A case of multiple aggressive osseous hemangiomas on bone scan

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A B S T R A C T
A 49-year-old male with worsening back and right leg pain was referred for bone scan imaging. Bone scan demonstrated multifocal expansile osteoblastic bony lesions, atypical for benign osseous hemangiomas, which are commonly cold on bone scan. Multisite bone biopsies were compatible with the diagnosis of multifocal osseous hemangiomas. This case illustrates that aggressive osseous hemangiomas, a rare subtype of hemangiomas, may have variable osteoblastic activity on bone scan, ranging from mild to severe uptake.

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Introduction

Intraosseous hemangiomas are relatively common skeletal lesions that typically arise within the vertebral bodies of the thoracic and lumbar spine, though they may arise in other locations [1,2]. While generally asymptomatic, more aggressive lesions may present with symptoms of pain, as well as neurological symptoms related to spinal cord and nerve compression [2]. We present an atypical case of multiple symptomatic biopsy-proven intraosseous hemangiomas of the axial and appendicular skeleton, presenting as "hot" lesions on Tc-99m methylene diphosphonate (MDP) bone scan.

Case report

A 49-year-old man with increasing right leg and back pain was referred to nuclear medicine for bone scan imaging. Previous medical history is notable for L1 and L2 vertebral bony lesions identified on prior outside hospital MRI of the lumbar spine imaging, which were biopsy proven hemangiomas (Fig. 1), and were previously treated with radiation therapy. Tc-99m MDP bone scan was performed (Fig. 2), which demonstrated multiple osteoblastic lesions throughout the axial and appendicular skeleton, atypical in appearance for benign intraosseous hemangiomas. Differential diagnosis includes multifocal malignant or metastatic process, vs multifocal atypical hemangiomas. Computed tomography (CT) of the chest, abdomen, and pelvis (Fig. 3A, B) demonstrates various sclerotic, lytic and mixed lesions throughout the axial and appendicular skeleton, again atypical in appearance for hemangiomas. CT imaging of the right lower extremity (Fig. 3C) demonstrates a large
Lytic lesion in the proximal tibia which was subsequently fixated with intra-medullary rod and screws to prevent a pathological fracture (Fig. 3D). Multisite tissue sampling (including of one the expansile rib, and proximal tibial lesions) was compatible with the diagnosis of benign intraosseous hemangiomas.

**Discussion**

Intraosseous hemangiomas are relatively common, benign vascular lesions that arise within bone. They are most common in middle-aged patients and are slightly more common in females than males [1]. As this patient was a middle-aged male with a known history of vertebral hemangioma, multiple intraosseous hemangiomas were a strong consideration. Hemangiomas are typically found within the vertebral bodies of the thoracic and lumbar spine, though they can arise at virtually any site within the axial or appendicular skeleton [1]. Though usually asymptomatic, patients may present with symptoms, depending upon the location. Symptoms are present in less than 1% of lesions and are generally related to increased bone activity that can lead to spinal cord and nerve compression [2,3]. In this case, as the lesions were particularly aggressive and osteoblastic in nature, the patient presented with increasing back and right leg pain, secondary to active lesion growth. While the lesions are often solitary, cases of multiple intraosseous hemangiomas have been reported, some of which have been extremely aggressive in nature [4,5]. Multiple symptomatic hemangiomas may present a diagnostic conundrum, as differential diagnoses of primary and metastatic bone tumors, as well as multiple myeloma, must be considered.

Intraosseous hemangiomas have a very heterogeneous appearance on a variety of imaging modalities, including on magnetic resonance imaging (MRI), CT and Tc-99m MDP bone scan. On MRI, intraosseous hemangiomas have high T1 (due to high fat content), and high T2 signal (due to high water content), as opposed to metastases which often have low T1 and high T2 signal [6]. On CT, axial imaging of vertebral hemangiomas often produces the classical “polka-dot” sign, with sagittal and coronal reconstructions demonstrating the “corduroy” sign, related to craniocaudally oriented, thickened trabeculae within the lesions [2]. The attenuation of the lesions...
Fig. 3 – (A) Axial contrast enhanced CT of the mid-thorax (bone window) shows an expansile, predominantly lytic lesion in the anterolateral aspect of the right fifth rib (arrow). (B) Contrast-enhanced CT of the upper abdomen (soft tissue window) shows multiple hypodense lesions in the spleen (arrows), likely representing splenic hemangiomas. (C) Coronal CT of the right knee (bone window) shows a large lucent lesion with central sclerosis in the proximal right tibial shaft (arrow). Tissue sampling of this lesion was consistent with benign osseous hemangioma. (D) Frontal radiograph of the right knee demonstrates intramedullary rod and screws fixation of the large lytic lesion within the proximal right tibial shaft (arrow).

may range from fat to soft tissue, and prior studies have suggested that attenuation and relative aggressiveness of the lesions are directly correlated [2,7]. The variable appearance of hemangiomas on CT can make diagnosis difficult. Additionally, while generally regarded to be “cold” appearing on bone scan, hemangiomas have been reported to be “hot”, with increased radiotracer uptake [2,8].

While some features are considered “typical” for intraosseous hemangiomas, the overall variable radiologic appearance can make diagnosis of the lesions difficult. Metastatic disease is an essential consideration in patients presenting with painful lesions that demonstrate increased radiotracer uptake on Tc-99m MDP bone scan, and careful consideration of risk factors is warranted in these situations.
Consideration of rare syndromes presenting with multiple intraosseous hemangiomas, such as skeletal angiomatosis, is also warranted, as these patients have been reported to present with multiple, symptomatic lytic lesions [9]. If multiple skeletal lesions demonstrate increased uptake on bone scan and have an atypical appearance on CT and/or MRI, bone biopsy may be needed to exclude malignancy or confirm the diagnosis of benign intraosseous hemangioma. Further studies are needed regarding the etiology and pathogenesis of aggressive intraosseous hemangiomas to better understand its radiologic appearance.

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