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CASE REPORTS

Recurrent Primordial Odontogenic Tumor: Epithelium-Rich Variant

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Abstract
Primordial odontogenic tumor (POT) is a rare, mixed odontogenic neoplasm composed of spindled and stellate-shaped cells in myxoid stroma resembling dental papilla, surfaced by cuboidal-to-columnar odontogenic epithelium. Most POTs present in the posterior mandible as a well-demarcated radiolucency associated with a developing tooth in children and adolescents. POT is treated conservatively with no recurrences documented to-date. To describe the clinicopathological features of a recurrent POT. A 19-year-old female presented with an asymptomatic swelling, and panoramic radiograph revealed a multiloculated radiolucency in the mandibular body and ramus, with buccal and lingual perforation. The tumor was composed of plump spindle and stellate cells in a delicately collagenous and myxoid stroma, surfaced by columnar epithelial cells with reverse nuclear polarization. There was extensive epithelial proliferation forming invaginations within the tumor mass and organoid/enamel organ-like structures with enameloid-like deposits, dentinoid, and dystrophic calcifications. This was similar to the POT that had been excised four years prior from the same location. The patient underwent hemi-mandibulectomy and currently is free of disease at a thirteen-month follow-up. This report describes the first recurrent POT exhibiting extensive epithelial proliferation.

Keywords Jawbones · Odontogenic tumors · Dental papilla · Inner enamel epithelium · Odontogenesis · Recurrent tumor

Introduction

Primordial odontogenic tumor (POT) is a rare mixed epithelial-mesenchymal neoplasm that was first described by Mosqueda-Taylor et al. [1] in 2014 and recognized by the World Health Organization in 2017 [2]. There are now twenty-three cases reported in the literature [1, 3–16]. This tumor has a predilection for children and adolescents and usually presents as a radiolucency in the mandible [17]. Histopathologically, POT consists of abundant fibromyxoid stroma with spindled and stellate-shaped cells resembling dental papilla. The tumor mass is surfaced by epithelium resembling enamel epithelium characterized by cuboidal to columnar cells with reverse nuclear polarization. Conventional treatment of POT is enucleation or excision with removal of the involved tooth, and there have been no reported recurrences [4, 17].

This is a report of a recurrent POT, the first that we are aware of in the English literature, emphasizing the clinicopathological characteristics and features that distinguish the current case from other reported POTs.

Case Report

A 19-year-old Hispanic female presented with a swelling on the right side of her face of 4-month duration. The patient had a POT excised four years ago in the same area, and this case had been previously reported in 2018 [7]. The patient had been lost to follow-up until this visit. Radiographic examination revealed a 4.5 × 4.0 cm well-demarcated,

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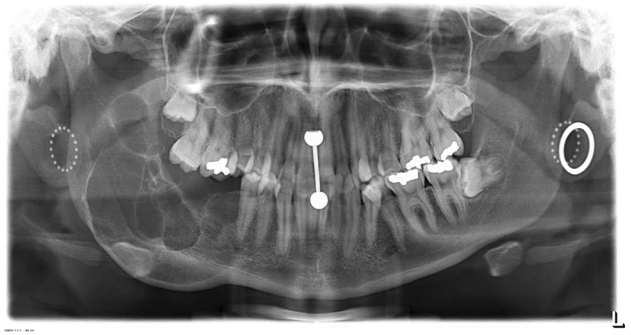


Fig. 1 Panoramic radiograph revealed a well-demarcated, multilocular radiolucency, measuring 5.5 × 4.0 cm in the right posterior mandible, in the area of missing right mandibular molars, extending to the right ramus and causing perforation of the superior cortex

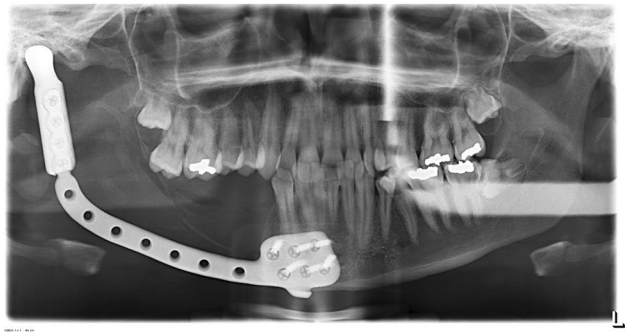


Fig. 2 Panoramic radiograph after tumor resection with the mandible reconstruction plate and condyle replacement

multilocular radiolucency in the area of missing right mandibular molars (removed during previous surgery) with extension to involve the entire right ramus (Fig. 1). The patient underwent right hemi-mandibulectomy from the right first premolar posteriorly with disarticulation. Mandibular reconstruction was performed, and a prosthetic condyle was placed (Fig. 2).

