

Maxillary dentigerous odontogenic cyst: the sinusitis simulator – case report

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Abstract

Cell degeneration of the enamel's starry reticulum gives rise to the primary cyst before the mineralized tissue is formed. This cyst appears in place of a normal, un-erupted or supra-numerous teeth and is frequently observed among young people in middle age. A 17-year-old boy, born and resident in Portugal, 2 months before hospitalization starts complaints of nasal obstruction on the right, and later the left, with progressive worsening and pain located in the dental arch and gums and appearance of swelling gingival pain with chewing in the last two weeks. On clinical examination there was a painful swelling of the right cheek, without any other inflammatory signs. Attached to the upper right maxillary dental arch there was a soft, vascularized and painful mass of 3 cm in diameter. Blood tests found hemoglobin 15.3 g/dL (reference: 10.3-13.5 g/dL), leukocytes 9110/uL (reference: 6-16/uL) and c-reactive protein 0.05mg/dL (reference: < 0.5 mg/dL). Computed tomography and magnetic resonance imaging described a 49x45x44 mm right maxillary odontogenic cyst, with an erosion and thinning of the external, posterior, and anterior wall of the maxillary sinus and left deviation of the nasal septum. The diagnosis of odontogenic cyst was made and the cyst was making an orbital compression and involving almost all maxillary sinuses. He was submitted to a videoendoscopic technique associated a Caldwell-Luc technique. This combined technique promotes a better view and control all borders of the lesion. The surgery completely removed the lesion and the supernumerary tooth. The follow up was done without complications. It is important, to bear in mind that in the presence of complaints associated with the sinuses and oral cavity, the need to exclude non-surgical pathology requires the performance of a detailed objective examination.

Keywords: Dentigerous cyst. Mouth. Sinusitis.

Quisto dentígero odontogénico: o grande simulador – relato de caso

Resumo

A degeneração celular do esmalte dá origem ao quisto primário antes da formação do tecido mineralizado. Este quisto aparece no lugar de um dente normal, não irrompido ou supranumerário e é frequentemente observado na segunda década de vida. Os quistos dentígeros podem ser diferenciados dos queratocitos, ameloblastos, fribroblastomas, fibromas, calcificação epitelial, tumores odontogénicos e adenomatoides e ameloblastomas. Apresentamos um adolescente de 17 anos, natural e residente em Portugal, previamente saudável. Queixas de obstrução nasal com agravamento progressivo e dor localizada à arcada dentária e gengiva direita com 2 meses de evolução. Ao exame objetivo apresentava tumefação dolorosa na hemiface direita, sem outros sinais inflamatórios.

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Received: 11-11-2023
Accepted: 13-11-2023
<https://pjp.spp.pt>

Available online: 07-02-2024
Port J Pediatr. 2024;55(1):53-57
DOI: 10.24875/PJP.M23000123

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Anexa à arcada dentária direita, objetivava-se uma massa mole, vascularizada e dolorosa com cerca de 3 cm de diâmetro. Laboratorialmente apresentava hemoglobina de 15,3 g/dL (referência: 10,3-13,5 g/dL), leucócitos de 9110/uL (referência: 6-16/uL) e proteína C-reativa 0,05 mg/dL (referência: < 0,5 mg/dL). A tomografia computadorizada e a ressonância magnética descreveram um quisto odontogênico do maxilar superior direito de 49x45x44mm, com erosão e afinamento da parede externa, posterior e anterior do seio maxilar e desvio esquerdo do septo nasal, com extensão intrassinusal, com compressão da órbita e envolvimento de quase a totalidade do seio maxilar. Procedeu-se à videoendoscopia associada à técnica de Caldwell-luz para a remoção do quisto, para otimização da análise da extensão da lesão. Na presença de queixas associadas aos seios da face e cavidade, importa excluir patologia cirúrgica através de um exame objetivo detalhado e minucioso, uma vez que poderá ter implicações prognósticas e terapêuticas.

Palavras-chave: Patologia da cavidade oral. Quisto odontogênico. Sinusite.

Keypoints

What is known

- Odontogenic cyst is not a rare disease, but at pediatric age is not so common.
- The patients present with slow-growing asymptomatic mass in mandible.
- It is obligatory to make this diagnosis when we are presented with sinusitis complaints.
- The treatment and follow-up are not consensual.

What is new

- Videoendoscopic technique associated a Caldwell-Luc technique can help to have a minimal sinuous wall damage.
- A multidisciplinary team is crucial to make the correct diagnosis, treatment, and follow-up.

Introduction

The embryologic process of tooth development is secondary to the invagination of oral epithelium into the jaws. This process has the main challenge to give rise to a tooth, but the odontogenic epithelium has the potential to persist after failing to break down following tooth eruption¹ and, when stimulated, may be the origin of cysts or tumors of odontogenic origin².

