Aneurysmal bone cysts of the spine

Report of 17 cases

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Seventeen cases of aneurysmal bone cyst of the spine are reported. Clinical manifestations, methods of investigation, differential diagnosis, treatment, and pathogenesis are discussed. The condition is essentially a benign lesion, affects a young age group, and grows rapidly, often producing a paravertebral mass. Occasionally it becomes acutely exacerbated, causing irreversible damage to the spinal cord. For these reasons, prompt diagnosis and timely treatment are important. Excision of the lesion is the treatment of choice but, if not feasible, thorough curettage and radiotherapy not exceeding 2000 rads are indicated. With a two-stage operation, excision of the lesion is often possible, and the stability of the vertebral column is assured with bone-graft fusions.

Key Words • aneurysm • bone cyst • spine • radiation therapy

The term “aneurysmal bone cyst” was first introduced by Jaffe and Lichtenstein in 1942. A more detailed description of the condition was reported in 1950. These lesions have been reported as subperiosteal giant-cell tumors, ossifying subperiosteal hematomas, aneurysmal giant-cell tumors, and benign bone aneurysms.

The lesion consists of unclotted venous blood under pressure filling communicating bone spaces, causing expansion and ballooning of the affected bone. Histologically, the walls of the spaces are lined by an indistinct epithelium, and the stroma consists of a brownish fibro-osseous meshwork with a few giant cells, extravasated blood, hemosiderin-laden phagocytes, and fields of osteoid and new bone formation. Aneurysmal bone cysts comprise 1.4% of all bone tumors. They can occur in any part of the skeleton, and about 20% of cases involve the spine. They primarily affect young people: 80% to 90% of the cases have occurred in individuals below the age of 20 years. Both sexes are equally afflicted. The lesion usually grows rapidly, with the duration of symptoms averaging 4 months. There may be rapid exacerbation of symptoms, and within a short time severe damage may result to the underlying structures. On the other hand, these bone cysts occasionally become quiescent, and instances of regression and incidental findings at necropsy have been reported.

We present 17 cases of aneurysmal bone cyst of the spine treated by us during the last 25 years. The clinical course of the patients is summarized in Table 1. The diagnosis was confirmed in all cases by histological examination of the tissues. Five cases are described in detail since they demonstrate important points in the clinical course and treatment. Case 1 was reported previously.

Illustrative Case Reports

Case 2

This 35-year-old man had complained of slight intermittent backache and abdominal pain for 3 months before admission. His physician could find no abnormality on clinical examination, and radiographs of the spine were normal. The attacks continued until 2 days before admission, when they became severe and constant, and the patient noted that his legs were weak. On admission, signs of cord lesion were found at the T-6 level. Radiographs of the spine demonstrated an expanding destructive lesion of the body of the T-6 vertebra. Myelography showed complete block at the T-6 level. During the night of his admission he became totally paraplegic.

At operation a few hours later, a brownish extradural...
TABLE 1
Clinical summary of 17 cases of aneurysmal bone cysts of the spine

