CASE REPORT

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Mucous retention cyst in maxillary sinus with expansion of maxillary tuberosity: Case Report

Abstract:

Mucosal retention cyst in the maxillary sinus is a benign entity that results from obstruction and dilation of seromucous glands ducts due to inflammation, infection or reaction of maxillary sinus mucosa to allergic processes. It is an asymptomatic lesion, usually identified in routine imaging tests requested by professionals. The present study aims to present a case of maxillary sinus mucosal retention cyst in a 63 years old patient who reported difficulty in adapting her total upper prosthesis and persistent pain in the left maxilla. The treatment was surgical enucleation.

Keywords: Retention cyst; Maxillary sinus; Oral diagnosis; Enucleation.

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INTRODUCTION

Classified as pathological cavity lined by epithelium a cyst is supported by fibrous connective tissue and may be filled by liquid, air or solid content regardless of its origin. Cysts considered non-odontogenic originate from the fusion lines of the bones or embryonic processes of the jaw, develop in the maxillofacial region and tend to progress slowly in size, probably in response to a slight increase in luminal hydrostatic pressure. despite a few cysts result in the inclusion of epithelium along the fusion lines of the embryonic processes, the majority of the cysts of the gnathic bones are lined by epithelial tissue derived from odontogenic epithelium¹.

Dome-shaped radiopaque shadows frequently seen on the floor of the maxillary sinus and sometimes incorrectly attributed to anthropoid mucoceles seem to represent the focal accumulation of inflammatory exudate that lifts the epithelial lining of the sinus and the periosteum away from the underlying bone, to form characteristic structures. Thus Its histological appearance is, the normal or inflamed lining of the maxillary sinuses².

Odontogenic cysts are frequently found in dental practice; in maxillary sinuses, the most frequent pathology is the mucosal retention cyst and occurs about 10.1% of cases³, the mean age of patients affecting being 29 years; the condition of the dentition and periodontics do not significantly influence the presence of mucous retention cysts. However, age and endodontic condition are associated with the location of the cyst. Regarding lesion size, the average size of the mucosal retention cysts in the maxillary sinus ranges from 6.28±2.93 mm⁴. More pathological findings in the maxillary sinus are reported in men than in women, having as more affected regions the medial wall and sinus floor⁵.

It's been reported that these cysts originate by the blockage of an antral seromucous gland, resulting in a cystic structure lined by ductal epithelium and filled by mucin. The majority of these lesions are asymptomatic, although there may be some slight sensitivity in the vestibular fundus region or, more rarely, a palpable vestibular expansion located in this region⁶. The mucosal retention cyst in the paranasal sinuses develops when drainage of the sinus is obstructed by inflammatory processes, trauma, surgery or tumor. The outcome of the secretion retention varies according to the anatomical location and size of the cyst having as consequence bone erosion and compression of adjacent structures⁷. The maxillary sinus retention cysts is expansive inflammatory cyst that presents radiographically as a radiopaque structures with a distinctly rounded edge that can be single or multiple located in the sinus wall and with absence of any cortical bone. It presents slow and limited growth in order to preserve mucosal integrity. Most maxillary sinus retention cysts feature spontaneous regression or remain stable without change in size at the long term⁸.

The diagnosis, is based on clinical history, physical and imaging examinations. Conventional panoramic radiographs may present with bone erosion or opacification; however, computed tomography (CT) is a valuable diagnostic method to the diagnosis of mucous cyst^{6,7}. On panoramic and periapical radiographs, the maxillary sinus mucosal retention cyst is hemispherical, homogeneous, radiopaque and well delimited. In most time, they are discovered incidentally found during the examination of images, they have a connection with the floor of the maxillary sinus and exhibit small clinical importance^{6,9}.

The differential diagnosis in cases of mucous retention cyst one should polyps, sinus lining hyperplasia as a result of an odontogenic infection, maxillary sinusitis and neoplasia arising within the soft tissues of the lining⁶⁻⁸. However, some of them can increase in size and cause obstruction of the natural ostium of the maxillary sinus. In addition, individuals with a lesion of size >20mm or bilateral cysts at premature diagnosis exhibit a higher risk of cyst growth⁷.

In a histopathological examination, the mucosal retention cyst presents limited by a columnar pseudostratified epithelium interspersed with occasional mucosal cells with minimally inflamed support elements. against the pseudocyst that doesn't showing of epithelial lining, deposits of mucous material surrounded by lightly compressed connective tissue are usually observed when it is not treated, although as they have limited growth potential, they are not destructive and most spontaneously rupture occur.

