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Evaluation of different treatment modalities for fractured and non-fractured simple bone cyst

A single-center review of 68 patients

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Abstract

Simple bone cysts (SBCs) occur most frequently in the proximal aspect of the humerus and femur in growing age and are associated with intercurrent pathological fractures in up to 87%. Therapeutic management of SBCs remains controversial. The aim of this study was to examine the outcome of conservative and various surgical treatment modalities considering the specific anatomic location and integrity of the SBC.

In this retrospective study, we analyzed 68 cases of SBCs who underwent a conservative or surgical treatment between 2009 and 2020 with a mean follow-up of 30.1 months. The epidemiological characteristics, complications, clinical, and radiographic outcome after conservative or surgical treatment were assessed.

The study includes 50 male (73.5%) and 18 female (26.5%) patients with a mean age of 9.1 years. The most common locations were the proximal humerus (69.2%, n = 47) and femur (16.2%, n = 11). In 43 cases (63.2%; upper limb n = 40, lower limb n = 3) a pathological fracture occurred. Fifty patients (73.5%; upper limb n = 40, lower limb n = 10) underwent a conservative treatment. In 11 cases (16.2.1%; upper limb n = 4, lower limb n = 7) an intralesional curettage and defect reconstruction with bone substitute without stabilization were performed. Five patients (7.3%; upper limb n = 4, lower limb n = 1) received an osteosynthesis, in two cases (2.9%; upper limb 1; lower limb 1) combined with an intralesional curettage and defect reconstruction with bone substitute. All 32 pathological fractures treated conservatively (upper limb n = 31, lower limb n = 1) healed within 6 weeks; 17/43 patients (39.5%) suffered at least one second fracture. After intralesional curettage and defect reconstruction with bone substitute local recurrence was observed in 5/13 cases (38.5%). Spontaneous consolidation, at least partially, was observed in three cases (4.4%) following conservative treatment after fracture. No relevant secondary angular or torsional deformity was observed after treatment.

The majority of SBCs can be treated conservatively, especially in the upper extremity. However, if a fracture is completely dislocated, joint affecting, unstable or open, surgical treatment is necessary. Load-dependent pain or the inability to mobilize timely after fracture can necessitate surgical treatment in SBCs affecting the lower extremity. Spontaneous resolution, especially after fracture, can be seen in rare cases.

Abbreviation: SBC = simple bone cysts.

Keywords: intralesional curettage, juvenile bone cyst, pathological fracture, recurrence, simple bone cyst, solitary bone cyst

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ND and ND contributed equally to this work.

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1. Introduction

Simple bone cysts (SBCs) represent tumor-like bone lesions, occurring most frequently in the proximal aspect of the humerus and femur in children and adolescents.[1] They are benign, single chambered, and fluid filled. The average age of onset is 8 to 14 years.[2] The male-to-female ratio is 2:1 to 4:1.[3] Pain caused by intercurrent pathological fractures is often the first symptom, and 63% to 87% of cysts are combined with pathological fractures.[4]

At present the treatment of SBCs is still under controversial discussion and various treatment options were described. As the cysts tend to resolve spontaneously after puberty[5] every surgical procedure has to be critically discussed with the patient and his family. Hardes et al regard a stable situation without clinical symptoms, lack of radiographic progression of the cyst or an incomplete healing as an acceptable intermediate result.[6] Among the therapeutical options for unfractured cysts are simple observation,[7] decompression of the cyst with screws or nails,[6] intracavitary injection of steroids and autologous red bone marrow,[8] intralesional curettage and defect reconstruction with bone substitute[9] or elastic intramedullary nailing[2] with or without use of bone marrow and/or bone substitute. In case of pathological fracture, conservative and different surgical treatment modalities are discussed.[7]

In this study, we analyzed the epidemiological characteristics, complications, the clinical and radiographic outcome after conservative or surgical treatment of SBCs with and without fracture. We want to comment on the questions whether a conservative treatment is adequate for SBCs considering the location (upper or lower limb), especially in case of pathological fracture and whether secondary axis deviation has to be expected and how many refractures may occur. We also examine the recurrence rates of SBCs after intralesional curettage and defect reconstruction with bone substitute to evaluate the benefit of an operation.