Odontogenic cysts (OCs) are intraosseous pathological cavities lined by an odontogenic epithelium and filled with fluid or semifluid content³⁻⁵. They comprise of up to 90%¹ of jaw cysts and are classified, according to the World Health Organization (WHO), as inflammatory or developmental⁶. The inflammatory group consists mainly of radicular and inflammatory collateral cysts, and the developmental group are dentigerous cysts (DTGs) and odontogenic keratocysts (OKCs). Immunohistochemistry is needed to make a final diagnosis^{7,8}.

It most commonly affects the anterior mandible, but other regions of the jaw can also be affected⁸. The patients present with slow-growing asymptomatic mass in the mandible^{7,8} that is usually an incidental radiographic finding, and most frequently in middle age⁸. However, when secondarily infected they can present with inflammatory signs due to compression of a neurovascular bundle^{1,7-9}.

Radiologically, the lesion may be unilocular or multilocular with borders that, although generally well defined, may occasionally show loss of cortical integrity¹⁰.

Histologically, they have a non-keratinized epithelium with a superficial layer of cuboidal or columnar cells¹¹.

The maxillary odontogenic cyst is a rare entity, namely in the pediatric population, that can simulate a respiratory tract infection, such as sinusitis. It can have an aggressive clinical course, behavior and recurrence rate. It is often mis-diagnosed.

The optimal treatment remains unclear. Extensive cysts involving the orbital or the inferior orbital nerve call for a conservative strategy¹². It consists of either decompression or marsupialization as a single modality or followed by cyst enucleation³. Even though cysts are usually treated by enucleation, they may grow extensively to involve the maxillary sinus cavity¹³. The surgery treatment can be accomplished with or without curettage and peripheral ostectomy¹⁴. It is important to notice that segmental resection, which provides free margin excision, may result in high mobility and cosmetic damages¹⁵. Segmental resection has the lowest recurrence rate because of the elimination of adjacent daughter cysts and surrounding mucosa. However, it has a high morbidity, including the loss of dentition, oroantral communication, chronic sinusitis and a need for extensive re-construction¹³. Namely, intervention of cysts found in the maxillary sinus may lead to oroantral fistula formation and chronic rhinosinusitis¹⁶. There are advantages when performing transnasal endoscopic sinus surgery such as avoidance of extensive tooth extraction and oroantral communication, better control of orbital floor and infraorbital



Figure 1. Gingival swelling on first superior right molar.

nerves, avoidance of transoral incisions and less post operative cheek swelling¹³.

The aim of this article is to show the importance of keeping in mind the differential diagnosis of upper respiratory tract infection, especially in pediatric age. This article reports a 17-year-old male with an odontogenic cyst who was admitted to the emergency department with swelling and pain in the upper right gingival area for two months.

Case report

A 17-year-old male, presented at the emergency department with a progressive bilateral nasal obstruction, local pain and change of skin color on arch and gums along with gingival swelling for 2 months (Fig. 1). On clinical examination, there was a painful swelling of the right cheek without any other inflammatory signs. Attached to the upper right maxillary dental arch there was a soft, vascularized, and painful mass of 3 cm in diameter. Blood tests found hemoglobin 15.3 g/dL (reference: 10.3-13.5 g/dL), leukocytes 9110/uL (reference: 6-16/uL) and c-reactive protein 0.05mg/dL (reference: < 0.5 mg/dL). Computed tomography and magnetic resonance imaging described a 49x45x44 mm right maxillary odontogenic cyst with an erosion and thinning of the external, posterior, and anterior wall of the maxillary sinus and left deviation of the nasal septum (Fig. 2). The diagnosis of odontogenic cyst was made. The patient was proposed to a video-endoscopic technique associated to a Caldwell-Luc technique with intrasinus extension.

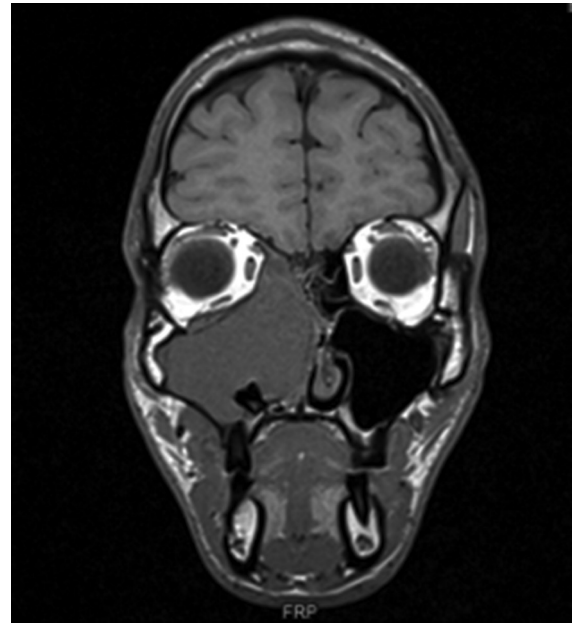


Figure 2. Right maxillary odontogenic cyst with erosion and thinning of the external, posterior and anterior wall of the maxillary sinus and left deviation of the nasal septum.

