ISSN 2674-8169

Odontogenic Choristoma in the Genian Subcutaneous Region of a Pediatric Patient: A Case Report

Vitória Somma Tessari[1], Davi Barbosa Portela[2], Hallana de Lima Siqueira[3], Heloísa de Colo[4], Alana Gabrieli Schvendtner[5], Christiane Elisa Ribas Batista[6], Eugênio Esteves Costa[7]

https://doi.org/10.36557/2674-8169.2026v8n7p5-14
Artigo recebido em 1 de Junho e publicado em 1 de Julho de 2026

ESTUDO DE CASO

ABSTRACT

Introduction: Choristomas are rare, benign tumor-like proliferations characterized by the development of histologically normal tissues in ectopic anatomical locations. Odontogenic choristomas, in particular, are uncommon findings in the maxillofacial region, often misdiagnosed as other neoplasms or reactive lesions. **Objectives:** This paper aims to report and clinically analyze a rare case of odontogenic choristoma located in the subcutaneous region of the cheek in a pediatric patient, deepening the discussion on its etiopathogenesis. **Methodology (Case Report):** A 12-year-old male patient presented with a palpable, painless, hard nodule in the left cheek, associated with chronic biting habits. Imaging (ultrasound) revealed a subcutaneous calcified lesion. An excisional biopsy was performed via an extraoral approach. **Results and Discussion:** Histopathological analysis confirmed the presence of immature bone trabeculae and acellular eosinophilic material compatible with dentinoid, immersed in dense connective tissue, establishing the diagnosis of odontogenic choristoma. The hypothesis of tissue metaplasia induced by chronic microtrauma versus embryological failure is discussed, as well as the absence of Klippel-Feil Syndrome signs in this specific case. **Conclusion:** Odontogenic choristoma must be included in the differential diagnosis of calcified nodular lesions of the maxillofacial region, especially in areas prone to local trauma. Complete surgical excision proved to be both therapeutic and diagnostic, with an excellent prognosis and no signs of recurrence.

Palavras-chave: Choristoma, Oral Surgery, Oral Diagnosis, Oral Pathology, Pediatric Dentistry

Instituição afiliada – [1] Doutoranda em Odontopediatria pela Universidade Federal do Paraná, [2] Cirurgiã – Dentista – Universidade Positivo – Curitiba Paraná, [3] Mestrando Faculdade Paulo Picanço, [4] Acadêmica Odontologia PUC Curitiba, [5] Mestre em PNE – UFP, [6] Mestre em Psicologia UFSC, [7] Doutor Odontologia, Coordenador Especialização Cirurgia e Traumatologia Bucomaxilo Facial Faculdade IPPEO

INTRODUCTION

From a stringent histopathological perspective, choristomas are defined as benign, tumor-like masses consisting of morphologically normal cells or tissues that develop in an ectopic, or anatomically incongruous, location (WEST; ATKINS, 1988). It is an academic imperative to distinguish them from hamartomas, which represent a disorganized developmental overgrowth of tissues that are natively expected to be present at the site of the lesion. In the maxillofacial territory, choristomas of any lineage are exceptionally rare entities (WEST; ATKINS, 1988).

Odontogenic choristomas (OC), in particular, denote the ectopic genesis of tooth-forming tissues (NOROOZI; ARORA, 2011; GUIDO et al., 2021). Clinically, an OC typically presents as a firm, circumscribed, and asymptomatic nodule. Because of its innocuous clinical presentation, clinicians frequently mistake it for other soft tissue neoplasms, calcified lymph nodes, or reactive foreign body granulomas (WEST; ATKINS, 1988). Therefore, rigorous histopathological examination remains the absolute gold standard for a definitive diagnosis.

The primary objective of this manuscript is to report a highly unusual clinical presentation of an odontogenic choristoma located in the subcutaneous region of the cheek in a 12-year-old pediatric patient. Furthermore, it aims to construct a robust theoretical framework through a literature review, elucidating the complex etiopathogenesis, differential diagnostic parameters, and optimal surgical management of this rare pathology.

