Original Article

Management of Unicameral Bone Cyst of Proximal Femur: Experience of 14 Cases and Review of Literature

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ABSTRACT

Objective: To assess the results of surgical treatment of unicameral bone cyst (UBC) involving the proximal femur

Design: Retrospective study of 14 cases of UBC of proximal femur

Setting: Al-Razi Orthopedic Hospital, Kuwait

Subjects and Methods: Fourteen cases of UBC seen and treated at Al-Razi hospital were included in the study. Their presentation and the method of treatment were recorded.

Intervention: Thirteen cases were treated surgically using intra-lesional excision (ILE). The cavity was filled with autogenous bone graft in three cases, hydroxyapatite matrix (HA) in eight cases, and combined autogenous graft and hydroxyapatite matrix in two cases. Internal fixation was carried out in six cases. External fixator was applied in one case from iliac bone to femur crossing the hip joint.

Main Outcome Measures: Patients were followed up post-operatively for an average period of 42 months (range = 9–120 months). They were observed for recurrence, complications and fracture healing.

Results: Recurrence was observed in one case while other cases showed healing of the cyst with consolidation and varying degrees of remodeling in one year's time. A case developed mal-union and growth arrest with subsequent shortening. Avascular necrosis and coxa vara was detected in another case. All the fractures healed in the usual expected time according to age.

Conclusion: UBC of the proximal femur exhibits unique characters and complications. Hydroxyapatite matrix is a useful and effective bone substitute. Post-excision stabilization of the cyst is recommended to avoid mal-union and to facilitate post-operative rehabilitation and earlier return to normal activities.

KEYWORDS: pathologic fracture, proximal femur, UBC

INTRODUCTION

Unicameral bone cyst (UBC) is defined as an atrophic degenerative osteolytic process consisting of a cavity filled with fluid and lined by a membrane[1]. The membrane is composed of cells staining positively with CD68, SDF-1, STRO-1, RANKL and express RUNX2. UBC cells show 24.2% of apoptosis significantly higher than 17.2% of trabecular bone cells[2]. Biochemical analysis of the cyst fluid showed bone-resorptive factors, i.e., prostaglandins, interleukin 1 and proteolytic enzymes[3].

UBC has highest incidence between 5 and 15 years[1] and 50% of upper femoral lesion are over 17 and their age as high as 54 years[4]. It usually arises in the metaphysis of long bones immediately beneath the growth plate and the most common location is proximal humerus followed by proximal femur[1,4-7] which accounts for 27% of cases[5]. UBC was the underlying lesion in 40% of pathological femoral neck fractures in children[8]. Avascular necrosis (AVN) of proximal femoral epiphysis and collapse of the articular surface was reported as a complication of UBC involving proximal femur[9-11].

Among the wide range of different modalities described for treatment of UBC are: radical excision in form of subperiosteal partial diaphysectomy and allograft[12], subtotal resection with[13] and without bone graft[14], curettage and bone graft[6], multiple drill holes[15,16], intra-cystic prednisolone injection[5,7,9] and recently, intra-medullary flexible nails[17,18]. Some authors focused their interest on UBC and other benign lesions located at proximal femur as this area exhibits unique characters and complications[8,19-23]. The aim of this study is to describe our experience with the results of surgical treatment of UBC involving proximal femur in Kuwait.

