THE AETIOLOGY AND TREATMENT OF SIMPLE BONE CYSTS

MASAKI CHIGIRA, SUSUMU MAEHARA, SATORU ARITA, EIICHI UDAGAWA

From Gunma University School of Medicine, Japan

The internal pressure of simple bone cysts was found to be slightly higher than the normal pressure of the bone marrow in the contralateral limb. The pressure within the cyst was measured during drilling with a Kirschner wire; it gradually decreased as the number of drill-holes increased. The PO₂ of the cyst fluid was markedly lower than that of either venous or arterial blood measured synchronously. It is suggested that venous obstruction in the bone is the likely cause of these cysts. Seven patients with simple bone cysts were treated by the multiple drill-hole method, and the clinical outcome was excellent. Multiple drilling may prove to be the treatment of choice for simple bone cysts in the younger patient, as it presents fewer hazards than other procedures.

Since Bloodgood’s classic article on cystic lesions in bone in 1910, an extensive literature has accumulated on this subject. The aetiology of the simple bone cyst is still open to speculation. Jaffe and Lichtenstein (1942) agreed with von Mikulicz that the lesion represents “some local disturbance in bone growth and development”, and they noted “simple bone cyst is not a tumor in the strict sense”. Broder (1968) reported two patients who had developed simple bone cysts with osteoblastic precursor lesions and suggested that vascular obstruction in a focal lesion might develop into a simple bone cyst. In 1960, Cohen showed that the chemical constituents of the fluid in these cysts were similar to those of serum and that injection of contrast medium into cysts could yield information pertinent to this hypothesis. No contrast medium could be shown leaving the cyst through its wall when a low injection pressure was used (Cohen 1970), and he therefore suggested that venous obstruction within the bone was the cause. But it is always difficult to prove that the venous drainage of a small area of tissue is deficient.

As direct evidence of venous obstruction in simple bone cysts is lacking, treatment is still debatable. Curettage and autogenous bone grafting have been used by many authors (Garceau and Gregory 1954; Maeda 1963), but the clinical results have not been satisfactory (Neer et al. 1973; Norman and Schiffman 1977). It seems necessary to identify the aetiology of simple bone cysts in order to establish treatment on a firm basis.

MATERIAL AND METHODS

Seven patients with simple bone cysts were treated at Gunma University Hospital by drilling multiple holes as described below. The internal pressure of the cyst was measured in four of the seven patients, and the partial pressures of oxygen and carbon dioxide were measured in three of the seven using an automatic gas analyser. The age, sex, location of the lesion and clinical response to treatment are shown in Table I.

Table I. Clinical details of the seven patients

<table>
<thead>
<tr>
<th>Case</th>
<th>Age (years)</th>
<th>Sex</th>
<th>Location of lesion</th>
<th>Time to consolidation (months)</th>
<th>Clinical response to treatment</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>9</td>
<td>F</td>
<td>Humerus</td>
<td>5</td>
<td>Good (partial recurrence)</td>
</tr>
<tr>
<td>2</td>
<td>14</td>
<td>M</td>
<td>Humerus</td>
<td>5</td>
<td>Excellent</td>
</tr>
<tr>
<td>3</td>
<td>8</td>
<td>M</td>
<td>Humerus</td>
<td>5</td>
<td>Fair (small recurrence)</td>
</tr>
<tr>
<td>4</td>
<td>11</td>
<td>M</td>
<td>Femur</td>
<td>5</td>
<td>Excellent</td>
</tr>
<tr>
<td>5</td>
<td>12</td>
<td>F</td>
<td>Humerus</td>
<td>5</td>
<td>Excellent</td>
</tr>
<tr>
<td>6</td>
<td>11</td>
<td>M</td>
<td>Tibia</td>
<td>4</td>
<td>Excellent</td>
</tr>
<tr>
<td>7</td>
<td>60</td>
<td>M</td>
<td>Femur</td>
<td>None</td>
<td>Poor</td>
</tr>
</tbody>
</table>

* Radiographic evidence of bone formation in the cyst.

