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CASE REPORT

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A case of peripheral odontogenic fibroma arising in the mandibular premolar region of a teenager

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Abstract

Peripheral odontogenic fibroma (POdF) is a rare, benign ectomesenchymal tumor. Herein, we report a case of a 15-year-old female patient who developed POdF in the mandible. The lesion was resected along with the periosteum. Histopathological findings revealed a small mass and cord-like epithelium. There was no recurrence 16 months postoperatively.

KEYWORDS

mandibular premolar region, odontogenic epithelium, peripheral odontogenic fibroma, teenager

1 | INTRODUCTION

Peripheral odontogenic fibroma (POdF) is a rare, benign ectomesenchymal tumor that most often occurs in adult female patient, frequently in the mandible and anterior maxilla.[1,2] Histopathologically, the tumor consists of fibrotic tissue with odontogenic epithelium and hard tissue showing varying degrees of calcification, but its expression is rare.[3,4]

Along with a review of the literature, here we report a case of POdF arising in the mandibular premolar region of a teenager.

2 | CASE STORY/EXAMINATION

A 15-year-old girl with a chief complaint of swelling in the right mandibular premolar region was referred to a local dental clinic for the treatment of dental caries and then to our hospital for treatment of the mandibular tumor. The patient first recognized a painless mass in her mandible at 6 years of age. Her medical history was significant for a history of surgery for congenital ear fistula. Intraoral examination revealed a well-defined, painless, elastic, firm mass, measuring 17×17 mm in size, between the right mandibular canine and first premolar (Figure 1).

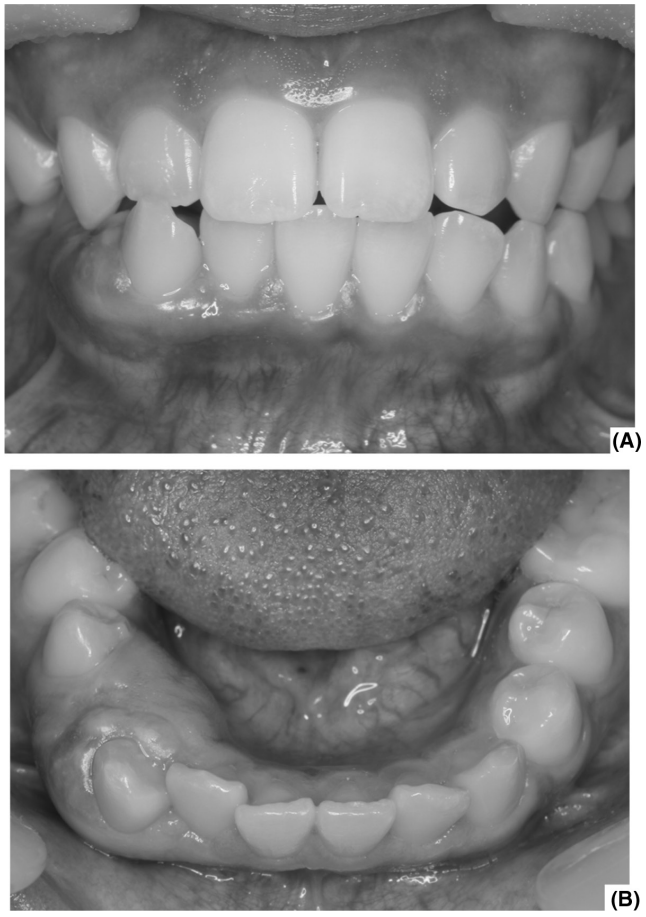

FIGURE 1 Intraoral view. (A,B) Intraoral view revealed a well-defined, painless, elastic, firm mass, measuring 17×17 mm in size, between the right mandibular canine and first premolar

