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Simple bone cysts of the proximal humerus presented with limb length discrepancy: A case report

Cing Syue Lin, Shang Ming Lin, Syang-Peng Rwei, Chin-Wen Chen, Tsung-Yu Lan

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Abstract

BACKGROUND

Simple bone cysts (SBC) are benign tumor-like bone lesions typically identified in children. While SBC may lead to growth disturbances or growth arrest, such cases are uncommon. The mechanisms behind these observations remain unclear. Additionally, research on the etiology of SBC remains inconclusive, and there has been no consensus on the appropriate timing and methodology for treatment.

CASE SUMMARY

Here, we present our experience in the successful surgical management of a 10-year-old girl with SBC, who presented with a pathological fracture complicated by malunion of the displaced fracture, varus deformity, and limb length discrepancy. We hypothesized two possible etiologies for the patient's growth arrest and subsequent humerus varus deformity: (1) Direct disruption of the physis by fluid from the cyst itself; and (2) damage to the epiphysis due to repetitive pathological fractures associated with SBC. In addressing this case, surgical intervention was undertaken to correct the proximal humerus varus deformity. This approach offered the advantages of simultaneously correcting angular abnormalities, achieving mild limb lengthening, providing definitive SBC treatment, and reducing the overall treatment duration.

CONCLUSION

As per current literature, acute correction of acute angular deformity in proximal humeral SBC is not well comprehended. However, in this specific case, acute correction was considered an optimal solution.

Key Words: Bone cyst; Solitary cysts; Benign; Humerus; Growth arrest; Limb length discrepancy; Case report

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Core Tip: We successfully operated on a 10-year-old girl with simple bone cysts (SBC), who presented with varus deformity, limb length discrepancy, and malunion resulting from a neglected pathological fracture. We proposed two potential causes for the humeral growth arrest and varus deformity in this patient: (1) Direct disruption of the physis by fluid from the cyst itself; and (2) damage to the epiphysis due to recurrent pathological fractures associated with SBC. Presently, the literature lacks a comprehensive understanding of the acute correction of angular deformity in proximal humeral SBC. However, in this specific case, acute correction was considered an ideal option.

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INTRODUCTION

Simple bone cysts (SBC), also known as solitary bone cysts are benign, fluid-filled tumor-like bone lesions commonly found in the proximal humerus (70%) or femur (25%) of children and adolescents[1,2]. While SBC may occasionally result in growth disturbances or growth arrest, such cases are uncommon. The underlying mechanisms behind this remain unclear[3]. This article presents a case involving a 10-year-old girl who initially complained of left arm pain attributed to an angulated old fracture, accompanied by limb length discrepancy. Additionally, a review of the current literature on SBC is included in this discussion.

CASE PRESENTATION

Chief complaints

A 10-year-old Chinese girl presented at our orthopedics clinic with a history of restricted left shoulder elevation and left upper limb length discrepancy spanning six months.

History of present illness

The patient experienced difficulty lifting her left arm during undressing, leading her mother to observe a noticeable length difference in the left arm. Concerned about these issues, the mother brought her to our clinic for evaluation.

History of past illness

The patient had no pre-existing medical conditions. No history of trauma, fever, local infection, or other precipitating factors in the left shoulder were noted.

Personal and family history

The patient's mother denied any family history of tumors.

Physical examination

During physical examination, it was observed that the patient's left arm was approximately 4-5 cm shorter than the right. The active range of motion (ROM) for the left shoulder was limited due to pain (Table 1). However, the distal circulation was found to be intact, and no focal neurological signs or symptoms were observed.

Laboratory examinations

Serum C-reactive protein levels were within the normal range, and routine blood and urine analyses showed no abnormalities. A tumor marker workup, which included alpha-fetoprotein, cancer antigen 125, carbohydrate antigen 19-9, prostate-specific antigen, and carcinoembryonic antigen, all revealed values within normal limits.

Imaging examinations

Initial imaging studies indicated an angulated old fracture at the proximal left humeral neck (Figure 1). Subsequent examination with magnetic resonance imaging (MRI) revealed marked deformity of the left humeral head and possible malunion of the displaced fracture in the left humeral neck. No evidence of cortical discontinuity, intraosseous pathologies, or periosteal reaction was noted (Figure 2).

Table 1 The preoperative and postoperative active range of motion of left shoulder

	Preoperative active ROM	Postoperative-1 month active ROM	Postoperative-1 year active ROM
Flexion	70	176	180
Abduction	80	175	180
External rotation	45	90	95
Internal rotation	10	65	75

ROM: Range of motion.

