Aneurysmal Bone Cyst of the Spine

BY R. CAPANNA, M.D.*, U. ALBISINNI, M.D.†, P. PICCI, M.D.†, P. CALDERONI, M.D.†, M. CAMPANACCI, M.D.†,
AND D. S. SPRINGFIELD, M.D.†, BOLOGNA, ITALY

From the Istituto Ortopedico Rizzoli, Bologna

ABSTRACT: The cases of twenty-two patients with an aneurysmal bone cyst of the spine above the sacrum were analyzed with regard to sex, age, site, symptoms, and radiographic findings. Four patients had extension of the lesion to the adjacent vertebra or rib, and twelve patients had neurological deficits. The primary treatment was either radiotherapy or surgery alone, or surgery and radiotherapy combined. No recurrences were found in patients who were treated with surgery alone or with surgery and radiotherapy, while three of the six patients who were treated with radiotherapy had a local recurrence, two of which were fatal. The patients with neurological deficits recovered after healing of the cyst.

Aneurysmal bone cyst is a benign bone lesion that is usually treated successfully by curettage when it involves the bones of the extremities. When it involves the spine it may be more serious because of the proximity of the lesion to the spinal cord. There is limited information in the literature regarding the treatment and the long-term results of the lesion in the spine. We reviewed the files at the Tumor Center of the Istituto Ortopedico Rizzoli. Twenty-two patients with an aneurysmal bone cyst of a vertebra were identified and their cases were analyzed.

Materials and Methods

We reviewed the files, and those of patients with the diagnosis of aneurysmal bone cyst were examined (198 patients observed between 1940 and 1982). Those with lesions in the cervical, thoracic, or lumbar spine were selected for this study (twenty-four patients). Only patients with a histologically confirmed diagnosis, adequate initial and follow-up radiographs, and at least two years of follow-up were considered (twenty-two patients). Two patients were excluded because of too-short a follow-up. The patients' charts were reviewed to determine the age, sex, date of diagnosis, initial complaints, physical findings on initial examination, treatment, and final results. The radiographs were reviewed to determine the exact location of the lesion and any evidence of extension, either extraosseous or to an adjacent bone. In eighteen patients tomograms and in three patients computed tomographic scans were also used. At follow-up, all of the patients had only plain radiographs made unless there was clinical or radiographic suspicion of a local recurrence. The lesion was considered radiographically healed when it filled with bone. Lesions with residual, small radiolucent areas surrounded by thick bone that remained stable on follow-up were also considered to be healed. Only lesions with a radiolucent area or soft-tissue mass that increased in size with time were considered to be recurrent.

Results

In the twenty-two patients whose cases were available for analysis, there was the usual increased incidence in female patients (thirteen female and nine male patients) and the majority of patients were less than twenty years old (Fig. 1). The lumbar spine was most commonly involved. Only one patient had had previous treatment (2600 rads of radiation). She was seen with a recurrence at the age of twenty, ten months after treatment. The lesion was located in the third lumbar vertebra.

Symptoms and Physical Findings

Twenty-one of the twenty-two patients had pain either in the back or due to involvement of a nerve root or the cord itself. One patient came to us with a painless paravertebral mass involving the twelfth thoracic vertebra and the adjacent rib. Four patients had nerve-root irritation, four had nerve-root compression, and four had central compression (cauda equina, two patients and spinal cord, two patients). Twelve patients had decreased motion of the spine. Two patients had scoliosis and one had kyphosis.

Radiographic Findings

The distribution of the lesions was as follows: three lesions each in the second cervical and second and third lumbar vertebrae; two lesions each in the first, fourth, and fifth lumbar vertebrae; one lesion each in the fifth cervical and seventh, eighth, eleventh, and twelfth thoracic vertebrae; and two lesions involving two adjacent vertebrae (the first and second and the seventh and eighth thoracic vertebrae). Two lesions, one in the seventh and one in the twelfth thoracic vertebra, showed involvement of an adjacent rib when the patients were first seen. The four lesions involving an adjacent vertebra or rib were all in the posterior elements of the thoracic spine, and the lesion had crossed to the adjacent bone either at the articulation between the posterior elements at the facet joint or at the articulation of the transverse process with the rib.
In seventeen patients the contour of the bone was expanded and there was disruption of the cortex. Ten patients had a reactive rim of bone surrounding the lesion while seven had a soft-tissue mass without radiographic evidence of a reactive rim.

