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Multifactorial retrospective analysis of pathological fracture risk and treatment strategies in unicameral bone cysts

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ABSTRACT

Unicameral bone cysts (UBCs) are benign fluid-filled bone lesions that primarily affect children and adolescents and carry a clinically relevant risk of pathological fracture. This study explored radiological and clinical variables associated with fracture and recurrence in a histologically confirmed retrospective cohort. Seventy-two patients with histologically confirmed UBCs treated between 2010 and 2021 were retrospectively analysed. Median follow-up was 38 months (range 24-132). Primary outcomes were pathological fracture and radiological recurrence. Because only eight fractures were observed, multivariable analyses were treated as exploratory. Pathological fractures occurred in 8/72 patients (11.1%). Fracture rates were 0/46 (0%), 2/20 (10%) and 6/6 (100%) for Capanna stages 1, 2 and 3, respectively ($p < 0.001$). Latent cysts had a higher fracture rate than active cysts (21.4% vs 4.5%; $p = 0.049$). Recurrence occurred in 7/71 evaluable patients (9.9%), exclusively in the interventional group (7/23, 30.4% vs 0/48; $p < 0.001$). In this retrospective, histologically selected cohort, higher Capanna stage, younger age, latent activity and diaphyseal localisation were associated with fracture risk. Given the limited number of fracture events, the mixed paediatric-adult population and the selection bias inherent to histological inclusion, these findings are best interpreted as exploratory and hypothesis-generating. Overall, they may be more compatible with structured, individualised risk assessment than with a prescriptive Capanna-guided treatment algorithm.

Keywords: Inicameral bone cyst, pathological fracture, recurrence, Capanna classification, cyst index, diaphyseal localisation, paediatric orthopaedics.

INTRODUCTION

Unicameral bone cysts (UBCs), also known as simple bone cysts, are benign osteolytic lesions accounting for approximately 3% of all primary bone tumours[1]. They arise predominantly within the first two decades of life, most commonly in the metaphyseal regions of the proximal humerus and femur, and carry a clinically significant risk of pathological fracture, which is the presenting feature in 15-50% of cases[2]. Historical concepts of cyst pathogenesis and decompression-based treatment have also been described[3].

The proximity of the cyst margin to the growth plate classifies lesions as active (<1 cm from the physis, biologically expanding) or latent (≥1 cm, migrated with skeletal growth)[1]. The Capanna radiological classification further stratifies aggressiveness by degree of cortical expansiveness and has been proposed as a guide to treatment escalation[4]. Natural-history studies have also highlighted the relationship between lesion behaviour and fracture risk[5]. Comparative paediatric treatment series have reported heterogeneous results across operative strategies[6,7].

Fracture-risk prediction based on imaging variables has also been investigated[8]. Recent pooled evidence underscores the heterogeneity of treatment outcomes across large cohorts[9]. Some authors have suggested that active upper-limb cysts may carry higher fracture risk, although anatomical context may modify this association[10]. The cyst index (CI), defined as the ratio

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of a projected radiographic cyst area estimate to the square of the diaphyseal diameter, quantifies structural cortical compromise and has been proposed as a threshold-based tool for surgical planning[11].

The aims of this retrospective cohort study were: (1) to identify variables associated with pathological fracture; (2) to characterise recurrence patterns; (3) to explore correlations between radiological indices; and (4) to describe treatment distribution in the context of lesion severity.

MATERIALS AND METHODS

Patients

This single-centre retrospective cohort study was conducted at Ataturk University Hospital, Erzurum, between March 2010 and March 2021. Four hundred and thirty-two patients with radiographic cystic bone lesions were screened. Following advanced imaging (MRI and/or CT) and histopathological confirmation by biopsy or surgical specimen, 72 patients with a definitive UBC diagnosis were included.

Inclusion criteria were histologically confirmed UBC, age 3-72 years at diagnosis, a minimum of 24 months of radiological follow-up, and complete clinical and imaging documentation. Exclusion criteria were other histopathological diagnoses (aneurysmal bone cyst, enchondroma, fibrous dysplasia, osteochondroma, intraosseous lipoma and giant cell tumour), loss to follow-up, and external referral without baseline data.

Histopathological confirmation was required in this retrospective screening cohort because the source population included a broad spectrum of cystic and lytic bone lesions with overlapping imaging appearances, and the study objective was to analyse only diagnostically secure UBC cases. We recognise that this inclusion strategy may preferentially capture lesions that were symptomatic, surgically treated or diagnostically uncertain, thereby introducing selection bias and limiting external validity. Ethical approval was granted by the Ataturk University Faculty of Medicine Ethics Committee (Meeting No: 06, Decision No: 14, 30.09.2021). Informed consent was waived because of the retrospective design.