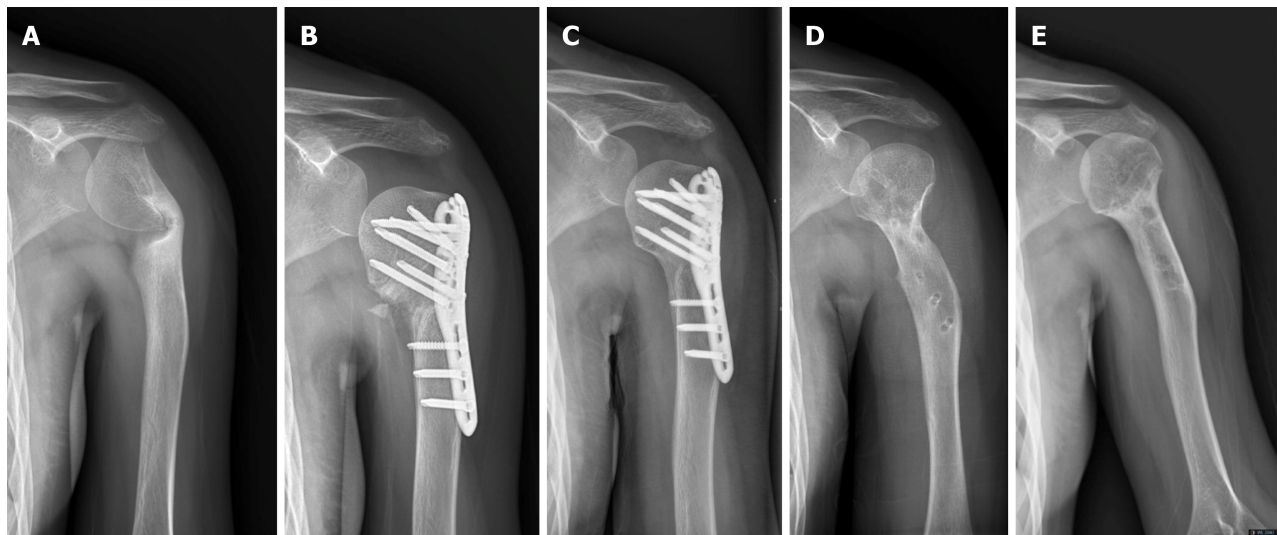


Figure 1 Series anteroposterior plain radiographs of the left humerus. A: Preoperative anteroposterior plain radiographs of the left humerus at initial presentation; B: 1-month postoperative anteroposterior plain radiographs of the left humerus; C: 1-year post-operative anteroposterior plain radiographs of the left humerus; D: Anteroposterior plain radiographs of the left humerus at 1-year post-removal of internal fixator; E: 1-year post-removal of internal fixator surgery.



Figure 2 Preoperative magnetic resonance images and pathological results of the patient. Preoperative T2-weighted magnetic resonance images of the left humerus showed a marked deformity of the humeral head and a potential malunion of the displaced fracture in the left humeral neck.

FINAL DIAGNOSIS

Considering the patient’s medical history, the final diagnosis indicated a case of left humerus SBC with varus malunion or suspected postinfection sequelae of osteomyelitis.

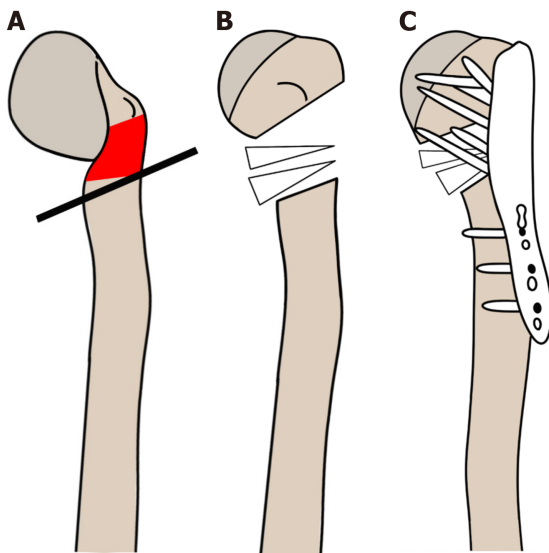


Figure 3 Surgical procedure for acute correction of the proximal humerus for the varus deformity. The red area indicates the location of the simple bone cyst (SBC). A: Osteotomy was performed in the most angulated area, removing the deformity site of the SBC; B: The gap was filled with allograft (donor femoral head); C: The proximal humerus was fixed with an anatomical locking plate (Depuy-Synthes®, Raynham, MA, United States) in a valgus position.

