ORIGINAL PAPER

Outcomes and complications of surgical treatment modalities for simple bone cysts of the humerus in children and adolescents

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Abstract

Purpose The aim of this study was to evaluate and compare different surgical treatment modalities for simple bone cysts (SBC) of the humerus regarding their effectiveness and recurrence rate.

Methods In this retrospective study, patients who received surgical treatment for previously untreated primary SBCs of the humerus were analyzed. Demographic data, cyst-specific as well as treatment-specific parameters, complications, treatment failures, and recurrence rates were collected and correlated with different treatment modalities. Observed procedures were categorized as open procedure ($n=20$) or osteosynthesis alone ($n=3$). For the open procedure group, four subgroups could be defined.

Results Twenty-three patients were included. The mean age at diagnosis was 11.6 ± 2.5 years, and the mean postoperative follow-up was 3.9 ± 2.6 years (range 1.0–10.3). After surgical intervention, a total of five (21.7%) patients showed at least one recurrence. Fracture occurred in three (13.0%) cases. The incidence of treatment failure was significantly higher in the curettage, allograft, adjuvants group, with five (83.3%) of six cases showing recurrence, than in the other subgroups ($\leq$ 25.0%) including the osteosynthesis alone group ($p=.024$).

For the open procedure group, the failure-free survival rates were 80.0% after two years and 50.4% after five years. For the three cases treated by osteosynthesis alone, no failures were observed.

Conclusion Open procedures showed similar failure rates except for the subgroup using curettage, allograft, and adjuvants which showed significantly higher treatment failure. Promising results were observed in the group which received solely osteosynthesis without cyst excision or filling, as no treatment failure was observed here.

Keywords Simple bone cyst · Unicameral · Bone cyst · SBC · Humerus · Pediatric

Introduction

Simple bone cysts (SBCs) are the most common benign cystic bone lesions in children [1]. These bone cysts are fluid-filled osteolytic lesions surrounded by a fibrous membrane [2]. SBCs usually present radiologically as central, well-marginated, radiolucent lesions, which are predominantly found in the long bones [3]. The proximal humerus (70%) followed by the proximal femur (25%) is most commonly affected in children or young adults [4]. SBCs may be asymptomatic and therefore are often discovered only as an incidental finding. However, cyst growth can result in thinning of the cortical bone around the cyst, which is associated with an increased risk of fracture [3]. This can lead to pain and limited range of motion. Most frequently (approximately 75%), the diagnosis is established upon presentation due to a pathologic fracture [1, 4]. The exact aetiology of SBCs remains unclear, which significantly hinders the development of targeted treatments [5]. However, a wide variety of therapeutic procedures for the surgical treatment of SBCs have been described [6, 7]. These procedures range from minimally invasive techniques such as percutaneous decompression, bone marrow injection, or steroid injection up to open procedures such as curettage with or without bone grafting (autologous/allogenic/synthetic) [7–11]. Another

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treatment approach is to decompress the cyst by means of osteosynthesis using titanium elastic nails (TEN) while simultaneously splinting the fracture-prone bone [12, 13]. However, some of these procedures showed high recurrence rates [14]. Self-healing rates of 11 to 15% in conservatively treated cysts after pathologic fracture have been reported in the literature, although possible complications such as refractures or axial malalignment should not be disregarded [5, 6, 15]. Since SBCs are benign bone lesions which are generally assumed to heal or self-limit with the completion of skeletal growth, it is difficult to establish a standardized indication for surgical treatment or the extent to which this should take place [16, 17]. In most cases, persistent pain, a pathological fracture or the need for a histological assessment are used as the factors indicating a surgical procedure [7]. Identifying the optimal surgical procedure for SBCs of the humerus in children and adolescents, its success rate and any prognostic parameters would also facilitate standardization of indications. However, detailed comparisons of therapeutic strategies are sparse, and a clear consensus regarding the ideal surgical treatment is still lacking. The aim of this study was to evaluate and compare different surgical treatment modalities for SBCs of the humerus regarding their effectiveness, complications, and recurrence rate.

