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CASE REPORT

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Osteochondromas gone pelvic: A case report of concurrent tibial osteoblastoma and incidental detection of multiple pelvic osteochondromas on bone scan

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ABSTRACT

In a unique clinical presentation, a 20-year-old female with a known left distal tibial osteoblastoma (previously treated via curettage) developed pain and weight-bearing difficulties in her left lower limb 6 months later. Subsequently bone scan revealed an unprecedented clinical presentation: residual activity in the treated osteoblastoma alongside additional foci of increased tracer uptake on skeletal scintigraphy, localized to the bilateral anterior superior and inferior iliac spines. Subsequent radiographs confirmed sessile bony outgrowths in these locations, consistent with multiple pelvic osteochondromas (an uncommon variant). Notably, these osteochondromas lacked associated radiolucencies or cortical destruction. Furthermore, an magnetic resonance imaging. confirmed the osteochondroma diagnosis. Interestingly, the patient declined surgical intervention for the asymptomatic osteochondromas, prioritizing ongoing treatment for the primary osteoblastoma. This case highlights the rare co-occurrence of these distinct tumor types in an uncommon usual pelvic location, emphasizing the importance of a patient-centered treatment approach based on patient preferences and clinical context.

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Introduction

Benign bone tumors are classified on their cellular composition and matrix production. The main categories are bone-forming, cartilage-forming, connective tissue, and vascular tumors. They are usually formed in growing skeletons and diagnosed incidentally or due to mild symptoms in children or young adults.

Osteochondromas (commonly called exostosis) are the most common benign neoplasms of bone, representing 10%–15% of all primary bone tumors and up to 50% of benign bone lesions [1], occurring in children or adults, the male-to-female ratio is 1.6:1. [2]. It is a cartilage-forming tumor that undergoes longitudinal growth via its chondrocytic activity. Osteochondroma is an exophytic growth from the bone that shows the diagnostic imaging features of cortical and medullary continuity with the underlying bone [1], presenting either as a pedunculated or a sessile mass (the latter being more common). The lesion is typically mushroom-shaped affecting the metaphysis of long bones such as the femur and tibia [3]. It usually affects bones that develop by enchondral ossification and rarely originates from bones that develop by intramembranous ossification such as the scapula, pubic ramus clavicle, and ribs [4].

Osteoblastoma is a rare, benign, osteoid-elaborating tumor, typically greater than 2.0 cm in diameter. Radiographically, they are highly variable and may be blastic, lytic, or mixed lytic-blastic. The lesions may be well-defined, exophytic, or very aggressive in radiographic appearance. In contrast, for osteoblastoma, the patient's age is typically 10–30 years, male-to-female ratio is usually 2:1. The spine and sacrum are the most common locations, but it can occur in any bone [2].

We report a rare case of a young girl known case of left distal tibial osteoblastoma (previously treated via curettage) with incidental finding of osteochondromas of bilateral anterior superior and inferior iliac spines on bone scan.

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Case Report

A 20-year-old female known left distal tibia osteoblastoma underwent curettage 5 times last on 20/7/2022. After 6 months patient developed complaints of pain and weight-bearing difficulties in the left lower limb. She was referred to our Department of Nuclear Medicine for a bone scan to evaluate for residual tumor.

Dynamic images of skeletal scintigraphy revealed increased perfusion and blood pool accumulation over the distal part of the left leg (Fig. 1), while delayed images showed abnormally increased tracer uptake over the distal part of the left tibia thus pertaining to findings of residual disease (Fig. 2a and b). Incidentally, foci of increased tracer uptake were also noted bilaterally, involving anterior superior and inferior iliac spines (Fig. 3).

There was no pain in the pelvis, no swellings appreciated on abdomino-pelvic examination, no history of trauma, and the general physical and systemic examinations were within normal limits. Regional lymph nodes were not enlarged. Anterior pelvic X-ray was advised and it revealed a sessile bony outgrowth from bilateral anterior superior and inferior iliac spines, without any evidence of focal radio lucencies or cortical destruction (Fig. 4).

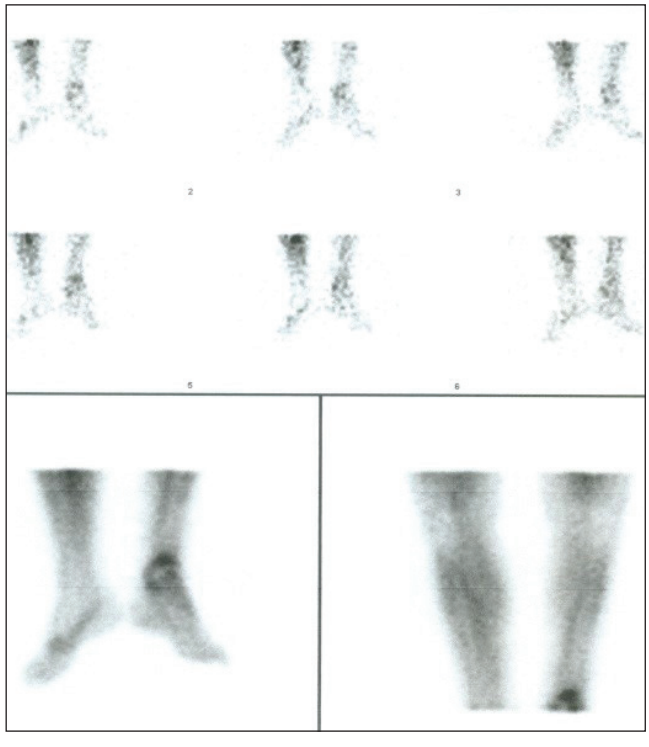


Figure 1. Dynamic imaging showing increased blood pool accumulation in the left distal leg.

