Osteoblastoma is a rare primary neoplasm of bone, usually categorized as benign bone tumor. It accounts for about 1% of primary bone tumors. Roughly 40% of osteoblastomas localize to the spine which may cause pain, scoliosis, or even neurologic deficit. Involvement of the sacrum by osteoblastoma is rare. We report a case of osteoblastoma in the sacrum with good result after treatment.

A 13-year-old male presented with a 6-month history of low back pain radiating from the right buttock to the right leg. Plain radiography, CT, and MRI of the lumbosacral spine revealed a lesion over the right ala of the sacrum. Biopsy confirmed a diagnosis of osteoblastoma. Superselective transarterial embolization was done before surgery to minimize bleeding. After surgery, his back pain improved, and was recurrence-free at the time of 3-year follow-up after treatment.

Case Report

Clinical presentation

A 13-year-old male patient presented with a 6-month history of chronic low back pain radiating from the right buttock to the right leg. His symptoms had aggravated gradually. Physical examination revealed a well-developed, well-nourished Asian young man with a chronically ill appearance. There was tenderness on palpation of the right hip and leg, but no neurological deficits and no limitation in range of motion.

Imaging and pathological findings

Plain radiography of the lumbosacral spine revealed a mixed osteosclerotic and osteolytic lesion over the right ala of the sacrum (Fig. 1a). Computed tomography (CT) of the sacrum revealed an expansile bone lesion in the right upper sacrum with radiolucent areas and a sclerotic rim (Fig. 1b). There was bone formation within the lesion and right S1/S2 foraminal stenosis was noted.
Osteoblastoma in the sacrum

(Fig. 1b). T1-weighted Magnetic resonance imaging (MRI) of the sacrum showed a large mass isointense to muscle with a sclerotic rim and a small hyperintense focus of recent hemorrhage (Fig. 2a). T2-weighted MRI revealed a hyperintense and heterogeneous mass with multiple dark foci representing bone or calcification (Fig. 2b). Contrast-enhanced MRI revealed heterogeneous enhancement with a thick sclerotic rim (Fig. 2c).

A direct bone biopsy was done via L5 hemi-laminectomy to identify the involvement of the S1 nerve. The gross appearance of the specimen was tan, white, and firm, without extensive vascularization. Microscopic study (Fig. 3) revealed haphazard proliferation of fibrovascular tissue with interlacing of osteoid cells.

There was osteoblastic rimming of the trabeculae without evidence of malignancy. Pathological diagnosis was osteoblastoma. The patient had surgery a month later.
Osteoblastoma in the sacrum

Prior to surgery, an angiogram was performed and it revealed a tumor supplied by a branch of right internal iliac artery. Therefore, a super-selective small tumor vessel transarterial embolization (TAE) was done to prevent uncontrolled bleeding during the surgery. Then, curettage of the tumor, bone grafting and lymphadenectomy was done via midline lower abdomen laparotomy. The surgeons found a 6 cm by 5 cm by 5 cm tumor in the S1 vertebra, right ala, and body.

Pathological examination of the tumor confirmed the diagnosis made at biopsy. A well-circumscribed tumor with a sclerotic rim revealed its slow-growing nature. Tumor cells were epitheloid with small nucleoli and several multinucleated cells. No obvious atypical mitoses were noted, while the lymph nodes showed lymphoid hyperplasia. The absence of lace-like osteoid or sheets of osteoblasts, together with the low mitotic index, are characteristic of osteoblastoma [5].

Post-operative course

The patient had post-operative follow-up exams immediately after surgery and at 1, 6, 12, 24, and 36 months. Follow-up plain radiography showed multiple radiopaque densities in the right sacral segment due to post-operative sclerotic change. The patient complained of morning back pain 2 months after surgery, but it gradually resolved. At 6 months post-surgery, MRI showed residual or recurrent expansile bone tumor in the right upper sacrum, with progressive massive peritumoral marrow edema. Bone scan at the same time showed no apparent interval change compared with a study at 1 month, and this was considered to be residual tumor. The Tc-99m methylene diphosphonate (MDP) whole body scan revealed a slight increase in radioactivity in the right upper sacral region with extension to the sacroiliac joint. Follow-up plain radiographies of the sacrum at 12, 24, and 36 months showed no interval change.

DISCUSSION

Our case is an expensile type osteoblastoma in sacrum. Its sclerotic ring implies its slow-growing nature. Although, the tumor has a blow out expansion to the adjacent tissues, it still has well-defined margin, and no moth eaten bony destruction nor soft tissue invasion as most malignant tumor such as osteosarcoma do. However, we can not totally exclude the possibility of the aggressive osteoblastoma and low grade osteosarcoma. According to a study by Tonai et al. in 1982, the diagnosis of aggressive osteoblastoma is based on radiographic appearance (cortical destruction), histologic pattern (highly cellular tissue with large plump osteoblasts predominating) and clinical course (rapid recurrence after curettage) [8]. Therefore, further follow up is necessary after surgery. Although aggressive osteoblastoma is more destructive and easily recurred, there has been no report of distant metastasis [8].