Analysis of the cyst fluid. Before measuring the internal pressure, one millilitre of fluid was withdrawn from the cyst. Arterial and venous blood samples were also taken. The fluid and blood were analysed by an automatic gas analyser (ABL-2, Radiometer A/S, Copenhagen, Denmark).

Measurement of internal pressure. Under general anaesthesia in the supine position, all patients were spontaneously ventilated with 33 per cent oxygen. The lesion
was viewed under an image intensifier, and cannulated using a steel needle (18 gauge), the cannula being connected with an electromanometer (P-231D, Gould Inc., Oxnard, California). Readings were taken on a two-channel monitor oscilloscope (2G47, SAN-EI Instrument Co., Tokyo), or on a pen-recorder (Rectigraph-8K, SAN-EI Instrument Co., Tokyo) for a permanent record. At the same time, the left radial artery was cannulated by inserting a small polyethylene cannula (18 gauge) in a distal-to-proximal direction, this cannula being connected with another electromanometer, so that simultaneous readings could be taken of the intra-arterial pressure. Once the system was completely connected, filled with heparinised saline and calibrated by a mercurial manometer, a stop-cock was turned to open the cannulae to the recording system. Internal pressure of the cyst was measured twice at different points within the same lesion. To avoid contamination with cells from the cyst, the normal bone marrow pressure was measured with another circuit.

**Treatment by multiple drill-holes.** All cysts were drilled with Kirschner wires (2.0 millimetres in diameter) percutaneously; the diaphysial side of the cyst wall was perforated in several places. Two or three Kirschner

---

![Fig. 1](image1.png)  
**Fig. 1** — The internal pressure (upper tracing) of this simple bone cyst was 33.0/30.0 mmHg. Figure 2 — The pressure of the normal bone marrow in the contralateral limb was 26.0/24.0 mmHg. The lower tracings in both figures are intra-arterial pressure readings in the radial arteries.

![Fig. 2](image2.png)

![Fig. 3](image3.png)  
**Fig. 3** — Changes in internal pressure of the cyst as a result of multiple drilling through the cyst wall (indicated by arrows). In this patient the internal pressure of the cyst (24.0/22.0 mmHg) fell to 8.0/6.0 mmHg during drilling. The blood pressure did not change during treatment. (The lower tracing is a continuation of the upper tracing.)
wires were left in situ after the operation to allow drainage of the fluid through the cyst wall.

RESULTS

Internal pressure of the cyst. As shown in Figures 1 and 2, internal pressure was slightly higher than the pressure in normal bone marrow in the contralateral limb (Table II). The intra-cystic pressure fell during the drilling (Fig. 3). The blood pressure did not change during the treatment.

Table II. Internal pressure of the simple bone cyst

<table>
<thead>
<tr>
<th>Case</th>
<th>Systolic/diastolic pressure (millimetres of mercury)</th>
</tr>
</thead>
<tbody>
<tr>
<td>3</td>
<td>22.0/19.0</td>
</tr>
<tr>
<td>4</td>
<td>33.0/30.0 (32.0/30.0)*</td>
</tr>
<tr>
<td>5</td>
<td>22.0/20.0</td>
</tr>
<tr>
<td>6</td>
<td>33.0/30.0 (26.0/24.0)*</td>
</tr>
<tr>
<td>Mean</td>
<td>27.5/24.8 (29.3/27.3)</td>
</tr>
</tbody>
</table>

* Contralateral normal bone marrow pressure.

Analysis of the cyst fluid. Although ventilation of the patients was good and was steady during the treatment, the PO₂ of the cyst fluid was lower than that of venous blood in the same patient (Table III). However, the PCO₂ of the cyst fluid was similar to pressures in arterial and venous blood. Base excess, pH and HCO₃ of the cyst fluid were not significantly different from those of arterial and venous blood.

