

Case Report

A Lytic Lesion in Proximal Phalanx of Hand: A Case Report and Diagnostic Approach

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Keywords

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Abstract

Lytic lesions arising in the hand can be confused with an enchondroma. Enchondroma is the most common tumor of the hand and can present with varied features. It often requires only observation. A dilemma arises when surgically treatable lesions like aneurysmal bone cyst (ABC) present in uncommon locations like the hand. To diagnose a lytic lesion in the hand, percutaneous biopsy is commonly done. But, percutaneous biopsy is unnecessary in enchondroma and may not be useful in conditions like simple bone cyst and ABC. In such situations, magnetic resonance imaging (MRI) can differentiate between the most frequent benign lesions of the hand thereby reducing the need for invasive procedures. We present a 25-year-old lady who presented with a painless right index finger swelling for the past 6 months. Radiographs revealed a lytic expansile lesion in the proximal phalanx of the hand. MRI showed multiple fluid-fluid levels. Curettage and autologous iliac crest bone grafting was done. Histopathology confirmed the diagnosis of an ABC. The patient was followed up for 12 months without any recurrence. We briefly review the paucity of literature on the diagnostic approach to benign lytic lesions of the hand.

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Introduction

Tumors of the hand are mostly benign painless lesions. Enchondromas are the most common primary bone tumors of the hand, comprising up to 70–90% of cases [1, 2]. Aneurysmal bone cysts (ABCs) are commonly found in long bones [3]. Atypical presentation

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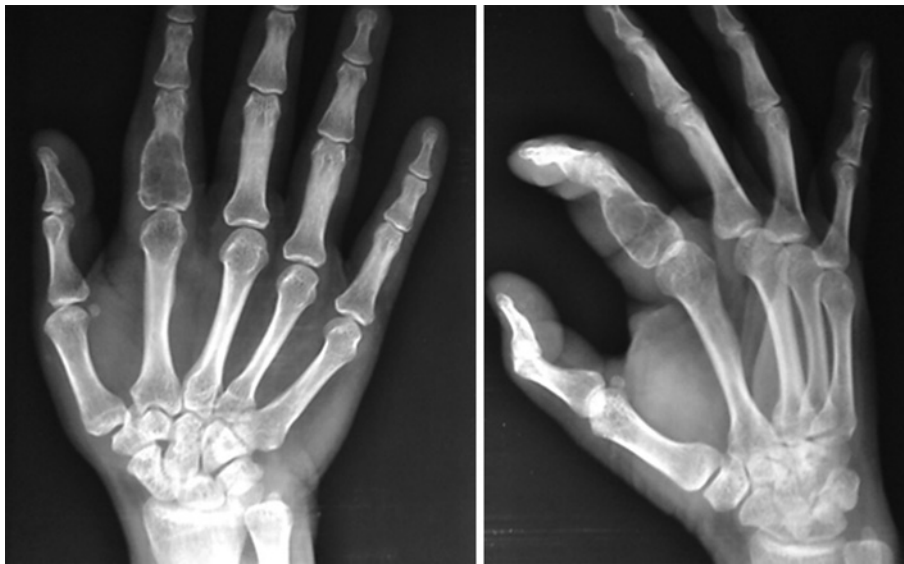


Fig. 1. Expansile lytic lesion of proximal phalanx of right index finger can be noted.

in hand may mimic other bone lesions of the hand like enchondroma [1]. Other bone tumors and tumor-like lesions in the hand include simple bone cyst (SBC), giant cell tumor (GCT), fibrous tumors, and vascular tumors [2]. Some of these benign tumors can have similar clinical and radiological presentations. However, the different natural courses of the diseases make an accurate diagnosis essential. A percutaneous biopsy can be done to diagnose the lesions, but they are invasive. Magnetic resonance imaging (MRI) can differentiate between the common benign tumors of the hand, but it is not definitive. A diagnostic approach to lytic lesions of the hand is lacking in the literature. We report a case of ABC in an uncommon location and briefly review the diagnostic approach to benign lytic lesions of the hand.

Case Report

A 25-year-old lady presented with a painless right index finger swelling for the past 6 months. The patient sustained a minor trauma 1 year back to the same finger. The swelling extended from the metacarpophalangeal joint to the proximal interphalangeal joint with restriction of terminal flexion at the joints. Plain radiographs revealed a lytic expansile lesion of proximal phalanx (Fig. 1). There was no periosteal reaction, calcification in the matrix, or soft tissue involvement. MRI showed multiseptate T2 hyperintense lesion with fluid-fluid levels (Fig. 2). Hemorrhagic fluid with granulation tissue was noted intraoperatively. Curettage and autologous iliac crest bone grafting was done (Fig. 3, 4). Histopathology confirmed the diagnosis of ABC (Fig. 5, 6). At 12-month follow-up, the clinical swelling was absent and radiograph shows good graft incorporation (Fig. 7).

