Indian Journal of Pathology and Oncology 2022;9(4):367–368

Content available at: https://www.ipinnovative.com/open-access-journals

Indian Journal of Pathology and Oncology

Journal homepage: www.ijpo.co.in

Case Report

Xanthogranulomatous osteomyelitis of Tibia in a case of 43 years old male patient: A rare entity diagnosed in a tertiary care hospital

Sarbashis Hota[1,*], Sukanya Ghosh[1], Urvee Sarkar[1]

[1] *Dept. of Pathology, R G Kar Medical College and Hospital, Kolkata, West Bengal, India*

ARTICLE INFO

Article history:
Received 11-11-2022
Accepted 26-11-2022
Available online 14-12-2022

Keywords:
Xanthogranuloma
Osteomyelitis
Touton giant cell

ABSTRACT

Xanthogranulomatous inflammation is a form of chronic inflammatory process with collection of foamy macrophages in sheets, often with presence of giant cell reaction. Here, we present a case of xanthogranulomatous osteomyelitis of tibia in a 43 years old patient, which is a rare pathological entity and possibly was reported first in 1984. The histopathological sections revealed dense collection of lympho-plasmacytic infiltrate in the marrow spaces with syncytial sheets of foamy macrophages admixed with numerous classical Touton giant cells.

1. Introduction

Xanthogranulomatous inflammation is a form of chronic inflammatory process with collection of foamy macrophages in sheets, often with presence of giant cell reaction. Classically, xanthogranulomatous inflammation is often reported in pathological conditions of gall bladder and kidney. An entity named juvenile xanthogranuloma is regarded as a variant of histiocytic neoplasm of hemato-lymphoid lineage. Here, we present a case of xanthogranulomatous osteomyelitis of tibia in a 43 years old patient, which is a rare pathological entity and possibly was reported first in 1984.[1]

2. Case Report

A 43 years old male sustained a closed fracture of Tibial shaft following a road traffic accident and open reduction with internal fixation was done in the Emergency OT along with implantation of Tibial nail. The post operative period was uneventful under high doses of parenteral antibiotic regimen. However, following the next visits, 6 weeks after the incident, the patient complained of loco-regional swelling, which soon gave rise to discharging sinuses. An X-ray was sought, which revealed the presence of sequestrum with collection inside the medullary cavity, surrounding the implant. A sequestrectomy & saucerisation along with removal of implant was planned; and the case was managed further by Illizarov's technique of external fixation. The curetted infected marrow material was sent for histopathological examination.

The histopathological sections revealed dense collection of lympho-plasmacytic infiltrate in the marrow spaces with syncytial sheets of foamy macrophages, obliterating the medullary cavity. Numerous classical Touton giant cells were evident, interspersed within the histiocytes. So, a diagnosis of Xanthogranulomatous osteomyelitis was made.

3. Discussion

In 1984, Cozutto[1] reported the first two cases of xanthogranlomatous osteomyelitis, one involving the first rib and the other involving tibial epiphysis. This particular entity is important to recognize, as clinically and

* *Corresponding author.*
E-mail address: sarbashishota94@gmail.com (S. Hota).

https://doi.org/10.18231/j.ijpo.2022.089

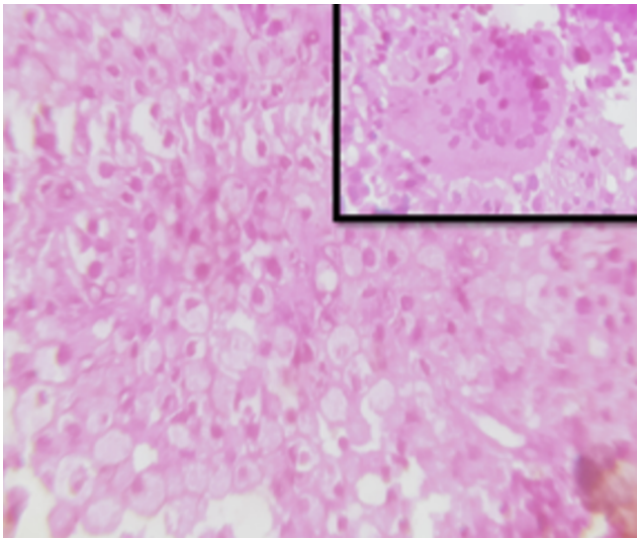

Fig. 1: Sheets of foamy macrophages, 400X, H & E [Touton giant cell inset]

