Aneurysmal Bone Cyst Involving the Vertebral Column

A Case Report*

RICHARD A. McARTHUR, M.B., AND ROBERT G. FISHER, M.D.
Departments of Neurosurgery and Orthopedics, Mary Hitchcock Memorial Hospital and Dartmouth Medical School, Hanover, New Hampshire

An aneurysmal bone cyst was first described by Jaffe and Lichtenstein in 1942. In 1950 Lichtenstein described the lesion as a localized tumor occurring in long bones and vertebrae, and characterized by "blow out distention" of the skeletal contour; the term "aneurysmal" is therefore partly descriptive. The term also derives from another feature of the lesions, namely, that the cyst consists of many vascular spaces that grossly resemble a blood-filled cavity.

Since the original description the pathology of this lesion has been well documented by many authors. Aneurysmal bone cyst is most common in children and young adults, but it can occur later in life. Dahlin et al. report 2 cases out of 26, and Lichtenstein 2 cases out of 9, occurring in the third decade, while in Sherman and Soong's series, there were 8 cases out of 43 who were over 30 years of age, the eldest being 61. The vertebral column is one of the common sites of involvement. It may appear in the vertebral body alone, the neural arch, or a combination of both. The neural arch is the most frequent site. The cyst causes localized distention and destruction of the affected bone and is limited peripherally by a thin shell. The radiographic features are distinctive: the cyst appears as an expansile lytic lesion surrounded by a fine cortical shell. However, if the vertebral body is involved, collapse of the body can obscure the classical features. The usual clinical features are those of a young person with pain and stiffness in some segment of the spine. As the lesion enlarges, there may be additional symptoms due to pressure on the cord or nerve roots. All these features appear in the following case.

Case Report

This 9-year-old boy had a 6 months' history of back pain radiating to the right buttock and calf, stiffness in the back, and a swelling in the right lumbar region. Two weeks prior to his admission, he had developed a limp and had noticed aggravation of his symptoms on coughing. There was no weight loss, sphincter dysfunction, fever, malaise, or past history of sepsis.

Examination. He was a well-developed boy who moved cautiously and walked with a slight limp on the right. The spine listed to the right with moderate para-vertebral spasm. There was firm, tender fullness on the right side of the fifth lumbar vertebra in the iliolumbar angle. Back motion was limited by aggravation of the back pain. Examination of the legs revealed a right-sided weakness of the extensors of the foot and toes. Examination of sensation and the deep tendon reflexes was normal. Straight leg raising was possible to 40° on the right and 55° on the left.

General examination was normal. In particular, there were no petechiae, cafe au lait spots, bone tenderness, or abdominal mass; the genitalia were normal.

Laboratory Investigations. Blood and urine analyses were normal as was the alkaline phosphatase. Lumbar puncture revealed clear cerebrospinal fluid of normal pressure with a negative Queckenstedt test. Cerebrospinal fluid examination was normal including a total protein of 22 mg. per cent. X-ray of the chest and an intravenous pyelogram were normal. X-ray of the lumbar spine showed an expanding lesion involving the pedicle of the right side of the fifth lumbar vertebra extending to the lamina and up to the superior articular process (Fig. 1). That the rate of growth was rapid could be seen by comparing the x-ray 2 months prior to that taken immediately before surgery. The lumbar myelogram revealed a right-sided filling defect at the L-5 level.

Operation. A presumptive diagnosis of aneurysmal bone cyst was made and exploration of the lesion was carried out. As the soft tissues on the right side were reflected, there was a gush of bloody fluid. The cyst which had been entered extended from the right side of the spinous process to the transverse process of L-5; it had destroyed the lamina completely, as high as the lower margin of the lamina of L-4, and downward to the upper margin of the dorsum of the sacrum which was slightly eroded. A frozen section of the cyst made at this stage confirmed the diagnosis. The roof of the cyst was then removed revealing that it had destroyed the articular facet of L-4 to 5 on the right and that anteriorly it had extended through the ligamentum flavum where it was found to be attached to the dural sac and the roots of L-5 and S-1. The 5th lumbar root was swollen. Curettage of the cyst was carried out. The tissue consisted of a reddish brown material surrounding a multilocular arrangement of spaces.

There was no evidence of instability of the spine and a routine closure was carried out. At no time was excessive hemorrhage encountered, the moderate blood loss being due to a steady ooze.

Postoperative Course. The patient was seen at 1 month, 2 months, and 4 months after surgery and complained of no pain. However, there was residual tenderness on palpation in the right lumbar region. Spinal

motion improved, but still was limited. His gait was guarded and the weakness of the toe extensors was improving. An x-ray 2 months after surgery showed evidence of sclerosis of the margins of the cystic cavity indicating early healing, and at 4 months considerable filling in with new bone had occurred (Fig. 2).

