Case Report

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Pathology Section

Cytodiagnosis of Telangiectatic Osteosarcoma in a Patient with Previous Fracture Humerus Surgery with Plating: A Case Report

SIMRAN KHAN[1], ARVIND BHAKE[2], GARIMA CHANDRAKAR[3], SHAKTI SAGAR[4]

ABSTRACT

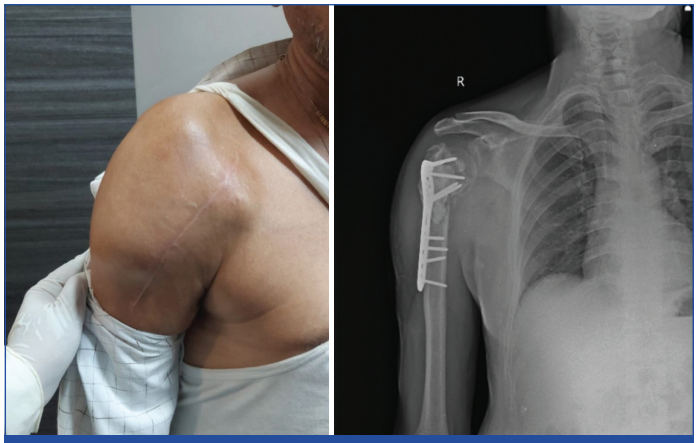
Telangiectatic Osteosarcoma (TO) is a rare variant of osteosarcoma, accounting for 0.4-12% of cases. It is a high-grade malignancy occurring in adolescence and the adult population. It commonly occurs in the bones of the limbs but rarely in the humerus. It is assumed to originate in stem cells of the mesenchyme or from transformed osteoblasts. The initial X-ray of TO would show an osteolytic lesion over the metaphysis of the long bone. However, the radiographic imaging techniques may not correlate with the histological variant of osteosarcoma. It has been rarely diagnosed and reported in the literature in terms of diagnostic modality using Fine Needle Aspiration Cytology (FNAC). Its cytological as well as histological diagnosis is challenging. However, the diagnosis of TO has numerous implications such as prognosis, differential diagnosis, and treatment options. The present clinical and pathological case images depict the unusual features of TO involving the upper humerus by an osteolytic lesion. Its clinical and diagnostic chronology is interesting due to the occurrence of TO in a case that underwent surgery for a fractured humerus with plating. The plate fixation in this particular case was surrounded by osteolytic tumour tissue extending into the soft tissue. The FNAC under Ultrasonography (USG) guidance was carried out. The smears of the aspirated material typically showed malignant spindle cell fragments with a few gaps of remnant sinusoids entrapping sparse red blood cells. The smears also showed osteoid material which was lacy in character. This case has several noteworthy features in the form of cytomorphological description of the telangiectatic variant of osteosarcoma, its occurrence following plating, and subsequent confirmation on histology and Immunohistochemistry (IHC).

Keywords: Cytology, Fine needle aspiration cytology, Osteolytic lesion

CASE REPORT

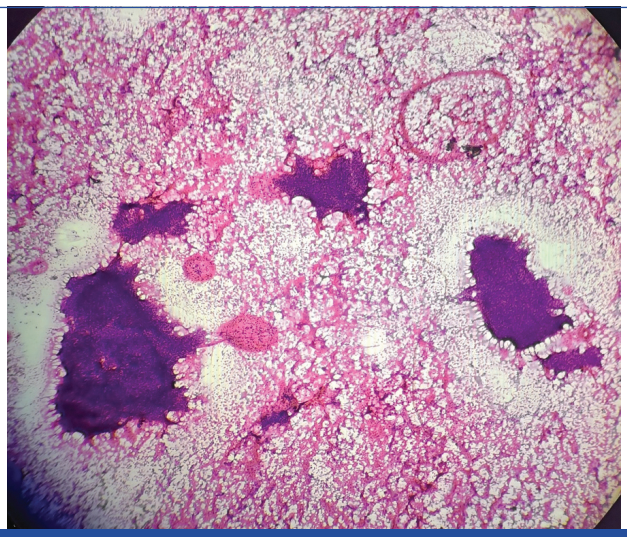
A 59-year-old male presented to the outpatient department of orthopaedics with a six-month history of pain and swelling in the right shoulder, which had gradually increased in size over the past month. Upon examination, diffuse swelling was observed throughout the right arm, with the skin appearing stretched and shiny. The size of the swelling was approximately 11×6 cm, extending up to the diaphysis of the humerus. Palpation revealed warmth, tenderness, and variability in consistency [Table/Fig-1]. Range of motion of the shoulder and elbow was severely restricted due to pain, with no evidence of distal neurovascular deficits. A previous surgery of open reduction and internal fixation had been performed, and a scar mark from this last surgery was also present. A provisional clinical diagnosis of a giant cell tumour was suspected based on X-ray findings. He was then referred to the Department of Radiology for an X-ray, which revealed ill-defined osteolysis and marrow signal alteration involving the right proximal humeral epimetaphysis and metadiaphysis with the involvement of the humeral head, greater tuberosity, lesser tuberosity, humeral neck, and proximal diaphysis. The lesion was mildly exophytic, predominantly bulging at the cortices, and causing marked thinning of the overlying cortex. The implant was seen in place [Table/Fig-2]. A differential diagnosis of primary osseous neoplasm and tendinosis was made.