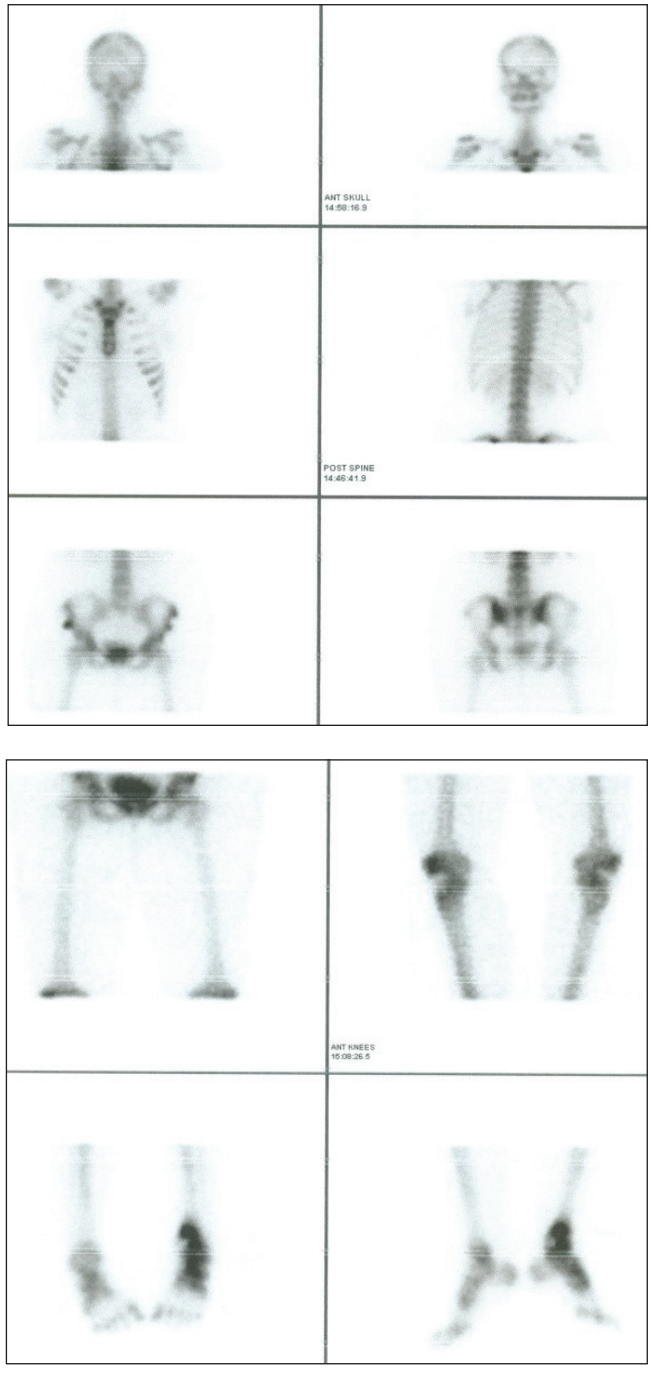


Figure 2. (a) Delayed static images showing normal distribution of the tracer in bones, increased tracer uptake seen over anterior iliac spines. (b) Static images showing increased tracer uptake in the left distal tibia representing residual tumor (Osteoblastoma).

The hematological and biochemical tests were within normal limits. The patient was advised magnetic resonance imaging (MRI) of the pelvis, which revealed exophytic masses from the anterior superior and inferior iliac spines largest 1.5 cm in size of the right anterior superior iliac spine. The osseous exostosis shows cortical and medullary continuity with the underlying bone. There is a 5 mm



Figure 3. Delayed static zoomed anterior and posterior pelvic images showing foci of abnormally increased tracer uptake over bilateral anterior superior and inferior iliac spines.



Figure 4. X-ray pelvis AP view showing sessile outgrowths from bilateral anterior superior and inferior iliac spines.

cartilaginous cap surrounding all osteochondromas. The patient opted for nonsurgical management of the osteochondromas prioritizing treatment for the symptomatic osteoblastoma.

This was, therefore, an interesting case as two coexistent tumors from different origins and a rare presentation of multiple osteochondromas were seen.

Discussion

Osteoblastoma and concurrent multiple osteochondromas of iliac spines are rare occurrences. To this date in the literature search, this is a first reported case. Another case previously reported had coexisting osteoblastoma and solitary osteochondroma of the humerus [5].

