciplinary bone tumor team the patient had another calcitonin treatment protocol. The patient is free of recurrence at 13-year follow-up with good functional outcome. He is currently 26 years old. He developed a physis growth arrest confirmed by the X-ray. He has a 2 cm leg length discrepancy without functional repercussion and full range of motion (Figure 6).

Figure 6: Radiograph after 13 years follow-up show no recurrence with 2cm leg length discrepancy.

Discussion

GCTs represent approximately 15-20% of non malignant bone lesions, with a higher rate of recurrence after curettage alone approaching 50% [1,2]. Those tumors remain rare before skeletal maturity (1.8% to 7.5%) [3]. There is no specific location for the child. Picci [4] reported six cases of giant cell tumors before skeletal maturity in the lower limb: four locations in distal femur, one case in proximal fibula and one location in proximal tibia. Minguella [5] has reported a hand location lesion with damage of the 3rd meta-

carpal. According to some authors an open physis does not prevent GCT from penetrating the epiphysis [6]. For other ones open physis restrict GCT to an almost exclusively metaphyseal location [7]. The aggressive nature of giant cell tumors has been well documented in adults. Similarly in children, Picci [4] noted five aggressive lesions in 6 cases. This same finding was reported by Dahlin [8]. This evolution explains the treatment performed by Picci [4] in two cases: resection of the upper extremity of the tibia and reconstruction with an allograft and knee arthrodesis in the second case. Recommendations for treatment are based on results from the retrospective analyses of non-randomized series from single or multiple institutions [9,10]. Authors advocate intra-lesional excision because it insures local control and maintains function [11,12]. The recommended curettage technique involves the opening of the bone through a large cortical window that allows visualization of the entire tumor cavity [12]. The epiphyseal extension does not allow conservation of the activity of the growth cartilage. Over the years, many different adjuvant therapies have been suggested. The purpose of these modalities is the control of microscopic disease in the reactive zone after curettage has removed the gross tumor. Harris [13] was the first to report the use of human calcitonin as a therapy for central giant cell granuloma (CGCG) of the mandible. Previous studies conclude that the effect of calcitonin on preventing local recurrence seems to be very limited. Therefore, they suggest that filling agents are not required after curettage of GCT [14,15]. Calcitonin therapy for GCTs in children has not been reported in the literature. Referring to our reported case evolution, lesions calcifications seem to be faster than in adults. This protocol was an interesting conservative alternative for the treatment of this localization. Age at diagnosis predicted recurrence of GCT [16]. This hypothesis is supported by studies showing inhibition of bone turnover with bisphosphonates reduced the risk of recurrence of GCT [17]. Previous studies also have shown location gender, tumor grade, and pathologic fracture did not influence recurrence [18]. In our practice, an average of two cures is performed for the treatment of giant cell tumors by the calcitonin protocol. The epiphyseal extension leads to physis damage and growth arrest explaining a 2 cm leg length discrepancy.

The patient is free of recurrence at 13 years follow-up, but a tumor recurrence remains possible. GCTs are exceptional in children. Despite their aggressiveness, the prognosis is generally good. A methodical approach for the purpose of the etiological diagnosis must be undertaken before any pathological fracture.

Conclusion

Although rare in skeletal immature patient, GCTs must be considered as one of the possible differential diagnosis of epiphyo-metaphyseal lesions in pediatric and adolescent population [18]. High rate of recurrence after isolated curettage and associated pathologic fracture constitute a challenge when treating GCTs and must include a multidisciplinary bone tumor team. A meticulous surgical procedure is mandatory in the treatment of GCTs, as in any other musculoskeletal tumor. Treatment depends on tumor location and bone immobilization can be associated to prevent fractures. Calcitonin protocol therapy for GCTs in children has not been reported in the literature. This calcitonin protocol therapy can be an interesting alternative treatment in this location and can be repeated although after initial recurrence.

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