Unicameral bone cysts managed with CHRONOS bone graft substitute: A case series

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ABSTRACT
A unicameral bone cyst (UBC) may require surgery if its location increases the risk for pathologic fracture, or if it is painful or increasing in size. This case series demonstrates that curettage and grafting with a bone graft substitute alone can result in healing of the UBC. Three children with UBC and pathological fractures were treated with curettage and grafting using bone graft substitute beta-tricalcium phosphate granules (CHRONOS). All three went on to full union of the fracture and good consolidation of the graft with small residual cysts (Neer/Cole grades II). Curettage and grafting using CHRONOS granules is an effective way of treatment of UBC. By using this method, we were able to avoid the necessity of obtaining bone graft in young patients. It shortened operative time and avoided donor site morbidity.

Keywords: Unicameral bone cyst, curettage, grafting, osteoconduction

INTRODUCTION
A unicameral bone cyst (UBC) is a benign membrane lined cavity located within a bone and is filled with fluid which accounts for 3% of all bone tumours. The proximal humerus is the most commonly affected site, followed by the proximal femur. UBC may remain asymptomatic unless fracture occurs.

There are various theories regarding aetiology and these include blockage of drainage of interstitial fluid in rapidly growing and remodeling areas of the cancellous bone, or obstruction within bone. 1, 2 Aberrations of chromosomes 4, 6, 8, 12, 16, and 21 with mutations associated with amino acid substitutions may also be associated with UBC. 3

Spontaneous healing of UBC may follow pathologic fractures. 4 However, surgery may be needed if the location of the cyst increases the risk for pathologic fracture, if it is painful, increasing in size, or has already fractured. The different surgical options include curettage and packing with freeze-dried, crushed, cortical-bone allograft, 5 subtotal resection and auto-grafting, 6 subtotal resection without grafting, 7 percutaneous aspiration and steroid instillation within UBCs, 8 or
percutaneous bone marrow injection.  

This case series reports on our experience with the management of UBC with curettage and grafting with a bone graft substitute.

**CASE SERIES**

Three children (2 Male and 1 Female) aged 15, 4 and 14 respectively, with UBC and pathological fractures were treated with curettage and grafting using bone graft substitute beta-tricalcium phosphate granules (CHRONOS). The affected sites were sub-trochanteric in two cases and one affected the mid shaft of the femur.

The surgical technique carried out involved making a small skin incision at the level of the cyst wall. The bone was reached after dissection and retraction of the soft tissues. The thin cortical wall was then punctured using a small curette under image intensifier guidance. Curettage of the lesion was carried out and material was sent for histopathological evaluation. The cyst cavity was then suctioned out. Grafting was then done using CHRONOS granules. The skin was re-approximated using interrupted sutures. Healing was confirmed using the Neer Cole 4-Grades rating scale (Table 1).  

All the three children presented after pathological fractures of femur through the bone cysts. All were managed initially with skin traction, but subsequently underwent cyst curettage and grafting using CHRONOS within two weeks of presentation.

One patient (Case 3) needed additional external fixator application after curettage and grafting. This was done given the location of cases.

<table>
<thead>
<tr>
<th>Grades</th>
<th>Radiological outcomes</th>
<th>Clinical outcomes</th>
<th>Results</th>
</tr>
</thead>
<tbody>
<tr>
<td>I</td>
<td>Complete obliteration of cyst</td>
<td>Cyst healed, no pain, no pathological fracture</td>
<td>Good</td>
</tr>
<tr>
<td>II</td>
<td>One or more cyst like areas present but good reestablishment of bone strength</td>
<td>Cyst healed, no pain, no pathological fracture</td>
<td>Good</td>
</tr>
<tr>
<td>III</td>
<td>Cyst visible but multi-locular and opaque</td>
<td>Cyst not healed, pathological fracture possible</td>
<td>Poor</td>
</tr>
<tr>
<td>IV</td>
<td>Cyst clearly visible and unchanged</td>
<td>Cyst not healed, pathological fracture possible</td>
<td>Poor</td>
</tr>
</tbody>
</table>

**Table 2: Summary of the three cases treated with CHRONOS graft.**

<table>
<thead>
<tr>
<th>Case 1</th>
<th>Case 2</th>
<th>Case 3</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (years)</td>
<td>15</td>
<td>4</td>
</tr>
<tr>
<td>Gender</td>
<td>Male</td>
<td>Male</td>
</tr>
<tr>
<td>Fracture</td>
<td>Left sub trochanteric</td>
<td>Right sub trochanteric</td>
</tr>
<tr>
<td>Initial management</td>
<td>Skin traction</td>
<td>Skin traction</td>
</tr>
<tr>
<td>Definitive treatment</td>
<td>Curettage &amp; graft</td>
<td>Curettage &amp; graft</td>
</tr>
<tr>
<td>Partial weight bearing after</td>
<td>4 Months</td>
<td>3 months</td>
</tr>
<tr>
<td>Full weight bearing after</td>
<td>5 months</td>
<td>4 months</td>
</tr>
<tr>
<td>2 years follow up</td>
<td>Neer/Cole grade II</td>
<td>Neer/Cole grade II</td>
</tr>
</tbody>
</table>
the fracture in the mid shaft of the femur. The external fixator was removed after 10 weeks without any complications.

Histopathological evaluation of the curettage specimens confirmed the cysts to be UBC in all the cases.

At two year follow-up, all three patients had full union of the fracture with good consolidation of the graft (Neer/Cole Grades II) and residual small cysts (Figures 1, 2 and 3). None of the patients developed any infections or other complications.

DISCUSSION

UBC is a benign, fluid-filled, radiolucent lesion that is typically found in either the proximal humerus or proximal femur but can appear in virtually any bone. It causes thinning of adjacent areas of bone resulting in fractures or pain from micro-fractures. When the cysts are adjacent to the growth plate, they are called active cysts. Cysts seen some distance from the growth plate are considered to be latent cysts. A UBC occurs most frequently in children aged 5-15 years, with an average age of approximately 9 years. UBC affects males approximately twice as often as females.

Aberrations of chromosomes 4, 6, 8, 12, 16, and 21 with mutations associated with amino acid substitutions of arginine for tryptophan or serine may be associated with UBC. Further studies are required to determine whether it is acquired or inherited and the dominance of this condition.
The main difficulty in treating UBC is the unknown aetiology of the condition. One of the theories is that production of cyst fluid causes bone expansion. Therefore decompressing the cyst and putting it into communication with the medullary canal improves blood flow through the cyst wall. This stimulates the periosteum to induce bone formation and stops bone destruction.  

We tried curettage and grafting with a bone graft substitute alone. In our series, curettage and grafting using CHRONOS granules was an effective way of treatment of UBC. All the patients had healed at two years. We were also able to avoid the necessity of obtaining bone graft therefore shortening operative time and avoiding donor site morbidity such as blood loss, pain and possibility of injury to the lateral cutaneous nerve of thigh. One study showed healing of the cyst in all the 31 patients in which CHRONOS was used. In yet another study, only one of 24 cysts in which CHRONOS was used did not heal well. No study reported any adverse effects. Our excellent results correlate with these authors.

In conclusion our study shows that curettage and grafting using CHRONOS pro-
vides good healing of the cyst and good results.

REFERENCES