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Case report

Pathology

Differential diagnosis of antral pseudocyst, surgical ciliated cyst, and mucocele of the maxillary sinus

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Abstract

Introduction

Pathologic alterations of the maxillary sinus, including antral pseudocysts, surgical ciliated cysts, and mucoceles of the maxillary sinus, may pose a diagnostic challenge. These conditions are often misdiagnosed, compromising the subsequent clinical and surgical approach. Although these lesions may present with similar clinical and imaginological features, their differential diagnosis is important for correct treatment planning owing to their distinct biological behaviour. The purpose of this paper is to report one case of each of these conditions, together with a discussion of their differential diagnosis and treatment plan.

Case report

In case 1, patient was a candidate for orthognathic surgery. A pre-operative cone beam computed tomographic showed a hyperdense domed-shaped image above the first and second molars, and with the provisional diagnosis of antral pseudocyst, no specific treatment carried out.

Case 2, patient complained of light intermittent discomfort on the left side of her maxilla for over a year and had a history of orthognathic surgery involving both jaws performed 19 years ago. Radiographic image showed a well-circumscribed hypodense area in the maxilla above the left first molar and an excisional biopsy was performed, which ultimately confirmed the diagnosis of surgical ciliated cyst.

In Case 3, patient presented with a history of a painful swelling on the left side of his face, which started 6 months earlier. Magnetic Resonance Image showed a hyperdense lesion, occupying the left maxillary sinus and an the surgical excision of the lesion, confirmed the mucocele of the maxillary sinus diagnosis.

Conclusion

Owing to their similar radiographic features and asymptomatic presentations, AP, SCC, and MMS can be misdiagnosed. Clinical signs and symptoms, patient history, and adequate radiographic evaluation are necessary to make accurate decisions regarding their initial diagnosis and subsequent treatment plans.

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Introduction

Different pathologic conditions can affect the maxillary sinuses and are frequently confused and misinterpreted. These lesions include the antral pseudocyst (AP), surgical ciliated cyst (SCC), and mucocele of the maxillary sinus (MMS). The variety of terms used for each of these lesions is itself suggestive of the fact that these entities are poorly understood[1,2].

In several cases, because of an improper differential diagnosis, the incorrect treatment plan is selected for that specific lesion.

In their early stages especially, AP, SCC, and MMS may be asymptomatic and present with similar radiographic features, which can cause misdiagnosis[3]. In some situations, a biopsy is necessary to confirm diagnosis, but in others, diagnosis relies solely on clinical and radiographic grounds[1,3]. Therefore, oral and maxillofacial surgeons, radiologists, and pathologists have to bear in mind features involved in the differential diagnosis of these cystic lesions for their correct treatment. The purpose of this paper is to report one case of each of these conditions and to discuss appropriate differential diagnosis and treatment.

Case report

Case 1: Antral Pseudocyst (AP)

A 19-year-old female patient presented to the Service of Oral and Maxillofacial Surgery of Clinics Hospital of the Universidade Federal de Minas Gerais (HC-UFMG) in Belo Horizonte, Brazil, as a candidate for orthognathic surgery. Her chief complaints were chewing dysfunction and dissatisfaction with her facial aesthetics.

The patient was medically fit and denied a history of any local or systemic disease. Upon clinical evaluation, maxillary retrognathism was diagnosed, but no extra-oral pathological alteration was observed. Intra-oral examination showed good periodontal health and a Class III malocclusion. No mucosal lesions were observed and the patient did not complain of any such symptoms. A cone beam computed tomographic (CBCT) scan was requested to complement the planning process for orthognathic surgery.

A hyperdense domed-shaped image was noted above the first and second molars, filling part of the right maxillary sinus floor (Figure 1). Since the patient had no signs or symptoms of maxillary sinus involvement, the provisional diagnosis was deemed AP, and it was proposed that no specific treatment for this be carried out. The previously planned orthognathic surgery was conducted without transoperative or postoperative complication. At the 10-month follow-up examination of the patient during the postoperative

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period, the patient had no clinical signs or symptoms suggestive of maxillary sinus pathology.

