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Giant cell tumor of bone at the proximal epiphysis of humerus in a skeletally immature patient: A case report

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ABSTRACT

INTRODUCTION: Giant cell tumor of bone (GCTB) in skeletally immature (SI) patients are rare benign lesions that have locally aggressive growth pattern and high risk of recurrence. The presence of GCTB at the proximal epiphysis of humerus in SI patients has never been described in literature.
PRESENTATION OF CASE: This report shows the case of a 10-year-old SI male who presented with a GCTB at the proximal epiphysis of humerus that was treated with curettage, cement and adjuvant therapy.
DISCUSSION: The presence of a lytic growing lesion at the proximal humerus in a SI patient should alert clinicians to consider GCTB in their differential diagnosis. The management of GCTB in SI patients is challenging for orthopaedic surgeons. Tumor resection with cementation and adjuvant therapy has been described as a method rationale to prevent the recurrence and preserve the joint function in SI patients with GCTB at the proximal epiphysis of humerus. Clinicians should continue to monitor these patients with radiographic imaging for possible recurrence, metastasis or growth plate injury.
CONCLUSION: Tumor resection with cementation and adjuvant therapy offers a treatment alternative to prevent the recurrence and preserve the joint function in SI patients with GCTB at the proximal epiphysis of humerus. The use of a prothesis in a SI patient should avoided if possible, to prevent implant-related complications and damage to the growth plate.

1. Introduction

Giant cell tumor of bone (GCTB) is a benign, aggressive tumor that occurs mostly at the epiphysis of long bones in skeletally mature patients [1,2]. It is estimated that 80% of GCTB cases occur in patients between 30–50 years of age [3]. The appearance of GCTB counts for less than 6% of all tumors in skeletally immature (SI) patients [2,4]. The metaphyseal area of distal femur, proximal tibia and distal radius are among the most common sites for its appearance [4].

The GCTB at the proximal humerus in SI patients has been reported in two previous cases with the involvement of both metaphyseal and epiphyseal area [4,5]. However, the sole appearance of a GCTB at the proximal epiphysis of humerus in a SI patient has not been reported before. The aim of this report is to illustrate the diagnostic and surgical challenges of GCTB of the proximal epiphysis of humerus in a SI patient.

2. Case Presentation

A 10-year-old male with no past medical nor family history presented with a one-year history of progressive pain, swelling, and reduced range of motion of the left shoulder. Initially, patient started complaining of intermittent shoulder pain with an 8/10 intensity, more prominently at night without preceding trauma nor weight changes. Patient was referred to our pediatric upper extremity clinic after an interventional radiologist performed a percutaneous biopsy that revealed GCTB.

During the physical examination, patient was anxious with left shoulder guarding. The left proximal humerus had a diffuse hard consistency mass with warmth, tenderness, visible vascularity and painful range of motion. Distal pulsation (+2) was palpable with adequate capillary refill. Neurological status was intact with an adequate gross sensation and two-point discrimination, and no paresthesia.

Preoperative radiographs showed an expansive lytic lesion at the left proximal humerus without any “soap bubble” appearance nor any visible fracture or dislocation. See Fig. 1. A magnetic

