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Periosteal Osteosarcoma of the Second Toe with Bone Marrow Involvement: A Rare Case

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Abstract: ***Periosteal Osteosarcoma is a variant that arises from the periosteal surface of bone without evidence of medullary involvement which exhibits distinct radiographic, and radiological features and biological behaviors that are different from the conventional Osteosarcoma. The treatment usually requires aggressive surgery with clear margins. We present a case of a 58-year-old man who presented with painful swelling of the second toe right foot. Clinico-radiological evaluation showed it as an osteolytic lesion following which underwent Ray amputation of second toe. Periosteal osteosarcoma usually affects young individuals in their 2nd and 3rd decade of life with predominantly involving long bones but is rare in small bones. Diagnosis requires a high index of suspicion for early management and better outcome of the patient.***

Keywords: periosteal osteosarcoma, bone, phalanx, MRI, foot

1. Introduction

Osteosarcoma (OS) is the most common primary malignancy of bone accounting for approximately 35% of all primary malignant bone tumors[1]. OS is predominantly a disease of the young, with classic presentations often involving long bones. Periosteal osteosarcoma (PO) is a variant that arises from the periosteal surface of bone without evidence of medullary involvement which exhibits distinct radiographic, and radiological features and biological behaviors that are different from the conventional OS[2]. As conventional OS, PO also affects young individuals in their 2nd and 3rd decade of life with predominantly involving long bones, tibia 40 %, femur 38% followed by ulna and humerus (5-10 %). However, PO can occur in older populations but is rarer in small bones, specifically the toes. Recognizing and diagnosing these tumors in atypical locations requires a high index of suspicion and a thorough understanding of their clinical presentation and radiologic characteristics.
We report a rare presentation of PO second toe in a middle-aged male.

2. Case Summary

A 58-year-old man came to us with a painful swelling in his second toe. The swelling had developed gradually over the past two months without any clear injury or trauma. He has diabetes, which is well-managed with oral medications. He does not give any history of fever, weight loss, decreased appetite or any other bony pain. He has no positive history of any previous malignancy or positive familial history.

On local examination, his right second toe appeared generally swollen. It was associated with mild pain but there was no increased warmth, open sores, or other visible signs of infection. Movements at IP (Interphalangeal) jts were painful but MTP jt (Metatarsophalangeal) was full and free with no distal neurovascular deficit.

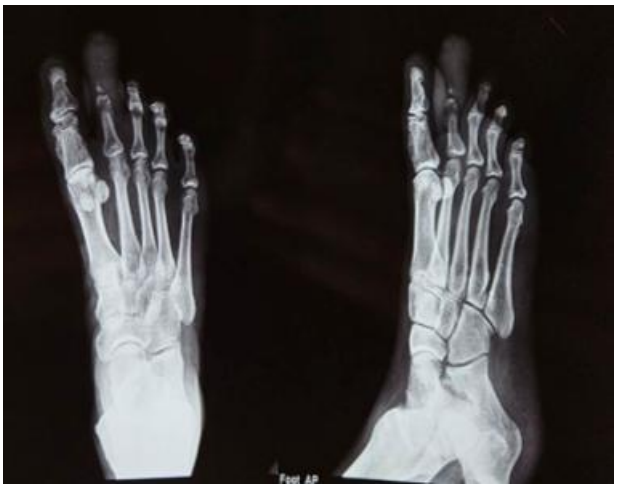

Figure 1: X-ray of the affected foot with osteolytic lesion involving the second toe.

An X-ray (Figure 1) showed bone loss (osteolysis) in the middle and end bones (phalanx) of the toe, while the joint connecting the toe to the foot (MTP jt) appeared normal.

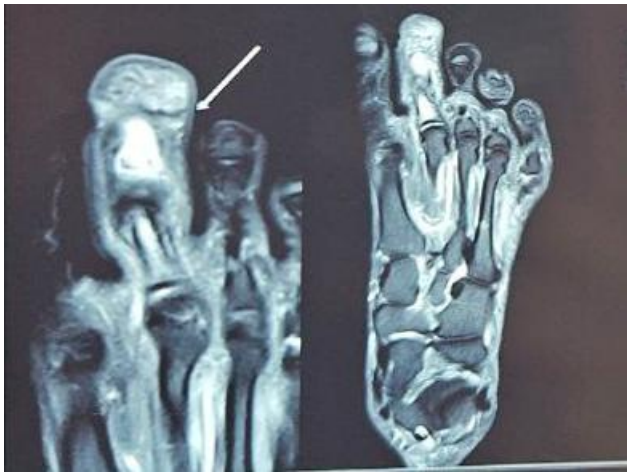

Figure 2: Pre-operative MRI of the affected foot shows an irregular, expansile osteolytic lesion involving the second toe.

Further imaging with an MRI (Figure 2) confirmed the presence of a destructive bone lesion centered in the middle phalanx. It extended into the joints at both ends of the toe (the proximal and distal IP joints), but again, the joint connecting the toe to the foot was not affected.

The patient was planned for surgery after obtaining informed consent and a pre-operative check-up. Second toe ray amputation was done and the specimen was sent for HPE. The postoperative period was uneventful. On gross examination, cut sections showed yellowish creamy colored hard areas that were eroding the bone. Bone was cartilaginous, discontinuous and easy to cut through. The overlying skin was intact with no ulceration.

Microscopic examination (Figure 3A and B) showed a tumor composed of atypical hyaline cartilage at the periphery, having myxoid area focally. The osseous component showed an intermediate-grade OS intermixed with a cartilaginous component.

3. Discussion

We report a case of a 58-year-old diabetic man with painful swelling over the second Toe of his right foot, which on radiological examination depicted an osteolytic lesion of the second Toe involving PIP and DIP joints but sparing MTP joint and other bones. The patient was managed with second Toe Ray amputation followed by histopathological examination (HPE) of the specimen. HPE suggested an intermediate-grade PO with clear margins. The post-operative period was uneventful, and the patient is currently under follow-up.

Three types of surface OS have been documented in the literature: Parosteal, Periosteal, and High-grade surface OS3. PO are described as intermediate-grade OS arising from the surface of bone. Though Ewing recognized PO for the first time in 1939, Unni et al coined the term “periosteal osteogenic osteosarcoma” and described its clinicopathological features145. Common locations for PO are around the knee6.

PO generally has a better prognosis than other forms of OS7. The prognosis of PO is intermediate between parosteal and high-grade surface OS and is superior to conventional OS. Recent long-term outcome data indicate that recurrence with progression to metastatic disease typically occurs within the first three years after presentation, suggesting that long-term disease-free survival is possible.

We treated this case with a Ray amputation of the second Toe with excision of the head of the metatarsal as the lesion had medullary involvement. After the diagnosis was confirmed, an oncologist consultation was obtained, and no adjuvant chemotherapy or radiotherapy was administered. There are not many reported cases of PO of small bones of the foot, and from our experience with this case, we believe this rare tumor involving small bones of the foot can be managed with wide surgical excision and follow-up.

4. Conclusion

PO is a rare tumor that involve the small bones of the foot. The presence of only a few reported cases in the literature underlines the importance of further investigation into this topic. In conclusion, each case should be given special consideration for multiple factors to prevent recurrence. Overall treatment depends on the surgeon’s experience and the clinical situation, but further research is needed into this rare tumor.

Conflict of interest:
The authors declare that we have no conflicts of interest.

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Patient consent for publication: Consent taken

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