Current Opinion
CO 4 (6), 914– 916 (2024)
Received 21 August 2024 | **Revised 22 September** 2024 | **Accepted 20 October** 2024 | **Online Available 05 November** 2024

https://doi.org/10.52845/currentopinion.v4i6.329

Current Opinion
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OPEN ACCESS

ISSN(O)2795-8639

ORIGINAL ARTICLE

Multifocal Intraosseous Hemangiomas: An Uncommon Site a Case Report

I.Arramach[1,*], F.Lamouime [1], M.Rhaouti [1], M.Lakranbi[1,2], Y.Ouadnouni[1,2], M.Smahi[1,2]

[1]Thoracic Surgery Department, Hassan Ii University Hospital, Fès

[2]Faculty of Medicine, Pharmacy, and Dental Medicine, Sidi Mohamed Ben Abdellah University (Fès)

***Corresponding Author: Ikram Arramach**

Résumé :
Multifocal intraosseous hemangioma, which affects multiple body parts, is rare and comprises about 1% of all bone tumors, most commonly affecting the spine. It is more prevalent in females and typically occurs in the second decade of life. Histologically, hemangiomas are classified into cavernous, capillary, venous, or mixed types. Radiographically, they present as well-defined lytic lesions with characteristic appearances on CT and MRI. Treatment varies based on symptoms and includes surgery, radiotherapy, and curettage, though spontaneous resolution can occur in asymptomatic cases.

Introduction

Intraosseous hemangiomas are benign vascular tumors composed of capillary-like blood vessels of varying calibers. These tumors are most commonly observed in the spine (30%–50%) and skull (80%) but can also be found in the calvarium, sternum, ribs, and long bones [1]. Intraosseous hemangiomas are rarely multifocal, typically presenting as a single lesion within one bone or region. Few reports have described multifocal intraosseous hemangiomas affecting different parts of the body, making diagnosis in such cases very challenging [2]. This case illustrates a multifocal intraosseous hemangioma and underscores the need to consider hemangioma as a differential diagnosis for multiple bone lesions.

Case presentation:

A 64-year-old woman was admitted to the thoracic surgery department with a chief complaint of intermittent sternal pain that is responsive to first-line analgesics. She did not have any obvious past or recent history of physical trauma. The medical histories of the patient and her family were unremarkable. The results of all laboratory studies were within normal reference ranges. The chest and pelvic CT images revealed a well-defined mass of the manubrium with mixed lytic and sclerotic features, with an oval shape, surrounded by peripheral bone sclerosis, without periosteal reaction or cortical erosion. The lesion shows homogeneous enhancement after contrast injection. CT examination of the lumbar vertebrae revealed a "polka-dot" sign with a slight compression fracture of the 10th thoracic vertebra.

On 18F-FDG positron emission tomography/CT (PET/CT), a small, well-defined, grid-like lytic lesion of the manubrium, exhibiting peripheral sclerosis without cortical involvement and no metabolic activity, suggests primarily a hemangioma. Additionally, there are associated angiomatous lesions in the cervical and thoracolumbar spine. Based on the clinical presentation and imaging results, metastatic diseases and multiple myeloma were excluded.

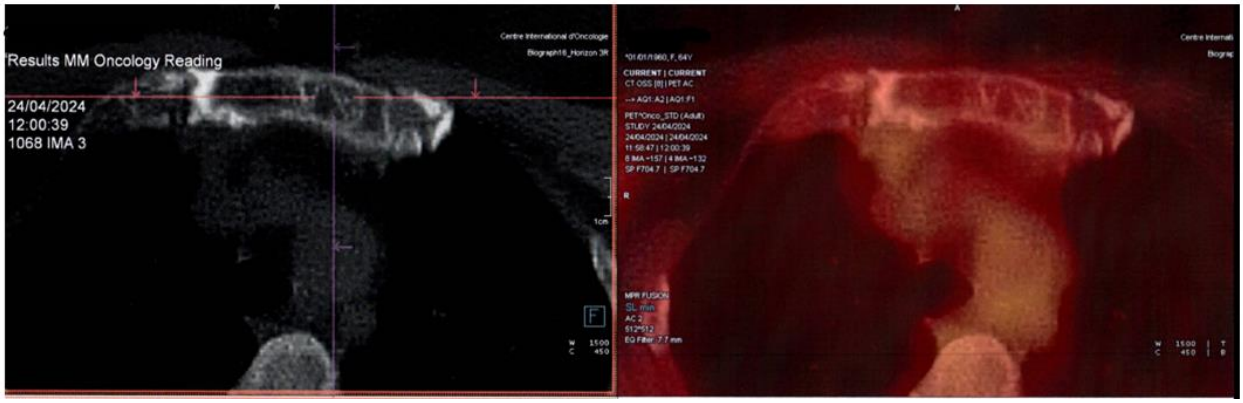

Figure 1: PET scan showing a small, well-defined, grid-like lytic lesion of the manubrium suggests primarily a hemangioma