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Age (yrs), Sex</th>
<th>Symptoms &amp; Signs</th>
<th>Duration of Symptoms</th>
<th>Vertebrae Affected</th>
<th>Treatment</th>
<th>Results</th>
<th>Follow-Up Period</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>13, M</td>
<td>pain, mass</td>
<td>1 yr</td>
<td>L-3</td>
<td>excision</td>
<td>good</td>
<td>6 yrs</td>
</tr>
<tr>
<td>2</td>
<td>35, M</td>
<td>backache, leg weakness</td>
<td>1 mo</td>
<td>T-6</td>
<td>curettage</td>
<td>poor</td>
<td>1 yr</td>
</tr>
<tr>
<td>3</td>
<td>22, F</td>
<td>backache, leg weakness</td>
<td>6 mos</td>
<td>T-3</td>
<td>curettage &amp; radiotherapy</td>
<td>good</td>
<td>14 mos</td>
</tr>
<tr>
<td>4</td>
<td>35, F</td>
<td>mass</td>
<td>2 yrs</td>
<td>C-6 &amp; C-7</td>
<td>curettage, radiotherapy</td>
<td>good</td>
<td>2 yrs</td>
</tr>
<tr>
<td>5</td>
<td>24, F</td>
<td>backache &amp; leg weakness</td>
<td>8 mos</td>
<td>T-11</td>
<td>biopsy &amp; radiotherapy</td>
<td>good</td>
<td>2 yrs</td>
</tr>
<tr>
<td>6</td>
<td>18, M</td>
<td>neck &amp; shoulder pain</td>
<td>2 mos</td>
<td>C-2 &amp; C-3</td>
<td>curettage &amp; radiotherapy</td>
<td>good</td>
<td>2 yrs</td>
</tr>
<tr>
<td>7</td>
<td>13, F</td>
<td>tender mass &amp; paraparesis</td>
<td>1 mo</td>
<td>T-1</td>
<td>radiotherapy</td>
<td>good</td>
<td>2 yrs</td>
</tr>
<tr>
<td>8</td>
<td>11, M</td>
<td>tender mass</td>
<td>4 mos</td>
<td>C-2</td>
<td>excision</td>
<td>good</td>
<td>—</td>
</tr>
<tr>
<td>9</td>
<td>15, M</td>
<td>leg weakness</td>
<td>3 mos</td>
<td>C-6</td>
<td>curettage &amp; radiotherapy</td>
<td>good</td>
<td>1 yr</td>
</tr>
<tr>
<td>10</td>
<td>20, M</td>
<td>leg weakness</td>
<td>3 mos</td>
<td>C-7 &amp; T-1</td>
<td>curettage</td>
<td>poor</td>
<td>—</td>
</tr>
<tr>
<td>11</td>
<td>12, M</td>
<td>tender mass</td>
<td>4 mos</td>
<td>L-2 &amp; L-3</td>
<td>curettage</td>
<td>good</td>
<td>1 yr</td>
</tr>
<tr>
<td>12</td>
<td>18, M</td>
<td>backache &amp; mass</td>
<td>3 mos</td>
<td>T-11</td>
<td>curettage, graft &amp; radiotherapy</td>
<td>good</td>
<td>2 yrs</td>
</tr>
<tr>
<td>13</td>
<td>19, M</td>
<td>mass &amp; pain</td>
<td>10 days</td>
<td>C-5</td>
<td>curettage, partial excision, &amp; fusion</td>
<td>good</td>
<td>5 yrs</td>
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<tr>
<td>14</td>
<td>16, F</td>
<td>low-back pain &amp; sciatica</td>
<td>3 mos</td>
<td>S-1</td>
<td>curettage</td>
<td>good</td>
<td>5 yrs</td>
</tr>
<tr>
<td>15</td>
<td>14, F</td>
<td>mass, paresthesia</td>
<td>8 mos</td>
<td>C-5</td>
<td>excision</td>
<td>good</td>
<td>2 yrs</td>
</tr>
<tr>
<td>16</td>
<td>20, F</td>
<td>backache</td>
<td>5 mos</td>
<td>L-3</td>
<td>two-stage operation &amp; graft</td>
<td>good</td>
<td>1 yr</td>
</tr>
<tr>
<td>17</td>
<td>45, M</td>
<td>neck pain &amp; hand &amp; leg weakness</td>
<td>8 mos</td>
<td>C-6</td>
<td>curettage, excision, &amp; bone graft</td>
<td>good</td>
<td>1 yr</td>
</tr>
</tbody>
</table>

mass was removed and the tumor in the T-6 vertebral body was curetted. Postoperatively, radiotherapy (2000 rads) was given. There was little improvement in the patient's neurological deficit. At 8 months after the operation the sensory level had descended to L-1, but there was no useful movement of the lower limbs.

**Case 11**

This 12-year-old boy had a slight backache for 1 month. Examination revealed a small tender paravertebral mass, and changes in the radiographs were suggestive of an aneurysmal bone cyst of the L-2 vertebra. Surgery was advised but refused by his parents. Three months later and 1 week before his second admission, the patient became paraplegic. There was little improvement following operation.

**Comment.** These two cases demonstrate that progressive neurological deficit with sudden total paraplegia can occur in patients with this benign lesion. They also demonstrate the poor prognosis once complete paraplegia has developed. A corollary is that, with persistent backache in young people, repeated radiological examinations are advisable.

**Case 12**

This 18-year-old man had complained of slight backache for 3 months. One week before admission, he was sitting on his motorcycle with his feet on the ground and leaning forward on the handle bar when a friend put his hand on his back to help himself to stand up. The patient suffered a transient paraparesis, and was unable to stand. Recovery occurred spontaneously in 1 minute.

