CASE REPORT

Case Report: A Primordial odontogenic tumor [version 1; referees: 1 approved]

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Abstract

Introduction: Primordial odontogenic tumors are a rare recently described mixed odontogenic tumor composed histopathologically of dental papilla like tissue and enamel organ like tissue. Only nine cases have been documented worldwide and we are reporting the tenth case which is from Egypt.

Clinical finding: A 2-year-old Egyptian boy that presented with an asymptomatic swelling of the mandible which appeared with multilocular radiolucency associated with an impacted developing tooth on a computerized tomography (CT) scan.

Diagnoses, interventions, and outcomes: The lesion was excised and diagnosed as a primordial odontogenic tumor. The patient was followed up for two years with no recurrence.

Conclusion: Differentiation of primordial odontogenic tumors from other odontogenic tumors, which resemble it histopathologically is crucial to avoid unnecessary aggressive treatment.

Keywords
Primordial, Mixed odontogenic tumor, Jaw tumor, Odontogenic.
Introduction
Primordial odontogenic tumor (POT) is a recently described mixed odontogenic tumor described in the last WHO classification of head and neck tumors. This tumor has been described as other entities in the past, because of its histological similarity to other odontogenic tumors as ameloblastic fibroma, odontogenic myxoma, and odontogenic fibroma and hyperplastic dental follicles.

Mosqueda-Taylor et al. described and denominated this novel lesion which did not fulfill the criteria of any of the previously classified odontogenic tumors by reporting the clinicopathological and immunohistochemical features of six cases diagnosed as primordial odontogenic tumor.

Primordial odontogenic tumors are characterized histologically by a variably cellular loose fibrous tissue with areas similar to the dental papilla, covered by cuboidal to columnar epithelium that resembles the internal epithelium of the enamel organ, surrounded at least partly by a delicate fibrous capsule.

Only nine cases have currently been reported, and we report an additional case of an primordial odontogenic tumor from Egypt.

Case report
A 2-years-old Egyptian boy referred to Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Cairo University in November 2015 with a fleshy swelling arising from site of marsupialization performed two months previous. By taking patient’s history we established the lesion arose as a painful swelling covered with normal mucosa causing obliteration of the vestibule with two months duration. Manual examinations of the regional lymph nodes were negative on examination. By computerized tomography (CT) scan, a multilocular radiolucent lesion was seen associated with an impacted developing tooth in the mandibular posterior area measuring 3cm × 4cm (Figure 1).

On aspiration, straw cystic fluid was noted. Complete surgical excision of the lesion with the impacted tooth was performed. And the excised lesion was sent to the Department of Oral and Maxillofacial Pathology, Faculty of Dentistry, Cairo University. The gross specimen showed a cystic lesion which showed areas of thickening. Hematoxylin and eosin stained sections revealed surface columnar and cuboidal epithelium covering a loose and myxoid fibrous tissue (Figure 2) and this specimen was diagnosed as a primordial odontogenic tumor. The patient was followed up for two years with no recurrence, and new bone formation was detected in the follow up radiographs (Figure 3).

Discussion
POT is a new entity first reported in a case series of 6 cases in 2014 described as benign mixed odontogenic tumor by Mosqueda-Taylor et al. Then another two cases were reported in 2015 and 2017 by Slater et al. and Ando et al. respectively then in 2018 Bajpai and Pardhe described another case. This novel lesion was added to the new WHO classification of odontogenic tumors.
Table 1 shows the clinicopathological and radiographic data of the nine documented cases. All reported patients were of young age group, ranging from 3–19 years with almost equal sex predilection and with posterior mandible as predominate site. All of these clinical finding are similar to this reported case.

All reported lesions were expansile and asymptomatic which are opposite to our case as it was painful during presentation, which may be the result of the previous marsupialization.

Radiographically, POT presents with a well-defined radiolucent lesion, either unilocular or bilocular, except in the case of Bajpai and Pardhe who reported a case that appeared multilocular.

All documented cases shared similar histopathological criteria proposed by Mosqueda-Taylor et al., as did our present case, where loose and myxoid connective tissue stroma resembles the dental papilla covered by columnar epithelium of a single layer, with the epithelium resembling the inner enamel epithelium.

Regarding the treatment approach, all previously documented lesions were treated with enucleation with different periods of follow up and reported no recurrence, in line with our presented case.