Discussion

Less than 5% of all ABCs occur in the hand and only half of these arise from the phalanges [2, 3]. Of all benign tumors of the hand, ABCs represent 1–8% as compared to enchondroma which is seen in 70–90% cases [1, 2]. ABC is generally considered reactive tumor-like lesion,

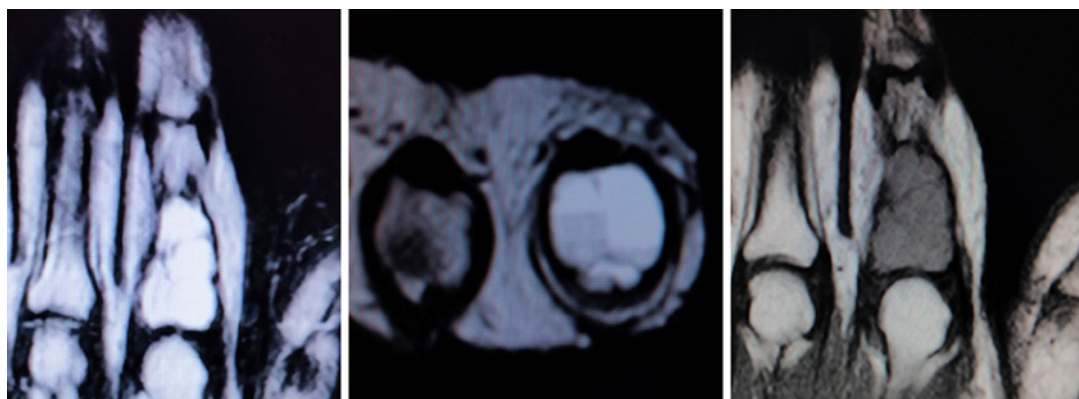


Fig. 2. MRI showing T2 coronal and axial sections showing hyperintense lesion. T1 section shows hypointense lesion. Fluid-fluid level is noted in the axial section and multiloculated lesion noted in the coronal sections. MRI, magnetic resonance imaging.



Fig. 3. Intraoperative picture after curettage and bone grafting.

but cytogenetic analysis suggests that it is a mesenchymal tumor [4, 5]. It is usually (80%) seen in patients <20 years of age and is rare after the fourth decade. Patients commonly present with swelling which may or may not be associated with pain. It is usually found in metaphysis or meta-diaphysis of long bones, the spine, and the pelvis in the decreasing order of frequency [4].

On plain radiography, an expansile lytic lesion with cortical thinning and septations giving rise to a multiloculated appearance is seen. A CT scan can detect pathological fractures. Cystic ABC presents with multiple fluid-fluid levels in T2WI of MRI, which can differentiate it with other tumors of the hand [6]. However, the solid variants of ABCs do not present with fluid-fluid levels on the MRI. In such cases, it can be confused with giant cell reparative granuloma (GCRG) which is similar to solid ABC in morphology and histopathology. A USP6 gene rearrangement study has suggested that most GCRGs of the tubular bones of limbs represent true ABCs, and the term GCRG should be used only in lesions arising from gnathic location [7].

The *differential diagnoses* for solitary lytic lesions of hand are as follows:

- Enchondromas are asymptomatic and are often detected incidentally. Symptoms may arise due to pathologic fractures or malignant transformation. Radiographs usually show chondroid matrix with calcifications but can present as pure lytic lesions in hands and feet. CT scan helps in detecting pathological fractures and calcifications. MRI shows characteristics of cartilaginous tumors [8].
- SBC is central, metaphyseal, less aggressive, and less expansile lesions when compared to ABC. Active cysts are seen in an age <10–12 years and have a single cavity with thin



Fig. 4. Immediate post-op radiograph. Autologous bone graft is filled in the cavity after thorough curettage.

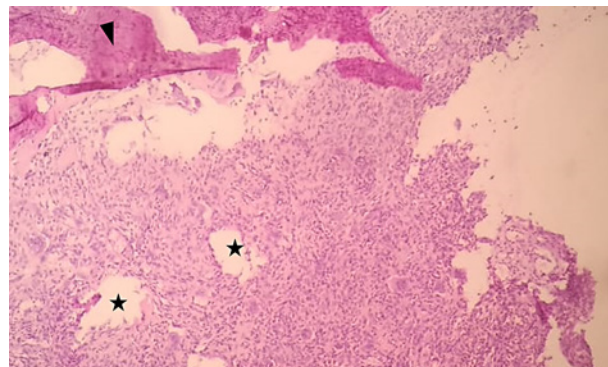


Fig. 5. Histopathological section showing fibro-collagenous tissue, proliferating spindle cells, multinucleated osteoclastic giant cells, blood vessels (star mark) with benign osteoid (arrowhead) (H&E. ×4).

