CASE REPORT

Multifocal osteoid osteoma of tibia

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SUMMARY

Multifocal osteoid osteoma of the bone is extremely rare. We report a 25-year-old man who presented with pain in the left leg since 11 months which was partially relieved by over-the-counter analgesics. Radiograph demonstrated two lytic lesions with surrounding sclerosis along the anterior cortex of the left tibia. Three-phase Tc 99m bone and CT scans confirmed the diagnosis of multifocal osteoid osteoma. The patient underwent surgical excision followed by protected weight bearing. The patient was asymptomatic at 6 months postoperatively. Multifocal osteoid osteoma needs to be considered in the differential diagnosis of multiple lytic lesions in the bone.

BACKGROUND

Osteoid osteoma is a benign bone-forming neoplasm which was first characterised by Henry L Jaffe.¹ It predominantly occurs in the first two decades of life.² Majority of the patients experience pain at the involved site, which is worse at night and is relieved by non-steroidal anti-inflammatory drugs (NSAIDs).² Some patients can present with growth disturbance, swelling or scoliosis.³ Diagnosing osteoid osteoma needs a high index of suspicion as small lesions can be easily missed on a plain radiograph. Furthermore, it is difficult to localise the exact site of pain in the paediatric age group. Advances in imaging modalities, such as bone scan and CT, have helped in early identification of these lesions. A classical osteoid osteoma would be a solitary, intracortical lesion usually occurring in long tubular bones with a significant cortical thickening.³ Multifocal osteoid osteomas are rare but these lesions do occur and their early recognition is important to guide an appropriate management.

CASE PRESENTATION

A 25-year-old male patient presented with a history of pain in the left leg for the past 11 months. The pain was insidious in onset, intermittent, occurred predominantly at night, progressively increasing, 6/10 on a visual analogue scale and was present over the anterior aspect of the left leg. It was non-radiating and there were no aggravating factors for pain. However, he got some relief with over the counter analgesics. There was no history of antecedent trauma and the remaining history and general examination did not reveal any significant abnormalities.

Local examination revealed diffuse tenderness over the anterior aspect of the left leg. There was no palpable swelling or wasting of the leg muscles.

The range of motion of the left knee and ankle joints was normal and there was no neurovascular deficit distally.

INVESTIGATIONS

Plain radiograph revealed two lytic bone lesions with surrounding sclerosis along the anterior tibial cortex (figure 1). The larger lesion was present at the junction of the upper and middle one-third and the smaller at the middle one-third of tibia. A three-phase Tc 99m bone scan was performed. In the skeletal phase, two focal areas of increased tracer uptake were noted with the distal lesion less intense than the proximal one (figure 2). The central nidus and the surrounding sclerosis of the bony lesions were apparent on the CT scan (figure 3). A diagnosis of a multifocal osteoid osteoma was made. Radiofrequency ablation (RFA) is the most favoured modality for treating osteoid osteomas at our institute.⁴ However, we had no experience in treating multifocal osteoid osteomas with RFA. The patient was explained regarding various treatment options and he opted for surgical excision.

Figure 1 Anteroposterior and lateral radiograph of the left leg of a 25-year-old man showing two lytic bone lesions with surrounding sclerosis along the anterior tibial cortex.

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To cite: Sampath Kumar V, Khan SA, Palaniswamy A, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2013-201712

Figure 2 Three-phase bone scan of the bilateral legs in the skeletal phase showing two focal areas of increased uptake with the proximal lesion showing more intense uptake than the distal one.

DIFFERENTIAL DIAGNOSIS

- ▶ Multiple myeloma
- ▶ Metastatic deposits in the bone
- ▶ Multifocal osteoid osteoma

Figure 3 Sagittal CT image of the left tibia depicting two cortical lytic lesions with surrounding sclerosis.

TREATMENT

The patient underwent surgical en bloc excision as a day care procedure. A shark bite excision of cortical lesions along with the surrounding sclerotic bone was performed under image guidance and the specimens were sent for histopathological examination (figure 4).

