J. Oral Diag. 2020; 05:e20200001.

CASE REPORT

Exuberant gingival lesion: a case of peripheral odontoma

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Abstract:

Odontomas are a benign lesion of odontogenic origin rarely found in deciduous dentition. We report herein a rare case of peripheral odontoma associated with an exuberant reactive gingival lesion. A two-year-old child was referred for evaluation due to asymptomatic gingival overgrowth. Intraoral examination revealed a firm consistency nodule in the superior anterior gingiva involving the left incisors. The radiographic image showed no alteration. The primary diagnostic hypothesis was a reactive proliferative process. An excisional biopsy was performed. Histopathological analysis revealed a peripheral complex odontoma associated with fibroepithelial hyperplasia. The patient completed treatment and was monitored with clinical and radiographic examinations for 16 months.

Keywords: odontoma; gingiva; pediatric dentistry

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Article received on April 30, 2020.
Article accepted on July 24, 2020.

DOI: 10.5935/2525-5711.20200001

INTRODUCTION

Odontomas are odontogenic tumors, which are considered nonaggressive, hamartomatous developmental malformations of dental tissues. Histologically, they are composed of variable amounts of enamel, dentin, cementum and pulp[1–3]. Their etiology is still unknown, although genetic and traumatic factors have been suggested as primary causes[4]. According to the latest taxonomy by the World Health Organization (WHO,2017), two types of odontomas exist: compound odontomas (single solid tumor mass) and complex (multiple tooth-like structures)[2].

Clinically, odontomas are classified as either intraosseous or extraosseous[1–5]. Intraosseous odontomas are asymptomatic intrabony lesions found in children with permanent dentition and are rarely found in deciduous dentition. They are identified during a routine radiological examination and sometimes can be associated with a disturbance in tooth eruption. In exceptional cases, the odontoma may spontaneously erupt into the oral cavity or cause other clinical complications[1–6]. An extraosseous or peripheral odontoma arising in the soft tissues is extremely uncommon and tends to exfoliate[7–10].

The ideal treatment option is surgical removal of the lesion in all cases, followed by histopathological examination of tissue with clinico-radiographic follow-up[1–4].

CASE REPORT

A two-year-old boy was referred to the pediatric dentistry clinic of the Federal University of Rio Grande do Sul for the examination of an overgrowth in the anterior region of his maxillary gingiva with 10 months duration. The patient was in good general health. His past medical and dental history did not reveal any significant events or trauma at the site. At the initial intraoral examination, a normal colored, sessile nodular lesion was identified in the upper anterior gingival mucosa involving the area of the left incisors. The area measured about 3 x 2cm and had a fibrous consistency upon palpation with no bleeding (Figure 1A). Periapical radiography revealed no bone alteration (Figure 1 B). Considering the clinical and radiographic findings, the main clinical diagnostic hypotheses were either a non-neoplastic proliferative process (inflammatory fibrous hyperplasia/irritative fibroma, pyogenic granuloma, peripheral ossifying fibroma or peripheral giant cell lesion) or a benign mesenchymal neoplasia. Following