Gross examination of the resected mandible showed a multilobulated tumor with perforation of the buccal and lingual cortical plates and extensive involvement of the ramus and the inferior portion of the coronoid process (Fig. 3, upper images). The tumor was a yellow to tan multilobulated mass with a bosselated surface, with gelatinous and solid components (Fig. 3, bottom image). Histological examination revealed a partially encapsulated tumor with both epithelial and primitive, and cellular ectomesenchymal components with cystic change, and myxoid stroma (Fig. 4A). The tumor had an undulating surface and was covered by epithelium that formed deep invaginations (typical appearance of a POT), but also contained organoid structures that resembled rudimentary enamel organ-like structures (Fig. 4B). The epithelium was composed of columnar cells with clear cytoplasm and ovoid-to-round nuclei exhibiting reverse nuclear polarization, resembling ameloblasts or enamel epithelium; it was sometimes associated with stellate reticulum; there was no evidence of nuclear atypia or mitotic figures (Fig. 4C). There was subepithelial cellular condensation of mesenchymal cells forming a cambium layer (Fig. 4C). The epithelium often formed interconnected trabeculae in a plexiform pattern surrounding coalescent hypercellular cambium zones. There was abundant enameloid and focal dentinoid deposition (Fig. 4D). The enameloid was associated with epithelial cells with moderate amounts of brightly eosinophilic cytoplasm. Wispy basophilic material reminiscent of enamel matrix was also present (Fig. 4E). The stroma was myxoid and variably collagenous, and the spindle cells had benign vesicular nuclei. Mitotic figures were not seen (Fig. 4F). These features were similar to the POT removed from the same site four years prior, except that the organoid/enamel organ-like structures were more abundant [7].

The epithelial cells demonstrated strong positivity for CK14, and the spindled and stellate-shaped cells within the ectomesenchymal condensation beneath the epithelium were positive for CD34 and CD56 (Fig. 5A-B). Nuclear positivity for MIB-1 was noted in less than 5% of the cells. Calretinin and BRAF were negative in the tumor cells. Based on the histologic features, a diagnosis of recurrent POT, epithelium-rich variant, was made. There was no evidence of recurrence at a thirteen-month follow-up.

Discussion

POT is a rare benign biphasic odontogenic tumor that was described in 2014 [1]. It has epithelial and mesenchymal components with the latter appearing to differentiate towards dental papilla. Since then, twenty-three cases of POT have been reported in the English literature [1, 3–16]. The age of patients with POT ranged from 2–19 with a mean of 11 years old, and there is equal sex predilection (Table 1). One of the cases was extra-osseous [14]. Most (82.6%) cases of POT presented with asymptomatic buccal and/or lingual cortical swelling with well-defined, unilocular (77.3%), multilocular (13.6%), or bilocular (9.1%) radiolucency associated with the crown of a developing tooth. The most commonly involved teeth in POT were the permanent third molars (12/23, 52.3%), primary molars (8/23, 34.8%), permanent first molars (1/23, 4.3%), permanent second premolars (1/23, 4.3%), and primary maxillary canines (1/22, 4.3%). All cases occurred in the posterior jawbones, with 69.6% involving the mandible. Radiographically, there was cortical expansion (36.4%), and tooth resorption (27.3%), and displacement (22.7%) to variable degrees. Two cases had small radiopacities within the radiolucency [8, 15]. Two cases reported cortical perforation, including the current case in both initial

