Peripheral compound odontoma: A rare case report and literature review

Ondina Karla Mousinho da Silva Rocha DDS, MSc1 | Caio César da Silva Barros DDS, MSc1 | Luiz Arthur Barbosa da Silva DDS, MSc, PhD1 | Erasmo Freitas de Souza Júnior DDS, MSc2 | Hécio Henrique Araújo de Morais DDS, MSc, PhD2 | Márcia Cristina da Costa Miguel DDS, MSc, PhD1

1Postgraduate Program in Dental Science, Federal University of Rio Grande do Norte, Department of Dentistry, Natal, Rio Grande do Norte, Brazil
2State University of Rio Grande do Norte, Department of Dentistry, Natal, Rio Grande do Norte, Brazil

Correspondence
Márcia Cristina da Costa Miguel, Federal University of Rio Grande do Norte, Department of Dentistry, Postgraduate Program in Dental Science. Av. Senador Salgado Filho, 1787, Lagoa Nova, CEP 59056-000, Natal, RN, Brazil
Email: mccmiguel@hotmail.com

Abstract
Peripheral odontoma is a very rare odontogenic hamartoma arising in soft tissues. Here, we report a case of peripheral odontoma in a pediatric patient and review the cases published in the literature. An 11-year-old male patient presented a nodular lesion in the anterior region of the palate for over 1 year. Under the clinical hypothesis of fibroma, an excisional biopsy was performed. Histopathological examination revealed the presence of tooth-like structures, formed by enamel, and dentin matrix, occasionally associated with the dental papilla and surrounding pulp tissue, thus, the histopathological diagnosis of peripheral odontoma was established. The patient has been undergoing follow-up for 6 months without any signs of lesion recurrence. Peripheral odontomas are uncommon lesions that usually affect young patients and display a preference for the maxilla and limited growth potential. The recognition of the clinical and histopathological features of the peripheral odontoma is indispensable for the establishment of its diagnosis.

KEYWORDS
compound odontoma, diagnosis, hamartoma, pediatrics

1 | INTRODUCTION

Odontomas are common, usually intraosseous, odontogenic hamartomas, which interfere with the eruption of permanent and deciduous teeth.1,2 However, these lesions may also develop in an extraosseous manner, in soft tissue, termed peripheral odontomas (PO).3–5

PO originate from the stimulation of epithelial dental lamina remains trapped in the soft tissues of the maxillomandibular complex.2 Clinically, these lesions present as asymptomatic nodules, with limited growth, and displaying absence of bone involvement.3,6 In addition, these odontomas can be classified as either compound or complex, based on their histopathological characteristics.2–5 PO are rare, and few cases have been reported in the literature.4 In this context, the aim of this study is to describe a PO case, emphasizing its clinical-pathological characteristics, considering the cases published in the literature to date.

2 | CASE REPORT

A female patient, 11 years old, presented an asymptomatic sessile nodular lesion in the anterior region of the palate for over 1 year, with normal color, of hardened consistency, exophytic, and measuring 0.3 × 0.3 × 0.3 cm (Figure 1A). Periapical radiography was requested to exclude a periapical abscess. However, no radiographic changes were observed. (Figure 1B). Thus, the clinical hypothesis of oral
**FIGURE 1** A, Clinical aspect—Nodular lesion in the anterior region of the palate (arrows); B, Periapical radiography without evidence of lesion.

**FIGURE 2** Histopathological features (H&E)—A—1000 μm, benign odontogenic tumor surrounded by predominantly dense fibrous connective tissue and oral mucosa lining epithelium; B—100 μm, a tooth-like structure composed by enamel matrix (asterisk) and mature dentin (arrow); C—50 μm, mass of immature dentin in fibrous connective tissue; Tooth-like structure associated with, D—100 μm, dental papilla (asterisk), and, E—50 μm, surrounding pulp tissue (asterisk); F—50 μm, dense fibrous connective tissue containing nests of odontogenic epithelium (arrows).
fibroma was raised, and an excisional biopsy was performed. During the transoperative period, no cortical bone resorption was observed. The histopathological analysis revealed the presence of tooth-like structures, formed by enamel and dentin matrix, occasionally associated with the dental papilla and surrounding pulp tissue (Figure 2). These structures were adjacent to the oral mucosal lining epithelium. Thus, a histopathological diagnosis of peripheral compound odontoma was established. The patient has been undergoing follow-up for 6 months without any signs of lesion recurrence.