2. Materials and methods

2.1. Patients' demographics and data acquisition

This retrospective study analyzed 68 cases of SBCs (50 males [73.5%], 18 females [26.5%]) with a mean age of 9.1 years (range 2–20) who were treated between 2009 and 2020 at our institution (Tables 1 and 2). The mean follow-up was 30.1 months (range 12–160). Epidemiological data, radiographic and histological examinations, different conservative and surgical treatment modalities, complications and local recurrence and refracture rates were assessed. Clinical information was obtained from the digital patient records (ORBIS, Agfa HealthCare GmbH, Mortsel, Belgium).

Imaging studies at time of presentation, including radiographs and MRI scans were analyzed in each case using a calibrated digital radiology system (Centricity^{TM} PACS, GE Healthcare, Chicago, IL and TraumaCad, Brainlab, Munich, Germany). In case of surgery the diagnosis was stated by (immuno)histological examination. Patients underwent follow-up with clinical and radiographic examinations (radiographs and MRI scans) at 4 weeks interval after a fracture and regularly at 6-month intervals for the following 2 years and thereafter once a year.

2.2. Conservative and surgical treatment

The initial treatment either consisted of a conservative approach or a surgical intervention comprising an intralesional curettage with filling up the cavity with bone substitute (silicate substitute calcium phosphate, Actifuse, Baxter, Deerfield, IL) and/or osteosynthesis. The treatment modality was determined on basis of joint decision making between the patient and the clinician depending on clinical symptoms, radiographic findings, and the necessity for histological diagnosis although no stringent algorithm was employed during the study period.

Table 1

Overview about the frequencies of location, fracture and kind of treatment of SBCs in the study group.

		Treatment						
			Surgical treatment					
Location	Fracture	Conservative treatment/observation	Curettage and bone substitute	Curettage. bone substitute and plate osteosynthesis	Plate osteosynthesis	Intramedullary nailing	Total	Total
Lower extremity	No	10 (58.8%)	6 (35.3%)	1 (5.9%)	0	0	7 (41.2%)	17
	Yes	1 (33.3%)	1 (33.3%)	0	1 (33.3%)	0	2 (66.6%)	3
Femur	No	5 (50.0%)	4 (40.0%)	1 (10.0%)	0	0	5 (50.0%)	10
	Yes	0	0	0	1 (100.0%)	0	1 (100.0%)	1
Tibia	No	2 (100.0%)	0	0	0	0	0	2
	Yes	0	1 (100.0%)	0	0	0	1 (100.0%)	1
Fibula	No	1 (100.0%)	0	0	0	0	0	1
	Yes	1 (50.0%)	1	0	0	0	0	2
Pelvis	No	2 (66.6%)	1 (33.3%)	0	0	0	1 (33.3%)	3
	Yes	0	0	0	0	0	0	0
Upper extremity	No	7 (87.5%)	1 (12.5%)	0	0	0	1 (12.5%)	8
	Yes	31 (79.4%)	3 (7.7%)	1 (2.6%)	0	4 (10.3%)	8 (20.5%)	39
Humerus	No	7 (87.5%)	1 (12.5%)	0	0	0	1 (12.5%)	8
	Yes	30 (76.9%)	4 (10.3%)	1 (2.6%)	0	4 (10.3%)	9 (23.1%)	39
Clavicle	No	0	0	0	0	0	0	0
	Yes	1 (100.0%)	0	0	0	0	0	1
Total	No	17 (68.0%)	7 (28.0%)	1 (4.0%)	0	0	8 (32.0%)	25
	Yes	32 (74.4%)	5 (11.6%)	1 (2.3%)	1 (2.3%)	4 (9.3%)	11 (25.6%)	43

Table 2

Patient demographics and SBCs characteristics comparing conservative and operative treatment.

