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OPEN
Romanian Journal of Rhinology, Vol. 7, No. 26, April-June 2017
DOI: 10.1515/rjr-2017-0013

CASE REPORT

Cystic lesions of the maxilla – clinical considerations and differential diagnosis

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ABSTRACT

Cystic lesions of the maxilla are benign entities with both odontogenic and non-odontogenic origins. The maxillary cyst is a benign tumor of the bones of the jaw, which has a membrane and contains a fluid, semi-solid or mixed (liquid / semi-gaseous) material. These often lead to deformities in the jaw area. Cases are specific by framing pathological rarity, etiology, pathogenesis and clinical symptoms.

Considering the large entity of cystic formations that can be found in the maxilla region, we selected two patients with cystic formations of the upper jaw, which were part of different pathological etiology categories, with special rarity occurring, evolutionary appearance and difficult to classify in terms of pathognomonic signs.

KEYWORDS: cysts, maxilla, nasolabial cyst, radicular cyst.

INTRODUCTION

Cystic lesions of the maxilla are benign entities with both odontogenic and non-odontogenic origins. Given the etiologic diversity and forms of pathological lesions of the maxillary bone, cysts have existed over time in many of these classifications. The most recent classification of the World Health Organization (WHO, 1992), updated and published in 2001, provides a good systematization of these entities[1]. The developmental cysts are divided into odontogenic cysts (keratocyst, follicular cyst, cyst rash, lateral periodontal cyst, gingival cyst of the adult, gingival cyst (alveolar) of new-born, calcified odontogenic cyst, glandular odontogenic cyst) and non-odontogenic cysts (nasopalatine cyst, median palatal cyst, nasolabial cyst, globulomaxillary cyst). Inflammatory cysts are represented by the radicular cysts, residual cysts or paradental cysts. Classification of the maxilla cysts includes also the pseudocysts[1].

Considering the large entity of cystic formations that can be found in the maxilla region, we selected two patients with cystic formations of the upper jaw, which were part of different pathological etiology categories, with special rarity occurring, evolutionary appearance and difficult to classify in terms of pathognomonic signs.

The nasolabial cyst is an extremely rare cystic mass, located paramedianly to the wing of the nose, in the nasal alar region, with an incidence of 0.7%. Nasolabial cysts are diagnosed frequently in female adults, in the fourth to fifth decade of life. From a clinical point of view, the lesion presents as a painless asymptomatic swelling in the nasolabial region. Common clinical features include a slowly growing painless mass, which results in the obliteration of the nasolabial sulcus, nasal vestibule and maxillary labial sulcus. The initial diagnosis and treatment are usually made in early stages because the lesion causes cosmetic problems; rarely does it achieve large dimensions[2].

Radicular cysts are the most common inflammatory cysts, which develop due to pulpal necrosis caused by caries or traumatic insult. The necrosis stimulates the epithelial cells rest of Malassez in the periodontal ligament to proliferate. Radicular and residual cysts are by far the most common cystic lesions in the jaws, representing 52.2% of jaw cysts and 62% of odontogenic cysts. They occur in all tooth-bearing areas of the jaws, about 60% being found in the maxilla and 40% in the

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mandible. A particularly high frequency can be found in the maxillary anterior region[3].

CASE REPORT

Case 1

A 45-year-old female patient referred to us for upper lip region swelling associated with the obliteration of the right nasolabial sulcus, asymmetry in the nasal valve aria and nasal obstruction (Figure 1). The swelling and tumefaction were evident for approximately 5 years, over which time the symptoms presented a slow progressive evolution.

On examination, the lesion was approximately 4 cm in diameter, with a soft and cystic consistency, mobile and fluctuant on palpation. Fullness of the right maxillary labial vestibule was present, between the maxillary incisors and right canine. The overlying skin and oral mucosa were normal in colour.

The anterior rhinoscopy and the nasal endoscopy revealed a partial deformation of the anterior part of the floor of the right nasal fossa (Figure 2).

The cranio-facial MRI showed a rounded, well-demarcated, homogeneous, low-density soft tissue lesion located in the right nasolabial region (Figure 3); characteristics of fluid mass in T1 (low intense) and T2 (bright) views.

The surgical treatment consisted in the removal of the cystic formation through sublabial approach, under general anaesthesia. A 5-cm incision was performed through the right gingivo-jugal sulcus (Figure 4), the

Figure 1 Nasal valve region asymmetry and obliteration of the right nasolabial sulcus.