Table III. PO₂ and PCO₂ analysis (in millimetres of mercury) in three patients

<table>
<thead>
<tr>
<th>Case</th>
<th>Intracystic</th>
<th>Intravenous</th>
<th>Intra-arterial</th>
</tr>
</thead>
<tbody>
<tr>
<td>PO₂</td>
<td>3</td>
<td>45.9</td>
<td>198.2</td>
</tr>
<tr>
<td></td>
<td>5</td>
<td>53.9</td>
<td>160.1</td>
</tr>
<tr>
<td></td>
<td>6</td>
<td>32.9</td>
<td>176.7</td>
</tr>
<tr>
<td>Mean</td>
<td></td>
<td>44.2</td>
<td>178.3</td>
</tr>
<tr>
<td>PCO₂</td>
<td>3</td>
<td>41.8</td>
<td>34.3</td>
</tr>
<tr>
<td></td>
<td>5</td>
<td>41.5</td>
<td>40.8</td>
</tr>
<tr>
<td></td>
<td>6</td>
<td>42.3</td>
<td>46.9</td>
</tr>
<tr>
<td>Mean</td>
<td></td>
<td>41.9</td>
<td>40.7</td>
</tr>
</tbody>
</table>

Clinical response to multiple drilling. Radiographs taken two to three months after treatment generally showed reconstruction of the cortical bone of the cyst wall, while the cystic cavity had become radiopaque. Three to five months after treatment there was new bone formation uniformly distributed throughout the cyst. "Healing", with complete disappearance of the cystic cavity, was generally observed within six to eight months (Figs 4 and 5).

In Cases 1 and 3, a small cyst reappeared in the same area four to five months after the Kirschner wire had been extracted. In Case 7, no clinical response could be observed within eight months, and the bone cyst was treated with curettage and bone grafting.

DISCUSSION

Aetiology. In 1904, Mönckeberg suggested that cystic lesions in bone represented the healing form of a giant-cell tumour or of osteitis fibrosa. In 1910, Bloodgood described benign bone cysts as cystic lesions with a fluid content. Phemister and Gordon (1926) thought that bone cysts were secondary to a form of osteomyelitis. Johnson and Kindred (1958), reviewing 400 cases, described a so-called parietal nubbin of tissue in the cyst lining that lay just outside the cyst wall or projected into the cyst; they called this an angiomyxofibrilar lipoma because of its four mesenchymal elements and they stated that if this nubbin was not removed it would be responsible for recurrence. Jaffe and Lichtenstein (1942) thought that the lesion was dysplastic and due to mechanical trauma at the epiphysial line, leading to defective enchondral bone formation.

However, the aetiology of simple bone cyst was still open to speculation. In 1960, based on a study of the fluid content of six bone cysts, Cohen proposed that a focus of fibrous tissue was formed in an area of rapid resorption and that this could represent the initial lesion. He showed that the fluid contents of simple bone cysts were similar to those of a transudate, and suggested that blockage of vessels in the area led to cyst formation. In 1970, Cohen showed that no contrast medium could be seen leaving the cyst through its wall when a low injection pressure into the cyst was used, although hourly radiographs showed that the cyst was still filled with contrast medium 24 hours after injection. He stated that venous obstruction.
in bone was the cause. Neer et al. (1973) stated that blockage of sinusoidal vessels in the area led to accumulation of interstitial fluid and cyst formation. He found that the alkaline phosphatase in the cyst fluid was 10 to 20 times higher than in venous blood, and suggested that this might be a measure of the osteoblastic repair and that the ratio of alkaline to acid phosphatase might be a good index of “activity”.

It is, however, always difficult to prove directly that the venous drainage of a small area of tissue is deficient. Unless there is major obstruction to venous return the usual copious collateral drainage masks the disturbance, especially when small veins rather than the large ones are involved. The most attractive explanation of simple bone cysts is venous obstruction in bone, but this has not yet been conclusively established. From our results it is clear that there were vascular channels in the bone cyst and obstruction of venous drainage through the cyst wall. The clinical response to multiple drill-holes seems to confirm that venous obstruction is the primary cause.

