Osteoblastoma of the Second Metatarsal:
A Case Report

N. K. Sferopoulos1*

1Department of Pediatric Orthopaedics, G. Gennimatas Hospital, 54635, Thessaloniki, Greece.

Author's contribution
This whole work was carried out by author NKS.

ABSTRACT

Aims: To present the clinical, radiographic and histopathological features of an osteoblastoma localized to the second metatarsal of a 50-year-old woman that was successfully treated with intralesional curettage.

Case Presentation: A 50-year-old woman presented with localized swelling and tenderness of the right forefoot of 3 years’ duration. She reported increased swelling and intense pain, worse at night, during the last 6 months. Imaging included plain radiographs, computed tomography (CT) and magnetic resonance imaging (MRI) and was indicative of a benign process. An expansile osteoblastic lesion was identified in the diaphysis of the second metatarsal on x-rays. A lytic bone lesion surrounded by an intact cortical rim with evidence of intralesional ossifications was evident on both CT scan and MRI. In the latter, there were also signs of bone marrow and soft tissue edema. The lesion was treated surgically with curettage and an osteoblastoma was diagnosed at biopsy. No further treatment was undertaken. No recurrence has occurred during a 7-year follow-up.

Discussion: Osteoblastoma is a rare benign osteoblastic neoplasm with a scarce localization to the metatarsals. Diagnosis is not difficult when the typical clinical and imaging features are present. Surgical treatment may offer intralesional curettage or wide resection to reduce recurrence rates in locally aggressive tumors, recurrent lesions, or in cases with suspected malignancy. In the reported case the lesion was misdiagnosed as an area of osteomyelitis. Definitive diagnosis was based on curettage-excision of the lesion indicating the typical histological pattern of osteoblastoma.

*Corresponding author: Email: sferopoulos@in.gr;
Conclusion: The clinical and radiographic appearance of osteoblastoma may be variable and can mimic other tumors or tumorous conditions. Differential diagnosis may be especially challenging and histological analysis of the biopsy specimens may be the diagnostic cornerstone.

Keywords: Benign tumor; osteoblastoma; metatarsal; foot.

1. INTRODUCTION

Osteoblastoma is a benign primary osteoblastic bone tumor. It is also called “giant osteoid osteoma” since it is usually characterized by the same architectural configuration but its nidus has a much larger diameter [1]. The most common type of the tumor is the conventional benign type, but it may also exhibit aggressive or malignant characteristics [2,3]. The most common sites of the lesion are the spine, the long bones and the ankle [4-6]; while localization to the metatarsals is extremely rare [7-9]. The lesion may present with a typical clinical and radiographic appearance but may also exhibit variable and often nonspecific characteristics [10]. Histopathological investigation is the key towards making a definitive diagnosis, although other benign tumors can mimic it, while differentiation of aggressive osteoblastoma from osteosarcoma may be challenging [11,12].

A 50-year-old woman with an osteoblastoma of the second metatarsal of the right foot is presented and the clinical, radiological and histological findings are presented and discussed.

2. CASE PRESENTATION

A 50-year-old woman was referred with a 3-year history of pain and swelling in the region of metatarsals of the right foot. She reported progressive increase of pain and swelling during the last 6 months that significantly restricted her from shoe wearing. She experienced severe, permanent pain that was accentuated with mobilization and ambulation and showed nocturnal exacerbation. Her symptoms were not relieved with acetylsalicylates or paracetamol but subsided for a few days following nimesulide's use. Her past medical history was unremarkable.

Blood tests were normal with the exception of a white cell count of 11,700 cells/mm$^3$ (normal values 4300-10800) that was associated with a neutrophil count of 60% (normal values 48-73), an erythrocyte sedimentation rate of 34mm in the first hour (normal values 1-20) and a C-reactive protein value of 1.5mg/dl (normal values 0-0.5mg/dl).

On physical examination the mass was localized to the distal half of the second metatarsal and was firm, painful and warm on palpation, but with no erythema. Both active and passive mobility at the metatarsophalangeal joints was painful and restricted.