	All	Conservative treatment	Surgical treatment
Sex			
Male	50	40	10
Female	18	10	8
Mean age	9.1	9.3	8.5
Affected extremity			
Lower extremity			
Fracture at presentation	3	1	2
No fracture at presentation	17	10	7
Mean no. of fractures	0.2	0.2	0.2
Upper extremity			
Fracture at presentation	39	31	8
No fracture at presentation	8	7	1
Mean no. of fractures	1.5	1.4	1.5
Location			
Metaphysis	59	42	17
Diaphysis	5	5	0
Mean cyst height (mm)	67.3	65.5	72.1
Mean cyst width (mm)	26.2	26.1	26.5
Mean cyst index	6.0	6.2	5.6
Mean distance from growth plate (mm)	18.8	19.6	16.5
Mean cortical thickness	4.2	4.0	4.7
Activity active	29	22	7
Latent	31	22	9

2.2.1. Non-fractured SBCs. In the majority of cases, especially in the upper limb, SBCs without pathological fracture (25; upper limb n = 9, lower limb n = 16) were managed by pure observation and watchful waiting with periodical clinical and radiographic examination once a year.

Curettage and defect reconstruction with bone substitute were conducted on the one hand due to load-dependent pain in the lower extremity and on the other hand because of recurrent fractures in the upper extremity. An additional non-locking plate was applied to prevent a fracture after extensive curettage and defect reconstruction in a femoral SBC (Fig. 1).

2.2.2. Fractured SBCs. In case of a humeral fracture, the adjacent joints were immobilized for 4 weeks using a one-part shoulder orthosis (Tricodur Gilchrist, BSN medical, Hamburg, Germany). In case of a clavicle fracture 4-week immobilization was achieved with a figure-of-8-splint (Tricodur Clavicula, BSN medical, Hamburg, Germany). Patients with fractures of the lower extremity were restrictedly mobilized on crutches with partial weight bearing without applying any casts or splints.

Elastic intramedullary nailing (Intramed, MK medical, Emmingen-Liptingen, Germany) was performed in humeral fractures (Fig. 2) and different types of locking and non-locking plates were used for fracture stabilization in the upper and lower extremity (Fig. 3).

2.3. Clinical assessment

2.3.1. Pain and function. Pain was documented as present or absent. Function was defined as unlimited if the patient was able to participate in full daily activity including contact sports. Limited function was defined if the patient was unable to participate in contact sports, but were still able to manage all other daily functions.

2.3.2. Secondary maltorsion. Torsion was assessed clinically by comparing the range of motion in the transversal plane of the affected side with the opposite site. A deviation in rotation of $>10°$ was considered as torsional deformity.

2.4. Radiographic assessment

Radiographs were evaluated in anteroposterior and lateral view at time of first presentation and after observation or intervention at last follow-up.

2.4.1. Extent of the SBCs. The cyst height and cyst width were determined by measuring the largest longitudinal and transverse diameter. The cyst index (CIX) was calculated as previously described by Kaelin and MacEwen.[10] Additionally, the smallest cortical thickness of the cyst was quantified.

2.4.2. Activity level of the SBCs. The distance from the growth plate to the cyst was measured in all cases with metaphyseal SBCs. According to Jaffé and Lichtenstein, the cyst was regarded as active if the distance was ≤ 10 mm and otherwise as latent.[11]

2.4.3. Healing status of the SBCs. The healing status of the cysts in terms of obliteration and increase of cortical thickness was classified as previously described by Neer et al and Capanna et al.[12,13] A cyst was classified as completely healed if it was filled with bone and the cortical margins had thickened. A cyst was classified as partially healed when it was consolidated in parts but there were still residual areas of radiolucency. A cyst was classified as recurrence when it had healed initially, but progressive areas of radiolucency and cortical thinning developed in the follow-up examinations.[14]

2.4.4. Fracture healing and secondary axis deviation. Osseous consolidation and recorticalization of a fractured SBC was assessed qualitatively on biplanar radiographs (Figs. 2 and 4).

Axis deviation in the coronal and sagittal plane at time of fracture and after treatment was evaluated by measuring the angle between the longitudinal axes of the proximal and distal segment (Figs. 3 and 4). A deviation of $>5°$ was considered as angular deformity.

