A Foot in the Right Direction: Metatarsal Osteoblastoma-A Rare Case and its Management

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Learning Point of the Article:

Non-vascularised fibular autograft is a reliable method for addressing the defect following excision of tumours in metatarsal bones.

Abstract

Introduction: Osteoblastoma is a rare, benign, bone-forming tumor accounting for <1% of all primary bone tumors. It has a predilection for the posterior elements of the spine and metaphysis and diaphysis of long bones. The occurrence of this tumor in the metatarsal region is rare. We report such the case of a metatarsal osteoblastoma which was treated with wide excision and non-vascularized fibular autograft: a reliable method of reconstruction.

Case Report: A 25-year-old woman presented with progressive pain and swelling over the right foot for 4 years. On examination, there was a gross swelling over the fourth metatarsal region over the dorsum of the foot. Radiographs revealed a osteoblastic lesion of the fourth metatarsal bone expanding into the intermetatarsal region. Magnetic resonance imaging (MRI) revealed an expansile altered signal intensity lesion which was hypointense on both T1 and T2 – weighted images with no soft-tissue component. With a working diagnosis of locally aggressive bone-forming tumor, she underwent wide excision of the tumor with reconstruction using a non-vascularized fibular autograft. Intraoperative samples sent for histopathological examination confirmed the diagnosis of osteoblastoma. After 2 years of follow-up, the patient is able to weight bear with no pain and imaging shows graft incorporation with no signs of recurrence.

Conclusion: Osteoblastoma of the metatarsal region can present a diagnostic conundrum to the treating clinician due to its rare nature. Proper evaluation and reconstruction at an early stage with wide excision and reconstruction with non-vascularized fibular autograft are a reliable treatment option.

Keywords: Osteoblastoma, metatarsal, fibular autograft.

Introduction

Osteoblastoma is a rare, benign, bone-forming tumor accounting for <1% of all primary bone tumors and 3.5% of all benign neoplasms of bone [1]. The neoplasm generally occurs in the second and third decades of life with preponderance for occurrence to the posterior elements of the spine and on the surfaces of long bones [2]. The tumor has a male preponderance (2:1). Osteoid osteoma and benign osteoblastoma have very

similar histological pictures presenting a diagnostic conundrum as they are different expressions of one pathological process. The radiological appearance is variable and differential diagnoses include: osteoid osteoma, aneurysmal bone cyst, infection, osteosarcoma, and metastasis [3]. The ankle and foot region even though being a common site for this tumor occurrence, the metatarsal region is rare with only few such previous case reports.

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Figure 1 AP and oblique radiographs of the right foot showing a well-defined eccentric lesion over the fourth metatarsal region.



Figure 2: (a-c) Computed tomography 3D reconstruction showing a well-defined, exophytic, and dense sclerotic lesion with osteoid matrix arising from the fourth metatarsal.

Case Report

A 25-year-old woman presented to the OPD with complaints of swelling and progressive pain over the right foot for the past 4 years. The swelling was initially small in size and gradually progressed to attain the present size and was associated with pain for the past 6 months on exertion like cycling. No history of previous trauma to the same site. There was no fever, chills, recent weight loss, or loss of appetite.

On examination, she had a gross swelling over the fourth metatarsal area with an overlying healed scar with minimal tenderness. There were no sinuses or discharge. The patient had pain while weight bearing on the right foot. The distal neurovascular status was intact.

The patient was initially treated at another hospital where biopsy of the swelling was done and histopathological examination revealed atypical osteoid proliferation. The working diagnosis was low-grade osteosarcoma/osteoid osteoma/osteoblastoma.

The initial baseline investigations were within normal limits. The radiograph of the right foot revealed a well-defined eccentric lesion with a homogeneous matrix over the distal two-thirds of the fourth metatarsal bone expanding into the intermetatarsal region (Fig. 1).

The CT revealed a well-defined, exophytic, lobulated, and dense sclerotic lesion with an osteoid matrix of size $4.6 \times 3.2 \times 3.6$ cm (AP × TR × CC) arising from the medial aspect of the distal shaft of the right fourth metatarsal bone (Fig. 2a,b,c). The lesion extends circumferentially around the shaft causing mild thinning of the shaft of third metatarsal.

The MRI revealed an expansile altered signal intensity lesion involving the fourth metatarsal bone appearing hypointense on both T1 and T2 weighted images with low-signal intensity internal areas with no associated soft-tissue component (Fig. 3a, b, c).

Based on clinical presentation and radiographic findings, the diagnosis of a locally aggressive bone-forming tumor was considered. The patient was planned for surgical intervention after obtaining the required consent.

Under general anesthesia, the tourniquet was inflated over the thigh to 220 mm Hg pressure. Through the dorsal approach, skin and soft tissue over the fourth metatarsal area was dissected. The tumor was carefully removed with a cuff of normal tissue at both ends. Intraoperative samples were sent for bone tissue culture and histopathological examination (Fig. 4). Non-vascularized fibular graft was harvested from the ipsilateral side to bridge the defect caused by tumor excision. The fibular graft was fixed with K-wire to the proximal phalanx and to the base of the fourth metatarsal. A below-knee cast was applied for 6-weeks. After 6 weeks, the patient underwent K-wire exit and was started on partial weight-bearing mobilization with the help of walker support.

The bacterial growth in the tissue samples provided for culture was negative. The diagnosis of osteoblastoma was supported by a histopathological examination of the excised tumor, which revealed a osteo-proliferative lesion with osteoblastic rimming and osteoid deposition (Fig. 5).

The patient was under follow-up for a total of 2 years, during which time she had no symptoms. On follow-up radiographs (Fig. 6), there were no signs of recurrence and the fibular graft had united to the metatarsal base.