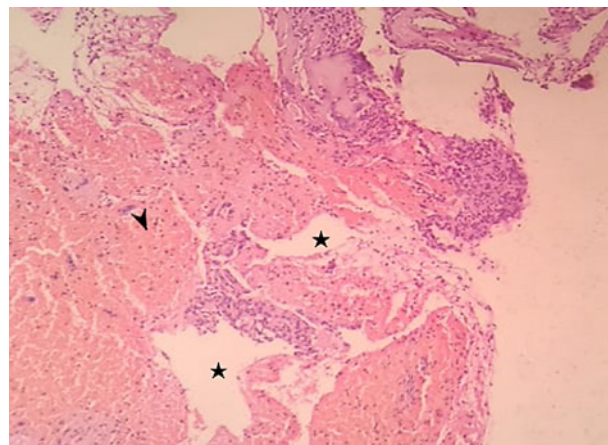


Fig. 6. Histopathological section showing areas of congestion (arrowhead) with dilated blood vessels (star mark), proliferating spindle cells, and fibro-collagenous tissue (H&E. ×10).

cortical bone. In contrast, inactive cysts are multiseptated, away from the growth plate (>5 mm), have a thick cortex, and usually occur after 12 years of age [8].

- GCT is a locally aggressive epiphyseal tumor occurring in long bones, usually between 20 and 40 years of age. The gross appearance of GCT is characteristic with hemorrhagic-cystic



Fig. 7. Radiograph at 12-month follow-up shows good incorporation of graft and remodeling of the phalangeal cortex.

areas and necrotic areas with reparative fibrosis. Enhancing solid components on MRI supports the diagnosis of GCT over ABC [8, 9].

- Fibrous dysplasia is commonly seen in long bones as an asymptomatic swelling which starts around puberty. A characteristic ground glass appearance is seen on plain radiographs [8].
- Fibrous cortical defect is more common in males (3:2) between 5 and 15 years of age and begins as an asymptomatic intracortical or subperiosteal lesion [8].
- GCRG is a solid tumor seen in gnathic locations. It is morphologically similar to solid variants of ABC [7]. GCRG is excluded in this patient based on the cystic gross appearance, presence of large blood vessels instead of small proliferating vessels on histology, and extra gnathic location.

MRI, though not definitive, can differentiate between the above lesions [9]. Biopsy is an invasive procedure, and it is unnecessary in enchondroma and cannot detect SBC. The diagnostic approach to benign lytic lesions of the hand is summarized in Figure 8.

Curettage with or without bone grafting remains the mainstay of treatment of ABC. The bone graft can be taken from olecranon process or distal radius if the required graft quantity is minimal. Adjuvant therapies include high-speed burr, phenol, cement, argon beam coagulation, and cryotherapy [10]. Radiotherapy and embolization have been reported to be successful, but the evidence is low. Newer methods of treating ABCs like sclerotherapy, bisphosphonates, percutaneous doxycycline, and RANKL inhibitors like denosumab have been promising, but need long-term results for wider acceptance [10]. ABC has a propensity for local recurrence, especially in the initial 2 years. Recurrence rates can be up to 30–50% in incomplete resections [6].

In conclusion, ABC can occur in atypical locations like hand. A solitary lytic lesion without calcification in a hand radiograph needs to be evaluated with MRI. The use of MRI can avoid