Methods

Ethical approval for this study was waived by the Ethics Committee of the City of Vienna because of the pure retrospective nature of this study. This study was completed in accordance with the Helsinki Declaration as revised in 2013.

In this monocentric retrospective study, the clinical charts of patients who underwent surgical intervention for a SBC of the humerus were examined. The inclusion criteria were an age at surgery of <18 years, the presence of a primary SBC in the humerus that had not previously received any treatment, confirmation of diagnosis by MRI or histologic examination, and a minimum follow-up of 12 months. Histological examination was conducted utilizing tissue samples obtained during the surgical procedure. The exclusion criteria were comorbidities associated with an increased risk of fracture and any previous humeral surgery.

The following parameters were obtained: sex, age, location, cyst volume (cm^3), minimum cortical thickness of the cyst (mm), previous fractures, type of surgical procedure, duration of postoperative immobilization, complications, recurrences, fractures, time until fracture (weeks), time until full load-bearing (weeks), and follow-up procedures.

To estimate the cyst volume as accurately as possible using two-dimensional radiographs in two different planes, the three measured diameters (length, width, height) of the

cyst were applied by using the equation for calculating the volume of an ellipsoid ($V = \frac{4}{3} \times \pi \times \frac{a}{2} \times \frac{b}{2} \times \frac{c}{2}$).

The initial surgical procedures were categorized as open procedure (curettage, allograft/synthetic bone graft/HA-cement, adjuvants/no adjuvants) and osteosynthesis alone (no further treatment of the cyst) and compared with respect to complications, fracture and recurrence rates, treatment failure, and time until full load-bearing of the treated limb. Under differentiated consideration of the open procedure group, four subgroups: (1) curettage, synthetic bone graft substitute, adjuvant; (2) curettage, allograft, adjuvant; (3) curettage, allograft; and (4) curettage, HA-cement could be defined. Curettage procedures were carried out under X-ray guidance to enhance the thorough removal of the cyst. Autologous bone marrow aspirate from the iliac crest was used as an adjuvant. Three experienced surgeons were involved in the surgical treatment of the patient population. The variety of different surgical techniques can be attributed to the continuous development of new products designed for the treatment and augmentation of bone cysts and to changes in surgeons' individual preferences over time. Recurrence was defined as progression of cyst growth or reappearance of a cyst after initial surgery. Treatment failure was defined as recurrence and/or fracture during the follow-up period. Primary outcome parameters were failure-free survival at two and five years and time until full load-bearing after initial procedure.

Statistical analysis

Statistical analysis was performed using IBM SPSS® 20.0. (IBM Deutschland GmbH, Ehningen, Germany). Descriptive statistics included mean (*M*), standard deviation (*SD*), median (*Md*), and interquartile range (*IQR*) for metric variables. For nominal scaled variables, frequencies (*n*) and the corresponding proportions (%) were calculated and presented. A cross-tabulation with chi-square testing was used to examine the association between treatment failure and treatment modality, which also required correction using Fisher's exact test. Non-parametric Kruskal-Wallis test was utilized to examine the difference of ordinal scaled data between more than two groups. Change in cyst volume was assessed by Wilcoxon matched-pairs signed-rank test. Kaplan-Meier functions were used to analyze failure-free survival as well as to assess the cumulative incidence of time until full load-bearing and were examined by log-rank testing. Cox proportional hazards model was utilized to examine the explanatory value of four predictors (age, cyst volume, pathologic fracture, and minimal cortical thickness—each at the time of diagnosis) for the occurrence of treatment failure (recurrence or fracture after intervention) to identify potential risk factors. These potential prognostic factors

were selected based on previously described study results [2, 6, 18].

