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Case Report

Multifocal Unicameral Bone Cysts in a Middle-Aged Patient Treated with Three Different Methods: A Rare Case Report

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Abstract

Background: Primarily diagnosed in the first two decades of life, unicameral bone cysts (UBCs) are benign cystic lesions of the bone. More than 90% of UBCs are located in the proximal femur or proximal humerus of skeletally immature patients, however in this case report we report a middle-aged patient with multiple lesions in unusual sites.
Case Report: A 42-year-old patient presented to our institute with multiple bony lesions in the right and left tibias, proximal radius, ulna, and the second metacarpus. The lesions were later pathologically proven to be UBCs. A multimodality treatment approach was selected including decompression-only, extended curettage, bone grafting, and prophylactic fixation based on the lesion size and anatomic location. All these methods proved to be effective with no disease relapse and complete radiographic obliteration of the cavities.
Conclusion: UBCs could occur in much older patients than generally believed and the proper method of treatment should be individualized based on the lesion's characteristics.

Keywords: Case Reports; Cysts; Bone Cysts

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Background

Initially reported by Virchow as "cystic structures" (1), unicameral bone cysts (UBCs), also known as "simple" or "solitary" bone cysts, are benign, fluid-filled cavities that principally emerge in tubular bones (2). Being the only genuine cysts of the bone, they are diagnosed predominantly in childhood and adolescence, with a peak at the age of 10, and if left untreated, more than 85% persist or enlarge (3). More than 95% of the lesions are diagnosed in long bones, and the proximal humerus and femur are the most prevalent sites accounting for almost 90% of these cases (1). The main etiology of these cysts is currently unclear. However, several reasons have been proposed as the possible etiology, including but not limited to a pathologic response to bone trauma (4) or an intraosseous synovial cyst that developed as a result of local venous obstruction as suggested by Cohen (5). Due to the benign essence of this type of cysts, patients are usually asymptomatic and accidentally diagnosed or referred with localized rather than systemic complaints such as pain or swelling. UBCs are almost always solitary lesions with few reports of multifocal cases (3). We herein report a rare case of multifocal UBCs in a middle-aged patient whose peculiarity lies in both the patient's age at presentation and the number and sites of lesions that, to the best of our knowledge, have no similar reports in the literature.

Case Report

Our patient was a 42-year-old woman who was referred to the outpatient clinic of our center in February 2018 with a chief complaint of persistent dull pain and mild local swelling in her left leg since a year ago. Her previous medical history was unremarkable except for mild hypertension controlled through lifestyle modification. She used no medications, denied any major trauma, and her past surgical history was also negative. The physical examination revealed mild bony tenderness in the left leg with normal neurovascular findings. A plain roentgenogram of the affected limb revealed a well-defined expanding cystic lesion in the tibial diaphysis with thin sclerotic margins and endosteal thinning but no periosteal reaction. The patient underwent tibial cyst resection and curettage, revealing a straw-colored fluid. No bone graft or bone graft substitute was utilized in the procedure. Histopathologic examination showed a thin fibrous membrane with a calcified, flakey cement-like bone matter with no signs of malignancy suggestive of a simple bone cyst. The post-operative period was uneventful, and shortly after that, the complete function of the limb returned. 14 months after the index surgery, the patient sought to consult for the same symptoms, this time in the right leg as well as her left forearm and left hand. A radiological skeletal survey revealed expansile cystic bony lesions in the tibial diaphysis, proximal ulna, radius, and second metacarpal bone with cortical thinning but no obvious destruction (Figure 1).

A whole-body technetium-99m bone scintigraphy was obtained and turned out to be "photopenic" in the affected areas as well as the whole bony skeleton. Blood works including erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP), alkaline phosphatase, serum calcium, phosphorus, and vitamin D levels were all within the normal limits. The rheumatoid panel was also negative; serum hormone levels, including parathyroid hormone and thyroid-stimulating hormone (TSH), were uniformly within the expected range.