Case 2: Surgical Ciliated Cyst of the Maxilla (SCC)

A 42-year-old female patient presented to the Service of Oral and Maxillofacial Surgery of HC-UFMG in Belo Horizonte, Brazil, complaining of light intermittent discomfort on the left side of her maxilla for over a year. The patient had a history of orthognathic surgery involving both jaws performed 19 years ago.

At the time of surgery, the maxillary segments were stabilised with steel wires. She denied a history of systemic disease and drug or tobacco use. No extra-oral alterations were observed.

Intra-oral examination revealed good periodontal health and no missing teeth or mucosal lesions. A CBCT scan was requested and showed a well-circumscribed hypodense area in the maxilla above the left first molar; the hypodense area was round in shape and communicating with the floor of the maxillary sinus at the superior part of the lesion (Figure 2).

Electric pulp tests showed that the left posterior maxillary teeth were vital. The provisional diagnosis was of SCC, and so an excisional biopsy was performed. A mucoperiosteal flap was lifted to enable a Caldwell–Luc approach to the maxillary sinus. A 1.5-cm-wide window of bone was removed from the maxilla above the right first molar apices.

This procedure exposed a fluid-filled membranous sac from which a clear straw-coloured fluid was then aspirated. Owing to the thin and friable cyst capsule, the cyst wall was perforated during surgery (Figure 3).

The entire cyst was then enucleated for histopathological analysis, which ultimately confirmed the diagnosis of SCC (Figure 4).

In the postoperative period, the patient presented without complications or complaints of the previous symptoms. A postoperative CBCT showed new bone formation in the region previously occupied by the cyst. No clinical or radiographic signs or symptoms were noted at the 14-month follow-up examination.

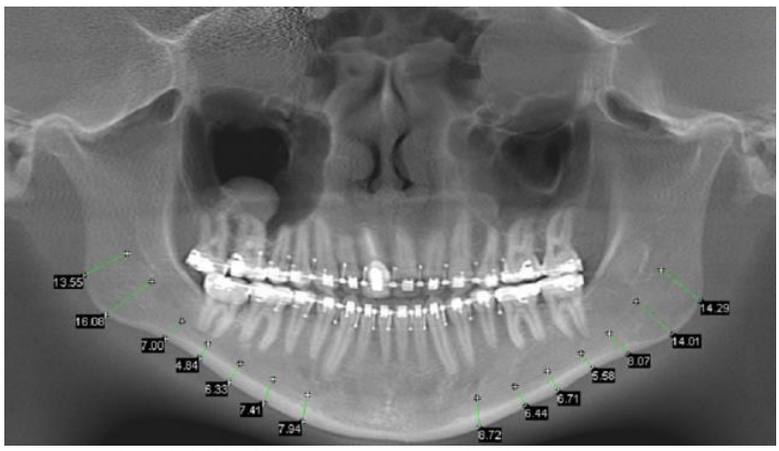

Figure 1: A well-defined hyperdense unilocular dome-shaped image above the right maxillary molars. Typical image of antral pseudocyst.