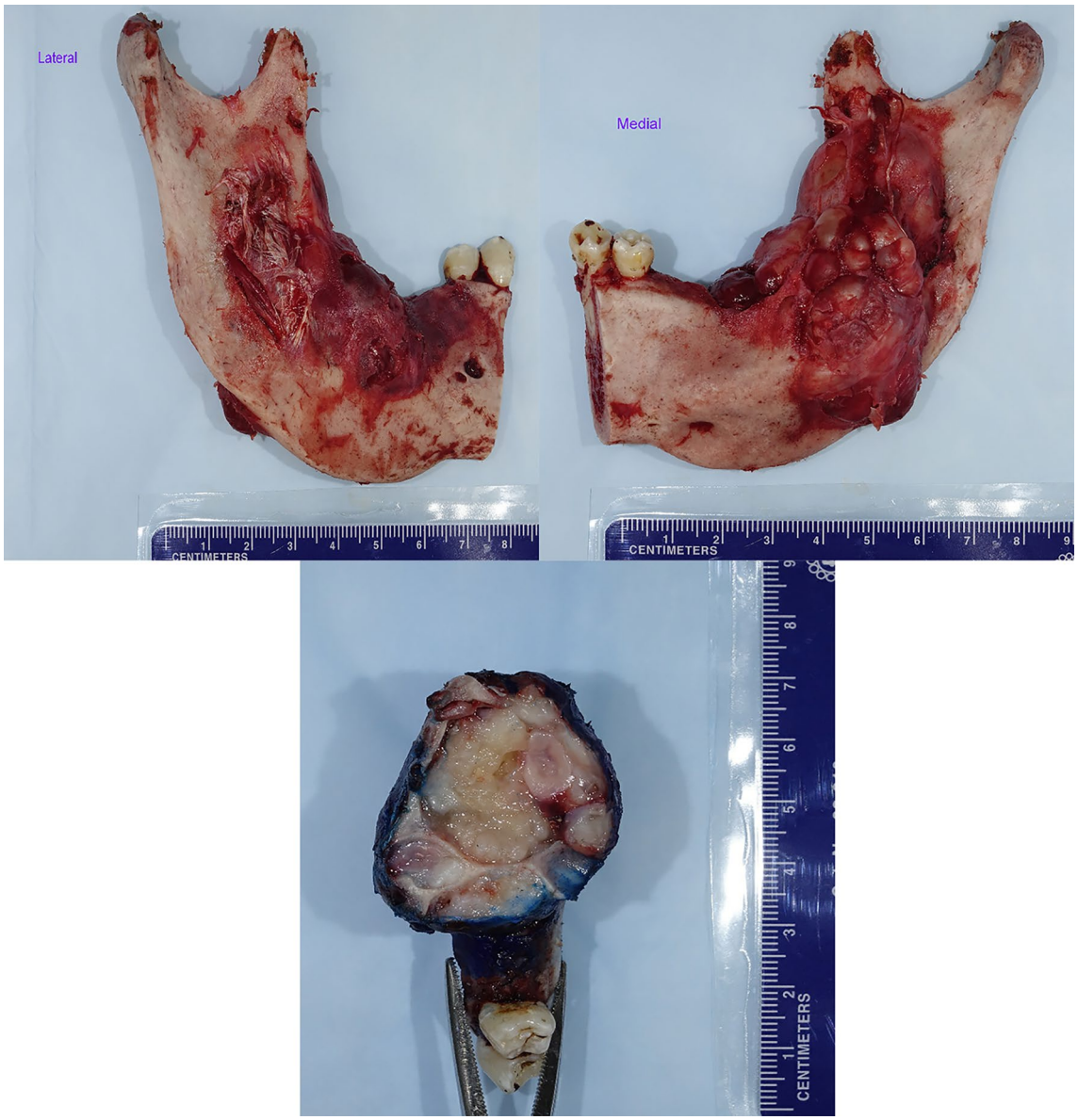
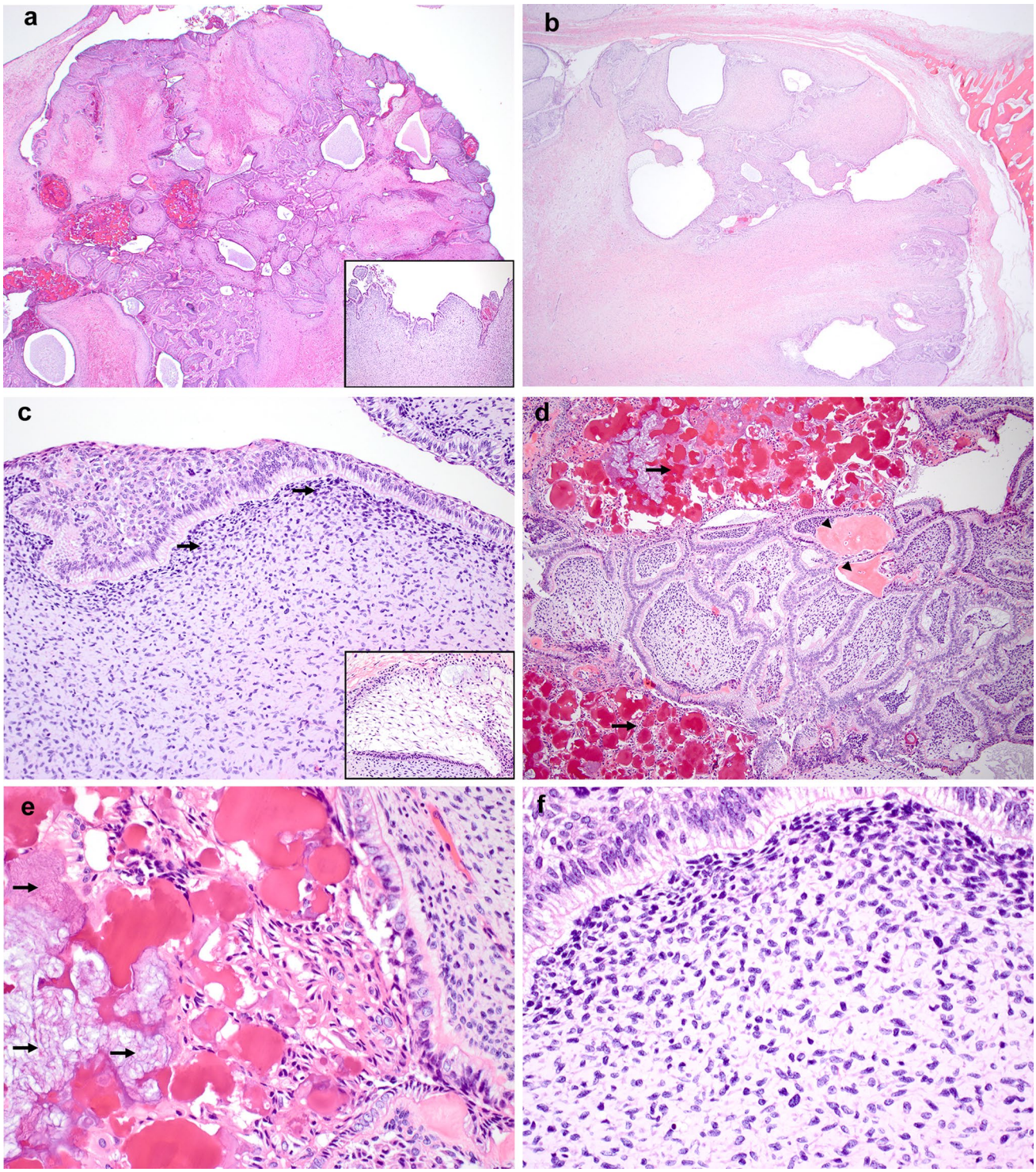


