

Surgical treatment of osteoblastoma : A report of 20 cases

Yener Saglik, Hakan Atalar, Yusuf Yildiz, Kerem Basarir, Cuneyd Gunay

From the Ankara University School of Medicine, Ankara, Turkey

Osteoblastoma is a locally aggressive osteoblastic lesion of bone with rare malignant transformation. We retrospectively evaluated 20 patients who were diagnosed and surgically treated for osteoblastoma in our institution. Their mean age was 22.5 years (range : 10 to 34). The most frequent location was in the posterior elements of the spine, followed by the long bones of the extremities and the talus. Intralesional curettage was the most common modality of treatment; it was used in 11 patients, with a 13% recurrence rate. This series demonstrates that osteoblastoma may be locally aggressive and may recur after removal. Although it is usually treated successfully with curettage, wide excision should be considered along with careful follow-up over the long term owing to the possibility of recurrence or malignant transformation.

Keywords : osteoblastoma ; benign bone tumours ; differential diagnosis ; surgical treatment.

INTRODUCTION

Osteoblastoma and osteoid osteoma are wellknown benign osteoblastic lesions of bone (16). They have similar histological features and probably correspond to different expressions of one pathologic process (5,12). They have come to be distinguished in terms of size, site and degree of ossification (1,4,25). Several authors define the lesion as osteoblastoma when its longest axis is longer than 1.5 cm (20,24). The term "benign osteoblastoma" has also been used for this tumour ; it was proposed independently by Jaffe and by Lichtenstein to define a vascular osteoid and bone-forming tumour containing numerous osteoblasts which occurred principally in the vertebral column (12). Osteoblastoma is a rare tumour which accounts for approximately 3.5% of all benign neoplasms of bone and for less than 1% of all bone tumours. The average age of incidence is between 15 and 20 years, and most patients are diagnosed before the age of 30 (8,12,25). Although osteoblastoma tends to occur in the axial skeleton, particularly in the posterior elements of the vertebral column, it has been reported in a variety of skeletal locations including the scapula, ulna and scaphoid (23,26,27).

Osteoblastomas have a tendency to increase in size, and their radiological appearance is variable (3,21). Most osteoblastomas are well contained,

Department of Orthopaedic Surgery and Traumatology, Fatih University, Ankara, Turkey.

■ Kerem Basarir, MD, Consultant.

Department of Orthopaedic Surgery and Traumatology, Hakkari State Hospital, Hakkari, Turkey.

Correspondence : Hakan Atalar, Alparslan Turkes Caddesi No : 57 06510 Emek / Ankara, Turkey.

E-mail: atalarhakan@yahoo.com

No benefits or funds were received in support of this study

[■] Yener Saglik, MD, Professor.

[■] Yusuf Yildiz, MD, Associate Professor.

Department of Orthopaedic Surgery and Traumatology, Ankara University, Ankara, Turkey.

[■] Hakan Atalar, MD, Assistant Professor.

[■] Cuneyd Gunay, MD, Resident.

^{© 2007,} Acta Orthopædica Belgica.

but occasionally they may extend into the soft tissue (5). The development of imaging techniques has increased the physician's ability to detect soft tissue expansion in osteoblastoma (21). In the differential diagnosis, the most important condition to be ruled out is osteoblastoma-like osteosarcoma (2,9). A recent study found immunohistochemical analysis of cyclooxygenase-2 to be useful in the differential diagnosis of osteoblastoma and osteosarcoma, because this enzyme was more reliably expressed in osteoblastoma cells (10).

The curative treatment for patients with osteoblastoma is surgery. The lesion can be removed with intralesional curettage or wide resection, depending on the clinical situation, location within the bone and suspicion of malignancy (8,21). Recurrence rates vary with the surgical approach, and wide resection is associated with lower recurrence rates (7,11,14). The purpose of this retrospective study was to analyse the clinical findings, radiological findings, and treatment results in 20 patients with osteoblastoma.

PATIENTS AND METHODS

During the period from March 1986 to December 2006, 22 patients at the authors' institutions were diagnosed with osteoblastoma. Of these patients, one did not have surgery, 21 underwent surgery and one was lost to follow-up after surgery, leaving 20 patients for analysis. Diagnoses were confirmed with histological examination of the excised lesion in all patients. Data obtained for each patient included age, gender, symptoms at presentation, skeletal location of the tumour, radiological features, diagnosis and treatment. The characteristics of the patients are summarised in table I.