The hydrodynamic character of atypical cysts (in adults and in the calcaneus) suggests that these may be different from the typical cysts in children. Abe et al. (1977) demonstrated that calcaneal cysts did not sustain pathological fractures and remained the same size for a long time. Several authors have stated that bone cysts in the adult respond well to curettage and bone grafting and do not recur (Maeda 1963; Neer et al. 1973; Norman and Schiffman 1977). In our series the clinical response of an atypical cyst to drilling was different from that of the typical cyst.

**Treatment.** The best approach to surgical management of simple bone cysts is still in doubt. Curettage and bone grafting have often been used, particularly for cysts in the humerus and femur, but a certain number have not healed (Neer et al. 1973). In children, the danger of damaging the epiphysis must be borne in mind (McKay and Nason 1977).

It is evident that the more radical the excision of the cyst and its contents, the lower the rate of recurrence. Fahey and O’Brien (1973) reported a success rate of 95 per cent in 20 patients treated by subtotal resection and bone grafting. However, obtaining a large volume of autogenous graft increases the potential complications. McKay and Nason (1977) reported that subtotal resection without grafting was highly effective for simple bone cysts; but that a large cyst needed grafting because of the risk of fracture.

The question, however, is whether the radical operation is really needed for the typical bone cysts in children. In 1954, Garceau and Gregory stated that healing could be expected in about 15 per cent of cases after pathological fracture of the cyst wall. However, several authors have reported that the cysts persisted and enlarged; more bone was absorbed and pathological fracture might follow (Clark 1962; Galasko 1974). Both Campanacci and Scaglietti thought that injection of a corticosteroid into the cyst cavity would cause resorption of transudates (Campanacci, De Sessa and Bellando Randone 1975; Scaglietti, Marchetti and Bartolozzi 1979). They demonstrated that the topical injection of methylprednisolone acetate was effective in bone cysts in children; however, no reasons were given for the effectiveness of their treatment. In 1981, Kuboyama et al. reported that repeated percutaneous drilling was highly effective, but that partial recurrence of the cyst frequently occurred. They were unable to explain why the cyst sometimes filled in after treatment. They agreed with Cohen’s hypothesis that venous obstruction was the primary cause and inferred that cyst fluid escaped through the drill-holes.

Our results clearly demonstrate that obstruction of venous drainage is the primary cause of simple bone cysts. The internal pressure of the cyst decreased during multiple drilling and we believe that this decrease is the essential feature. In Cases 1 and 3, recurrence was observed after the removal of the Kirschner wires, although radiographs showed that the cysts had been filled by bony tissue. It seems that the Kirschner wire plays an important role in preventing recurrence.

There are two possible explanations for the effectiveness of the multiple drill-hole method. First, that the Kirschner wire, being a foreign body, may provoke an immunological response which somehow helps to prevent recurrence; this, however, seems unlikely. Secondly, that the Kirschner wires keep the holes in the cyst wall open, and permit fluid to escape through the narrow spaces around the wires.

Whether it is necessary to leave the Kirschner wires in position is a different matter. Kuboyama et al. (1981) demonstrated that drilling alone promoted healing, since in their patients the Kirschner wires were withdrawn as soon as drilling was completed. Companacci et al. (1975) and McKay and Nason (1977) also showed that fenestration was highly effective in normalising the hydrodynamic abnormality. Whether the wires are left in, or withdrawn immediately, it seems clear that the multiple drill-hole method is the best treatment of simple bone cysts in youth; the operative risk is minimal and the clinical result is excellent.

We would like to thank Dr Atsuko Heshiki, Associate Professor of Radiology, Gunma University Hospital, and Dr Tsunao Oogimi, Director of Orthopaedic Surgery, Maebashi Red Cross Hospital, for their help.

**REFERENCES**


THE AETIOLOGY AND TREATMENT OF SIMPLE BONE CYSTS