SUBJECTS AND METHODS

The current study represents a retrospective
<table>
<thead>
<tr>
<th>Case No.</th>
<th>Age at presentation</th>
<th>Sex</th>
<th>Side</th>
<th>Presentation</th>
<th>Work up</th>
<th>Location of the lesion</th>
<th>Location of the fracture</th>
<th>Displacement</th>
<th>Treatment</th>
<th>complications</th>
<th>Follow up months</th>
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<tbody>
<tr>
<td>1</td>
<td>14</td>
<td>F</td>
<td>Lt</td>
<td>Pathological fracture</td>
<td>C.T</td>
<td>Inter –subtrochanteric</td>
<td>Basotrochanteric</td>
<td>Displaced</td>
<td>ILE + local adjuvant + HA + DHS + Screw + Spica</td>
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<tr>
<td>2</td>
<td>14</td>
<td>M</td>
<td>Lt</td>
<td>Pathological fracture</td>
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</tr>
<tr>
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<td>8</td>
<td>M</td>
<td>Lt</td>
<td>Pathological fracture</td>
<td>Neck</td>
<td>Transcervical</td>
<td>Undisplaced</td>
<td>ILE + HA +KW’S + spica</td>
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<td>4</td>
<td>24</td>
<td>M</td>
<td>Rt</td>
<td>Pain, limp, wasting of quadriceps</td>
<td>CT+ Bone scan</td>
<td>Head &amp; neck</td>
<td>-</td>
<td>ILE+ Auto grafts + Ext fixator</td>
<td>Nil</td>
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<td></td>
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<td>5</td>
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<td>M</td>
<td>Rt</td>
<td>Pain, limp, wasting of quadriceps</td>
<td>CT+ Bone scan</td>
<td>Intertrochanteric</td>
<td>-</td>
<td>ILE + local adjuvant + HA + Auto graft + DHS + Screw</td>
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<td>Lt</td>
<td>Pain</td>
<td>C.T</td>
<td>Inter –subtrochanteric</td>
<td>-</td>
<td>Auto graft + DHS</td>
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<td>7</td>
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<td>Lt</td>
<td>Pain</td>
<td>C.T</td>
<td>Head &amp; neck</td>
<td>-</td>
<td>ILE+ Auto grafts + Spica</td>
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<td>60</td>
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</tr>
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<td>8</td>
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<td>M</td>
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<td>Pain</td>
<td>-</td>
<td>Inter Trochanteric</td>
<td>-</td>
<td>ILE + HA + Auto grafts + spica</td>
<td>Recurrence</td>
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<tr>
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<td>Inter-trochanteric</td>
<td>Undisplaced</td>
<td>ILE + HA + spica</td>
<td>Nil</td>
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<td>Inter-trochanteric</td>
<td>Undisplaced</td>
<td>ILE + HA + spica</td>
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<tr>
<td>11</td>
<td>7</td>
<td>F</td>
<td>Rt</td>
<td>Pathological fracture</td>
<td>-</td>
<td>Neck + Inter –subtrochanteric</td>
<td>Inter-trochanteric</td>
<td>Undisplaced</td>
<td>ILE + local adjuvant + HA +Spica</td>
<td>Shortening+ mal union +growth arrest</td>
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<td>M</td>
<td>Rt</td>
<td>Pain &amp; limitation of hip movements</td>
<td>-</td>
<td>Inter –subtrochanteric</td>
<td>-</td>
<td>ILE + local adjuvant + HA +Spica</td>
<td>Nil</td>
<td>33</td>
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<tr>
<td>13</td>
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<td>M</td>
<td>Rt</td>
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<td>C.T</td>
<td>Inter –subtrochanteric</td>
<td>Inter-trochanteric</td>
<td>Undisplaced</td>
<td>ILE + local adjuvant + HA + DHS</td>
<td>Nil</td>
<td>30</td>
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<tr>
<td>14</td>
<td>7</td>
<td>M</td>
<td>Rt</td>
<td>Pathological fracture</td>
<td>-</td>
<td>Neck + inter –subtrochanteric</td>
<td>Neck</td>
<td>Displaced</td>
<td>Conservative</td>
<td>AVN, Coxa vara,shortening</td>
<td>85</td>
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Rt = Right, Lt = Left, ILE = Intra-lesional excision, HA = Hydroxyapatite, DHS = Dynamic hip screw, AVN = avascular necrosis, KW = Kirschner wire
analysis of 14 cases of UBC involving proximal femur treated at Al-Razi Orthopedic Hospital, Kuwait during the period from 1990 through 2003 (Table 1). Analysis was a thorough review of medical records, including medical history, clinical examination, work-up, operative details and follow-up at the clinic.

Age at presentation ranged from 5 - 30 years with an average of 13 years. Ten cases were male and four were female. All cases were symptomatic. Pathological fractures were the presenting symptoms in five cases. The other cases presented with pain, various degrees of limp, limited hip movement and quadriceps wasting. The right side was involved in eight cases. Radiographic examination was the basic imaging technique for all the cases, whereas CT was done for selected cases (case # 1, 4, 5, 6, 7 and 13) to assess bony destruction and bone scan for cases with doubtful diagnosis (case # 4 and 5). In most cases the lesion was located in the intertrochanteric area with varying degrees of extension toward the neck and subtrochanteric region. The head and neck were involved in two cases (case # 4 and 7). The neck was affected in one case (case # 3). In one case (case # 14) diagnosis was based on typical X-ray appearance and clinical course. In the other cases histopathological study of the curetted material confirmed the diagnosis.

