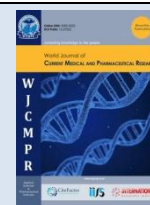




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TREATMENT OF PATHOLOGICAL FRACTURE FOLLOWING UNICAMERAL BONE CYST IN UNUSUAL LOCATION OF PROXIMAL RADIUS: RARE CASE REPORT

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Article History	Abstract
Received on: 07-04-2023 Revised on: 19-05-2023 Accepted on: 09-06-2023	Unicameral bone cysts (UBCs) are benign bone lesions predominantly affecting children and teenagers. This case report highlights the rare occurrence of a simple bone cyst in the proximal radius in a 29 year-old woman with a pathological fracture. Although the proximal radius is an uncommon site for UBC occurrence, the surgical approach required special care due to the complex anatomical structures near the fracture site. The surgical management of UBCs aims to prevent or manage pathological fractures, facilitate cyst healing, prevent recurrence, and reduce the risk of refracture. Understanding the treatment options and involving patients in the decision-making process are crucial for achieving the best possible results in managing UBCs with pathological fractures. The decision to pursue non-operative or operative treatment should be made in consultation with healthcare professionals, considering factors such as clinical presentation, radiological features, and symptoms.
	Keywords: Unicameral Bone Cyst, Simple Bone Cyst, UBC, Proximal Radius, Bone Mass.
	

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Introduction

Unicameral bone cysts (UBC) are also known as solitary or simple bone cysts and are the only true cysts of the bone [1]. UBC is a non-cancerous growth that predominantly affects children and teenagers, accounting for approximately 3% of primary tumors in the first two decades of life [2-4]. It is more common in males than females with a ratio of 3:1 [5]. UBC most usually develops in the metaphyseal-diaphyseal region of the growth plate of tubular bones, particularly in the humerus and femur [3, 6]. UBC is frequently asymptomatic and may spontaneously regress; however, symptoms may include pain, swelling, or stiffness in the nearest joint, and pathologic fracture is often the initial sign of the lesion [3, 6, 7]. UBC are often characterized as active (adjacent to the epiphyseal growth plate and separated by 0.5 cm) or latent (moving away

and separating from the growth plate by a normal region of cancellous bone) [7-9].

The exact pathogenesis of UBC remains unclear, but several hypotheses have been suggested, including disturbance in local bone growth, pre-existing lesions, intramedullary hemorrhages, and small nests of synovial cells trapped in an intraosseous position [3, 10]. The most popular pathogenic idea is Cohen's proposed occlusion of intramedullary venous drainage, which occurs in a rapidly growing and changing region of cancellous bone [11, 12].

Treatment for UBC has evolved over time and can include injection, decompression, and combined surgical techniques, depending on the purpose of restoring bone strength, cortical thickness, and cyst obliteration. Healing of UBC is classified by Neer and Capanna based on these criteria [13,14]. Non-operative management is recommended for asymptomatic patients who are not at risk of pathological fracture, while non-operative treatment with immobilization is appropriate for patients who have developed a pathological fracture in the upper extremity due to UBC [15]. UBCs in the proximal femur can result in pathologic fractures with serious complications, including osteonecrosis, varus malunion, and growth arrest of the proximal femoral physis [16].

Case Presentation



Figure 1 Lateral view radiograph of the forearm. There was a well-defined, geographic radiolucent lesion with narrow transition zone on the proximal part of radius bone.

A 29 years-old woman came to our emergency ward, with chief complaint of intractable pain, after falling on standing height with outstretched left hand. The pain was felt on her left forearm near the elbow joint. From physical examination there was a deformity and swelling on the proximal part of the forearm, and local tenderness on the proximal part of the forearm, especially on the ulnar side. An X-ray was later performed and we found a pathological fracture on the proximal part of radius, it was suspected due to a benign bone lesion. A surgery was later planned, the goal was to remove the mass as clean as possible, then to reduce and fixate the fracture site ensuring the stability and strength. We used postero-lateral approach to gain access to the lateral side of the forearm, then we isolated posterior interosseus nerve. An access to the pathological fracture was established and curettage of the content was performed, the sample was later sent to the histopathological department. The space left by the curettage was later filled with an autologous cancellous bone graft taken from the patient's proximal tibia and to support the strength of the bone we used a plate and screws. The finding from the histopathology department confirmed the mass was simple bone cyst.