TREATMENT

Surgical intervention was recommended for this case of left humerus SBC with varus malunion or suspected postinfection sequela of osteomyelitis. The procedure involved placing the patient under general anesthesia in a semi-sitting position. The anterior approach was utilized to expose the proximal metaphysis and diaphysis of the left humerus. Osteotomy was performed at the most angulated area, and curettage over the lesion site was performed, with the specimens sent for pathological study. The proximal humerus was then fixed with an anatomical locking plate (Depuy-Synthes, Raynham, MA, United States) in a valgus position and the gap was filled with an allograft (donor femoral head) (Figure 3). The patient has not received any additional medication therapy beyond the surgical intervention.

OUTCOME AND FOLLOW-UP

Postoperatively, the patient was immobilized in a sling for 2 wk. The left shoulder length discrepancy was reduced by 2 cm. After 1 month, she exhibited full active ROM in the left shoulder, with no reported wound complications or infection (Table 1 and Figure 4). Follow-up radiographs indicated no progression of the lesion; the osteotomy site showed solid union at 1 month and 1 year postoperatively (Figure 1). The histological report revealed degenerated cortical bone and cartilage tissue, with fibrous cystic wall with fibrin-like materials, cholesterol clefts and surrounding some new bone formation (Figure 5), leading to the diagnosis of SBC. While no pain or discomfort was reported, the implant was removed one year later based on the parents' preference. Subsequent radiographs showed that the left proximal humerus had healed with union and was well-aligned (Figure 1). One year following implant removal surgery, radiographic assessment revealed favorable remodeling and alignment of the left proximal humerus. Physical examination showed full active ROM and normal muscle power in the left shoulder (Table 1 and Figure 1). She exhibits the ability to execute motor milestone tasks in accordance with her chronological age. Furthermore, the length discrepancy between both hands was maintained at a 2 cm difference.

DISCUSSION

SBC are benign metaphyseal lytic lesions predominantly occurring in males[3]. Despite research, the etiology of SBC remains inconclusive[4,5]. Most cases of SBC are asymptomatic and are discovered incidentally. Spontaneous resolution of SBC occurs in only about 5% to 10% of cases[6,7]. Symptomatic cases are often associated with pathological fractures (63%-87%)[1,8] and less than 15% of cysts resolve spontaneously after a fracture occurs[5,9]. Presently, there is no consensus or established guidelines on the optimal timing and method of treating SBC[2,6,7,10]. To the best of our knowledge, the acute correction of angular deformity in proximal humeral SBC is not yet fully understood.

The primary differential diagnoses for pathological fractures in children aged 5–10 years include SBC, aneurysmal bone cysts (ABC), non-ossifying fibroma, and osteomyelitis[11]. In our patient, the lesion was located in the proximal humerus, consistent with the predisposing location of SBC. As our patient exhibited no signs of inflammation, bruises, or swelling, and had not received antibiotic treatment, chronic osteomyelitis was ruled out. Given the similar clinical symptoms of SBC, ABC, and non-ossifying fibroma, imaging and pathology played a crucial role in differentiating these



Figure 4 Two-week postoperative photos of the patient. A: Full active range of motion of the left shoulder; B: About 2-3 cm length discrepancy on the affected side.

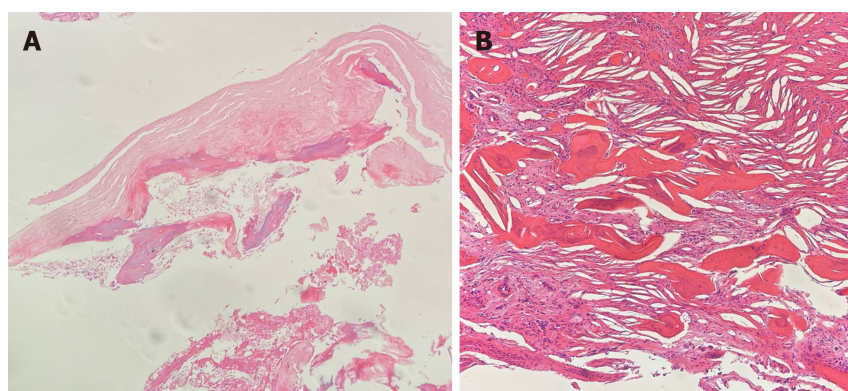


Figure 5 Pathological results of the patient. A: Histological results showed fibrous cyst wall with fibrin-like material and calcification; B: Histological results showed cystic wall with cholesterol clefts and surrounding some new bone formation, all of which may be indicative of a diagnosis of a simple bone cyst.

diseases.

