

# The Role of Surveillance in Predicting Fracture in Pediatric Patients with Incidentally Discovered Unicameral Bone Cysts: Is it worth it?

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**INTRODUCTION:** Unicameral bone cysts (UBCs) make up only 3% of all bone lesions, and 85% of these lesions occur in children or adolescents. UBCs may present with fracture or as an incidental finding. The focus of this study is on those that present as an incidental finding with respect to the value of surveillance in determining patients that will fracture and/or require surgical intervention. Surveillance of incidentally discovered UBCs is utilized to identify a point in the lesion progression where a patient may be at risk for pathologic fracture. Previous research has not clearly delineated an optimal longitudinal risk assessment protocol for incidentally discovered UBCs in pediatric patients. For any given anatomic location, there has been no prior comparison of fracture rates per anatomic site overall vs those discovered incidentally. The proximal humerus and proximal femur are common pediatric locations for UBC, but the likelihood of fracture for those that are discovered incidentally is unknown. We hypothesized that (a) the risk of fracture for incidentally discovered UBC is less than the overall percentage of UBC that present with pathologic fracture for any given anatomic site, (b) no particular clinical characteristics gained from surveillance would predict subsequent fracture, and (c) resources devoted to surveillance for incidentally discovered lesions may not be warranted.

**METHODS:** A retrospective chart review was accomplished for all patients with UBC diagnosed between January 1, 2012, and February 1, 2021. The ICD-10 codes for solitary bone cysts and bone lesions yielded 1661 patients who were further filtered using the inclusion criteria of being younger than 18 years of age, possessing radiographic or clinical documentation of a UBC diagnosis, and having at least one follow-up. One-hundred twenty UBCs were identified, 67 as incidental findings and 53 presenting with a pathologic fracture. Locations of the 53 pathologic fractures were recorded for calculation of comparative fracture risk per anatomic site overall vs that for incidentally discovered lesions (Figure 1b). Of the 67 patients' incidental UBC lesions, there were 44 males and 23 females with a median age of 13 years (mean:12.14, range: 2-17). Incidental UBCs occurred in the calcaneus (25), femur (21), humerus (9), ilium (6), fibula (4), tibia (1), and ischium (1). Lesions were incidentally discovered due to injury-related acute pain (22), pain attributed to the lesion (17), unrelated musculoskeletal diagnosis (11), or were asymptomatic (15), had indeterminate pain (1), or had unknown symptoms (1). Initial visit symptoms were examined as a possible clinical characteristic with predictive value of fracture or surgery. The imaging modalities leading to incidental discovery included X-ray (62), MRI (3), and CT (2). The two primary endpoints were subsequent fracture or surgery following incidental UBC diagnosis.

**RESULTS SECTION:** Mean follow-up (FU) duration for the incidentally diagnosed UBCs was 15.23 months (SD: 14.57, range: 0.23-62.63). Over the course of follow-up, 13 of 67 patients (19.4%) went on to have prophylactic surgery at a median 2.53 months (mean: 3.40, range: 0.57-9.07) after initial diagnosis for pain attributed to the lesion (10/13), increased lesion size, and perceived increased fracture risk. Also, over the course of FU, two (3.0%) pathologic fractures occurred and were treated non-operatively. One pathological fracture was through a midshaft humeral UBC due to trauma from a fall 29.8 months after discovery, while the other was through a UBC in the ilium due to low impact trauma 26.6 months after discovery. Over the course of FU, the 67 patients with incidentally discovered UBCs underwent 221 clinical visits, 227 imaging tests (138 X-ray, 74 MRI, 4 CT, 6 bone scan), three core biopsies, and two open biopsies. There was a median of 3 clinical follow-up visits per patient (mean: 3.30, range: 0-10) and a median three follow-up tests per patient (3.39, 1-12). A median of two X-rays (2.06, 0-8) and one MRI (1.10, 0-4) were obtained per patient. Furthermore, the UBCs that present with fractures seem to contrast to the incidentally discovered in the same bone locations. Comparing initial presenting fracture rates to incidentally discovered UBCs, 80.4% of humeral lesions presented with fracture while only 11.1% of incidentally discovered subsequently fractured. The difference between femoral lesions was 30.0% vs 0%. No ilium or calcaneal UBC presented with a fracture, and only 1/6 (16.7%) ilium incidental UBC subsequently fractured. All 25 calcaneal lesions were found incidentally. In addition, 3/9 (33.3%) of the incidentally discovered humeral lesions, 7/21 (33.3%) femoral lesions, 2/25 (8.0%) calcaneal lesions, and the one tibial UBC underwent further prophylactic surgery. No ilium, fibular, or ischium UBC underwent further surgery.

**DISCUSSION:** Considerable resources were devoted to the follow-up of these lesions during which time 2 pathological fractures occurred (one of which was due to a high impact trauma), and an additional 19.4% underwent surgery to prevent pathologic fracture and/or treat lesional pain. Furthermore, UBC bone location data supports our hypothesis that the risk of fracture for incidentally discovered UBC is less than the overall percentage of UBC that present with pathologic fracture for any given anatomic site. Utilization of clinical features may also prove beneficial in predicting future surgery as 10/17 (58.8%) of patients presenting with lesional pain went on to receive eventual prophylactic surgery while only 1/17 (5.9%) went on to fracture. The potential number of preventable fractures, the sum of lesions receiving prophylactic surgery and the two with eventual fracture, in our cohort is 15 (22.0%). The report by Urukawa et al. of 95 incidental UBCs was not restricted to pediatric patients, making this the first study to analyze incidentally discovered UBCs solely in pediatric patients [1]. One other study by Ahn et al. [2] formulated UBC imaging characteristics in pediatric patients to identify the patients at risk of fracture, but these studies have not established whether prospective use of those criteria is of any clinical relevance in incidentally discovered UBCs. We conclude that it is likely not necessary for most patients that present with an incidental UBC to undergo routine surveillance, unless the lesion is discovered in association with lesional pain on the first visit, as this could indicate need for future prophylactic surgery.

**SIGNIFICANCE/CLINICAL RELEVANCE:** Routine UBC surveillance has been considered a crucial aspect of management given the frequency with which fractures occur in some anatomic sites such as the humerus, but the rate of subsequent fracture and prophylactic surgery in UBCs that are incidentally discovered was only 19.4% of patients. Surveillance may be warranted for lesional pain on presentation due to increased risk for future prophylactic surgery.

**REFERENCES:** [1] Urukawa et al, 2014. BMC Musculoskeletal Disorders. 15:159. [2] Ahn et al, 1994. International Orthopaedics. 18(1).

**IMAGES AND TABLES:**