Figure 2 Nasal endoscopic examination – partial deformation of the anterior part of the floor of the right nasal fossa.

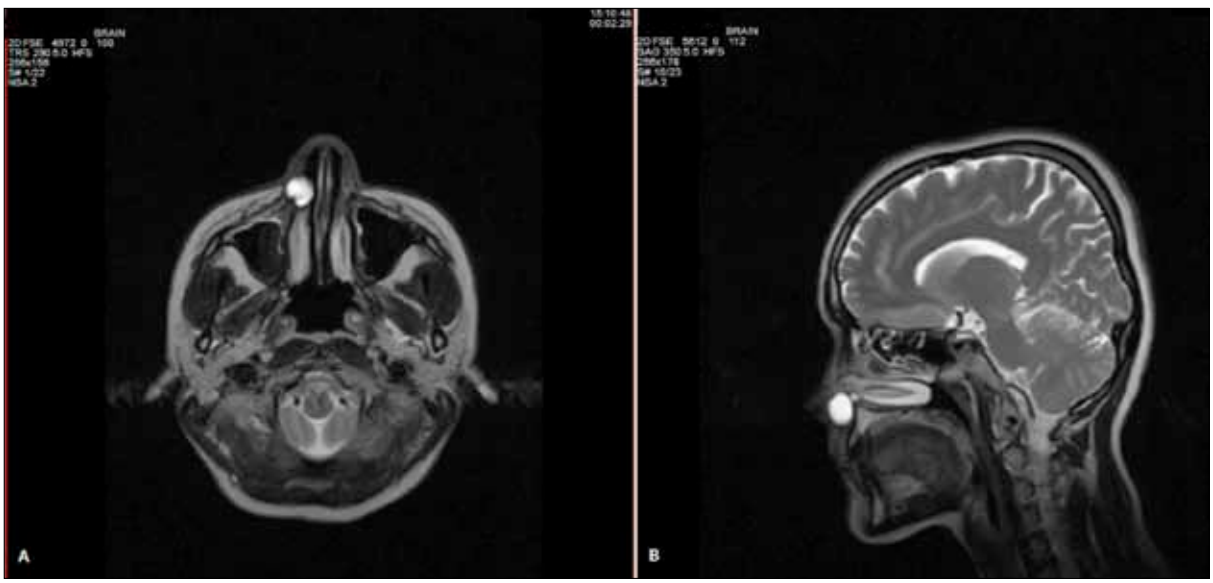

Figure 3 Cranio-facial MRI, axial **(A)** and sagittal **(B)** slices, shows a well-demarcated, rounded, homogeneous, low-density soft tissue lesion in the nasolabial region.

Figure 4 Intraoperative view – incision in the right gingivolabial sulcus and dissection of the cystic formation.

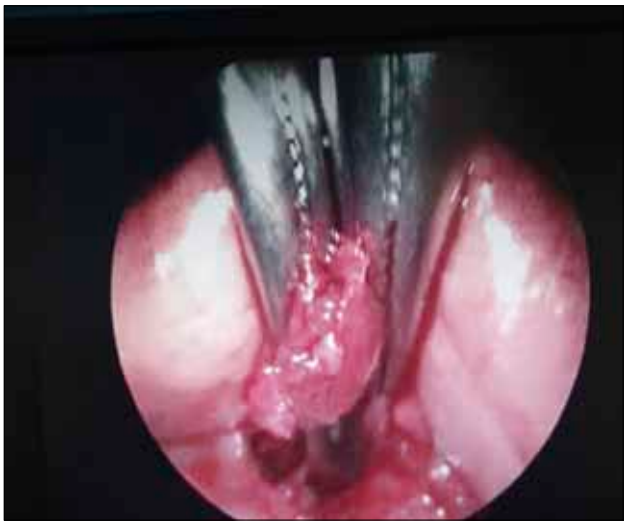

Figure 5 Intraoperative view of the excised specimen.