Plain radiographs of the foot showed an ostoblastic bone lesion localized to the distal portion of the diaphysis of the second metatarsal with a thinned and expanded cortex, but an intact cortical rim and an intramedullary border of bone sclerosis (Fig. 1).

Computed tomography and MRI revealed an intramedullary radiolucent central portion with several faint areas of internal ossification and a peripheral margin of sclerotic bone. There
was no evidence of periosteal reaction or cortical destruction (Figs. 2, 3a). Magnetic resonance imaging also demonstrated bone marrow edema of the second metatarsal and significant soft tissue edema around the second metatarsal (Fig. 3b). The most likely diagnosis as indicated from the clinical, laboratory and imaging findings was considered to be subacute osteomyelitis, although a diagnosis of osteoblastoma could not be excluded.

Fig. 1. Anteroposterior radiograph of the right foot in a 50-year old woman indicating an expanding, osteogenic tumor of the second metatarsal with benign features, such as circumscription and sclerotic margination.
Fig. 2. CT scan image demonstrated an expansile tumor with a central ossified nidus surrounded by a sclerotic rim

Surgical biopsy of the lesion was performed. Exposure was through a longitudinal incision along the diaphysis of the second metatarsal. The cortex over the lesion was fenestrated and curettage was performed (Fig. 4). The removed material consisted of fragments of red gritty tissue.
Fig. 3. MRI images showed an osteogenic lesion with a central nidus associated with both intraosseous (a) and surrounding soft tissue (b) edema.
Fig. 4. Intraoperative radiograph following removal of the nidus

Histological examination of the obtained specimens revealed an osteoblastic tumor, composed of anastomosing immature bone trabeculae, lined by a single layer of osteoblasts, and separated by a fibrovascular stroma (Fig. 5a). A central sclerotic nidus formed by dense sclerotic woven bone was evident (Fig. 5b). The margins of the host bone-tumor surface were sharp, and well circumscribed. There was no evidence of mitoses or atypical changes of the osteoblasts. The above histological features were consistent with osteoblastoma. The histological findings of the soft tissue envelope indicated fibrous tissue with few lymphocytes and skeletal muscle fibers.
Following surgery, the ankle was immobilized in a below-knee splint in neutral position for 2 weeks. The patient was allowed free weight-bearing after complete relief of pain at 6 weeks post-surgery.

Fig. 5. Microscopically, inter anastomosing trabeculae of woven bone lined by a single layer of osteoblasts were separated by a fibrovascular stroma (a), while the nidus was formed by dense sclerotic woven bone (b)
There were no complications or complaints during follow-up. She remained pain free and had no gait problems. Swelling slowly diminished and radiographs indicated considerable remodeling of the shaft of the second metatarsal over the following 7 years (Fig. 6).
3. DISCUSSION

Osteoblastoma is an uncommon benign osteoid tissue-forming tumor with clinical, radiographic and histological similarities to osteoid osteoma [1,13,14].

Compared with osteoid osteoma, osteoblastoma occurs in a slightly older population, pain is not worse at night and is less likely to be relieved with nonsteroidal anti-inflammatory drugs, usually involves the posterior elements of the vertebral column and exhibits an intramedullary localization when involving the long bones [15]. Radiographic appearance may indicate a well-defined, expansile, radiolucent, central ossified nidus associated with a surrounding reactive new bone formation. CT and bone scans are useful to demonstrate the osteoblastic nature of the lesion and to localize it. MRI may not present any specific characteristics but it is helpful to determine intraosseous or soft tissue extension of the tumor [13,16].

Differential diagnosis should include: osteoid osteoma, subacute osteomyelitis (Brodie’s abscess), aneurysmal bone cyst, giant cell tumor and osteosarcoma [16].

The basic histological pattern in osteoblastoma may be similar to osteoid osteoma. The absence of atypia and mitoses as well as the normal appearance of osteoblasts are important features to distinguish osteoblastoma from a low-grade osteosarcoma [11,12].

