

CASE REPORT

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Aggressive osteoblastoma of ilium: Diagnosed on FNAC

Lubna Khan, MK Gupta, PK Singh, Asha Agarwal

ABSTRACT

Introduction: Aggressive osteoblastoma is a rare tumor which has a higher growth potential than conventional osteoblastomas and high risk of recurrence. **Case Report:** An aggressive osteoblastoma of ilium diagnosed on cytology which had three recurrences. Osteolytic lesion of hemate bone occurred in a 23-year-old patient. Tumor was excised and diagnosed as osteoblastoma on histopathology. First recurrence was managed with excision and implantation of rods, and bony chips from right iliac bone. Second recurrence was similarly managed by implantation of bony chips from the other iliac bone. Below elbow amputation was performed following third recurrence. After eleven months from the last surgery patient developed swelling of iliac bone and pulmonary metastasis. **Conclusion:** The case highlights the biological diversity of aggressive osteoblastoma and its potential for implantation at the site of surgery and distant metastasis. The case illustrates the difficulties which may be encountered in differentiating between benign and malignant form of osteoblastoma, and between aggressive osteoblastoma and osteosarcoma.

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INTRODUCTION

Osteoblastoma is an uncommon primary neoplasm of the bone. It has clinical and histological manifestations similar to those of osteoid osteoma; therefore, some consider the two tumors to be variants of the same disease, with osteoblastoma representing a giant osteoid osteoma. However, an aggressive type of osteoblastoma has been recognized, making the relationship less clear.

There is continuing debate in the literature regarding recurrent aggressive osteoblastic lesions with epithelioid osteoblasts, as to whether these represent benign, locally aggressive lesions [1] or osteosarcomas [2].

Aggressive osteoblastoma has unpredictable clinical course. A case of aggressive osteoblastoma is presented which had three recurrences along with implantation of tumor at surgical site and pulmonary metastasis over a period of 3 years and 4 months.

CASE REPORT

A 23-year-old male, presented with mild pain at the dorsum of hand around wrist joint. Thereafter gradually increasing 2x2 cm, hard, non mobile and non tender swelling appeared around the same region over a period

of 2 months. On X-ray osteolytic lesion of hemate bone was detected. Excisional biopsy was performed and it was diagnosed as osteoblastoma on histopathology. After 11 months 2x3 cm, hard, fixed non tender tumor recurred for which patient was reoperated and rods were implanted along with bony chips taken from right iliac bone. Unfortunately, 2x3 cm hard fixed non-tender tumor again recurred after a year for which patient was reoperated and rods were implanted along with bony chips which were now taken from left iliac bone. Lesion was diagnosed as osteoblastoma with active osteoblastic activity on histopathological investigation.

Third recurrence of tumor (4x4 cm) occurred after five months and level of alkaline phosphatase was raised (343 IU/L). Below left elbow joint amputation was performed. Patient was lost in the follow up. After 1 year 2 months, patient again reported in the outpatient department with 6x5 cm swelling in right iliac region which had developed over a period of three months. Retrospective history suggested asymptomatic period of 11 months followed by development of a small swelling in right iliac region (Figures 1 and 2). Level of alkaline phosphatase had increased further (840 IU/L) during 1 year. The digital pulmonary skiagram showed multiple round radiopaque shadows in the upper lobes of both the lungs suggestive of metastasis. Fine needle aspiration cytology (FNAC) was done from right iliac swelling.

On FNAC highly cellular smears consisting of oval osteoblastic cells with eccentric nuclei and dense eosinophilic cytoplasm were obtained. Some of these cells had characteristics 'hof'—a feature of activated osteoblasts. Multinucleated giant cells were abundant in the smears with occasional mitotic figures (Figure 3). Atypical mitosis and nuclear pleomorphism was evident. Cytomorphologic features, size more than 4 cm and frequent recurrence of previous lesions favored the diagnosis of aggressive osteoblastoma, which was later confirmed by histopathological examination.

DISCUSSION

Osteoblastoma is a benign lesion, which is differentiated from osteoid osteoma by the large size of the nidus (> 1.5 cm), the absence of surrounding reactive new bone formation and lack of intense pain [3]. Microscopically, osteoblastoma is well demarcated from the surrounding bone and is composed of haphazardly deposited trabeculae of woven bone rimmed by osteoblasts and scattered osteoclasts within richly vascular stroma. Cytomorphologically, the osteoblasts in conventional tumors are ovoid or round with eccentric nuclei and moderate amount of eosinophilic cytoplasm. Mitoses are uncommon and necrosis is usually absent [4]. Aggressive osteoblastoma is very rare tumor that is considered as borderline between benign osteoblastomas and osteosarcomas [5]. They have a higher growth potential than conventional osteoblastomas and are typically more than 4 cm in size with high risk of recurrence [5]. They are distinguished microscopically from the ordinary osteoblasts because



Figure 1: Clinical photograph: Swelling of right ilium.

