Dentigerous cyst in the maxilla: case report in a pediatric patient

Cisto dentígero em maxila: relato de caso em paciente pediátrico

Quiste dentígero en el maxilar: informe de un caso en un paciente pediátrico

Abstract
Dentigerous cysts are the second most common odontogenic cysts of the jaws, and sometimes inhibit the eruption of teeth. Almost all of the dentigerous cyst encloses the crown of an unerupted tooth and the radiolucent area is attached to the tooth at the cementoenamel junction. Dentigerous cyst is more common in male patients and most commonly develop in the second and third decades of life. Clinically, a localized swelling of the alveolar bone can be detected, however, dentigerous cysts can be asymptomatic during a long period of time, leading to a significant destruction of bone. The conventional treatment plan is cyst removal and marsupialization. Marsupialization therapy can be useful to promote the spontaneous eruption of the involved tooth within the cyst. However, tooth eruption does not always occur spontaneously after marsupialization. This article aims to report a clinical case of the diagnosis and treatment of a dentigerous cyst in the maxilla associated with an unerupted canine in a pediatric patient and discussion of the treatment performed.

Keywords: Dentigerous cyst; Tooth unerupted; Multidisciplinary approach.
extirpación del quiste y la marsupialización. La terapia de marsupialización puede ser útil para promover la erupción espontánea del diente afectado dentro del quiste. Sin embargo, la erupción del diente no siempre ocurre espontáneamente después de la marsupialización. Este artículo tiene como objetivo reportar un caso clínico del diagnóstico y tratamiento de un quiste dentígero en el maxilar asociado a un canino no erupcionado en un paciente pediátrico y la discusión del tratamiento realizado.

**Palabras clave:** Quiste dentígero; Diente no erupcionado; Planteamiento multidisciplinar.

1. **Introducción**

La erupción dental resulta del intercambio de varios pasos entre el epitelio oral y el tejido mesenquímico. La interacción anormal del tejido durante el desarrollo puede eventualmente resultar en la erupción ectópica del diente. (Buyukkurt et al., 2010). La erupción ectópica puede ser el resultado de uno de estos 3 procesos: enfermedad del desarrollo, actividad iatrogénica, o proceso patológico como un tumor o quiste. (Patil et al., 2019)

Los quistes dentigeros son considerados el segundo grupo más común de quistes odontogénicos de los arcos y pueden inhibir la erupción de los dientes. La mayoría de los quistes dentigeros están localizados en contacto íntimo con la corona de un diente no erupcionado y la zona radiolúcida está en contacto con el diente en la unión cemento-esmalte. Aparece como una lesión odontogénica radiolúcida bien definida que involucra la corona de un diente no erupcionado, causando dificultad en su erupción. Este quiste está asociado con dientes que desarrollan impacción, como terceros molares y cánones, y también pueden afectar dientes supernumerarios y hasta odontomas. (Ustuner et al., 2003)

Los quistes dentigeros involucrando dientes impacientes usualmente desplazan estos dientes hacia posiciones ectópicas. En la maxilar, estos dientes pueden ser desplazados hacia el seno maxilar. Los quistes dentigeros progresan lentamente y se desarrollan durante varios años sin mostrar síntomas. Cuando el seno maxilar es invadido, los síntomas suelen ocurrir después de varios años. Las secuelas de estos quistes pueden llevar a obstrucción del seno. (Lacerda et al., 2018)

La mejor hipótesis para la formación de un quiste dentiger es la acumulación de fluido, originándose de la presión aumentada ejercida por un diente que está erupciando en la foliculo, ya sea entre la capa reducida o entre las capas del órgano esmalte. (Contar et al., 2015)

Los quistes dentigeros son más comunes en pacientes masculinos y pueden desarrollarse en la segunda y tercera década de la vida. Clínicamente, un bulbo localizado en la mandíbula puede ser detectado, sin embargo, los quistes dentigeros pueden ser asintomáticos durante un largo período de tiempo, llevando a una destrucción significativa de la mandíbula. (Ustuner et al., 2003)

Los quistes dentigeros se caracterizan por áreas radiolúcidas uniloculares asociadas con las coronas de dientes no erupcionados al nivel de la unión cemento-esmalte. La lesión radiolúcida es bien definida y circunscrita con un borde sclerótico (radiopaco). (Meningaud et al., 2006)

Los quistes dentigeros pueden tener infección secundaria, bordes irregulares, o mal definidos. Las raíces de los dientes adyacentes pueden mostrar resorción o desplazamiento causado por la presión del quiste dentigeroso. Los quistes dentigeros más grandes pueden tener una apariencia multilocular y deben considerarse en el diagnóstico diferencial de ameloblastoma. (Meningaud et al., 2006)

Microscópicamente, un epitelio cístico no queratinizado de hasta 2 a 3 capas es observado. La cápsula fibrosa está dispuesta de manera suelta y puede tener islas de células epiteliales odontogénicas inactivas. Los quistes dentigeros inflamados pueden presentar epitelio cístico multilaminar. (Patil et al., 2019)

