INTRODUCTION

Mucoceles are slow growing benign lesions, filled with mucus and lined with epithelium. They are commonly found in the frontal sinus and are relatively rare in the maxillary sinus (< 1%) [1]. They gradually expand via a dynamic process of bone resorption and resultant new bone formation [2]. Most cases of maxillary mucoceles are the result of infection, neoplasm, allergy, trauma or surgery. The most common surgery associated with maxillary mucoceles is the Caldwell-Luc procedure. Symptoms may be delayed for several years after the surgery.

Maxillary mucoceles occurring after a maxillary osteotomy are relatively rare. We report a case of a 37-year-old woman with giant maxillary mucocele that occurred 17 years after a Le Fort I advancement surgery with resultant extensive destruction of the maxilla.

Keywords: mucocele; Le Fort osteotomy; postoperative complications; maxillary sinus

CASE DESCRIPTION

We present the case of a 37-year-old woman who suffered from severe electrical burns to the mouth at the age of 2 resulting in maxillary hypoplasia and upper lip contractures.

The patient underwent numerous reconstructive surgeries including a Le Fort I advancement procedure 17 years earlier followed by an Abbe-Estlander flap 10 months later for upper lip reconstruction. The patient was a nonsmoker and was not on any medication.

The patient recently presented to our department for a one month history of tooth mobility. She was otherwise healthy and had no pain, fever, nasal discharge or epistaxis. On physical examination she only had mobility of the upper left premolars and molars.

A panoramic radiograph revealed the presence of a cystic bony process in the maxillary alveolar ridge crossing the midline with destruction of the upper left premolars and molars roots. A computerized tomography (CT) confirmed the presence of a giant cystic mass occupying the maxillary alveolar ridge with well-defined sclerotic borders and bone remodeling (Figure 1). A 3D reconstruction of the CT scan revealed a severe erosion of the anterior maxillary wall with the remaining alveolar ridge only held in place by the previously placed titanium plates and screws (Figure 2).

Surgery was undertaken to remove the lesion. A maxillary gingival incision was performed. Directly upon incision 20 ml of a thick greenish fluid was expressed. A giant mucocele occupying the maxilla and extending to the right maxillary sinus was found. The anterior wall of the maxilla was extensively eroded.
and the remaining bone was only held in place by the previously placed titanium plates and screws. The diseased mucosa was removed and sent to pathology, which confirmed the diagnosis of maxillary mucocele.

The titanium plates and screws from previous surgery were considered important for the support of the maxillary bony pillars. They were thus left in place. No postoperative complications were noted and the patient was discharged two days later.

DISCUSSION

Mucoceles are chronic mucus-filled expanding sacs that occur with similar frequency in both sexes [5]. They originate from infection, neoplasm, allergy, trauma or surgery [6]. The most common surgery associated with maxillary mucoceles is the Caldwell-Luc procedure [3]. Symptoms may be delayed for several years after the surgery. Thio et al. reported a case of maxillary mucocele occurring 15 years after a maxillary advancement procedure [4]. Due to increasing pressure from accumulation of mucus, the mucocele expands in the least resistant path and erode through bone protruding to adjacent structures [7]. Maxillary extension into the alveolar ridge was demonstrated in our patient.

Microscopically, the maxillary mucocele is a true cyst, lined by pseudostratified columnar ciliated epithelium with mucous cells [8]. The main differential diagnoses of maxillary mucoceles include retention cysts, surgical ciliated cysts and antral pseudocysts [9]. Retention cysts are usually small and result from obstruction of the ducts of the seromucinous gland of the sinus. Not unlike mucoceles of the maxillary sinus, surgical ciliated cysts are secondary to trauma, surgery, or damage to the maxillary sinus leading to entrapment of the sinus mucosa. In case of large lesions, the differential diagnosis from a giant mucocele of the maxilla may not be possible. Antral pseudocysts are the most common cysts of the maxillary sinus. They are dome-shaped collections of inflammatory exudate caused by infections or allergic sinusitis of the maxillary sinus [9].

Many theories are suggested to explain the pathogenesis of iatrogenic maxillary mucoceles.

One claims that tissue reaction to foreign material may cause enough inflammation to impair drainage of maxillary sinus and result in mucocele formation many years later [4]. Another theory suggests that entrapment of mucosa is the origin of maxillary mucoceles occurring as a complication of Caldwell-Luc surgery [10].

In our case the maxillary hypoplasia which resulted from severe burns was corrected by a Le Fort I advancement procedure. The fragile scarred tissue at the time of the osteotomy may have been entrapped in the line of fracture resulting in mucocele formation. The sac then slowly expanded into the alveolar ridge over many years.

CONCLUSION

Maxillary mucoceles occurring after maxillary advancement have only been reported once. We demonstrated that these sacs can expand into the maxillary alveolar ridge resulting in its near total destruction. With time, mucoceles may be very aggressive locally and one must be aware of this rare and late complication of such surgeries.
REFERENCES