Plain radiographs were available for all patients. In 12 patients, computerised tomography (CT) and in 7 patients magnetic resonance imaging (MRI) was done as part of the diagnostic workup. The average duration of follow-up was 50.6 months (range : 20 to 192). One patient (Patient 9) had undergone intralesional curettage before being referred to our institution, and presented recurrence.

Intralesional curettage followed by bone grafting of the cavity was the most common surgical procedure. If the stability was not acceptable, such as in the removal of multiple posterior elements in the spine or in the extensive resection of a long bone, fixation with an implant was performed. For example, in Patient 13, after wide excision of the tumour, the distal fibula was reconstructed with a nonvascularised autograft from the patient's contralateral fibula, which was stabilised with a plate and screws (fig 1).

RESULTS

Of the 20 patients treated surgically for osteoblastoma who continued in follow-up, seven were female and 13 were male, with an average age of 22.5 years (range : 10 to 34). The spine was the most common location followed by the femur and talus. The most common presenting symptom was pain, present in all patients, followed by a tender mass in four patients. Pain was radicular in nature in two of six patients with vertebral involvement. Pathological fracture and paraesthesia were presenting complaints in one patient each. The mean period of follow-up was 50.6 months (range : 20 to 192).

Intralesional curettage was the preferred mode of treatment, used in 11 patients, and wide resection was used in three patients. Six patients with involvement of posterior elements of the spine underwent marginal excision. In the curettage group, the cavity was filled with bone graft in seven patients (five autograft, two allograft), with cement in one patient and with bone graft plus cement in one patient. Simple curettage without bone graft or cement was used in two patients (table I).

Recurrence was observed in three patients (table I). A 29-year-old male who underwent curettage at another center was referred to our institution within the first year after surgery and was treated with intralesional curettage and autogenous bone grafting (Patient 9, table I). In the second patient (Patient 1), recurrence was observed 10 months after facet excision for L2 posterior element involvement. Surgery for the recurrent tumour was not accepted by the patient and she was thereafter lost to follow-up. The third patient with a recurrent lesion was a 30-year-old female who had originally presented with pathological fracture of the humerus (Patient 16). Curettage with bone autograft was performed for the initial lesion, and 12 months later non-union and recurrence of the



Fig. 1. — Patient 13. (a) This preoperative plain radiograph shows an expansile and lytic lesion at the distal fibula. (b) Preoperative CT shows destruction of the cortex and ossification in the lesion. (c) After wide excision of the osteoblastoma, the distal fibula was reconstructed with a nonvascularised autograft from the contralateral proximal fibula. The graft was fixed to the tibia with a screw, which was removed two months later. (d) This follow-up plain radiograph taken 3 years after surgery shows complete fusion of the graft and fibula.



Acta Orthopædica Belgica, Vol. 73 - 6 - 2007

Follow-up	26	45	22	32	41	63	20	26	45	30	67	54	192	28	33	156		44	29	43	36	-
Complication									Myositis ossificans			Transient peroneal nerve palsy				Pseudoarthrosis &	limitation of range of motion					
Recurrence/treatment	No surgery								Curettage, autograft							Excision, autograft						
Reconstruction	Posterior instrumentation	Posterior instrumentation	Posterior instrumentation	ı	Posterior instrumentation	1	Autograft-cement	Autograft	Autograft	Autograft	Cement	1	Fibular autograft		Allograft	Autograft		Allograft	1	Autograft	Autograft	
Primary treatment	Facetectomy	Laminectomy	Laminectomy	Laminectomy	Laminectomy	Laminectomy	Curettage	Curettage	Curettage	Curettage	Curettage	Resection	Resection	Curettage	Curettage	Curettage		Curettage	Curettage	Curettage	Resection	
Location	L2	L2	L5	T11	L3	C5	Acetabulum	Acetabulum	Femoral neck	Femoral neck	Distal femur	Fibula	Distal fibula	Tibia	Proximal tibia	Humerus		Talus	Talus	Talus	Metatarsal	
Radiology	CT	CT	CT	CT	MRI	CT	CT, MRI	CT, MRI	CT, MRI			CT, MRI	CT		CT				CT, MRI		MRI	
Complaint	Pain	Pain, paraesthesia	Pain	Pain	Pain	Pain	Pain	Pain	Pain	Pain	Pain	Pain, mass	Pain	Pain, mass	Pain	Pain,	pathologic fracture	Pain	Pain, mass	Pain	Pain, mass	omography
Sex	ц	ц	X	М	Σ	ц	ц	М	М	н	М	X	М	н	М	ц		Μ	Σ	М	М	rized t
Age	24	34	24	16	29	17	15	20	29	25	18	16	20	22	22	30		10	33	27	22	ompute
No	-	5	3	4	5	9	7	8	6	10	11	12	13	14	15	16		17	18	19	20	CT : c