Before proceeding with the FNAC procedure, the patient underwent haematological investigations and a coagulation profile assessment. The results indicated a normal haematological profile. The FNAC procedure was performed under ultrasound guidance using a 22-gauge needle attached to a 10 mL syringe. The aspiration was carried out from the proximal humerus on the right-side. The wet smears were stained with haematoxylin and eosin and Papanicolaou stain, whereas the dried smears were stained with Giemsa.

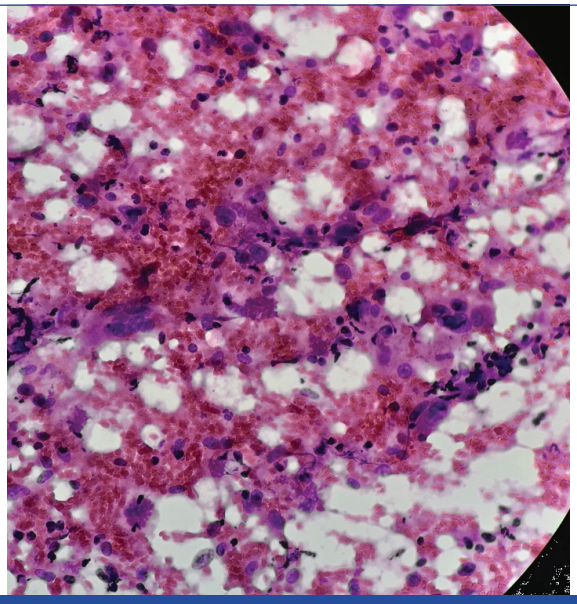


[Table/Fig-1]: Clinical image of the shoulder showing nodular and irregular swelling over the upper end of humerus with mark of previous surgery.
[Table/Fig-2]: X-ray showing osteolytic lesion involving upper end of humerus and its head with plate of previous surgery. (Images from left to right).

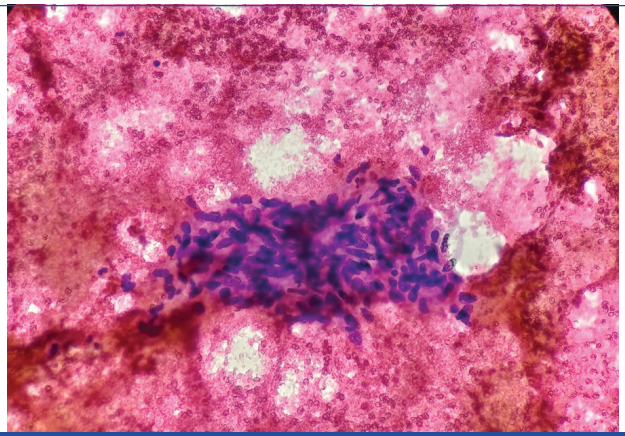
The smears were cellular. They showed multiple cell fragments consisting of pleomorphic as well as spindle nuclear cells with merging cell bodies. At places, these cell fragments offered the look of a mosaic appearance with red blood cells in between. The cells carried hyperchromatic pleomorphic nuclei with irregular chromatin condensation and infrequent nucleoli. At places, angular and elongated nuclei were also present. The cytoplasm was scant to modest and appeared pink magenta or pale eosinophilic. The smears also showed isolated mononuclear giant cells and multinucleate tumour giant cells with frequent mitosis. Rare cell fragments showed pale staining, pink material appearing like a sheet of osteoid. In rare places, osteoclastic giant cells and nuclear smudging were observed. The cytomorphology was suggestive of TO [Table/Fig-3-5]. The pathological differential diagnosis were malignant spindle cell sarcoma, giant cell tumour, and Aneurysmal



[Table/Fig-3]: FNAC of Telangiectatic Osteosarcoma (TO) shows the fragments of predominantly spindle malignant cells with nuclear atypia (Papanicolaou stain, 10x).