OUTCOME AND FOLLOW-UP

The histopathology was consistent with the diagnosis of osteoid osteoma for both the lesions. The patient was kept non-weight bearing for 6 weeks, followed by partial weight bearing mobilisation as tolerated gradually progressing to complete weight bearing. At the 6-month postsurgery, the patient was completely asymptomatic and the follow-up radiographs revealed a well-healed excision site and no evidence of recurrence.

DISCUSSION

Osteoid osteoma is a benign bone tumour of unknown aetiology. The central lytic area of osteoid osteoma is called the nidus around which the sclerotic bone is laid.³ 'Multifocal' osteoid osteoma refers to the presence of more than one lesion within the same bone whereas 'synchronous multicentric' osteoid osteoma denotes the simultaneous presence of osteoid osteoma in two different bones.⁵ When a single lesion had more than one nidus, it is termed as an 'osteoid osteoma with multicentric nidus'.⁶ The lesion is painful due to the high level of prostaglandin production, particularly PGE2 and PGI2. Mungo *et al*⁷ studied the cyclooxygenase (COX) expression in osteoid osteoma specimens and showed that these tumours predominantly express COX-2. NSAIDs relieve pain by inhibiting this prostaglandin production.

Spontaneous regression of the osteoid osteoma has been described.³ However, patients need to take long-term analgesics which may cause systemic side effects. CT guided radiofrequency ablation has an established role in the treatment of osteoid osteoma at accessible sites and the success rate varies between 76% and 100%.^{8,9} Surgical excision aided by

Figure 4 Postoperative anteroposterior and lateral radiographs of the left leg following wide excision of both lesions.

intraoperative fluoroscopy is the gold standard in the treatment. Besides en bloc excision, a burr-down technique has been described where the sclerotic cortex is burred up to the nidus, the cavity curetted and the wall is again burred thoroughly.³ Localising smaller lesions can be challenging, and intraoperative nuclear imaging has been described for the same.¹⁰ Surgical excision carries the risk of pathological fracture as the cortical defect acts as a stress riser. Multifocal osteoid osteomas can weaken the bone further and the limb needs to be protected for a longer duration. It was for this reason that our patient was kept under non-weight bearing mobilisation protocol for 6 weeks.

The differential diagnoses for multifocal lytic lesions in the bone include Langerhans cell histiocytosis, multiple myeloma and leukemic and metastatic deposits. Benign bone tumours/tumour-like lesions that are known to occur at multiple sites include fibrous dysplasia, enchondroma, osteochondroma, haemangiomatosis and fibromatosis. Multifocal osteoid osteoma is extremely rare. Three cases were reported in the literature, one each in the humerus, proximal femur and fifth lumbar vertebra.^{2 11 12} Synchronous multicentric osteoid osteomas have been reported in three patients.^{5 13 14} In paediatric population, this lesion needs to be differentiated from Langerhans cell histiocytosis. Zmurko *et al*¹⁵ reported a case of solitary osteoid

osteoma of femur that progressed to a lesion with multicentric nidus at 1 year.

To the best of our knowledge, this is the first report of multifocal osteoid osteoma of the tibia. Our case report highlights the importance of considering a diagnosis of multifocal osteoid osteoma when dealing with multifocal lytic lesions of the bone. It also emphasises the need to evaluate these lesions with higher modalities such as CT and Tc 99m bone scans.

Contributors SR, SAK, VSK and AP were part of the operating team that performed surgery. VSK authored the manuscript under the guidance of coauthors. AP did the preoperative workup of the patient.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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Learning points

- Osteoid osteoma needs to be considered in the differential diagnosis of multifocal lytic lesions of the bone.
- Whenever a solitary osteoid osteoma is diagnosed, we recommend radiological examination of the entire length of the bone to look for other lesions.
- Failure to recognise the second lesion may result in persistent pain despite adequate treatment of the diagnosed osteoid osteoma.
- Additional investigative modalities such as three-phase bone scan may be necessary to rule out other aetiologies of multifocal lytic lesions.

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