Letter to the Editor

Primordial Odontogenic Tumor of Mandible; A Case with Proposed Diagnostic Criteria

Dear Editor

Primordial odontogenic tumor (POT) has been included as a new entity in a recent WHO (2017) classification of odontogenic tumors under benign mixed mesenchymal and epithelial tumors.[1] Although there is controversy regarding the true nature of POT as a newly-recognized embryonal neoplasm of immature dental tissue exhibiting progressive growth potential or just a histopathological variant of ameloblastic fibroma, or odontogenic myxoma/fibroma.[2] Only 8 cases of POT have been reported in the literature so far.

A 17-year-old boy referred to our institution with a painless swelling of the left lower back region of his jaw that he had been suffering from for 6 months. Family history and medical history of the patient was not contributory to his present symptom. Intra–oral examination revealed a soft tissue gingival growth extending from tooth #34 to #36 measured about 3×2 cm. The swelling was firm and too hard with no signs of discharge and ulceration. A panoramic radiograph revealed that a large multilocular well defined radiolucent lesion of right posterior mandible extending from the mesial root of #34 to the angle of the mandible surrounding unerupted #38. Resorption of the roots of #35, #36, and #37 was noted. The periphery of the lesion was well defined and not surrounded by any sclerotic border. The internal aspect of the lesion was multilocular with large size locules (Figure 1A).

Based on the radiographic examination, a provisional diagnosis of unicystic ameloblastoma was made.[3] Incisional biopsy was done but the resected tissue was found to be insufficient to make a histopathological diagnosis. Based on the suggestion of the surgeon, the incisional biopsy was not repeated.

Enucleation of the lesion was done along with the extraction of #46 under general anesthesia; the ID canal was not preserved. The defect was restored by reconstruction plates. The follow-up period of 6 months was uneventful. The resected specimen was sent for histopathological evaluation.

Histopathological examination of hematoxylin staining soft tissue sections revealed a loose and myxoid connective tissue stroma covered by columnar epithelium of a single layer, the epithelium resembled inner enamel epithelium (Figure 1B and Figure 2).

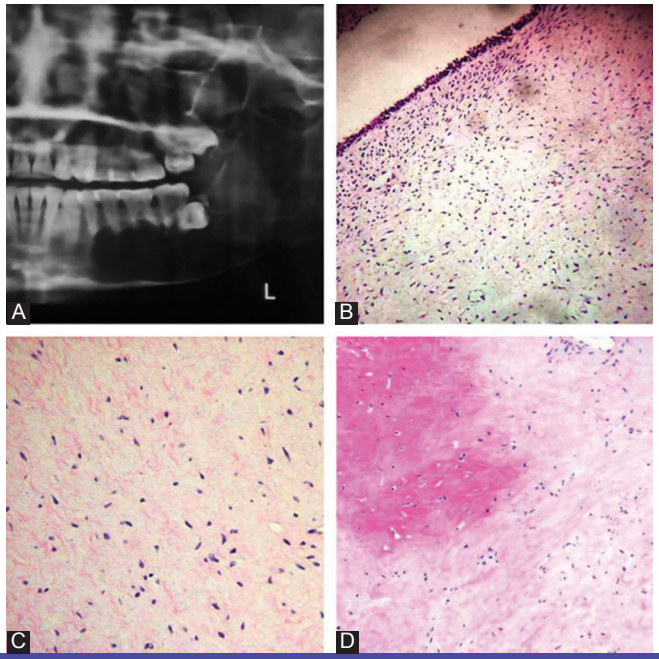

Figure 1: (A) Radiolucent osteodestructive lesion of left posterior mandible enclosing the crown of unerupted third molar. (B) Loose fibrillar connective tissue stroma surrounded by columnar epithelium resembling inner enamel epithelium (Hematoxylin and Eosin staining ×20). (C) Loose and fibrilar stroma composed of stellate-shaped fibroblasts (Hematoxylin and Eosin staining ×40). (D) Connective tissue stroma shows myxoid areas resembling dental papilla (Hematoxylin and Eosin staining ×40).