Results

This study included 23 cases who underwent surgical treatment regarding a SBC of the humerus. Of these, 16 (69.6%) were male. The right side was affected more frequently, with a total of 14 (60.9%) cases observed. The mean age at diagnosis was 11.6 ± 2.5 ($Md = 11.6$, IQR 10.2; 12.6) years, The mean postoperative follow-up was 3.9 ± 2.6 (range 1.0–10.3) years. In the study population, the most affected site was the humeral diaphysis at 52.2%. The definitive diagnosis was confirmed by histologic examination in six (26.1%) cases and by MRI in five (21.7%) cases. In 12 (52.2%) cases, both procedures were utilized to confirm the diagnosis. At the time of diagnosis, 14 (60.9%) out of 23 patients presented with a fracture around the humeral cyst. A total of 11 (47.8%) cases had a previous fracture prior to initial presentation, with five (21.7%) patients having both a fracture at initial presentation and a previous fracture. No fractures were evident prior to intervention in three (13.0%) cases. The median cyst volume at diagnosis, calculated from radiographs, was 24.2 (IQR 15.6; 34.4) cm^3 (Table 1).

Observed procedures were categorized as open procedure (n=20) or osteosynthesis alone (n=3) using TEN. Postoperatively, all patients received immobilization. The median time of immobilization was three (IQR 2.1; 4.0, min 1.7–max 6.3) weeks. After the initial surgical intervention, treatment failure (recurrence or fracture) was observed in seven (30.4%) cases. A total of five (21.7%) patients showed at least one recurrence. The complication rate after initial surgery, excluding recurrences, was 17.3%. In three (13.0%) cases, a fracture occurred, two of which required further surgical intervention. The median time until fracture occurred after surgical intervention was 36.4 weeks. Table 2 shows the distribution of the different therapy methods, the number of recurrences, and the complications in a detailed breakdown.

The incidence of treatment failure (recurrence or fracture) was significantly higher in group 2 (curettage, allograft, adjuvant), with five (83.3%) of six cases, than in the other subgroups ($\leq 25.0\%$) including the osteosynthesis alone group ($p = .024$) (Table 3).

**Table 2** Frequencies and percentages of therapeutic procedures, recurrences, and complications

	Cases (n=23), n (%)
Osteosynthesis alone	
TEN	3 (13.0%)
Open procedure	
Curettage, allograft, adjuvants	6 (26.1%)
Curettage, allograft	6 (26.1%)
Curettage, synthetic bone graft substitute, adjuvants	4 (17.4%)
Curettage, HA-cement	4 (17.4%)
Number of recurrences	
0	18 (78.3%)
1	3 (13.0%)
2	2 (8.7%)
Complications	
Infection	1 (4.3%)
Fracture	3 (13.0%)

Table 1 Characteristic values of biometric and clinical parameters regarding the two treatment procedures

Parameter	Osteosynthesis alone	Open procedure	Total
Group size	3	20	23
Demographics			
Male, n (%)	3 (100%)	13 (65.0%)	16 (69.6%)
Age at diagnosis (y), $M \pm SD$	11.4 ± 1.2	11.6 ± 2.7	11.6 ± 2.5
Follow-up (y), Md (IQR)	2.8 (2.3; 5.0)	3.5 (1.4; 4.8)	3.4 (1.5; 4.8)
Localization			
Epiphysis, n (%)	0	1 (5.0%)	1 (4.3%)
Metaphysis, n (%)	1 (33.3%)	5 (25.0%)	6 (26.1%)
Meta-diaphysis, n (%)	0	4 (20.0%)	4 (17.4%)
Diaphysis, n (%)	2 (66.7%)	10 (50.0%)	12 (52.2%)
Fracture			
Prior, n (%)	2 (66.7%)	9 (45.0%)	11 (47.8%)
At diagnosis, n (%)	3 (100%)	11 (55.0%)	14 (60.9%)
Cyst size			
Est. volume (cm^3), Md (IQR)	9.1 (8.8; 23.9)	24.8 (17.4; 34.9)	24.2 (15.6; 34.4)
Min. cort. thickness (mm), $M \pm SD$	1.1 ± 0.4	1.0 ± 0.2	1.0 ± 0.3