El tratamiento convencional es la extirpación del quiste a través de la marsupialización. Este método de tratamiento puede ser útil para promover la erupción espontánea del diente involucrado. No obstante, la erupción del diente no es necesariamente espontánea después de la marsupialización. (Aoki et al., 2018)

Este artículo se propone reportar un caso clínico de la evaluación y tratamiento de un quiste dentiger en la maxilar asociado a un cánion no erupcionado en un paciente pediátrico y discusión del tratamiento realizado.
2. Methodology
This clinical case report is part of a descriptive observational study, which in addition to discussing the proposed treatment, also aimed to discuss the topic through the bibliographic survey carried out (Pereira, A. S., 2018) and thus contribute to the scarce scientific literature on the subject.

Thus, representing a descriptive and qualitative content. The case was conducted according to clinical and professional ethics. The patient reported in the study has a personal data sheet and anamnesis, where he authorized the use of images, clinical, radiographic, and socioeconomic data for educational and research purposes, and with this signed a Free and Informed Consent Form and the Informed Consent Form. Consent of the Person's Participation as a Subject, which were duly read and explained.

3. Case Report
Patient E.F.O, 10 years old, male, was accompanied by his parents to the Oral and Maxillofacial Surgery Outpatient Clinic of the Leforte Group, referring volume increase in the right maxilla region for 4 months and with progressive worsening. Clinical examination revealed an extra-oral swelling in the middle third of the right face, with erasure of the nasolabial groove. Intraoral examination revealed bulging of the vestibule in the region of tooth 13, which had not erupted and the patient reported pain on palpation.

A computed tomography scan of the face was requested for further evaluation. Upon the patient's return, a hypodense, circumscribed lesion with a well-defined halo of approximately 28.15 x 18.50 mm was observed in the right pre-maxilla region, with complete involvement of tooth 13 within the lesion. According to the clinical and imaging features, the diagnostic hypothesis was a dentigerous cyst. (Figure 1 and 2)

Figure 1 - A computed tomography scan axial of the face showing a hypodense, circumscribed lesion with a well-defined halo of approximately 28.15 x 18.50 mm was observed in the right pre-maxilla region.

Source: Authors.
**Figure 2** - A computed tomography scan sagital of the face showing a hypodense, circumscribed lesion with a well-defined halo was observed in the right pre-maxilla region.

![CT Scan](image1)

Source: Authors.

It was decided to perform the surgical procedure under general anesthesia for enucleation of the cystic capsule and exodontia of tooth 13, due to malposition and making orthodontic traction unfeasible. (Figure 3)

**Figure 3** - The surgical procedure under general anesthesia for enucleation of the cystic capsule and exodontia of tooth 13.

![Surgical Procedure](image2)

Source: Authors.

The procedure was performed under general anesthesia, with infiltrative local anesthesia in the right upper sulcus with lidocaine, incision and displacement of the mucoperiosteal flap to access the cystic capsule, followed by enucleation of the cystic capsule and exodontia of tooth 13. (Figure 4)
Figure 4 - The incision and displacement of the mucoperiosteal flap to access the cystic capsule, followed by enucleation of the cystic.

![Figure 4](source.png)

Source: Authors.

The surgical specimen was sent for pathological anatomic examination with the following description, presence of thin cystic epithelium of approximately 3 non-keratinized layers. Scattered mucous cells and fibrous capsule with small odontogenic epithelial islands.

We opted for curettage of the surgical site to remove the cystic capsule, extraction the tooth and refinement of the remaining bone borders. Repositioning and inter papillary suture of the flap were performed. (Figure 5)

Figure 5 - The cystic capsule and tooth 13 after extration.

![Figure 5](source.png)

Source: Authors.

The patient is being followed up as an outpatient, and a computed tomography of the 6-month postoperative period shows satisfactory bone formation at the site of the enucleation of the dentigerous cyst and no signs of recurrence. (Figure 6)

Patient does not report symptoms and remains under outpatient follow-up.
Figure 6 – The computed tomography of the 6-month postoperative period shows satisfactory bone formation at the site of the enucleation of the dentigerous cyst and no signs of recurrence.

Source: Authors.