Radiography and conventional tomography are usually enough for differential diagnosis between osteoblastoma and osteosarcoma. However, low grade and intracortical osteosarcoma are sometimes very difficult to differentiate from osteoblastoma radiographically, especially when it exhibiting a geographic bony destruction without apparent periosteal reaction nor soft tissue mass [5]. Biopsy is usually needed for diagnosis but sometimes osteoblastoma can be difficult to differentiate from low grade osteosarcoma by histology, particularly if there is limited biopsy material [5]. Clinicians must combine clinical presentation, history, radiological findings, and pathological findings in order to achieve reliable diagnosis.

MRI and CT studies were done for the patient because its complex anatomic location. We would like to know whether there is cortical breakthrough, soft tissue invasion, or soft tissue mass implying a potential malignancy. Besides, MRI and CT provide more information of the extension of the tumor and

Figure 3. Microscopic findings (HE stain) (100 ×) shows haphazard proliferation of fibrovascular tissue with interlacing of osteoid. A single layer of osteoblastic rimming of the trabeculae is seen without permeation (arrows). No atypical cells were found, suggesting that this is not osteosarcoma.
Osteoblastoma in the sacrum

its relation with the surrounding tissues which is necessary for preoperative staging. However, since the osteoblastic matrix containing tumors all present with low signal intensity in MRI, MRI provides less information about the matrix of tumor than plain film. Therefore, specific diagnosis is difficult to make based on MRI study.

Differential diagnosis of osteoblastoma should include osteoid osteoma, giant cell tumor, aneurysmal bone cyst, bone abscess, and enchondroma. Osteoblastoma rarely has a radiographic nidus, and involves the vertebral column more often than does osteoid osteoma. In addition, osteoid osteoma is rarely larger than 1 cm [7]. A giant cell tumor typically arises at the distal femur or proximal tibia of a young adult after epiphyseal closure, and rarely shows mineralization radiographically. If the lesion involves the end of a long bone or the body of a vertebra, a giant cell tumor should be considered first [7]. A bone abscess is usually marked by a serpentine tract which is almost never seen in osteoblastoma. An aneurysmal bone cyst sometimes is similar to osteoblastoma but lacks central opacities. An enchondroma usually display a calcified matrix of dots, rings, and arcs. Most sacral tumors are benign, and diagnosis can occur late in the disease course [1]. In a retrospective study of osteoblastoma, the average duration of pain was 13.9 months [9].

The treatment of choice for osteoblastoma is surgery to excise the tumor. Curettage combined with chemo-or thermocauterization can provide a complete cure for benign lesions. Although it is possible that radiotherapy can induce malignant change of an osteoblastoma, radiotherapy is often used for the adjuvant therapy of residual tumor or for inoperable tumor [9]. The recurrence rate of tumor in the sacrum can be as high as 67%, but tumor recurrence usually occurs in the context of failure to get adequate tumor exposure and full excision [7]. Incomplete curettage of osteoblastoma is associated with recurrence [10].

In summary, we presented a rare case of osteoblastoma with sacral involvement. It is crucial to distinguish osteoblastoma from osteosarcoma and aggressive osteoblastoma. Regular follow-up for patients with sacral lesion showing equivocal imaging is necessary in order to achieve correct diagnosis of osteoblastoma.

REFERENCES
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骨母細胞瘤：病例報告

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骨母細胞瘤（osteoblastoma）是一種罕見的良性原發性骨瘤，佔所有原發性骨瘤的百分之一。大約有四成的骨母細胞瘤發生在脊椎，而我們要報告的病例是發生於薦骨，一個更為罕見的位置。

一位 13 歲的男孩因為持續 6 個月由右側臀部延伸到腿部的疼痛來求診。腰薦部的 X 光顯示有一病兆在右側薦骨的翼部。電腦斷層掃描及磁振造影檢查後便進一步作切片，結果顯示該病兆為骨母細胞瘤。由於該腫瘤易出血所以在術前先做選擇性的動脈血管栓塞術以減少術中的出血。術後病人的下背痛改善了，並且之後 3 年的追蹤也沒有復發的現象。

骨母細胞瘤是一種罕見的良性原發性骨腫瘤，通常發生在脊椎可能會造成疼痛，脊椎側彎，甚至是神經學的症狀。易有復發的情形，通常手術治療後病人的症狀都能改善。薦骨的骨母細胞瘤十分罕見，也會造成一般常見的慢性下背痛，因此將其報告出來作為鑑別診斷的參考。