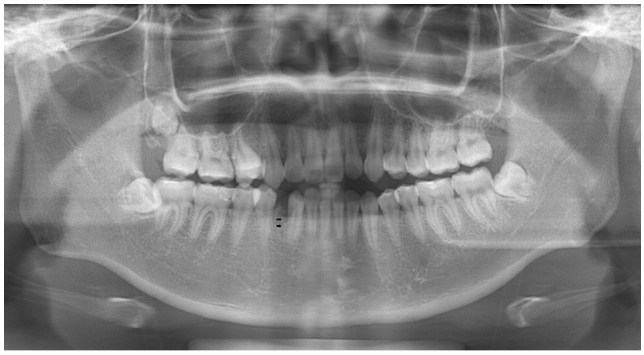

FIGURE 2 Panoramic X-ray. Panoramic radiography showed a radiolucent area in the alveolar bone between the right mandibular canine and first premolar

Panoramic radiography showed a radiolucent lesion in the alveolar bone at the same location (Figure 2). In addition, sandy calcifications were observed inside the lesion. Computed tomography (CT) revealed compressive bone resorption of the alveolar bone between the mandibular right canine and first premolar (Figure 3). Magnetic

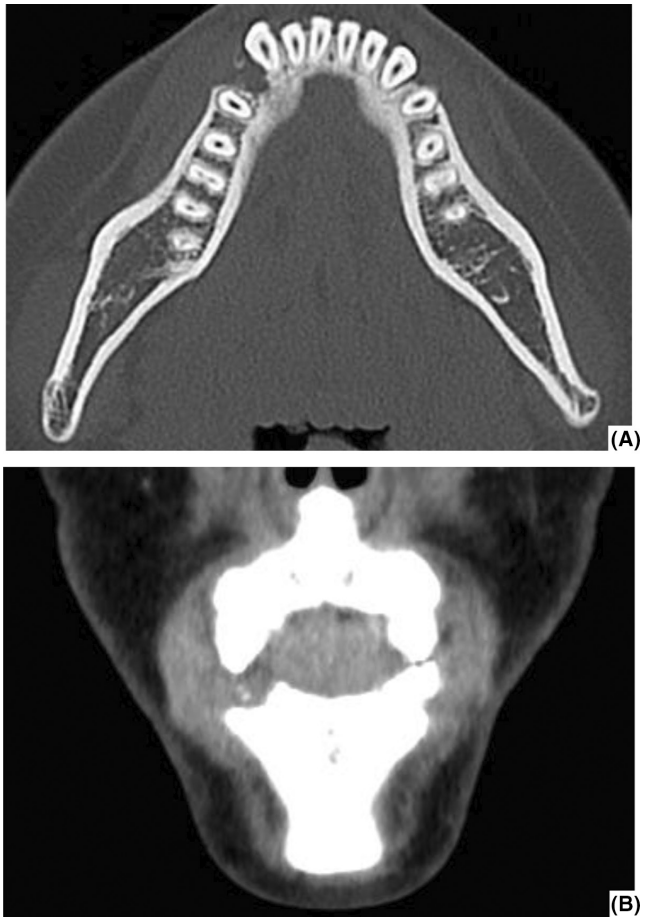

FIGURE 3 Computed tomography findings. (A,B) Compressive bone resorption of the alveolar bone between the right canine and first premolar of the mandible

resonance imaging (MRI) revealed endophytic characteristics of a well-defined enhancing mass on the right side of the mandible on T2-weighted imaging (Figure 4).

A biopsy was performed under local anesthesia, and a benign tumor with fibroblastic proliferation was diagnosed. Tumor resection was performed under general anesthesia. At the time of resection, a definitive diagnosis could not be obtained by biopsy and other tumors such as epulis, which originate from the periosteum, could not be ruled out. Hence, the entire tumor was detached from the surrounding bone surface and resected along with the periosteum. A bone depression was observed around the tumor, and the bone surface was smooth. The tumor was 17×17 mm in size and had a smooth surface. Histopathological examination revealed spindle-shaped fibroblast-like cells that spread subcutaneously. Scattered cord-like and lump-like epithelial components were observed in the fibrous tissue near the base of the tumor (Figure 5A). No atypical cells or nuclear fission were observed. Cementum- or bone-like calcifications were observed in the deep area of the tumor. Epithelial components resembled the epithelial cell rests of Malassez and