Thus, only followed up clinically and radiographically is necessary⁶. The formation of the mucosal retention cyst is attributed to a combination of sinus obstruction and inflammation. Regarding the treatment, it can be use enucleation technique, completely removing the cyst. And in cases of large cysts, marsupialization technique can be used. It has an excellent prognosis and relapse rarely occurs after complete removal¹.

CASE REPORT

A 63 year-old female patient, from Buritama (SP), was referred to department of surgery and bucomaxilofacial trauma clinic for São Paulo State University (UNESP), School of Dentistry, campus Araçatuba (FOA) in October 2013, complaining of pain in face.

During the anamnesis, the patient reported pain in the left maxilla, in the tuberosity region. The patient has reported having suffered a car accident with facial trauma for 19 years, when she loss of all dental elements of the left maxilla due. The extra-oral examination, did not demonstrate change in facial symmetry or swelling.

During the intra-oral examination, there was a small swelling in the maxillary tuberosity region of the left side (Figure 1). On palpation, the lesion had a firm consistency and showed a normal appearance.



Figure 1. Intraoral view, swelling in left maxillary tuberosity.

Shortly after evaluation, as the first procedure, a fine needle aspiration (FNA) procedure was performed in this region, using a 20 mL disposable syringe, to rule out some suspicions and assist in the lesion treatment. It was noticed that the fluid was thick because of the difficulty in performing the puncture and had a yellowish color, increasing the hypothetical diagnosis of cyst. The material was stored and sent to the laboratory for cytopathological examination, which returned with the diagnosis of maxillary sinus lesion. After a few weeks the result was confirmed as cystic lesion.

Simultaneously with the cytopathological examination, imaging exames were obtained through panoramic radiography (Figure 2) and computed tomography (CT) (Figure 3), where it was verified, in the left maxillary sinus, a well-defined, rounded, homogeneous radiopaque zone was observed, occupying about half of the maxillary sinus.



Figure 2. Preoperative Panoramic Radiography.



Figure 3. CT Scanner, front cut histopathological analysis.

It had not implying any effect on adjacent bone walls and floor. In the computed tomography of the paranasal cavity, it was evident a hyperdense mass in the left maxillary sinus. A 3D image was requested for further details of the cystic lesion (Figure 3).

After the analysis of the imaging exames, it was propose a surgery for the removal of the lesion that was scheduled at Santa Casa de Misericórdia de Araçatuba, two months after the initial consultation. Preoperative blood tests were requested: complete blood count, coagulogram, prothrombin time and time of partially activated thromboplastin, glycemia, triglycerides, urea, creatinine, TGO, TGP, urine, all of which turned to be normal.

Thus, enucleation of the lesion and bone curettage were chosen. The surgical procedure was performed under general anesthesia, patient was in DDH (horizontal dorsal decubitus), performing nasotracheal intubation, oropharyngeal plug and ocular protection with micropore, intrapsychic and extra-buccal antisepsis with depleting PVPI and topical 10%. For anesthetic used, 0.5% bupivacaine hydrochloride solution containing 1: 200,000 epinephrine diluted in 1:1 distilled water in a fundal region of the phonix in the posterior region of the left maxilla was used. After that an incision was performed with blade number 15 on the collar and (Newman), in the left maxilla with the mucoperiosteal detachment (Figure 4), the buccal wall of the maxillary sinus was opened, then the enucleation of the cystic lesion was done (Figure 5), followed by bone curettage with the curette and the collection of material for the biopsy (Figure 6).



Figure 4. Incision and surgical flap.



Figure 5. Enucleation of the lesion.



Figure 6. A sterile absorbable gelatin sponge (Gelfoam) was used for hemostasis in the surgical cavity.

A sterile absorbable gelatin sponge (Gelfoam) was used for hemostasis in the surgical cavity (Figure 7) after performing curettage on the ostium of the maxillary sinus. The replacement and passive closure was obtained with 4–0 vicryl threads. After the suture, the pharyngeal "packing" was removed, the patient extubated, and the surgical procedure completed.



Figure 7. Histopathological lamina of the cystic lesion of mucous retention in the maxillary sinus.

The biopsy was sent for histopathological analysis (Figure 8). that has confirmed the result of a mucosal retention cyst in the maxillary sinus (Figure 9).

