Case Report

Osteoid osteoma of the spine: surgically correctable cause of painful scoliosis

Helmut Haibach,* MD
Corinne Farrell,† MD
Robert W. Gaines,‡ MD

Osteoid osteoma is a small benign neoplasm of bone that may occur in any part of the skeleton.[1] When present in the spine it is often associated with pain and scoliosis.[2,3] Surgical removal usually leads to prompt relief of the painful muscle spasm and correction of the scoliosis. The importance of the small bone lesion, therefore, lies in its recognition as a cause of painful scoliosis that is surgically correctable by simple removal.

In many cases the lesion is recognized only after years of unsuccessful attempts to correct painful scoliosis with braces.[4] Repeated radiographic studies including computed tomography may be required to detect the small, elusive lesion. Although osteoid osteoma is an uncommon cause of scoliosis, a thorough search for this lesion in cases of painful scoliosis can spare patients years of frustration and disability.

Case reports

Case 1

A 16-year-old well-muscled boy had had scoliosis and unremitting back pain centred at the thoracolumbar junction for 18 months. The pain was unchanged by activity but relieved by acetylsalicylic acid. The spine was curved to the left in the thoracolumbar region. With forward bending there was marked rigidity and moderately severe bilateral paravertebral muscle spasm.

Radiographs demonstrated scoliosis convex to the left and tomograms a radiolucent area in the right pedicle of the 10th thoracic vertebra corresponding to a focal scintigraphic site of increased activity. Resection of the lesion completely relieved the pain, and the scoliosis resolved spontaneously in 2 to 4 weeks. The surgical specimen had the histopathologic features of osteoid osteoma.

Case 2

A 13-year-old girl had had low back pain and scoliosis for approximately a year. There was mild to moderate scoliosis of the entire spine convex to the left, with a thoracic hump 2 cm high. Tomography revealed a single radiolucent area 5 mm in diameter in the body of the first lumbar vertebra (L1) (Fig. 1). Previous bone scintigraphy had shown minimally and diffusely increased activity in the thoracolumbar region and thus had not localized the lesion. After resection of the left pedicle of L1 the lesion was removed by thorough curettage of the surrounding bone. There was prompt relief from pain and progressive straightening of the spine. Eighteen months later the patient was still symptom free. The surgical specimen had the histopathologic features of osteoid osteoma.

Case 3

An 8-year-old girl had had scoliosis for 2 years and lumbar pain for 3 months. She had been treated with braces by her local orthopedic sur-

*From the departments of *†Pathology and ‡Surgery, University of Missouri-Columbia and †Central Washington University, Ellensburg, Wash.*

Reprint requests to: Dr. Helmut Haibach, Department of Pathology, University of Missouri-Columbia, Number One Hospital Dr., Columbia, MO 65212, USA

geon. Thoracolumbar scoliosis convex to the left was evident. With forward bending the left paravertebral area was higher than the right. Radiographs with tomography (Fig. 2) revealed sclerosis of the second lumbar vertebra (L2) involving the right transverse process, pedicle, lamina and superior articular process and a small radiolucency in the lamina. Scintigraphy showed a small focus of increased activity in the upper lumbar spine corresponding to the radiolucent area.

Two operations were performed. In the first, a red nidus 1 cm in diameter was curetted from the lamina; in the second, 18 months later, a new expansile lesion was removed by extirpation of the right transverse process, pedicle and superior articular process. During 3 years of follow-up there was progressive straightening of the spine and no pain.

The first surgical specimen, approximately 2 g of red to tan bony fragments, contained sclerotic bone and nidus, the latter consisting of equal amounts of narrow, disorganized bone trabeculae and loose vascular connective tissue (Fig. 3). The second specimen consisted of several bony fragments measuring up to 2 × 3 cm with identical histopathologic findings.