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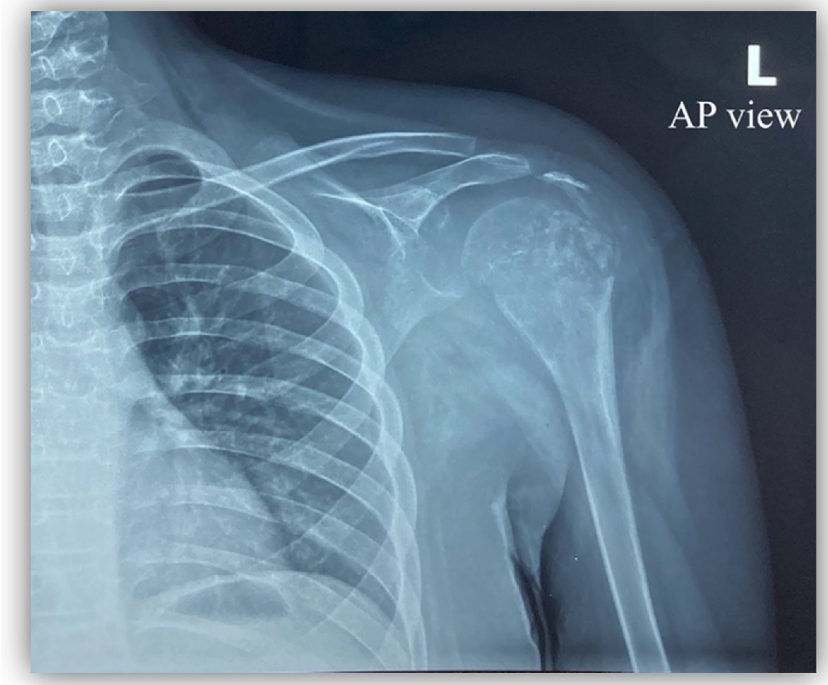

Fig. 1. Anteroposterior view of left shoulder of a skeletally immature patient before surgery.

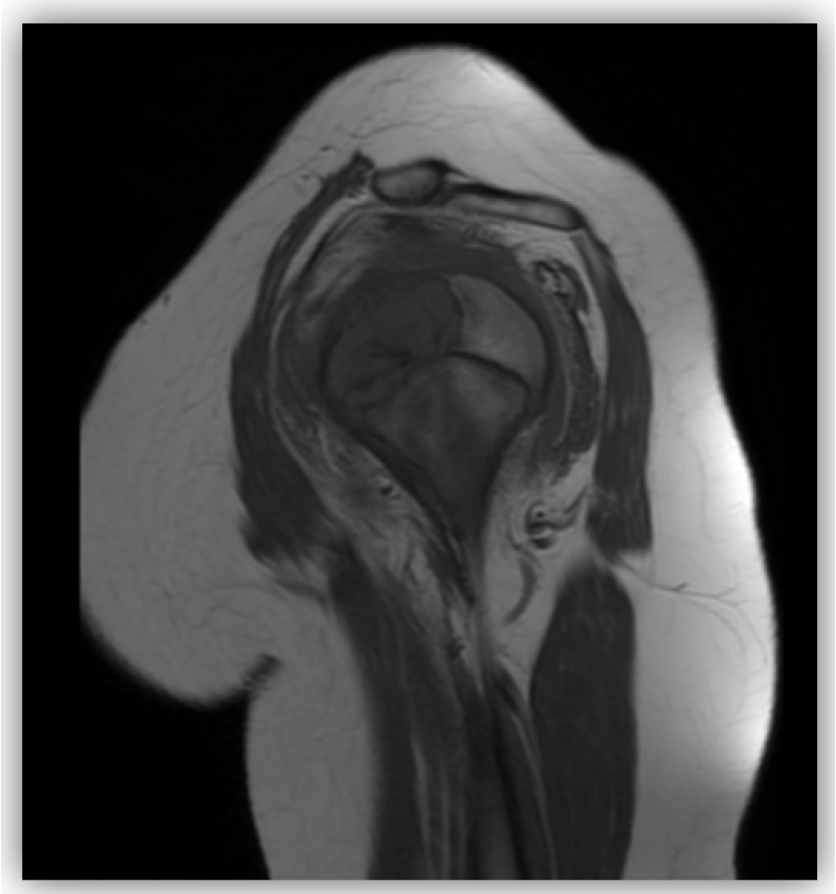

Fig. 2. Magnetic resonance imaging of left shoulder of a skeletally immature patient before surgery.

