



Artigo original

PERIPHERAL ERUPTED COMPOUND ODONTOMA IN THE ANTERIOR MAXILLARY REGION

Odontoma composto periférico erupcionado em região anterior da maxila

Winícius Arildo Ferreira Araújo¹, Natália Galvão Garcia², Déborah Rocha Seixas¹, Letícia Cunha Borges³, Eduardo Sanches Gonçalves^{4*}

RESUMO

O odontoma é o mais prevalente entre os tumores odontogênicos, ocorrendo principalmente em homens na primeira década de vida. Entretanto, o odontoma extraósseo ou periférico é extremamente incomum e desconhecido pelos cirurgiões-dentistas. Nesse sentido, o objetivo do presente artigo é relatar um caso incomum de odontoma composto periférico que erupcionou espontaneamente. O caso envolveu um paciente do sexo masculino, de 17 anos, que se queixava de erupção dentária em local errado. A lesão era uma estrutura semelhante a dentículos e assintomática na região anterior da maxila. O exame clínico e de imagem contribuiu para estabelecer o diagnóstico de odontoma composto erupcionado. A cirurgia de remoção foi feita sob anestesia local sem intercorrências. No acompanhamento, foram avaliadas cicatrização satisfatória e ausência de recidiva. Diante disso, conclui-se que o odontoma composto erupcionado na região anterior da maxila, embora raro, possui relevância clínica para os cirurgiões-dentistas. Sendo assim, o diagnóstico precoce é essencial para um melhor prognóstico, principalmente na região estética.

Palavras-Chave: Odontoma Composto; Tumor Odontogênico; Cirurgia; Diagnóstico Oral.

ABSTRACT

Odontoma is the most prevalent among odontogenic tumors, it mainly occurs in men in the first decade of life. However, extraosseous or peripheral odontoma is extremely uncommon, being unaware by surgeons. In this sense, the purpose of the present paper is to report an unusual case of peripheral compound odontoma erupted spontaneously. The case involved a 17-year-old male patient, who complained of a tooth eruption in the wrong location. The lesion was a tooth-like structure and asymptomatic in the anterior maxillary region. The clinical and imaging examination contributes to establishing the diagnosis of erupted compound odontoma. The removal surgery was made under local anesthesia without intercurrent. In follow-up, satisfactory healing and no recurrence were evaluated. Given this, it is concluded that the compound odontoma erupted in the anterior region of the maxilla, although rare, has clinical relevance for dental surgeons. Since that, early diagnosis is essential for better prognosis, especially in the aesthetic region.

Key words: Compound Odontoma; Odontogenic Tumor; Surgery; Diagnosis, Oral.

1. Postgraduate Program in Oral and Maxillofacial Surgery, Bauru School of Dentistry, University of Sao Paulo, Bauru, Sao Paulo, Brazil.

2. Professor, University Center of Lavras (UNILAVRAS), Lavras, Minas Gerais, Brazil.

3. Dentist, Morgana Potrich Faculty (FAMP), Mineiros, Goiás, Brazil.

4. Associate Professor, Department of Surgery, Stomatology, Pathology and Radiology, Bauru School of Dentistry, University of São Paulo, Bauru, Sao Paulo, Brazil.

*Correspondence:

Eduardo Sanches Gonçalves, DDS, MS, PhD.

University of São Paulo

Alameda Dr. Octávio Pinheiro Brisolla, 9-75 - Vila Regina, Bauru - SP, 17012-230

Tel. + 55 14 3235-8000.

E-mail: eduardogoncales@usp.br



INTRODUÇÃO

Odontoma is the most common odontogenic tumor with a nonaggressive behavior, usually intraosseous, it is considered a lesion that rises from malformation of dental tissue development (rest of Serres) ^{1,2}. According to the World health organization (WHO), odontomas are classified related to the cell proliferation, can be classified as complex or compound, intraosseous or extraosseous ^{1,3}.