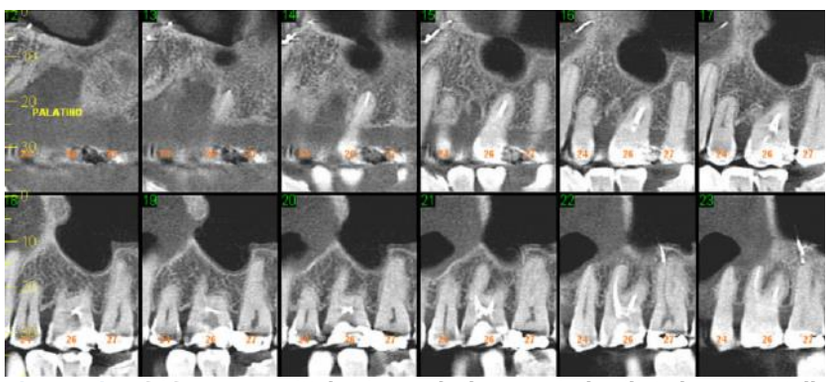

Figure 2: CBCT parasagittal view of the surgical ciliated cyst. Well-circumscribed round-shaped unilocular hypodense area, observed inside the maxillary bone, communicating with the maxillary sinus, in a patient with a history of orthognathic surgery.

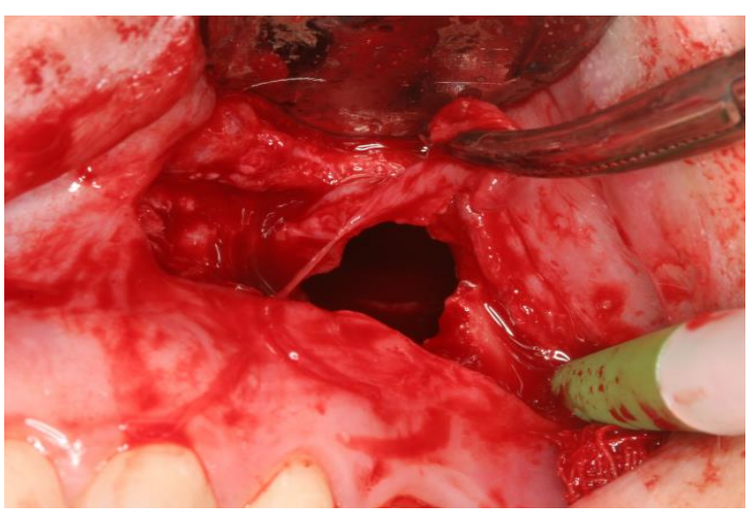

Figure 3: Caldwell–Luc approach for excisional biopsy of surgical ciliated cyst. The thin and friable capsule may disrupt during surgery.

Case 3: Mucocele of the Maxillary Sinus (MMS)

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A 59-year-old male patient presented to the Service of Oral and Maxillofacial Surgery of HC-UFMG in Belo Horizonte, Brazil, complaining of a painful swelling on the left side of his face, which started about 6 months earlier.

The patient denied history of any other disease or of drug or tobacco use. Upon extra-oral clinical examination, a discreet swelling of the left side of his face was noted. Intra-oral examination revealed partially edentulous maxillary and mandibular arches, as well as a mild swelling with shortening of the vestibule in the posterior region of the left side of the maxilla, which was painful on palpation. As the patient presented to our Service with a magnetic resonance image (MRI) of the head and neck region, no further investigations were requested.

The MRI showed a hyperdense lesion, with homogeneous borders, occupying the left maxillary sinus. The images also suggested that the lesion was highly intensive, suggesting bony expansion, erosion, and invasion into the nasal cavity (Figure 5).

An incisional biopsy was performed under local anaesthesia. Histopathological examination of the cyst showed a fibrous capsule lined with pseudostratified columnar epithelium, which transitioned to non-keratinised squamous epithelium.

The diagnosis was MMS. The patient was placed under general anaesthesia, and a mucoperiosteal flap was raised to access the maxillary sinus. An osteotomy, measuring about 2.5 cm in width of the anterior wall of the maxilla was performed to approach the lesion. Despite the friable cyst capsule, the lesion was removed completely (Figure 6).

Recovery was uneventful, with resolution of the deleterious signs and symptoms. The final histopathological evaluation confirmed the initial diagnosis of MMS (Figure 7). After a follow-up of 10 months, no sign of recurrence was observed.

