



Periosteal osteoblastoma of the distal femur

Ahmet KAYA, Taskin ALTAY, Murat SEZAK, Hasan OZTURK

From Izmir Tepecik Training and Research Hospital, Izmir, Turkey

Periosteal osteoblastoma is a rare benign osteoblastic bone tumour located on the cortical bone. We report a case of periosteal osteoblastoma located at the distal femur in a 31-year-old man. Clinical, radiological and histopathologic findings are described and differential diagnosis is discussed. Despite its rarity, periosteal osteoblastoma should be considered as a possible diagnosis together with periostitis ossificans, periosteal chondroma, periosteal osteosarcoma and parosteal osteosarcoma when confronted with a superficial bone lesion.

Keywords : periosteal osteoblastoma ; benign bone tumour ; osteoblastoma.

INTRODUCTION

Osteoblastoma is a rare bone tumour which represents approximately 1% of all primary bone tumours and 3% of all benign bone tumours (10). Its histopathology is similar to that of the nidus of osteoid osteoma, but it is larger in size (6). It is usually located in the medullary cavity of flat bones and long bones, and periosteal locations are rare (7). Lichtenstein and Sawyer first described the periosteal location of osteoblastoma in 1964 (3). Since that time approximately about 30 cases have been reported, but there are only a few studies about periosteal osteoblastoma with detailed clinical, radiological and histopathological descriptions (2,3,9).

We report a case of periosteal osteoblastoma located at the distal femur and review the literature

with respect to clinical, radiological and histopathological criteria ; we also discuss the differential diagnosis.

CASE REPORT

A 31-year-old man presented to our clinic with gradually increasing pain at his right lower posterior thigh over the past six months. Pain was moderate and constant and was not completely alleviated by rest and also not by non steroid anti-inflammatory drugs (NSAIDs). Heavy physical exercises increased the pain and could not be performed easily. There was no history of trauma, and no other medical problem. At physical examination, slight local tenderness was observed at the distal part of the posterior aspect of the affected thigh. Range of

■ Ahmet Kaya, MD, Associate Professor of Orthopaedics and Traumatology.

■ Taskin Altay, MD, Associate Professor of Orthopaedics and Traumatology.

■ Hasan Ozturk, MD, Deputy Chief of 1st Orthopaedics and Traumatology Clinic.

Department of Orthopaedics and Traumatology, Izmir Tepecik Training and Research Hospital, Izmir, Turkey.

■ Murat Sezak, MD, Assistant Professor of Pathology.

Department of Pathology, Ege University, Faculty of Medicine, Bornova – Izmir, Turkey.

Correspondence : Ahmet Kaya, 2023 Sok. Nu : 2/3 Bostanlı 35540 Izmir, Turkey. E-mail : drahmetkaya@yahoo.com

© 2009, Acta Orthopædica Belgica.

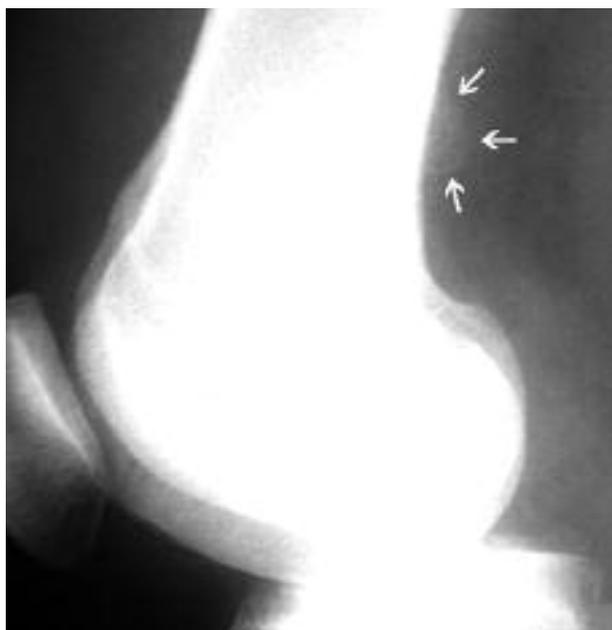


Fig. 1a. — Lateral radiographic view of the distal femur



Fig. 1b. — Anteroposterior radiographic view of the distal femur.

