ANEURYSMAL BONE CYSTS OF THE SPINE

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Aneurysmal bone cysts are uncommon lesions, especially in the spine. Seventy-eight cases have been previously documented in the English literature and an additional fourteen cases are now reported. There is a definite predilection for the lumbar region and the neural arch is the part of the vertebra most commonly affected. It is recommended that treatment should consist of total excision or when this is not possible, curettage. Radiotherapy should be reserved for those few cases where operation is inadvisable.

In 1942 Jaffe and Lichtenstein described two cases of a peculiar blood cyst which they termed an aneurysmal bone cyst. They recognised that the nature of the lesion was not clear, but they were of the opinion "that the blood-filled cavity should be regarded as simply a large venous pool". The term aneurysmal was used to denote the "blow out" radiographic appearance which resembles the saccular protrusion of the walls of an aneurysm and also because cystic blood-filled spaces are encountered at operation. The name has been generally accepted, though indeed it is neither an aneurysm nor a bone cyst.

![Graph showing age distribution of eighty-one patients with aneurysmal bone cysts](image)

Age distribution of eighty-one patients with aneurysmal bone cysts showing that this is a condition mainly affecting teenagers.

Aneurysmal bone cysts are uncommon and Dahlin (1967) found twenty-six (1.4 per cent) in 2000 primary bone tumours at the Mayo Clinic. The lesion has been reported in almost every bone, with a high incidence in the spine. This has been variously recorded from 3 per cent of sixty-six cases (Biesecker et al. 1970) to 20 per cent of ninety-five cases (Tillman et al. 1968). The aetiology of this condition is unknown and much debated. In the spine these lesions often present problems in diagnosis and management, and there is frequently a neurological involvement.

Because of the rarity of this condition and the small size of any one series, it was decided to analyse the clinical features of all adequately documented cases of aneurysmal bone cyst of the spine in the English literature. Twenty-seven authors have reported seventy-eight cases. To these we add fourteen cases of our own, a total of ninety-two. The details of the latter are summarised in Table I. In certain respects a proportion of the previously reported cases are incompletely documented.

**DIAGNOSIS**

**Clinical features**

**Sex incidence.** In all large series of aneurysmal bone cysts from the whole skeleton there is a slight predominance of female patients. Among our own patients with aneurysmal bone cyst of the spine, and also those in the literature, 57 per cent were female and 43 per cent male.

**Age.** Aneurysmal bone cyst is primarily a condition of the second decade. The incidence in the spine is the same as in the appendicular skeleton. In eighty-one patients with spinal lesions for whom there was sufficient information, the average age was 16.6 years (Fig. 1).

**Site.** Aneurysmal bone cysts have been described at every level of the spinal column except the coccyx (Fig. 2). Jaffe (1958) considered there was no bias in spinal distribution but in this series, taking into account the number of vertebrae in each region, there was a definite predilection for the lumbar spine (Fig. 2). These lesions are often not confined to one vertebra and six of our...
Table I. Case histories of fourteen patients

