CASE STUDY

105. Diagnostic Challenges in Maxillary Telangiectatic Osteosarcoma: A Rare Case

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Histologically, telangiectatic osteosarcoma is defined by blood-filled cystic spaces lined by high-grade malignant cells. This appearance leads to frequent diagnostic confusion with an aneurysmal bone cyst on both imaging and pathological examination.

Owing to its infrequency, aggressive behavior, and nonspecific clinical features, this variant poses significant diagnostic difficulties.

The Case: A 37-year-old woman presented with a rapidly enlarging right cheek swelling of 1 month's duration. It is associated with fresh blood coming out of the mouth. Later she reported a further increase in the swelling size, eventually impairing eating and speech. There is no history of trauma, systemic diseases, prior malignancies, or previous surgeries apart from dental prosthetic implants. On physical examination, there was a large, firm, right cheek swelling, non-pulsatile, non-compressible, without skin changes.

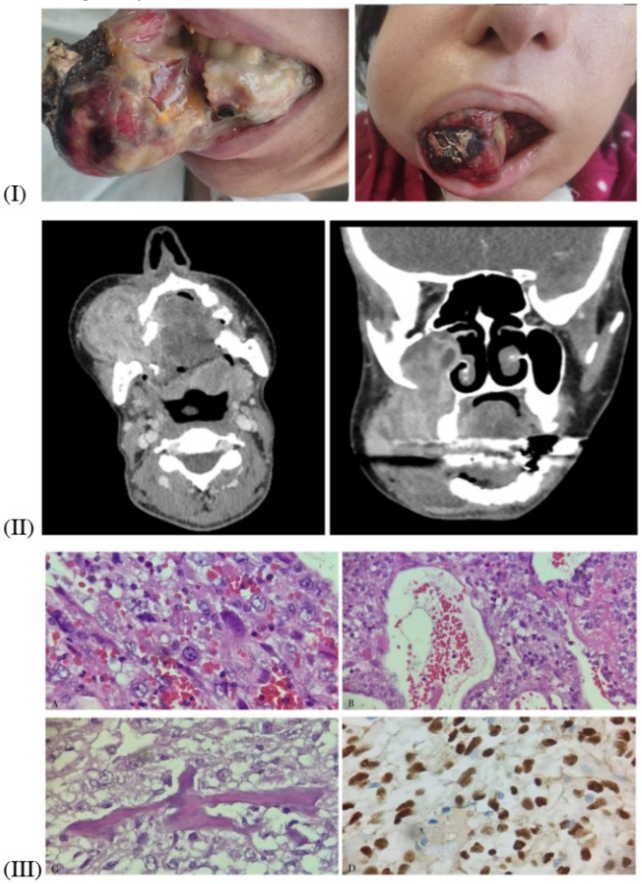
Computed Tomography (CT) scan showed a heterogeneously enhancing mass lesion occupying most of the right maxillary sinus, measuring about 7x5 cm, showing few foci of calcifications compatible with detached teeth, demonstrating aggressive features such as adjacent bone erosions and destruction of the alveolar maxillary ridge inferiorly and maxillary sinus wall posteriorly. In addition to exerting mass effect upon masticator muscles, it also has medial extension into the nasal and oral cavities, displacing the tongue into the contralateral side, causing airway narrowing. The patient was assessed by the maxillo-facial surgeons, who recommended tracheostomy to secure the airways.

An initial biopsy result revealed an inconclusive, poorly differentiated malignant neoplasm, given the aggressive clinical course and inconclusive results; repeat histopathological examination and immunohistochemistry was done and revealed a highly cellular malignant neoplasm predominantly composed of atypical spindle cells, with prominent vascularity and scattered areas of lace-like osteoid matrix production by the neoplastic cells. These features, along with the immunohistochemistry test, were suggestive of a poorly differentiated malignant neoplasm showing spindle and epithelioid features, consistent with telangiectatic osteosarcoma.

The diagnosis and treatment options were discussed with a multidisciplinary team and the patient, and the plan involved chemotherapy with MAP protocol (Methotrexate + Doxorubicin + Cisplatin) followed by surgical resection.

Conclusion: Very few cases exist regarding telangiectatic osteosarcoma, which is a unique type of osteosarcoma, and its occurrence in the maxillary sinus is even rarer. The diagnostic challenge arose during clinical judgement, histopathological, and radiological examinations, and the rapid progression of our patient's neoplasm during one month, causing profound impairment in swallowing and speech function and airway compromise, emphasizes the tumor's aggressive potential and necessitates early recognition of such complex craniofacial cancers that require a multidisciplinary approach involving airway management, oncology assessment, and function restoration and preservation, which shapes a framework for similar scenarios, especially in resource-limited healthcare settings.

Figure 1. Clinical, Radiological, and Histopathological Findings of an Oral Malignancy.



Legend: (I) Osteosarcoma of the maxilla. (II).Contrast enhanced CT of the face and neck showing a heterogeneously enhancing irregular soft tissue mass centered in the right maxillary bone, obliterating the right maxillary sinus, and extends into the oral cavity, oropharynx, temporal space, nasal cavity and the cheeks with erosion of the regional bones of the right maxilla and mandible; as tumor involved right mandibular ramus, medial and inferior walls of right maxillary sinus. No evidence of major vascular luminal narrowing. (III) A+B- Highly cellular malignant neoplasm predominantly composed of atypical spindle cells with hyperchromatic, pleomorphic nuclei and frequent mitotic figures. Prominent vascularity is observed, characterized by numerous thin- and thick-walled blood vessels distributed throughout the lesion. C- Scattered areas of lace-like osteoid matrix production by the neoplastic cells are evident. D- SATB4 immunostaining firms the osteoblastic differentiation.

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