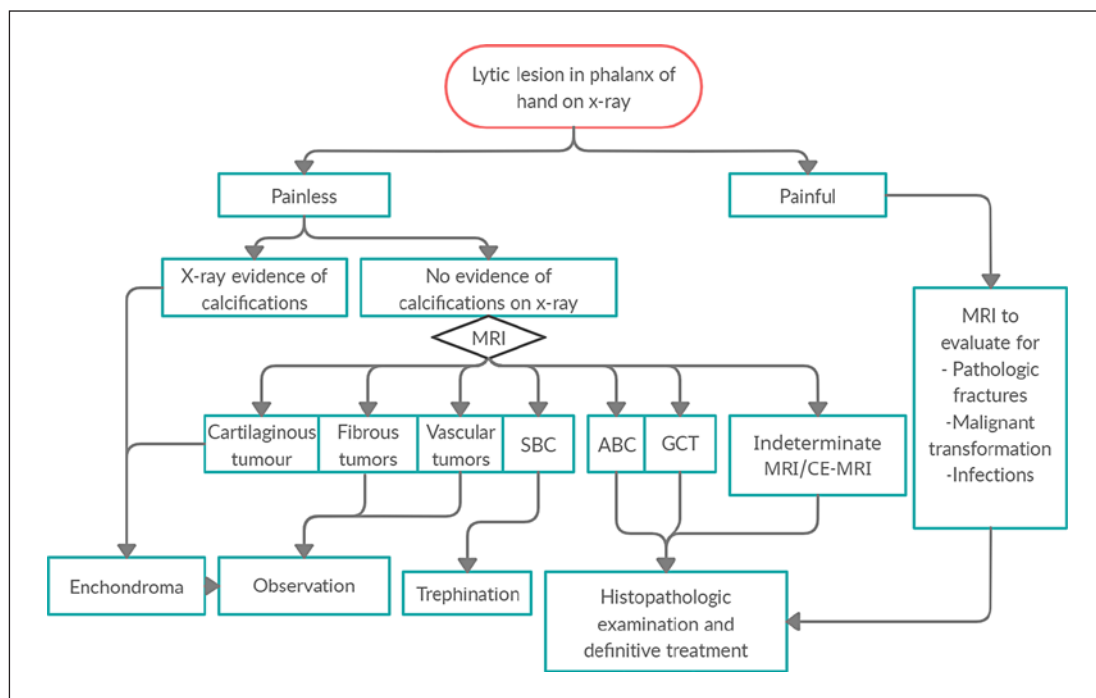


Fig. 8. Diagnostic approach for a lytic lesion of hand. Plain MRI can differentiate most solitary bone lytic lesions. In indeterminate cases, CE-MRI is useful in differentiating active versus inactive lesions, cartilaginous versus fluid based, and high-flow versus stagnant lesions. If the lesion is indeterminate even after CE-MRI, biopsy is indicated. MRI, magnetic resonance imaging; CE-MRI, contrast enhancement of MRI.

unnecessary surgeries in conditions such as enchondroma, SBC, nonossifying fibroma, and fibrous dysplasia.

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Statement of Ethics

A written informed consent of the patient has been obtained to publish their case report and images.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Contribution Details

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References

- Henderson MM, Neumeister MW, Bueno RA Jr. Hand tumors. *Plast Reconstr Surg*. 2014 Jun;133(6):814e–21e.
- Simon MJ, Pogoda P, Hövelborn F, Krause M, Zustin J, Amling M, et al. Incidence, histopathologic analysis and distribution of tumours of the hand. *BMC Musculoskelet Disord*. 2014 Dec 1;15(1):182.
- Melamud K, Drapé JL, Hayashi D, Roemer FW, Zentner J, Guermazi A. Diagnostic imaging of benign and malignant osseous tumors of the fingers. *Radiographics*. 2014 Nov;34(7):1954–67.
- Cottalorda J, Bourelle S. Modern concepts of primary aneurysmal bone cyst. *Arch Orthop Trauma Surg*. 2007 Feb 1;127(2):105–14.
- Oliveira AM, Perez-Atayde AR, Inwards CY, Medeiros F, Derr V, Hsi BL, et al. USP6 and CDH11 oncogenes identify the neoplastic cell in primary aneurysmal bone cysts and are absent in so-called secondary aneurysmal bone cysts. *Am J Pathol*. 2004 Nov 1;165(5):1773–80.
- Jansen J, Terwey B, Rama B, Markakis E. MRI diagnosis of aneurysmal bone cyst. *Neurosurg Rev*. 1990 Jun 1; 13(2):161–6.
- Agaram NP, LeLoarer FV, Zhang L, Hwang S, Athanasian EA, Hameed M, et al. USP6 gene rearrangements occur preferentially in giant cell reparative granulomas of the hands and feet but not in gnathic location. *Hum Pathol*. 2014 Jun 1;45(6):1147–52.
- Campanacci M. *Bone and soft tissue tumors*. 2nd ed. Wien: Springer-Verlag; 1999. p. 73–463.
- Stacy GS, Peabody TD, Dixon LB. Mimics on radiography of giant cell tumor of bone. *AJR Am J Roentgenol*. 2003 Dec;181(6):1583–9.
- Park HY, Yang SK, Sheppard WL, Hegde V, Zoller SD, Nelson SD, et al. Current management of aneurysmal bone cysts. *Curr Rev Musculoskelet Med*. 2016 Dec 1;9(4):435–44.