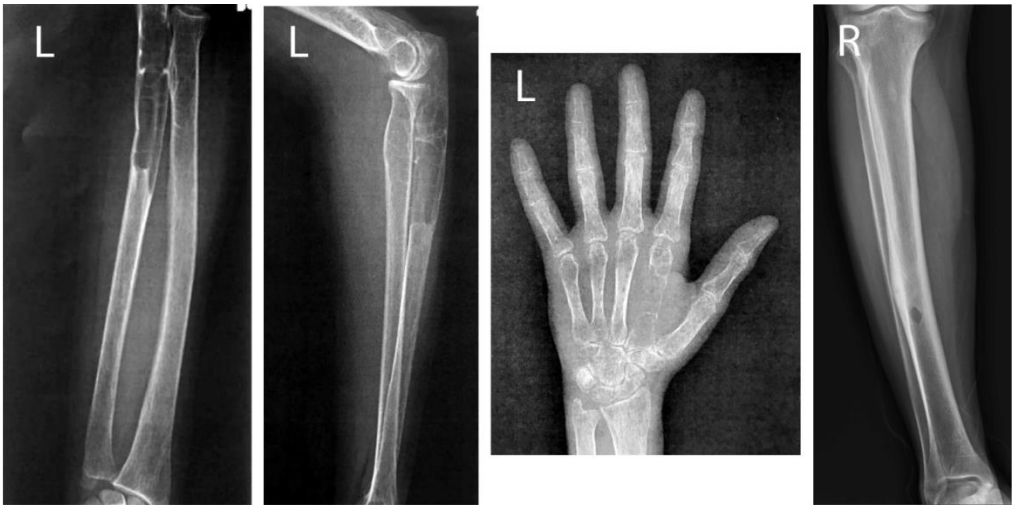

Figure 1. Roentgenograms of the left upper limb and right lower limb showing cystic bony lesions

Serum and urine protein electrophoresis was normal. Due to the multiple numbers of cysts, a surgical treatment plan was chosen. The right tibial diaphyseal cyst was treated with extended curettage using electrocautery with no bone graft materials. The metacarpal cyst was approached via a dorsal incision, and due to the large lesion, a fibular strut allograft was cut to the exact size of the lesion and placed longitudinally inside the cystic lesion after curettage. No type of internal fixation was utilized to hold the allograft in place, and instead, a volar wrist and hand splint was applied. Considering the large dimensions of the proximal ulnar cyst and the high probability of a secondary pathologic fracture, it went through fibular strut allograft augmentation and prophylactic fixation with a 3.5-mm anatomical proximal ulnar locking plate following decompression and curettage through a direct posterior approach (Figure 2).

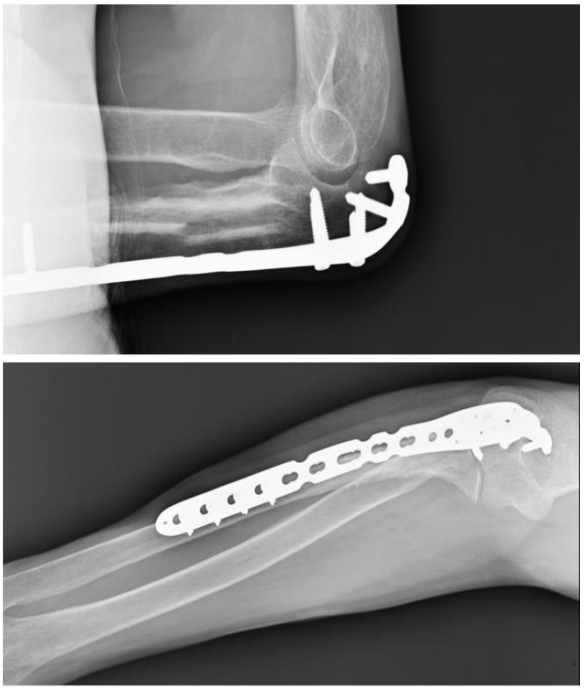

Figure 2. Proximal ulnar lesion prophylactically fixed and augmented using fibular allograft

During this procedure, the radial neck cyst was decompressed by making two drill holes through the lateral wall of the ulnar cyst under image intensification with no further interventions. Histopathologic and cytological fluid examination of all four anatomic sites, namely the right tibia, left ulna and radius, and the left second metacarpus, showed no signs of malignancy with features consistent with simple bone cysts. A 24-month follow-up of the patient was uneventful with no signs of relapse and near-complete obliteration of the cavities on control radiographs (Figure 3). She is currently symptom-free and doing well.