Fig. 3 Upper left and right hemi-mandible: Multilobulated tumor with buccal and lingual cortical perforation. Bottom: The gross specimen consisted of a yellow-tan solid mass with a papillary surface measuring 4 cm × 3.8 cm in the greatest dimensions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◂**Fig. 4** **A** The tumor was partially encapsulated and consisted of both epithelial and cellular ectomesenchymal components with cystic spaces, and myxoid stroma (H&E, original magnification × 40), Inset: the tumor showed an undulated surface lined by columnar epithelium (H&E, original magnification × 100). **B** The surface epithelium formed deep invaginations and organoid structures that resemble rudimentary enamel organ-like structures. Note the fibrous capsule of the tumor and cortical perforation (H&E, original magnification × 40). **C** The epithelium consisted of columnar epithelial cells with clear cytoplasm and reverse nuclear polarization with no evidence of nuclear atypia or mitotic figures with subjacent ectomesenchymal condensation (cambium layer) (arrows) (H&E, original magnification × 200), Inset: Some areas showed stellate reticulum-like tissue (H&E, original magnification × 200). **D** The epithelium formed interconnected trabeculae that surrounded the ectomesenchymal component that consisted of cellular spindled and stellate-shaped cells in a delicately collagenous and myxoid stroma with subepithelial condensation. Extensive enameloid deposition (arrows) and focal dentinoid like material (arrowheads) were present (H&E, original magnification × 100). **E** Brightly eosinophilic material resembling enameloid globules are associated with epithelial cells with abundant eosinophilic cytoplasm, and there is wispy basophilic material reminiscent of enamel matrix (arrows) (H&E, original magnification × 400). **F** The ectomesenchymal component of POT was myxoid with spindle cells in exhibiting benign vesicular nuclei, without cellular atypia or mitotic figures (H&E, original magnification × 200)

and recurrent presentations (Table 1) [7, 12]. The maximum dimension ranged from 1.2 to 9.0 cm with a mean of 3.8 cm.