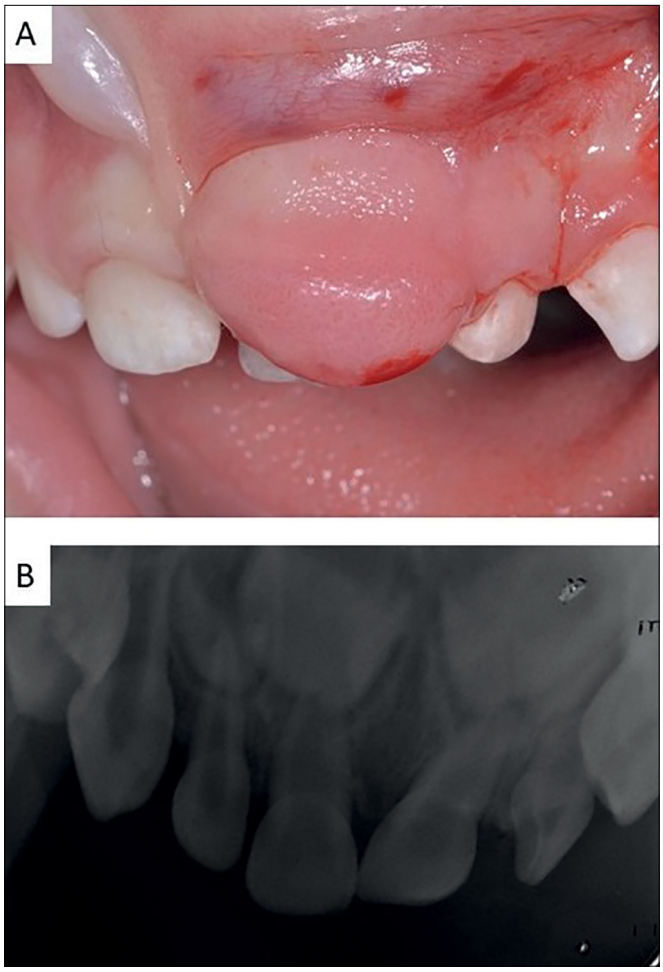

Figure 1. (A) Clinical presentation showing a nodular lesion in the upper anterior gingival mucosa involving the area of the left incisors. (B) Periapical radiography revealed no bone alteration.

those hypotheses, an excisional biopsy was performed with local anesthesia. During the surgical procedure, the lesion was easily detached from the tooth and gum. In addition, reabsorption of the maxillary vestibular cortical bone was observed. A small mineralized tissue segment was detached and was sent with the removed lesion for histopathologic examination. A delicate curettage with a periodontal curette was performed at the interface between the tooth and bone tissue with the intention of eliminating tissue remnants and preventing recurrence. Due to a small amount of local gingival tissue available for suturing, associated with the patient's behavioral issues, surgical cement was used (Periobond®) to assist in hemostasis and wound healing.

The microscopic examination of excised soft tissue surgical specimens revealed an unencapsulated lesion (Figure 2A). The oral mucosa was covered by hyperplastic keratinized stratified squamous epithelium

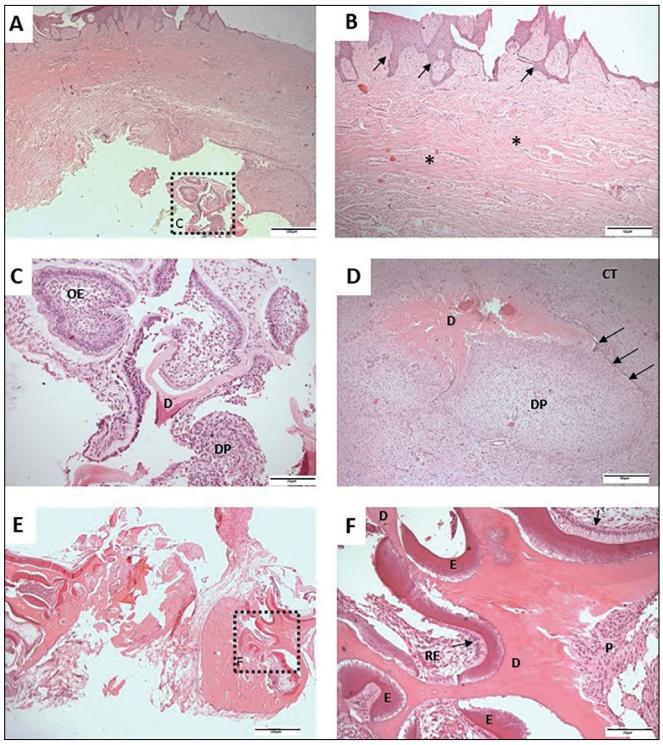

Figure 2. Histopathological features of the peripheral odontoma and fibroepithelial hyperplasia. (A) Oral mucosa covered by hyperplastic keratinized stratified squamous epithelium. Dense fibrous underlying connective tissue with scarce chronic inflammatory infiltrate. Also, a newly forming odontoma is present in the deep of connective tissue (HE, x40, original magnification). (B) Hyperplastic epithelium (arrows) and increased collagen deposition (asterisk) (HE, x100, original magnification). (C) Forming odontoma represented by cords of odontogenic epithelium (OE), dental papilla (DP) and dentin (D) deposition (HE, x200, original magnification). (D) In a close relationship with the oral mucosa connective tissue, an immature peripheral odontoma could be observed. The dental papilla (DP) is partially surrounded by odontogenic epithelium (arrows). Dentin (D) is also observed (HE, x100, original magnification). (E) A low-power view of decalcified samples revealing a complex odontoma with a haphazard deposition of odontogenic tissues (HE, x40, original magnification). (F) Irregular dentine masses (D), enamel (E) matrix surrounded partially by enamel epithelium. Stellate reticulum (SR) of enamel organ, elongated epithelial cells similar to ameloblasts (arrows), enamel matrix (E) and dental papilla can be observed. (HE, x200, original magnification).