motion, especially flexion of the right knee, was somewhat limited. Nearly full extension could be achieved, with increased pain. There were no other abnormal findings on local or systemic examination. Laboratory tests were normal. Lateral radiographs of the thigh showed a 2.5×1.5 cm expansile, well defined, rounded lesion with amorphous density arising from the posterior aspect of the distal femur (fig 1a). The anteroposterior radiograph showed that the lesion was surrounded by osteosclerosis and new bone formation (fig 1b). Whole body technetium bone scan revealed increased uptake only at that site. There was a well-defined mass on the posterior aspect of the femur on CT: the lesion was located in the posterior cortex of the distal femur, and no cortical destruction or medullary involvement was detected (fig 2).

On MRI, the center of the lesion was isointense with adjacent muscles and was surrounded by a thin high-density area on the axial T1 image (fig 3a). Mixed low-density and isointense areas were observed on sagittal T1 images (fig 3b). T2 STIR images showed oedema in perilesional muscles and medullary bone (fig 3c). The lesion displayed high

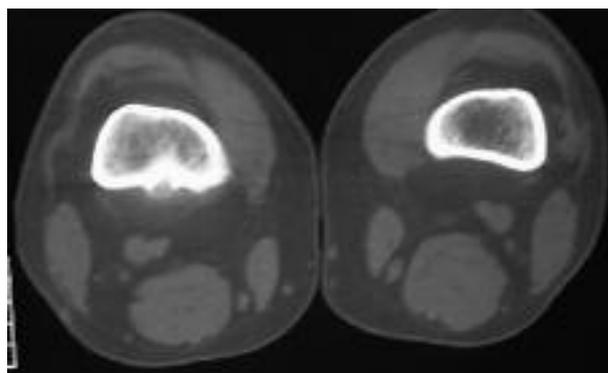


Fig. 2. — CAT-scan of both distal femora

signal intensity on postcontrast and T2 images (fig 3d, e).

As the clinical and imaging findings were compatible with a wide range of tumours especially parosteal osteosarcoma, we decided to perform an excisional biopsy. The lesion was removed en bloc with surrounding normal bone.



Fig. 3a. — Axial T1 image



Fig. 3b. — Coronal T1 image



Fig. 3c. — Coronal STIR T2 image

On histopathologic examination, the tumour was characterized by osteoid and woven bone surrounded by osteoblasts, and was well limited from the surrounding normal bone tissue. Osteoblast-like cells and dilated small capillaries were seen in the intertrabecular spaces. Based on these findings, a number of possible diagnoses were excluded, and the tumour was determined to be a periosteal osteoblastoma (fig 4a, b).

The patient was well and disease free at final examination 16 months postoperatively. There was no indication of recurrence on plain radiographs, CT-scan and MRI.

DISCUSSION

Osteoblastoma is an uncommon benign bone-producing tumour. It is generally seen in the second and third decades of life and it is the only benign



Fig. 3d. — Postcontrast coronal T2 image



Fig. 3e. — Postcontrast axial T2 image

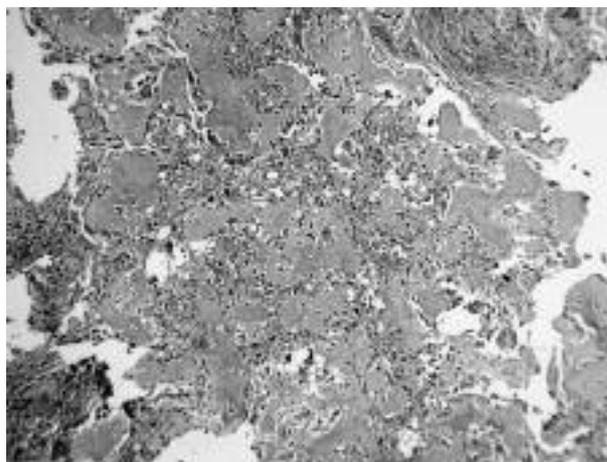


Fig. 4a. — In the central part of the lesion, irregular bony trabeculae are seen, rimmed with osteoblasts, osteoblast-like cells, with occasional osteoclasts and fibroblasts; small dilated capillaries are seen in intertrabecular spaces. Nuclear pleomorphism and atypical mitoses are not noted.