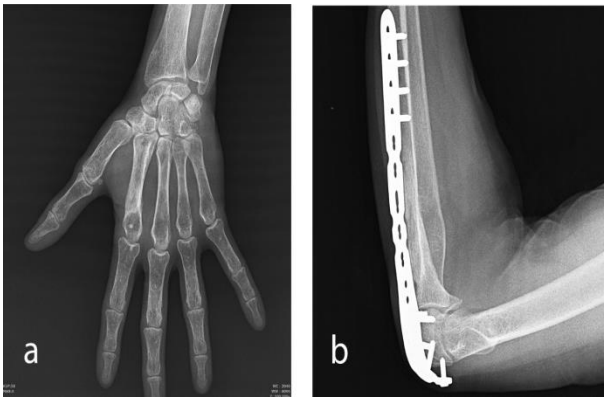

Figure 3. Postoperative images of the patient 12 months after the surgery

Discussion

We described a case of multiple simple bone cysts in an adult treated utilizing different modalities. The epidemiology of UBCs warrants attention. UBCs account for approximately 3% of all biopsied bone tumors (6). More than 91% of simple bone cysts usually occur in the second and third decades of life, though there are rare case reports in the literature diagnosed in patients aged up to 53 years (7). Our case was first presented in her fifth decade of life. Not being a true neoplasm, simple bone cysts are often serendipitously diagnosed on radiographs obtained for other reasons (8) or present with nonspecific and mostly local symptoms such as the case with our patient, unless a secondary pathologic fracture is rendered.

Universally, on a plain X-ray, UBCs happen as lytic, **z**bone with an attenuated but unpenetrated cortex. Computed tomography (CT)-scan and magnetic resonance imaging (MRI) could be useful in unusual areas with complex bony anatomy, such as the pelvis and spine (9).

The exact etiology of UBCs remains widely imprecise. Historically, they were thought to be a pathologic response to bone trauma (4), but more recent theories include an increased intramedullary pressure by virtue of venous obstruction and resultant interstitial fluid accumulation (10). There might also be a contributing genetic factor, such as FUS-NFATC2 or EWSR1-NFATC2 fusions, especially in the younger population (11, 12).

Diversified strategies are currently available for managing UBCs, such as intralesional injections, decompression, curettage and bone grafting, and a combination of such techniques (13). Each treatment method is to be selected on a patient-based approach, and none is considered unquestionably superior to the others. Treatment aims to prevent or manage pathologic fracture, promote cyst healing, and prevent cyst recurrence and re-fracture (2). Since the first report of Scaglietti et al. on 90% of "favorable" results with intralesional steroid injections (14), this technique became one of the most prevailing methods of treatment with varying degrees of relapse (13).

We did not use steroid injections in the management of our patient. Decompression and curettage is another time-honored management technique for UBCs. In the present case, the tibial cyst curettage was performed by creating a cortical window after multiple drill holes in the cyst wall sufficient to view the entire cavity. After aspiration of the fluid, the fibrous membrane lining the cyst wall was curetted. Although many authors reported a relatively high risk of failure and recurrence with this method (15), this was not the case with our patient who showed no signs of cyst activity on follow-up visits. Of note is that the relatively large radial neck lesion was completely healed only after decompression by drill holes with no curettage.

Combined techniques include the utilization of both mechanical techniques and biological agents to improve healing rates. Jamshidi et al. used a locking plate and fibular strut-graft augmentation in the reconstruction of a UBC of the proximal femur in the pediatric population (16). We used the same combined technique for the large proximal ulnar cyst in our patient to prevent a secondary pathologic fracture, which is a common complication seen with these lesions (1, 17).

Conclusion

Although primarily believed to be a disease of the immature skeleton, the solitary bone cyst should be kept in mind as a differential diagnosis in much older patients that present with musculoskeletal symptoms. Considering the lesions' characteristics such as size and anatomical location is the key in selecting the right treatment modality out of the many available options to minimize the risk of complications and recurrence. We also could not recognize an underlying systemic cause for our patient's multiple cystic lesions, which might be a subject of interest for further studies.

Conflict of Interest

The authors declare no conflict of interest in this study.

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