Histologically it was described like, the mucosal retention cyst that is composed of columnar epithelial lining of the cylindrical, ciliated pseudostratified type. The adjacent connective tissue is fibrous-elastic, called the mucosa-specific tunica. Amid the fibers lies the fundamental substance of the connective tissue matrix rich in carbohydrates and proteins.

Besides that, the polymorphonuclear cells, lymphocytes, plasma cells and macrophages are present in inflammatory states and blood vessels appear in small numbers. Another find was Scaly glandular elements of the sero-mucous gland type that were found dispersed. Mucous cells are located around the ostia of the ducts, while the serous ones are located in the bottom of the acini⁶.

The patient has been followed up over a period of two years and there were no complications such as paresthesia or any sign of recurrence of the lesion (Figure 10).

DISCUSSION

Paranasal sinuses are described in the literaure as air cavities that communicate with the nasal cavity



Figure 8. Material for the biopsy was sent for histopathological analysis.



Figure 9. Panoramic Radiography (2-year follow-up).



Figure 10. Intraoral vision after 2 years.

through channels and ostia^{1,10}. These sinuses are covered by a thin mucous membrane that adheres to the periosteum, and the ciliated epithelium helps to drain the secretions formed in the nasal cavity. The maxillary sinuses can present diverses shapes and sizes, depending on several factors such as facial type, age and number of teeth present. These sinuses tend to encroach (pneumatize) the spaces left by the teeth that are progressively extracted, becoming quite developed in edentulous patients¹¹.

Cysts are asymptomatic lesions of chronic inflammatory nature, which affect all races, both genders, and can occur at any age.

The mucous retention cyst (mucocele) of the maxillary sinus is a phenomenon resulting from the retention of mucous present in the mucous glands of the maxillary sinus lining epithelium, being detected as a radiographic finding in the dental examinations by image¹²⁻¹⁴.

These cysts, because they are chronic lesions, have slow and asymptomatic growth, and are detected most of the times by routine radiographic exames⁹; despite that most mucosal retention cysts are asymptomatic, when symptoms such as headaches, periorbital or facial pain, repeated infections of the paranasal sinuses, and/ or nasal obstruction are present, surgical treatment may be necessary¹⁵.

On a literature review, Kanagalingam et al.¹² observed that the patient presented pain in the left maxilla region and underwent panoramic radiography and computed tomography, and surgical treatment was performed. It is agreed that mucosal retention cysts in maxillary sinuses are benign injuries that result from obstruction of the duct from seromucous gland, which in turn causes mucus accumulation and cystic dilatation of the gland.

The incidence of mucosal retention cyst was reported to be around 29.4% (150 cases). The average age was 37.9 years (17-76), and the male-female ratio was 1.7:1. Twenty-five patients (16.6%) demonstrated bilateral cysts in the maxillary sinus, while there were 118 (78.6%) unilateral cases, 54 (45.8%) with mucosal retention cyst in the right maxillary sinus, and 64.2%) in the left maxillary sinus.

Some patients had cysts in more than one sinus, according to Schalek et al.¹³, (56.25%) were men and 28 (43.75%) were women, with a mean age of 40.4 years (range 18-70 years)¹⁸. In 34 patients, the cyst was located in the right maxillary sinus in 28 patients of the left maxillary sinus, and in two cases the cysts were bilateral¹⁹; predominance of the male gender was found to be the case with 2:1 predisposition (31.96% vs. 14.44%)²⁰.

The etiology of this pathology is controversial. It is believed that allergic processes may play a role in the formation of sinus mucoceles²¹, and the possible relationship with dental and periodontal problems, mainly through infections that penetrate the sinus giving origin to the mucous cyst. Moreover, many report that a history of trauma and dental infectious have been correlated with mucous retention cysts formation^{22,23}. There are reports of a patient who lost all dental elements of the left jaw due to automobile accident followed by an onset of sinus mucous cystic lesion⁸.

Treatment for the mucosal retention cysts with decompression and marsupialization were first suggested in 1892, yet at the time considering the definitive treatment for cysts the removal of the epithelium overlying the bone with the cyst. If possible suturing should be performed in order to maintain the opening of the cyst in decompression/marsupialization. The argument at that time was to open the cyst^{24,25}.

Marsupialization of large cystic lesions demonstrated a 47% decrease in size, resulting in a much simpler enucleation²⁶. Marsupialization allows a less invasive approach, with excellent results, avoiding exhaustive traumatic procedures²⁷.