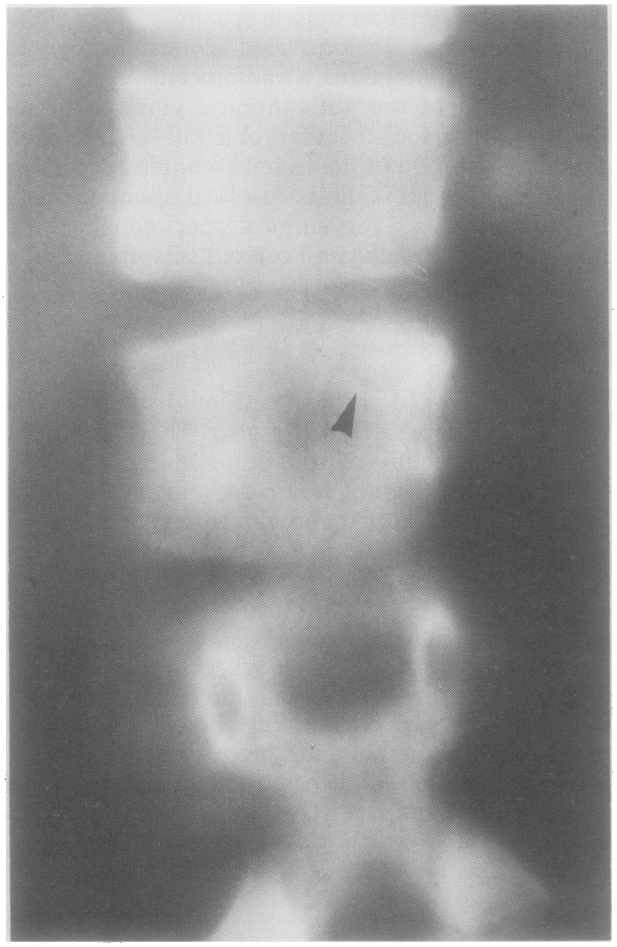

Fig. 1 — Case 2: Tomogram of lower thoracic and upper lumbar spine at 8.1 cm shows radiolucency in body of first lumbar vertebra (arrow), with denser core, suggesting nidus.

Discussion

Several reviews and series of osteoid osteoma of the spine have been published in the last three