Variables

Demographic data comprised age (continuous and dichotomised at <18 years) and sex. Radiological variables were: (1) cyst activity, active (margin <1 cm from the growth plate) or latent (≥1 cm), applied primarily in skeletally immature patients; in adults with closed physes, activity status was retained as a pragmatic radiographic descriptor rather than a validated biological classification; (2) Capanna stage, categorised as stage 1, 2 or 3; and (3) cyst index (CI), calculated from a projected radiographic area estimate obtained by trapezoidal approximation on orthogonal radiographs, divided by the squared diaphyseal diameter. Thresholds of >4.0 for the humerus and >3.5 for the femur were considered to indicate increased mechanical fracture susceptibility.

Anatomical localisation (metaphyseal, diaphyseal or epiphyseal) was classified by the same two observers.

Outcomes

The primary outcome was pathological fracture at any time during follow-up. Secondary outcomes were radiological recurrence, defined as re-expansion of the cyst cavity beyond its post-treatment boundaries or reappearance of a lytic defect at the same site on serial plain radiographs irrespective of symptoms, and the treatment modality applied.

Statistical analysis

All analyses used SPSS version 25.0. Normality was tested by Shapiro-Wilk; all continuous variables were non-normal (all $p < 0.001$). Continuous variables are presented as median (IQR) and compared with the Mann-Whitney U test for two groups or the Kruskal-Wallis test for three or more groups. Categorical variables are presented as n/N (%) and compared using the chi-square or Fisher exact test, as appropriate. Correlations between continuous or ordinal radiological variables were assessed using Spearman rho.

For fracture analysis, only eight events were available. Univariate associations therefore constituted the primary analysis, and any multivariable modelling was considered exploratory. A Firth penalised logistic regression restricted a priori to two clinically motivated predictors (age, continuous, and Capanna stage, ordinal) was used only to reduce small-sample bias and to avoid over-interpretation of sparse-event data. Two-sided p values <0.05 were considered statistically significant.

RESULTS

Cohort characteristics

The cohort comprised 32 males (44.4%) and 40 females (55.6%). Age at diagnosis ranged from 3 to 72 years (median 25.5, IQR 17.0-40.0), and 21 patients (29.2%) were younger than 18 years. Median follow-up was

38 months (range 24-132). Demographic, radiological and treatment characteristics are summarised in Table I; bone distribution is shown in Table II.

The most frequently affected bones were the femur (14/72, 19.4%), humerus (12/72, 16.7%), calcaneus (11/72, 15.3%) and tibia (10/72, 13.9%). Anatomically, 50 lesions (69.4%) were metaphyseal, 18 (25.0%) diaphyseal and 4 (5.6%) epiphyseal. By Capanna stage, 46/72 lesions (63.9%) were stage 1, 20/72 (27.8%) stage 2 and 6/72 (8.3%) stage 3. Cyst activity was active in 44/72 patients (61.1%) and latent in 28/72 (38.9%). Median CI was 1.3 (IQR 0.5-3.8). Inter-observer ICC for CI was 0.91 (95% CI 0.86-0.95).

Pathological fractures

Pathological fractures occurred in 8/72 patients (11.1%). The distribution across Capanna stages was 0/46 (0%) in stage 1, 2/20 (10%) in stage 2 and 6/6 (100%) in stage 3 ($\chi^2 = 53.775$; $p < 0.001$). Fracture rates were also higher in latent than in active cysts (21.4% vs 4.5%; $p = 0.049$) and in diaphyseal than in metaphyseal/epiphyseal lesions (33.3% vs 3.7%; $p = 0.002$). These findings are reported as associations within this retrospective cohort and are best interpreted cautiously given the small number of fracture events. Median CI was higher in fractured cases (3.8, IQR 2.6-5.2) than in non-fractured cases (1.2, IQR 0.4-3.5), but this did not reach statistical significance ($U = 152.0$; $p = 0.101$). The pre-specified CI threshold (>4.0 for the humerus or >3.5 for the femur) had a sensitivity of 62.5% (5/8) and a specificity of 89.1% (57/64) for fracture in this cohort.

In the exploratory Firth penalised logistic regression (age plus Capanna stage), Capanna stage was associated with fracture (penalised OR per stage increase 9.2; 95% profile-likelihood CI 2.8-31.4; $p = 0.003$), and younger age showed a trend towards higher fracture risk (penalised OR per year 0.94; 95% CI 0.88-1.00; $p = 0.061$). Because only eight

Table I. — Demographic, radiological and treatment characteristics of the cohort.