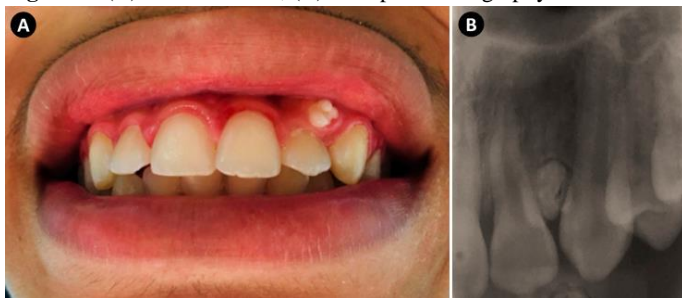
The extraosseous or peripheral odontomas present as asymptomatic nodular lesions, with slow growth and no bone involvement, tending to exfoliate, and rarely erupt. They usually are associated with permanent or temporary tooth eruption disturbances, identified in routine radiographic examination, having a higher prevalence in children^{1,2}.

While there is information available about odontomas in literature, there are only a limited number of case reports regarding specific peripheral compound odontomas in the anterior region of the maxilla. Therefore, the aim of the present paper is to report an unusual case of peripheral compound odontoma (PCO) erupted spontaneously in the anterior maxillary region.

CASE REPORT

The patient, a 17-year-old male, presented complaining about a “tooth eruption in the wrong location”. The lesion appeared one year before the consult and remained asymptomatic. The patient stated that there was no history of harmful habits, previous trauma, or infection in the area surrounding the lesion. His medical history was non-contributory.

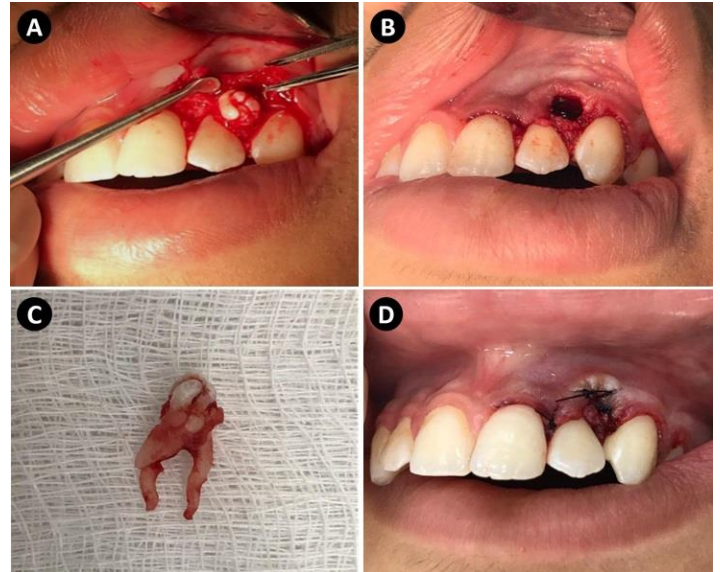
Figure 1: (A) Intraoral view; (B) Periapical radiography.



Fonte: os autores, 2024.

During the intraoral examination, denticles were found exposed in the gingiva located between the permanent maxillary lateral incisor and canine teeth on the anterior left side. (Figure 1). The radiographic analysis showed that the teeth had an irregular shape, comprising of a crown, root and pulp, but without any signs of bone involvement. (Figure 2).

Figure 2: (A) Surgical site showing the denticule to be removed; (B) Excision of the denticule; (C) The excised denticule; (D) The surgical site after removal of odontoma.



Fonte: os autores, 2024.

A compound odontoma was clinically diagnosed in the gingiva, and after being surgically removed under local anesthesia, there was not any evidence of bone erosion beneath the tumor (Figure 3). After the surgery, there were no complications and the suture was removed seven days later. At the time, the surgery site has healed satisfactorily and, since then, had no recurrence.

Figure 3: One-month follow-up after the surgery.



Fonte: os autores, 2024.

It's presented in Table 1 all the cases published and diagnosed as peripheral odontoma. The research included the terms “peripheral odontoma”, “gingival odontoma”, “erupted odontoma” and “soft tissue odontoma” as keywords in Pubmed, Medline, and Google School databases, and resulted only in 20 papers dating from 1989 to 2023. Were excluded articles with diagnosis of complex odontoma and one article from the list due to absence of enough information to join the table.