2.5. Assessment of perioperative complications

Perioperative complications such as delayed wound healing, infection, and neurovascular damage were recorded.

2.6. Statistical analysis

Statistical analysis was performed with the use of SPSS software, Version 26 (IBM, Armonk, NY). Continuous variables such as age and time of follow-up were described using mean and range. The Wilcoxon test was used to compare axis deviation of fractured bones before and after conservative treatment.

3. Results

3.1. Epidemiological results

The most common location was the proximal metaphyseal humerus (61.8%, n = 42), followed by the proximal metaphyseal femur (16.2%, n = 11) and the diaphyseal humerus (7.4%, n = 5). In 3 cases (4.4%), SBCs were located in the proximal or distal metaphyseal fibula, in 3 further cases in the pelvis (4.4%), in 2

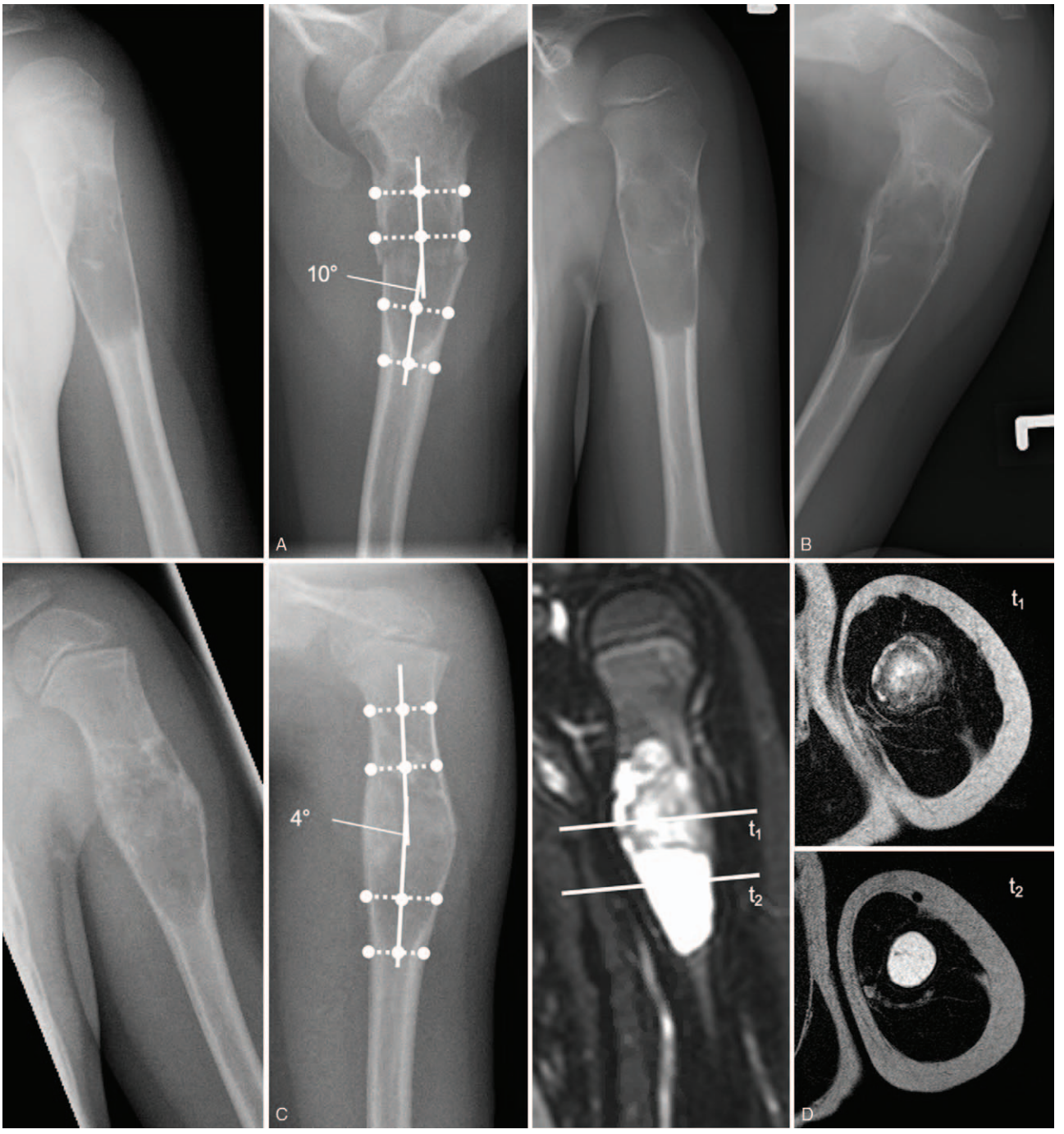

Figure 1. Imaging of a male patient with SBC of the proximal humerus with pathological fractures. (A–D) Anteroposterior radiographs showing the fractured SBC at the age of 8 years (A), the immediate postoperative Status after elastic intramedullary nailing (B), complete consolidation of the fracture 8 weeks after surgery (C) and the persistent SBC after removal of the implants 5 months after surgery (D). (E–H) Anteroposterior radiographs showing a refracture of the persistent SBC at the age of 12 years and continuous growth of the proximal humerus (E), complete consolidation of the fracture after 8 weeks of conservative treatment (F) and continuous growth of the proximal humerus and the persistent SBC (G–H).

cases (2.9%) in the distal metaphyseal tibia, in one case in the proximal metaphyseal tibia (1.5%) and in one case in the clavicle (1.5%) (Table 1). No SBC affected the epiphysis.

Forty-three/sixty-eight patients (63.2%) suffered a pathological fracture. In 34 cases the proximal humerus and in five cases the diaphyseal humerus were affected. A fracture was observed once in the proximal femur, the proximal fibula, the distal tibia, and the clavicle, respectively (Table 1).

Fifty/sixty-eight patients (73.5%) underwent conservative treatment. Among those were 33 fractures of the humerus (Fig. 4) and each one fracture of the proximal fibula and the clavicle (Table 1).