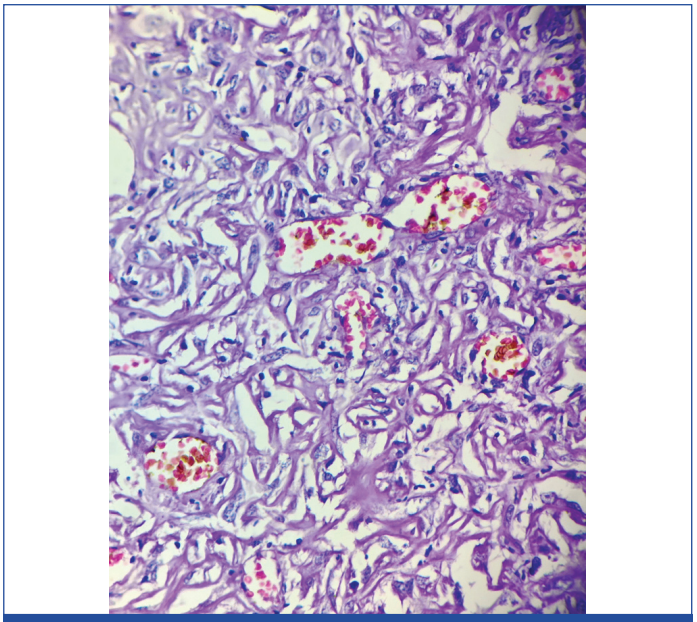
[Table/Fig-4]: FNAC of Telangiectatic Osteosarcoma (TO) shows the tumour giant cells, pleomorphic cell population in isolation and scant osteoid (Papanicolaou stain, 40x).



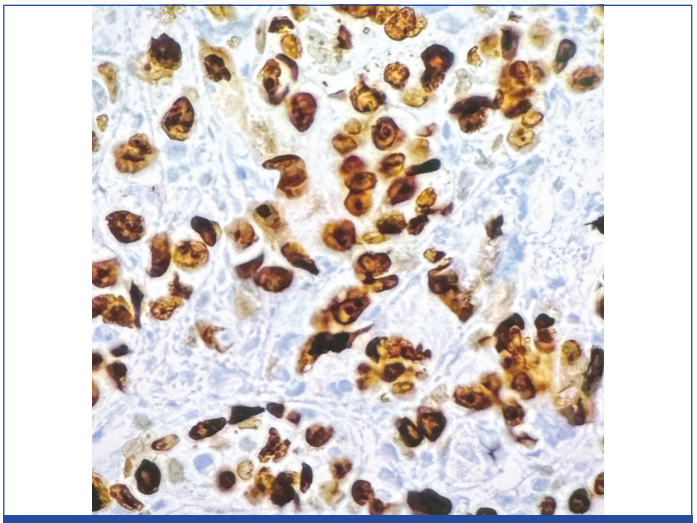
[Table/Fig-5]: FNAC of Telangiectatic Osteosarcoma (TO) shows intercellular reminiscent sinusoidal pattern and extravasated RBC (Papanicolaou stain, 40x).

Bone Cyst (ABC). A part of the aspirate was processed as a cell block. The section showed typical malignant spindle cells with a sinusoid pattern, red blood cells, and lacy osteoid. Hence, the morphological features were suggestive of TO on histopathology [Table/Fig-6]. Immunohistochemistry (IHC) for Ki-67 was performed, showing a high Ki-67 index of 60% [Table/Fig-7].

Following surgery, a medical oncologist's opinion was obtained, and adjuvant chemotherapy was initiated. After closely monitoring the patient for two weeks, he was able to resume his daily activities and was subsequently discharged. He was advised to return to the healthcare centre for a follow-up visit 15 days after discharge.



[Table/Fig-6]: Histopathological analysis of Telangiectatic Osteosarcoma (TO) shows typical spindle malignant cells with sinusoid pattern, RBCs, and lacy osteoid (H&E, 40x).



[Table/Fig-7]: Immunohistochemistry (IHC) for Ki-67.

DISCUSSION

TO represents a rare subtype of osteosarcoma, comprising approximately 0.4-12% of all occurrences [1,2]. Accounting for less than 10% of total osteosarcoma cases, this variant is characterised by its high-grade malignant nature. Believed to originate from mesenchymal stem cells or transforming osteoblasts, TO exhibits a male-to-female ratio of 2:1, with the typical age range being 11-29 years [1,3]. A literature search on the Google search engine yielded limited cases of TO of the humerus, with most diagnosis confirmed through histopathological examination [1]. While the diagnosis of TO through FNAC has been reported, such occurrences are exceptionally rare [4-7].