and dense fibrous underlying connective tissue with scarce chronic inflammatory infiltrate (Figures 2A and 2B). The deepest part of the sample areas had an island and cords of odontogenic epithelium within the surrounding fibrous connective tissue. Also, dental papilla and deposition of dentin matrix were observed (Figures 2A and 2C). In another area, an immature/initial forming odontoma could be detected. Please note that this lesion had developed in the oral mucosa connective tissue. Dental papilla surrounded partially by odontogenic epithelium and dentin were observed (Figure 2D). The decalcified section showed a complex odontoma characterized by haphazard deposition of irregular dentine masses, including an enamel matrix surrounded partially by enamel epithelium and dental papilla (Figures 2E and 2F).

The final histopathologic diagnosis was complex odontoma and fibroepithelial hyperplasia. Postoperatively there were no complications. The patient was followed at intervals of 15 days (Figure 3A), 60 days (Figure 3B), and 180 days (Figure 3C). A radiographic evaluation at 180 days showed no alteration (Figure 3D).

DISCUSSION

Odontoma is classified as an odontogenic tumor having an epithelial and mesenchymal origin, however, it is more widely understood as a developmental malformation of dental tissues. It is one of the most prevalent odontogenic tumors commonly diagnosed between the first and second decade of life[1–3]. However, extraosseous lesions (peripheral odontomas) are quite rare and reports of them in the literature are limited[7–10]. Here, we present a case of peripheral complex odontoma associated with a reactive growth of soft tissue leading to the formation of fibroepithelial hyperplasia.

Clinically, peripheral odontomas are usually diagnosed between the first and the second decade of life[7–9] in general, 10 years earlier than its central counterpar[9,11]. There is no gender predilection[7,9,11]. The most prevalent site is the maxillary anterior region[7,11] with many reports describing the lesion as appearing in a lingual position[7]. Most peripheral odontomas are relatively small, rarely exceeding 1cm in diameter and, over time, some of them probably exfoliate[11]. In the present case, we have reported that the

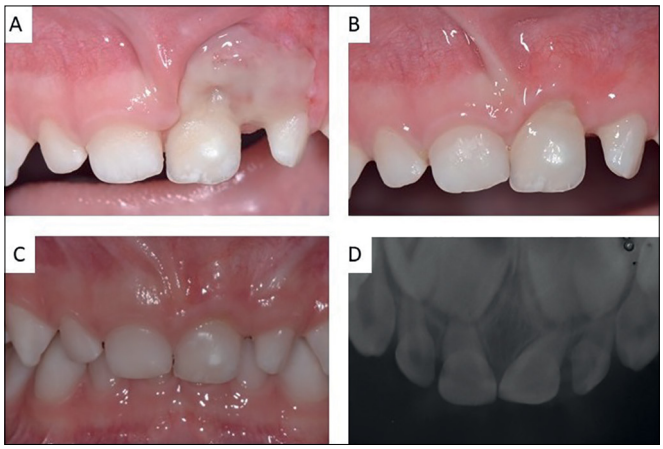

Figure 3. Clinical and radiographic follow up. (A) Clinical aspects after 15 days of surgical procedure exhibiting ulcerative area. (B) Clinical aspect after 60 days showing a small gingival defect. (C) After 180 days, an evaluation showed excellent healing of the oral mucosa. (D) Periapical radiograph after 180 days showing an absence of bone lesions.

clinic-demographic characteristics of the patient agree with the literature except for the vestibular position of the lesion[7–12].