In conclusion, this is the first report case of POT from Egypt after it was defined in the latest WHO classification. Differentiation between POT and other closely resembling odontogenic
Table 1. Clinicopathological and radiographic data of the nine documented cases of primordial odontogenic tumor. M: Male; F: Female; RL: Radiolucent; UL: Unilocular; ML: Multilocular; mm: millimeter.

<table>
<thead>
<tr>
<th>Study</th>
<th>Age</th>
<th>Gender</th>
<th>Site</th>
<th>Clinical Picture</th>
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</thead>
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<tr>
<td>Mosqueda-Taylor et al.</td>
<td>18 years</td>
<td>M</td>
<td>Posterior mandible</td>
<td>Asymptomatic buccal swelling</td>
<td>RL, UL, well defined, 45 x 40 mm</td>
<td>Enucleation, 20 years, uneventful</td>
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<td>Mosqueda-Taylor et al.</td>
<td>16 years</td>
<td>M</td>
<td>Posterior mandible</td>
<td>Asymptomatic, buccal and inferior mandibular cortical bone expansion.</td>
<td>RL, UL, well defined, 55 x 50 mm</td>
<td>Lost to follow-up</td>
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<tr>
<td>Mosqueda-Taylor et al.</td>
<td>16 years</td>
<td>M</td>
<td>Posterior mandible</td>
<td>Asymptomatic buccal swelling</td>
<td>RL, UL, well defined, 65 x 50 mm</td>
<td>Enucleation, 10 years, uneventful</td>
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<td>Mosqueda-Taylor et al.</td>
<td>3 years</td>
<td>F</td>
<td>Posterior mandible</td>
<td>Asymptomatic buccal and lingual bony expansion.</td>
<td>RL, biloculated, well defined, 90 x 70 mm</td>
<td>Enucleation, 9 years, uneventful</td>
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<tr>
<td>Mosqueda-Taylor et al.</td>
<td>13 years</td>
<td>F</td>
<td>Posterior mandible</td>
<td>Asymptomatic buccal swelling.</td>
<td>RL, biloculated, well defined, 80 x 50 mm.</td>
<td>Enucleation, 3 years, uneventful</td>
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<tr>
<td>Mosqueda-Taylor et al.</td>
<td>3 years</td>
<td>F</td>
<td>Posterior maxilla</td>
<td>Asymptomatic buccal and palatal bony swelling.</td>
<td>RL, UL, well defined, 35 x 30 mm.</td>
<td>Enucleation 6 months, uneventful</td>
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<td>Slater et al.</td>
<td>19 years</td>
<td>M</td>
<td>Posterior mandible</td>
<td>Asymptomatic buccal and lingual bony swelling</td>
<td>RL, UL, well defined, 25 x 19 mm</td>
<td>Enucleation, 6 months uneventful</td>
</tr>
<tr>
<td>Ando et al.</td>
<td>8 years</td>
<td>F</td>
<td>Posterior maxilla</td>
<td>Asymptomatic, buccal swelling</td>
<td>RL, UL, well-defined, 16 x 15 mm</td>
<td>Enucleation, 16 months, uneventful</td>
</tr>
<tr>
<td>Bajpai and Pardhe</td>
<td>17 years</td>
<td>M</td>
<td>Posterior mandible</td>
<td>Asymptomatic buccal swelling</td>
<td>RL, ML, well defined, 30 x 20 mm</td>
<td>Enucleation, 6 months, uneventful</td>
</tr>
</tbody>
</table>

tumors is crucial, especially in the case of odontogenic myxomas as it is a more aggressive tumor and requires more aggressive treatment.

The clinical, radiographical and histopathologic data of the nine previously documented cases in addition to our case will be useful to differentiate this new tumor from other odontogenic tumors, which resemble it histopathologically, to avoid unnecessary aggressive treatment modalities.

Consent
Written informed consent for publication of clinical details and images was obtained from the patient’s parent.

References

Data availability
Dataset 1: Raw histological image. A photomicrograph of Hematoxylin and eosin (H&E) stained sections showing primitive connective tissue stroma covered by columnar epithelium, (x200). 10.5256/f1000research.14735.d20221

Competing interests
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This is a well-written case report describing a rare entity which was recently described. I would encourage the authors to add more histopathological details and elaborate on the discussion section mainly focusing on important differentiating points with histopathological mimics.

Please click here for the annotated PDF of this article.

Is the background of the case's history and progression described in sufficient detail?
Yes

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?
Yes

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?
Yes

Is the case presented with sufficient detail to be useful for other practitioners?
Yes

**Competing Interests:** No competing interests were disclosed.

**Referee Expertise:** Neuropathology, Surgical Pathology

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