OSTEOID OSTEOMA AND BENIGN OSTEOBLASTOMA OF THE SPINE

ALICI E., M.D. PINAR H, M.D. KARAKAŞLI A, M.D.

Summary: *12 cases of osteoid osteoma (n:8) and benign osteoblastoma (n:4) of the spine were reviewed at an average of 3.9 years (range, six months to 10 years) after surgical removal. There were eight boys and four girls with an average age at the onset of symptoms of 12.8 years (range, 5 to 18 years). The average delay between the onset of symptoms and definitive diagnosis was 23 months (range, 12 to 48 months). All patients with osteoid osteoma of thoracolumbar spine presented with painful scoliosis and one patient with cervical involvement presented with torticollis. All the lesions were situated in the posterior elements. At follow-up all the patients were free of pain. Only one patient could not achieve full spinal mobility.*

Osteoid osteoma is a benign bone-forming tumor characterized by Jaffe in 1935. It constitutes 11% of all benign bone tumors (5). Its reported incidence in the spine has ranged from zero to 25 per cent, with an average of about 10 per cent (11, 24). The term "benign osteoblastoma" was independently proposed by Jaffe (1956) and Lichtenstein (1956). It is an uncommon tumor that accounts for fewer than 1 per cent of all bone tumors (9). Howewer, more than 40 per cent of the reported cases have been located in the spine (12).

A scoliosis can arise in response to a painful osseous lesion situated unilaterally in the spine or in the posterior end of the ribs. Benign osteoblastoma and osteoid osteoma are the most common causes of scoliosis provoked by pain 514). Pain provoked scoliosis is not a common condition. Only several isolated cases have been published (1, 4, 7, 10, 11, 13, 14, 18). The largest of these series includes 15 patients (13, 18).

Indeed, osteoid osteoma and osteoblastoma of the spine are rare. Only a few studies except for some isolated ones have been reported to date (2, 7, 8, 11, 12, 13, 20).

The purpose of this study is to evaluate the clinical presentation and the results of surgical treatment of osteoid osteoma and osteoblastoma of the spine.

MATERIALS and METHODS

Twelve patients with osteoid osteoma or osteoblastoma of the spine treated surgically have been reviewed. There were eight cases of osteoid osteoma and four of osteoblastoma. Eight patients were boys and four girls, with an average age at the onset of symptoms of 12.8 years (range, 5 to 18 years). The average delay between the onset of symptoms and diagnosis was 23 months (range, 12 to 48 months). The average age at presentation was 14.5 years (range, 6 to 20 years). Seven ceses involved the lumbar spine, three the sacrum, one the thoracic spine, and one the cervical spine. The posterior elements were involved in all the cases. The three cases with sacral involvement were osteoblastomas. Prior to peresentation, two patients had been treated for lumbago, and four had been treated for herniated nucleus pulposus. In one of these, rectal examination was also done for a possible coccycodynia. The remaining six patients had not received any specific diagnoses. The clinical data are summarised in Table I and II.

Table 1. Osteoid osteoma.

Case	Age at onset of symptoms (years)	Interval to Diagnosis (months)	Sex	Side	Vertebra	Site
1	11.5	18	F	R	L2	Sup. facet
2	12	24	M	R	L2	Sup. facet
3	14	24	M	L	L1	Pedicle
4	5	12	F	L	C3	Sup. facet
5	10	12	F	L	L3	Inf. facet
6	10	24	M	L	L4	Pedicle
7	14.5	30	M	R	L1	Inf. facet
8	16	48	M	R	T11	Sup. facet

Table 2. Osteoblastoma.

Case	Age at onset of symptoms (years)	Interval to Diagnosis (months)	Sex	Side	Vertebra	Site
1	16	24	M	R	S1	Lamina
2	15.5	18	M	R	S1	Lamina
3	10.5	18	M	R	S1	Lamina
4	18	24	F	R	L5	Lamina

The presenting complaint was pain in all the patients. Pain was persistent in eight, intermittant in one patient. Three patients had pain only at night. Four of

Department of Orthopedics and Traumatology, Dokuz Eylül University Hospital, İzmir.

the patients with persistent pain stated that their pain became worse at night. Aspirin either taken prior to or after presentation provided good symptomatic relief in all patients. None of the patients showed evidence of abnormal neurological function on examination.