Grossly, all POT cases, including the current case, showed a solid, multilobulated mass with focal gelatinous texture [17]. Histopathologically, POT is well-circumscribed, and a few cases showed, at least partial encapsulation by a thin fibrous band, although in many cases this feature was not described. There are two key histopathological features of POT: 1) Abundant delicately collagenous, fibromyxoid tissue with many plump spindled and stellate-shaped fibroblasts resembling dental papilla; 2) Odontogenic epithelium composed of cuboidal to columnar cells with clear cytoplasm and reverse nuclear polarity resembling the inner enamel epithelium, present at the tumor periphery. Stellate reticulum-like areas were present adjacent to the odontogenic epithelium in almost half of the cases (11/23; 47.8%) (Table 2). The majority of the cases (16/23; 69.6%) showed cell-rich mesenchymal condensation beneath the odontogenic epithelium forming a cambium layer. The cells here were CD34 positive. Surface undulation and epithelium invaginations were also noted (13/23; 56.5%) (Table 2). Unlike other cases, the current case showed florid epithelial invagination and proliferation in a plexiform and interlacing pattern, even in the original tumor from four years ago (Fig. 4D); this is similar to one other case reported by Zeng et al.[12]. Six cases (26.0%) exhibited dystrophic calcifications and five cases (21.7%) enameloid globules, including the current case in both the original and recurrent tumor [5, 7, 8, 11, 12, 15, 16]. Two cases exhibited a prominent, clear cell population within the tumor [1, 7] while one case contained dentinoid [12]. The current case is the only reported case with rudimentary organoid or rudimentary enamel organ-like structures in both initial and recurrent presentation characterized by odontogenic epithelium with mesenchymal condensations often seen before hard tissue is deposited [7].

The histological differential diagnoses for POT include dental papilla, odontogenic myxoma, archegonous cystic odontoma (primordial odontogenic cyst with induction phenomenon), ameloblastic fibroma, and ameloblastic fibro-odontoma. Dental papilla has similar mesenchymal stroma but is surfaced by odontoblasts, which are not epithelial in nature; it also does not attain the size of POT (mean size of 3.8 cm) (Fig. 1) [17]. Odontogenic myxoma is a mesenchymal odontogenic tumor with occasional inactive, scattered odontogenic epithelial islands and is not surfaced by epithelium [18]. There is only one case of archegonous cystic odontoma reported so far in the literature [19]. It is also referred to as a primordial odontogenic cyst with induction phenomenon because it develops in the area of a missing tooth, with hard tissue deposition. Unlike POT, archegonous cystic odontoma is purely cystic, although it is possible that it may be the cystic form of a POT [19].

The two most important differential diagnoses are ameloblastic fibroma (AF) and ameloblastic fibro-odontoma (AFO), the latter reclassified by WHO in 2017 as an immature odontoma [2]. These are biphasic tumors composed of a mesenchymal proliferation of spindle and stellate cells with fibrous and myxoid stroma, similar to the stroma seen in POT. The epithelial component of AF and AFO is composed of islands and strands of ameloblastomatous epithelium in a follicular pattern that exhibits reverse polarization of basal cells with central stellate reticulum. In contrast, POTs are surfaced by a single layer of columnar/cuboidal epithelium with reverse nuclear polarization and adjacent stellate reticulum in about half (47.8%) of cases. Occasionally, the epithelial component invaginates into the underlying mesenchymal stroma. AF does not form hard tissues while AFO produces enamel matrix, dentin, dentinoid, and cemento-osseous