Characteristic	n/N	%
Sex		
Male	32/72	44.4
Female	40/72	55.6
Age at diagnosis		
<18 years	21/72	29.2
≥18 years	51/72	70.8
Pathological fracture		
Yes	8/72	11.1
No	64/72	88.9
Cyst activity		
Active	44/72	61.1
Latent	28/72	38.9
Anatomical localisation		
Metaphyseal	50/72	69.4
Diaphyseal	18/72	25.0
Epiphyseal	4/72	5.6
Capanna stage		
Stage 1	46/72	63.9
Stage 2	20/72	27.8
Stage 3	6/72	8.3
Treatment group		
Conservative	48/72	66.7
Interventional	24/72	33.3
Radiological recurrence*		
Present	7/71	9.9
Absent	64/71	90.1

*One patient was excluded from recurrence analysis because of incomplete follow-up.

Table II. — Skeletal distribution of unicameral bone cysts.

Bone	n/N	%
Femur	14/72	19.4
Humerus	12/72	16.7
Calcaneus	11/72	15.3
Tibia	10/72	13.9
Pelvis	6/72	8.3
Fibula	6/72	8.3
Metacarpal	3/72	4.2
Lunate	2/72	2.8
Metatarsal	2/72	2.8
Ulna	1/72	1.4
Talus	1/72	1.4
Scaphoid	1/72	1.4
Capitate	1/72	1.4
Phalanx	1/72	1.4
Scapula	1/72	1.4

fractures were observed, this model is best regarded as exploratory rather than definitive.

Radiological index correlations

Capanna stage was inversely correlated with age (rho = -0.539; $p < 0.001$), indicating higher-stage lesions in younger patients. CI correlated positively with Capanna stage (rho = 0.618; $p < 0.001$) and with cyst area (rho = 0.678; $p < 0.001$). Median CI increased significantly across stages (stage 1: 0.9; stage 2: 3.8; stage 3: 4.1; $H = 27.665$; $p < 0.001$; stage 1 vs 2, $p < 0.001$; stage 1 vs 3, $p = 0.005$).

Treatment

Treatment modalities are summarised in Table I. Conservative management (observation or biopsy with observation) was used in 48/72 patients (66.7%), whereas operative interventions (curettage with or without grafting/fixation, steroid-based injection or elastic intramedullary stabilisation) were used in 24/72 (33.3%). Surgical management was more frequent in stage-3 lesions (6/6, 100%) than in stage-2 lesions (7/20, 35.0%) or stage-1 lesions (11/46, 23.9%; $\chi^2 = 12.542$; $p = 0.005$). These treatment distributions are descriptive and reflect clinician judgement in relation to lesion severity, symptoms and perceived mechanical risk.

Recurrence

Radiological recurrence was identified in 7/71 evaluable patients (9.9%); one patient was excluded because of incomplete follow-up. All seven recurrences were observed among patients who had undergone an intervention (7/23, 30.4% vs 0/48, 0%; $p < 0.001$). Because treatment allocation was non-random and severity-driven, these recurrence distributions are best interpreted descriptively and not as evidence of comparative treatment effect.

In univariate recurrence analysis, younger age (median 18 years in recurrent vs 28 years in non-recurrent; $p = 0.036$) and male sex (OR 9.36; 95% CI 1.06-82.46; $p = 0.038$) were associated with recurrence. Capanna stage showed a non-significant trend ($p = 0.090$). These findings are exploratory and may reflect case-mix rather than independent biological determinants.

DISCUSSION

This retrospective study of 72 histologically confirmed UBC patients suggests that fracture risk may cluster in lesions with higher Capanna stage, younger patient age, latent activity and diaphyseal localisation. These associations should nevertheless be interpreted cautiously. The small number of fracture events limits precision, and the present dataset is better suited to exploratory clinical pattern recognition than to definitive prediction modelling.

The marked distribution of fractures across Capanna stages in our cohort supports the clinical relevance of structural severity, but does not justify using stage alone as a stand-alone management algorithm. Because Capanna stage itself incorporates cortical expansion and structural compromise, part of its association with fracture likely reflects definitional overlap with mechanical fragility rather than wholly independent prognostic discrimination. This cautious interpretation is broadly consistent with the findings