On admission, a paravertebral mass was found on the left side at T-11. The mass was firm, measured 4 x 6 cm, and was tender to the touch. The neurological examination was normal. Radiographs of the spine demonstrated an expansile destructive lesion involving the body and lamina of T-11. To reenact the incident on the motorcycle, just before the operation the patient was placed in a prone position with his legs bent at the hip and knees. Gentle but sustained pressure was applied on the mass by the palm of the hand, and within a few seconds first the left and then the right leg became weak.

At surgery, an eggshell-like capsule was found 2 cm below the skin; it was opened and a large amount of unclotted dark blood escaped. The cavity and the lesion in the vertebral body were curetted. There was profuse bleeding which was controlled by packing the area for 2 to 3 minutes with gauze soaked in hydrogen peroxide solution. The cavity in the vertebral body was partially filled with a bone graft. Postoperative radiotherapy (2000 rads) was given. When examined 2 years later, the patient was free of symptoms and there was no evidence of a recurrence.
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Case 16

This 20-year-old woman had suffered backache for 5 months. At first the pain was slight and localized to the low back, but it gradually increased in severity and extended into the posterior aspect of both thighs, becoming worse with activity. Six weeks before admission she noticed gradually increasing weakness of the legs, and on admission she was unable to walk. Examination revealed a left paravertebral mass at the L2-4 level. Pressure on the mass increased the pain, especially in the buttocks. No bruit was audible on auscultation of the mass. Atrophy, loss of tendon reflexes, and impairment of sensation were noted below the L-2 distribution. Plain radiographs demonstrated destruction of the L-3 vertebral body on the left side, with involvement of the lamina. A mass extending anteriorly and covered by a fine shell of bone could be seen at the L2-3 level on the lateral films. Myelography demonstrated a complete block at the L-3 level.

This patient was treated by a two-stage operation. At the first operation, the involved left lamina and pedicle were removed through a posterior approach. Brown friable tissue covering the dura was exposed, and was removed with little bleeding. Curettage of the vertebral body resulted in profuse bleeding, which was controlled by packing for 2 to 3 minutes with gauze soaked in hydrogen peroxide solution. The patient was transfused with 1500 ml of blood.

After the operation a plaster-of-Paris cast was applied. Recovery of function was rapid and she was able to walk 2 weeks after the operation. At the second operation 6 weeks later, the abdominal mass was exposed through a transperitoneal approach. The mass was lying to the left of the midline and was covered by a thin shell-like bone capsule; it was completely removed. The vertebral body and intervertebral discs were again curetted. Fusion of L2-4 was performed with a fibular bone graft. When examined a year later, the patient was free from symptoms and in good health.

Comment. This case demonstrates that, even when both the anterior and the posterior elements of a vertebra are involved, complete excision of the lesion is possible with a two-stage operation. To ensure the stability of the spine, application of a suitable bone graft is necessary.

Case 17

This 45-year-old man was admitted with an 8-month history of pain in the lower part of the neck. During the 4 weeks before admission he had noted some weakness in his right hand and both legs. Examination revealed weakness of grasp in the right hand with a positive Hoffman's sign, loss of abdominal reflexes, brisk knee and ankle jerks, and equivocal plantar responses. There was no sensory deficit and no sphincter disturbance. Plain radiographs of the cervical spine showed a destructive and expansile lesion of the C-6 vertebral body (Fig. 1 left). Computerized tomography (CT) demonstrated that the lesion involved the right pedicle and lamina (Fig. 2), and myelography showed a complete block at C-6. Scintigraphy of the skeleton did not reveal any abnormality apart from the C-6 lesion.

At operation through an anterior approach, the mass replacing the body of the vertebra was removed and the pedicle and anterior part of the right lamina were thoroughly curetted. A graft from the iliac crest was employed to replace the vertebral body. When examined 1 year later, the patient was free of symptoms and in good health. Radiographs of the cervical spine showed satisfactory fusion of the vertebrae (Fig. 1 right).

Analysis of Cases

These 17 patients included 10 males and seven females (Table 1). They ranged in age from 11 to 45

FIG. 1. Lateral radiographs of the cervical spine in Case 17. Left: Preoperative film demonstrating an expansile lesion of the body of C-6. The honeycomb appearance of the lesion is characteristic of an aneurysmal bone cyst. Right: Radiograph 1 year after the operation. Satisfactory fusion of the vertebrae is shown.