In 13 cases, five of these with a fracture, intralesional curettage with defect reconstruction using bone substitute was performed. One case with a pathological humeral fracture and one case of a non-fractured femoral SBC received an additional plate osteosynthesis (Fig. 1). One fractured SBC of the proximal femur was

Figure 2. Imaging of a 6-year-old female patient with SBC and pathological fracture of the proximal femur. (A) Anteroposterior and lateral radiographs showing the fractured SBC. (B) Anteroposterior and lateral radiographs 6 weeks after fixation by transphyseal non-locking blade plate without curettage of the cyst and without surgical correction of the 20° axis deviation in the lateral plane. (C) Anteroposterior and lateral radiographs 1.5 years after operation showing complete consolidation of the fracture, partial sclerosis of the SBC and continuous growth of the femoral neck. (D–F) Anteroposterior and lateral radiographs 1.5 (D), 2.5 (E), and 3 (F) years after operation showing continuous growth of the femoral neck, complete correction of the sagittal angular deformity (1°), but a persistent and progressive SBC.

treated with a plate osteosynthesis without curettage (Fig. 3). Four patients (5.8%) with pathological fractures of the proximal humerus were treated with elastic intramedullary nailing (Fig. 2).

3.2. Results of treatment and complications

3.2.1. Fracture healing and refracture rate. All pathological fractures treated conservatively (n = 32) and surgically (n = 11) healed within 6 weeks; 17/43 patients (39.5%) developed at least one second fracture during follow-up (Fig. 2). The mean number of fractures per patient was 1.8 (77 fractures in 43 patients). There were no differences concerning the refracture rate between the affected location (upper or lower limb) and the treatment modality (conservative or surgical) ($P = .852$).

Figure 3. Imaging of a 6-year-old male patient with SBC and pathological fracture of the proximal humerus. (A) Anteroposterior and lateral radiographs showing the fracture with 10° axis deviation in the lateral plane. (B) Anteroposterior and lateral radiographs 4 weeks after conservative treatment showing progressive consolidation of the fracture. (C and D) Anteroposterior and lateral radiographs (C) and MRI scans (D) 12 months after fracture showing partial sclerosis of the SBC and complete consolidation of the fracture.