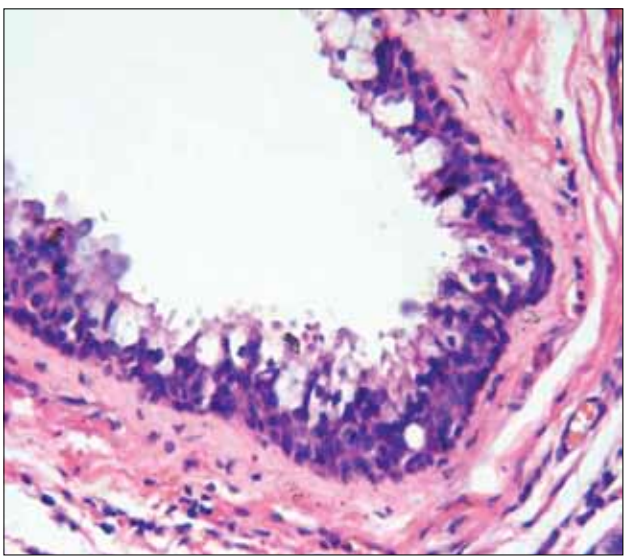

Figure 6 Histopathological examination - cystic structure lined with pseudostratified columnar and stratified squamous epithelium.

cystic formation was dissected up to the piriform aperture and separated from the labial mucosa, the overlying skin and the nasal mucosa.

After the curettage of the contiguous hard and soft tissue, the wound was closed primarily.

On the seventh day postoperatively, the patient presented an inflammatory reaction at the right upper lip level, with remission under anti-inflammatory treatment.

The follow-up was performed after one and two months and showed a favourable evolution of the patient.

Microscopic evaluation of the excised specimen (Figure 5) revealed a cystic structure lined with pseudostratified columnar and stratified squamous epithelium (Figure 6), confirming the diagnosis of nasolabial non-odontogenic cyst.

Case 2

A 30-year-old male presented for important deformity of the anterior left maxillary wall, inflammation and swelling of the left cheek region (Figure 7), accompanied by recurrent episodes of suborbital pain, mild nasal obstruction and odontalgia. Palpation showed a semi-hard consistency imprecisely defined structure at the level of the left maxillary sinus region.

Oral clinical examination revealed an important corono-radicular destruction on tooth #26. To determine the actual extension and borders of the lesion, a cone beam computerized tomography (CBCT) was performed. On the axial, coronal, and sagittal CBCT images, a radiolucent lesion that invaded the entire left maxillary sinus, with partial destruction of the anterior wall of the maxillary sinus, was detected (Figure 8).

Under general anaesthesia, the patient underwent a surgical procedure consisting in a combined ENT and oro-maxillo-facial technique, which included surgical enucleation of the cyst. A crevicular incision was made in the sublabial region, between the 24th and 27th teeth, and a full-thickness modified mucoperiosteal

Figure 7 Deformation of the left cheek region.

