

Case Report

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

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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

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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-

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.

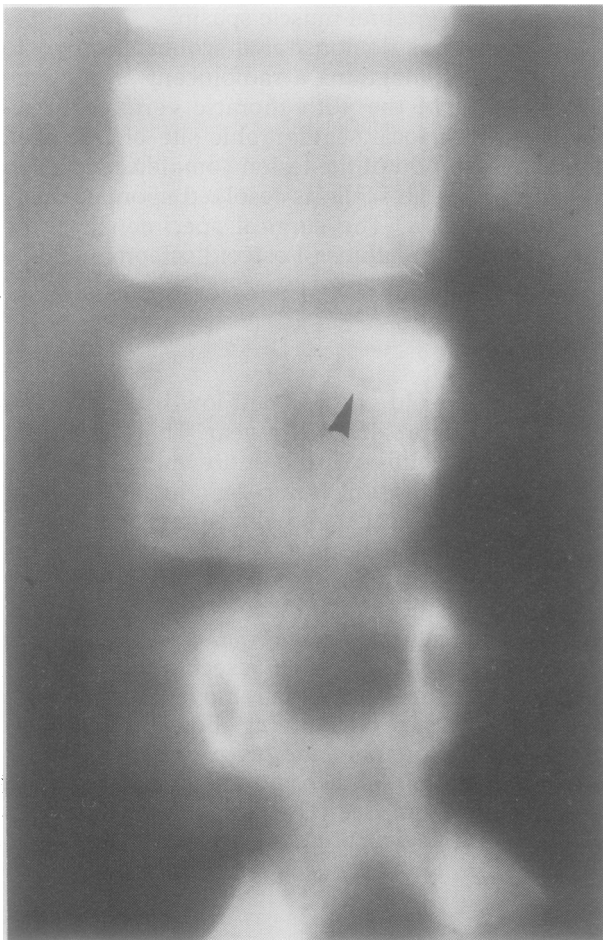


Fig. 1 — Case 2: Tomogram of lower thoracic and upper lumbar spine at 8.1 cm shows radiolucency in body of first lumbar vertebra (arrow), with denser core, suggesting nidus.

Discussion

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

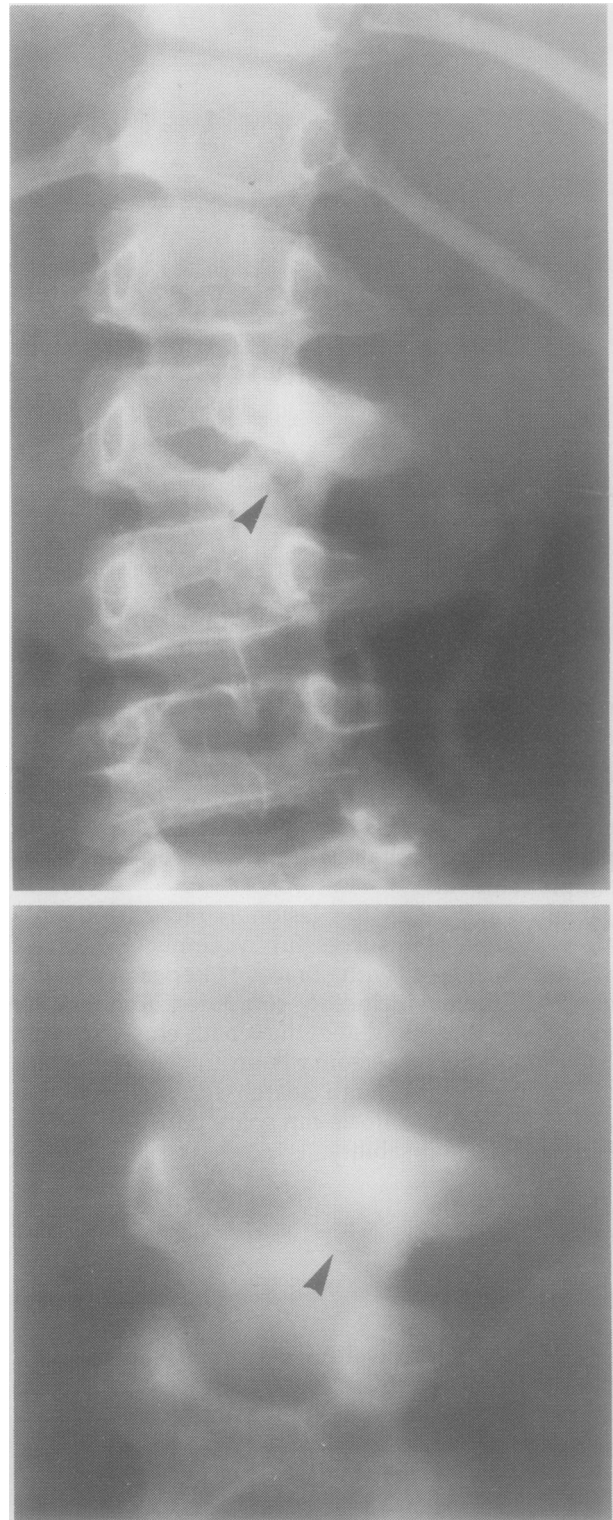


Fig. 2 — Case 3: Radiograph in top panel shows scoliosis of lumbar spine convex to left and sclerosis of second lumbar vertebra (L2). Tomogram in lower panel at 4.0 cm shows sclerosis of L2 involving right transverse process, pedicle, lamina and superior articular process. Arrows indicate central radiolucency 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 tools⁸ and outcome of surgery;⁹ others address the delineation from osteoblastoma.¹⁰ From these and reports of individual cases¹¹⁻¹⁹ 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,