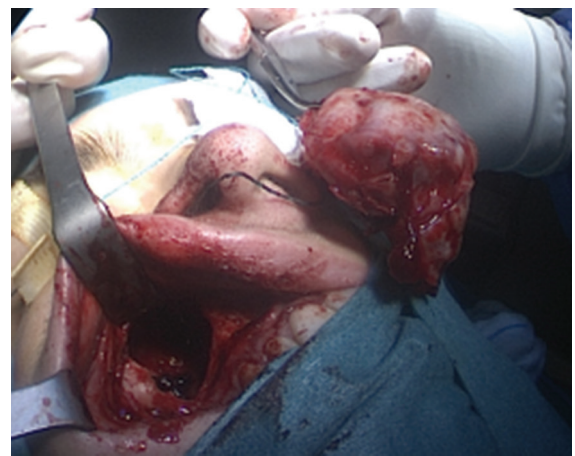


Figure 3. Excised odontogenic cyst - Caldwell-Luc approach.

The cyst was causing orbital compression and involved almost completely the maxillary sinus. This combined technique promotes a better view and control all borders of the lesion. The lesion and the supernumerary tooth were completely removed during surgery. This endoscopic approach provides a better control of the sinus (Fig. 3). Pathological examination was

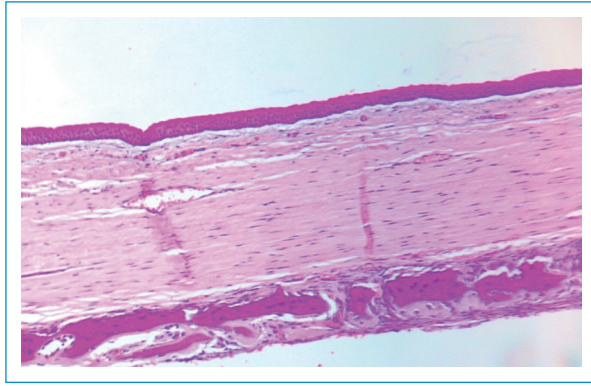


Figure 4. Non-keratinized pavement epithelium cyst with fibrous wall and without bone erosion.

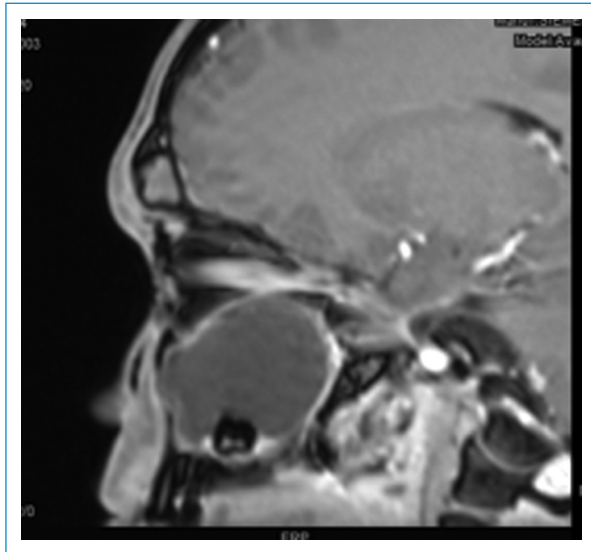


Figure 5. Odontogenic cyst around the crown of the teeth.

compatible with a dentigerous odontogenic cyst surrounded by non-keratinized epithelium (Fig. 4). The follow-up was done without complications.

Discussion

Pediatric odontogenic cysts occur more frequency in middle age, and are rare in pediatric age. Dentigerous cyst accounts for 24% of cases^{1,5} (Fig. 5). They are usually asymptomatic^{5-8,10-13}, unless if secondarily infected, like in the present case, with nasal obstruction and pain, which made a good exam important to make the differential diagnosis of upper respiratory tract infection.

Odontogenic cysts can mimic several tumors or vascular disease and can lead to consequences such as pathologic fractures⁵. The diagnosis is based on its distinct clinical and radiographic characteristics; TC is helpful to characterize correctly.

The treatment is not linear, because extraction of mandibular molar can have a significant static and tooth impact. The choice of treatment should take into account various factors, including the patient's age, size and location of the cyst, soft tissue involvement, history of treatment and the histological variant of the lesion. The goal is to choose the treatment modality that will completely eradicate the lesion with the lowest possible risk of recurrence and the least morbidity. In the present case, considering the age and associated symptoms, the videoendoscopic technique associated to a Caldwell-Luc technique with intrasinus extension was the option.

Some recommendations describe that these patients should be kept under follow-up for a minimum of 3 years¹⁷, but no guidelines were previously made and more knowledge is necessary.

Funding

None.

Conflicts of interest

None.

Ethical disclosures

Protection of human and animal subjects. The authors declare that no experiments were performed on humans or animals for this study.

Confidentiality of data. The authors declare that they have followed the protocols of their work center on the publication of patient data.

Right to privacy and informed consent. The authors declare that no patient data appear in this article.

Use of artificial intelligence for generating text. The authors declare that they have not used any type of generative artificial intelligence for the writing of this manuscript, nor for the creation of images, graphics, tables, or their corresponding captions.

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