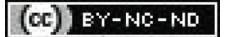


A Case of Aneurysmal Bone Cyst of Fourth Metacarpal Bone: A Rare Entity at Unusual Location

AJIT KUMAR SINGH¹, MADHUSMITA MOHANTY², URMILA SENAPATI³

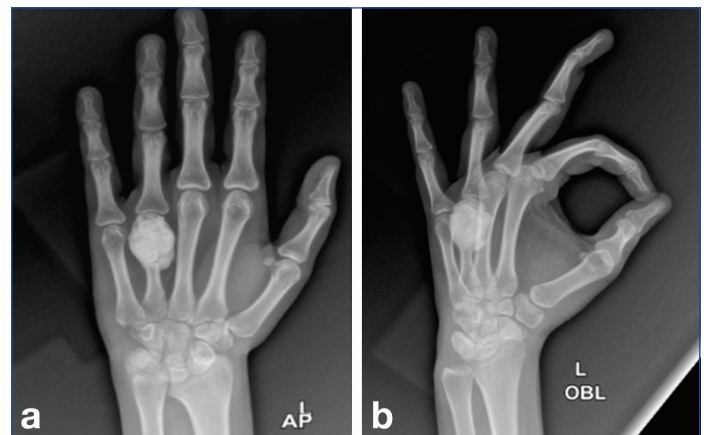
ABSTRACT

An Aneurysmal Bone Cyst (ABC) is a locally aggressive, rapidly growing, rare benign bone tumour that typically occurs in children and early adulthood. It usually occurs in the first and second decades of life, with a female predominance. The long bones of the lower extremities and spine are common sites for ABC, with rare involvement of the small bones of the hand. An 18-year-old female presented to the hospital with progressive swelling over her left hand for 4 to 5 months, associated with pain. Local examination elicits pain and tenderness over the 4th metacarpal bone with restricted movement at the Metacarpophalangeal (MCP) joint. A histopathological diagnosis of ABC was made due to the presence of osteoclastic giant cells lining a cyst, along with areas showing a pool of haemorrhage and stromal cells. The postoperative period remained uneventful, with no complaints of pain and good healing of the operation site noted during follow-up. Metacarpal bones are rare sites for ABCs.

Keywords: Bone tumours, Giant cell lesion, Metacarpophalangeal

CASE REPORT

An 18-year-old female presented to the hospital with progressive swelling over her left hand for 4 to 5 months, associated with dull aching pain that increases with movement. Local examination elicited pain and tenderness over the 4th metacarpal bone with movement restriction at the MCP joint. The overlying skin was found unremarkable with a mild local rise in temperature. General body build, blood pressure, and pulse were within normal limits. Haematological and biochemical investigations are shown in [Table/Fig-1]. X-ray examination of the left hand showed an expansile lytic lesion with ballooning over the epiphysis and metaphysis of the 4th metacarpal bone [Table/Fig-2a,b], leading to a provisional diagnosis of Giant Cell Tumour (GCT) and ABC. The patient was managed surgical management, with excision performed under local anaesthesia, and the sample was sent for histopathological study.



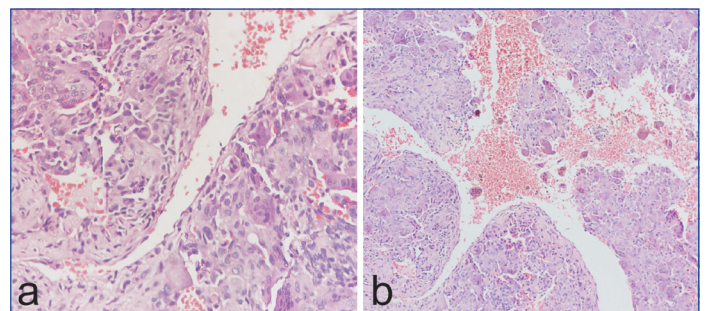
[Table/Fig-2]: a,b) X-ray left hand showing expansile lytic lesion in 4th metacarpal bone.

Parameters	Value (Reference range)
Haemoglobin	11.3 gm/dL (12-15 gm/dL)
TRBC	$3.8 \times 10^6/\mu\text{L}$ ($4.5-5.5 \times 10^6/\mu\text{L}$)
PCV	34.3% (36-46%)
MCV	87.5 fL (83-101fL)
MCH	28 pg (27-32 pg)
MCHC	31.2 gm/dL (31.5-34.5 gm/dL)
RDW-CV	12.8% (11.6-14.0%)
TPC	$159 \times 10^3/\mu\text{L}$ ($150-410 \times 10^3/\mu\text{L}$)
TWBC	$9 \times 10^3/\mu\text{L}$ ($4-10 \times 10^3/\mu\text{L}$)
Neutrophil	67% (40-80%)
Lymphocyte	24% (20-40%)
Monocyte	07% (2-10%)
Eosinophil	02% (1-6%)
Basophil	0 (0-1%)
Serum calcium	9.9 mg/dL (8.6-10.3 mg/dL)
Serum Urea	30.3 mg/dL (12.0-42.0 mg/dL)
Serum creatinine	0.6 mg/dL (0.6-1.1 mg/dL)

[Table/Fig-1]: Laboratory investigations findings.