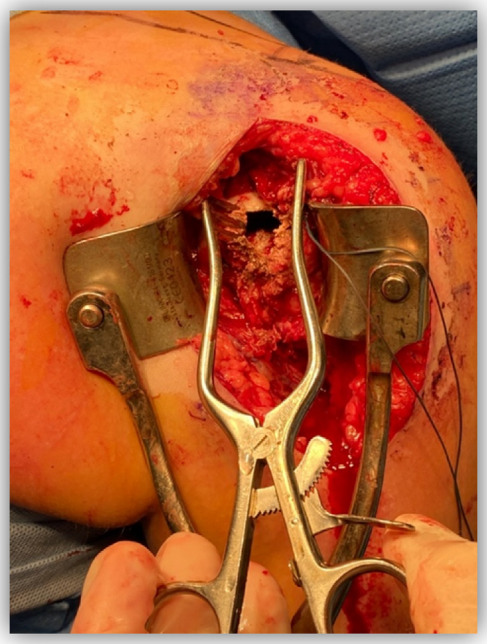

Fig. 3. Left proximal humerus osteotomy of a skeletally immature patient with giant cell tumor of bone.

resonance image (MRI) showed a primary heterogenous lesion surrounding the epiphyseal area of the lateral proximal humerus; with a minor involvement of periarticular subchondral bone, and displacement of the adjacent tendon and muscles. See Fig. 2. Chest radiographs and computerized tomography (CT) scan did not presented any evidence of pulmonary metastasis. Based on the previous biopsy and imaging, surgery was schedule for tumor resection by a pediatric upper extremity orthopaedic surgeon with more than 10 years of experience.

At surgery, a deltopectoral approach was used to demarcate the tumor. A synovectomy and interval split lateral to bicep tendon was performed to access the greater tuberosity. A friable grayish tissue in the periarticular area of subacromial and subdeltoid bursa was seen with intact cortical bone of greater tuberosity. Limited to no extension to metaphyseal was seen with intact subchondral bone at periarticular humeral head. Extensive curettage and bone saucerization of atypical tissue was performed with augmentation therapy of phenol through the bone window. See Fig. 3. Shoulder reconstruction was deferred as the joint appeared to be stable intraoperatively. Proximal humerus cavity was then filled with four milliliters of cement. Wound was closed in layers without complication. See Fig. 4. Finally, the shoulder was immobilized with a Velpeau Arm sling. Histopathological examination showed a red to brown, soft friable tissue (measuring 3.0 × 0.4 × 0.2 cm) with hemosiderin laden mononuclear cells; confirming the diagnosis of GCTB.

During the two-week postoperative visit at the outpatient pediatric orthopaedic clinics, patient started on rehabilitation therapy with active-assisted mobilization. Lastly, the six-month follow up evaluation showed that the patient had no guarding and reported significant pain improvement. Despite a limited improvement of active ROM, full passive ROM was preserved. Follow up radiographs showed filled cavity of epiphysis and subchondral bone of articular humeral head without evidence of recurrence. Patient is currently evaluated every 6 months for recurrence or metastasis of GCTB. See Fig. 5.

This manuscript was structured according to the Updating Consensus Surgical Case Report (SCARE) Guidelines [6].

3. Discussion

This report illustrates the rare appearance of a GCTB of the proximal epiphysis of humerus in a SI patient. Giant cell tumor of bone in SI patients are rare with an incidence that ranges from 1.8% to 5.6% of cases [2]. Most of the GCTB cases reported in SI patients have occurred at the metaphysis of femur, tibia and distal radius in their late teens [7,8].

The appearance of GCTB at the proximal humerus in SI patients have been mentioned in two different reports [4,5]. In 2007, Puri et al. described a seventeen-year-old patient with GCTB of proximal humerus who required an excision and reconstruction with a prothesis [4]. Later on, Jung et al., illustrated the appearance of a multicentric GCTB in a thirteen-year-old who had an initial GCTB at the epiphysis and metaphysis of proximal humerus treated with curettage and cementation [5]. This patient developed a GCTB metastasis at the distal femur and proximal tibia 14 months after initial surgery [5].