3.2.2. SBC healing and recurrence rate. After intralesional curettage and defect reconstruction with bone substitute complete healing of the cyst was observed in five cases (33.3%, two in the proximal humerus with fracture, one in the distal tibia with fracture, one in the pelvis without fracture). Partial healing according to Capanna[13] was achieved in three cases (25.0%, two in the proximal femur without fracture, one in the distal metaphyseal fibula without fracture). In five further cases (33.3%, three in the proximal humerus, one with fracture, two in the proximal femur without fracture [Fig. 1]) a recurrence of the cyst was observed.

Spontaneous healing, at least partially, was observed in two cases (2.9%) after pathological fracture without surgical treatment (Fig. 4) and in one case (1.5%) under simple observation of the cyst without pathological fracture.

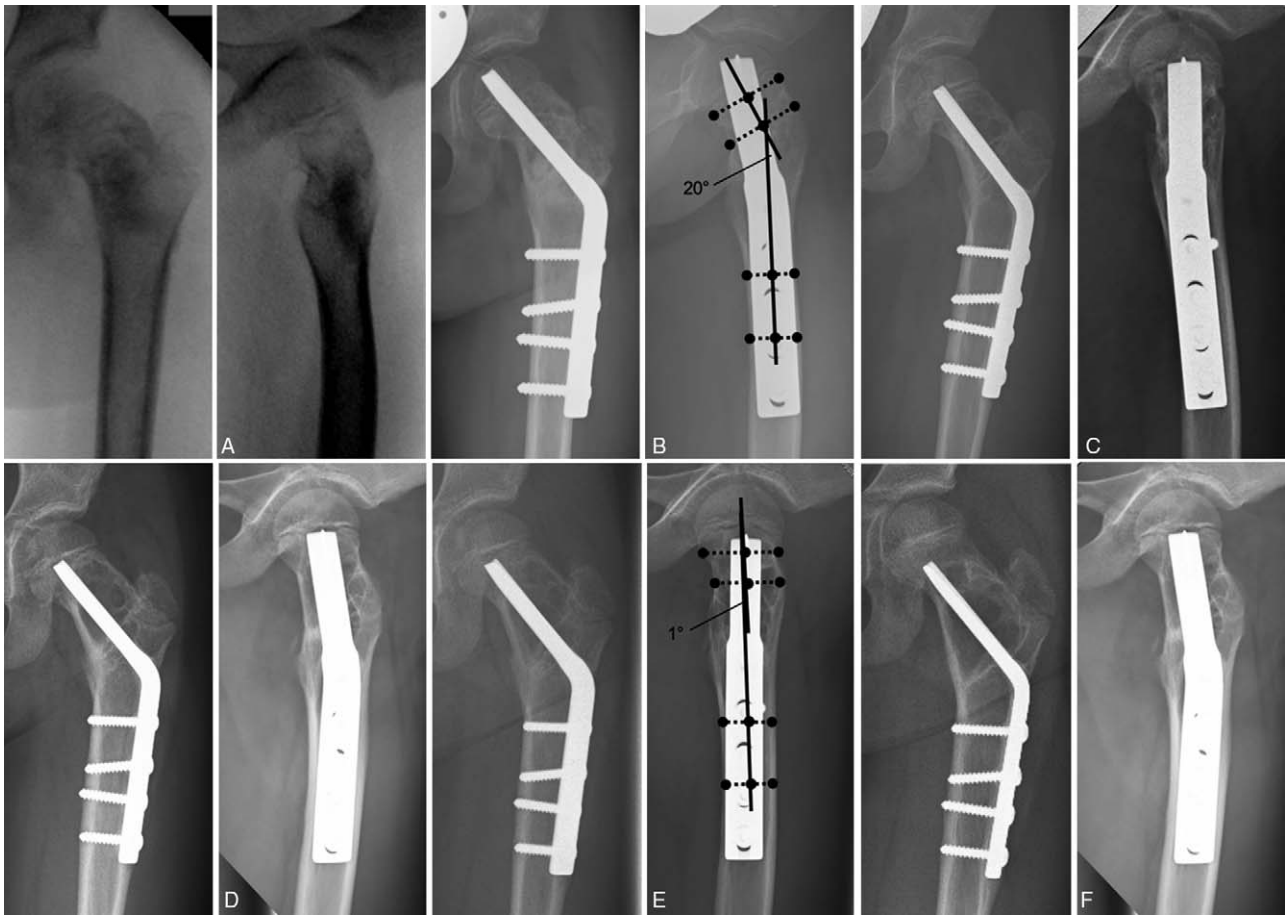

Figure 4. Imaging of a 5-year-old male patient with SBC of the proximal femur. (A) Anteroposterior and lateral radiographs showing the non-fractured SBC. (B) Anteroposterior and lateral radiographs 1 year after intralesional curettage of the SBC, defect reconstruction with bone supplement and fixation by non-locking reconstruction plate. (C) Anteroposterior and lateral radiographs 1 year after operation with initial signs of local recurrence of the former curetted SBC. (D) Anteroposterior and lateral radiographs 5 years after operation showing further progression of the SBC.

3.2.3. Complications and axis deviations. No perioperative complications were recorded. No secondary angular deformity occurred under conservative (Fig. 4) and surgical (Fig. 3) treatment ($P=.776$). The mean axis deviation was 3.1° in anteroposterior view and 4.2° in lateral view. No clinically noticeable arm or leg length discrepancy was detected.

3.2.4. Pain and function. Only one patient (1.6%) complained of persistent pain with functional impairment at the last follow-up due to local recurrence after surgical treatment of a humeral SBC.

4. Discussion

In our study, male patients represented three-quarters, female patients one-quarter of the cohort; the mean age was 9.1 years which is in accordance to other studies.[2] As previously described,[1] the most common locations for SBC manifestation are the proximal humerus and the proximal femur. The proximal humerus, followed by the proximal femur and the diaphyseal humerus were also the most common locations in this study. In this cohort most SBCs were localized in the metaphysis consistent with previous observations.[15]