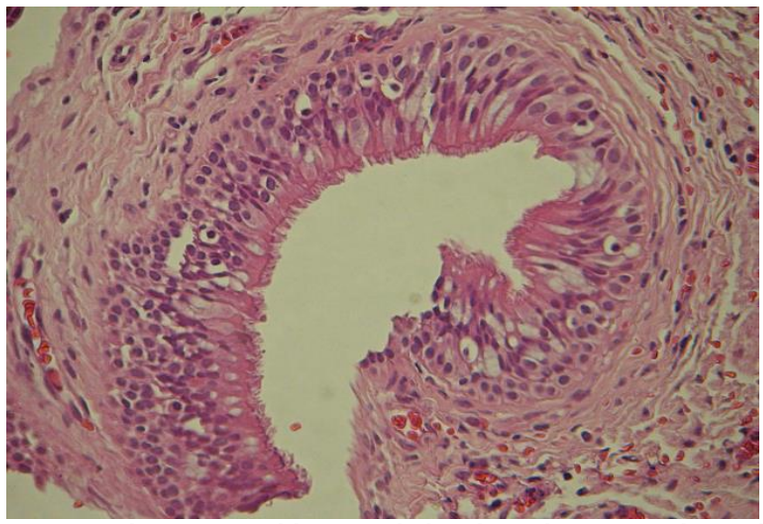

Figure 4: Histologic picture of the surgical ciliated cyst showing pseudostratified columnar ciliated epithelium containing mucous cells.

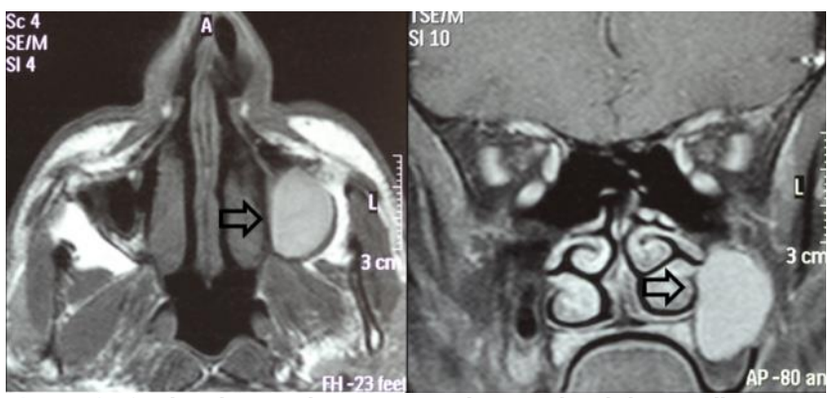

Figure 5: Axial and coronal MRI views of mucocele of the maxillary sinus showing a homogeneous and well-circumscribed lesion, with a high signal image. The black arrow points to initial invasion of the nasal cavity.

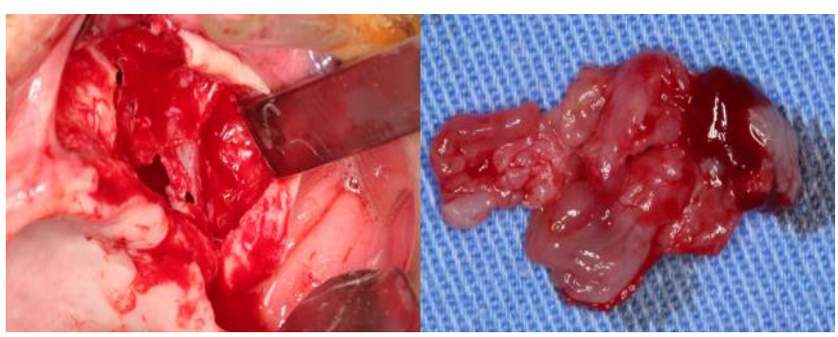

Figure 6: Mucoperiosteal flap and creation of a window to access the maxillary sinus for complete removal of the lesion.