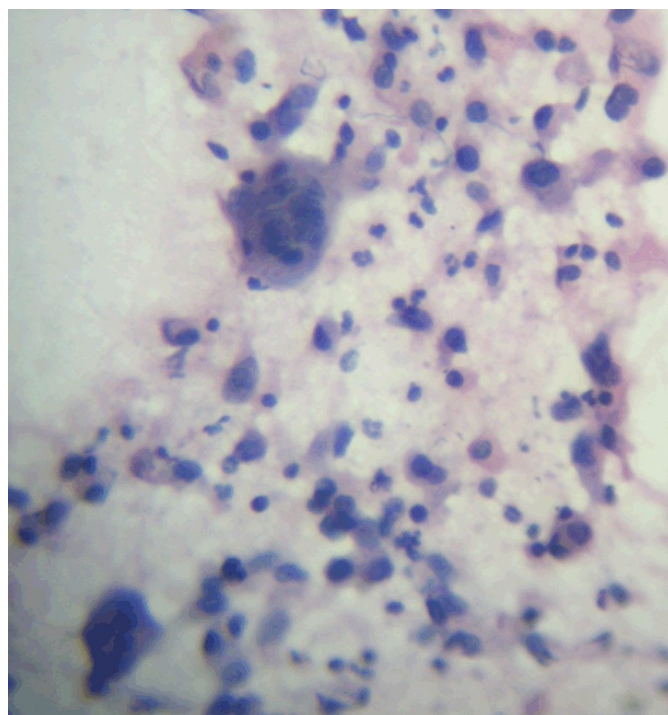


Figure 2: X-Ray showing lytic lesion of right iliac bone.

of the presence of wider or more irregular trabecula, which is bordered by epithelioid appearing osteoblasts, by the focal lack of a trabecular pattern of the osteoid proliferation [1]. Aspirates from aggressive osteoblastoma are highly cellular and the osteoblasts are two to three times larger than the conventional osteoblasts and have abundant eosinophilic cytoplasm with vesicular nuclei and prominent nuclei (epithelioid osteoblasts) [4]. The presence of epithelioid osteoblasts is the main differentiating point between conventional osteoblastoma and aggressive osteoblastoma [5] (Table 1).

The complex presentation of this case presented two diagnostic dilemmas. One was whether to consider this tumor as aggressive osteoblastoma or as low grade/well differentiated osteosarcoma. Morphologic features, involvement of short and flat bones, long duration of illness and diagnosis of osteoblastoma in hemate bone were in favor of diagnosis of aggressive osteoblastoma. Aggressive osteoblastoma is differentiated from

conventional osteosarcoma due to low mitotic rate and the absence of the following features: lace-like osteoid, permeation of surrounding intertrabecular spaces and atypical mitoses [6]. Aspirates from osteosarcoma are highly cellular. Osteosarcoma cells are spindled, oval, rounded, or pleomorphic, and show high degree of cellular atypia. Intracellular and/or extracellular osteoid may be present [5] (Table 2).

Table 1: Differentiation between osteoblastoma and aggressive osteoblastoma.

	Osteoblastoma	Aggressive Osteoblastoma
Size	Benign More than 1.5 cm	Borderline between osteoblastoma and osteosarcoma More than 4 cm
Cytomorphology	Osteoblasts are round or ovoid with eccentric nuclei and moderate amount of cytoplasm	Aspirates are highly cellular, osteoblasts are 2-3 times larger than conventional osteoblasts (epithelioid osteoblasts)
Histopathology	Haphazardly deposited trabeculae of woven bone rimmed by osteoblasts and scattered osteoclasts with in richly vascular stroma Mitosis rare	Wider and more irregular trabeculae bordered by epithelioid osteoblasts. Mitosis present. High risk of recurrence

Table 2: Difference between aggressive osteoblastoma and low grade / well differentiated osteosarcoma.