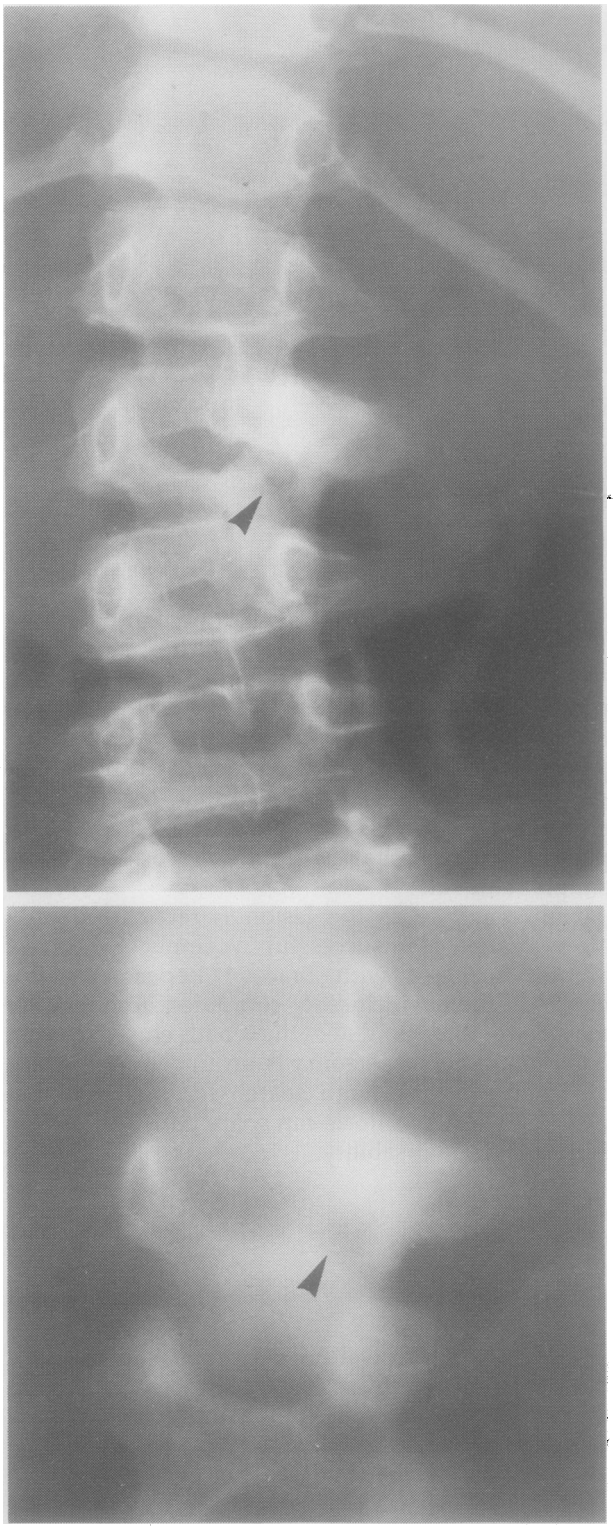

Fig. 2 — Case 3: Radiograph in top panel shows scoliosis of lumbar spine convex to left and sclerosis of second lumbar vertebra (L2). Tomogram in lower panel at 4.0 cm shows sclerosis of L2 involving right transverse process, pedicle, lamina and superior articular process. Arrows indicate central radiolucency in lamina.

decades, each with different emphasis. Some describe symptoms and the distribution in the axial skeleton;[3,5-7] later reports discuss improved diagnostic tools[8] and outcome of surgery;[9] others address the delineation from osteoblastoma.[10] From these and reports of individual cases[11-19] a fairly consistent picture emerges.

Approximately 10% (0% to 25%) of osteoid osteomas occur in the spine,[3,5,6] almost 60% being in the lumbar, 27% in the cervical, 12% in the thoracic and 2% in the sacral region.[3] Two-thirds to four-fifths of these are diagnosed in patients 10 to 20 years old. Males predominate 2:1 to 4:1.[1,5] Scoliosis is associated in at least two-thirds of the cases,[3] and the osteoid osteoma is almost always on the concave side. According to Janin and associates,[3] who reviewed 82 cases, the parts of any vertebra most likely to be involved alone are, in order of decreasing frequency, the lamina (in one-third of cases), the pars interarticularis, the pedicle and the transverse process.

The clinical presentation is rather uniform.[7] Pain is the most prominent symptom in almost all cases and is usually classified as severe. It is increased by motion and described as radicular in half of the cases. Rarely is the spinal canal narrowed, in contrast to cases of osteoblastoma. Salicylates offer relief in one-third of the cases. Stiffness of the spine as a symptom is underrated, according to Kirwan and colleagues.[4] Most importantly, in most cases the diagnosis is delayed from months to years (7 to 24 months in two-thirds of the patients).[4]