Pathological study demonstrated the classical features of an aneurysmal bone cyst (Fig. 3). The stroma of the cyst had a mature fibrous tissue containing bone spicules and osteoid tissue; throughout this connective tissue stroma were many good sized blood lakes. These blood lakes were lined in places with endothelial cells but had no muscularis layer in their walls. Scattered throughout the stroma in relation to the blood lakes were many multinuclear giant cells and foamy macrophages. The giant cells seen in the lesion were smaller and had fewer nuclei than those found in a giant cell tumor. There was no evidence of malignancy either in the stroma or in the blood vessels.

**Discussion**

As Lichtenstein\(^6\) pointed out, there is a lack of awareness that the same pathological entity of aneurysmal bone cyst not only occurs in the long bones but also in the vertebral column. Frequently the lesion has been interpreted as a giant cell tumor, a sarcoma, or a hemangioma of a vertebra, and with treatment had a favorable outcome.

It should be stressed that this lesion is a benign entity; in fact it is not considered to be a true neoplasm, but rather a vascular malformation.\(^5,6\)

It also should be stressed that it occurs in the young age group and it is common in the vertebrae. Lichtenstein had 13 cases of aneurysmal bone cyst out of 50 which involved the vertebral column\(^9\) and Dahlin’s figures show 5 cases out of 26.\(^4\)
In dealing with these vertebral lesions, prompt diagnosis and treatment is indicated for they may enlarge rapidly and produce pressure symptoms of a serious nature. The duration of symptoms in 7 vertebral lesions studied by Beeler et al. averaged 3 to 7 months; the 6 month duration of our case is comparable. Pain, swelling, and limitation of motion usually precede the onset of neurological signs which ultimately can be profound when cord and nerve root pressure become marked.1 This particularly occurs in those lesions which cause collapse of a vertebral body or expansion of the inner aspect of the neural arch.

The treatment recommended is surgical curettage1,2,8,14 after the establishment of the diagnosis by histological study. This has proved successful even when there has been evidence of nerve root or cord pressure; often treatment results in a reversal of the neurological signs.13 One of the features of the aneurysmal bone cyst, as has been pointed out by Barnes,1 is that there is a tendency to heal even after incomplete removal; hence
Aneurysmal Bone Cyst of Vertebral Column

Fig. 3. Aneurysmal bone cyst. Mature fibrous stroma containing large blood lakes (A), and scattered small giant cells (B).

curettage is all that needs to be done. Radiotherapy also has been used successfully in the treatment of vertebral lesions. Beeler et al. treated 7 vertebral lesions by this method obtaining good results and no malignant degeneration. However, the current literature favors radiotherapy only for those lesions in the vertebral column which by x-ray appear to be inaccessible to surgery or when thorough curettage becomes impracticable due to surgical difficulties. Thus occasionally radiotherapy is used as a supplement to incomplete surgery. A note of warning has been sounded by Lichtenstein who has recorded a case of post-irradiation sarcoma occurring 6 years after the initial treatment.

The clinical recognition of an aneurysmal bone cyst of the vertebral column is of considerable practical importance. The myelogram may be helpful (see Fig. 4). The lesion is benign, responds readily to treatment, and can usually be distinguished from malignant lesions of the vertebral column. One benign lesion which must be distinguished is the hemangioma which usually involves the vertebral body exclusively at 2 or 3 adjacent levels. Radiographically, the hemangioma can be distinguished by the presence of vertical striations, in the lytic area, and histologically one sees a large number of vascular channels in a loose connective tissue stroma without any of the features of the aneurysmal bone cyst. Other benign lesions which must be considered in the differential diagnosis are benign osteoblastoma, a solitary focus of fibrous dysplasia, and giant cell tumor. These tumors may be difficult to differentiate except on a histological basis.

Summary

Aneurysmal bone cyst is a benign lesion which
occurs in the vertebral column as well as the long bones. This case report illustrates the characteristic clinical and radiological features and also the excellent response to treatment. We have pointed out that this lesion should be readily recognized and treated early in order to avoid the hazards of cord and nerve root pressure.

We wish to thank Dr. O. S. Staples of the Department of Orthopaedics for allowing us to present this case, Dr. Reginald K. House of the Department of Pathology who gave considerable help with preparation of the slides, and Dr. William C. MacCarty, Jr., of the Department of Radiology who was most helpful in revising the roentgenographic material.

References