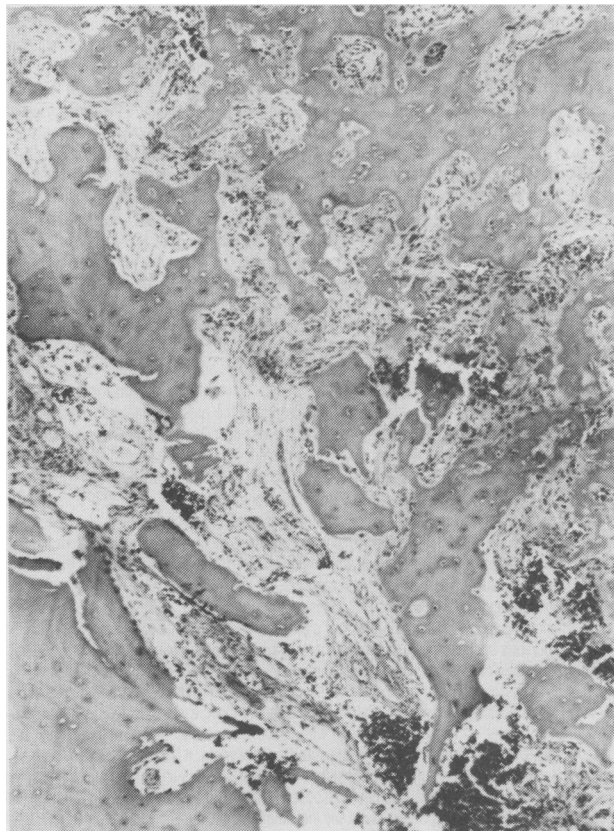


Fig. 3 — Case 3: Microscopic view of fragment of curettage specimen shows nidus in right upper corner, separated by band of loose vascular connective tissue from dense bone in left lower corner (hematoxylin-eosin; $\times 100$).

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

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 Wilson⁵ 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 scintiscans¹⁶).