Table 3 Frequencies and percentages of the occurrence of treatment failure and time until full load bearing in respect to the different procedures
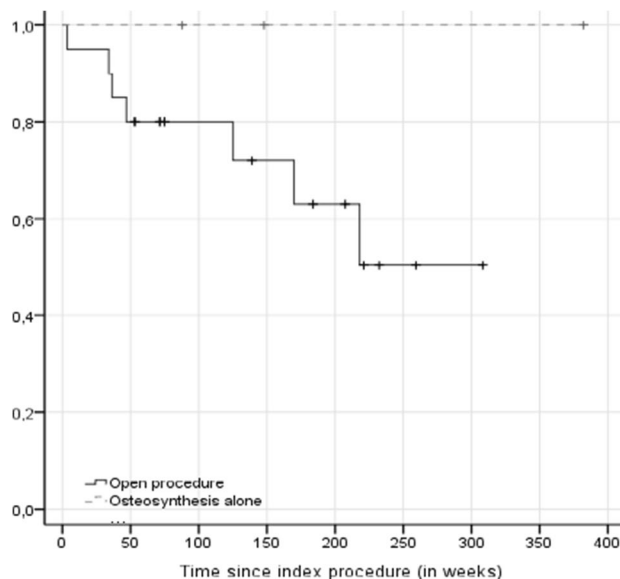
	Procedure					Total
	Osteosynthesis alone	Curettage, synthetic bone graft substitute, adjuvant	Curettage, allograft, adjuvant	Curettage, allograft	Curettage, HA-cement	
Total	3	4	6	6	4	23
Failure	0	1 (25.0%)	5 (83.3%)	1 (16.7%)	0	7 (30.4%)
Time LB (weeks), *Md*	4.1	3.0	5.9	7.1	5.6	4.9
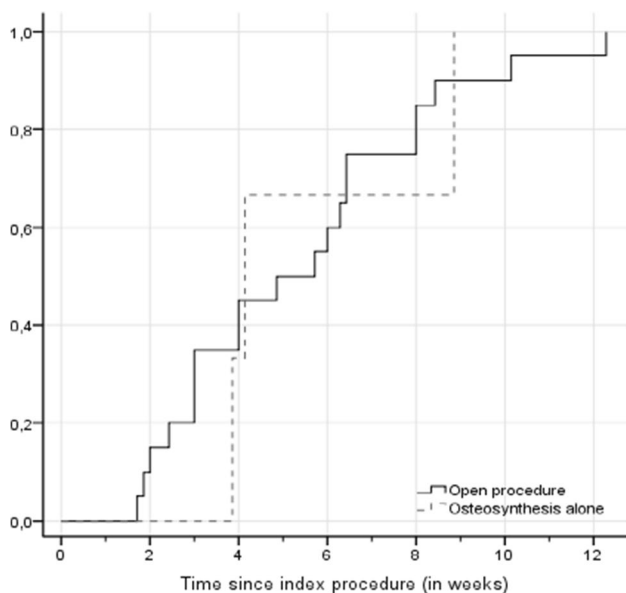
Fig. 1 Cumulative incidence of time until full weight bearing

Fig. 2 Kaplan-Meier function of failure-free survival considering treatment modality

The median time to full load-bearing after surgery was 4.9 (*IQR* 3.0; 7.2) weeks. Comparison of the groups in terms of time to full load-bearing using Kruskal-Wallis testing showed a non-significant result (*p* = .256). Full load-bearing of the operated limb was possible after four weeks in 45% and after eight weeks in 85% of the open procedure group. In the osteosynthesis alone group, it was possible in 33% after four weeks and 67% after eight weeks (Fig. 1). No significant difference in the progression of postoperative load-bearing between the two groups was detected by log-rank testing (*p* = .903).