TRBC: Total red blood cells, PCV: Packed cell volume, MCV: Mean corpuscular volume, MCHC: Mean corpuscular haemoglobin concentration, RDW-CV: Red cell distribution width-coefficient of variation, TPC: Total platelet count, TWBC: Total white blood cell

Grossly, laboratory received multiple tissue fragments ranging in size from 0.3 to 4.0×2.0×0.4 cm. Histopathological examination {Haematoxylin and Eosin (H&E)} revealed multiple tissue fragments showing large cysts mostly filled with pools of haemorrhage. Osteoclastic giant cells and stromal cells lining the cysts, along with fibrous stroma and inflammatory cell infiltration. A few fragments also contained sheets of stromal cells along with osteoclastic giant cells [Table/Fig-3a,b]. So, the pathologists give a diagnosis of ABC of the 4th metacarpal bone. Postoperatively, the patient is doing well, and follow-up reveals no pain at the operation site with good healing.



[Table/Fig-3]: a,b) Histology (H&E staining 100X, 400X) showing osteoclastic giant cell lining a cyst with extensive haemorrhage and stromal cells.

DISCUSSION

The ABCs are rare, locally aggressive, benign bone tumours that typically affect the long bones of the lower extremities and spine, with the small bones of the hand being an uncommon location [1,2]. They usually occur in the first and second decades of life and predominantly affect females [2]. The present case report describes an ABC affecting the 4th metacarpal bone in an 18-year-old female, which is a rare presentation. A review of the literature reveals that ABCs of the metacarpal bones are indeed uncommon, with only a few published case reports. Nanda SN et al., reported a case of an ABC involving the 3rd metacarpal bone in an 11-year-old boy [2]. Similarly, De Vitis R et al., conducted a systematic review of ABCs in the hand and found that the metacarpal bones were involved in only four out of 32 cases [3]. More recently, Pena-Burgos EM et al., reported a series of 14 cases of primary ABCs in the hands and feet, with three cases involving the metacarpal bones [4].

Compared to the cases reported in the literature, the patient presented with the more common symptoms of pain, slow-growing swelling, and restricted joint movement [1,2,4]. It can present with a pathological fracture, in contrast with the present case. The radiological findings of an expansile, lytic lesion with internal septations were also consistent with the typical appearance of ABCs [4]. Histopathologically, the present case showed the hallmark features of ABCs, including osteoclastic giant cells, stromal cells lining the cystic spaces, and areas of haemorrhage [2-4].

The exact aetiology and pathogenesis of ABCs are not fully understood, with various theories proposed, such as local haemodynamic disturbances leading to venous thrombosis and arteriovenous shunting [5], or as a secondary reaction to a pre-existing bone lesion [3]. Recent studies have identified specific chromosomal translocations and the overexpression of insulin-like growth factor-1 as potential contributors to the development of ABCs [6-9].

Differentiating ABCs from other giant cell-containing lesions, such as GCT of bone and Telangiectatic Osteosarcoma (TO), can be challenging based on clinical and radiological features alone [4,10]. However, careful histopathological examination can help distinguish these entities. GCTs typically show sheets of stromal cells with osteoclastic giant cells, while in ABCs, the cystic spaces and haemorrhage are more prominent [10]. ABCs and TOs reveal similar clinical and radiological features. TO can also occur in hand and foot bones. Pathological fracture is more common in TO compared to ABC due to rapid growth and infiltration into surrounding bone. TOs, on the other hand, exhibit an osteoid matrix, nuclear atypia, and increased mitotic activity, contrasting with the more benign histology of ABCs [4].

Due to the rarity of ABCs in the metacarpal bones, there is no established treatment protocol. In the present case, as in the majority of reported cases, complete surgical excision was the treatment of choice [3,4,6]. Careful follow-up is essential, as local recurrence is a known complication of ABCs [4,6].

CONCLUSION(S)

The present case report highlights the rare occurrence of an ABC in the 4th metacarpal bone of an 18-year-old female. A combination of clinical, radiological, and histopathological findings is crucial for the accurate diagnosis of this uncommon entity. Sometimes, other tumours can be presented as ABC-like features near to the tumour due to secondary changes. So, complete sampling of the specimen should be done to rule out other neoplastic lesions. Surgical excision with bone grafting is the treatment of choice. Prompt surgical management and close follow-up are essential to monitor for potential recurrence.