Surgical treatment was employed in all cases with exception of one case (case # 14) which was referred from another hospital. The basic surgical technique in all cases was intra-lesional excision (ILE). Local adjuvant therapy was used in six cases (case # 1, 2, 5, 11, 12 and 13) which consisted of painting with phenol 40%, cleaning with alcohol 90% and irrigation with normal saline. The cavity was filled with autogenous bone graft in three cases (case # 4, 6, and 7), hydroxyapatite matrix (HA) in eight cases (case # 1, 2, 3, 9, 10, 11, 12, and 13) and combined autogenous graft and hydroxyapatite matrix in two cases (case # 5 and 8). Internal fixation was carried in the form of dynamic hip screw (DHS) in five cases (case # 1, 2, 5, 6 and 13) and KWs in one case (case # 3) which was removed in six weeks time. External fixator from the iliac bone to femur crossing the hip joint was applied in one case (case # 4).

RESULTS

Follow-up ranged from 9 -120 months with an average of 42 months. With exception of one case which showed recurrence 9 months after surgery (case # 8), the minimum follow-up was 24 months. All other cases showed healing of the cyst with consolidation and varying degrees of remodeling in one years time. The hardware was removed in three years time in three cases (case # 1, 6 and 13). Case # 11 developed mal-union and growth arrest of greater tuberosity growth plate with subsequent shortening. This required raising the heel and did not need any surgical procedure at the last clinical assessment. AVN and coxa vara were detected in another case (case # 14). She was 14 years old at last follow-up. Clinical evaluation showed fair range of movement with occasional hip pain. Radiographic examination reported incongruent congruity of the involved hip joint. She needs long follow-up to detect the onset of disabling degenerative changes which dictate reconstructive procedures. All the fractures healed in the usual expected time according to age.

DISCUSSION

The basic surgical technique employed in the current study was ILE in the form of curettage and cleaning of the wall by power burr. In order to improve the margin and to decrease recurrence rate, local adjuvant therapy was applied earlier through this study in six cases (case # 1, 2, 5, 11, 12, and 13). 40% phenol was applied with a cotton tipped applicator and was removed by lavage with alcohol. No recurrence was reported in those cases. Phenol was first applied to UBC by Neer et al in 1966. However, because of lack of recent evidence supporting the use of phenol in UBC, we did not use it in later cases.

We used autogenic bone graft alone to fill resultant cavity in three cases (case # 4, 6, 7). Neer et al reported surgical treatment of 129 cases of UBC by curettage (ILE) and bone graft. They evaluated 24 out of 31 cases located in the proximal femur and reported recurrence in four cases (17%). The results of allograft were compared to autograft used to fill the defect after curettage of 93 cysts located in proximal femur and humerus. In 35 cases treated with autograft, 21 cases (60%) were excellent, six cases (17%) showed residual defect and eight cases (23%) required re-operation. In 58 cases treated with allograft; 28 cases (48%) were excellent, 12 cases (48%) showed residual defect, and 18 (31%) cases required re-operation. Thus autograft was slightly better and therefore indicated in recurrence, when there is a sufficient quantity of bone which can be conveniently taken to fill the defect. Allograft is indicated for the pediatric age group with large cyst. Campanacci et al did not find any relation between type of bone graft and rate of recurrence and pointed out the importance of packing of the cyst well as residual empty spaces might be a source for recurrence.

HA was reported to have been used to fill the cavity of benign bone tumor and lesions after surgical excision. Out of 22 cases of UBC treated
by curettage followed by packing of the cavity with high-porosity HA, complete healing without cyst recurrence occurred in 18 (78%) cases. New bone surrounding HA was radiologically detected within an average of 2.3 months[26]. In the current study HA matrix was used alone to fill the curetted cavity in eight cases and combined with autogenic bone in two cases. All ten cases healed and showed varying degrees of remodeling without recurrence (Fig. 1a, b, and c).

Recurrence was reported in one case (7%) in the current study (case # 8). A 5-year-old boy presented with UBC involving the inter-trochanteric region of left femur. He was treated by ILE and packing of the cyst cavity with a mixture of HA matrix and autogenous bone graft. Early signs of recurrence were detected by the ninth month. It was reported that recurrence following surgical treatment was more frequent in patients under the age of 10 years, and age was a more reliable prognostic factor than the proximity of the cyst to growth plate[4,6].

In 1974, Scaglietti et al described minimally invasive empiric injection of UBC with methylprednisolone. He reported 72 cases out of which 11 were located in the proximal femur with a healing rate of 96%[7]. Capanna et al reviewed 90 cases of UBC treated by intra-cystic injection of methylprednisolone out of which 20 were located at proximal femur. He reported 80% satisfactory results and there was a need for two to six injections per patient in order to achieve healing. Recurrence after cyst consolidation was observed in 12 patients (13%). Pathological fracture developed in seven patients during the course of treatment. One of them was a displaced fracture at proximal femur four months after the onset of treatment which required open reduction and internal fixation. A limb-length discrepancy was observed in two cases without prior surgery or pathological fracture. One of them was at the proximal femur[9].