For the open procedure group, the failure-free survival rates were 80.0% after two years and 50.4% after five years. For the three cases treated by osteosynthesis only, no failures were observed (Fig. 2). However, no significant difference in failure-free could be observed between the two groups by log-rank test (*p* = .278).

In model testing, no significant explanatory value for the criterion treatment failure could be detected among the four predictors (age, pathologic fracture, cyst volume,

minimal cortical thickness—each at the time of diagnosis), *p's* ≥ .228.

Cyst volume was reduced/improved in 20 (87.0%) cases from the time of diagnosis to the time of the last follow-up, while there was an increase/deterioration in three (13.0%) cases (*p* < .001; suggesting a moderate effect, *r* = .49). Of the 20 cases in which a reduction in cyst volume was observed, nine cases (39.1%) showed complete, healing of the cyst, with no apparent residual lesion. The median cyst volume was reduced from 24.2 (*IQR* 15.6; 34.4) cm^3 at diagnosis to 0.5 (*IQR* 0; 5.9) cm^3 at last follow-up. Overall, there was a median reduction of 20.4 (*IQR* 5.7; 27.8) cm^3.

Discussion

In this retrospective study, the outcome of 23 cases who underwent surgical treatment regarding a SBC of the humerus was analyzed; the different treatment procedures were compared with respect to treatment failure,

complications, fracture rates, and time until full load-bearing, and potential prognostic parameters were investigated. Combined open procedures involving curettage and the application of diverse filling materials exhibited comparable rates of treatment failure (≤ 25.0%) except for the subgroup curettage, allograft, and adjuvant which showed a significantly higher treatment failure (83.3%). Promising results were observed in cases treated exclusively with osteosynthesis using TENs without cyst excision or filling, as no treatment failure was seen in this particular cohort. Comparison of the groups in terms of time until full load-bearing revealed no substantial differences. It should be emphasized that the potential prognostic predictors for recurrence or refracture described in previous literature (age, pathologic fracture, cyst volume, minimal cortical thickness—all assessed at the time of diagnosis), could not be confirmed in the present study and did not provide any explanatory value for the outcome [2, 6, 18].

In the present study, the most frequently affected localization of the humerus was the humeral diaphysis (52.2%). This is a remarkably high proportion in contrast to the literature, where SBCs have predominately been documented at the humeral metaphysis. For instance, Green et al. reported a diaphyseal localization in 33% of cases in their 2019 analysis [6]. Similarly, Kadhim et al.'s 2016 publication indicated a diaphyseal involvement in 25% of cases [14]. These findings aligns with the study by Teoh et al. from 2010, wherein the prevalence of SBCs in the humeral diaphysis was noted at 18.8% [2]. Kadhim et al. showed in a systematic review and meta-analysis that the humeral diaphysis is implicated in a mere 14.3% of humeral SBC cases [17]. To which extent the different localizations on the humerus have an influence on the healing rate is still insufficiently supported by research.

In 14 of the 23 patients (60.9%), a pathological fracture existed in the region of the humeral cyst at the time of diagnosis. Only three cases (13.0%) did not present with a prior history of fracture at their initial assessment. This finding is consistent with existing literature, where proportions ranging from 69% to as high as 100% have been documented [1, 2, 6, 19, 20]. One plausible hypothesis for this high fracture rate is that the proximity to the growth plate in the humerus may lead to a deceleration in bone growth. Consequently, cyst expansion may outpace metaphyseal growth, culminating in a progressive weakening of the bone structure [20]. Another possible explanation could be that the increasing cortical thinning remains asymptomatic more often due to the reduced mechanical load on the upper extremity until a fracture occurs in the context of a trauma or sports participation.