radiologically it can mimic neoplasm from every respect. A long array of differential diagnoses like Langerhans cell histiocytosis, Erdheim Chester disease, chronic recurrent multifocal osteomyelitis(CRMO), even metastatic renal cell carcinoma may get included in the list.

Borijan et al[2] reported a multifocal case of xanthogranulomatous osteomyelitis in a 14 years old Afgan male, involving the humerus and the fibula. Kamat et al[3] and Solooki et al[4] have described two similar cases involving distal part of tibia, in a 13 year and 15 year old boy, respectively.

Pathak S et al[5] reported a case of xanthogranulomatous osteomyelitis of hip in a 43 years old female patient, which simulated TB hip. The patient also gave a history of ATD intake during the last five months.

Vankalakunti et al[6] reported a similar case in a 50-year-old post menopausal female manifesting as an expansile osteolytic lesion of ulna, while Baisakh M R et al[7] reported bilateral occurrence of this pathology in a 21-year-old female, involving the lower end of femur and upper end of tibia. The entity presented as lytic lesions of bone on x-ray.

Our patient is now under high doses of parenteral antibiotic regimen, which may be adjusted further after arrival of the culture-sensitivity report. The inflammatory markers are gradually showing signs of improvement and patient is clinically improving according to our expectation.

4. Conclusion

The interesting entity of xanthogranulomatous osteomyelitis can be clinically vexing for the orthopaedic surgeon. Accurate diagnosis is necessary for planning further line of management as well as for overall outcome of the patient.

5. Source of Funding

None.

6. Conflict of Interest

None.

References

1. Cozutto C. Xanthogranulomatous osteomyelitis. *Arch Pathol Lab Med*. 1984;108(12):973–6.
2. Borijan A, Rezaei F, Eshagi MA, Shemshaki H. Xanthogranulomatous Osteomyelitis. *J Orthop Traumatol*. 2012;13(4):217–20.
3. Kamat G, Gramapurohit V, Myageri A, Shettar C. Xanthogranulomatous osteomyelitis presenting as swelling in right tibia. *Case reports in pathology*. 2011;.
4. Solooki S, Hoveidaei AH, Kardeh B, Azarpira N, Salehi E. Xanthogranulomatous osteomyelitis of the Tibia. *Ochsner J*. 2019;19(3):276–81.
5. Pathak S, Gautam, Prince PC, Bagtharia P, Sharma A. A rare case report on xanthogranulomatous osteomyelitis of hip mimicking tuberculosis and review of literature. *Cureus*. 2019;11(10):e5921.
6. Vankalkunti M, Saikia UN, Matthew M, Kang M. Xanthogranulomatous osteomyelitis of ulna mimicking neoplasm. *World J Surg Oncol*. 2007;5:46. doi:10.1186/1477-7819-5-46.
7. Baisakh MR, Kar MR, Agarwal A, Mohapatra N. Xanthogranulomatous osteomyelitis mimicking neoplasm: a rare entity. *Indian J Pathol Microbiol*. 2016;59:253–4.

Author biography

Sarbashis Hota, Senior Resident

Sukanya Ghosh, Senior Resident

Urvee Sarkar, Assistant Professor

Cite this article: Hota S, Ghosh S, Sarkar U. Xanthogranulomatous osteomyelitis of Tibia in a case of 43 years old male patient: A rare entity diagnosed in a tertiary care hospital. *Indian J Pathol Oncol* 2022;9(4):367-368.