METHODOLOGY

This manuscript delineates a descriptive observational study structured as a clinical case report, conducted in strict adherence to the ethical principles governing medical research. The clinical, imaging, and histopathological data were meticulously acquired from the medical records of a patient treated at the Pediatric Dentistry Clinic of Universidade Positivo. The surgical intervention comprised an extraoral excisional biopsy, followed by standard histopathological processing and microscopic evaluation

to establish the definitive diagnosis.

Concomitantly, to substantiate the academic discussion, a narrative literature review was executed. The theoretical framework was built upon selected peer-reviewed articles focusing on the pathophysiological mechanisms, epidemiological prevalence, and syndromic associations of osseous and odontogenic choristomas within the head and neck region.

LITERATURE REVIEW

To comprehend the nature of Odontogenic Choristomas, one must delve into their epidemiological distribution and morphological variations. A review of the current literature elucidates that the posterior third of the tongue—specifically near the foramen cecum—is the most frequently documented intraoral site for osseous or odontogenic ectopic manifestations (WEITZNER, 1986; BENAMER; ELMANGOUSH, 2007). The manifestation of these tissues in the buccal mucosa or genian (cheek) spaces is exceedingly scarce, representing a significant diagnostic challenge (NOROOZI; ARORA, 2011; GUIDO et al., 2021). The ectopic presence of these tissues, remarkably, is not strictly confined to the oral cavity; documented cases have demonstrated involvement in extraoral structures, such as the periocular and palpebral regions (JAKOBIEC et al., 2009; MEN et al., 2019; ARENS et al., 2019).

From a histological standpoint, the spectrum of OC is broad. As observed by Noroozi e Arora (2011) and Liu et al. (2013), the microscopic architecture can range from primitive, unorganized dentinoid and osteoid matrices to the presence of fully formed ectopic dental organs featuring mature dentin, enamel matrices, and functional pulp tissue (GUIDO et al., 2021).

The etiopathogenesis of choristomas represents a focal point of intense academic debate, pivoting primarily on two distinct pathophysiological hypotheses (ARENS et al., 2019):

1. **The Embryological Hypothesis:** This theory postulates a developmental anomaly. During the complex invaginations, fusions, and cellular migrations characteristic of embryonic facial development, pluripotential cells from the odontogenic apparatus may become displaced into heterotopic spaces, such as the cheek or

ocular regions (JAKOBIEC et al., 2009; MEN et al., 2019). Once displaced, these epithelial rests lie dormant until an unidentified biological trigger initiates their differentiation into calcified odontogenic tissues (NOROOZI; ARORA, 2011).

2. **The Metaplastic / Traumatic Hypothesis:** Conversely, Vered, Lustig e Buchner (1998) proposed a reactive mechanism. They suggest that chronic mechanical irritation—such as continuous friction during deglutition for lingual lesions, or chronic cheek biting habits—acts as a catalyst. This persistent inflammatory stimulus can induce a metaplastic cascade within the pluripotent mesenchymal cells of the local connective tissue, ultimately promoting endochondral ossification and the formation of dentinoid structures (VERED; LUSTIG; BUCHNER, 1998).

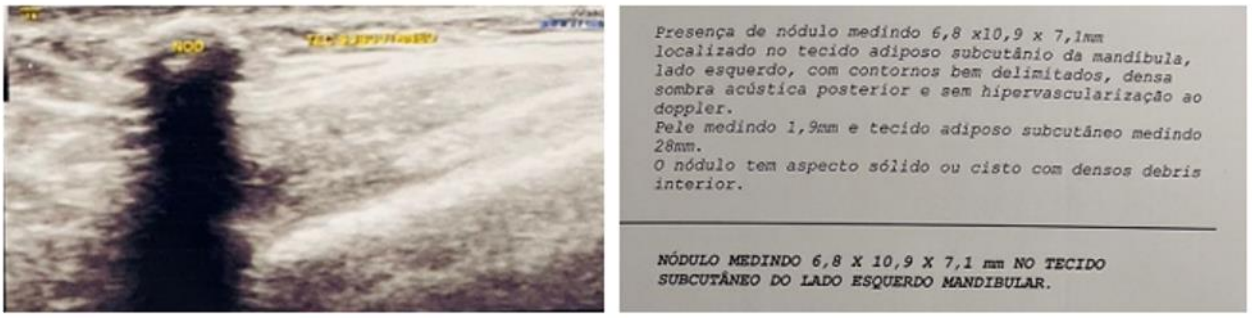
Furthermore, it is a clinical imperative to recognize syndromic associations when evaluating ectopic calcifications. Ectopic bone masses and odontogenic choristomas have a well-documented correlation with Klippel-Feil Syndrome (DOUGLAS; MOOS; HISLOP, 1992). This complex congenital condition is primarily characterized by the abnormal fusion of two or more cervical vertebrae, resulting in restricted neck mobility and a low posterior hairline (TRACY; DORMANS; KUSUMI, 2004). Gaitán-Cepeda, Quezada-Rivera e Ruíz-Rodríguez (2003) explicitly detailed a case where an osseous choristoma in the cheek served as a soft-tissue manifestation in a patient diagnosed with Klippel-Feil Syndrome, emphasizing the need for systemic evaluation in such presentations.