Marsupialization is especially useful, because the conduits releive pressure within the cystic cavity^{28,29}, more than that the marsupialization lowering damage risk of the inferior alveolar neurovascular bundle in such cases because when the inferior alveolar canal is in a very critical position with a mandibular cyst, thus decreasing the cystic size before enucleation.

The procedure has a high success in decreasing cyst size prior to enucleation and is useful to avoid extensive surgery. Decompression may be performed through a small opening in the cyst and holding it open with a drain of some type^{30,31}.

It is believed that most maxillary sinus retention cysts regress spontaneously or do not show any significant change in long-term size⁹, than the treatment of sinus mucoceles is only the radiographic follow-up, through panoramic radiographs and only in extreme symptomatic cases is the excision of the lesion indicated²³.

Medical follow-up is indicated only in patients with recent allergic histories. As it has been observed that these cysts can remain stable in size for many years, yet they can also rupture and disappear altogether in a relatively short time without any surgical intervention. In addition, unlike odontogenic cysts, it is very difficult for expansion of the bony walls of the sinus antrum to occur even when the lesion completely occupies the maxillary sinus³⁰. Regarding cystic recurrences, they may be related to cyst characteristics at the cellular level or difficulty in complete surgical removal³². In the clinical case described in this article, the treatment chosen was surgical, because the lesion was symptomatic²³; the patient reported pain in the left jaw region and was unable to use her prosthesis, so she has opted for enucleation of the mucosal retention cyst. Marsupialization of the lesion is indicated when it presents some risk to the integrity of the patient or of adjacent structures^{30,31} in this case, there were no risk of damage to any structure.

CONCLUSION

The maxillary sinus mucosal retention cyst is a usually asymptomatic condition, with a higher prevalence of the male gender, restricted to the maxillary sinus and discovered at random in routine imaging exames. The above-described case is not a typical presentation of a maxillary sinus mucocele, mainly because there was an expansion of the maxillary tuberosity, pain complaint and the inability of the patient to fit her upper prosthesis. Thus, the clinical management indicated was surgical enucleation, followed by adequate postoperative followup for two years, without recurrence of the lesion.

REFERENCES

- 1. Neville BW. Patologia oral & Maxilofacial. Rio de Janeiro: Guanabara Koogan; 2016.
- 2. Gardner DG. Pseudocysts and retention cysts of the maxillary sinus. Oral Surg Oral Med Oral Pathol. 1984;58:561-7.
- Rege IC, Sousa TO, Leles CR, Mendonça EF. Occurrence of maxillary sinus abnormalities detected by cone beam CT in asymptomatic patients. BMC Oral Health. 2012;12:30.
- 4. Yeung AWK, Tanaka R, Khong PL, von Arx T, Bornstein MM. Frequency, location, and association with dental pathology of mucous retention cysts in the maxillary sinus. A radiographic study using cone beam computed tomography (CBCT). Clin Oral Investig. 2017. [Epub ahead of print]
- 5. Vogiatzi T, Kloukos D, Scarfe WC, Bornstein MM. Incidence of anatomical variations and disease of the maxillary sinuses as identified by cone beam computed tomography: a systematic review. Int J Oral Maxillofac Implants. 2014;29:1301-14.
- Moon IJ, Kim SW, Han DH, Shin JM, Rhee CS, Lee CH, et al. Mucosal cysts in the paranasal sinuses: long-term follow-up and clinical implications. Am J Rhinol Allergy. 2011;25:98-102.
- 7. Gothberg KA, Little JW, King DR, Bean LR. A clinical study of cysts arising from mucosa of the maxillary sinus. Oral Surg Oral Med Oral Pathol. 1976;41:52-8.
- 8. Wang JH, Jang YJ, Lee BJ. Natural course of retention cysts of the maxillary sinus: long-term follow-up results. Laryngos-cope. 2017;117:341-4.
- 9. Allard RH, van der Kwast WA, van der Waal I. Mucous antral cysts. Review of the literature and report of a radiographic survey. Oral Surg Oral Med Oral Pathol. 1981;51:2-9.
- Bulgarelli AF, Silva ABM, Paulino SM, Watanabe PCA, Pardini LC. Cisto de retenção do seio maxilar - considerações para o clínico geral. Rev Assoc Paul Cir Dent. 2002;56:178-81.

