

INTRAOSSIOUS LESION OF PHALANX: TWO SIMILAR CASES BUT NOT THE SAME

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Abstract

Intraosseous lesions at phalanges are rare. They frequently present with pain and swelling. Fortunately, the majority of the lesions are benign. However, some lesions are destructive and early interventions are required. We report two cases of similar presentations of swelling and discomfort at the little finger for six months. The lytic lesions involved the whole middle phalanx with cortical breach sparing the joints. A provisional diagnosis of osteomyelitis was made, although unlikely. Bone biopsy was performed early to plan for definitive treatment and surgery. Patient 1 was diagnosed for intraosseous gout whereas Patient 2 for epithelioid hemangioma. Both were benign destructive bone lesions. Thus, we counselled the patients for curettage of lesion, bone grafting and spanning external fixation in view of extensive lesion. Patient 1 defaulted treatment. Patient 2 had an uneventful surgery. She regained her grip strength. In two years follow up, there was no evidence of infection, recurrence or malignant transformation.

Keywords: Gout, Epithelioid Hemangioma, Osteomyelitis of Phalanges, Benign Bone Lesions

Introduction

Swellings in hands are commonly benign. Common intraosseous lesions are enchondroma, giant cell tumour and osteomyelitis. Eventhough benign, some lesions could be aggressive and warrants for intervention. We report two rare cases of similar presentations of swelling and discomfort at little finger for six months duration.

Cases

Patient 1 is a 54-year-old retired male journalist, left hand dominant. Patient 2 is a 77-year-old female, home maker, right hand dominant. Both patients had swelling and discomfort at the little finger of their respective dominant hand. The pain and swelling had progressively worsened over six months, hence explaining the need for medical attention. They did not present with any history of trauma nor other systemic illness. On examination, there was a circumferential fusiform swelling at the middle phalanx region sparing both distal and proximal interphalangeal joints (Figure 1, 2). It was slightly warm and tender. Motion at the proximal interphalangeal joint (PIPJ) was restricted;

10° fixed flexion deformity with 70° flexion limited by swelling.



Figure 1: Clinical photo and radiograph anteroposterior view of Patient 1



Figure 2: Clinical photo and radiograph anteroposterior view of Patient 2

Radiographs showed similar findings of an expansile lytic lesion over the middle phalanx with cortical breach preserving the joint (Figure 1, 2). Osteomyelitis was initially suspected but in the absence of previous bony injury, a biopsy was performed to confirm the diagnosis. Bone biopsy was performed for both patients with similar technique. An incision (1 cm) was made over mid-dorsum of the middle phalanx and a core needle biopsy sample was taken distal to the central slip insertion. However, the results were not the same. Patient 1 was diagnosed with intraosseous gout (Figure 1) and patient 2 with epithelioid hemangioma (Figure 2, 3).

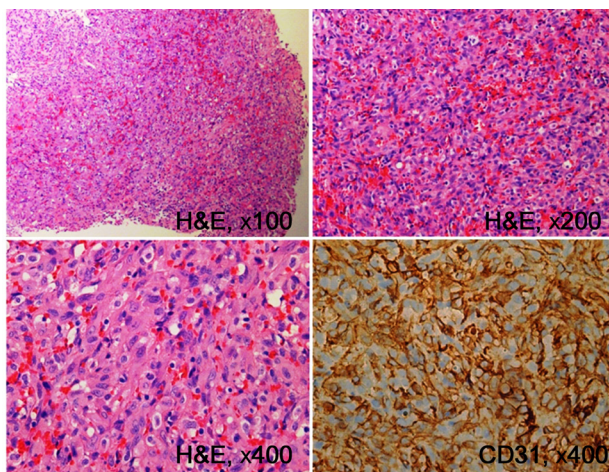


Figure 3: Lesion appears vascular in nature with many red blood cells seen (H&E, x100), many congested vascular spaces/ channels lined by plump endothelial cells which appear epithelioid (H&E, x200), plump epithelioid endothelial cells with no increase in mitosis and small inconspicuous nucleoli (H&E, x400), plump endothelial cells which appear epithelioid stain positive (CD31, x400)

Both patients were counselled for curettage, bone grafting (synthetic) and external fixation of phalanges (spanning the middle phalanx). However, patient 1 defaulted treatment. In patient 2, surgery was uneventful (Figure 4). External fixator was removed after two months and physiotherapy commenced for improvement of joint movements.