RESULTS AND DISCUSSION

A 12-year-old male patient presented for a routine dental evaluation at the Pediatric Dentistry Clinic (Universidade Positivo). While the primary complaint was unrelated to any intraoral lesion, systematic physical examination revealed a firm, nodular mass palpable in the subcutaneous tissue of the left cheek. Anamnesis confirmed a full-term birth without complications and no history of facial trauma. However, a critical piece of clinical history was obtained: the patient's guardians reported the long-standing presence of the mass and noted the child's chronic, habitual behavior of biting the affected cheek area.

An ultrasound imaging requested preoperatively revealed a well-defined, calcified lesion in the subcutaneous adipose tissue of the left cheek, presenting dense posterior acoustic shadowing. (Figure 1).

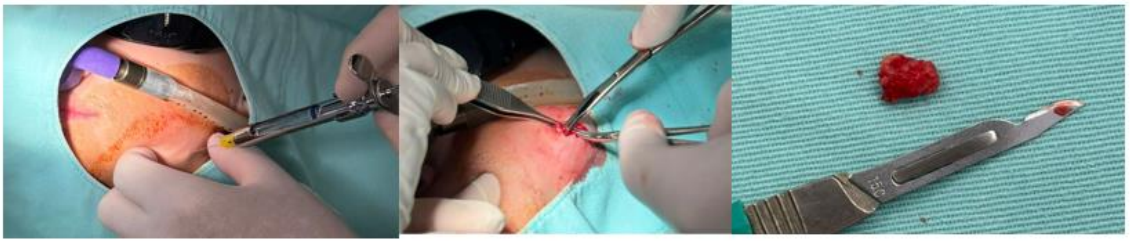
Figure 1 – Imaging Examination: Preoperative Ultrasound and Report.



Fonte: Autores.

Under extraoral local anesthesia and an intraoral buccal nerve block, an excisional biopsy was performed via an extraoral approach using a cold scalpel, allowing for the complete removal of the mass without capsular rupture (Figure 2).

Figura 2 – Intraoperative.

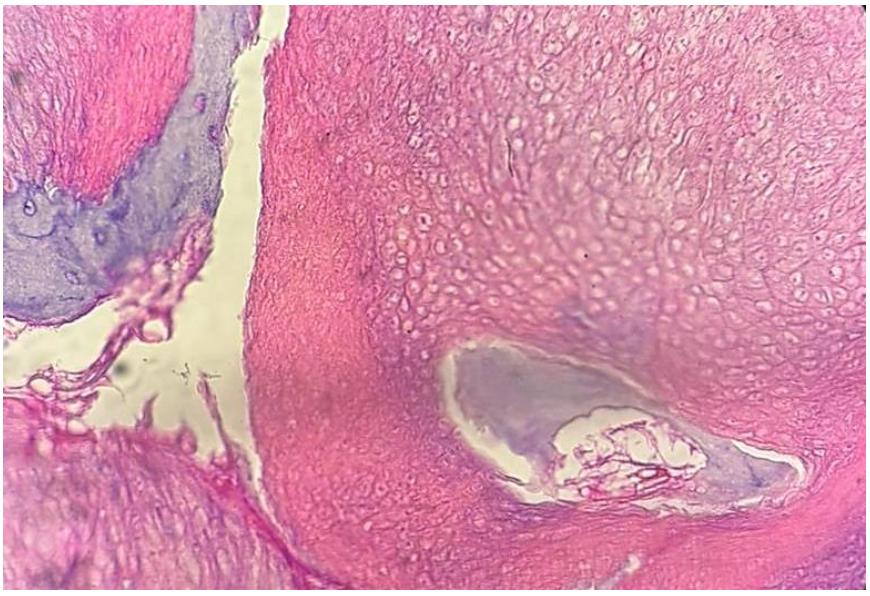


Fonte: Autores.