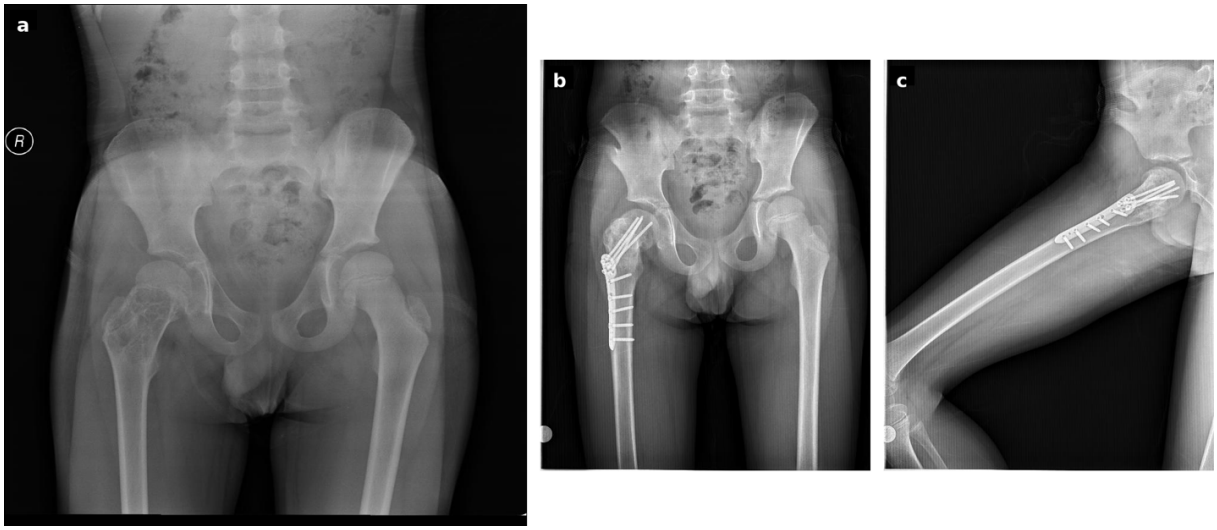

Fig. 1 — Representative case of a unicameral bone cyst in the proximal femur, managed with curettage, iliac crest autologous bone grafting and internal fixation. (a) Preoperative anteroposterior pelvic radiograph showing an expansile lytic lesion of the proximal femur with cortical thinning and loss of medullary architecture, consistent with a high-risk lesion. (b) Postoperative anteroposterior radiograph confirming satisfactory bone graft position and plate-and-screw fixation with appropriate axial alignment. (c) Follow-up radiograph at 24 months demonstrating graft incorporation, cortical reconstitution and maintained hardware position without recurrence.

