Osteoid-Osteoma of the Ilium Associated with Malignant Hypertension in an Adult Female

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Abstract

Osteoid-osteoma, a benign bone tumor, is rare in the ilium bone. Its incidence is uncommon after third decades of life, and can represent a significant diagnostic challenge, resulting in a delay of treatment.

We present a 38 years old lady who presented with history of severe right gluteal pain, more during night, and was relieved with non-steroidal anti-inflammatory drugs. Malignant hypertension was associated co-morbid condition without any underlying cause. Plain radiograph was initially normal and her diagnosis was delayed for more than six months until computed tomography (CT) scans and magnetic resonance imaging (MRI) showed a circumscribed lesion of the right ilium suggestive of Osteiod Osteoma.

Minimally invasive flouroscopy guided curettage was done, and histopathological findings confirmed the diagnosis of osteoid-osteoma. After surgery, the pain was significantly relieved and, surprisingly, the hypertension was controlled, and she was discharged on single antihypertensive drug in maintenance dose.

Key words: iliac bone, gluteal pain, malignant hypertension, Osteoid osteoma

Introduction

Osteoid-osteoma, first described by Jeff [1] is a small, benign osteoblastic tumor consisting of highly vascularised nidus of connective tissue surrounded by sclerotic bone.[1,2] It is commonly distributed in the long bones, such as the femur and tibia, and is more frequently found during second and third decades of life with male predominance.[2,3,4] Clinically, there is severe night pain which gets relieved by salisylate group of non-steroidal anti-inflammatory drugs (NSAIDs) [4,5,6]. Radiographically, the typical finding is a small radiolucent central nidus, usually less than 1.5 cm [7]. Pelvic bone osteoid-osteoma is a rare entity, and typically present with gluteal pain [8]. Radiographic findings and the clinical presentation of pain may lead to a mistaken diagnosis of prolapsed intervertebral disc, sacroilitis or osteomyelitis.[2,3,9,10] En-block surgical excision classically has been the treatment of choice [4,11], but, recently, minimally invasive modalities have begun to supplant surgical management of osteoid-osteoma [9]. There has been no report on associated co-morbid condition like hypertension, with Osteoid Osteoma. We report a very rare case of pelvic bone (Ilium) osteoid-osteoma in relatively older female patient with associated malignant hypertension.

Case History

A 38 years old lady presented to our emergency with complaint of severe pain on right gluteal region for last 2 months. Pain was continuous type and was only slightly relieved with NSAIDS. She had history of on/off pain in same side for last 5 to 6 months and was treated with NSAIDS at other centers without relief. There was no significant past medical or surgical history. She did not have history of trauma or fever. Pain was non-radiating and bilateral lower limb neurology was intact. However, her pain visual analog score (VAS) was 10/10 and she was

bedridden due to pain. On general examination, increase in blood pressure was detected and the hypertension was of malignant type.

The laboratory findings were within normal limits except raised C- reactive protein (CRP). Plain radiograph, antero-posterior view, of the pelvis showed mild sclerosis in right ilium bone at the level of sciatic notch (Figure 1). MRI with T1 weighing demonstrated diffuse low signal intensity in inferior aspect of right iliac vbone, adjacent to the sacro-iliac joint and, high signal on T2 weighted image (Figure 2). CT scan images clearly demonstrated small well defined central lytic lesion (nidus) surrounded by sclerosis (Figure 3).