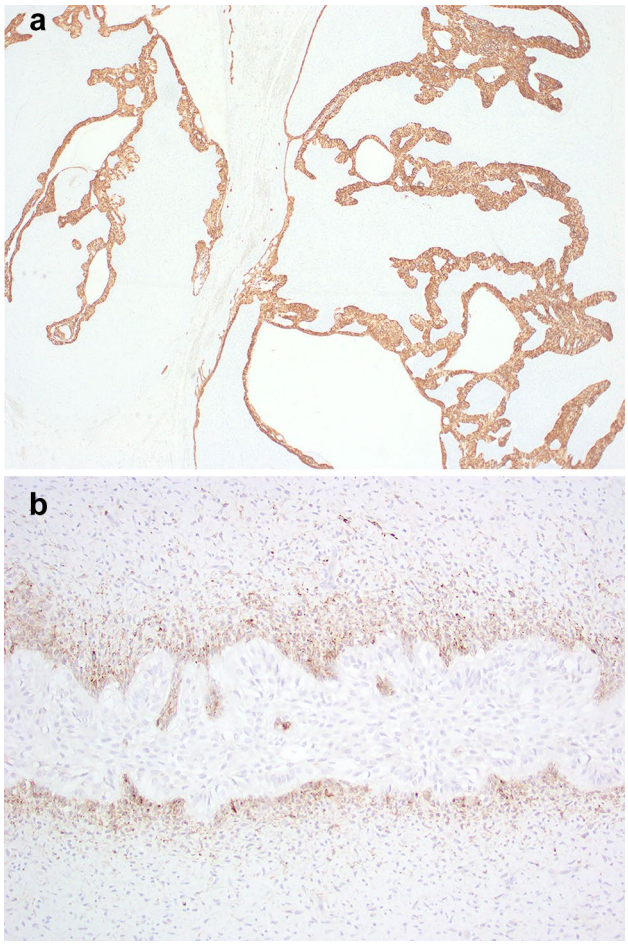

Fig. 5 **A** The study for CK14 showed strong cytoplasmic positivity in the epithelium (IHC, original magnification × 40). **B** The study for CD34 showed cytoplasmic positivity in the spindled and stellate-shaped cells in the ectomesenchymal condensation beneath the epithelium (IHC, original magnification × 200)

material. POT does not generally exhibit any dental tissue components except for five cases of POT that showed very focal enameloid deposition [7, 8, 11, 12] and one case that showed dentinoid deposition [12]. This current case showed both enameloid and dentinoid very focally.

Passador-Santos et al.[20] recently published a case of dental germ tumor in a 16-year-old female with well-defined mixed radiolucency associated with impacted right first mandibular molar. Histological examination revealed areas of cystic spaces lined by thin non-keratinized squamous epithelium, consistent with a dentigerous cyst. Within the lumen are POT-like areas with "dental germ-like formations" which resembled the rudimentary enamel organ-like structures noted in our case. In addition, there was extensive calcification and enameloid deposition. This might be the second case of an epithelium-rich variant of POT with extensive calcification and enameloid deposition. The patient was treated with tumor enucleation and curettage, and extraction of the first mandibular molar. The patient was free of disease after three years. Additionally, Zeng et al. [12] reported a case of an unusual mixed odontogenic tumor in a 12-year-old female, with features of POT that exhibited similar features to the current case, including florid epithelial proliferation and enameloid deposition with dystrophic calcification. Their case also showed dentinoid deposition. The patient was followed up for 13 years, with no sign of recurrence.

Sun et al.[21] reported a case of POT in a 10-year-old male that presented between the left mandibular canine and first premolar. This was a 0.5 cm radiolucency and the histology showed dental papilla-like tissue, enamel epithelium with stellate reticulum, and dentine deposition more consistent with a developing odontoma, as was noted by others [22]. Sato et al.[14] reported an intriguing case of peripheral POT in a 3 year-old-male that presented as gingival swelling associated with a radiolucency around an unerupted right maxillary second deciduous molar. This appears to be the only such case in the literature.

Most reported POT cases were treated with either enucleation/curettage (13/23; 56.5%) or tumor excision (9/23; 39.1%). In one case, the patient underwent partial mandibulectomy because the initial diagnosis was odontogenic myxoma [8]. None of the reported cases of POT showed recurrence on follow-up (range 6 months-20 years), except the current case where the patient exhibited recurrent tumor four years after the initial presentation.

This is a report of a POT, a rare mixed/biphasic odontogenic tumor that recurred four years after excision. The striking difference between the original and recurrent tumors that differentiates this POT from others that have been reported in the literature is the florid epithelial proliferation that forms organoid/rudimentary tooth germ-like structures with enameloid and dentinoid deposition. It may be that such epithelium-rich variants may be more likely to recur, but this will only become clearer as more cases are published.