In this case patient was undergoing treatment for osteoblastoma and multiple exostosis were determined on a bone scan. Osteochondroma may occur in any bone that is formed by enchondral bone growth. It is intimately associated with the growth plate. One theory of pathogenesis describes a focus of ectopically oriented physis growing at right angles to the longitudinal axis of the bone [2]. When multiple lesions are present, osteochondromas exist as part of a syndrome known as hereditary multiple osteochondromas which is related to mutations in EXT1 and EXT2 genes [4].

Pelvic osteochondromas are rare; however, the crestal border of the ilium, vertebral border of the scapula, and ends of the clavicle are not unusual sites [6]. It is hypothesized that these tumors represent growth plate cartilage that has displaced from the metaphysis [7]. Most of the patients present with a painless bony swelling. However, they may also present with signs and symptoms of lumbar nerve root compression, symptoms related to local pressure, and mass effect. Most commonly, osteochondroma manifests as a slowly expanding, hard, painless mass [8].

Inheritance pattern is autosomal dominant which can cause multiple exostosis or isolated lesions [9]. They usually present during the period of rapid skeletal growth and cease to grow once maturity is reached [10]. Malignant transformation of the cartilage cap into a secondary Chondrosarcoma varies from less than 1% for solitary osteochondromas to 2%–5% in cases of hereditary multiple exostosis [11]. Plain radiographs are often diagnostic. They show a "trumpet-shaped deformity" due to the metaphyseal widening and accounting for 88.2% of the cases [12].

Ultrasound helps to determine the thickness of the cartilaginous cap. If it continues to grow after skeletal maturity, malignant transformation should be considered [2]. MRI is the imaging modality of choice for evaluating the thickness of the cartilaginous cap. Normally, the cap is only a few millimeters thick in adults and any thickness more than 2 cm should be viewed suspiciously [7].

Definitive diagnosis is usually established on histopathological examination. The presence of a hyaline cartilaginous cap covering the bone is diagnostic. Most of the osteochondromas can be managed by observation alone. Surgical treatment in the form of en bloc resection is usually indicated for pain, cosmetic reasons, neurovascular compromise, abnormal growth, skeletal deformity, decreased motion of the adjacent joint, or in cases with evidence of malignant transformation.

Osteoblastomas are rare, benign bone tumors composed of active osteoblasts that produce osteoid and immature lamellar bone. They are frequently described as large (>1.5–2 cm) osteoid

Table 1. Osteoblastoma cases reported in literature.

Study	Ethnicity	Age	Gender	Location	Management	Complications
Alkan et al. [13]	Turkey	17	Male	Right proximal tibia	Biopsy done NOF with osteoblastoma of tibia	Not mentioned
Hani et al. [14]	French	30	Male	Distal tibia rt	Curettage	None
Görgün et al. [15]	Turkey	13	Male	Proximal left tibia	Curettage bone grafting	Malignant transformation to osteosarcoma after 2 years with pleural mets resulting in death
Mortazavi et al. [16]	Irani	20	Female	Proximal tibial	Excision	None, no complications up to 16 months
Miszczyk et al. [17]	Polish	55	Male	Proximal tibia	Excision	Post-op 1 year recurrence with malignant transformation EBRT done
Miszczyk et al. [17]	Polish	24	Female	Distal shaft	Nonradical surgery	EBRT done after 4 months due to severe pain, recurrence
Miszczyk et al. [17]	Polish	16	Male	Proximal tibia	Curettage	No complication after 39 months
O'Connell et al. [18]	USA	47	Male	Multiple lesions involving the left tibia and fibula	Above knee amputation	No complications, pt. followed up to 5 years

osteomas, although osteoblastomas tend to grow rather than regress, and they are less often painful compared to osteoid osteomas. While the malignant transformation of osteoblastomas is rare, they can be aggressive and have a high rate of recurrence following treatment [5]. Periosteal reaction and cortical thinning and expansion are common. Aggressive lesions can also cause cortical destruction reported in literature with complications, listed in Table 1.

The cells of both tumors consisted of cells of different origins types namely osteoblasts and chondrocytes therefore pathogenesis is unclear. Osteochondromas could be due to mutation of EXT1/EXT2 genes. The genesis of osteoblastoma is a less clear process as it is poorly understood on a molecular basis.

Following a thorough discussion of the risks and benefits, the patient, in collaboration with the care team, prioritized treatment for the symptomatic osteoblastoma and opted for nonsurgical management of the asymptomatic osteochondromas. While this approach aligned with the patient's preferences and immediate medical needs, it did preclude definitive characterization of the osteochondromas via biopsy or surgical exploration. This asymptomatic presentation is typical for osteochondromas.

Conclusion

We report this case due to its rare co-occurrence of two tumors (Osteoblastoma and Osteochondromas) of separate cell origin, multifocality of osteochondromas, and its uncommon location at anterior superior and inferior iliac spines incidentally detected on skeletal scintigraphy.

List of Abbreviations

EXT, Exostosin glycosyltransferase; MRI, Magnetic resonance imaging.

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