Small Bone Islands: Unusual Clinical Symptomatology

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abstract

Bone islands are usually considered benign, stable, nonprogressive lesions, radiographically characterized by an ovoid, round, or oblong homogeneously dense and sclerotic focus in the spongiosa, with a preference for the long bones and the pelvis. Benign solitary bone islands are usually believed to be asymptomatic, with no necessity for treatment. Symptomatic bone islands reported in the literature are characterized by a diameter greater than 2 cm, belonging to the category of giant bone islands.

The authors report a rare case starting from a painful symptomatology and involving dense sclerotic bone lesions of less than 2 cm, thus falling into the category of small bone islands. The patient underwent a surgical resection to achieve complete recovery. Normally, small bone islands do not explain the pain in patients who present with symptomatology when they are detected during radiographic studies. Based on this case, the authors believe that even small bone islands can be the cause of symptomatology and justify a surgical procedure if all medical investigations suggest no other possible causes of the pain.

Figure: Bone scan demonstrating increased uptake in the area of the lesion.

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An enostosis or bone island is defined as a focus of compact (cortical) bone within the cancellous bone, representing a failure of resorption during endochondral ossification. Typically asymptomatic and found as an incidental lesion, a bone island is diagnosed largely on the basis of its characteristic radiographic features. With a reported frequency of approximately 1%, bone islands can be found in most parts of the skeleton but have a preference for the pelvis, femur, and other long bones. Most symptomatic bone islands reported in the literature are giant, measuring more than 2 cm in diameter. Small and asymptomatic bone islands are observed as incidental findings and do not interfere with the differential diagnosis or necessitate treatment. In contrast, recognizing giant symptomatic bone islands and differentiating them from more significant primary or metastatic tumors is important. Reports of small symptomatic bone islands measuring less than 2 cm in diameter are rare. The current authors found only 1 other case of this entity.

The purposes of this article are to present the case of a symptomatic small bone island, to discuss the clinical presentation and the management of this lesion once pertinent differential diagnostic workup ruled out a nonsignificant bone lesion such as a primary or metastatic tumor, and to provide a brief review of the literature.

**CASE REPORT**

A 10-year-old boy reported a 2-year history of nocturnal left hip pain with a recent limp and decreased walking distance. Physical examination revealed a painful left hip with decreased range of motion. Radiographs and computed tomography scan of the left hip showed a 1.2×1.2×0.8-cm anterolateral, sclerotic, dense lesion with brush borders in the proximal femoral epiphysis (Figures A-D). The lesion was in contact with the epiphyseal growth plate and the articular cartilage of the femoral head, which was subluxated 4 mm laterally compared with the right femoral head. A bone scan demonstrated an increased uptake in the area of the lesion (Figure E). Magnetic resonance imaging showed low signal intensity on both T1- and T2-weighted sequences, characteristic of cortical bone (Figure F). All of these findings were consistent with a bone island. Due to the persistence of pain in the left hip, subluxation, and decreased range of motion, and because no other cause of these symptoms was evident, the authors decided to perform a transmetaphyseal curettage of the lesion. Histologic findings of the curetted lesion were consistent with bone island. After 1 month, the pain completely disappeared, and the hip achieved full range of motion. At 4-year follow-up, the patient was well and reported no left hip pain. No epiphysiodysis of the upper femoral physis occurred.

**DISCUSSION**

A bone island is a completely benign, discrete focus of compact bone with Haversian systems located within the cancellous bone. Mirra defined it as misplaced, hamartomatous cortical bone. Likely congenital or developmental in origin, a bone island is a benign condition closely related to osteopoikilosis. It can be found in almost any bone in the body except the cranial vault and has a predilection for the long bones and pelvis. The spine is rarely involved, accounting for only 3 (1.4%) of 209 bone islands reviewed by Onitsuka; these vertebral bone islands involved the thoracic and lumbar segments. Bone islands are more common in adults and show no sexual predilection. On radiographs, they are ovoid, round, or oblong, with the long axis parallel to the cortex of the affected bone. Bone islands are homogeneously dense and sclerotic in the cancellous bone with distinctive radiating bony streaks known as thorny radiation or pseudopodia.

The radiological differential diagnosis of a bone island includes osteoma, osteoid osteoma, osteoblastoma, enchondroma, medullary bone infarct, fibrous dysplasia,
healed nonossifying fibroma, and unusual osteosarcoma. Of the bone forming and fibro-osseous lesions mentioned, perhaps the most important diagnosis to exclude is osteosarcoma, particularly the low-grade central variety. In the current case, the radiologic findings were consistent with a benign lesion; however, a biopsy was performed because the authors had already performed a resection curettage of the bone island with a therapeutic purpose.

Bone islands usually do not demonstrate increased uptake of a tracer on bone scan. However, several reports in the literature have described histologically proven bone islands marked by increased radionuclide uptake on bone scan, as in the current case. The exact pathomechanism of this phenomenon is uncertain. Sickles et al speculated that increased radionuclide activity is the result of either the large size or the continued growth of a lesion because uptake of radiopharmaceutical is roughly proportional to the volume of a lesion. Greenspan et al correlated the radiographic and pathologic findings in 6 cases of bone islands and suggested that the increased tracer uptake observed in 2 bone islands appeared to be directly related to the higher degree of bone remodeling and osteoblastic activity that these lesions exhibited in comparison with the scintigraphically “cold lesions.”

The majority of bone islands are between 0.1 and 2 cm in greatest diameter, and they are usually asymptomatic. Symptomatology has been reported with bone islands that attain a size of 2 cm or greater in any diameter. Eight cases of symptomatic giant bone island have been reported in the literature. These bone islands had a mean diameter of 7.3 cm (range, 2.5-10.5 cm). Symptomatology related to small bone islands has only been reported once, in a 69-year-old woman who had a history of breast cancer 5 years prior to bone island diagnosis. Pain was located in the proximal tibia. Radiographic studies showed a dense sclerotic lesion measuring 1.5×1.4 cm. The pathology after en bloc resection confirmed the diagnosis of bone island.

Most bone islands do not require treatment after the diagnosis has been established; however, when there is persistent pain, even with small bone islands, surgical treatment may be helpful. In the current case, the patient’s pain did not respond to medical treatment. All investigatory examinations were negative except for the presence of a bone island confined to the proximal femoral epiphysis. Because the patient’s symptomatology could not be explained by any pathology other than a bone island, the authors decided to perform a transphyseal resection with a trocar. One month after the intervention, the patient was completely asymptomatic, proving that the source of this symptomatology was the small bone island. The presence of a small island in a relatively small bony cavity, such as vertebrae or a proximal femoral epiphysis, is probably associated with pain that can be induced by hyperpression in this cavity. On opening the cavity and resecting the lesion, the local hyperpression, as well as the symptomatology, disappears.

**CONCLUSION**

When a patient presents with painful symptomatology and a small bone island is found in the symptomatologic region, the bone island should not be considered the source of the pain unless the pain persists and there is no other reason for it. If that is the case, the hypothesis should be that the bone island is responsible for the symptomatology. This was proven by the current patient’s full recovery after removal of the bone island. The diagnosis of bone island should be based on the morphologic features of the lesion on plain radiography, computed tomography, and magnetic resonance imaging rather than on any clinical circumstances.

**REFERENCES**

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