- 11. Sawada K, Yanagi K, Sakurai Y, Imai T. Three cases of sphenoid sinus retention cyst of the headache. Otolaryngol Head Neck Surg (Tokyo). 2005;48:174-81.
- 12. Kanagalingam J, Bhatia K, Georgalas C, Fokkens W, Miszkiel K, Lund VJ. Maxillary mucosal cyst is not a manifestation of rhinosinusitis: results of a prospective three-dimensional CT study of ophthalmic patients. Laryngoscope. 2009;119:8-12.
- Schalek P, Otruba G, Hornáčková Z, Hahn A. Mucosal maxillary cysts: long-term subjective outcomes after surgical treatment. Eur Arch Otorhinolaryngol. 2013;270:2263-6.
- González Ortin M. [Salivary cysts of the maxillary sinus. One case report. An Otorrinolaringol Ibero Am]. 1996;23:589-95. Spanish.
- 15. Ruprecht A, Batniji S, el-Neweihi E. Mucous retention cyst of the maxillary sinus. Oral Surg Oral Med Oral Pathol. 1986;62:728-31.
- 16. Aydın E, Yerli H, Tanrıkulu S, Hizal E. Mucosal cysts of the maxillary sinus in solid organ transplant population: computerised tomography follow-up results. Balkan Med J. 2013;30:305-8.
- 17. Myall RW, Eastep PB, Silver JG. Mucous retention cysts of the maxillary antrum. J Am Dent Assoc. 1974;89:1338-42.
- Casamassimo PS, Lilly GE. Mucosal cysts of the maxillary sinus: A clinical and radiographic study. Oral Surg Oral Med Oral Pathol. 1980;50:282-6.
- 19. Halstead CL. Mucosal cysts of the maxillary sinus: report of 75 cases. J Am Dent Assoc. 1973;87:1435-41.
- 20. Partsch C. Über Kieferzysten. Dtsch Monatszeitschrift Zahnheilkd. 1892;10:271-304. German.
- 21. Partsch C. Behandlung Kieferzysten. Dtsch Monatszeitschrift für Zahnmed. 1910;28:252-60.
- 22. Clark P, Marker P, Bastian HL, Krogdahl A. Expression of p53, Ki-67, and EGFR in odontogenic keratocysts before and after decompression. J Pathol Oral Med. 2006;35:568-72.

- 23. Borgonovo AE, Di Lascia S, Grossi L, Maiorana C. Two-stage treatment protocol of keratocystic odontogenic tumour in young patients with Gorlin-Goltz syndrome: marsupialization and later enucleation with peripheral ostectomy. A 5-year-follow-up experience. Int J Pediatr Otorhinolaryngol. 2011;75:1565-71.
- 24. Bland PS, Shiloah J, Rosebush MS. Odontogenic keratocyst: a case report and review of an old lesion with new classification. J Tenn Dent Assoc. 2012;92:33-6.
- 25. Pogrel MA. Decompression and marsupialization as a treatment for the odontogenic keratocyst. Maxillofac Oral Surg Clin North Am. 2003;15:415-27.
- 26. Eyre J, Zakrzewska JM. The conservative management of large odontogenic keratocysts. Br J Oral Maxillofac Surg. 1985;23:195-203.
- 27. Nakamura N, Mitsuyasu T, Mitsuyasu Y, Taketomi T, Higuchi Y, Ohishi M. Marsupialization for odontogenic keratocysts: long-term follow-up analysis of the effects and changes in growth characteristics. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2002;94:543-53.
- Costa C, Costa CAC, Ferreira ETT, Oliveira JX. Cisto mucoso do seio maxilar: apresentação de dois casos. Rev Inst Ciênc Saúde. 1992;10:63-5.
- 29. Finkelstein MW, Hellstein JW, Lake KS, Vincent SD. Keratocystic odontogenic tumor: a retrospective analysis of genetic, immunohistochemical and therapeutic features. Proposal of a multicenter clinical survey tool. Oral Surg Oral Med Oral Pathol Oral Radiol. 2013;116:75-83.
- 30. Politano GT, Manetta IP, Araújo VS, Aguiar JMRP, Brianez N, Echeverria S, et al. Cisto radicular- relato de caso clínico. ConScientiae Saúde. 2009;8:129-32.
- Bermejo A, Aguirre JM, López P, Saez MR. Superficial mucocele: report of 4 cases. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 1999;88:469-72.
- 32. Schalek P., Otruba G., Hornáčková Z., Hahn A. Mucosal maxillary cysts: long-term subjective outcomes after surgical treatment. EurArchOtorhinolaryngol, 2013. p. 2263-6.