The findings of this study demonstrate a notable reduction in cyst volume in 87% of cases. Furthermore, complete resolution of the cyst without visible residual signs was observed in 39.1% of patients. This significant improvement underlines the potential of the applied surgical treatment approaches and contrasts with previously described results where cyst expansion persisted despite medical intervention [21]. It is important to note, that this metric does not show the number of procedures required to achieve this outcome. To offer more robust evidence and facilitate comparability across various approaches, treatment failure, defined as the occurrence of fracture or recurrence subsequent to the initial surgical procedure, was adopted as an outcome parameter. The current study showed a rate of 30.4%. This is comparable with other studies. The investigation by Traub et al. in 2016 documented a surgical procedure failure rate of 26.1%, although it is worth noting that this study included a broader spectrum of bone cyst locations beyond the humerus [3]. A more differentiated evaluation of the various treatment modalities in the current study revealed only marginal disparities in terms of the restoration of full limb function postoperatively. However, a quite different pattern emerged when the individual techniques were assessed in terms of their treatment failure. It has been shown that the cohort subjected to curettage, allograft, and adjuvant involving autologous bone marrow aspirate from the iliac crest exhibited an exceptionally high treatment failure rate of 83.3%, manifesting substantially inferior outcomes compared to all other therapeutic interventions under investigation. In the literature, diverse failure rates for open surgical intervention at the humerus have been documented, reaching up to 64% [4, 16, 17, 22]. A plausible explanation for this may be found in the work of Canavese et al. from 2011, wherein the three therapeutic approaches of curettage, bone marrow injection, and the injection of methylprednisolone were compared. This investigation revealed that the bone marrow group exhibited notably suboptimal healing rates, measuring at 21% [23]. This assumption is underlined by the work of Gundle et al. who explored the application of bone marrow aspirate and demineralized bone matrix in humeral SBCs. Following a singular administration, the healing rate stood at a mere 22% [24].

Promising outcomes were observed in the cohort exclusively treated with osteosynthesis using TENs (Fig. 3). This procedure has the advantage of a minimally invasive approach and does not require excision of the cyst. The intramedullary nails break and open the cyst while concurrently stabilizing the fracture-prone bone, thus mitigating the risk of refractures This intervention facilitates continuous decompression of the cyst and drainage of cystic fluid, a mechanism described in the literature as a plausible explanation for the therapeutic efficiency of this technique [10, 25–27]. In the present study, no treatment failure was detected for this group, and a swift return to full load-bearing capacity was achieved in 4.1 weeks (Fig. 3). In a 2008 workup by Masquijo et al., a cure rate of 92.3% was described after intramedullary splinting for SBC of the

Fig. 3 **a** Preoperative antero-posterior X-ray showing **a** SBC located at the left humeral diaphysis in a 12-year-old boy with a pathologic fracture. **b** X-ray taken 2 weeks postoperatively, demonstrating progressive callus formation. **c** X-ray obtained 20 months postoperatively, after the removal of the metal implants, showcasing bony consolidation of the cyst

humerus. It should be noted that this work also compared healing rates between the humerus and femur. Superior outcomes were seen when the cyst was situated in the upper extremity, with a cure rate of 92.3% as opposed to 84.2% in the lower extremity [10]. Excellent results have also been reported by Glanzmann et al., Roposch et al., and Sanctis et al. with cure rates of 90.9%, 93.8%, and 100%, although it should be noted that these studies included both humeral and femoral cysts [12, 28, 29]. In a study conducted by Wang et al. in 2021, the cohort receiving intramedullary stabilization with TENs exhibited a notably lower level of postoperative pain compared to the comparison group, which was treated with curettage and filling alone [30]. Despite the excellent results, however, it should not be ignored that this procedure entails a secondary, although typically minor, intervention, namely the removal of the implanted material.