	Aggressive osteoblastoma	Low grade/ well differentiated osteosarcoma
Involvement	Short and flat bone	Long bone
Duration	Long	Short
Mitotic rate	Low	High
Atypical mitosis	Absent	Present
Lace like osteoid	Absent	Present
Permeation of surrounding bones	Absent	Present

Second problem was to establish whether the tumor developed de novo, or was it a metastasis of osteoblastoma, or weather it developed due to implantation during surgery. The implantation theory was given weightage due to the development of tumor in the iliac bone following sampling for bone chips.

The histopathological study of biopsy from right iliac region showed abundant osteoid with numerous empty as well as nucleated osteocytic lacunae separated by lacy osteoid matrix. The osteoid had numerous blotchy areas of calcification and a few entrapped dysplastic osteoblastic as well as spindle shaped cells (Figure 4). A small island of proliferating osteoblasts, showing well



Figure 3: FNAC: Osteoblasts with characteristic 'hof' and multinucleate osteoclastic giant cell (H&E, x500).

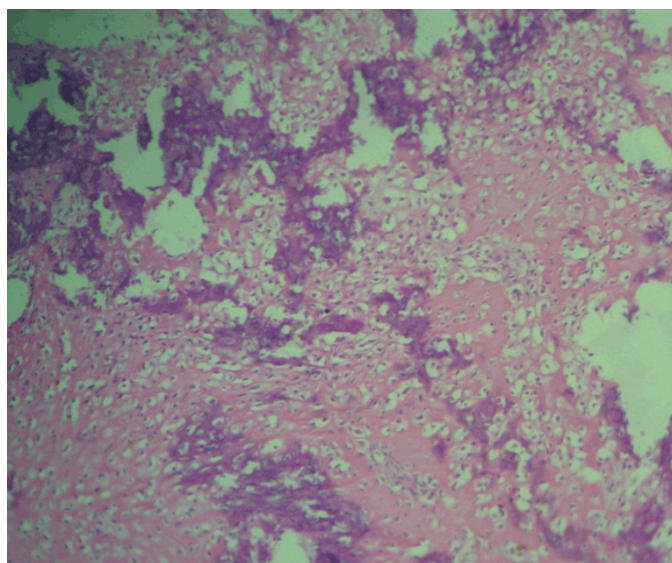


Figure 4: Section showing abundant osteoid with proliferation of immature plump osteoblasts and areas of calcification. (H&E, x250).

defined darkly stained nucleus and abundant deeply eosinophilic cytoplasm was seen at the periphery. Another small area showed proliferating spindle shaped fibroblastic type cells with aggressive appearing nuclei. Occasional multinucleated osteoclastic giant cells as well as a few small bi and tri nucleated giant cells were also appreciated. These findings favored the diagnosis of aggressive osteoblastoma.

Mark et al. [7] have reported two cases of aggressive osteoblastoma exhibiting diverse biological behavior. One was in iliac bone of 15 years old male patient. The patient died despite curettage, extensive chemotherapy, and radiotherapy. On autopsy distant metastases were detected. The other case was in femur of 12 years old female patient. Amputation was recommended following recurrence, but the patient declined and radiation therapy was given. Patient survived up to next 14 years on radiotherapy [7]. Two unusual cases have been reported in maxillary bone. Both the cases were initially diagnosed as osteoblastoma, but later it was suggested as a malignant transformation of osteoblastoma into osteosarcoma [8]. One case has been reported in calcaneum of a 29-year-old female, which was initially diagnosed as benign osteoblastoma but after recurrence, revised diagnosis of aggressive osteoblastoma was established on histology [9]. Osteoblastoma with conversion to osteosarcoma should be considered as a separate tumor entity distinguished from genuine osteosarcoma [10]. However, it is very difficult to differentiate between aggressive osteoblastoma and low grade/well differentiated osteosarcoma [11] both radiologically and histologically. The presence of epithelioid osteoblasts, trabecular osteoid, prominent osteoclastic activity, absence of cartilage, well demarcated woven bone-host bone interface and low mitotic activity are some of the histological criteria to differentiate it from osteosarcoma.

CONCLUSION

These cases highlight the biological diversity of osteoblastoma like lesions and illustrate the difficulties which may be encountered in attempting to differentiate between benign and malignant forms of osteoblastoma and between aggressive osteoblastoma and osteosarcoma.

Author Contributions

Lubna Khan – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article and revising it critically for important intellectual content, Final approval of the version to be published

MK Gupta – Substantial contributions to acquisition of data, analysis and interpretation of data, Drafting the article, Final approval of the version to be published

PK Singh – Substantial contributions to conception and design, Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Asha Agarwal – Substantial contributions to conception and design, Analysis and interpretation of data; Revising the article critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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