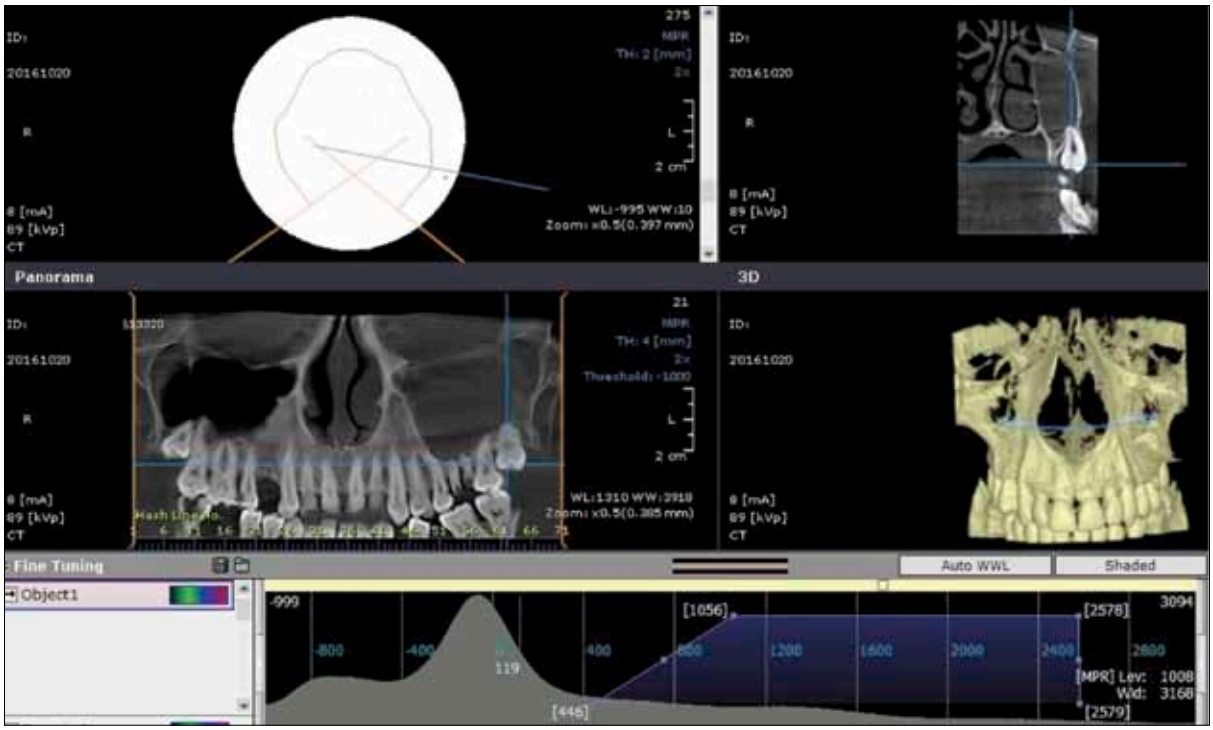

Figure 8 CBCT – radiolucent mass invading the entire left maxillary sinus.

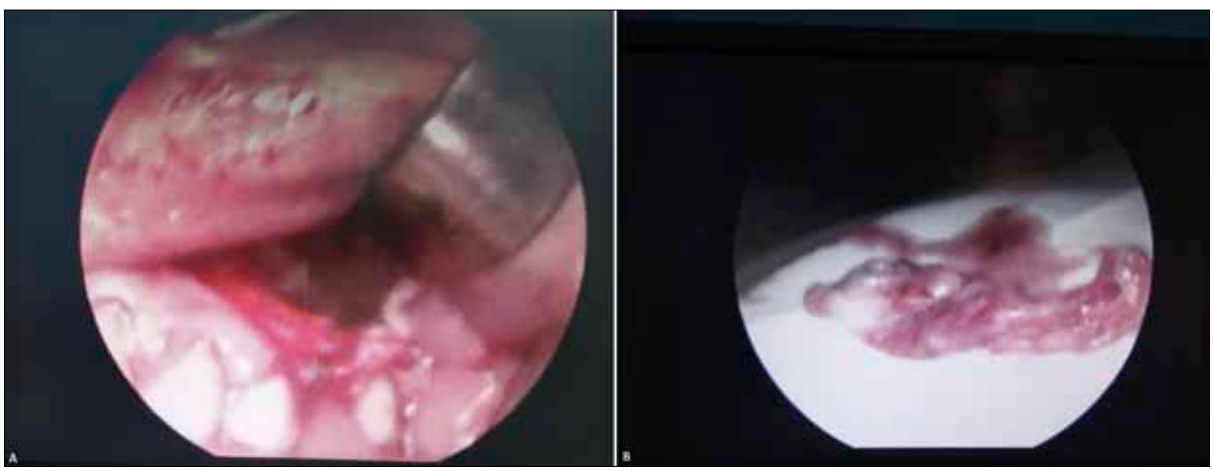

Figure 9 A, B. Intraoperative view.

Ochsenbein-Luebke flap was reflected. We performed a Caldwell-Luc procedure, enucleation of the cystic lesion, tooth 26 removal and an alveoloplasty (Figure 9).

Hemostasis was realized using an intrasinusal dressing maintained for 48 hours. Flap closure was done with 4-0 Vicryl. Postoperatively, the patient received antibiotics and analgesic drugs for 3 days.

The follow-up was performed 1 and 2 months after surgery, showing a favourable evolution with no recurrence signs.

The histopathological examination indicated an important inflammatory reaction composed of plasma cells, lymphocytes, neutrophils and leukocytes in the intraepithelial and subepithelial areas of the cystic structure and nodules of opaque yellow material, representing cholesterol. The cyst was lined by a nonkeratinizing stratified squamous epithelium of varying thickness (Figure 10). The final diagnosis was radicular cyst.

DISCUSSIONS

Even if they have different origins, the multiple types of maxillary cysts show a similar clinical picture; differentiation is induced by location, expansion, growth direction, or the occurrence of complications.