Based on the study results and their comparison with existing literature, a proposed treatment algorithm for SBCs of the humerus can be outlined (Fig. 4). First-time pathologic fractures without significant axial deviation may be managed conservatively. Surgical intervention becomes necessary if the fracture leads to notable axial deviation. After the occurrence of a second pathological fracture, surgical treatment is recommended in all cases. Additionally, surgical management should be considered even in cases without prior pathological fractures, if there is evidence of cyst progression or pain. While promising results have been observed with the use of TENs for stabilization without cyst removal and filling, definitive recommendations are currently not possible due to insufficient data. Nonetheless, the initial outcomes appear highly encouraging.

This study is not without its limitations, primarily stemming from its retrospective design and the relatively small

Fig. 4 Treatment algorithm for unicameral bone cysts of the humerus
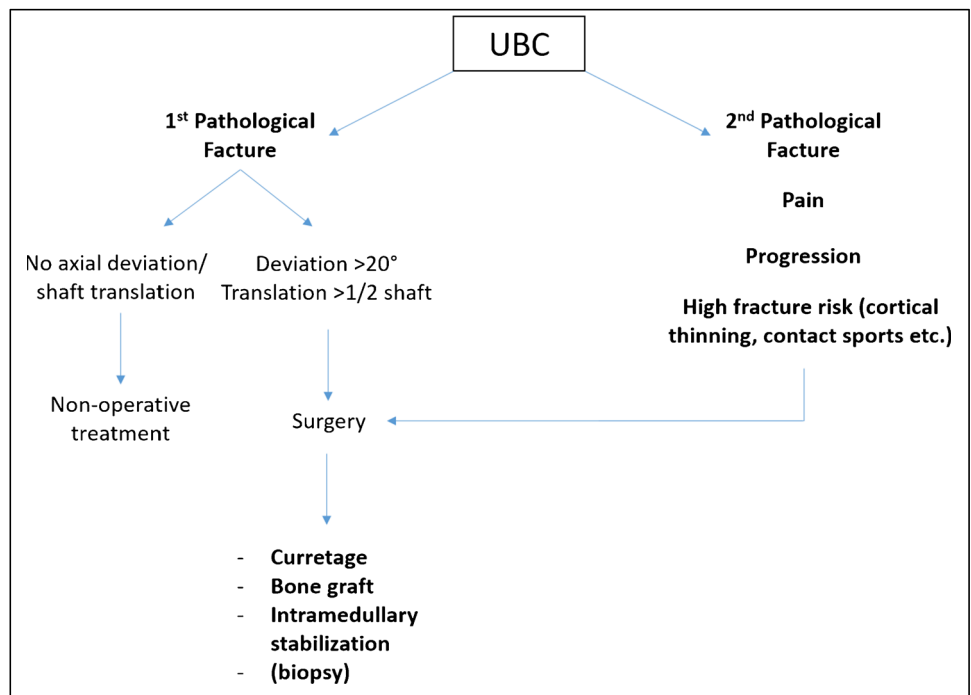
sample size for each individual treatment modality. The abundance of diverse therapeutic approaches, along with their ongoing refinement for treating SBCs, presents a significant challenge in amassing larger cohorts for a comprehensive comparative analysis of these methodologies. The minimum follow-up period was established at 12 months. While many patients were monitored for considerably longer durations, there exists the potential for overlooking a recurrence that may have occurred after the conclusion of the follow-up assessments. Nevertheless, given that all patients at our institution are informed of this possibility prior to the conclusion of treatment, the associated risk is deemed to be relatively low.

In summary, open procedures demonstrated comparable failure rates across most subgroups, except for the subset employing curettage, allograft, and adjuvant, which exhibited a notably elevated treatment failure rate. A noteworthy contribution towards establishing a consensus on the optimal surgical treatment can be provided by the promising results achieved, though in a small subset of cases, through a strategy exclusively centered on breaking and splinting of the cyst via TENs, as this approach showed a complete absence of treatment failures.

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