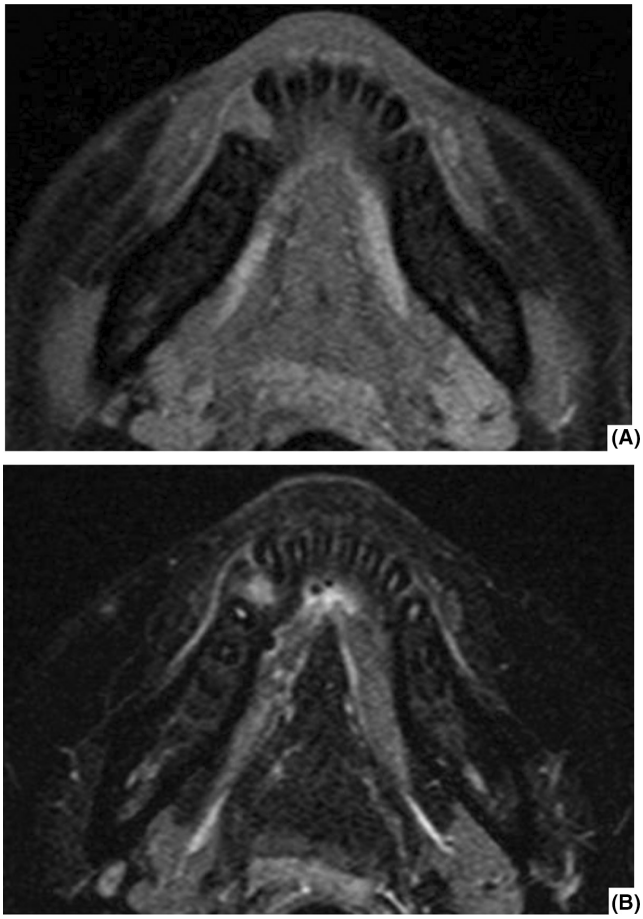

FIGURE 4 Magnetic resonance imaging findings. (A,B) Endophytic appearance of a well-defined enhancing mass on the right side of the mandible on T2-weighted images

Hertwig's epithelial root sheath and were positive for cytokeratin (CK) CK19 staining (Figure 5B,C). There were no signs of postoperative recurrence after 16 months.

3 | DISCUSSION

Gardner explained that odontogenic fibromas can be subdivided into central and peripheral variants. The peripheral variant, POdF, can be histopathologically differentiated from peripheral ossifying fibroma.[5] POdF is a rare odontogenic tumor with gradual growth that presents as a firm, elastic, smooth gingival mass.[1,3,4]

The onset age of POdF is 5 months to 84 years, and there is seemingly a peak in middle age, specifically, during the forties.[1,2,6,7] The most common sites to develop POdF are known to be the anterior maxilla and mandible in adults, while some reports show that POdF specifically occurs more frequently in the mandibular canine to premolar region.[7,8] A total of 25 reports of POdF occurring in patients younger than 19 have been published, of which twelve reported POdF in the mandible (Table 1).[6,7,9–18]