REFERENCES

- [1] Deventer N, Toporowski G, Gosheger G, de Vaal M, Luebben T, Budny T, et al. Aneurysmal bone cyst of the foot: A series of 10 cases. *Foot Ankle Surg.* 2022;28(2):276-80. Doi: 10.1016/j.fas.2021.03.002. Epub 2021 Mar 6. PMID: 33715952.
- [2] Nanda SN, Tripathi S, Shiraz SM, Warriar S. Aneurysmal bone cyst of 3rd metacarpal, management and follow-up: A case report. *J Orthop Case Rep.* 2018;8(2):09-12. Doi: 10.13107/jocr.2250-0685.1024. PMID: 30167402; PMCID: PMC6114219.
- [3] De Vitis R, Vitiello R, Perna A, Passiatore M, Cipolloni V, Pripp C, et al. Hand's aneurysmal bone cyst: A rare localization. Case report and systematic literature review. *Orthop Rev (Pavia).* 2020;12(Suppl 1):8658. Doi: 10.4081/or.2020.8658. PMID: 32913594; PMCID: PMC7459386.
- [4] Pena-Burgos EM, Serra-Del Carpio G, Tapia-Viñe M, Iglesias Urraca C, Cordero García JM, Ortiz-Cruz EJ, et al. Primary aneurysmal bone cyst of hands and feet: A series of 14 cases. *Ann Diagn Pathol.* 2023;66(6):152169. Doi: 10.1016/j.anndiagpath.2023.152169. Epub 2023 Jun 1. PMID: 37295038.
- [5] Althof PA, Ohmori K, Zhou M, Bailey JM, Bridge RS, Nelson M, et al. Cytogenetic and molecular cytogenetic findings in 43 aneurysmal bone cysts: Aberrations of 17p mapped to 17p13.2 by fluorescence in situ hybridization. *Mod Pathol.* 2004;17(5):518-25.
- [6] Mejbel HA, Zein-Sabatto B, Wei S, Siegal GP. An aneurysmal bone cyst harboring a novel ACSL4::USP6 fusion gene. *J Orthop Sci.* 2024;29(2):690-94. Doi: 10.1016/j.jos.2023.05.014. Epub 2023 Jun 20. PMID: 37349178.
- [7] Oliveira AM, Hsi BL, Weremowicz S, Rosenberg AE, Dal Cin P, Joseph N, et al. USP6 (Tre2) fusion oncogenes in aneurysmal bone cyst. *Cancer Res.* 2004;64(6):1920-23. Doi: 10.1158/0008-5472.can-03-2827. PMID: 15026324.
- [8] Mendenhall WM, Zlotecki RA, Gibbs CP, Reith JD, Scarborough MT, Mendenhall NP. Aneurysmal bone cyst. *Am J Clin Oncol.* 2006;29(3):311-15. Doi: 10.1097/01.coc.0000204403.13451.52. PMID: 16755186.
- [9] Leithner A, Lang S, Windhager R, Leithner K, Karlic H, Kotz R, et al. Expression of insulin-like growth factor-I (IGF-I) in aneurysmal bone cyst. *Mod Pathol.* 2001;14(11):1100-04. Doi: 10.1038/modpathol.3880443. PMID: 11706070.
- [10] Ellison DA, Sawyer JR, Parham DM, Nicholas R Jr. Soft-tissue aneurysmal bone cyst: Report of a case with t(5;17)(q33;p13). *Pediatr Dev Pathol.* 2007;10(1):46-49. Doi: 10.2350/06-03-0070.1. PMID: 17378626.

PARTICULARS OF CONTRIBUTORS:

1. Senior Resident, Department of Pathology, Kalinga Institute of Medical Sciences, Bhubaneswar, Odisha, India.
2. Associate Professor, Department of Pathology, Kalinga Institute of Medical Sciences, Bhubaneswar, Odisha, India.
3. Professor, Department of Pathology, Kalinga Institute of Medical Sciences, Bhubaneswar, Odisha, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:

Madhusmita Mohanty,
Duplex 15, Somu Villa, Nandan Vihar, Near Northen Heights Apartment,
Bhubaneswar-751024, Odisha, India.
E-mail: drmadhu80@gmail.com

AUTHOR DECLARATION:

- Financial or Other Competing Interests: None
- Was informed consent obtained from the subjects involved in the study? No
- For any images presented appropriate consent has been obtained from the subjects. NA

PLAGIARISM CHECKING METHODS: [Jain H et al.]

- Plagiarism X-checker: Feb 19, 2024
- Manual Googling: Mar 14, 2024
- iThenticate Software: Apr 13, 2024 (8%)

ETYMOLOGY: Author Origin

EMENDATIONS: 7

Date of Submission: **Feb 17, 2024**

Date of Peer Review: **Mar 04, 2024**

Date of Acceptance: **Apr 15, 2024**

Date of Publishing: **Jun 01, 2024**