<table>
<thead>
<tr>
<th>Case number</th>
<th>Sex</th>
<th>Age (years)</th>
<th>Duration of symptoms</th>
<th>Symptoms</th>
<th>Signs</th>
<th>Radiological findings</th>
<th>Level</th>
<th>Site and extension in vertebra</th>
<th>Treatment</th>
<th>Result and follow-up</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>M</td>
<td>9</td>
<td>4 months</td>
<td>Back pain after injury</td>
<td>Tenderness. Restricted movement</td>
<td>Lytic lesion</td>
<td>L4</td>
<td>Spinous process—lamina and pedicles</td>
<td>Partial excision and curettage</td>
<td>Full recovery, 8 years</td>
</tr>
<tr>
<td>2</td>
<td>M</td>
<td>12</td>
<td>1 week</td>
<td>Back pain after injury</td>
<td>Tenderness. Restricted movement</td>
<td>Lytic lesion</td>
<td>L4</td>
<td>Transverse process</td>
<td>Excision</td>
<td>Full recovery, 7 years</td>
</tr>
<tr>
<td>3</td>
<td>F</td>
<td>10</td>
<td>2 months</td>
<td>Neck pain</td>
<td>Lower motor neurone signs in upper limb</td>
<td>Expanding lytic lesion</td>
<td>C7</td>
<td>Pedicle and transverse process</td>
<td>Curettage</td>
<td>Full recovery, 7 years</td>
</tr>
<tr>
<td>4</td>
<td>F</td>
<td>8</td>
<td>2 weeks</td>
<td>Back pain. Urinary incontinence</td>
<td>Almost paraplegic</td>
<td>Plain films thought normal. Myelogram—complete block</td>
<td>T5</td>
<td>Lamina—soft tissues</td>
<td>Partial excision and curettage</td>
<td>Full recovery, 4 years</td>
</tr>
<tr>
<td>5</td>
<td>F</td>
<td>9</td>
<td>6 months</td>
<td>Neck pain</td>
<td>Lump in neck</td>
<td>Large lytic lesion. Collapse of body. Displacement of vertebral artery</td>
<td>C3</td>
<td>Body—pedicle and transverse process</td>
<td>Curettage</td>
<td>Full recovery, 2 years</td>
</tr>
<tr>
<td>6</td>
<td>M</td>
<td>25</td>
<td>3 months</td>
<td>Back pain</td>
<td>Tenderness. Profound paraparesis</td>
<td>Erosion of pedicles and lamina. Myelogram—complete block</td>
<td>L1</td>
<td>Spinal process—lamine and pedicles</td>
<td>Excision. Recurrence 2 months later. Repeat laminectomy + 3000 rads</td>
<td>Full recovery, 1 year</td>
</tr>
<tr>
<td>7</td>
<td>F</td>
<td>16</td>
<td>5 months</td>
<td>Back pain</td>
<td>Local tenderness</td>
<td>Pathological fracture</td>
<td>T12</td>
<td>Body and pedicle</td>
<td>Curettage and grafting</td>
<td>Full recovery, 3 years</td>
</tr>
<tr>
<td>8</td>
<td>F</td>
<td>11</td>
<td>2 months</td>
<td>Thigh pain</td>
<td>Tenderness. Muscle spasm</td>
<td>Lytic lesion. Compression body</td>
<td>T12/L1</td>
<td>Body—neural arch</td>
<td>Needle biopsy unsuccessful. Open biopsy. 3500 rads in 4 weeks</td>
<td>Full recovery, 2 years</td>
</tr>
<tr>
<td>9</td>
<td>F</td>
<td>15</td>
<td>1 month</td>
<td>Neck, arm pain</td>
<td>Torticollis. Lower motor neurone signs in upper limbs</td>
<td>Lytic lesion</td>
<td>C5/6</td>
<td>Entire vertebra C6</td>
<td>Biopsy + 1500 rads</td>
<td>Full recovery, 1 year</td>
</tr>
<tr>
<td>10</td>
<td>M</td>
<td>36</td>
<td>6 months</td>
<td>Back pain</td>
<td>Almost paraplegic</td>
<td>Lytic lesion</td>
<td>T2/3</td>
<td>Spinal process—lamina and pedicles</td>
<td>Partial excision</td>
<td>Full recovery, 2 years</td>
</tr>
<tr>
<td>11</td>
<td>M</td>
<td>22</td>
<td>6 weeks</td>
<td>Back pain</td>
<td>Almost paraplegic</td>
<td>Lytic lesion</td>
<td>T6/7</td>
<td>Pedicle—lamina</td>
<td>Partial excision + 3400 rads 3 months later</td>
<td>Residual back pain. 4 years</td>
</tr>
<tr>
<td>12</td>
<td>M</td>
<td>6</td>
<td>6 months</td>
<td>Leg pain</td>
<td>Lower motor neurone signs in leg</td>
<td>Lytic lesion. Myelogram—large filing defect</td>
<td>L5</td>
<td>Pedicle—sacrum</td>
<td>Partial excision</td>
<td>Full recovery, 6 years</td>
</tr>
<tr>
<td>13</td>
<td>F</td>
<td>24</td>
<td>8 months</td>
<td>Neck pain</td>
<td>Lump felt. Lower motor neurone signs in upper limbs. Upper motor neurone irritation in lower limbs</td>
<td>Lytic lesion</td>
<td>C6/7</td>
<td>Spinal process—soft tissues, lamina and pedicles</td>
<td>Partial excision</td>
<td>Full recovery, 17 years</td>
</tr>
<tr>
<td>14</td>
<td>F</td>
<td>18</td>
<td>2 months</td>
<td>Back pain</td>
<td>Local tenderness</td>
<td>Lytic lesion</td>
<td>T10</td>
<td>Body</td>
<td>Curettage</td>
<td>Full recovery, 6 months</td>
</tr>
</tbody>
</table>

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fourteen patients were affected at more than one level (see Figs. 7 and 8).