Campanacci et al compared 178 cases of UBC treated by curettage and bone graft to 141 cases treated by methylprednisolone injection. The recurrence rate was 33 and 15% respectively. Pathological fracture developed in two cases during injection treatment and in another 11 cases after recurrence of the cyst. AVN of the femoral head was observed in one case treated by injection therapy[5]. It was found that healing response to intra-lesional corticosteroid injection is unpredictable and usually incomplete even after multiple injections. The failure rate in the weight-bearing bone is high[27].

Percutaneous injection of autogenous bone marrow was described for treatment of UBC. The results of single injection of bone marrow into UBC were reported in eight cases, four out of them in the proximal femur. Healing according to Capanna criteria[9] was complete in one case, incomplete in six cases, no response in one case and there was no recurrence[28]. In a series of 79 consecutive patients with UBC, the results of aspiration and bone marrow injection were compared with those of aspiration and injection of steroids. The author reported that no advantage could be shown for the use of marrow injection over steroid injection in treatment of UBC[29].

Autogenous bone marrow injection was combined with allogenic demineralized bone matrix for treatment of UBC in 23 patients five of them were in proximal femur. The average time for
pain relief was five weeks, and the average time until patients returned to full unrestricted activity was six weeks. A second injection was required because of recurrence in five patients (22%) whereas pathological fracture occurred in one case\textsuperscript{[30]}. Similar results were reported after injection of demineralized bone graft without bone marrow, suggesting that the use of bone marrow may not be necessary to achieve good results\textsuperscript{[31]}. Based on venous obstruction as a theory for development of UBC, Chigira \textit{et al} introduced the treatment by multiple drill holes. He reported on seven cases of UBC. Two were located in the femur. Healing was observed within six to eight months. One of the cysts located in the femur required curettage and bone graft because of poor healing\textsuperscript{[15]}. The same technique was applied to 23 cases of UBC. Nine cysts were located in the femur. Recurrence was observed in 15 cases\textsuperscript{[16]}. Roposch \textit{et al} evaluated the results of flexible intra-medullary nail in the treatment of UBC in 32 cases. The cyst was located in the proximal femur in nine patients. Recurrence was observed after removal of the nail in two cases, one of which was located in proximal femur. Change of the nail was required in nine cases, three located in the femur. A varus deformity of proximal femur developed in five cases after consolidation of the cyst\textsuperscript{[17]}. In series of 12 cases of UBC of proximal femur treated by flexible intra-medullary nail, complications were reported in three cases. Perforation of the nail through the lateral cortex led to coxa vara in one case. The nail was removed and the femoral neck was fixed with a plate. The nail was changed as it became too short for the growing child in two cases\textsuperscript{[18]}. In the current series there was a seven year old girl (case # 14) who presented with pathological neck fracture through UBC involving the femoral neck and inter-trochanteric area. She was treated conservatively in form of skin traction followed by hip spica. The fracture healed and the cyst consolidated, but she developed coxa vara and AVN of the femoral head (Fig. 2a, b, c). In a series of 20 children with pathological fracture of proximal femur treated conservatively, the fracture was displaced in eight cases. All of the displaced fractures healed but with coxa vara and AVN in one child, coxa vara in a second and coxa breva in a third child. All of 12 undisplaced fractures healed without deformity or avascular necrosis, but they required from 1 - 7 injections of prednisolone for healing of the cyst. Re-fracture was observed in six children 2 - 5 years after presentation\textsuperscript{[21]}. After pathological fracture through UBC the early radiological appearance suggests progressive healing of the cyst, but complete healing of the cyst seldom takes place without operative treatment\textsuperscript{[22]}. Hence the proximal femoral cyst needs to be surgically treated to avoid mal-union and persistence of the cyst\textsuperscript{[19,33]}. Wai \textit{et al} reported 11 cases of pathological fractures of proximal femur secondary to benign lesion. All cases were treated with curettage, high speed burring and reconstruction using a mixture of allo- and autogenic graft, and a fixed angle implant. All fractures healed without local recurrence\textsuperscript{[23]}. Both location of the cyst and the amount of bone loss dictate whether fixation can stabilize the fracture and type of fixation best suited for this purpose. Based on remaining bone in the femoral neck beneath the growth plate, bone in the lateral proximal femur