Table I. — Demographic and clinical details of 20 patients with osteoblastoma

Acta Orthopædica Belgica, Vol. 73 - 6 - 2007

tumour were observed. The patient then underwent repeat surgery with wide excision and structural autogenous fibular grafting with internal fixation. She was followed up for a further 10 years with no sign of recurrence. The overall complication rate was 15% (three patients, table I). The complications were limitation in the range of motion (Patient 16, passive flexion and abduction at the shoulder joint limited to 80°); transient superficial peroneal nerve palsy (Patient 12, following proximal fibular resection, resolved in 6 months) and myositis ossificans (Patient 9, gluteal region, after treatment for recurrent lesion). The overall recurrence rate in our series was 10% for patients whose initial surgery was performed at our institution.

DISCUSSION

The cases of osteoblastoma in this series account for 1.7% of the benign bone tumours diagnosed in our department during the same period, which is slightly lower than the 3-4% of all benign bone tumours reported in the literature (8,13). The female-to-male ratio in our series (7 to 13) is consistent with previous findings, in which osteoblastoma has occurred roughly twice as frequently among males (8). Similar to previous studies, the average age was around 20 years (22.5 years in our patients) and the majority of patients (87% in our study) were diagnosed before age 30 (21,24,25).

Osteoblastomas generally grow slowly with few symptoms, but aggressive lesions may cause severe symptoms such as soft tissue oedema, joint stiffness and contracture (5). Pain is the most common presenting symptom, as was the case in our series. Spinal involvement may cause deformity (21). Lesions may also be discovered incidentally (7,21,25). Spinal deformity or incidental lesions were not encountered in our series.

Osteoblastoma tends to occur in the axial skeleton, particularly in the posterior elements, with lesions there accounting for 40% of the total, followed by those in the long bones of the lower extremity, which account for 20% (16). The proportions in our series, which were 6/20 for spinal lesions and 8/20 for lesions in long bones of the lower extremity, are in agreement with the literature (4,12,25). Other locations in our series such as the talus and metatarsus have also been reported in other studies (5,25).

Imaging techniques generally show osteoblastomas as being osteolytic tumours ; however, due to osteoid and bone components, osseous type mineralisation may be seen (5). Several authors have argued that osteoblastoma has no characteristic radiographic appearance, but a number of radiographic features have been found helpful in making the diagnosis (6,11,25). The lesions are usually well circumscribed, central to slightly eccentric in long bones, rounded or ovoid, and expansile (7,14). A shell of reactive bone separating the tumour from the underlying bone has been reported to occur in nearly half of the patients (16).

CT and MRI are often useful to show the extent of the tumour and its relation to local anatomy, particularly for tumours in the spine, where routine radiographs may be difficult to interpret. In an imaging study of osteoblastoma, Youssef et al (28) concluded that while CT is more useful in diagnosing osteoid osteomas, MRI can be reserved for equivocal cases. We obtained CT and/or MRI in more than two-thirds of our patients. MRI showed soft tissue and surrounding bone oedema in all of our patients and provided valuable information about the neighbouring anatomical structures, and thereby helped in the planning for effective tumour removal. However, since no MRI features characteristic of osteoblastoma have been described, CT demonstrating the osteoblastic nature of the lesion is still more commonly preferred, as was the case in our series (28).