It has been noted that the neural arch is the most common site. In an analysis of seventy-four cases, 40 per cent occurred in the bodies and 60 per cent in the pedicles, transverse processes, laminae and spinous processes (Fig. 3). Twenty-four were confined to a single anatomical region of the vertebra and this localisation was evident in two of our own cases.

Of the seventy-one patients for whom there was adequate information, local pain was the presenting symptom in 72 per cent. A mass was reported in 23 per cent. Eight patients were paraplegic and thirteen had serious long tract signs usually described as incomplete paraplegia or tetraplegia, a total of 29 per cent. A further 18 per cent had some root symptoms and signs.

**Radiological features**

These have been well described by Sherman and Soong (1957), Slowick, Campbell and Kettelkamp (1968) and many others and vary according to the site and maturity of the lesion. In the spinous or transverse processes where cortical restraint on expansion is minimal the eggshell-thin "aneurysmal" edge is seen (Figs. 4 and 5). In the bodies more advanced lesions may show partial collapse (Fig. 6) or erosion of contiguous vertebral bodies and adjoining ribs. A classical lesion is characterised by an expansile, osteolytic cavity often containing fine strands of bone and surrounded by an eggshell of blown-out cortex. Eight of our cases illustrate these features.

Destruction of an entire vertebral body (Fig. 7) may progress to vertebra plana (Schachar and Edwards 1974). An aneurysmal bone cyst may cross the posterior apophysical articulations and so be mistaken for a malignant neoplasm (Murray and Jacobson 1971). Tomography may be particularly informative in delineating the extent of vertebral involvement before operation. A myelogram should be carried out as soon as there is evidence of cord impairment—from above and below when there is complete block. An irregular, extradural defect is the most usual finding (Fig. 8).

Schobinger and Stoll (1957) noted a diffuse opacity of the cystic area during the venous phase of angiography in one of their cases. Lindbom et al. (1961) found evidence of an arteriovenous shunt in three cases. Billings and Werner (1972) emphasised the diagnostic value of this procedure: the arteries supplying the
lesions are normally formed and peripherally distributed. Indistinct pools of contrast media are visible within the cyst structure and are felt to reflect the sinusoidal distribution of the thin-walled vascular spaces seen histologically. Selective arterial catheterisation can be a useful guide to the blood supply of the cord and the extent of the mass (Fig. 9).
Differential diagnosis

The radiographic findings should allow a differential diagnosis from eosinophilic granuloma, haemangioma of bone, osteoblastoma, fibrous dysplasia and secondary malignancy, for example malignant lymphoma, leukaemia and neuroblastoma.

The predilection of aneurysmal bone cysts for the posterior elements of the vertebrae and the age at which they occur are atypical of secondary bone tumours.

Needle biopsy was attempted in two of our fourteen patients and was successful in only one. In the other it resulted in neurological deterioration presumably because of extradural haemorrhage. As discussed later, where open biopsy has occasionally cured the condition there seems little justification for needle biopsy which is likely to be negative and possibly dangerous.

OPERATIVE FINDINGS

Findings at operation vary and probably depend on the stage of development of the aneurysmal bone cyst. The eggshell-thin cyst of subperiosteal new bone which delimits the lesion is usually readily discernible. In some there is intense bleeding once the very thin outer shell of the bone has been removed. A variable amount of soft tissue will be present. It may be soft, brownish, fleshy and vascular, perhaps indicating a more actively developing phase, or it may be more cystic with a smooth, grey-purplish mottled appearance and thin trabeculae of grey tissue partitioning the interior, which contains unclotted blood. The lesion may appear to be invading muscle or soft tissue (Cases 4 and 13), or so surround the theca (Case 10) that it cannot really be separated from it without excessive bleeding. Characteristically the bony shell is continuous with the cortex of the adjacent bone. There is, therefore, a variable amount of solid tissue containing vascular channels and blood-filled cysts of varying size, depending on the state of development of the lesion. Bleeding appears to come from the soft tissue lining the cysts and may be profuse and difficult to control until all the lining has been removed.