Table 1 Summary of clinical and radiographic features of published cases of POT

	Age in years / gender	Location	Radiographic presentation	Treatment	Follow up
Mosqueda-Taylor et al. [1]	18/M	Left posterior mandible	RL, UL, well defined, 4.5 × 4.0 cm, surrounding the crown of the third molar	Enucleation and tooth extraction	20 years, NED
	16/M	Left posterior mandible	RL, UL, well defined, 5.5 × 5.0 cm, surrounding the crown of the third molar with jaw expansion	Enucleation and tooth extraction	LFU
	16/M	Left posterior mandible	RL, UL, well defined, 6.5 × 5.0 cm, surrounding the crown of the third molar	Enucleation and tooth extraction	10 years, NED
	3/F	Left posterior mandible	RL, BL, well defined, 9.0 × 7.0 cm, surrounding the crowns of the second deciduous and first permanent molars with jaw expansion, and tooth displacement	Enucleation and teeth extraction	9 years, NED
	13/F	Left posterior mandible	RL, BL, well defined, 8.0 × 5.0 cm, surrounding the crown of the third molar	Enucleation and tooth extraction	3 years, NED
	3/F	Left posterior maxilla	RL, UL, well defined, 3.5 × 3.0 cm, surrounding the crowns of second deciduous and first permanent molars with jaw expansion and tooth displacement	Enucleation and teeth extraction	6 months, NED
Slater et al. [3]	19/M	Right posterior mandible	RL, UL, well defined, 2.5 × 1.9 cm, surrounding the crown of the third molar with jaw expansion and tooth resorption	Excision and tooth extraction	7 months, NED
Ando et al. [4]	8/F	Left posterior maxilla	RL, UL, well defined, 1.5 × 1.5 cm, associated with crown of the first deciduous molar with jaw expansion and tooth displacement	Enucleation	16 months, NED
Mikami et al. [5]	5/M	Right posterior mandible	RL, UL, well defined, 1.5 × 0.5 cm, associated with crown of second deciduous molar	Excision and tooth extraction	7 months, NED
Amer et al. [6]	2/F	Right posterior mandible	RL, ML, well defined, 3.0 × 4.0 cm, associated with the crown of deciduous molar with jaw expansion	Excision and tooth extraction	2 year, NED
Pardhe et al. [9]	17/M	Left Posterior mandible	RL, ML, well defined, 9.0 × 5.0 cm associated with the crown of the third molar with multiple teeth resorption	Enucleation with tooth extraction	6 months, NED
Almazyad et al. [7]	15/F	Left posterior mandible	RL, ML, well defined, 3.5 × 2.0 cm, associated with the crown of the third molar with cortical perforation and tooth resorption	Excision and tooth extraction	Recurrence after 4 years
	18/M	Left posterior mandible	RL, UL, well defined, 1.2 × 0.7 cm, associated with the crown of the third molar	Curettage and tooth extraction	20 months, NED
Bomfim et al. [11]	4/M	Left posterior mandible	RL, UL, well defined, 3.0 × 2.0 cm, associated with the crown of the deciduous second molar with jaw expansion and tooth resorption	Excision and tooth extraction	LFU
Teixeira et al. [10]	13/F	Left posterior mandible	RL, UL, well defined, 3.0 × 4.0 cm, associated with the crown of the third molar	Excision	Not mentioned
Poomsawat et al. [8]	17/F	Left posterior mandible	RL, UL, well defined, 3.0 × 2.5 cm associated with the crown of the third molar with internal radiopaque material	Partial mandibulectomy	18 months, NED

Table 1 (continued)

	Age in years / gender	Location	Radiographic presentation	Treatment	Follow up
Zeng et al.[12]	2/M	Right posterior maxilla	RL, UL, well defined, 2.5 × 2.2 cm, associated with the crown of first deciduous molar with jaw expansion and perforation	Enucleation	11 months, NED
	12/F	Right posterior maxilla	RL, UL, well defined, 3.5 × 2.5 cm, associated with the crown of the third molar with multiple teeth resorption	Excision	13 year, NED
Delgado-Azanero et al. [13]	12/F	Right posterior mandible	RL, UL, well defined, 3.5 × 3.0 cm, associated with the crown of second premolar with tooth displacement and resorption	Enucleation with tooth extraction	15 months, NED
	13/F	Left posterior mandible	RL, UL, well defined, 2.5 × 2.0 cm, associated with the crown of third molar	Enucleation	5 years, NED
Kayamori et al. [15]	10/M	Right posterior maxilla	RL, UL, well defined, 1.7 × 1.5 × 1.0 cm, associated with the crown of deciduous second molar with jaw expansion, and internal radiopaque material	Enucleation	30 months, NED
Naina et al. [16]	14/M	Right anterior maxilla	RL, UL, well defined, 3.0 × 2 cm, associated with crown of the canine with jaw expansion	Excision	3 years, NED
Sato et al.[14]*	3/M	Right posterior maxilla	RL, ill-defined, 1.3 × 1.5 cm, associated with second deciduous molar	Excision	3 years, NED
Possible cases of POT					
Passador-Santos et al.[20]	16/F	Right posterior mandible	RL, UL, well defined, 3.0 × 5.0 cm associated with second molar with multiple radiopacities	Enucleation and curettage	3 years, NED