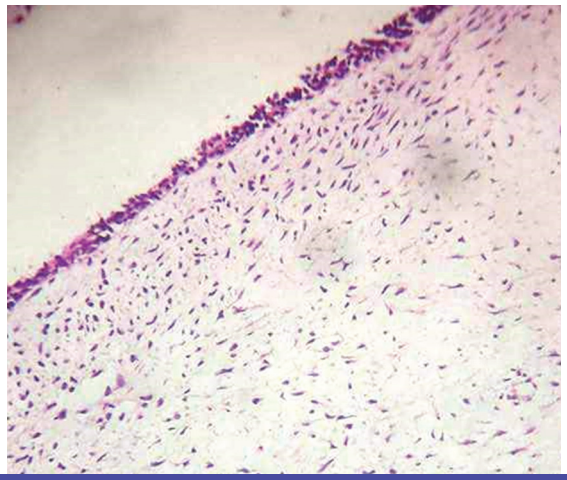

Figure 2: Loose, myxoid, and fibrilar connective tissue stroma, surrounded by columnar epithelium (Hematoxylin and Eosin staining ×40).

Table 1: A proposed-diagnostic criterion for POT based on the clinical, histopathological, and immunohistochemical criteria.[1,2,4]

S. No	Diagnostic criteria
1	Clinically, the lesions are asymptomatic, located in tooth-bearing areas, and closely associated with erupting teeth favoring their odontogenic origin.
2	Histopathologically, lesions are composed of loose fibrous connective tissue stroma having stellate-shaped fibroblasts with areas of myxoid degeneration resembling dental papilla covered by peripheral columnar epithelium which itself resembles inner enamel epithelium (covering of columnar epithelium is absent in odontogenic myxomas and odontogenic fibromas).
3	Immunohistochemically, epithelium shows positive expressions of cytokeratins (CK), especially CK5, CK14, and CK19, and mesenchymal tumor cells show positive expression of nestin.

The connective tissue stroma was loose, myxoid, and comprised of numerous stellate-shaped fibroblasts, resembling dental papilla (Figure 1C and D). Based on the histopathological examination, a final diagnosis of POT was rendered. A written consent of the patient was obtained for the publication of this case.

POT is a rare odontogenic neoplasm and is included as a separate entity in the latest WHO classification of odontogenic tumors (2017).[1] POTs are histopathologically characterized by a loose connective tissue stroma resembling dental papilla, lined by a single layer of columnar cells.[4] The differential diagnoses of POT includes ameloblastic fibroma and odontogenic fibroma/myxoma.[1] An review of the literature revealed that only eight cases of POT had been published, of which seven were published before the revised classification of odontogenic tumors was proposed and only one case was published after it was recognized as a separate entity by the WHO.[5,6] Mosqueda-Taylor and colleagues reported six cases of POT in a case series in 2014 [4] after which, Slater and colleagues reported a case of POT in 2016.[7]

In January 2017, the classification of odontogenic tumors, cysts, and maxillofacial bone tumors was published. Ando and colleagues published a case of POT after its inclusion as a separate entity in 2017.[1] Hence, this is only the second report of POT after it was recognized as a separate entity by the WHO; furthermore, this is the first report of POT from India.

An literature review did not reveal proper diagnostic criteria for POTs; hence a diagnostic criterion is proposed here, which is based on clinical, histopathological, and immunohistochemical criteria (Table 1).

The differentiation of POTs from odontogenic myxomas is mandatory, considering the fact that odontogenic myxomas are more aggressive tumors and require more aggressive mode of treatment. We hope that the diagnostic criteria proposed here will be useful for oral pathologists to diagnose and differentiate this new and uncommon entity from its closest resembling tumors in order to avoid a more aggressive treatment approach.

Conflict of Interest: None declared.

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