CBCT FEATURES OF UNUSUAL MUCOCELE OF MAXILLA

ABSTRACT

Mucoceles are a common injury of the oral mucosa, contained in the group of pathologies of the salivary glands. Besides this type of injury, it is important that health professionals become aware of the occurrence of lesions in other areas who also receive the name of mucoceles. These pathologies are benign paranasal sinus lesions, cyst-like structures, lined with a secretory respiratory mucosa of pseudostratified columnar epithelium. Despite its benign appearance, mucoceles exhibit aggressive behavior and may expand and cause erosion in surrounding tissues, leading to severe bone destruction. Here, we report a case of a unusual mucocele in midline of the maxilla with invasion of paranasal sinuses after orthognathic surgery, mainly focused on tomographic features.

KEYWORDS: Mucocele; Computed tomography; Cone-Beam computed tomography; Maxillofacial surgery.

RESUMO

Mucoceles são lesões comuns da mucosa oral, contidas no grupo de patologias das glândulas salivares. Além deste tipo de lesão em tecido glandular, é importante que os profissionais de saúde tenham-se conscientes da ocorrência de lesões em outras áreas que também recebem o nome de mucoceles. Estas patologias são lesões benignas nos seios paranasais, com aspecto de estruturas císticas, revestidas com uma mucosa respiratória secretora de epitélio colunar pseudoestratificado. Apesar de sua aparência benigna, mucoceles apresentam comportamento agressivo e podem se expandir e causar erosão nos tecidos circundantes, levando à destruição óssea grave. Relatamos o caso de uma mucocele incomum na linha média da maxila com invasão de seios paranasais após cirurgia ortognática, com foco principalmente em características tomográficas.

PALAVRAS-CHAVE: Mucocele; Tomografia Computadorizada; Tomografia Computadorizada por Feixe Cônico; Cirurgia Maxilo-Facial.

1 INTRODUCTION

Mucoceles are a common injury of the oral mucosa, contained in the group of pathologies of the salivary glands. They result from the rupture of a duct with consequent spillage of mucin in the soft tissues, and are often associated...
with minor trauma. Besides this type of injury, it is important that health professionals become aware of the occurrence of lesions in other areas who also receive the name of mucoceles. These pathologies are benign paranasal sinus lesions, cyst-like structures, lined with a secretory respiratory mucosa of pseudostratified columnar epithelium. They grow slowly, are locally aggressive, and may be the result of accumulation and retention of mucous secretions in the sinus caused by the loss of draining properties of the mucous epithelium of the sinus. The majority occur in the frontal sinus (60 per cent) followed by 30 per cent in the ethmoid sinuses. Only 10 per cent are found in the maxillary sinuses and rarely are located in the sphenoid sinuses. Despite its benign appearance, mucoceles exhibit aggressive behavior and may expand and cause erosion in the bone. Among complications, amaurosis is the most feared, but in a fortunately way, it occurs with low frequency, and its occurrence with maxillary sinus mucoceles is rare. Although the diagnosis may be suggested by the clinical presentation, past medical history, or physical examination, Computed Tomography (CT) is necessary to accurately analyze the regional anatomy and the extent of the lesion. Recently cone-beam computed tomography (CBCT) systems have become available, which are specifically designed to image of hard tissues in the maxillofacial region.

The purpose of this paper is to describe a case of a giant mucocele in midline of the maxilla with invasion of paranasal sinuses after orthognathic surgery, focused on the tomographic features.

2 CASE REPORT

A 49-year-old caucasian patient was referred to maxillofacial surgeon complaining of respiratory distress and painful increase of volume in the anterior region of maxilla. The patient had undergone an orthognathic surgery 8 years earlier. A CBCT was performed, and the tomographic analysis depicted an extensive destruction in midline of the maxilla associated with hyperdensity of soft tissue in the region of nasal cavity and nasopharynx, with invasion of paranasal and maxillary sinuses. Four metal plates linked to previous orthognathic surgery was seen at the facial mid third. There was large bone resorption involving the region of the anterior nasal spine, some areas of the hard palate and medial walls of the maxillary sinus. A large destruction of the nasal septum and dislocation of the osteomeatal complex and infundibulum was seen as well, with loss of alveolar bone in the buccal plate of anterior tooth of the maxilla. The lesion measures are about 34.25 mm in latero-lateral direction, 25.50 mm in the superior-inferior direction, and 28.75 mm anteroposteriorly. Under general anesthesia, surgical excision of the lesion was performed and the histopathological slides diagnosis confirmed the hypothesis of traumatic mucocele of the paranasal and maxillary sinuses.

3 DISCUSSION

A mucocele is an epithelial lined mucus-containing sac completely filing a paranasal sinus and capable of expansion by virtue of a dynamic process of bone resorption and new bone formation.
on. They can cause facial pain, headache, nasal obstruction, diplopia, decreased visual acuity, displacement of the eyeball, facial swelling and meningitis, depending on the anatomic area involved. In cases of maxillary mucoceles, the clinical symptoms are due to sinus expansion into the nose, mouth and orbit resulting in upward displacement of the eye, proptosis and swelling of the cheek. As depicted in our case, there was a huge destruction, despite the benign character of this, with the involvement of the paranasal sinus and destruction of the anterior nasal spine, areas of the hard palate and medial walls of the maxillary sinus, as can be seen in Figure 1.