Mild to modirate spinal stiffness due to paravertebral muscle spasm was noted in 9 patients (75 per cent). All patients with osteoid osteoma having thoracic or lumbar involvement had scoliosis at the time of presentation. In all cases the lesion was located on the concave side of the curve which was average 14.3° (range, lo to 22°. One case involving the cervical spine was associated with a torticollis. The only deformity in patients with osteoblastoma was a slight pelvic obliquity in one patient (L5 involvement).

In 7 patients the presence and site of the tumor was determined by plain radiographs and technetium bone scans. Computerised axial tomography was used in addition in five patients to confirm the exact site and extent of the lesion.

The treatment consisted of surgical removal of the lesions. Stabilization was not necessary. The followup evaluation consisted of physical and radiographic examination.

RESULTS

The average followup was 3.9 years with a range from six months to 10 years. All patient noted dramatic relief of their pain within several days after the operation. At followup all the patients were free of pain. All patients except one had full spinal mobility. Forward flexion was limited moderately in the patient with osteoblastoma in L5. All patients stuted that they had returned to their normal daily activities.

At followup, the scoliosis had completely resolved in all patients. No recurrence was noted in plain radiographs.

DISCUSSION

This study demonstrates that osteoid osteoma of the thoracolumbar spine should be suspected in a patient with painful scoliosis and spinal stiffness. In the present study, seven patients with thoracolumbar involvement all had painful scoliosis. A painful torticollis, as the case in this study, should draw one's attention to a possible osteoid osteoma of the cervical spine. Scoliosis has been demonstrated in more than 50% of the patients affected with benign ostecblastoma of the thoracolumbar spine or ribs (12). None of the cases of osteoblastoma in the present study had scoliosis. This may be dua to sacral localization in three cases. Furthermore, the number is small to draw any conclusions.

At routine clinical examination, these atypical and rare causes of scoliosis are easily neglected. As reported in other studies (7, 8, 11), there was a significant delay in this study was 23 months. This delay was reported to be 13.7 months and 19 months in Keim and Reina's (7) and in Kirwan et al.'s (8) studies, respectively. Six of our patients (50%) had previous misdiagnoses, and the other 50 per cent had been treated without a spesific diagnosis. In Keim and Reina's study (7) eight of nine osteoid osteoma cases had previous misdiagnoses. 72 per cent of the patients reported by Kirwan et al. (8) had consulted three or more specialists. These figures suggest that the diagnosis of spinal osteoid osteoma or osteoblastoma is frequently not suspected.

The difficulty in making the diagnosis, especially of osteoid osteoma, could be due to the fact that it may produce pain before it is roentgenographically detectable (4, 17). Anothen reason is that the nidus is difficult to demonstrate on plain roentgenograms (5, 16, 21, 22). Spinal osteoblastoma may also be difficult to see on plain radiographs (5), but in contrast to osteoid osteoma radiographs are usually sufficient to confirm the diagnosis. Because of the difficulties with plain radiographs, radionuclide scans and computerised tomography have become the most useful and reliable tools for making the diagnosis (5, 8, 16, 26). Bone scans are positive and especially useful in recognizing the presence of the lesion and thus directing radiographic investigation. An important clue may be the fact that almost all the lesions are located at or around the apex of the curve and on the concave side as seen in our cases (1, 8, 14). Computerised tomography allows precise localisation of the nidus and distinguishes the center of the lesion from the surrounding solerosis and thus, is most helpful in directing the surgical treatment (5, 8, 16). as a result, it allows less radical and more accurate surgical excision. Radionuclide scanning in addition to plain radiographs was used in all of our cases. Computerised tomography was needed in five cases (four osteoid osteoma, one osteoblastoma7? More recently we have used magnetic resonance imaging in some of our cases not included in this study. Obviously, it has a definite place in the diagnosis and surgical treatment of these lesions.

None of our patients had radicular pain, but preoperative radiculograms are recommended in cases with

objective neurologic signs. Despite radicular pain, the radiculograms may be normal with no evidence of nerve root compression at operation (7, 8, 11), and in this situation there is no indication to explore the vertebral canal (8).