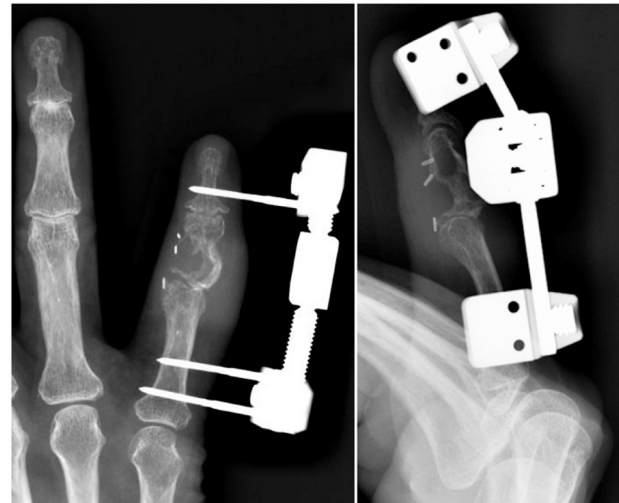


Figure 4: Cross PIPJ external fixator to maintain stability of the bone post-surgery

Six month post-surgery, she regained her grip strength (MRC 4+). Range of motion at PIPJ was 10° to 70°; similar to pre-operative findings. Radiographs at 18 months follow up showed consolidation of more than 50% of the intraosseous lesion (Figure 5). There was no evidence of infection, recurrence or malignant transformation at two years follow-up.



Figure 5: Post-operative 18-months radiographs showing more than 50% of consolidation

Discussion

Isolated intraosseous gouty tophus and epithelioid hemangioma at phalanges are rare (1-7) and aggressive (1, 3-5). They mimic other common lytic lesions of phalanx clinically and radiographically (1-9).

A suspicion of intraosseous gout can be made if patient was diagnosed for gout or hyperuricemia previously (2, 4, 8). About 10% of patients with gout presents with tophi (8) and common depositions are at articular cartilage, subarticular region, tendon sheath and bursae (2). In some cases, intraosseous lesion could be the first manifestation (1, 6) as seen in our case. Further investigations frequently revealed hyperuricemia. Patient 1 presented with neither gouty attacks nor systemic disorders. He presented with merely discomfort not pain at lesion as described by other authors (1, 2, 4, 6, 8). His lesion involved the whole shaft of middle phalanx sparing the joints.

Similarly, patient 2 also presented with discomfort rather than pain as described by other authors (3, 5, 7, 9). In epithelioid hemangioma, usually patients present with skin and subcutaneous lesions (5, 9). Patients who had lesions specifically at phalanges, mostly presented with multiple foci (3, 5, 7). In our case, both entities were not found. Dannaker *et al.* reported that their patient had history of trauma and history of exposure to polymerized vinyl chloride which could be the precipitating factors (5). In our patient, there was neither history of trauma nor chemical exposure.

Thus, we found that a clinical diagnosis was impossible for both cases. Radiological imaging were similar in both cases. None of the authors were able to diagnose intraosseous gout and epithelioid hemangioma after radiographs and magnetic resonance imaging (MRI) of the lesions. Generally, all authors agreed that there were no radiographic feature which was pathognomonic and they mimic tumor or infection (1-9).

Diagnoses in all patients were established intraoperatively. In intraosseous gout, the lesions were found to be chalky white (1, 4, 8) or plaque-like (2) whereas in epithelioid hemangioma the features were like dark brown jelly (9) or raspberry (3). Final diagnoses were confirmed by histopathological examination (1-9). Therefore, biopsy is crucial in planning definitive treatment in both cases.

Most studies suggested excision or curettage to reduce the urate load followed by anti-gout medications (1-4) and to prevent further destruction (6-8). In a sesamoid bone gouty tophus, complete resection of the bone is possible (4) where less morbidity is expected. Lamovec *et al.* reported two cases of epithelioid hemangioma at the distal phalanx and another in the distal half metatarsal which they amputated in view of multiple foci in the same digit (3). Amputation is a fair option in cases of extensive intraosseous lesions with joint destructions. Dannaker *et al.* opted for radiation therapy in view of multiple skin lesions with multiple bone involvement (5).

In intraosseous lesions of the long bones and phalanges, grafting could provide structural stability and promote healing (2, 3, 6-8). Patients presenting with extensive lesions, consequently suffer pathological fractures (2). In Patient 2, we augmented the middle phalanx with a spanning external fixator as there was a cortical breach and an impending fracture. Most studies reported that there were no recurrence after surgery in two to six years follow up (1-4, 6, 7, 9).

Conclusion

In patients presenting with the similar complaint of sudden onset digital swelling with bone destruction, different diagnoses of intraosseous gout and epithelioid hemangioma should be considered. Curettage with bone grafting is the treatment of choice in intraosseous gout and epithelioid hemangioma of the phalanges. External fixations are recommended in extensive lesions with or without fracture as it provides additional stability.

Competing interests

The authors declare that they have no competing interests.

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