Although most of the GCTB are benign, they can be locally aggressive, damaging nearby structures, especially in peri-articular lesions [2–4]. In the past, GCTB was categorized radiographically by the involvement of cortex and the tumor margin definition using the Campanacci grading system, which is now considered less useful for staging [9,10]. Nowadays, clinical evaluation, x-rays and contrast enhanced MRI are the most important imaging modalities to diagnose, perform local staging, evaluate response to systemic treatment, and detect local recurrence in this condition [10]. Additional imaging such as chest x-ray and CT scans are also recommended for detection of possible metastasis [10].

Differential diagnoses can include chondroblastoma, brown tumor of hyperparathyroidism, telangiectatic osteosarcoma among others; based on their radiographic findings [10]. The histologic presentation of GCTB rule out these differential diagnoses; showing benign neoplastic lesions consisting of mononuclear histiocytic cells, multinucleated giant cells that resemble osteoclasts, or neoplastic stromal cells that are the main proliferating cell population [11].

Early management of GCTB is key for joint and growth preservation among SI patients. The ability to destroy bone and metastasize, makes surgery the standard treatment for GCTB [12]. The surgical treatment goals of a GCTB in SI patients is similar to the skeletally mature patients; focusing on tumor removal and preserving adjacent joint function [12]. Depending on the extent, GCTB treatments include curettage with or without cementation, bone grafting or adjuvants, segmental resection and reconstruction with an endoprosthesis [12,13]. Although there is no consensus about GCTB treatment in SI patients, the use of adjuvants has shown to decrease the risk of GCTB recurrence among skeletally mature patients. [7,9,14]. Lastly, the use of neoadjuvant therapies, such as Denosumab, are now starting to become new treatment options for locally advanced GCTB [13]. However, the guidelines of this therapy are currently restricted for skeletally mature patients [13]. In our study, we avoided the use of a prothesis to prevent implant-related complications and damage to the growth plate. Even though we used curettage with cementation and adjuvant therapy (phenol irrigation) based on the surgeon preference, we recognize that there is a risk that our patient will be predispose to a pathologic fracture, growth plate restriction or neurovascular injury [15].

Although the appearance of GCTB at the proximal epiphysis of humerus in a SI patient is rare, it should be considered in the differential diagnosis of any increasing proximal humerus lesion [4,5]. Once the GCTB diagnosis is established, an adequate tumor resection with adjuvant therapy could prevent the recurrence and preserve its joint function. Clinicians should continue to monitor these patients with radiographic imaging for possible recurrence, metastasis or growth plate injury.