Surgery is the curative treatment for patients with osteoblastoma, but may depend on the tumor’s size, anatomic location and the ability to resect the entire lesion. Most lesions can be successfully treated with intralesional curettage. En bloc resection is necessary in recurrent and aggressive lesions, which appear with bone destruction and extension into soft tissues. Recurrences do seldom occur more than 2 years post-operatively [17-20].

The rarity of the presented case reflected not only to the involvement of the second metatarsal of a 50-year-old female but also to the 3-year-long history of clinical symptoms and local signs. Both clinical and laboratory findings led to a differential diagnosis dilemma between subacute osteomyelitis and osteoblastoma. Radiological features, although indicative of a benign bone lesion, most likely an osteoblastoma, misled to the diagnosis of osteomyelitis due to the intraosseous and soft tissue edema adjacent to the lesion that was evident on the MRI images. It has already been reported that bone tumors, such as osteoid osteoma and osteoblastoma, produce a matrix that may mineralize (central ossified nidus) and simulate on imaging a bone sequestrum [21]. A few distinguishing radiographic findings have been presented to differentiate subacute osteomyelitis and osteoblastoma, but they are not pathognomonic [22-24].

Bone marrow and soft tissue edema are observed with MRI following trauma, osteomyelitis and malignant bone tumors [25]. The detection of increased prostaglandin levels in aggressive or malignant tumor tissues offers a possible explanation of the MRI detected edema around some particular tumors [26]. Prostaglandins are important mediators of normal bone metabolism and it is reasonable that benign bone forming tumors, such as osteoid osteoma and osteoblastoma [27] may also synthesize a large amount of prostaglandins that may lead to a prominent peritumoral edema [28].

It seems, therefore, reasonable to consider that no specific surgical measures should be undertaken for the reactive soft tissue mass associated with an osteoblastoma.
Histology in the reported patient revealed typical features of a conventional osteoblastoma. Meticulous intralesional curettage was proved sufficient for the treatment of the osteoblastoma, since no recurrence of the lesion has appeared 7 years post-operatively.

4. CONCLUSION

Histological analysis of the biopsy specimens was the cornerstone of the diagnostic investigation in the reported patient. In addition, intralesional curettage was proved sufficient for the surgical treatment of the lesion.

CONSENT

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

ETHICAL APPROVAL

Not applicable.

COMPETING INTERESTS

Author has declared that no competing interests exist.