FIG. 2. Computerized tomography scan in Case 17 showing involvement of the pedicle and lamina.
years. The average age was 20.5 years, but only three patients were over 25 years old. The cervical spine was involved in eight cases, the thoracic spine in five, the lumbar spine in three, and the sacrum in one case.

In 94 cases of aneurysmal cyst of the spine reviewed by Hay, et al., 57% were female and the average age was 16.6 years. The cervical spine was affected in 22%, the thoracic vertebrae in 33%, the lumbar spine in 31%, and the sacrum in 13% of the cases. Although in most of the reported cases the posterior elements of the vertebrae were the site of the lesion, in our patients the anterior and posterior elements were equally affected. The vertebral body was involved in 12 cases, and the spinous process and lamina in five cases. In four cases, two vertebrae were involved. Aneurysmal bone cysts are probably the only benign lesions that involve adjacent vertebrae. In our series, the rate of growth of the lesion varied from case to case. In some patients, irreparable cord dysfunction occurred acutely. Duration of the first symptom before active treatment was initiated varied from 1 month to 2 years (average 6 months), and the duration of the symptom that precipitated admission varied from a few hours to 2 years (average 3 months).

Symptoms and Signs

The main symptoms in aneurysmal bone cysts of the spine are: pain, presence of a mass that is often tender to the touch, and symptoms secondary to spinal cord compression. Pain was present in nine of our cases, a mass in nine cases, and neurological symptoms in 12 cases, including three with total paraplegia. Pain is usually slight and intermittent at first, but becomes severe and constant when the lesion expands rapidly. Occasionally, pain is due to pressure on the nerve roots, as in one of our patients (Case 14) who complained of severe sciatica.

Nine patients presented with a mass; five of the lesions were in the cervical area, two in the thoracic area, and two in the lumbar region. In the five cases with cervical lesions, the mass was posterior in four and anterior, simulating a thyroid neoplasm, in one. The posterior masses are usually paravertebral, but the spinous process lesion in Case 15 was in the midline (Fig. 3). The mass is usually no more than 4 to 6 cm in diameter with variable consistency and is tender to the touch. Pressure on the mass may produce temporary signs of cord and root compression, as in Case 12.

Neurological symptoms varied from slight paresthesia to complete paraplegia. Recovery occurred following decompression in all cases except those presenting with complete paraplegia. Rapid neurological deterioration may occur, and in such instances surgery is indicated even more urgently to prevent irreversible damage to the spinal cord.

Methods of Investigation

Plain radiographs are of great value in diagnosis of aneurysmal bone cyst of the spine. Usually a destructive lesion is shown with expansion and trabeculae (Fig. 3), and some sclerosis of the surrounding bone is often present. Tomography may demonstrate the lesion more clearly. Angiography demonstrates the blood supply to the lesion along with pathological circulation and occasional arteriovenous shunts (Fig. 4). Myelography and CT scans demonstrate the extent of the lesion, and scintigraphy is helpful in the differential diagnosis when metastatic disease is a consideration. Visualization of the aneurysmal bone cyst by injection of an opaque substance into the cavities has been accomplished, but there is some risk associated with insertion of the needles. Needle biopsy has been carried out in at least two cases but one of those patients developed

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Fig. 3. Radiograph in Case 15 showing expansion of the C-5 spinous process due to an aneurysmal bone cyst.

Fig. 4. Angiography in Case 13 demonstrating an abnormal circulation with arteriovenous shunts.
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an extradural hematoma.\textsuperscript{12} Final diagnosis always rests with histological examination of the tissue.

Differential Diagnosis

Aneurysmal bone cysts of the spine must be differentiated from giant-cell tumors, hemangiomas, hydatid cysts, fibrous dysplasia, osteosarcoma, and metastatic lesions. Giant-cell tumor is rarely seen in patients younger than 20 years; it occurs mostly in the sacrum, and has no cavernous vascular spaces.\textsuperscript{11} Histologically, the giant cells are bigger and more numerous. Vertebal hemangioma is differentiated by its characteristic radiographic picture. The cavernous type affects the vertebral body, shows no ballooning, and does not involve neighboring vertebrae.\textsuperscript{21} Computed tomography with enhancement will easily differentiate an aneurysmal bone cyst, which contains blood, from hydatid disease of the bone, where the cysts contain lower-density fluid.