No patient had involvement of the vertebral body alone. All patients had involvement of the posterior elements. Twelve patients had asymmetrical involvement of the body and posterior elements (pedicle and transverse process in two patients; pedicle and lamina in one patient; pedicle, transverse process, and lamina in four patients; pedicle, transverse process, articular processes, and lamina in four patients; and pedicle, transverse process, lamina, and spinous process in one patient). Two patients had symmetrical involvement of the body and posterior elements, so that the entire vertebra was destroyed (Fig. 3). Eight patients had no involvement of the body of the vertebra. Of these, three had unilateral involvement of the posterior arch (pedicle, transverse process, lamina, and adjacent rib in two patients and the entire hemiarch, including the articular and spinous processes, in one patient) and five had bilateral involvement of the posterior arch (spinous process and both laminae in two patients; spinous process and both laminae and pedicles in one patient; and the entire posterior arch, including the articular processes of two adjacent vertebrae, in two patients).

_Treatment and Follow-up_

One patient with a lesion at the second cervical vertebra had only a biopsy, but the lesion progressed postoperatively, and the patient complained of increasing pain. She had no neurological deficit and underwent a posterior fusion of the first, second, and third cervical vertebrae. The pain was relieved and she refused additional treatment. The lesion, initially located in the body and pedicles of the second cervical vertebra, slowly enlarged postoperatively, progressively involving the laminae, the spinous process, and part of the autogenous graft that had been used for the posterior fusion. Three years later the lesion stabilized spontaneously, and the patient had no complaints twenty-one years after treatment.

Six patients had only irradiation treatment (2600 to 5000 rads). Three of these patients responded well clinically and radiographically, and had no complications over the follow-up period of two years. The other three had complications. An eighteen-year-old man, in whom a lesion in the fourth lumbar vertebra compressed the cauda equina, had a recurrence nine months after receiving 3800 rads and was re-treated with 4000 rads, achieving healing of the cyst and neurological recovery. Seven years later, he sustained a pathological fracture of the fourth lumbar vertebra with a posterior subluxation of the third lumbar vertebra, causing a kyphotic deformity of the spine. Since side-bending radiographs showed a segmental spinal instability, first a posterior and then an anterior arthrodesis was performed to correct the deformity and to stabilize the spine. He had only occasional mild discomfort after eleven years and eight months of follow-up, and had no neurological deficit.

A second patient, a twenty-year-old woman, had a recurrence in the third lumbar vertebra and signs of compression of the cauda equina (no weight-bearing ability, urinary incontinence, and weakness of the quadriceps, tibialis an-
An aneurysmal bone cyst involving the posterior elements of two adjacent vertebrae (first and second thoracic) in a fourteen-year-old girl. The cyst healed after curettage and radiation therapy.

terior, and extensor digitorum muscles) eleven months after 2600 rads had been given as treatment. The body and posterior area (pedicle, lamina, and transverse and superior articular processes) on the right side of the third lumbar vertebra were involved. Curettage and anterior and posterior arthrodesis was then done. This patient had a second recurrence five months later, and a second curettage was done. One year later a third recurrence appeared, which was also treated by curettage. A fourth recurrence (one year after the third) was treated with 4000 rads. The patient then had a large intra-abdominal mass displacing the ureter and was paraplegic, and the lesion did not respond to the therapy. The patient died of the complications of radiotherapy (cystitis, septicemia, and endocarditis) four years and ten months after she was first seen.

A forty-six-year-old man, whose lesion was in the body, pedicle, and transverse process of the fifth cervical vertebra, had signs of root compression and was treated with 3500 rads. He improved clinically but had extension of the lesion to the fourth and sixth cervical vertebrae. Additional treatment was refused until neurological signs of compression of the cord began to develop, fifteen and a half years after the original treatment. The patient, having become tetraplegic, then underwent posterior decompression without biopsy in another hospital, but died in the immediate postoperative period. No autopsy was done and no material was obtained for histological examination. Therefore, whether this case represents a recurrence or a radiation-associated sarcoma is not known. This patient was the only one who had extension of the lesion to other bones after the initial treatment.