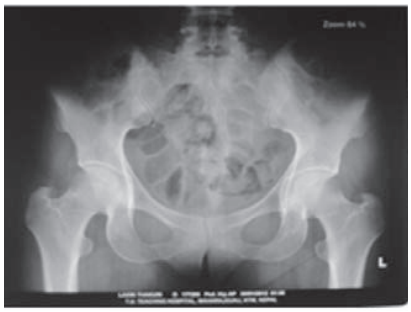

Figure 1: Plain X-Ray Pelvis AP View

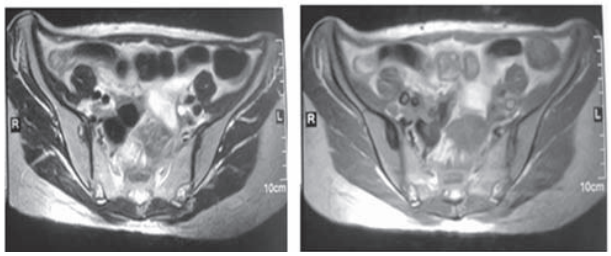

Figure 3: MRI T1 and T2 weighted images of the pelvic bone

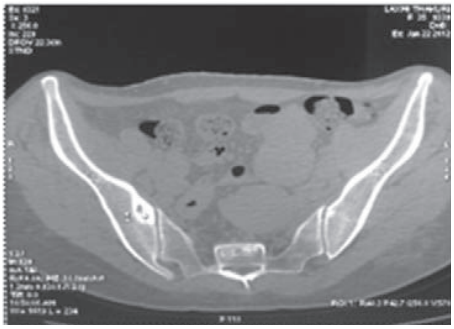

Figure 4: CT Scan of the pelvic bone

The clinical and radiological diagnosis was osteoid-osteoma. Raised blood pressure was controlled by use of combination anti-hypertensive drugs: amlodipine, enalapril and hydrochorthiazide. She underwent percutaneous flouroscopy guided extended curettage. Histopathology revealed circumscribed nidus composed of anastomosing bony trabeculae rimmed focally by osteoblasts. The spaces between the trabeculae show capillary proliferation and marrow fragments consistent with osteoid-osteoma (Figure 5). Her pain significantly relieved within 48 hours of the surgery, with pain VAS of 2/10. Surprisingly, her blood pressure also became normal and she was put on maintenance dose of single anti-hypertensive drug, amlodipine 2.5mg OD on 5th post operative day. After 6 weeks of the surgery she was back to her regular work and activities free of pain. On one year follow up, she was pain free and her blood pressure was under control needing no medication.

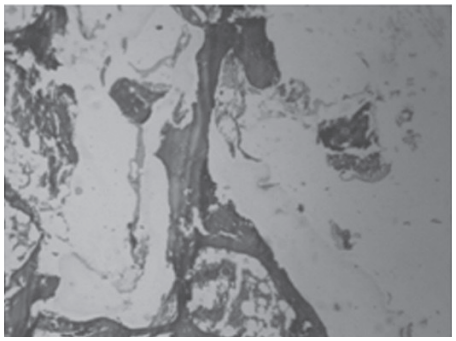

Figure 5: Histopathology finding typical of Osteoid Osteoma

Discussion

Osteoid-osteoma is a painful, small, benign osteoblastic tumor of the bone and is fairly common and accounts for about 10% to 12% of all benign bone tumours. It is relatively rare after 30 years of age and in female.[3] Diaphysis of the long bones, particularly femur and tibia is the most common site. However, it can occur in unusual sites and can cause delay in diagnosis. [12] Pelvic bone is a rare site for Osteoid Osteoma constituting only 2.7% of it.[7,8,9,13]

The clinical symptoms depend on the location of the lesion. The chief complaint is usually mild to moderate pain at the site of the lesion. The duration of symptoms may range from 3months to 2 years before the patients seek medical attention. The pain is localized and may be worse with movement, but sometimes improves with activity. It is more severe at night and typically awakes patient from sleep.[3] The pain may be caused by the presence of nerve fibers in the nidus. Prostaglandin production may contribute to the increased vascular pressure that may stimulate afferent fibers around the nidus that produce pain.[4]

The diagnosis of osteoid-osteoma is often elusive because of variability and unusual location of the tumor or lack

of classic radiographic findings [5,9,11,12]. There is scarce report on pelvic bone osteoid-osteoma, and most have patients younger than 30 years [8,13]. Our case, a lady of 38 years age presented in ER with agonizing back/gluteal pain for last two months, as was suggested [12,14] prolapsed intervertebral disc was the provisional diagnosis with sacroilitis, gluteal abscess as differential diagnosis. Her blood investigations showed no features of inflammations.

Delay in the diagnosis of osteoid-osteoma is not uncommon. The pathognomonic osteoid nidus is not always seen on conventional radiographic imaging, and pain may mimic many other conditions depending on tumor location.[5,10,11] Our case has symptoms for six months but was not diagnosed till she had agonizing pain. Therefore, whenever there is high index of suspicion different imaging modalities has to be done for confirmation of the diagnosis. [3,5,7,11] Among the different modalities of imaging CT scan provides, as in this case also, accurate tumor sizing and localization for percutaneous treatment.

Pain relief is the main goal of the treatment and surgical excision classically has been the treatment of choice [3,5,10] as the symptoms of osteoid osteoma typically resolve only after a prolonged period. Recently, less invasive modalities, including CT-guided excision, arthroscopic excision, cryoablation, and thermoablation with radiofrequency or laser, have begun to supplant surgical management of osteoid-osteoma, resulting in a decrease in the need for definitive surgical treatment .[5,15,16,17]

Malignant Hypertension as a co-morbid condition is not reported in Osteiod Osteoma. It is difficult to suggest malignant hypertension as a cause or effect of Osteiod Osteoma. Our case had malignant hypertension needing triple anti-hypertensive drugs preoperatively. Surprisingly, there was immediate reduction in blood pressure after the surgery suggesting its association. On one year of post operative follow up, cardiologist have discontinued antihypertensive drugs after maintenance dose of single antihypertensive drug.

In summary, our case not only represents a rare presentation of pelvic osteoid osteoma, in a woman older than age 30, but also the first case with associated malignant hypertension.

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