Radiographic findings in our case revealed an old, angulated fracture at the proximal left humeral neck (Figure 1). Although the characteristic cystic lesions of SBC were not visibly evident on radiography and MRI, the possibility of SBC could not be ruled out. Furthermore, MRI showed a smoother sclerotic junction with the absence of fat stranding or inflammation in the surrounding soft tissue (Figure 2), suggesting fluid reabsorption and healing. These findings, combined with the histological report, were indicative of a benign cause.

A pathological fracture occurred in association with the benign lesion, leading to the subsequent displacement of the fracture and resulting in a malunion at the left humeral neck. The histological report showed fibrous cystic wall with fibrin-like materials, cholesterol clefts and surrounding some new bone formation, consistent with SBC. Considering the patient's history, the results suggest fibrosis with chronic inflammation, supporting a diagnosis of SBC. Pathologic fractures can often remain asymptomatic until the moment of fracture or may occur after lesions that cause prodromal pain. However, they typically do not present with accompanying symptoms.

As such, we posit that this patient experienced an initially asymptomatic pathologic fracture due to low-energy trauma, resulting in a delayed diagnosis and treatment. The lack of a proper diagnosis and timely intervention contributed to the development of malunion in the old, displaced pathological fracture at the left humeral neck, along with subsequent issues of limb length discrepancy and angular deformity.

On rare occasions, SBC may lead to growth disturbances, and, the underlying mechanism remains a subject of debate. Some evidence suggests that the cyst fluid itself may directly disrupt the physis, causing growth disturbances[12-14]. Other potential processes include: (1) Fractures through the cyst, damaging the physis and resulting in growth arrest; (2) direct extension of the cyst through the physis; and (3) iatrogenic damage to a developing physis due to surgical removal

