Fine-Needle Aspiration of Spinal Osteoblastoma in a Patient With Lymphangiomatosis

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Diagnosis of osteoblastoma by fine-needle aspiration (FNA) is rare, as in most patients, excisional biopsy is performed. We report a case of FNA diagnosis of a spinal osteoblastoma in a lymphangiomatosis patient. The patient had a history of lymphangiomatosis since birth with lymphangiomas removed from various locations, and had radiograph evidence of multifocal vertebral involvement. He presented with an enlarging C3 posterior element vertebral lesion. Clinically and radiologically, it was suspicious for lymphangiomatosis involvement. A computed tomography (CT)-guided FNA yielded a moderately cellular specimen. Lesional cells exhibited plasmacytoid features, fine chromatin, smooth nuclear membranes, and binucleation, in a background of occasional osteoclastic cells, spindle cells, and osteoid matrix. A cytological diagnosis of osteoblastoma was favored and confirmed with follow-up surgical biopsy. We believe that this is the first case reporting an osteoblastoma diagnosed by FNA in a lymphangiomatosis patient. We describe the clinical and cytological findings of osteoblastoma and its differential diagnosis.

Osteoblastoma (OB) is a benign tumor representing less than one percent of all primary bone tumors and over three percent of benign primary tumors. It has an affinity for young males primarily in the second decade. The tumor most commonly involves the vertebral column, particularly the posterior or dorsal elements, as well as the sacrum. It also occurs in the long bones (with a slight metaphyseal over diaphyseal predominance), jaw, and craniofacial bones; however, any bone can be affected.1,2 Symptoms consist of dull, progressive bone pain that is more generalized than that of osteoid osteoma, and less likely to be relieved by aspirin or other analgesics. Rarely, systemic symptoms can occur, including weight loss, fever, and anemia.3 Spinal lesions may present with neurologic symptoms as a result of mechanical compression, as well as scoliosis.1,4 Because of its capacity for progressive growth with local bony destruction creating clinical symptoms as well as a potential for aggressive behavior, complete surgical excision is the preferred treatment.5,6 Osteoblastoma is not known to be associated with any syndrome.

Case Report

A 15-yr-old male with a significant past medical history of diffuse lymphangiomatosis presented for evaluation of neck pain that was slowly progressing over the past year. The pain was described as sharp, and worsened with activity but relented with ibuprofen. There was occasional concomitant right upper arm numbness and tingling without weakness. No other neurological symptoms were present. Physical examination showed tenderness to palpation of the right neck without an apparent mass. A complete neurological examination was normal.

His past medical history was notable for diffuse lymphangiomatosis at birth, and numerous lymphangiomas removed from various locations in the thorax and extremities over the years, including the mediastinum, skull, humerus, tibia, femurs, and pelvic region. Clinical and radiological examinations were notable for several intra-abdominal and thoracic lesions. Multiple lytic lesions in the thoracic and lumbar spine involving the vertebral bodies were noted as well. He received treatment with α-2 Interferon, with stabilization of his lymphangiomas several years before his latest presentation.

An initial CT scan 6 mo earlier revealed a 0.9 cm enhancing soft tissue focus with no aggressive features (believed to represent a lymph node), associated with the C3 vertebral body on the right side. This mass progressed to a 2.9 cm lesion on a follow-up scan 5 mo later. The latter CT showed an expansile lytic mass arising from the lateral posterior elements at C3 with evidence of bony irregularity, poorly defined margins, and infiltration into the ver-
terial body (Fig. 1). On MRI, the mass was homogeneous and had a T1 signal slightly higher than the muscle with an intermediate-to-high T2 signal. A clinical working diagnosis of recurrent lymphangiomatosis with concern for degeneration into a more aggressive process was made.

A fine-needle aspiration (FNA) was performed showing a moderately cellular specimen in a background of blood. The cells exhibited plasmacytoid features with round nuclei having somewhat fine chromatin and smooth nuclear membranes, and distinct, single nucleoli. There was a moderate amount of pale-blue to blue cytoplasm on Papanicolaou and Diff Quick stains, respectively, with a perinuclear “Hof” and peripheral cytoplasmic condensation (Fig. 2A). Occasionally, these cells were binucleated. Benign appearing osteoclastic giant cells and spindle cells were present in small number (Fig. 2B and C). The cell block revealed cellular groups, with an eosinophilic matrix interspersed between the cells consistent with osteoid (Fig. 2D). A cytologic diagnosis of an osteoblastic neoplasm, favoring osteoblastoma, was made. Excision of the tumor with C2-3 fusion was performed. Histologic diagnosis confirmed osteoblastoma (Fig. 3).

Discussion

We report a case of FNA diagnosis of OB in a lymphangiomatosis patient with a spinal lesion. In fact, the clinical and radiological impressions favored recurrent lymphangiomatosis. CT scans for lymphangiomas in bone are widely varied typically revealing a single, often multiloculated, sharply delineated, cystic/solid mass with a uniform, homogeneous appearance having no periosteal reaction in adjacent bone. However, the radiologic appearance can exhibit irregular cyst walls with calcification, as well as inhomogeneous patterns leading to a broad differential diagnosis. Such a differential diagnosis includes tumors of vascular origin, aneurysmal bone cyst, giant cell tumor, fibrous dysplasia, chondroma or chondrosarcoma, as well as osteoblastoma and osteosarcoma.

The FNA findings in this case were consistent with an osteoblastoma as opposed to the findings of cystic fluid, with lymphocytes and histiocytes in lymphangioma. Confirmation of the diagnosis was made by follow-up excisional biopsy. We believe that this is the first reported case of the osteoblastoma in a lymphangiomatosis patient. FNA provided a diagnosis leading to a different clinical treatment and management. The significance of association of osteoblastoma and lymphangiomatosis is not clear. One could speculate that there may be an etiologic relation. For
example, it has been proposed that osteoid osteoma (OO), a tumor with very similar morphologic features with osteoblastoma, may derive from an aberrant vascular development.10

Osteoblastoma was originally described in 1932 by Jaffe and Mayer.1 The age range for osteoblastoma is wide, encompassing those as young as 5 to those 45 or even 75 yr of age.6 OB occurs within the bone cortex in ~2/3 of the cases, with an average size of 3.2 cm on radiographic stud-

From a practice point of view, FNA shows good to excellent correlations to histological sections. When com-
pared to radiographic accuracies, FNA compares favorably suggesting FNA can be of value in the standard diagnostic workup of a bony tumor, especially when other clinical or radiographic parameters do not agree.4,15

In summary, we believe this is the first case of osteo-
blastoma discovered in association with lymphangiomi-
atosis. Whether a causative relationship is present is not
known; however, the possibility of a vascular link in
osteoid osteoma etiology as speculated in the literature
may provide insight to the relationship between the two
conditions.

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