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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and generally occurs during the first 4 weeks of therapy. It is usually mild and disappears within a few days of dosage reduction, short term treatment with an antihistaminic agent, and/or discontinuing therapy; remission may occur even if captopril is continued. Pruritus without rash occurs in about 2% of patients. Between 7 and 10% of patients with skin rash have shown an eosinophilia and/or positive ANA titers. A reversible associated pemphigoid-like lesion, and photosensitivity, have also been reported. **Allergic:** Angioedema of the face, mucous membranes of the mouth, or of the extremities has been observed in approximately 0.1% of patients and is reversible on discontinuance of captopril therapy. Serum sickness and bronchospasm have been reported. One case of laryngeal edema has been reported. **Cardiovascular:** Hypotension may occur; see WARNINGS and PRECAUTIONS [Drug Interactions] for discussion of hypotension on initiation of captopril therapy. Tachycardia, chest pain, and palpitations have each been observed in approximately 1% of patients. Angina pectoris, myocardial infarction, Raynaud's syndrome, and congestive heart failure have each occurred in 0.2 to 0.3% of patients. Flushing or pallor has been reported in 0.2 to 0.5% of patients. **Alterations in Taste:** 2% of patients receiving ≤ 150 mg/day of CAPOTEN developed a diminution or loss of taste perception. At doses > 150 mg per day, 7% of patients experienced this effect. Taste impairment is reversible and usually self-limited (2 to 3 months) even with continued drug administration. Weight loss may be associated with the loss of taste. The following have been reported in about 0.5 to 2% of patients: **Gastrointestinal:** gastric irritation, abdominal pain, nausea, vomiting, diarrhea, anorexia, constipation, aphthous ulcers, peptic ulcer. **CNS:** dizziness, headache, malaise, fatigue, insomnia, paresthesia. **Others:** dry mouth, dyspnea, cough, alopecia, impotence, loss of libido, disturbed vision, and itching and/or dry eyes. **Altered Laboratory Findings:** Elevations of liver enzymes have been noted in a few patients but no causal relationship to captopril use has been established. Rare cases of cholestatic jaundice, and of hepatocellular injury with or without secondary cholestasis, have been reported. Elevation of BUN and serum creatinine may occur, especially in patients who are volume-depleted or who have renovascular hypertension. In instances of rapid reduction of longstanding or severely elevated blood pressure, the glomerular filtration rate may decrease transiently, resulting in transient rises in serum creatinine and BUN. Small increases in the serum potassium concentration frequently occur, especially in patients with renal impairment (see PRECAUTIONS). **DOSE AND ADMINISTRATION** CAPOTEN (captopril) should be taken one hour before meals. **DOSAGE MUST BE INDIVIDUALIZED. Adults: Hypertension:** Initiation of therapy requires consideration of recent antihypertensive drug treatment, the extent of blood pressure elevation, salt restriction, and other clinical circumstances. If possible, discontinue the patient's previous antihypertensive drug regimen for one week before starting CAPOTEN. The initial dose of CAPOTEN is 25 mg t.i.d. If satisfactory reduction of blood pressure has not been achieved after one or two weeks, the dose may be increased to 50 mg t.i.d. The dose of CAPOTEN in hypertension usually does not exceed 50 mg t.i.d. Therefore, if the blood pressure has not been satisfactorily controlled after 1 to 2 weeks at this dose (and the patient is not already receiving a diuretic), a modest dose of a thiazide-type diuretic (e.g., hydrochlorothiazide, 25 mg daily) should be added. The diuretic dose may be increased at 1 to 2 week intervals until its highest usual antihypertensive dose is reached. In a patient already receiving a diuretic, CAPOTEN therapy should be initiated under close medical supervision (see WARNINGS and PRECAUTIONS [Drug Interactions] regarding hypotension), with dosage and titration of CAPOTEN as noted above. In severe hypertension, if further blood pressure reduction is required, the dose may be increased to 100 mg t.i.d., and then, if necessary to 150 mg t.i.d., while continuing the diuretic. The usual dose range is 25 to 150 mg t.i.d. A maximum daily dose of 450 mg CAPOTEN should not be exceeded. For patients with accelerated or malignant hypertension, when temporary discontinuation of current antihypertensive therapy is not practical or desirable, or when prompt titration to more normotensive blood pressure levels is indicated, diuretic should be continued but other concurrent antihypertensive medication should be stopped and CAPOTEN dosage promptly initiated at 25 mg t.i.d., under close medical supervision. When necessitated by the patient's clinical condition, the daily dose of CAPOTEN may be increased every 24 hours under continuous medical supervision until a satisfactory blood pressure response is obtained or the maximum dose is reached. In this regimen, addition of a more potent diuretic, e.g. furosemide, may also be indicated. Beta-blockers may also be used in conjunction with CAPOTEN therapy, (see PRECAUTIONS - Drug Interactions) but the effects of the two drugs are less than additive. **Heart Failure:** Initiation of therapy requires consideration of recent diuretic therapy and the possibility of severe salt/volume depletion. In patients with either normal or low blood pressure, who have been vigorously treated with diuretics and who may be hyponatremic and/or hypovolemic, a starting dose of 6.25 or 12.5 mg t.i.d. may minimize the magnitude or duration of the hypotensive effect (see WARNINGS, [Hypotension]). For these patients, titration to the usual daily dosage can then occur within the next several days. For most patients the usual initial daily dosage is 25 mg t.i.d. After a dosage of 50 mg t.i.d. is reached, further increases in dosage should be delayed, where possible, for at least two weeks to determine if a satisfactory response occurs. Most patients studied have had a satisfactory clinical improvement at 50 or 100 mg t.i.d. A maximum daily dose of 450 mg of CAPOTEN should not be exceeded. CAPOTEN is to be used in conjunction with a diuretic and digitalis. Therapy must be initiated under very close medical supervision. **Dosage Adjustment in Renal Impairment:** Because CAPOTEN is excreted primarily by the kidneys, excretion rates are reduced in patients with impaired renal function. These patients will take longer to reach steady-state captopril levels and will reach higher steady-state levels for a given daily dose than patients with normal renal function. Therefore, these patients may respond to smaller or less frequent doses. Accordingly, for patients with significant renal impairment, initial daily dosage of CAPOTEN should be reduced, and smaller increments utilized for titration, which should be quite slow (1 to 2 week intervals). After the desired therapeutic effect has been achieved, the dose should be slowly back-titrated to determine the minimal effective dose. When concomitant diuretic therapy is required, a loop diuretic (e.g., furosemide) rather than a thiazide diuretic, is preferred in patients with impaired renal function. CAPOTEN is removed by hemodialysis. **AVAILABILITY** CAPOTEN (captopril) is available as tablets containing: 25 mg of captopril - white, square, quadrisect scored on one side and imprinted CAPOTEN 25 on the other. 50 mg of captopril - white, oval, biconvex with a partial bisecting score and SQUIBB imprinted on one side and imprinted CAPOTEN 50 on the other. 100 mg of captopril - white, oval, biconvex with a partial bisecting score and SQUIBB imprinted on one side and imprinted CAPOTEN 100 on the other. **Storage:** Store at room temperature. Protect from moisture. Keep bottles tightly closed. **Product monograph available to physicians upon request.**

The following table which is based on theoretical considerations may be useful as a guide to minimize drug accumulation.

Creatinine Clearance (mL/min / 1.73 m ²)	Dosage Interval (Hours)
> 75	8
75-35	12-24
34-20	24-48
19-8	48-72
7-5	72-108
	(3 to 4.5 days)

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