In the differential diagnosis of osteoblastoma, the following should be considered : osteoid osteoma, osteosarcoma, Brodie's abscess, aneurysmal bone cyst and giant cell tumour (5). Osteoblastoma lesions are by definition larger than those of osteoid osteoma, and a central nidus is usually not present in osteoblastoma (19). The bone formation is usually of a benign type and of a solid type in contrast to the aggressive periosteal reaction seen in osteosarcoma. The reactive bone surrounding osteoblastoma has been reported to be less when the lesion is in the cancellous bone of the spine, ilium or talus compared to lesions in the cortex of bones such as the metacarpals or the femur (16). Brodie's abscess resembles osteoblastoma, but in the former, bonetype radiodensities are not visible within the lesion (5). For aneurysmal bone cyst and osteoblastoma, the spine is a common site of involvement, and a blown-out bone may bee seen on imaging in both conditions (5). Histopathological investigation is then needed for the diagnosis. Around the epiphyses of long bones, osteoblastoma resembles giant cell tumour radiologically, but in contrast to giant cell tumour, peritumoral sclerosis is generally seen in osteoblastoma (5). A small percentage (<1%) of osteoblastomas have been reported to undergo malignant transformation (18,22). Consistent with this, no malignant transformations or metastases to the lungs were encountered in our series.

The basic microscopic pattern in osteoblastoma is one of numerous osteoblasts producing osteoid and woven bone, and is virtually the same as in osteoid osteoma (20,21). Osteoblastoma is distinguished by its aggressive characteristics, its extension into soft tissue and its size (3). A longest-axis length of at least 1.5 cm is accepted by some authors as part of the definition of osteoblastoma (17,25). Histologically, it is not always easy to distinguish osteoblastoma from rare lesions such as osteoblastoma-like osteosarcoma. In this context it has been reported that expression of cyclooxygenase 2 is more prominent in osteoblastoma cells (10). For the diagnosis of osteoblastoma in our department, histological examination combined with clinical and radiological features, including a criterion of 1.5 cm or more for the length of the tumour's longest axis are used, and these are the basis of the study reported here.

Most benign tumours of bone can be successfully treated with intralesional curettage alone. Use of a burr can aid in extending the curettage, particularly in areas that are difficult to access. We prefer to use cautery in difficult-to-reach areas, because the generated heat may help to destroy tumour cells. Curettage can be followed by bone grafting or bone cementing. In aggressive lesions marginal or wide resection is necessary (5). Radiotherapy should be used only when complete removal of the tumour is impossible and the tumour continues to grow, with the risks of radiotherapy, including postradiation sarcoma, being kept in mind (15,21,22).

Recurrence rates of up to 21% following firsttime surgical treatment of osteoblastoma have been reported (7,11,14). Wide resection has lower recurrence rates when compared with intralesional procedures (7,11,14). In our series, there were no recurrences in the patients who were treated with wide resection of the tumour. Recurrence was observed in one of six patients who underwent marginal excision, and in two of 11 patients who underwent intralesional curettage. The overall recurrence rate in our series was 10%, which is comparable to other studies (7,11,14).

In conclusion, in the treatment of patients with osteoblastoma, wide resection should be considered whenever it is anatomically possible. However, although intralesional curettage appears to be associated with higher rates of tumour recurrence, it can also be effective. Postoperatively, patients require long term follow-up due to the possibility of tumour recurrence and malignant transformation.

REFERENCES

- 1. Bettelli G, Capanna R, van Horn JR, Ruggieri P, Biagini R, Campanacci M. Osteoid osteoma and osteoblastoma of the pelvis. Clin Orthop 1989; 247: 261-271.
- 2. Bonar SF, McCarthy S, Stalley P et al. Epiphyseal osteoblastoma-like osteosarcoma. Skeletal Radiol 2004; 1:46-50
- 3. Boriani S, Capanna R, Donati D, Levine A, Picci P, Savini R. Osteoblastoma of the spine. Clin Orthop 1992; 278:37-45.
- 4. Byers PD. Solitary benign osteoblastic lesions of bone. Osteoid osteoma and benign osteoblastoma. Cancer 1968 ; 22:43-57.
- 5. Campanacci M. Osteoblastoma. In: Campanacci M, Enneking WF, editors. Bone and Soft Tissue Tumours. 2nd edition. Springer, New York, 1999 ; pp 415-433.
- 6. Dahlin DC, Johnson EW. Giant osteoid osteoma. J. Bone Joint Surg 1954 ; 36-A : 559-572.
- 7. Frassica FJ, Waltrip RL, Sponseller PD, Ma LD, McCarthy EF Jr. Clinicopathologic features and treatment of osteoid osteoma and osteoblastoma in children and adolescents. Orthop Clin North Am 1996; 27: 559-574.
- 8. Golant A, Lou JE, Erol B, Gaynor JW, Low DW, Dormans JP. Pediatric osteoblastoma of the sternum : a new surgical technique for reconstruction after removal :

case report and review of the literature. *J Pediatr Orthop* 2004; 3: 319-322.