Discussion

Antral Pseudocyst (AP)

The AP is the most common cyst or pseudocyst of the maxillary sinus. Some authors have noted a seasonal variation in its prevalence, claiming it to be more common in the winter because of infection of the upper airways[4].

The AP is a collection of inflammatory serum exudate that may have been caused by one of the following

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conditions: periapical or periodontal odontogenic infection, infection of the sinus or allergic sinusitis[5].

The accumulation of this inflammatory exudate occurs beneath the periosteum, forcing the sinusal lining away from the bone. Since APs are usually asymptomatic, they are frequently detected upon routine radiographic examination. The radiographic presentation of the AP is of a solitary dome-shaped, radiopaque mass situated in the floor of the sinus.

Microscopic examination of the AP reveals sinus mucosa displaced by cystic fluid. The mucosa is covered by pseudostratified columnar ciliated epithelium resting on a superficial layer of oedematous loose connective tissue, with chronic inflammation of variable intensity[1]. As the AP does not have specific or characteristic histopathological features, its diagnosis is not possible without appropriate clinical and radiographic information[1].

Surgical Ciliated Cyst of the Maxilla (SCC)

First described by Gregory and Shafer[6], the SCC is reported to be most common in Japan. The SCC does not affect the entire sinus, at least initially. This cyst occurs secondary to surgery, trauma, or damage to the maxillary sinus, all of which may result in the formation of scar tissue and resultant entrapment of the sinus mucosa[3]. Therefore, the lining of the cyst originates from the maxillary sinus mucosa itself.

The SCC is often asymptomatic, especially in early stages. Because of its potential to be locally aggressive, some patients may complain of swelling, pain, or discomfort in the maxillary region, as the cyst progresses. A comprehensive patient history helps in guiding the initial diagnosis. A history of a previous Caldwell–Luc procedure, or other sinus surgery is frequently reported[7].

Radiographically, the SCC appears as a round and well-circumscribed radiolucent or fairly radiopaque lesion[3].

As the cyst progresses, perforation, and expansion into the maxillary sinus is seen. In case of large lesions, the differential diagnosis from a MMS may not be possible. CBCT or MRI images may help elucidate the cyst boundaries and its relation with surrounding structures[8].

Microscopically, the SCC is a true cyst, lined by pseudostratified columnar ciliated epithelium with mucous cells, though squamous epithelium may be observed in some cases[3].

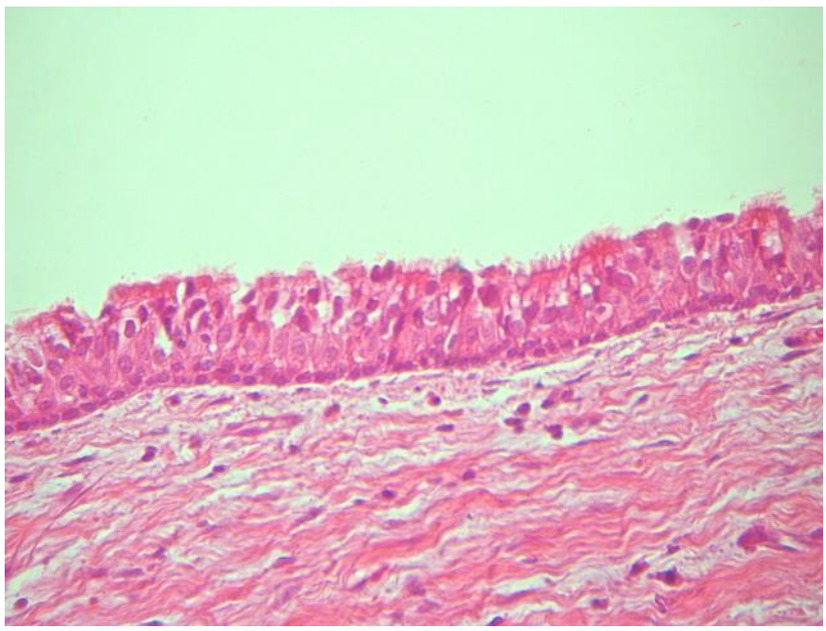

Figure 7: Histologic picture of mucocele of the maxillary sinus showing pseudostratified columnar ciliated epithelium.