Research on the genesis of the maxillary cyst starts with the medical history, that gives us data about the

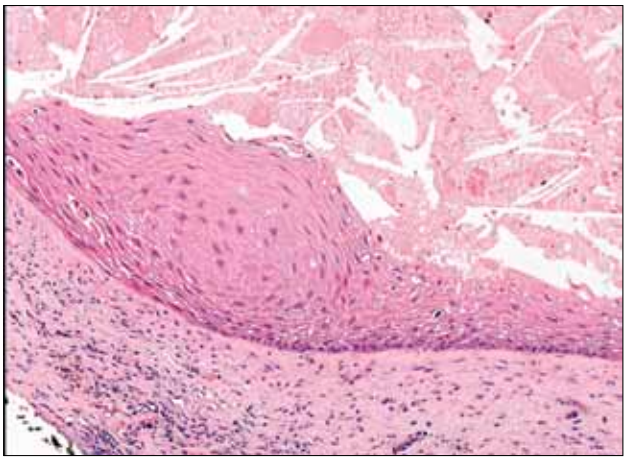

Figure 10 Cyst lined with nonkeratinizing stratified multilayered squamous epithelium.

existence or not of symptoms onset (acute inflammatory phenomena, mucous or cutaneous fistulas, pain, paresthesia, bone deformities, etc.) and their characteristics. Then, the clinical examination gives complementary means of diagnosis[4].

The nasolabial cyst is a developmental nonodontogenic type of soft tissue cyst occurring in the nasal alar region. Since its original description performed by Zukerkandl in 1882, the nasolabial cyst has been known by other names such as nasal-vestibular cyst, nasoglobular cyst and nasoalveolar cyst[5]. Since 1953, the lesion has been known as the Klestadt's cyst, after the scientist who performed complex studies on this pathology[2].

There are two hypotheses regarding the pathogenesis of the nasolabial cyst. According to the first one, the cyst is derived from the epithelial cells retained in the mesenchyme after fusion of medial and lateral nasal processes during the 4th week of intrauterine life. The second hypothesis suggests the persistence of epithelial remnants from the nasolacrimal duct extending between the lateral nasal process and the maxillary prominence[2,5].

Nasolabial cysts diagnosis is established mostly by the clinical findings. Bidigital palpation can reveal a fluctuating swelling between the floor of the nasal vestibule and the gingivolabial sulcus. Nasolabial cysts do not present any finding on the plain radiographs, except when they cause significant maxillary bone erosion.

Computed tomography (CT) and magnetic resonance imaging (MRI) scans may reveal the cystic nature of these lesions, their relation to the nasal alae and the maxillary bone, as well as bone involvement. In our patient, the MRI examination facilitated the diagnosis.

The differential diagnosis has to be made with odontogenic lesions (canine space abscess, follicular cysts, periodontal and residual cysts, and salivary gland neoplasms)[6]. In the literature, there is only one case of carcinoma progressing from a nasolabial cyst reported. A special attention should be taken to the differentiation of the infected nasolabial cysts with the furuncle of the nasal vestibule floor.

Histopathologically, the cyst is lined with pseudostratified columnar epithelium and, occasionally, a stratified squamous epithelium[1,6].

In our patient, the diagnosis was based on clinical and radiological findings, which described all of the characteristics of the nasolabial cyst, and it was confirmed by histopathology. Also, all the features of this rare entity were similar with the literature.

The origin of the radicular cyst epithelium lies with rests of Malassez, which are epithelial remnants of Hertwig epithelial root sheath that lie dormant within the periodontal ligament. The pathogenesis of the radicular cyst is an inflammation-stimulated cyst secondary to the pulpal infection and necrosis of the periapical tissues. The inflammatory cells secrete lymphokines to neutralize, immobilize and degrade bacteria. These reactions induce bone resorption due to the elaboration of interleukin-1 and osteoclast-activating factors. During the same process many other factors are released either directly or indirectly act as epithelial growth factors, stimulating the proliferation of the rests of Malassez in the periapical granuloma. As the epithelial cell mass enlarges, the central cells become distant from their blood supply and break down, forming a cyst. The cyst continues to enlarge due to epithelial proliferation in the lining and to the hydrostatic pressure. The osmotic gradient favours transudation of fluid into the cyst's lumen, causing further resorption of the surrounding bone[7].