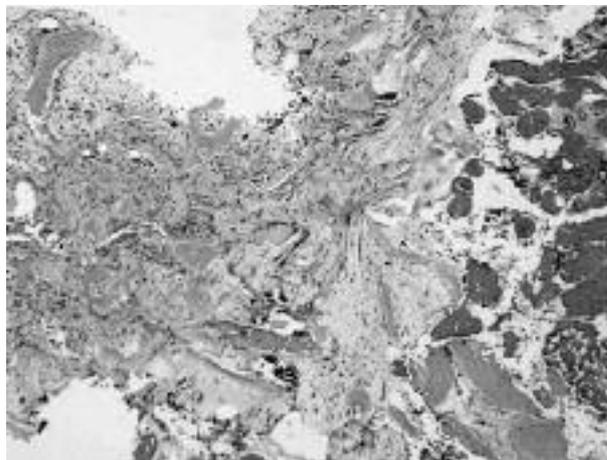


Fig. 4b. — In the peripheral part of the lesion, irregular bony trabeculae are rimmed with osteoblasts; the stromal cells show neither pleomorphic nuclei nor mitoses. The lesion is pushing away normal skeletal muscle.

tumour which has a marked predilection (over 40% of the cases) for the vertebral column, including the sacrum. Long bones come next, with approximately one third of the cases. In the long bones it is generally located in the metaphysodiaphyseal region, and the proximal femur is the most frequent site of involvement. Periosteal osteoblastoma occurs twice more frequently in male than in female patients.

The tumour may be central or eccentric or, very rarely, periosteal (1). Almost 30 cases have been reported as periosteal osteoblastomas to date (6,9). There are no pathognomonic clinical or radiological features that could allow for a positive diagnosis of periosteal osteoblastoma (4). Severe pain, worse at night, and relief from aspirin are not typically seen as with osteoid osteoma (8).

The ages of the patients with periosteal osteoblastomas reported in literature have ranged from 7 to 51 years, with a mean age about 20 years (6,9). Our patient was 31 years old. Radiological features of periosteal osteoblastoma are either a heavily mineralized mass or a radiolucent lesion with central calcification, on the surface of the diaphysis or metaphysis of a long bone. The lesion arises from the surface of the bone, with no evidence of cortical destruction, and is commonly associated with cortical thickening and periosteal new bone formation (4,6).

The lesions are well-circumscribed, radiolucent and usually expansive, and may display a thin shell of peripheral new bone. The intense perilesional sclerosis seen in osteoid osteoma is absent. Adjacent soft tissues may display mildly increased density, and there may be intralésional spotty mineralisation close to the cortex (7).

There are only a few studies about the NMR appearance of periosteal osteoblastoma. Kawaguchi *et al* reported that the lesion is isointense to muscle on T1-weighted images and displays high signal intensity on T2-weighted images (2), Sultzbacher *et al* reported a significant signal enhancement following contrast administration (9). Nakatani reported isointensity to muscle on T1-weighted images and a mixture of low and high signal intensity on T2-weighted images, with the surrounding soft tissues showing high signal intensity on T2-weighted STIR images, suggesting muscle oedema (7). In the present case we observed all the above mentioned findings.

Lesions in a periosteal location include a wide spectrum of tumours and pseudotumours, which causes difficulties in diagnosis (9). This spectrum includes myositis ossificans, osteochondroma, avulsive cortical irregularity syndrome, periosteal chondroma, periostitis ossificans, osteoid osteoma,

parosteal osteosarcoma, periosteal osteosarcoma, high grade surface osteosarcoma, periosteal chondrosarcoma. One fourth of the Mayo Clinic cases were considered as malignant tumours before histopathologic investigation (8).

Although histologically all lesions with cartilaginous tissue can be excluded, the differential diagnosis of periosteal osteoblastoma still may be difficult. Open biopsy was reported to be the procedure of choice to establish the diagnosis of osteoblastoma and in particular to distinguish it from other tumours that may demonstrate osteoblastic activity, especially osteosarcoma (8). Parosteal osteosarcoma is a usually low-grade malignant tumour that develops predominantly on the surface of long bones in an exophytic pattern. The lesion usually occurs in the third or fourth decade of life. More than 80% of parosteal osteosarcomas are located at the posterior aspect of the distal femur.