Discussion

Jaffey and Mayer in their foundational paper in 1932 described an osteoblastic osteoid tissue-forming tumor of the metacarpal in which they described osteoblastoma as an osteoblastic tumor that possessed a great capacity to produce osteoid tissue [4]. It was termed as Jaffey–Mayer tumor of the metacarpal bone. After its first description, the tumor was described in many



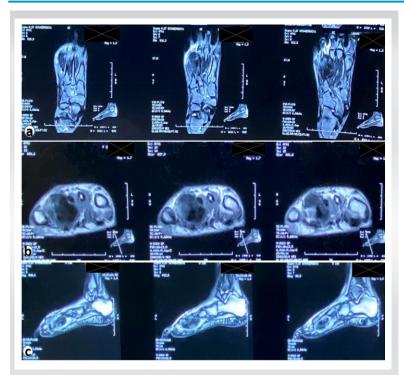




Figure 3: (a-c) Coronal, axial, and sagittal sections of magnetic resonance imaging of the left foot showing expansile hypointense lesion in T1 and T2 weighted imaging.

Figure 4: Gross appearance of the excised tumor.

papers under different terminology including "osteogenic fibroma", "giant osteoid osteoma" putting forward the abundance of fibrous stromal elements and striking similarity histologically to osteoid osteoma, respectively. In 1956, the currently accepted terminology of benign osteoblastoma was proposed by Jaffe and Lichtenstein independently in different papers highlighting the osteoblastic origin and relatively nonaggressive course when compared to osteosarcoma [5].

Osteoblastomas have been reported in almost all bones with a predilection for flat and membranous bones. The posterior elements of the spine were the most common site of tumor occurrence, followed by long bones, jaw, and foot and ankle region. Talus is the most common site of osteoblastoma in the foot and ankle region [6]. The cardinal symptom with which the patients presented to the OPD was gradually progressive pain. Localized swelling was another common complaint when the swelling was close to the surface. Pain relief obtained with aspirin and nocturnal aggravation of pain which is a characteristic feature of osteoid osteoma was not present in majority of patients.

Tumor size at presentation was quite variable depending on the duration of swelling but majority of the swellings were >2 cm. Osteoblastoma has the tendency for enlargement and progression and has been at times reported to be locally aggressive and has the potential to undergo malignant change. Recurrence – persistence rate of 9.8% has been observed among osteoblastomas following curettage and marginal excisions [7].

Radiologically, the neoplasm on plain radiograph presents with a mineralized matrix, reactive sclerosis, and abundant periosteal new bone formation. Younger lesions tend to be radiolucent, whereas they tend to ossify as they become mature [2]. Furthermore, osteoblastomas generally lack the halo of sclerotic bone which is generally associated with osteoid osteoma this is attributed to the fact that osteoblastomas which occur in cancellous bone and osteoid osteoma which occur in cortical bone respond differently to the same pathologic process. At presentation, more than 30% of osteoblastomas have a radiologic appearance that is non-specific in pointing to a diagnosis.

Histopathological picture of osteoid osteoma and osteoblastoma is strikingly similar with only minimal differences being observed in multiple studies [8, 9]. Quantitatively, stroma is present in a greater amount and osteoid to a lesser amount when compared to osteoid osteoma. Osteoblast lining, giant cells, and capillaries showed no significant difference. Qualitative comparison showed that the stroma of osteoblasts always showed active proliferation and osteoblastic differentiation. McLeod et al. [2] had arbitrarily proposed 1.5 cm as the cutoff for differentiating between osteoid osteomas and osteoblastomas. Size, radiographic appearance, and cytologic pattern will throw light on the diagnosis of osteoblastoma when in doubt.

Most of these lesions can be successfully treated with wide resection including a cuff of normal tissue. Jackson et al. have



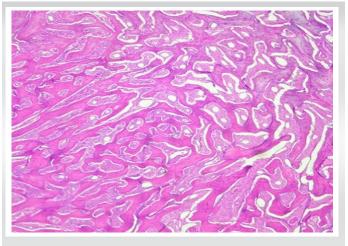


Figure 5: H and E stained section of the excised specimen.

reported that there have been no recurrences following en bloc resections [7]. The need to address the defect following resection in appendicular skeleton, especially in the foot and ankle weight-bearing region can be addressed with the help of fibular autograft. Hammad et al. report a case of first metatarsal osteoblastoma which was treated with non-vascularized fibular autograft [10]. However, in that case, the tumor had involved the metatarsal head causing osteoarthritic changes in the metacarpophalangeal joint. But in our case, even though the tumor was diaphyseal we were unable to preserve the head. Follow-up of 2 years in our case showed no residual pain, swelling, or recurrence.

Conclusion



Figure 6: 2-year follow-up radiograph of the right foot.

Osteoblastoma in the metatarsal bones is quite rare. While evaluating the patient clinically and radiologically, there are numerous differential diagnoses to be taken into account. The mainstay in resolving this diagnostic conundrum is histopathological analysis of the excised tissue. A dependable therapeutic strategy is a wide excision followed by reconstruction with non-vascularized fibular autograft – a foot in the right direction.

Clinical Message

Foot swellings that progress over time are often overlooked by patients unless they cause severe pain. The clinician while evaluating must go all-in to diagnose and treat the swelling using the armamentarium at his disposal. Prompt diagnosis, treatment, and reconstruction with reliable methods will ensure the patient's ability to walk without pain.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given the consent for his/ her images and other clinical information to be reported in the journal. The patient understands that his/ her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict of interest: Nil Source of support: None

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Consent: The authors confirm that informed consent was obtained from the patient for publication of this case report

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