(A)



(B)

Figure 2 AP view (A) and lateral view (B) radiograph of the forearm. Postoperative radiographs were conventionally taken after open reduction and internal fixation (ORIF) on the proximal part of radius bone. Operative treatment with plate fixation of pathological fracture aims to immediate rigid stabilization, facilitate early mobilization, and also as a pain relief.

Discussion

Unicameral bone cyst is a benign lesion of the bone and first identified by Virchow in 1876. It's mostly diagnosed in childhood. UBC usually found in adolescent to the second decade of life. The lesion itself usually diagnosed later than the common predilection due to the relatively asymptomatic nature of the lesion, usually as an incidental finding or when a pathological fracture occur. This type of tumor accounts for

approximately 3% of total bone tumors with unknown etiology [17,18,19].

The most common site of UBC are proximal humerus and proximal femur, it accounts up to 90% of the cases reported, which in our case the location on proximal radius is rather an uncommon site for the lesion to occur. This site also rather challenging to manage, due to its close proximity to major vessels and nerve near the elbow joint. The approach we

used was a postero-lateral approach where we can access the fracture site while isolating the posterior interosseus nerve. Then, we proceed to do the curettage of the lesion, and then filling it with an autologous cancellous bone graft taken from the patient's proximal tibia. And to ensure the stability of the fracture we later implanted a plate and screw fixation. While some data said the healing rate was as low as 25% to 36%, we took consideration of the patient's age which is older than the average, which usually leads to better healing outcome up to 90% healing rate.

Most often, the diagnosis of UBC is made through radiologic and pathologic examinations. The indication of surgical intervention in the patient of this case report to treat the pathological fracture. Plain radiographies are commonly used for radiologic diagnosis. Radiologically, a cyst appears as a translucent, well-defined, lytic lesion located along the central axis of the metaphysis. Sometimes, a fragmented bone cortex can be observed within the cyst cavity, known as a "fallen fragment." This particular sign is not specific to cysts and is present in approximately 20% of cases. Computed tomography (CT) is not useful for diagnosis, in cases where the diagnosis is uncertain. Advanced imaging techniques can provide valuable information to distinguish a unicameral bone cyst (UBC) from similar lesions. Computed tomography (CT) scans can reveal thinning of the cortical bone, fluid density within the lesion, and occasionally the fallen fragment sign. Magnetic resonance imaging (MRI) is another diagnostic method used, but it is not routinely employed. It is typically reserved for cases requiring additional attention in surgical planning, such as when the cyst is in close proximity to metaphyseal growth plates. Magnetic resonance imaging (MRI) demonstrates specific characteristics of UBC, such as a hypointense signal on T1-weighted images and a hyperintense signal on T2-weighted and fluid-sensitive sequences. MRI can also visualize any septations that may be present, and peripheral enhancement can be observed on post-contrast images. In the presence of a fracture, T1-weighted images may show a hypersignal due to blood, while T2-weighted and fluid-sensitive sequences can display marrow edema, periosteal reaction, and surrounding edema as hyperintense signals. Fluid levels may be present, particularly in cases involving a pathological fracture. MRI is particularly valuable for evaluating suspected insufficiency fractures and determining if the cyst affects the growth plate or causes disruption. Simple bone cysts are benign lesions that do not require routine biopsy and treatment in asymptomatic cases. Although there are many different procedures for treatment, such as subtotal resection, curettage, repair with bone graft, and intralesional steroid or saline injection, none are widely accepted treatments [20,21,22].

The primary aim of surgical intervention in UBC cases is to prevent or manage pathological fractures, facilitate cyst healing, prevent cyst recurrence, and reduce the risk of refracture. Various treatment modalities are currently suggested for UBC management, including observation, steroid injection, autologous bone marrow injection, curettage and bone grafting, demineralized bone matrix application, and autogenous bone marrow injection. Specific treatment decisions should be made by medical professionals based on individual patient characteristics and circumstances. The

treatment approach for UBCs depends on several factors, including the clinical presentation, radiological features, and the presence of symptoms [23,24].