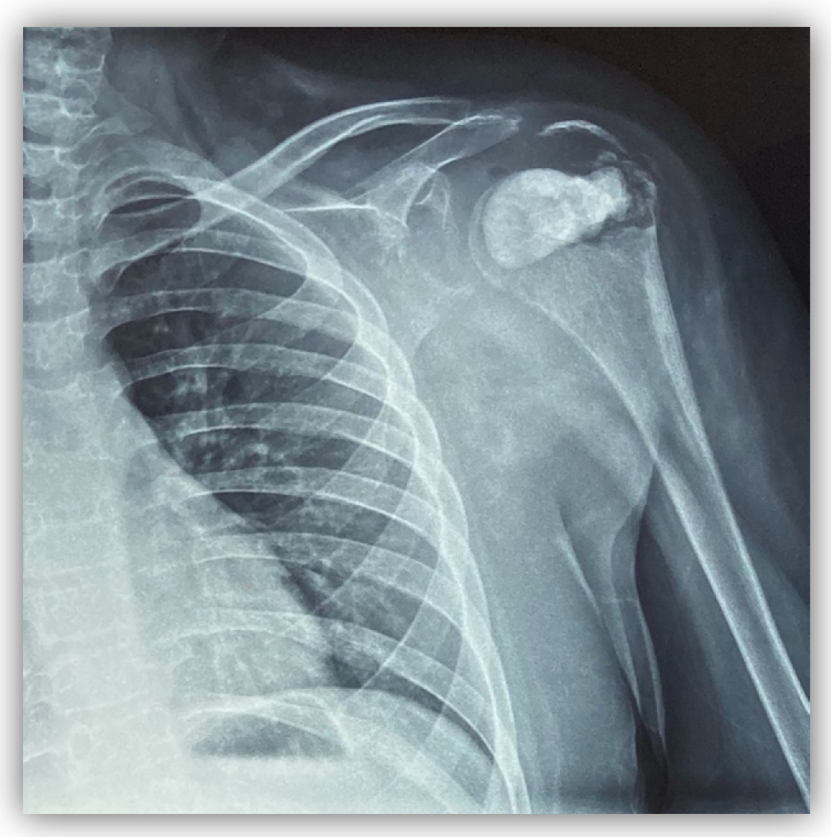

Fig. 4. Anteroposterior view of left shoulder of a skeletally immature patient immediately after surgery.

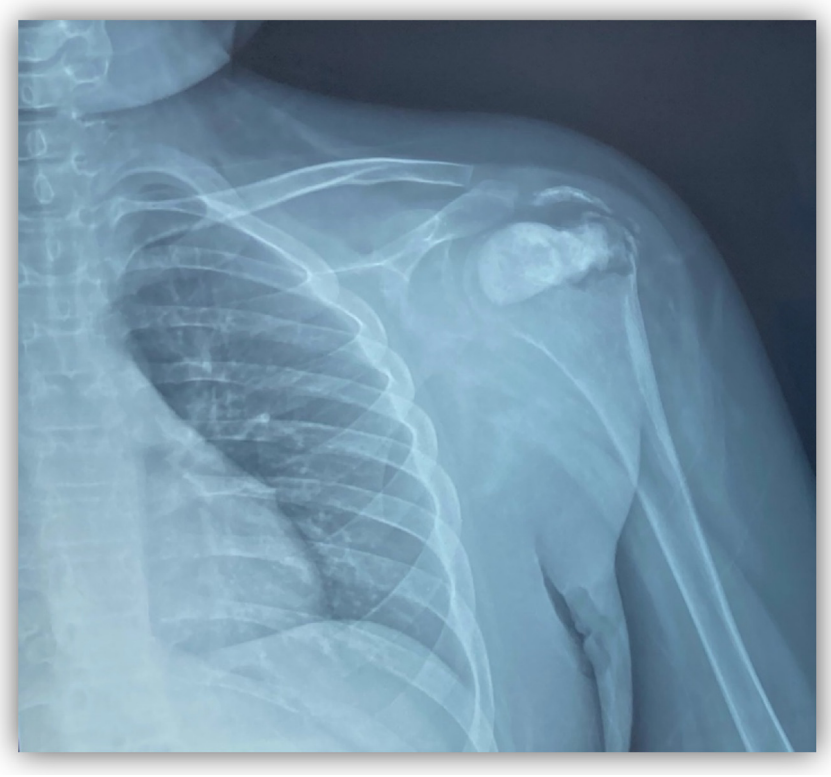

Fig. 5. Anteroposterior view of left shoulder of a skeletally immature patient immediately at six-month follow-up evaluation.

Declaration of Competing Interest

The authors declare no conflict of interest.

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Ethical approval

The University of Puerto Rico Institutional Review Board (IRB) has determined that case reports do not meet the DHHS definition of research with human subjects as the federal regulation (45 CFR 46.102(d)) and are exempt from ethical approval. The procedure performed in this study adhere to the tenets of the Declarations of Helsinki.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of consent is available for review by editor-in-chief of this the journal on request.

Author contribution

(1) Substantial contributions to the conception or design of the work; or the acquisition, analysis, or interpretation of data for the work: [*Manuel A. Ramírez-González, MD; Gerardo Olivella MD, MPH; Norman Ramírez, MD; Antonio Soler-Salas, MD; Eric Astacio, MD; Juan Bibiloni, MD; Christian Foy-Parilla MD*].
(2) Drafting the work or revising it critically for important intellectual content: [*Manuel A. Ramírez-González, MD; Gerardo Olivella MD, MPH; Norman Ramírez, MD; Antonio Soler-Salas, MD; Eric Astacio, MD; Juan Bibiloni, MD; Christian Foy-Parilla MD*].
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Registration of research studies

N/A.

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