Osteoid osteoma and osteoblastoma in the spine have a predilection for the posterior elements (1, 5, 7, 8, 11, 18); the laminae, pedicles, transverse and spinous processes are most frequently involved and the vertebral body spared. In other words, the majority of these tumors occur in the region of the base of the transverse process (8). The sites of involvement were the same in this study. The authors believe that sacral involvement in three of four cases of osteoblastoma has to be emphasized, because in a review done by Dias and Prost (2), only five cases of sacral osteoblastoma out of sixtyfour were reported between 1935 and 1970.

The natural history of osteoid osteoma remains controversial. Spontaneous resolution of pain and healing of the lesions have been reported 515, 25)., although histologic confirmation of the diagnosis has never been provided in such reports. Sabanas et al (19). reported that the natural history of spinal osteoid osteoma may be one of spontaneous remission in some cases. Despite this fact, a lesion should be remeved when it is diagnosed (5, 7, 8). Local removal of reactive bone without removal of the nidus may decompress the lesion and relieve symptoms in some cases (24). However, complete extirpation of the nidus is the most predictable way to cure osteoid osteoma and should be the goal of surgical intervention. As many as 28% recurrence rates have been reported (23). Intralesional resection or curettage has the highest, and en bloc resection the lowest recurrence rate. Complete relief of symptoms in all patients indicates that we could achieve complete excision of the lesions. an important reason for selecting surgical treatment is the fact that the scoliosis, at first considered secondary to the paravertebral muscle spasm can become structural while waiting for possible and unpredictable spontaneous remission. This is especially important in growing children. In the growing child, the age at onset of symptoms, the duration of sympoms, the severity of the scoliosis at the time of operation and the time interval to skeletal maturity have a direct bearing on whether the scoliosis will resolve after removal of the painful stimulus. On the other hand, those patients presenting around skeletal maturity will undergo complete resolution of the scoliosis (7, 14, 18). In the present study there were no persistent scolioses. a possible explanation may be that the preoperative curves were less than 20° except for one case (22°). It should be noted that in the aforementioned studies, the preoperative curves which persisted or increased were more than 20° to 30°.

Marginal resection is curative for benign osteoblastoma (3). Incisional procedures such as curettage are associated with a high recurrence rate (6), but Marsh et al. (12) reported a high rate of cure, even if curettage was incomplete due to the location of the tumor. ırradiation following marginal resection was not used in any of our patiens, as there are few data in the literature to document its effect on this tumor. Radiation therapy is rarely indicated for treatment of benign osteoblastoma (5). Although the patient population is small (n:4), no recurrences indicated that the lesions were completely resected.

Intraoperative imaging by radiographs or radionuclide scanning have been recommonded for precise localization of osteoid osteoma and osteoblastoma during surgery (5). This was not necessary in any of our patients. Computerized tomography and magnetic resonance imaging obviates the use of intraoperative imaging in most cases.

Spinal fusion was not performed in our cases. Fusion should be performed only if the spine is rendered unstable by the surgery (1, 5, 7). The presence of structural changes in the spine is another indication for coorection and stabilization (1).

An underlying osteoid osteoma or osteoblastoma of the spine must be strongly suspected in a patient presenting with marked spinal stiffness and a painfl scoliosis in the second decade of life. Plain radiographs of the painful area should be made and especially the vertebra at the apex of the curve should be studied carefully. The use of technetium bone scanning and computerized axial tomography ensure accurate diagnosis and precise localization of the lesion. Patients respond very well when the lesion is excised completely.

REFERENCES

1. Akbarnia, B.A., Rooholamini, S. A.: Scoliosis caused by benign osteoblastoma of the thoracic or lumbar spine. J.Bone Joint Surg. 63-A: 1146-1115, 1981.
2. Dias, L.S., Frost, H.M.: Osteoblastoma of the spine. A review and report of eight new cases. Clin. Orthop. 91: 141-151, 1973.
3. Enneking, W. F.: Musculoskeletel Tumor Surgery. New York, Churchill Livingstone, pp: 1035-1042, 1983.