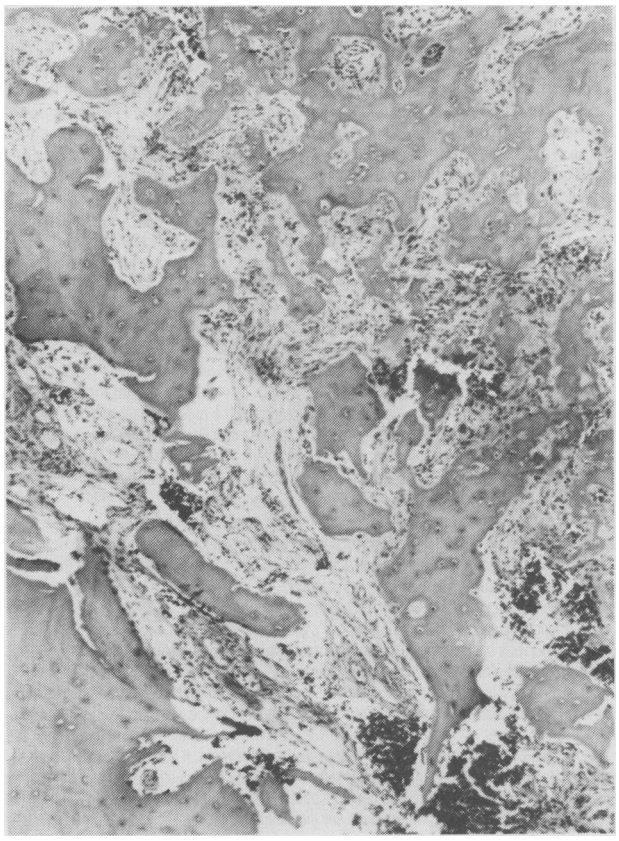

Fig. 3 — Case 3: Microscopic view of fragment of curettage specimen shows nidus in right upper corner, separated by band of loose vascular connective tissue from dense bone in left lower corner (hematoxylin-eosin; ×100).

Conventional tomography often fails to detect an osteoid osteoma because of the complex anatomy of the vertebra. Computed tomography is successful more often and with contrast enhancement may demonstrate increased vascularity in radiolucent areas corresponding to the nidus. The differential diagnosis of such "blushes" includes, besides neoplasms, abscess formation.[2,3] If the diagnosis is, instead, "reactive sclerosis and bony hypertrophy", which may be a separate entity or an osteoid osteoma that could not be visualized and resolved spontaneously, unnecessary surgery will be prevented.[19]

Scintigraphy may show a focal increase in the uptake of bone-seeking agents.[15] A negative scan has been reported in a single case.[12] In one of our cases scintigraphy with technetium Tc 99m phosphonate also failed to localize the lesion.

The preferred surgical approach to osteoid osteoma of the spine is curettage rather than en bloc resection: the latter can interfere with the stability of the spine and necessitate grafting and fusion. Removal of the entire nidus is usually accomplished by curettage alone and is considered essential for cure of the symptoms.[3] However, a second operation may prove necessary, as in our case 3. The persistence of this patient's pain after the first operation and the proximity of the second lesion to the first raise the question whether the second lesion represented a recurrence. Several observations weigh against this possibility: careful inspection of the site of the first lesion revealed no remnant of tumour; the second lesion was at least 1 cm from the site of the first; although several cases of recurrent osteoid osteoma of the spine have been described,[11] curettage was curative in most; and osteoid osteomas in two different bones have been reported.[6] MacLellan and Wilson[5] reported on two patients requiring two operations and one patient requiring three.

Osteoid osteomas of the spine share the pathological features of osteoid osteomas elsewhere in the skeleton.[5,14,16,17] On radiographs most osteoid osteomas measure 0.3 to 1.0 cm in diameter. The colour is usually described as red or pink, and occasionally as yellow, the consistency as soft granular or gritty. The typical lesion consists of three components: a softer bony nidus (bone trabeculae with interposed osteoid and loose vascular connective tissue) surrounded by loose vascular connective tissue (corresponding most likely to the radiolucent ring occasionally seen [Fig. 1]), in turn surrounded by sclerotic hard bone (usually dense and reactive and corresponding to the sclerosis seen on radiographs and probably the increased accumulation of bone-seeking agents seen on scintiscans[16]).

By convention, lesions that on tomograms are less than 2.0 cm in diameter are called osteoid

osteomas and larger lesions osteoblastomas.[10] However, because osteoid osteoma and osteoblastoma are similar in light microscopic and ultrastructural features, alternative terms have been proposed: "circumscribed osteoblastoma" and "genuine osteoblastoma" respectively.[10]

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