Many radicular cysts are symptomless, being discovered when periapical radiographs are performed[8]. Radicular cysts are probably the most common cause of swelling of the jaws and usually have a progressive slow evolution. At first, the enlargement is bony hard but as the cyst increases in size, the covering bone becomes very thin. Expansion of the cyst can cause an erosion of the floor of the maxillary sinus. The internal structure of the cyst is homogeneous and radiopaque relative to the sinus cavity.

It is very rare to encounter odontogenic cysts that have reached a very large size like in our presented case. Indeed, the odontogenic keratocyst (OKC), dentigerous cyst and traumatic bone cyst might reach such exceptional wideness. In general, radicular cysts tend to grow slowly and do not reach large sizes. However, they may enlarge to occupy an entire quadrant of the jaws[2,9-11]. The cyst in our case reached very large dimensions by invading a major part of the left maxillary sinus. Radicular cysts are discovered either by bone deformation and inflammation, or, like in our case, by chance, during routine radiographic examination[11].

The radicular cysts generally grow slowly and extend either to the anatomical neighbouring such as sinuses, nasal cavity, nasal vestibule, or palate[11]. The extension of the odontogenic cysts to the maxillary sinuses is in relation with the proximity of the lesion to the sinuses. The type of the lesion has no effect on the extension. Infected cysts show symmetric expansion and diffuse to the spaces like sinuses or nasal cavity at the points where the bone is weak. Because of this, they remain asymptomatic and do not cause asymmetry unless sinuses are completely occupied[11]. Similarly, the cyst in the presented case was extended to the maxillary sinus.

In a large cyst, like the one we described above, when it is in relation with the maxillary sinuses, the panoramic radiography is not a sufficient imaging technique. CBCT has some advantages over panoramic radiography like high bone detail, information upon the dimension of the lesion and the relation of the cyst to nearby structures (paranasal sinuses, orbital and nasal cavity). It is also useful during the postoperative evaluation[12].

The differential diagnosis of the radicular cysts is made with**:** odontogenic keratocyst (OKC), adenomatoid odontogenic tumor, ameloblastoma, ameloblastic fibroma[3]. OKC is predominately located in the posterior body of the mandible and rarely seen in the maxilla. It shows minimal bone expansion and the involved teeth are kept vital. It may have a smooth round or oval shape identical to other cysts and usually it has internal septa that give a multilocular appearance[8]. In our patient, the cyst had well-defined borders, it was unilocular without internal septa and it had devital relevant tooth. In opposition to the cyst in our case, the adenomatoid odontogenic tumor is often associated with a missing tooth. Furthermore, internal radioopacities develop in most of the cases. Predominantly, ameloblastoma develops in the posterior region of the mandible and when it occurs in the maxilla, the third molar region is involved. From that point, it may extend to the maxillary sinus and nasal floor. The border of the lesion is often curved. While small lesions are uniform radiolucent, greater lesions have multilocular appearance[8]. The lesion in our case emanated from the maxillary incisor region and it was unilocular. Furthermore, ameloblastic fibroma usually develops in the premolar-molar area of the mandible, and it often has a relationship with an unerupted tooth. The internal structure may be multilocular[8].

The histopathological examination, pulp vitality tests, long-term follow-up are useful for a concrete and correct diagnosis.

The treatment of the radicular cysts has to take into consideration the type and the volume of the lesion. Small cysts usually heal up with successful endodontic treatment. In large cysts with sinus extension, which maintain a minimum separation between the bone cyst wall and the sinus mucosa, a radical cure of the maxillary sinus is not needed. But, in case bone erosion is present, the radical cure of the maxillary sinus is indicated, like in the case we presented.

CONCLUSIONS

Cystic lesions of the maxilla can have both odontogenic and nonodontogenic origin. Considering the multiple types of cystic lesion, a correct diagnosis cannot be determined only on symptoms, clinical and radiological evaluation, and it has to include a histopathological examination.

From our point of view, the two cases we presented are relevant because of the lack of clinical symptoms and the presence of important bone lesion. Through this presentation we sustain the necessity of the radiologic information, especially CT or MRI scans, in order to have a complete correlation between the extension of the lesion, the nasal and sinus cavity and the teeth. This information is crucial in order to recommend the proper surgical treatment technique. A complete removal of the cystic mass is needed to prevent recurrences.

Conflict of interest: The authors have no conflict of interest.

Contribution of authors: All authors have equally contributed to this work.

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