Macroscopically, the specimen was a stony, yellowish mass measuring 0.9 × 0.8 × 0.6 cm. Histopathological analysis (Figure 3) revealed a dense, sparsely cellular connective tissue matrix containing scattered mononuclear inflammatory cells. The defining

characteristic was the presence of irregularly distributed trabeculae of immature woven bone and distinct areas of an acellular, eosinophilic matrix morphologically compatible with dentinoid material. These findings established the definitive diagnosis of Odontogenic Choristoma.

Figure 3 – Acellular eosinophilic material compatible with dentinoid, trabeculae of immature bone tissue within sparsely cellular dense connective tissue.



Fonte: Autores.

When evaluating the demographic profile, OC is predominantly reported in female patients and is classically diagnosed during early adulthood (GAITÁN-CEPEDA; QUEZADA-RIVERA; RUÍZ-RODRÍGUEZ, 2003; GUIDO et al., 2021). Consequently, the manifestation of this lesion in a 12-year-old male patient renders the present case epidemiologically singular. Furthermore, the anatomical location in the genian subcutaneous space is highly atypical, deviating from the classic lingual dorsum presentation (WEITZNER, 1986; BENAMER; ELMANGOUSH, 2007).

The primitive tissue architecture observed histologically in our patient suggests an early or arrested stage of ectopic odontogenesis, contrasting with reports of fully formed ectopic teeth (LIU et al., 2013; GUIDO et al., 2021). The etiology of this specific lesion strongly aligns with the *Metaplastic/Traumatic Hypothesis* (VERED; LUSTIG; BUCHNER, 1998). The active clinical history of chronic cheek biting reported by the patient's

guardians provides a tangible mechanical stimulus that likely triggered the metaplastic ossification cascade within the buccal connective tissue.

In terms of systemic evaluation, a rigorous physical assessment of the pediatric patient effectively ruled out any morphological features or systemic signs indicative of Klippel-Feil Syndrome, isolating the choristoma as a localized, non-syndromic event (DOUGLAS; MOOS; HISLOP, 1992; TRACY; DORMANS; KUSUMI, 2004).

Therapeutically, the consensus in surgical pathology dictates that conservative yet complete surgical excision is the treatment of choice, serving as both a diagnostic necessity and a definitive cure (WEST; ATKINS, 1988; ARENS et al., 2019). Meticulous soft-tissue dissection is required to prevent iatrogenic damage to adjacent neural structures, a precaution also highlighted by Liu et al. (2013). Odontogenic choristomas possess an overwhelmingly favorable prognosis. They exhibit no known tendency for malignant transformation, and recurrence is practically nonexistent following adequate enucleation (NOROOZI; ARORA, 2011). In our clinical follow-up at 4 months post-operation, the patient exhibited perfect tissue healing with no clinical or ultrasonographic signs of recurrence.

FINAL CONSIDERATIONS

Odontogenic choristoma is an exceedingly rare, benign entity that challenges standard diagnostic paradigms in oral and maxillofacial pathology. Despite its rarity, it must be systematically included in the differential diagnosis of calcified, asymptomatic nodular lesions in the head and neck region, particularly when located in areas susceptible to chronic microtrauma. Complete surgical excision remains the definitive standard of care, providing the necessary histopathological confirmation and ensuring an excellent prognosis without recurrence.

ACKNOWLEDGEMENTS

Above all, the authors render their deepest gratitude to God for the guidance, strength, and wisdom bestowed throughout this academic and professional journey. We also extend our most sincere thanks to our families for their unconditional love, boundless patience, and unwavering support, which constituted the fundamental pillars

for the completion of this work.