\begin{figure}[h]
\centering
\includegraphics[width=\textwidth]{image1.png}
\caption{Case # 14 shows pathological fracture treated conservatively}
\end{figure}

\begin{figure}[h]
\centering
\includegraphics[width=\textwidth]{image2.png}
\caption{Healing with coxa vara and AVN (AP view)}
\end{figure}

\begin{figure}[h]
\centering
\includegraphics[width=\textwidth]{image3.png}
\caption{Healing with coxa vara and AVN (lateral view)}
\end{figure}
(lateral buttress) and skeletal maturity, Dormans et al classified pathological fracture of femoral neck in children into three types with six subtypes and described the best fixation for each type[19].

Case # 1 presented with displaced pathologic baso-trochanteric fracture. It was type III-B according to classification proposed by Dormans et al[19]. Treatment was in form of ILE, local adjuvant therapy, and filling the cavity with HA. The fracture was stabilized by DHS and anti-rotation screw. The hardware was removed two years after surgery. At 47 month follow-up there was excellent function, healing of the fracture without deformity and complete consolidation of the cyst (Fig. 3a, b, c).

DHS was also applied to stabilize the fracture after surgical treatment in two cases classified as sub-type III-B (case # 2 and 13). Case # 3 presented with undisplaced pathological fracture neck and was classified as II-A[17]. The fracture was fixed after surgical treatment with multiple KWs supplemented with hip spica (Fig. 4a, b).

Case # 11 was a seven year old girl who presented with pathological fracture through UBC involving the neck, sub- and inter-trochanteric areas. After surgical treatment it was difficult to stabilize the fracture because there was not enough bone between the cyst and capital epiphysis and deficient lateral buttress (Fig. 5a, b, c). This case was classified as II-B[19]. The cyst consolidated and the fracture healed but with mal-union, shortening and growth arrest of the greater trochanter.

Few studies were published to assess the fracture risk in cases of UBC. Kaelin et al devised a cyst index to assess the risk for fracture and found that in femoral fracture the average index was 4.4 (SD = 0.75), the lowest index was 3.6 and there was no fracture in cysts with index lower than 3.5[24]. Shih et al defined the criteria for impending fracture secondary to UBC of proximal femur as 2.5 cm or larger lytic area in the upper femur, a lesion involving half of the femoral neck or more and expansile lesion with thin cortex and progressive deformity[22].

Jaffe et al reported management of seven patients with benign lesion of the femoral head and neck. The diagnosis was fibrous dysplasia in four cases, aneurysmal bone cyst in two cases, and UBC in one case. Curettage and autogenous fibular strut graft in conjunction with a sliding hip screw were carried out in six cases. One case was treated by curettage and strut graft. The results were excellent
in five cases, good in one case and fair in one case. AVN of the femoral head developed in a case with pathological neck fracture through aneurysmal bone cyst[20]. Shih et al treated 35 cases of benign lesion of the femoral neck and inter-trochanteric area and 11 out of them were UBC. The lesions were curetted, filled with cortical strut allo-graft and autogenic iliac cancellous graft and fixed with sliding hip screw. They reported excellent results[22].

In the present study six cases presented with hip pain without fracture. We tried to apply Dorman’s classification[19] for selection of fixation after surgical treatment. Type III-B matched two cases (case # 5 and 6) where DHS was applied. The femoral head was completely destroyed by the cyst in two cases (case # 4 and 7, Fig. 6a, b, Fig. 7a). This type was not included in Dorman’s classification and was difficult to fix. Case # 4 was stabilized by external fixator extending from the iliac bone to proximal femur crossing the hip joint (Fig. 7 b). The treatment
was designed to achieve healing of the cyst and to prevent collapse of the articular surface. Our clinical assessment at 24 months follow-up reported minimal pain and good range of movement. Radiographic examination showed consolidation of the cyst with mild narrowing of joint space (Fig. 7c). At that time no active intervention was indicated. However, when disability develops reconstructive procedures for the hip will be considered accordingly.

CONCLUSION

UBC of the proximal femur needs special consideration as it exhibits unique characteristics and complications. Each case should be evaluated individually. HA matrix is useful and effective bone substitute particularly in the pediatric age group where there is limited supply of autogenic bone graft. Post-excision stabilization of the cyst is recommended to avoid mal-union and to facilitate post-operative rehabilitation and earlier return to normal activities. Choice of internal fixation depends on the location of the cyst, amount of bone loss and skeletal maturity. The surgeon, the patient and parents should be worried about the possible complications.

REFERENCES