Different treatment options of SBCs are controversially discussed in literature, mainly regarding the particularities of the affected limb, possible growth plate involvement and the presence and type of a pathological fracture.[2,6–9] Kadhim et al conclude that complete healing of humeral SBCs is challenging to achieve irrespective of the treatment modality.[16]

Hardes et al recommended an observing approach in case of small, inactive cysts.[6] Green et al observed similar outcomes in a comparative study of humeral SBCs with conservative or surgical treatment.[17] In our study, one-quarter of the patients were only periodically monitored without specific therapeutic intervention as the cysts were asymptomatic and did not show any expanding growth. Nevertheless, the absolute volume of SBCs increased due to the natural bone growth of children and adolescents.

About three-quarters of the patients with pathological fractures in this study were treated conservatively. Ambacher et al did not see the necessity for surgical intervention in any case of SBC. In uncomplicated cysts with or without pathological fracture the authors rather underlined the possibility of spontaneous healing.[7] In our study, all conservatively treated fractures healed without complications and showed no relevant secondary axis deviation.

However, Ambacher at al considered surgical treatment in completely dislocated, joint affecting, instable or open fractures.[7] In such cases, elastic intramedullary nailing was described as surgical treatment option with or without the use of bone substitute.[2] Irrespective of the affected limb Rapp et al recommended elastic intramedullary nailing in combination with artificial bone substitute and autologous platelet rich plasma to treat pathological fractures due to SBCs.[18] Zhang et al saw significant advantages in bone healing and recurrence rates in a comparative study between the treatment of SBCs by intralesional curettage with and without additional nailing in favor of the combined technique.[2] In our cohort in four cases with fractured SBCs of the proximal humerus elastic intramedullary nailing without additional intralesional curettage was performed. These fractures healed within 4 weeks but the SBCs did not resolute and patients suffered from persistent disease with one individual even sustained a second fracture after removal of the elastic nails. Whereas Roposch et al[19] recommended elastic intramedullary nailing for fractured and non-fractured SBCs, DeSanctis and Andreacchio reported about the risk of recurring pathological fractures after hardware removal after intramedullary nailing in case of partially or non-healed UBCs.[20] Thus, we agree with Zhang et al that patients might benefit from an additional curettage when performing intramedullary nailing. However, the number of patients treated with elastic intramedullary nailing in this study is very low.