In some cases, observation may be recommended when UBCs are incidentally discovered in asymptomatic patients and the affected bone remains structurally stable without significant weakening. This means that close monitoring is conducted to ensure that the cyst doesn't cause any adverse effects or progress further [23,24].

For cases where intervention is necessary, various treatment options are available. Steroid injections, such as corticosteroids, are sometimes used to treat UBCs. The injection of steroids into the cyst may help promote healing and reduce inflammation. Autologous bone marrow injection involves aspirating bone marrow from the patient's own body and injecting it into the cyst. This procedure aims to introduce cells that can promote bone regeneration and healing. Autologous bone marrow injection involves aspirating bone marrow from the patient's own body and injecting it into the cyst. This procedure aims to introduce cells that can promote bone regeneration and healing. Curettage and bone grafting is a surgical technique commonly used for UBC treatment. It involves scraping the cyst's lining (curettage) and filling the cavity with a bone graft, which can help stabilize the bone and promote healing. Demineralized bone matrix (DBM) is another treatment option. DBM is derived from cadaveric bone and contains various proteins and growth factors that promote bone healing. It can be used alone or in combination with other procedures, such as curettage. Open surgery with radical cyst resection and bone grafting is considered a definitive treatment for UBCs. This procedure involves surgically removing the entire cyst and any surrounding abnormal tissue. The resulting void is then filled with a bone graft to facilitate healing and restore bone strength. It's important to note that the decision to pursue non-operative treatment with immobilization should be made in consultation with a healthcare professional who can assess the specific circumstances of the fracture and determine the most appropriate course of action. Regular follow-up is essential in both cases of non-operative and operative intervention to monitor the progression of the UBC and evaluate the effectiveness of the chosen treatment approach. If there are any changes or worsening of symptoms, further intervention may be required [23-27].

In this case report, we did the curettage of the lesion, and then filling it with an autologous cancellous bone graft. The use of bone grafting after curettage and decompression is beneficial in providing structural support and promoting healing through osteoconductive and osteoinductive properties. Although bone grafts and substitutes can easily incorporate into the unicameral bone cyst (UBC), there is still a possibility of recurrence or persistence. Different types of grafts, including autograft, allograft (cancellous and cortical strut), and synthetic bone substitutes, have been documented in the literature with similar outcomes. No particular graft type has a clear advantage, but bone substitutes are attractive due to their unlimited supply, absence of donor site morbidity, and no risk of disease transmission. Additionally, certain bone substitute formulations can offer immediate structural support

to the UBC. Numerous studies on curettage and grafting of UBCs can be found in the literature, although most of them are case series or have low-level evidence. Despite the varying degrees of success, virtually all graft types have shown some level of effectiveness in promoting healing and reducing recurrence rates compared to decompression and curettage alone [28].

Unicameral bone cyst (UBC), are indeed benign masses that can cause destruction of bone tissue. Simple bone cysts in the proximal part of the radius in this case report are considered rare, challenging and adds complexity to the surgical approach. The individualization of treatment and surgical approach is crucial to ensure the best possible outcome and to minimize the risks associated with the procedure, especially given the complex anatomy and structures surrounding the proximal part of the radius near the elbow joint. It is essential to involve the patient and their family in the decision-making process and provide them with detailed information about the benefits, risks, and alternatives to surgical intervention.

Conclusion

A pathological fracture following a SBC, usually found incidentally during radiological test, in our patient after a trivial injury. The site of SBC in our patient is rather uncommon and need a special care due to the complex anatomical structures near the fracture. The prognosis of the patient usually is better with increasing age since the lesion is latent. The goals of the treatment are to make sure that the cavity is clean, to promote bone healing, and a stable fixation.

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Conflict of interest

All authors declare no conflict of interest.

Ethical Statement

The patient provided written informed consent for this case to be published.

Informed consent

Written informed consent was obtained from the patient.

Authors' contributions

FL, THB, and AEW designed the study. FL, THB retrieved the data. FL, THB, and AEW analyzed the data descriptively. FL, THB, and AEW wrote the manuscript. All authors approved the final version of the manuscript.

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