Table 1: Clinical cases of peripheral odontoma related in the literature in the period of 1989 to 2023.

Author	Gender/age	Localization	Diagnostic	Erupted in oral cavity
Present case, 2023	M/17 yo	Maxilla, anterior gingiva	Compound odontoma	Yes
Soluk-Tekkeşin et al. ¹⁰ 2022	F/12 yo	Maxilla, anterior gingiva	Compound odontoma	No
Shi et al. ¹¹	F/11 yo	Maxilla, anterior gingiva	Compound odontoma	No
Da Silva Rocha et al. ¹ 2020	F/11 yo	Maxilla, palatal region	Compound odontoma	No
De Oliveira et al. ¹²	M/30 yo	Maxilla, anterior region	Compound odontoma	Yes
Bernardes et al. ¹³ 2016	M/12 yo	Maxilla, anterior gingiva	Compound odontoma	No
Michaliszyn, ⁹ 2016	F/10 yo	Maxilla, palatal region	Compound odontoma	Yes
Mehta et al., ⁸ 2014	M/22 yo	Maxilla, anterior region	Compound odontoma	Yes
Hanemann et al. ⁷ 2013	F/15 yo	Maxilla, anterior gingiva	Compound odontoma	Yes
Nadendla, ¹⁴ 2013	F/18 yo	Mandible, lingual anterior region	Compound odontoma	Yes
Mikami et al. ¹⁵ 2013	M/9 months	Maxilla, palatal region	Compound odontoma	No
Friedrich et al. ¹⁶ 2010	M/3 yo	Maxilla, palatal region	Compound odontoma	No
Silva et al. ¹⁷ 2009	M/8 months	Maxilla, palatal region	Compound odontoma	No
	M 5 months	Maxilla, palatal region	Compound odontoma	No
Ide et al. ¹⁸ 2008	F/7 yo	Mandible, lingual posterior region	Compound odontoma	No
Kintarak et al. ¹⁹ 2006	F/13 yo	Maxilla, palatal region	Compound odontoma	No
Ide et al. ²⁰ 2000	M/39 yo	Maxilla, anterior gingiva	Compound odontoma	No
Ledesma-Montes et al. ²¹ 1996	F/3 yo	Mandible, lingual posterior region	Compound odontoma	No
Castro et al. ²² 1994	M/6 yo	Mandible, lingual posterior region	Compound odontoma	No
Giunta and Kaplan. ²³ 1990	M/5 yo	Maxilla, palatal region	Compound odontoma	No
	M/21yo	Mandible, lingual posterior region	Compound odontoma	No

Abbreviations: M, male; F, female; yo, years-old. **Fonte:** os autores, 2024.

DISCUSSION

The etiology of odontoma is unknown. Some authors have suggested that odontoma can be associated with local trauma, inflammatory or infectious process, odontoblastic hyperactivity, or hereditary conditions such as Gardner syndrome and Hermanns Syndrome^{1,2,4}. In case of peripheral odontomas, it is believed that the entrapment of epithelial dental lamina rests (rests of Serres) located supraperiosteally in the gingiva, by an uncertain stimulus, may develop a peripheral tumor^{1,5,6}.

The present case, along with approximately twenty other compound odontomas reported in the literature, is an example of how peripheral odontomas are extremely uncommon.

This information is summarized in Table 1. Clinically, peripheral odontoma presents as an asymptomatic nodular lesion, showing firm consistency, normal color when submucous, with slow growth. When erupted, they present as tooth-like structures^{1,4}, as seen in the present case.

The mechanism of erupting odontoma remains uncertain⁷⁻⁹. Being extremely rare, little is known about erupted odontomas. Some authors reported that odontoma eruption could occur when there is an absence of erupted

teeth, or severe bone resorption. The most acceptable eruption hypothesis of the case presented is due to the mechanism behind the bone remodeling of the jaw and the increase in tumor size⁷⁻⁹. Hanemann et al. (2013)⁷, proposed that denticles from a peripheral compound odontoma might be present in the gingiva, as shown in this case, may have a propensity towards exfoliation due to its growth over time, rather than a real tooth eruption process, in contrast to what may be expected. In the current research, this is the sixth documented case of a peripheral odontoma being erupted.