4. Discussion

The vast majority have involvement with the crown of an unerupted tooth and the radiolucent area is closely attached to the tooth at the junction of the cementum and enamel. (Ustuner et al., 2003)

Cysts occur due to accumulation of fluid between the crown of the unerupted tooth and the follicular epithelium (reduced enamel epithelium). (Patil et al., 2019)

The dentigerous cyst may exhibit irregular margins or ill-defined borders, and it is possible to observe the roots of adjacent teeth a resorption or displacement caused by the pressure of the cysts during their development. Large cysts can have a multiloculated appearance and can be considered in the differential diagnosis of ameloblastoma. There are three types of radiographic features usually noted in dentigerous cysts: central, lateral, and circumferential. The most found radiographic feature is a radiolucent cavity that surrounds the crown of an unerupted tooth. The lateral variant, on the other hand, is characterized by a radiolucent appearance observed laterally along the root and encompasses part of the crown of the unerupted tooth. In the circumferential variant, it is a real challenge to diagnose a dentigerous cyst, because the radiolucent cavity involves the entire tooth. Most practitioners are aware of the fact that the radiolucent cavity is associated with the crown of an unerupted tooth extending to the level of the cemento-enamel junction. (Meningaud et al., 2006)

Microscopically, a non-keratinized epithelium composed of 2 to 3 thin layers is observed. Scattered mucous cells are observed and a fibrous capsule is freely arranged and may present small odontogenic epithelial islands of inactive appearance. In inflamed dentigerous cysts it is possible to observe a non-keratinized cystic epithelium with several layers and hyperplastic. Some associations to dentigerous cysts are: ameloblastoma due to the possibility of a potential transformation of nests of odontogenic epithelial cells, mucoepidermoid carcinoma, characterized as a malignant tumor of the salivary glands which can arise through the alteration of mucous cells observed in the lining epithelium of the cyst, and squamous cell carcinoma of the lining epithelium. (Gadbail et al., 2012) (Thompson, 2018)

According to Babu and Santosh (2020) we can include unicystic ameloblastoma as a differential diagnosis, ameloblastic fibroma can also be considered when in younger individuals. However, adenomatoid odontogenic tumor can be considered with differential diagnosis of dentigerous cyst due to pericoronal radiolucency observed mainly in upper or lower canines. However, mucoceles, retention cysts and pseudocysts are also included in the differential diagnosis when a lesion is observed in the maxillary sinus and generating maxillary expansion. (Ustuner et al., 2003)(Dağistan et al., 2007) (Babu & Santosh, 2020)

The lack of correlation between the expression of apoptosis-related factors and cell proliferation markers in dental follicles suggests that the stage of impaction and pericoronal radiolucency did not significantly correlate with the expression of
apoptosis-related factors or cell proliferation markers. The Ki-67-LI marker of dental follicles with positivity in epithelium during proliferation was significantly higher than the Ki-67-LI of epithelium with other morphological characteristics, indicating a high proliferative potential. Dental follicles with inflammatory changes had significantly higher ssDNA-LI and Ki-67-LI than Ki-67-LI of epithelium with other morphological features, indicating a high proliferative potential. Dental follicles with these characteristics suggest that inflammatory changes regulate cell turnover of the epithelial components of the dental follicle. (Edamatsu et al., 2005)

Marsupialization is a surgical procedure that reduces the intracystic pressure and gradually leads to cyst reduction. This procedure causes a cavity in the cyst wall to drain the liquid content of the cyst, and sometimes the cyst wall is sutured to the oral mucosa. The benefit of this method is the ability to promote eruption of the tooth associated with the cyst, with or without orthodontic traction. However, its limitations are the long postoperative duration of treatment and the inconvenience of keeping the wound open. (Ertas & Yavuz, 2003)

It is a consensus that marsupialization and orthodontic traction are the best treatment option for the patient with a dentigerous cyst that includes an unerupted tooth. Orthodontic traction of unerupted teeth is usually performed after marsupialization, when there is enough space for eruption. Some studies have been developed to establish the adequate moment for orthodontic traction. The results obtained were that the spontaneous eruption of the tooth associated with the cyst is predictable within 3 months after marsupialization; however, orthodontic traction may be required for a longer period of time in some cases. (Karamanis et al., 2006) (Caliento et al., 2013)

We believe that enucleation is an effective type of treatment, as is marsupialization; however, when it is observed that there is no possibility of dental traction, as in the case described in this article, we opt for enucleation for a faster and more resolutive treatment.

The prognostic factors for eruption of a cyst associated tooth after marsupialization are debatable. Some authors have reported that numerous factors affect tooth eruption, including patient age, depth, inclination, stage of root formation (open or closed apex) and space availability. (Sumer, Baş, & Yıldız, 2007) In contrast, other authors state that these elements are not significant and do not affect tooth eruption. (McCrea, 2009) Primarily, the goal of the treatment method is marsupialization and tooth eruption with or without the use of orthodontic traction. The explanation for the failure of orthodontic traction is that the tooth axis with severe lingual tipping of the canine may prevent the eruption of the canine. (Aoki et al., 2018)

5. Conclusion

The treatments described in the literature, such as marsupialization, are very effective and have been used in surgical procedures with satisfactory results. However, we believe that enucleation is a type of effective treatment, as well as marsupialization; however, when it is observed that there is no possibility of dental traction, as in the case described in this article, we opted for enucleation for a faster and more resolutive treatment. However, we believe that more prospective studies related to the comparison of success rates are needed, when comparing the types of treatment available for this pathology.

References