* peripheral/extra-osseous lesion

RL radiolucent, *UL* unilocular, *NED* no evidence of disease, *LFU* lost to follow up, *BL* biloculated, *ML* Multilocular

Table 2 Summary of histopathological features of published cases of POT

	Stellate reticulum	Mesenchymal condensation	Surface undulation/ epithelial invaginations/ proliferation	Product deposition	Other features
Mosqueda-Taylor et al. [1]	Absent	Present	Surface undulation	Not seen	Fibrous capsule
	Present	Absent	None	Not seen	
	Absent	Absent	Epithelial invaginations	Not seen	Clear cell population
	Absent	Present	Epithelial invaginations	Not seen	
	Present	Absent	Surface undulation and epithelial invaginations	Not seen	Fibrous capsule
	Absent	Present	Surface undulation	Not seen	
Slater et al. [3]	Present	Present	None	Not seen	
Ando et al. [4]	Present	Present	Epithelial invaginations with papillary configuration	Not seen	
Mikami et al. [5]	Absent	Present	None	Dystrophic calcifications	
Amer et al. [6]	Absent	Absent	Surface undulation	Not seen	
Pardhe et al. [9]	Absent	Present	None	Dystrophic calcifications	
Almazyad et al. [7]	Present	Present	Epithelial invaginations, papillary configuration and extensive epithelial proliferation	Enameloid, dystrophic calcifications	Fibrous capsule, clear cell population, and rudimentary enamel organ
	Absent	Present	None	Not seen	
Bomfim et al. [11]	Present	Present	Epithelial invaginations	Enameloid, dystrophic calcifications	
Teixeira et al. [10]	Present	Present	None	Not seen	
Poomsawat et al. [8]	Present	Present	Surface undulation	Enameloid, dystrophic calcifications	Fibrous capsule
Zeng et al.[12]	Absent	Present	Papillary configuration	Enameloid	
	Present	Present	Surface invaginations, papillary configuration and extensive epithelial proliferation	Dentinoid and enameloid deposition and dystrophic calcifications	
Delgado-Azanero et al. [13]	Present	Present	None	Not seen	
	Absent	Present	Surface undulation	Not seen	
Kayamori et al. [15]	Present	Absent	Surface undulation and invaginations	Dystrophic calcifications	Fibrous capsule
Naina et al. [16]	Absent	Absent	None	Dystrophic calcifications	Fibrous capsule
Sato et al.[14]*	Absent	Absent	None	Not seen	Unable to evaluate the epithelial lining
Current case	Present	Present	Surface invaginations, papillary configuration and extensive epithelial proliferation	Dentinoid, enameloid, enamel matrix like material	Fibrous capsule, and rudimentary enamel organ
Possible cases of POT					
Passador-Santos et al.[20]	Present	Present	Surface invaginations, papillary configuration and extensive epithelial proliferation	Dentinoid, Enameloid, dystrophic calcifications	Reduced enamel cell-like population

*peripheral/extraosseous

Author's Contributions David Collette provided clinical information, radiographic imaging and follow up information of the case reported. Dahua Zhang provided gross images and histological slides. Asma Almazyad contributed substantial in data acquisition, data interpretation and manuscript writing and drafting. Sook bin Woo has drafted and revised manuscript for intellectual content.

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Availability of Data and Material Upon request with no patient identifiers.

Declarations

Conflict of interest No conflict of interest to disclose.

Ethical Approval All study procedures were performed in accordance with the Declaration of Helsinki, 1964 and our Institution Research Committee Regulations.

Consent for Participate The patient is aware and consented of the previous and current publications.

Consent for Publication All co-authors read and approved the manuscript.

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