Also, Table 1 shows that most cases of peripheral odontoma previously described were seen in men, most commonly in the first decade of life. On the other hand, women were mostly committed in second decade of life. The maxilla was the most frequent location in both sexes, with a higher prevalence in the palate region, followed by the gingiva^{1-3,6-8}. All information described, as gender, age, location, clinical presentation, and size of the lesion corroborate to the present case.

Due to its rare occurrence, lack of pathognomonic clinical features of peripheral odontoma, and resemblance to any localized gingival overgrowth, peripheral odontoma,

when unerupted, is often difficult to diagnose clinically. Both complex odontoma and compound odontoma can develop peripherally, however when the compound erupted odontoma occurs, supernumerary tooth can be included in the differential diagnosis. Thus, the complementary imaging exam becomes essential in the diagnosis of these lesions. The treatment of choice in odontoma cases is surgical excision, thus the early treatment is necessary for minimizing the invasive interventions ^{1,7}.

Considering the rare occurrence of peripheral odontomas and the difficult to diagnose clinically, the present case is the sixth report of a peripheral odontoma erupted in the oral cavity. Therefore, dentists must be aware that peripheral odontomas can eventually emerge in the mouth, obstructing esthetics.

CONCLUSION

It is concluded that the compound odontoma erupted in the anterior region of the maxilla, although rare, has clinical relevance for dental surgeons. Since that, early diagnosis is essential for better prognosis, especially in the aesthetic region.

REFERENCES

1. da Silva Rocha OKM, da Silva Barros CC, da Silva LAB, Souza Júnior EF, Morais HHA, da Costa Miguel MC. Peripheral compound odontoma: A rare case report and literature review. *J Cutan Pathol*. 2020;47(8):720-724. doi:10.1111/cup.13676
2. Custódio M, Araujo JP, Gallo C de B, Trierveiler M. Gingival complex odontoma: A rare case report with a review of the literature. *Autops Case Reports*. 2018;8(1). doi:10.4322/acr.2018.009
3. Wright JM, Vered M. Update from the 4th Edition of the World Health Organization Classification of Head and Neck Tumours: Odontogenic and Maxillofacial Bone Tumors. *Head Neck Pathol*. 2017;11(1):68-77. doi:10.1007/s12105-017-0794-1
4. Koneru A, Surekha R, Vanishree M, Hamsini A, Hunasgi S. Rare gingival odontoma: Report of a case and review of literature. *J Dr NTR Univ Heal Sci*. 2014;3(2):133. doi:10.4103/2277-8632.134889
5. Barba LT, Campos DM, Rascón MMN, Barrera VAR, Rascón AN. Aspectos descriptivos del odontoma: revisión de la literatura. *Rev Odontológica Mex*. 2016;20(4):272-276. doi:10.1016/j.rodex.2016.11.009
6. Manor Y, Mardinger O, Katz J, Taicher S, Hirshberg A. Peripheral odontogenic tumours—differential diagnosis in gingival lesions. *Int J Oral Maxillofac Surg*. 2004;33(3):268-273. doi:10.1006/ijom.2003.0508
7. Hanemann JA, Oliveira DT, Garcia NG, Santos MR, Pereira AA. Peripheral compound odontoma erupting in the gingiva. *Head Face Med*. 2013;9(1):15. doi:10.1186/1746-160X-9-15
8. Mehta D, Raval N, Udhani S, Parekh V, Modi C. An Unusual Case Report of Erupted Odontoma. *Case Rep Dent*. 2013;2013:1-3. doi:10.1155/2013/570954
9. Michaliszyn GF. Odontoma composto : relato de caso. *Repositório Digit UFRGS*. Published online 2016. <http://hdl.handle.net/10183/150298>