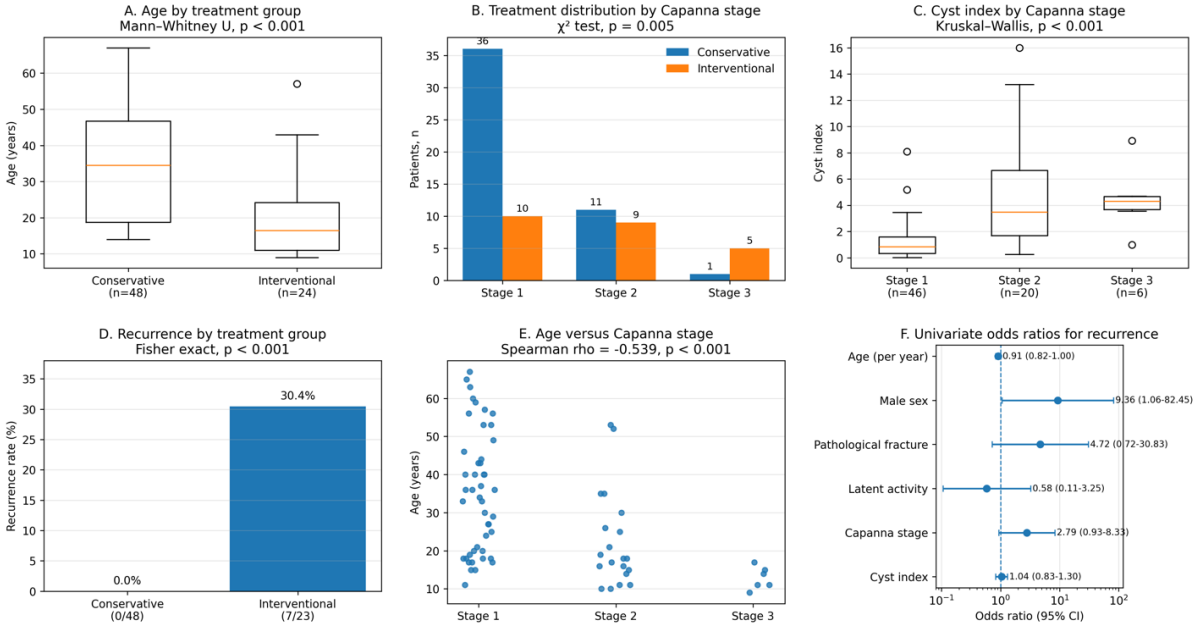

Fig. 2 — Statistical analysis panels for 72 patients with unicameral bone cysts. (A) Age by treatment group; Mann-Whitney U, p < 0.001. (B) Treatment distribution by Capanna stage; χ² test, p = 0.005. (C) Cyst index by Capanna stage; Kruskal-Wallis test, p < 0.001. (D) Recurrence rate by treatment subgroup; all 7/71 recurrences occurred in the interventional group (Fisher exact test, p < 0.001). (E) Age versus Capanna stage; Spearman rho = -0.539, p < 0.001. (F) Forest plot of univariate odds ratios for recurrence; male sex was significantly associated (OR 9.36; 95% CI 1.06-82.46; p = 0.038). CI, confidence interval; IQR, interquartile range.

reported by Pireau et al., who identified the bone cyst index as the most informative imaging predictor of fracture risk, and by Docquier and Delloye, who framed treatment selection in children around radiographic fracture-risk assessment rather than fixed procedural rules[12]. In that sense, our data may be more useful for refining risk estimation than for prescribing a single stage-based intervention strategy.

The association between latent activity and fracture warrants a more nuanced interpretation. Some

paediatric series, particularly in upper-limb lesions, have reported higher fracture risk in active cysts close to the physis[10]. Our finding in the opposite direction does not necessarily contradict that literature. In a mixed paediatric-adult cohort, the active/latent classification is inherently less specific once the physes are closed, and activity status may interact with anatomical site and mechanical loading. In addition, many latent lesions in our cohort were diaphyseal or otherwise structurally vulnerable, which may have shifted the observed risk signal away from physeal proximity and toward local mechanical context. Accordingly, this association may represent a cohort-specific signal shaped by mixed-age inclusion and should not be interpreted as evidence overturning established paediatric biological paradigms without external validation. This interpretation remains hypothesis-generating and should be confirmed in larger multicentre cohorts.

The non-significant cyst index finding also deserves careful contextualisation. Directionally, the higher median CI in fractured patients is in keeping with prior radiographic studies, including the report by Pireau et al., which emphasised structural imaging markers in fracture-risk assessment[8]. The absence of formal statistical significance in our cohort is likely explained by limited power, as only eight fracture events were available for analysis. Moreover, CI correlated positively with Capanna stage and inversely with age, suggesting that these variables may capture overlapping aspects of lesion severity. Under such circumstances, lack of statistical significance should not be equated with lack of clinical relevance; rather, CI may still provide complementary information when interpreted alongside age, stage and anatomical site.

Treatment allocation was strongly influenced by lesion severity, location, symptoms and perceived mechanical risk. Accordingly, the higher recurrence frequency in the interventional group should not be interpreted as evidence of treatment-related harm or lower efficacy; it more likely reflects confounding by indication and closer surveillance in higher-risk lesions. Recent pooled analyses and paediatric treatment series likewise emphasise heterogeneity of outcomes across interventions, arguing against simple one-variable treatment rules[6,7,9]. The present data therefore describe treatment patterns and outcome distributions within a real-world retrospective cohort, but do not support causal comparison between management strategies[13,14].

Several limitations must be acknowledged. The small fracture event count limits multivariable precision and precludes stable predictive modelling. The histopathological inclusion criterion, while improving diagnostic certainty, likely selected a subgroup enriched for diagnostically uncertain, symptomatic or surgically treated lesions, thereby limiting external validity. The mixed paediatric-adult design also restricts direct interpretation of the active/latent descriptor across all patients.

In summary, higher Capanna stage, younger age, latent activity and diaphyseal localisation were associated with fracture in this cohort, but these findings are best viewed as exploratory rather than prescriptive. Taken together with the existing literature, the present data may support an individualised, risk-adapted approach in which Capanna stage, cyst index, age and anatomical context are considered jointly, rather than using any single radiographic variable as a sole determinant of prophylactic intervention.

Ethics approval: Approved by the Ataturk University Faculty of Medicine Ethics Committee (Meeting No: 06, Decision No: 14, Date: 30.09.2021). The study was conducted in accordance with the Declaration of Helsinki. Informed consent was waived because of the retrospective design.

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Authors' contributions: Y.S.D. conceived and designed the study, collected and analysed the data, and drafted the manuscript. B.K., S.T. and K.T. contributed to data collection and critical revision. All authors approved the final version.

Data availability: Available from the corresponding author on reasonable request.

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