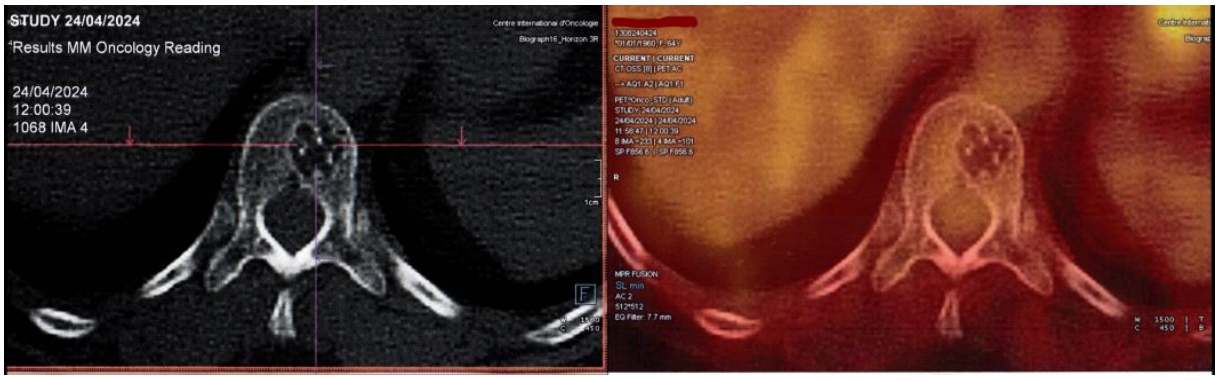

Figure 2: PET scan showing an angiomatous lesion in the thoracolumbar spine.

Considering that the patient is minimally symptomatic and the pain is well-managed with analgesics, we opted for a watchful waiting approach. After one year of follow-up, the patient remains stable.

Discussion:

Intraosseous hemangioma is a rare benign vascular tumor, comprising approximately 1% of all bone tumors and most commonly affecting the spine [3]. Involvement of the sternum has been rarely reported. The incidence is higher in females compared to males, with a female-to-male ratio of 2:1, typically occurring in the second decade of life [4]. Histologically, hemangiomas can be classified as cavernous, capillary, venous, or mixed, based on the type of vascular involvement. Cavernous hemangioma is the most frequently observed type in peripheral bones, representing about half of the reported cases. Capillary hemangioma, though less common, accounts for approximately 10% of all cases according to the literature [5]. The pathogenesis of intraosseous hemangioma remains elusive. Some researchers consider it to be a true neoplasm, while others hypothesize that it results from trauma. Numerous studies have documented cases where patients reported a history of trauma prior to the development of an intraosseous hemangioma [4]. However, in our case, there was no history or predisposition to trauma. The clinical presentation of intraosseous hemangioma is highly variable according to the size, number, and location of the tumor [2]. It is usually asymptomatic; however, large tumors may cause neuropathic pain and dyspnea. Pleural effusion and thoracic outlet syndrome have also been described [6]. A malignant origin should be suspected in cases of rapid growth or invasive radiological features.

The radiographic appearance of hemangiomas is not pathognomonic and can be challenging for clinicians and radiologists to interpret accurately [5]. Hemangiomas may exhibit a characteristic sunburst-like appearance. They generally present as well-defined lytic lesions with a coarse trabecular pattern on plain radiographs. On CT scans, hemangiomas often display a polka dot or honeycomb appearance, resulting from coarse trabeculae interspersed with low-attenuation fat [7]. MRI images of intraosseous hemangiomas

typically show variable signal intensities on T1-weighted sequences and high signal intensities on T2-weighted sequences, due to the presence of water in the stagnant blood within the hemangioma [8]. Previous studies have described PET/CT imaging as an accurate method for the pre-operative staging of bone and soft tissue sarcomas [9]. Hemangiomas are considered metabolically stable benign tumors on PET/CT, with the SUVmax for FDG in 16 hemangiomas, including both soft tissue and osseous tumors, reported to range from 0.73 to 1.67 [10].

The management of intraosseous hemangioma is guided by the symptomatology in each affected region [8]. Therapeutic options include surgery, radiotherapy, curettage, and embolization. Although hemangiomas exhibit responsiveness to radiotherapy, the potential long-term adverse effects, such as malignancy, regional growth impairment, and scarring, make it a less favorable treatment modality [4]. Curettage carries a significant risk of uncontrollable hemorrhage [11]. In many cases, as a bone hemangioma matures, spontaneous degradation may occur, with vascular tissue being gradually replaced by fibrous tissue, leading to self-resolution [8]. Consequently, asymptomatic patients should be managed with vigilant monitoring rather than invasive intervention, as was the case with our patient.

Conclusion:

Multifocal intraosseous hemangiomas affecting different body parts are rare, and involvement of the sternum is an unusual condition that should be included in the differential diagnosis of chest wall tumors. The majority of patients with hemangioma present with asymptomatic and incidental radiographic findings. These lesions are typically non-life-threatening and generally do not require treatment, particularly if they are asymptomatic, with close follow-up being sufficient.

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