Fibrous dysplasia of the vertebra affects the same age group, usually includes more than one vertebra, often with visible involvement of the skull, grows slowly until the skeletal growth ceases, and is painless. Osteosarcoma and metastatic lesions produce much more pain, are seen in older age groups, and do not show any ballooning in the radiographs; nevertheless, in some cases only a biopsy can differentiate the two conditions.

Treatment

Total excision of the lesion is the treatment of choice, but it is not always feasible. We were able to excise the lesion in five cases. Curettage was carried out in 11 cases (followed by radiotherapy in seven), and in one case radiotherapy was the sole method of treatment after biopsy of the lesion. Bone grafting and fusion are often indicated following excision or thorough curettage of the lesion when both the anterior and posterior elements are involved, or when the body of the vertebra must be removed.\textsuperscript{2} 19

Radiotherapy is definitely of value and it is even claimed to be superior to surgery.\textsuperscript{7,20} There are two drawbacks to radiotherapy: malignant changes and post-irradiation myelopathy. It appears that radiotherapy not exceeding 2000 rads is safe. Embolization of the feeding vessels could be of value in aneurysmal bone cysts of other parts of the skeleton, but there is danger of interference with the blood supply to the cord in lesions of the spine.

Recurrence

There has been no recurrence in our cases, but follow-up monitoring is often not possible in Iran for social and geographic reasons. On the other hand, if there is a recurrence of symptoms, patients nearly always return to the original surgeon. In the series reported by Hay, et al.,\textsuperscript{12} of patients with aneurysmal cysts of the spine, there were two recurrences among 34 patients treated by partial excision and radiotherapy, and two recurrences among nine patients treated by radiotherapy alone. Recurrence of an aneurysmal bone cyst is unusual after 2 years and rare after 4 years.\textsuperscript{22} It is more likely to occur in patients younger than 15 years, and then appears within a few months of the operation. Even in those cases, the prognosis is good, as there is usually no recurrence after a second curettage. It is likely that, if at the first operation all the spaces were opened and curedt, there will be no recurrence.

Pathogenesis

There is no general agreement regarding the pathogenesis of aneurysmal bone cyst. Lichtenstein\textsuperscript{16} believed that: “the condition apparently results from some persistent local alteration in haemodynamics leading to increased pressure. The anomalous circulation could conceivably result from intrasosseous shunts.” These shunts have been demonstrated by angiography in several cases.\textsuperscript{23} The pressure in cavities of aneurysmal bone cysts has been measured by insertion of needles, and arterial, venous, and capillary pressures have been demonstrated.\textsuperscript{2} Besides the presence of anomalous circulation in these lesions, other factors are of importance. Nearly one-third of the cases appear with some other pathology, such as fibrous dysplasia, osteoblastoma, and chondromyxoid fibroma.\textsuperscript{4} Some authors believe that the condition arises from a preexisting pathology,\textsuperscript{6,14} and trauma is often mentioned in case histories. There were two instances of trauma in our 17 cases, and several cases of aneurysmal bone cysts associated with fractures have been reported.\textsuperscript{9} The most interesting case was in a newborn infant.\textsuperscript{10} Although aneurysmal bone cysts are not tumors, they may enlarge rapidly.

We suggest\textsuperscript{1} that aneurysmal bone cysts may be due to development of a hemodynamic vicious circle in a bone that is the site of a congenital vascular anomaly. This cycle is triggered by trauma or the development of other pathology which interferes with venous drainage of the area. The rate of growth of the lesion is adversely dependent upon the venous drainage. This hypothesis would explain the presence of communicating spaces containing unclotted venous blood under pressure, and ballooning of the affected bone. It would also explain why curettage of the lesion and even taking a biopsy may arrest or cure the condition in some cases. If this hypothesis is correct, the recurrence after the curettage relates to isolated cavities not being opened during the operation. In the same way, it can be presumed that radiotherapy cures the condition by its effect on small arteries, thus diminishing the blood supply to the lesion.

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Manuscript received January 15, 1985. Accepted in final form April 15, 1985. Address reprint requests to: Nosrat O. Ameli, M.D., 42 Hi Mount Drive, Willowdale, Ontario M2K 1X5, Canada.