4. Freiberger, R.H.: Osteoid osteoma of the spine: a cause of backache and scoliosis in children and youğn adults. Radiology, 75: 232, 1960.
5. Healey, J. H., Ghelman, B.: Osteoid osteoma and osteoblastoma. Current concepts and recent advances. Clin. Orthop. 204: 76-85, 1986.
6. Jackson, R.P.: Recurrent osteoblastoma: A review. Clin. Orthop. 131: 229, 1978.
7. Keim, H.A., Reina, E.G.: Osteoid osteoma as a cause of scoliosis. J.Bone Joint Surg. 57-A: 159, 1975.
8. Kirwan, E. O'G., Hutton, P.A.N., Pozo, J.L., Ransford, A.O.: Osteoid osteoma and benign osteoblastoma of the spine. J.Bone Joint Surg. 66-B: 21-26, 1984.
9. Lichtenstein, L., Sawyer, W.R.: Benign osteoblastoma. Further observations and report of twenty additional cases. J.Bone Joint Surg. 46-A: 755-765, 1964.
10. Lindholm, T.S., Snellman, O., Osterman, K.: Scoliosis caused by benign osteoblastoma of the spine. A report of three patients. Spine, 2: 276-281, 1977.
11. MacLellan, D.I., Wilson, F.C., Jr.: Osteoid osteoma of the spine. A review of the literature and report of six new cases. J. Bone Joint Surg. 49-A: 111-121, 1967.
12. Marsh, B.W., Bonfiglic, M., Brady, L.P., Enneking, W.F.: Benign osteoblastoma: Range of manifestations. J. Bone Joint Surg. 57-A: 1-9, 1975.
13. Mehta, M.H., Murray, R.O.: Scoliosis provoked by painful vertebral lesions. Skeletal Radiol. 1: 223, 1977.
14. Mehta, M.H.: Pain provoked scoliosis. Observations on the evolution of the deformity. Clin. Orthop. 135: 58-85, 1978.
15. Mobey, E.: The natural course of osteoid osteoma. J. Bone Joint Surg. 33-A:166, 1951.
16. Nelson, O.A., Greer, R.B.: Localization of osteoid osteoma of the spine using computerized tomography. J.Bone Joint Surg. 65-A: 263-265, 1983.
17. Prabhakar, B., Reddy, D.R., Dayananda, B., Rao, G.R.: Osteoid osteoma of the skull. J.Bone Joint Surg. 54-B: 146-148, 1972.
18. Ransford, A.O., Pozo, J.L., Hutton, P.A.N., Kirwan, E.O'G.: The behaviour pattern of the scoliosis asscoiated with osteoid osteoma of osteoblastoma of the spine. J. Bone Joint Surg. 66-B: 16-20, 1984.
19. Sabanas, A.O., Bickel, W.H., Moe, J.H.: Natural history of osteoid osteoma of the spine. Review of the literature and report of three cases. Am. J. Surg. 91:880-889, 1956.
20. Schajowicz, F., Lemos, C.: Osteoid osteoma and osteoblastoma. Acta Orthop. Scand. 41: 272, 1970.
21. Sherman, M.S.: Osteoid osteoma associated with changes in adjacent joint. Report of two cases. J. Bone Joint Surg. 29: 483-490, 1947.
22. Sherman, M.S.: Osteoid osteoma. Review of the literature and report of thirty cases. J.Bone Joint Surg. 29: 918-930, 1947.
23. Shulman, L., Dorfman, H.D.: Nerve fibers in osteoid osteoma. J.Bone Joint Surg. 52-A: 135, 1970.
24. Sim, F.H., Dahlin, D.C., Beabout, J.W.: Osteoid osteoma.
Diagnostic problams. J. Bone Joint Surg. 57-A: 154-159, 1975.
25. Vickers, C.W., Pugh, P.C., Ivins, J.C.: Osteoid osteoma. A 15 year followup of an untreated patient. J.Bone Joint Surg. 41-A: 357, 1959.
26. Winter, P.F., Johnson, P.M., Hilal, S.K., Feldman, F.: Scintigraphic detection of osteoid osteoma. Radiology. 122: 177-178, 1977.