REFERENCES

1. Dahlin DC, Johnson EW Jr. Giant osteoid osteoma. J Bone Joint Surg Am. 1954;36(3):559-72.
2. Revell PA, Scholtz CL. Aggressive osteoblastoma. J Pathol. 1979;127(4):195-8.
3. Schajowicz F, Lemos C. Malignant osteoblastoma. J Bone Joint Surg Br. 1976;58(2):202-11.
4. Berry M, Mankin H, Gebhardt M, Rosenberg A, Hornier F. Osteoblastoma: A 30-year study of 99 cases. J Surg Oncol. 2008;98(3):179-83.
5. Temple HT, Mizel MS, Murphey MD, Sweet DE. Osteoblastoma of the foot and ankle. Foot Ankle Int. 1998;19(10):698-704.
6. Wu KK. Osteoblastoma of the foot. J Foot Surg. 1988;27(1):92-4.
7. Ellis BI, Shier CK, Gaba AR, Ohorodnik JM, Haggar AM. Case report 538. Osteoblastoma of the second metatarsal. Skeletal Radiol. 1989;18(3):228-32.
8. Hoeffel JC, Segal P, Abadou H, Adnet JJ. Osteoblastoma of the first metatarsal bone. Rofo. 1989;151(4):506-7.
9. Segal P, Hoeffel JC, Abadou H, Dehoux E, Adnet JJ, Herbent P. Osteoblastoma of the 1st metatarsal bone. Apropos of a case. J Radiol. 1987;68(8-9):533-5.
10. Ruggieri P, McLeod RA, Unni KK, Sim FH. Osteoblastoma. Orthopedics. 1996;19(7):621-4.
11. Lucas DR, Unni KK, McLeod RA, O'Connor MI, Sim FH. Osteoblastoma: Clinicopathologic study of 306 cases. Hum Pathol. 1994;25(2):117-34.
12. Della Rocca C, Huvos AG. Osteoblastoma: varied histological presentations with a benign clinical course. An analysis of 55 cases. Am J Surg Pathol. 1996;20(7):841-50.
13. Atesok KI, Alman BA, Schemitsch EH, Peyser A, Mankin H. Osteoid osteoma and osteoblastoma. J Am Acad Orthop Surg. 2011;19(11):678-89.
14. Barlow E, Davies AM, Cool WP, Barlow D, Mangham DC. Osteoid osteoma and osteoblastoma: Novel histological and immune histochemical observations as evidence for a single entity. J Clin Pathol. 2013;66(9):768-74.
15. Atesok Ki, Alman BA, Schemitsch EH, Peyser A, Mankin H. Osteoid osteoma and osteoblastoma. J Am Acad Orthop Surg. 2011;19(11):678-89.
16. Campanacci M. Osteoblastoma. In: Campanacci M, Enneking WF, editors. Bone and soft tissue tumors. 2nd ed. New York: Springer; 1999.
17. Tonai M, Campbell CJ, Ahn GH, Schiller AL, Mankin HJ. Osteoblastoma: Classification and report of 16 patients. Clin Orthop Relat Res. 1982;(167):222-35.
18. Saglik Y, Atalar H, Yildiz Y, Basarir K, Gunay C. Surgical treatment of osteoblastoma: a report of 20 cases. Acta Orthop Belg. 2007;73(6):747-53.
19. Papagelopoulos PJ, Galanis EC, Sim FH, Unni KK. Clinicopathologic features, diagnosis, and treatment of osteoblastoma. Orthopedics. 1999;22(2):244-7.
20. Fechner RE, Mills SE. Tumors of the bones and joints. In: Rosai J, editor. Atlas of tumor pathology. 3rd ed. Washington: Armed Forces Institute of Pathology; 1993.
21. Jennin F, Bousson V, Parlier C, Jomaah N, Khanine V, Laredo JD. Bony sequestrum: A radiologic review. Skeletal Radiol. 2011;40(8):963-75.
22. Mahboubi S. CT appearance of nidus in osteoid osteoma versus sequestration in osteomyelitis. J Comput Assist Tomogr. 1986;10(3):457-9.
23. Grey AC, Davies AM, Mangham DC, Grimer RJ, Ritchie DA. The ‘penumbra sign’ on T1-weighted MR imaging in subacute osteomyelitis: Frequency, cause and significance. Clin Radiol. 1998;53(8):587-92.
24. Gulati Y, Maheshwari AV. Brodie's abscess of the femoral neck simulating osteoid osteoma. Acta Orthop Belg. 2007;73(5):648-52.
25. Yamamura S, Sato K, Sugiuara H, Katagiri H, Ando Y, Fukatsu H, Iwata H. Prostaglandin levels of primary bone tumor tissues correlate with peritumoral edema demonstrated by magnetic resonance imaging. Cancer. 1997;79(2):255-61.
26. Ara G, Teicher BA. Cyclooxygenase and lipoxygenase inhibitors in cancer therapy. Prostaglandins Leukot Essent Fatty Acids. 1996;54(1):3-16.
27. Wold LE, Pritchard DJ, Bergert J, Wilson DM. Prostaglandin synthesis by osteoid osteoma and osteoblastoma. Mod Pathol. 1988;1(2):129-31.
28. Crim JR, Mirra JM, Eckardt JJ, Seeger LL. Widespread inflammatory response to osteoblastoma: The flare phenomenon. Radiology. 1990;177(3):835-6.