The present case is very intriguingly because the patient presented with a rapidly growing, single gingival lesion with no osseous alteration identified by a periapical radiograph. Based on these aspects our main provisional hypothesis was a non-neoplastic proliferative process (inflammatory fibrous hyperplasia/irritative fibroma, pyogenic granuloma, peripheral ossifying fibroma or peripheral giant cell lesion) or benign mesenchymal neoplasia. These hypotheses agreed with the Tamiolakis et. al (2018)[13] study that evaluated 1,187 localized gingival enlargements and showed that 85.17% of gingival lesions were reactive in origin. In addition, these authors reported that in the first decade of life, the most common lesions in gingiva were pyogenic granulomas, peripheral ossifying fibromas and peripheral giant cell lesions. In addition, reviewing the reports about peripheral odontoma, it is possible to observe that this lesion is frequently misdiagnosed clinically due to other common reactive soft tissue lesions as mentioned above[9,14].

On radiographic evaluation, complex odontomas (intra or extraosseous) may be observed as a dense radiopaque mass, whereas compound odontomas appear as several malformed teeth[1,3,5]. In the present case, no radiographic alteration was detected in a periapical exam; this is similar to other cases of reported peripheral odontomas[12,15]. This could occur because an odontoma radiological appearance can vary according to the developmental stage of the odontoma and its amount of calcification. In the first stage, radiolucency can be detected due to a lack of calcification (soft odontoma); in the intermediate stage, partial calcification is seen, while in the third stage odontoma demonstrates radiopacity due to complete calcification with thin radiolucent periphery. In the present case, the two-year-old patient had an odontoma that probably was in the initial/intermediate stage or superposed to the permanent teeth image. This reinforces the importance of histopathological examination of all lesions.

Based on clinical and radiographic aspects we decided to perform an excisional biopsy. During the surgical procedure, all of the soft tissue lesion was removed, and a very small fragment of calcified tissue was observed. Our interpretation was that this calcified tissue could be produced by the lesion. This can occur in cases of peripheral ossifying fibroma and peripheral giant cell lesions. Or it could represent a remnant fragment of vestibular cortical bone. Surprisingly, the histopathological exam demonstrated a complex odontoma associated with fibroepithelial hyperplasia. In the soft tissue analysis, the presence of dental tissue outside the alveolar process (odontoma in formation) could be detected. The interaction of epithelial-mesenchymal odontogenic tissue forming a dentin and enamel matrix in the oral mucosa connective tissue was found, leading to the diagnosis of peripheral odontoma as previously described[7,8,10,12]. The small calcified tissue revealed a complex odontoma. Taking into consideration all of the clinical, radiograph and histopathological aspects we infer that this is a new case of peripheral odontoma.

The exact mechanisms involved in the pathogenesis of peripheral odontoma and fibroepithelial hyperplasia in the present case are not clear. For an odontoma, it has been suggested that some possible factors, such as genetic basis, infections, inflammation, trauma or even hyperactivity of odontoblasts, are involved. However, some authors have suggested that dental lamina and its remnants (rests of Serres) could be persistent and play a role in the development of peripheral odontomas based upon the capacity of epithelial–mesenchymal interactions[9,12]. No trauma or infection were reported in the medical history of the patient. We believed that the peripheral odontoma in the present case was in its early development stage. Although peripheral odontomas have a limited potential for growth, some cases reinforce the idea that if the lesion had not been surgically removed it might have erupted into the oral cavity. The eruptive mechanism of peripheral odontoma remains uncertain. In the present case, we believe that a peripheral odontoma stimulates the growth of adjacent oral mucosa tissue promoting the formation of fibroepithelial hyperplasia.

The treatment of peripheral odontomas consists of simple surgical excision and the possibility of recurrence is very low[9–12,14,15]. In the present case, no signs of recurrence were observed after xx months. Also, complete oral mucosa and bone tissue healing were detected. It is essential for oral health professionals to be familiar with peripheral odontoma. We concluded that even in rare lesions such as peripheral odontoma, if the professionals follow all the steps of the diagnostic process including meticulous history, clinical examination, and radiographic and histopathological tests, the correct diagnosis will be established, and the patient will receive optimal treatment.

CONSENT

Written informed consent was obtained from the patient for publication of this Case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

CONFLICT OF INTEREST

All authors declare no conflict of interest

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