**Figure 1** - Tomographic features of unusual mucocele. (A) Axial, (B) sagittal and (C) coronal views depicting a destruction in the anterior maxilla with invasion of paranasal sinuses. A huge soft tissue hyperdensity is seen at the midline, involving anterior nasal spine, hard palate, extending laterally to paranasal sinus. It is possible to see the huge size of the lesion, with involvement of both maxillary sinus and nasal septum. Destruction of middle turbinates is also seen, with superior displacement of osteomeatal complex and infundibulum.

The aetiology of mucoceles is not clear. Fifty per cent of these patients have a history of prior infection, 25 per cent of trauma and 10 per cent a prior allergic history. They may result from infection, trauma including previous surgery, fractures and bullet wounds or tumours. Reports of development of ciliated cysts arising in non-conventional locations, and its relationship with orthognathic surgery has been seen in the literature. Transplanted respiratory epithelium can be attached to sinonasal material, proliferating in favorable healing sites, leading to uncommon development of cysts. In our case, in the 3D images of CBCT (Figure 2), can be observed the presence of surgical plates used for orthognathic surgeries, showing the occurrence of a surgery prior to the appearance of the lesion of mucocele. During orthognathic surgery, after the achievement of osteotomy, may be rupture and migration of glandular cells of the paranasal sinuses that can cause the formation of the lesion. Given the features of the lesion...
(well demarcated margins, clear separation of bone lying, homogeneus stroma), despite the local aggressive behaviour in this particular case, and the presence of endodontically treated tooth (central incisors) was seen, making the hypothesis of radicular cyst as first tomographic diagnosis and probable cause of lesion. Another tomographic hypothesis that could be rised was the nasopalatine duct cyst, but this lesion is somewhat rare and related only about to 1% of the maxillary cysts. Only after histopathological evaluation, the final diagnosis of mucocele was emitted. After confrontation with clinical history, surgical aspect during the removal and slow growth of the lesion, a rupture/entrapment of respiratory epithelium during orthognatic surgery was credited as etiological cause of the mucocele.

Figure 2 - CBCT 3D images (volume rendering): (A) and (B) depicting the presence of surgical plates used in previous orthognatic surgery. An total of four metal plates is seen at the anterior walls of maxillary sinus and alveolar process of maxilla. Destruction of nasal septum and facial mid third is evidentiated, with extension to superior region of nasal fossa. Aditionally, the loss of buccal wall in anterior teeth is seen, and the amount of bone destruction caused by compression of the mucocele is demonstrated.

Figure 3 - Aspect of mucocele during the surgical excision (A). The image shows the bone destruction and the final aspect after the removal of the lesion. The soft tissue aspect of the lesion is remarkable, with the clear depiction of a capsule. (B) Cavity and bone aspect after complete removal of the lesion.
Maxillary sinus involvement must be carefully assessed because orbital damage and the spreading of associated infections could lead to local and systemic compromise to the patient. Even in cases of benign pathologies, suspicion of maxillary sinus involvement means that radiographic evaluation is mandatory. Mucoceles of maxillofacial complex are well-known for its benign histological nature, without specific symptoms. Otherwise, in some instances, aggressive behaviour could be seen and aggression to neighboring structures as orbit and brain could lead to serious complications and revealed by ophtalmic or intracranial complications. Diagnostic of such lesions is based on imaging, being CT and MRI the gold standard\textsuperscript{11}.

The anatomy of the paranasal sinuses is complex and shows great variation. When there is the formation of a mucocele, the bone erosion and the consequent destruction of anatomical parameters, makes it difficult to understand the local anatomy. Consequently, it is necessary to perform a radiological assessment before planning surgery. Mucoceles can be easily diagnosed by computed tomography (CT)\textsuperscript{12}. CBCT is one of the main resources for dentistry nowadays. With spreading and increasing of volumetric imaging, is mandatory to all practitioners to be familiar with all the structures contained within a scanned area, as well as with the anatomy of the nasal cavity and common anatomic variations and pathologic entities\textsuperscript{13}.

4 CONCLUSIONS

In our case, cone beam computed tomography proved to be an excellent diagnostic tool, which due to its access easier to the dentist can be consolidated as one method of choice for diagnosis of several diseases of maxillofacial complex. Tomographic analysis revealed a large lesion, extending from the anterior maxilla to the posterior region of the nasal cavity, with resorption of nasal septum and involvement of the medial walls of right and left maxillary sinuses. With the presence of metal plates, the history of previous ortognathic surgery, even with the presence of endodontic treated teeth, mucoceles are an option to be considered in the tomographic diagnostic of lesions with such appearance. Despite its benign behaviour, serious complications could occur, with high morbidity and implications for the patients. In this regard is mandatory that the oral and maxillofacial radiologist keep in mind such conditions, avoiding delay in the diagnostic procedures and surgical treatment. Hence, in this context, the CBCT proved to be crucial for planning optimal surgical procedure, elucidating the diagnosis and allowing a full delineation of the lesion.
REFERENCES