Six patients had curettage and irradiation treatment (2500 to 4800 rads) and one patient had a partial resection, curettage, and irradiation treatment (3000 rads). None of these patients had a recurrence or any complication. One was followed for only eighteen months and the others were followed for more than four and a half years (range, four years and seven months to twenty-four years).

Seven patients had curettage and one had partial resection and curettage. There were no recurrences or complications in this group. All of these patients were followed for more than five and a half years (range, five years and seven months to fourteen years).

All of the patients who were seen with localized back pain or root irritation were essentially asymptomatic and neurologically normal at follow-up. Three of the four patients with initial signs of root compression (with lesions in the third lumbar, fourth lumbar, and second thoracic vertebrae) had complete regression of the neurological abnormalities. The fourth patient, who had involvement of the fifth cervical vertebra, as previously discussed, died with tetraplegia sixteen years after the initial diagnosis.
Two patients were seen with central compression of the cord. Both patients had an aggressive lesion, involving the posterior elements of two adjacent vertebrae (the seventh and eighth thoracic vertebrae) in one and the posterior elements of one vertebra (the seventh thoracic vertebra) and the adjacent rib in the other. At admission, both patients were unable to walk and had complete paraplegia, hyposthesia, hyperreflexia, and a positive Babinski sign. The treatment consisted of decompression laminectomy, curettage, and radiation therapy. The patient with a lesion in the seventh thoracic vertebra and the adjacent rib had complete neurological recovery postoperatively except for a persistent hyperreflexia. The patient with a lesion of the seventh and eighth thoracic vertebrae improved postoperatively, and at follow-up six years later he had complete recovery of sensitivity and sphincter control and residual weakness of the flexor-extensor muscles of the foot, and was able to walk with crutches.

Two patients were seen with compression of the cauda equina. The one with involvement of the fourth lumbar vertebra had complete neurological recovery but, as previously discussed, had a late pathological fracture of the body of the vertebra that required an anterior and posterior arthrodesis. The other patient, who had involvement of the third lumbar vertebra, also previously discussed, had some initial neurological recovery but regressed, and after multiple recurrences this patient died paraplegic.

**Discussion**

The spine is one of the common locations for aneurysmal bone cysts (11 per cent of the lesions in the files of our center). The lumbar vertebrae are most often affected. The patients are usually younger than twenty years old and have complaints of back or root pain, usually with a decrease in the range of motion of the spine and a neurological deficit.

Involvement of the posterior elements (pedicle or lamina, or both) was always present, and no patient had involvement of the vertebral body alone. About 40 per cent of the patients (eight of twenty-two) had involvement of only the posterior elements; in these patients the lesion always involved the lamina and the spinous process and fre-
The appropriate treatment for aneurysmal bone cysts of the spine is controversial, particularly as regards the role of irradiation\textsuperscript{14}. The results of treatment in our patients suggested that there was no benefit from the addition of irradiation to curettage or to partial resection and curettage. All patients who had curettage or partial resection and curettage, whether or not irradiation was added, had an excellent result, whereas irradiation without surgery was associated with a poor result in half of the patients.

We therefore recommend resection of the lesion when it involves only the transverse or spinous process, and we advise curettage for the other, more central lesions. When there is involvement of the body, we recommend careful curettage, through an anterior approach if necessary, with decompression of the nerve root or cord if either is compromised. The addition of postoperative irradiation is logical if, at operation, some infiltrative tissue remains in the patient and cannot be removed.

Neurological recovery from deficits and radiographic healing are to be expected in most patients (Figs. 4-A through 5-B). Persistent radiolucencies within the treated lesion are not necessarily of concern but should be followed. If there is no progression of a persistent radiolucent area, no treatment is necessary. If the lucency increases in size, repeat curettage is then the recommended treatment. The lesion can progress slowly or recur after many years. Patients should therefore be followed for at least several years after the treatment.

References