Furthermore, we wish to express our profound gratitude to the patient and his legal guardians for their full cooperation and for granting informed consent for the publication of this clinical case, thereby significantly contributing to the scientific literature in oral pathology. We also extend our appreciation to the clinical staff at the Pediatric Dentistry Clinic of Universidade Positivo for their initial assistance.

Special recognition is owed to the Instituto Paranaense de Pesquisa e Ensino Odontológico (IPPEO), particularly highlighting the clinical and surgical practice environment in Mafra, Santa Catarina, for fostering rigorous academic development. Finally, we are deeply indebted to Dr. Eugenio Esteves Costa for his continuous scientific coordination, exceptional guidance, and invaluable support throughout the research and academic structuring of this manuscript.

REFERENCES

1. ARENS, P.; ULLRICH, A.; OLZE, H.; UECKER, F. C. Extraoral Osseous Choristoma in the Head and Neck Region: Case Report and Literature Review. **Case Reports in Otolaryngology**, [S. l.], v. 2019, p. 8532356, 28 maio 2019.
2. BENAMER, M. H.; ELMANGOUSH, A. M. Lingual Osseous Choristoma: Case report and review of literature. **Libyan Journal of Medicine**, [S. l.], v. 2, n. 1, p. 46-48, 1 mar. 2007.
3. DOUGLAS, P. S.; MOOS, K. F.; HISLOP, W. S. Abnormal bone masses in Klippel-Feil syndrome. **British Journal of Oral and Maxillofacial Surgery**, [S. l.], v. 30, n. 6, p. 382-386, dez. 1992.
4. GAITÁN-CEPEDA, L. A.; QUEZADA-RIVERA, D.; RUÍZ-RODRÍGUEZ, R. Osseous choristoma of the oral soft tissue: Case report. **Medicina Oral**, [S. l.], v. 8, n. 3, p. 220-223, maio/jul. 2003.
5. GUIDO, G.; BENEDETTI, S.; ROSSI, F.; GENNARO, P. Odontogenic choristoma embedded in the cheek of an old patient. **BMJ Case Reports**, [S. l.], v. 14, n. 10, p. e245733, 13 out. 2021.
6. JAKOBIEC, F. A.; NGUYEN, J.; MANDELL, K.; FAY, A. Complex palpebral odontogenic choristoma: A reappraisal of the origin of teeth-bearing periocular lesions. **American**

Journal of Ophthalmology, [S. l.], v. 147, n. 3, p. 531-543.e1, mar. 2009.

7. LIU, Y.; HUANG, Y.; YU, T.; LI, L. Teeth erupted from the buccal mucosa: simple odontogenic choristoma or accessory teeth? **Journal of Oral and Maxillofacial Surgery**, [S. l.], v. 71, n. 11, p. 1834.e1-1834.e4, nov. 2013.
8. MEN, C. J.; WU, F.; LEE, B. W.; LIN, J. H.; KORN, B. S.; KIKKAWA, D. O. Odontogenic choristoma presenting as dermolipoma. **Orbit**, [S. l.], v. 38, n. 6, p. 492-494, dez. 2019.
9. NOROOZI, A. R.; ARORA, E. Odontogenic choristoma: report of a case. **Journal of Oral and Maxillofacial Surgery**, [S. l.], v. 69, n. 12, p. 3006-3009, dez. 2011.
10. TRACY, M. R.; DORMANS, J. P.; KUSUMI, K. Klippel-Feil syndrome: clinical features and current understanding of etiology. **Clinical Orthopaedics and Related Research**, [S. l.], n. 424, p. 183-190, jul. 2004.
11. VERED, M.; LUSTIG, J. P.; BUCHNER, A. Lingual osteoma: a debatable entity. **Journal of Oral and Maxillofacial Surgery**, [S. l.], v. 56, n. 1, p. 9-13, jan. 1998.
12. WEITZNER, S. Osseous choristoma of the tongue. **Southern Medical Journal**, [S. l.], v. 79, n. 1, p. 69-70, jan. 1986.
13. WEST, C. B. Jr.; ATKINS, J. S. Jr. Choristomas of the intraoral soft tissues. **Otolaryngology-Head and Neck Surgery**, [S. l.], v. 99, n. 5, p. 528-530, nov. 1988.