Roentgenographically, periosteal osteoblastoma is characterized by a heavily ossified lesion arising from the cortex, but no obvious signs of periosteal reaction are observed (7). In our case, the most probable diagnosis before the histopathologic examination was parosteal sarcoma. Parosteal sarcoma consists of a spindle-cell stroma with minimal cytological atypia and rare mitoses, associated with long trabeculae of osteoid and woven bone and it is also characterized by the absence of zone phenomena (7,9). The background stromal elements lack a prominent vascular network; giant cells are not present. In addition the tumour invades the adjacent soft tissues and underlying bone (3,5). Absence of cartilage is one of the histologic features that distinguish osteoblastoma from osteosarcoma. Rare instances of an otherwise typical histology of osteoblastoma associated with a cartilaginous component have however been reported. Although cartilage may be found in an osteoblastoma-like osteosarcoma, the cellular atypia and permeative growth pattern are features that support the presence of a malignant process. Differentiating a periosteal osteoblastoma from parosteal sarcoma is critical, since parosteal sarcoma requires a more aggressive treatment (5,6,10).

Although its histopathologic features are similar to those of osteoid osteoma, osteoblastoma is larger

in size, and radiologic criteria are different. It has a geographic contour, it is expansile and usually nonprogressive in appearance. It may present as a blastic, lytic or mixed density lesion (4). From a clinical viewpoint, night pain predominance is not clear as compared with osteoid osteoma. The pain is not relieved with aspirin or other NSAIDs.

CONCLUSION

Although less frequent than other benign and malignant tumours, periosteal osteoblastoma should be considered in the differential diagnosis when confronted with a proliferative periosteal lesion. The distinction between periosteal osteoblastoma and parosteal osteosarcoma is essential. Clinical, radiological and pathological findings should be evaluated altogether for diagnostic accuracy, a principle that also applies to any tumour.

REFERENCES

1. **Campanacci M.** *Bone and Soft Tissue Tumors*. 2nd ed. Piccin Nuova Libreria and Springer Verlag, Padova and New York, 1999.
2. **Kawaguchi K, Oda Y, Miura H et al.** Periosteal osteoblastoma of the distal humerus. *J Orthop Sci* 1998 ; 3 : 341-345.
3. **Lichtenstein L, Sawyer WR.** Benign osteoblastoma : further observations and report of twenty additional cases. *J Bone Joint Surg* 1964 ; 46-A : 755-765.
4. **Manaster BJ.** Musculoskeletal tumor imaging. In : Putman CE, Ravin CE (eds). *Textbook of Diagnostic Imaging*. 2nd ed. W.B. Saunders, Philadelphia, 1994.
5. **Mirra JM, Picci P, Gold RH.** *Bone Tumors. Clinical, Radiological and Pathologic Correlation*. 1st ed. Lea & Febiger, Philadelphia, 1989.
6. **Mortazavi SMJ, Wenger D, Asodallahi S et al.** Periosteal osteoblastoma : report of a case with a rare histopathologic presentation and review of the literature. *Skeletal Radiol* 2007 ; 36 : 259-264.
7. **Nakatani T, Yamamoto T, Akisue T et al.** Periosteal osteoblastoma of the distal femur. *Skeletal Radiol* 2004 ; 33 : 107-111.
8. **Simon MA.** Musculoskeletal tumors. In : Everts CMcC (ed). *Surgery of the Musculoskeletal System*. 2nd ed. Churchill Livingstone, New York, 1990.
9. **Sulzbacher I, Puig S, Trieb K, Lang S.** Periosteal osteoblastoma : a case report and a review of the literature. *Pathol Int* 2000 ; 50 : 667-671.
10. **Unni KK.** Benign osteoblastoma (giant osteoid osteoma). In : Unni KK. (ed). *Dahlin's Bone Tumors. General Aspects and Data on 11087 Cases*, 5th edition. Lippincott Raven, Philadelphia, 1996.