Generally, one should question if intramedullary nailing without curettage of the cyst is advisable in case of an uncomplicated fracture of the upper extremity. As every fracture in our study healed without secondary axis deviation under conservative treatment timely, the necessity of an operative treatment only for stabilization of the fracture without addressing the underlying cyst is questionable.

Intralesional curettage with filling up the resulting cavity with bone substitute was the most common surgical procedure in our cohort. Curettage and defect reconstruction with bone substitute in cases without pathological fracture were performed due to load-dependent pain in the lower extremity, due to frequent fractures of the humerus and on request of the patient and its family in the upper extremity. After intralesional curettage and defect reconstruction with bone substitute complete healing of the cyst was observed in one-third of the cases, while a recurrence was detected in 41.7% of the patients. This recurrence rate is slightly higher compared to the study by Zhang et al that found a recurrence rate of 31.2% in their curettage group without additional nailing.[2] Traub et al reported no recurrence of SBCs after curettage and local steroid application in a small cohort of eight patients. In contrast, the authors described failure rates after initial treatment of 36.6% with steroids, 50.0% with intramedullary nailing alone and 21.4% with intramedullary nailing and additional steroid application.[1] The recurrence rate in this study is in accordance with previous studies that reported recurrence rates of up to 55.0% after intralesional curettage.[15,21]

Different studies investigating the outcome after surgical treatment of SBCs demonstrated a high number of local recurrences after curettage with or without usage of adjuvants. The lowest number of recurrences was reported by Traub et al investigating a small cohort of only eight patients treated by combination of intralesional curettage and local steroid application.[1] However, a study with a larger number of patients is necessary to prove the benefit from a combination of intralesional curettage and local steroid application.

This study has several limitations due to its retrospective, observative character and diversity of the patient cohort. To identify potential risk factors of the treatment prospective groupwise and comparative study designs are needed. The patients and compared groups were not randomized and exclusively descriptive statistical analysis were conducted.

5. Conclusions

Unfractured SBCs of the upper extremity can normally be treated conservatively by observation. Regarding the uncomplicated healing potential and the lack of relevant secondary axis deviations most pathological fractures caused by SBCs of the upper extremity can also successfully be treated conservatively. However, if the fracture is completely dislocated, joint affecting, unstable or open, surgical treatment is advisable.

In SBCs affecting the lower extremity surgical treatment is recommended in most cases with pathological fracture to quickly regain mobility and to prevent secondary axis deviations. Contrary to the upper limb surgical stabilization can also be considered in unfractured SBCs of the lower extremity due to load-dependent pain and increased risk of fracture.

In these conditions intralesional curettage and defect reconstruction with bone substitute might be preferable. However, the considerable risk of local recurrence needs to be considered. Spontaneous healing, especially after fracture, can be seen in rare cases.

Author contributions

Niklas Deventer, M.D. (Contribution: study conceptualization, data acquisition and curation, manuscript preparation – original draft)
Nils Deventer, M.D. (Contribution: study conceptualization, data acquisition and curation, manuscript preparation – original draft)
Georg Gosheger, M.D. (Contribution: supervision, resources, manuscript preparation – review & editing)
Marieke de Vaal, M.D. (Contribution: data curation, statistical analysis, manuscript preparation – review & editing)
Tymoteusz Budny, M.D. (Contribution: data acquisition and curation, manuscript preparation – review & editing)
Adrien Frommer, MD (Contribution: data acquisition and curation, manuscript preparation – review & editing)
Bjoern Vogt, M.D. (Contribution: project administration, study conceptualization, data acquisition and curation, manuscript preparation – review & editing)
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Data curation: Niklas Deventer.
Formal analysis: Nils Deventer.
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