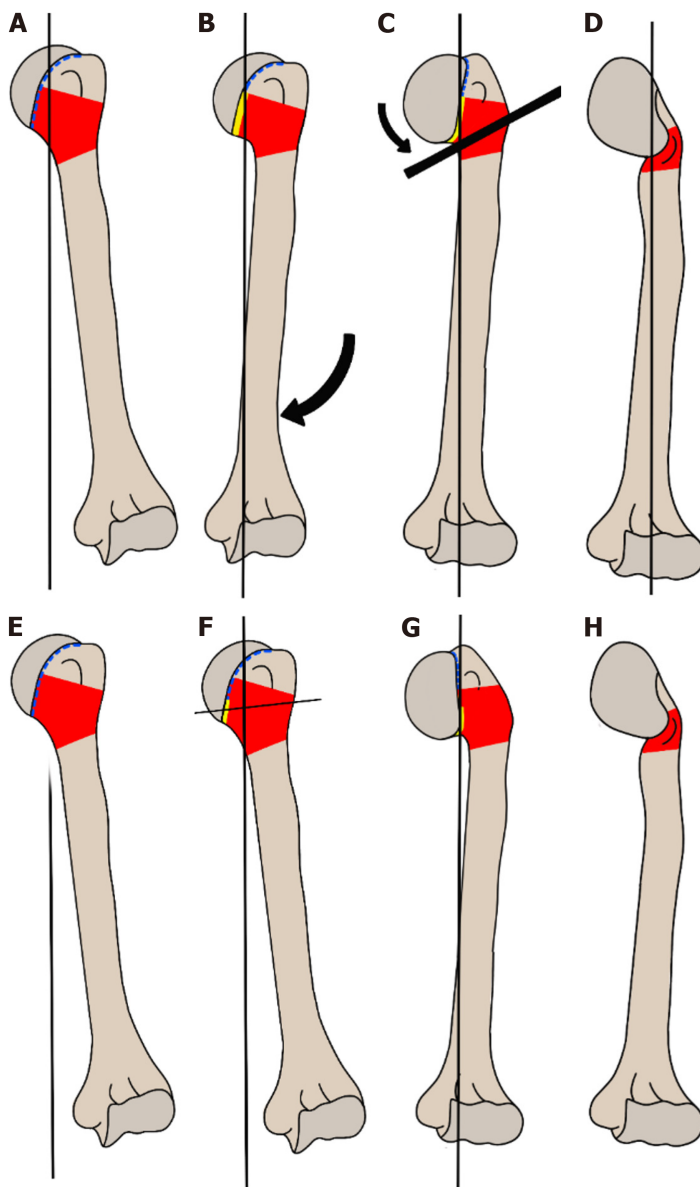


Figure 6 Two hypotheses for the etiology of our patient with simple bone cyst who presented with a pathological fracture, angular deformity, and limb length discrepancy. The red area represents the location of simple bone cyst (SBC); the blue dotted line represents the epiphysis; and the yellow line indicates the affected epiphysis. A–D: The first hypothesis. A: The proximal humeral simple bone cyst extends through the physis on the medial side; B: The SBC subsequently caused growth arrest and varus deformity of the humerus; C: Due to a proximal humerus fracture, additional varus angulation of the humeral head occurs; D: SBC presenting with a pathological fracture, complicated with malunion of the displaced fracture, varus deformity and limb length discrepancy. E–H: The second hypothesis. E: A proximal humeral simple bone cyst develops; F: Repetitive pathological fractures due to SBC damaged the medial epiphysis of the proximal humerus and varus humeral head angulation was established as a consequence; G: The humerus develops varus deformity and a length discrepancy due to a damaged epiphysis; H: SBC presenting with a pathological fracture, complicated with malunion of the displaced fracture, varus deformity and limb length discrepancy.

of a cyst in proximity[6,12–18]. Regardless of the etiology, growth disturbances ultimately result in angular deformity and/or limb length discrepancy in the affected limb. In the case of our patient, limb length discrepancy was evident before surgical treatment, making it less likely that her injury was related to iatrogenic damage.

We proposed two hypotheses to explain the etiology of our patient's condition. First, we considered the possibility of direct extension of the SBC through the epiphysis on the medial side, or the direct disruption of the physis by the cyst fluid itself, leading to growth arrest and subsequent varus deformity of the humerus. Given that the proximal physis contributes to approximately 80% of the humerus' development, any disturbance in this region can result in significant upper limb length discrepancy[6]. The additional varus angulation of the humeral head occurred as a sequela of the proximal humerus fracture (Figure 6).

The second hypothesis proposed was that the epiphysis was damaged by repetitive pathological fractures due to SBC. The bone affected by SBC is weakened and prone to pathological fractures, which can, in turn, damage the epiphysis. As a consequence of these pathological fractures, varus humeral head angulation developed. The muscles pulling on the proximal humerus allowed pathological fractures to heal with an increasing varus deformity, giving rise to the typical complaint of severe shoulder ROM limitations in abduction and flexion. Furthermore, due to a defective medial epi-

physis, the humerus developed further varus deformity and a length discrepancy (Figure 6).

We presented two surgical intervention options for the patient. The first option involved acute correction of the proximal humerus varus deformity. The alternative was distraction osteogenesis with external fixation, such as Ilizarov or Taylor spatial frame, combined with internal lengthening nails for limb lengthening and angle correction. Following careful consideration, the parents opted for the first treatment due to its shorter course, while recognizing the inherent limitation in correcting limb length discrepancy. Considering the nearly closed proximal humeral physis in the patient, the likelihood of developing subsequent length disparities was deemed low. Minor discrepancies in arm length, up to 5 or 6 cm, are generally well-tolerated without causing significant functional constraints, given the primary role of the upper extremity, which is non-weight-bearing[18,19].

Thus, the surgical treatment of acute correction for the proximal humerus varus deformity was performed. This approach offers the advantage of simultaneously correcting angular abnormalities, providing mild limb lengthening, definitive cyst therapy, and a shortened course of treatment. Consequently, we believe that the acute correction of the varus deformity was the optimal option in this case.

CONCLUSION

In conclusion, we presented a case of SBC presenting with a pathological fracture, complicated with malunion of the displaced fracture, varus deformity, and limb length discrepancy. The discussion included differential diagnoses of SBC, alongside a comprehensive review of the current literature. In our case, the patient's limb length discrepancy and varus deformity emerged as complications of the SBC and pathological fracture. It underscores the importance of close follow-up in patients with SBC to monitor the potential occurrence of growth arrest.

FOOTNOTES

Author contributions: Lan TY, Lin SM, Rwei SP, and Chen CW contributed to conceptualization and supervision; Lin CS contributed to data collection, software, paper review, manuscript writing and editing; Lan TY contributed to clinical examination, surgery, original draft preparation and editing; All authors have read and agreed to the published version of the final manuscript.

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