3 | DISCUSSION

The etiology of odontoma is unknown, however, some authors describe the possible association with local trauma in the primary dentition, inflammatory and/or infectious processes, odontoblastic hyperactivity, and alterations in the genetic component associated or not with hereditary conditions (Gardner syndrome and Hermanns syndrome).7,8

It is believed that PO development, as well as the peripheral odontogenic tumors, is because of the entrapment of epithelial dental lamina remains (rests of Serres) located supraperiosteally in the gingiva. These epithelial remnants appear to retain the ability to interact with the mesenchyme, leading to PO development.2,5,8,9 However, little is known about the stimuli that can activate this epithelium and trigger this odontoma.5

According to Koneru et al4 and Satish et al,7 one possible hypothesis is that the rests of Serres undergo genetic alteration that results in interference of the mechanism of control of the formation of tooth, as well as stimulating the mesenchymal differentiation that leads to the development of the odontoma. From this, nests of odontogenic epithelium may or may not proliferate and develop singly as multiple tooth-like structures or as random conglomerates of dental tissue.

Odontomas can be classified according to their clinical presentation into central (intraosseous), peripheral (extraosseous), and erupted odontoma. The central odontoma is the most frequent, followed by the erupted odontoma, while the PO is extremely rare.8 Only 18 PO cases have been reported in PubMed, MEDLINE, Scopus, Web of Science, and Scielo databases (January 14, 2020) (Table 1).

As shown in Table 1, most previously described PO were diagnosed in males (58.8%) during the first two decades of life, most commonly in the first decade (58.8%). The maxilla was the most frequent location (76.4%), with a higher prevalence in the palate region, followed by the gingiva. Clinically, in most of the reported cases, the lesions presented asymptomatic nodular swelling, showing firm consistency, normal color, and slow and limited growth (range: 0.25-1.5 cm). Data on age, location, clinical presentation, and size of the lesion corroborate the findings of the present case.

Because of its rarity, PO is seldom considered as a differential diagnosis of gingival lesions.5 The cases described in Table 1 support this view, since they investigated frequent gingival lesions, including fibrous hyperplasia, periodontal abscess, pyogenic granuloma.
Peripheral ossifying fibroma, and congenital epulis (in cases of natal lesions), as the diagnostic hypothesis of the PO. Besides that, radiographic images are useful for directing the diagnosis of oral lesions such as odontoma. In this way, periapical radiographs are often requested; however, they are not so appropriate or helpful for diagnosing PO.3,4 Santos et al20 reported that cone beam computed tomography plays an important role in the identification and diagnosis of odontomas, as it allows a better resolution analysis and an accurate assessment of the relationship between the lesion and adjacent tissues. In the present case, the radiograph showed no changes that indicated the presence of a lesion. Because of the clinical hypothesis of oral fibroma, complementary imaging tests were not requested.

Histopathologically, depending on the degree of differentiation, PO, as well as its intraosseous counterpart, can be categorized as either a compound or complex odontoma.3 As in most literature reports, the present case was classified as a compound PO. In addition to these morphological characteristics, it is important to note that cases reported in the literature displayed the involvement of fibrous connective tissue, usually presenting odontogenic epithelium nests, and superficial stratified squamous epithelium.

When the PO is classified as a compound type, it is important to distinguish it from supernumerary teeth. Some authors believe that these dental anomalies are because of the same pathological process that results in PO, and the similar histopathological appearance of both lesions supports this hypothesis.21 However, although this assumption is applicable in some cases, supernumerary teeth do not present tumor growth.5,20 Furthermore, mixed odontogenic tumors may also aid in the differential diagnosis of this pathology.1

Peripheral cases of mixed odontogenic tumors, such as ameloblastic fibroma, have been previously reported.12 However, this lesion differs from odontoma because it does not microscopically exhibit the formation of dental hard tissue.22 It is important to emphasize that epithelium-rich odontoma may be a diagnostic challenge because it exhibits histopathological similarities with the ameloblastic fibro-dentinoma and ameloblastic fibro-odontoma.5 However, the 2017 World Health Organization classification reports that the formation of dental hard tissue for these lesions is a stage of maturation. Thus, these lesions were grouped into the odontoma and represent a spectrum of its development.22

The treatment of choice for PO is local excision. To date, no recurrence cases have been reported after complete lesion removal. An accurate diagnosis is indispensable to avoid excessive and inadequate treatments, thus, recognize clinical and histopathological features is essential in this context.

4 | CONCLUSIONS

PO are uncommon lesions that usually affect young patients and display a preference for the maxilla, and limited growth potential. In addition, some odontogenic lesions present a peripheral variant, indicating a differential diagnosis from other common extranodal lesions in the oral cavity. Thus, this report is significant concerning the rarity of these lesions and the low number of cases published in the literature and provides information that aids in the recognition of clinical and histopathological PO characteristics in order to establish appropriate diagnoses.

CONFLICT OF INTEREST

None of the authors has any conflicts of interest, financial or otherwise, in connection with the preparation of this manuscript.

ORCID

Caio César da Silva Barros https://orcid.org/0000-0003-3103-3588
Erasmo Freitas de Souza Junior https://orcid.org/0000-0003-1714-9226

REFERENCES