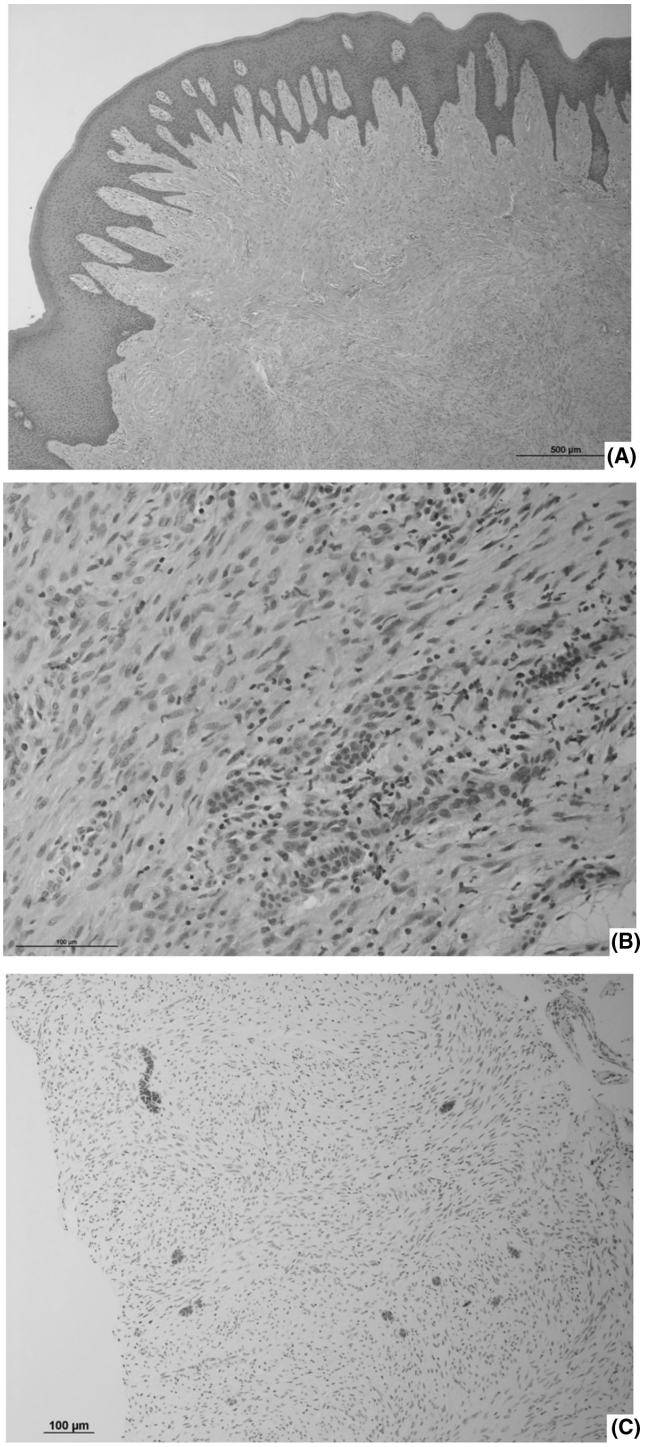

FIGURE 5 Histopathological findings. (A) Scattered cord-like and lump-like epithelial components were observed in the fibrous tissue near the base of the tumor (H-E staining, ×40). (B) Cementum- or bone-like calcifications were observed (H-E staining, ×400). (C) Epithelial components resembled the epithelial cell rests of Malassez and Hertwig's epithelial root sheath and were positive for CK19 (CK19 staining, ×40)

Among those, two cases, including the present case, reported POdF occurring in the mandibular premolar region of teenagers.[10]

TABLE 1 Reports of peripheral odontogenic fibroma occurring in patients under 19 years of age

Case No.	Study	Year	Age	Sex	Location	Hard tissue
1	Nakashiro et al.[9]	1966	1	F	Maxilla incisor	–
2	Hasegawa et al.[10]	1975	11	M	Mandibular premolar	Cementum like
3	Weber et al.[15]	1992	8	F	Maxilla and mandibular incisor	–
4		1992	3	M	Maxilla and mandibular incisor	–
5	Fujimura et al.[11]	1992	3	M	Mandibular incisor	–
6	Daley et al.[7]	1994	13	M	Mandibular molar	Unknown
7		1994	12	F	Maxilla incisor	Unknown
8		1994	14	M	Maxilla molar	Unknown
9	Katano et al.[12]	1998	2	M	Mandibular molar	–
10	Siar et al.[6]	2000	5 M	Unknown	Unknown	Unknown
11		2000	4	Unknown	Unknown	Unknown
12		2000	9	Unknown	Unknown	Unknown
12		2000	11	Unknown	Unknown	Unknown
14	Kawano et al.[13]	2003	7	F	Maxilla molar	–
15	Martelli et al.[16]	2006	4 M	F	Maxilla incisor	–
16		2006	12	F	Maxilla molar	Unknown
17		2006	15	M	Mandibular molar	Unknown
18		2006	12	F	Maxilla incisor	Unknown
19	Alaeddini et al.[17]	2010	15	F	Mandibular	Bone like
20		2010	12	M	Mandibular	Cementum Like
21		2010	8	M	Mandibular	–
22		2010	9	M	Mandibular	–
23	Becker et al.[18]	2015	3	F	Mandibular incisor	–
24	Sugano et al.[14]	2017	3	F	Mandibular molar	–
25	Current case	2021	15	F	Mandibular premolar	Bone like