Malignant lytic bone tumours raise suspicion for TO, which closely mimics conventional osteosarcoma in radiographic and clinical characteristics. Radiologically, TO exhibits similarities with ABC and giant cell tumours, posing diagnostic challenges for radiologists. X-ray imaging often reveals enlarged septa and enhancing nodular patches surrounding a non enhancing haemorrhagic component, aiding in diagnosis. These areas frequently yield diagnostic specimens with cytologic atypia, and stromal cells exhibiting strong mitotic activity [8-10].

A study conducted by Pinto RGW assessed the utility of FNAC in diagnosing osteosarcomas [6]. This study underscored FNAC's effectiveness in diagnosing multicentric or multifocal osteosarcoma, providing valuable insights into its diagnostic role across various subtypes of this rare malignancy. Current treatment protocols advocate for neoadjuvant chemotherapy followed by surgical resection. With the implementation of neoadjuvant chemotherapy, the prognosis for TO patients has markedly improved, now comparable to that of patients with conventional osteosarcoma [11-13].

The FNA diagnosis of osteosarcoma has been reported in the past. The existence of TO in a patient treated for a fractured humerus is not reported in the literature. Present study reported the unique diagnostic cytomorphological characteristics (FNAC) enabling the diagnosis of TO. The rarity of its occurrence in a patient previously operated on for a fracture of the upper humerus with a plate at the site due to TO has not been reported in the past. Therefore, the cytodiagnostic comparisons of the reports of TO have failed to be made with other studies [14]. The present case offers a great learning experience, and the subsequent histopathology endorsed the cytodiagnosis of TO in the present case. The IHC of TO is similar to conventional osteogenic osteosarcoma with a high Ki-67 index [4,15].

TO is extremely rare, as evidenced by the limited cases found in the literature [Table/Fig-8] [1,15]. The current case, diagnosed through FNAC and later confirmed by histological analysis of the excised specimen, represents a rare instance of this disease. Survival rates for TO are better than those for conventional osteosarcoma when treated with neoadjuvant chemotherapy and surgery. These patients did not experience any recurrence of the condition [Table/Fig-8].

Authors	Publication year	Place of study	No. of cases discussed	Finding of case
Thamizharasan L [1]	2019	India	Case of 34-year-old male	The case underscores the rarity and accurate diagnosis. The patient presented with swelling in his right arm after surgery which was ultimately diagnosed as TO.
Jeyarani G et al., [15]	2021	India	Case of six-year-old female	The patient presented with fracture of left proximal humerus after trauma.
Present case	2024	India	59-year-old male	The present case came with swelling of right shoulder with previous fracture humerus surgery with plating and was confirmed as TO.

[Table/Fig-8]: Published cases of Telangiectatic Osteosarcoma (TO) [1,15].

CONCLUSION(S)

This case study highlights the rarity and complexity of diagnosing TO, a rare variant of osteosarcoma that presents significant diagnostic challenges due to its uncommon occurrence and atypical radiological and histological presentations. The use of FNAC under ultrasound guidance proved instrumental in identifying the unique cytological features of the disease, such as malignant spindle cells and lacy osteoid material, despite the limitations typically associated with radiological imaging in correlating with the histological subtype of osteosarcoma. FNAC of the bone tumours with adequate cell yield enables the diagnosis of not only osteosarcoma but also its variant TO. The successful identification and confirmation through FNAC and subsequent histological and immunohistochemical analyses underline the importance of a comprehensive diagnostic approach.

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PARTICULARS OF CONTRIBUTORS:
1. Junior Resident, Department of Pathology, Datta Meghe Institute of Medical Sciences, Sawangi (Meghe), Maharashtra, India.
2. Director Professor, Department of Pathology, Datta Meghe Institute of Medical Sciences, Sawangi (Meghe), Maharashtra, India.
3. Junior Resident, Department of Pathology, Datta Meghe Institute of Medical Sciences, Sawangi (Meghe), Maharashtra, India.
4. Junior Resident, Department of Pathology, Datta Meghe Institute of Medical Sciences, Sawangi (Meghe), Maharashtra, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Dr. Simran Khan,
Department of Pathology, Jawaharlal Nehru Medical College, Sawangi (Meghe), Maharashtra-442107, India.
E-mail: sabhatsimrankhan416@gmail.com

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