PATHOGENESIS

Many theories have been postulated over the years and include the following: a vascular disturbance of bone (Lichtenstein 1953; Donaldson 1962; Slowick et al. 1968); a secondary manifestation of a pre-existing condition (Jaffe 1958); a reparative process (Coley 1949; Thompson 1954); trauma and subsequent haemorrhage (Barnes 1956); skeletal haemangioma (Wieberdink 1953; Hadders and Oterdoom 1956); and a haematoma which may have arisen as the result of leakage from a congenital haemangioma or trauma (Aegerter and Kirpatrick 1975).

While easily recognisable, the typical and classical appearance of an aneurysmal bone cyst consists of anastomosing cavernomatous spaces (Dahlin 1967). Varying sections may contain a densely cellular, compact structure with plump stromal cells, multinucleate giant cells and thin-walled blood vessels, or a preponderance of fibrous tissue with enlarging vascular spaces.

TREATMENT

Including our own, seventy-nine cases from the literature were analysed (Table II).

Treatment is a controversial matter. Nevertheless it is clear that this is a benign condition in which disappearance has occurred spontaneously (Sherman and Soong 1957) and following biopsy only (Murray and Jacobson 1971). Despite these observations, surgical treatment short of complete excision is followed by a high incidence of recurrence (MacCarty et al. 1961; Verbiest 1965). When complete excision is not feasible some authors (Barnes 1956; Beeler, Helman and Campbell 1957; Neuhauser 1956) advocate simple curettage of the lesion plus bone grafting if necessary, though bone grafting did not appear to have influenced the incidence of recurrence. A recurrence is best treated
by further partial or total excision with or without curttage.

Of the thirty-four patients treated by partial excision of the lesion and radiotherapy, two had recurrences which were treated successfully by total and partial excision respectively. Nine patients were treated by radiotherapy alone, one of whom had a recurrence (MacCarty et al. 1961).

**PROGNOSIS**

Despite the obvious difficulties associated with neurological involvement and inaccessibility of the lesions, the prognosis is excellent and no different from that of cysts elsewhere in the skeleton (Nobler, Higinbotham and Phillips 1968; Tillman et al. 1968).

All ten patients with recurrences recorded were free of symptoms after further treatment. Follow-up varied from three months to fourteen years. Tillman et al. (1968) found that the incidence of recurrence throughout the skeleton lessened with age, but in this series the average age for the ten patients with recurrences was 18.9 years. Recurrences usually occur rapidly and in these ten patients the interval from initial treatment to recurrence varied from one to twelve months with an average of four months, suggesting incomplete initial excision.

**DISCUSSION**

Since 60 per cent of these lesions affect the posterior elements of the vertebrae, a midline posterior approach provides ready access to allow excision of the cyst and, where indicated, decompression of the theca.

Myelopathy is an accepted complication of radiotherapy to the spine (Palmer 1972), though we cannot detect such a case after treatment for aneurysmal bone cyst. The exact incidence of radiation myelopathy is difficult to determine, but an estimate by Palmer (1972) based on the reported clinical series gives an incidence of 2.9 per cent with a latent period of up to five years. Radiation myelopathy may also occur even when the dosage falls below the established safety limits (Asscher and Anson 1962). Lichtenstein (1953), Sabanus et al. (1956) and Tillman et al. (1968) have reported sarcomatous change developing in six aneurysmal bone cysts after radiotherapy, three of which were in the spine. For these reasons we disagree with Nobler et al. (1968) who advocate radiotherapy with 2000 to 3000 rads as a primary method of treatment.

**REFERENCES**


Jaffe, H. L., and Lichtenstein, L. (1942) Solitary unicameral bone cyst with emphasis on the roentgen picture, the pathological appearance and the pathogenesis. Archives of Surgery, 44, 1004–1025.