- **9. Hermann G, Klein MJ, Springfield D, Abdelwahab IF.** Osteoblastoma like osteosarcoma. *Clin Radiol* 2004 ; 1 : 105-108.
- Hosono A, Yamaguchi U, Makimoto A *et al.* Utility of immunohistochemical analysis for cyclo-oxygenase 2 in the differential diagnosis of osteoblastoma and osteosarcoma. *J Clin Pathol* 2007; 60: 410-414.
- **11. Jackson RP.** Recurrent osteoblastoma : a review. *Clin Orthop* 1978 ; 131 : 229-233.
- **12. Lichtenstein L.** Benign osteoblastoma : category of osteoid- and bone-forming tumours other than classical osteoid osteoma which may be mistaken for giant-cell tumour or osteogenic sarcoma. *Cancer* 1956; 9: 1044-1052.
- **13. Lichtenstein L, Sawyer WR.** Benign osteoblastoma : further observations and report of twenty additional cases. *J Bone Joint Surg* 1964 ; 46-A : 755–765.
- 14. Lucas DR, Unni KK, McLeod RA, O'Connor MI, Sim FH. Osteoblastoma : clinicopathologic study of 306 cases. *Hum Pathol* 1994 ; 25 : 117-134.
- 15. Mark RJ, Poen J, Tran LM, Fu YS, Selch MT, Parker RG. Postirradiation sarcomas : A single-institution study and review of the literature. *Cancer* 1994; 73 : 2653-2662.
- **16. Marsh BW, Bonfiglio M, Brady LP, Enneking WF.** Benign osteoblastoma : range of manifestations. *J Bone Joint Surg* 1975 ; 57-A : 1-9.
- 17. McLeod RA, Dahlin DC, Beabout JW. The spectrum of osteoblastoma. *Am J Roentgenol* 1976 ; 126 : 321-325.
- **18. Merryweather R, Middlemiss JH, Sanerkin NG.** Malignant transformation of osteoblastoma. *J Bone Joint Surg* 1980 ; 62-B : 381-384.

- **19. Miller SL, Hoffer FA.** Malignant and benign bone tumours. *Radiol Clin North Am* 2001; 39: 673-699.
- 20. Nemoto O, Moser RP Jr, Van Dam BE, Aoki J, Gilkey FW. Osteoblastoma of the spine : A review of 75 cases. Spine 1990 ; 15 : 1272-1280.
- **21. Ozaki T, Liljenqvist U, Hillmann A** *et al.* Osteoid osteoma and osteoblastoma of the spine : experiences with 22 patients. *Clin Orthop* 2002 ; 397 : 394-402.
- 22. Seki T, Fukuda H, Ishii Y, Hanaoka H, Yatabe S. Malignant transformation of benign osteoblastoma : A case report. *J Bone Joint Surg* 1975 ; 57-A : 424-426.
- **23.** Theologis T, Ostlere S, Gibbons CL, Athanasou NA. Toxic osteoblastoma of the scapula. *Skeletal Radiol* 2007 ; 3 : 253-257.
- 24. Tonai M, Campbell CJ, Ahn GH, Schiller AL, Mankin HJ. Osteoblastoma : classification and report of 16 patients. *Clin Orthop* 1982 ; 167 : 222-235.
- 25. Unni KK. Benign Osteoblastoma. In: Dahlin's Bone Tumours. General Aspects and Data on 11,087 Cases. 5th ed. 1996. Lippincott-Raven, Philadelphia, pp 131-142.
- 26. Van Giffen N, De Smet L. Osteoblastoma of the proximal ulna, an unusual case of ulnar wrist pain : A case report. *Acta Orthop Belg* 2005 ; 71 : 736-739.
- 27. Xarchas KC, Leviet D. Osteoblastoma of the carpal scaphoid : Frequency and treatment. Acta Orthop Belg 2002; 68: 532-536.
- **28.** Youssef BA, Haddad MC, Zahrani A *et al.* Osteoid osteoma and osteoblastoma : MRI appearances and the significance of ring enhancement. *Eur Radiol* 1996; 3 : 291-296.