Imaging usually depicts sand-like calcifications inside the mass and compressive resorption of the surrounding bone.[17] However, there have been no reports of MRI findings of POdF to date. In our case, we reported the first MRI findings of POdF: an endophytic appearance of a well-defined enhancing mass on the right side of the mandible on T2-weighted images.

Histopathologically, POdF is characterized by odontogenic epithelium scattered to various degrees in the fibrous substrate.[3,5,7] It is classified into epithelium-poor or epithelium-rich subtypes according to the amount of dentin epithelium.[19] In addition, bone-like, dentin-like, or cementum-like hard tissue formation is usually observed, with a reported frequency of 28.3% of bone-like tissue and 15.2% of cementum-like tissue.[6] In the present case, the lesion was described as being histopathologically rich in cellular components as well as a mixture of small mass and cord-like epithelial components similar to the epithelial cell rests of Malassez and Hertwig's epithelial root sheath. In addition, bone-like calcifications were observed in the deep areas of the tumor.

To date, there have been only four reports of POdF with hard tissue inside the lesion among teenagers.[17] Although few reports have been published on hard tissue formation in POdF, it has been suggested that the odontogenic epithelium inside the lesion may induce hard-tissue formation, which requires a lengthy time to develop in teenagers.[17] Thus, it has been speculated that the frequency of calcification in POdF might be low.

Some reports of CK-positive cells in the odontogenic epithelium of POdF exist.[20] In the present case, CK19-positive epithelial components resembled the epithelial cell rests of Malassez and Hertwig's epithelial root sheath.

Central odontogenic fibromas originate from the periodontal ligament or dental follicle according to the 2005 World Health Organization classification, while other authors have reported the possibility of periodontal ligament origin.[19] On the contrary, POdF

is hypothesized to originate from the periodontal ligament, dental follicle, and dental papilla.[17] We believe the periodontal ligament or dental follicle to be the origin of the tumor in this case for the following reasons: The tumor was observed during the tooth-replacement period, tumor growth was observed from the neck of the adjacent tooth, fibroblast growth around the tumor did not resemble that of dental papilla, and cells similar to the epithelial cell rests of Malassez and Hertwig's epithelial root sheath were present.

The rate of POdF recurrence after surgery is reportedly low.[17,21] However, some studies have reported an early recurrence rate as high as 50% (29/58 cases).[2,22,23] Moreover, Patel S, et al[22] reported that the recurrence rate was higher in POdF when the periosteum was not resected during tumor removal; hence, they recommended resection of the periosteum for these tumors. In our case, the results of the biopsy did not completely rule out other tumors; hence, the periosteum was also resected.

4 | CONCLUSIONS

In this case, we resected a POdF with the periosteum and preserved the adjacent tooth; there were no signs of recurrence 16 months after the operation. We also reported the first MRI findings of POdF. Thus, this case demonstrates a novel imaging finding to help clinicians diagnose POdF and consider resection of the periosteum during treatment to minimize the risk of recurrence.

AUTHOR CONTRIBUTIONS

Katsuhisa Sekido and Kie Yamashiro involved in manuscript preparation. Masaharu Tatetsu, Masashi Harada, and Michiko Okita involved in patient management. Yasushi Hariya involved in manuscript preparation and patient management.

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CONFLICT OF INTEREST

The authors have no conflicts of interest to declare.

DATA AVAILABILITY STATEMENT

Data sharing is not applicable to this article as no new data were created or analyzed in this study.

CONSENT

Written informed consent was obtained from the patient and her parent to publish this report in accordance with the journal's patient consent policy.

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