Mucocele of the maxillary sinus (MMS)

Poor drainage of the sinus, resulting from any condition that obstructs the ostium, such as inflammatory processes, allergic reactions, or malignant diseases, may result in the formation of a MMS[2,3].

Although mucoceles are relatively frequent in the paranasal sinuses, especially the frontal and ethmoidal sinuses, in the maxillary sinus they are rare, accounting for less than 10% of paranasal sinus[9].

The clinical signs and symptoms of MMS depend mostly on its development stage. In its early stages, patients may be asymptomatic or present with complaints of headache, nasal congestion, swelling, or slight intermittent pain. With further development of the cystic lesion, facial swelling, nasal discharge, and periorbital or dental pain may occur, because of pressure from the cyst. In cases in which the mucocele invades the orbital floor, it can cause ocular displacement, nerve compression, and proptosis[10,11]. Infection can even convert a mucocele in a pyocoele[12].

On conventional panoramic radiographs, MMS presents as a rounded radiopaque mass, involving the sinus floor or entire maxillary sinus. Since MMS is usually invasive, bone erosion with medial extension into the nasal cavity is frequently seen. CBCT or MRI may be helpful to delimitate the lesion and evaluate the possible damaged surrounded structures, which will also help ascertain the proper surgical approach. The histopathologic features of MMS are similar to SCC of the maxilla.

Differential Diagnosis

The main differential diagnoses of AP include sinus polyps, retention cysts, and mucoceles. Except when it develops in the floor of a sinus, sinus polyps are often multiple and pendulous in

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appearance because of the effect of gravity; on the other hand, AP is usually solitary or bilateral[1]. Additionally, while the sinus mucosa adjacent to sinus polyps is thickened by oedema, with formation of fluid-filled spaces, and the polyps are generally irregular shaped and pendulous, the fluid in AP accumulates beneath the sinus mucosa and periosteum and forms a characteristic dome-shape[1].

Retention cysts are formed because blockage of the ducts of the seromucinous gland of the sinus can cause dilatation of the duct[1]. These cysts are usually small and may not even be evident clinically or radiographically. However, when they are large, they may have the same appearance as AP and therefore differential diagnosis is not possible on a purely radiographic basis. Finally, while retention cysts may be multiple and located next to the sinus ostium, pseudocysts are usually solitary and affect the floor of the sinus[1].

In Case 1, no cortical erosion or bone invasion by the AP was observed. As neither biopsy nor surgical procedures are necessary to diagnose AP, follow-up of the patient is recommended. The differential diagnosis of SCC depends on its developmental phase. If no association to the maxillary sinus is identified, the main diagnosis is of one of the several odontogenic benign lesions of the maxilla. In the initial stages, SCC may mimic AP, polyps of the sinus, radicular cysts or residual cysts of the maxilla[13].

In cases in which perforation and expansion of the maxillary sinus are detected; however, other hypotheses, such as salivary gland tumours, odontogenic tumours, cysts, and malignant conditions should be considered.

In Case 2, the initial provisional diagnosis of SCC was made on the basis of the history of orthognathic surgery 19 years ago, after which the sinus mucosa that was entrapped in the maxilla during healing of the Le Fort I osteotomy developed the SCC.
Like SCC, the differential diagnosis of MMS depends on its developmental stage at the time of evaluation. If no bone erosion is found, MMS must be differentiated from the most common conditions that affect the maxillary sinus, such as AP, chronic sinusitis, and sinus polyps. Other benign lesions that should also be included in the differential diagnosis are odontogenic and salivary gland tumours[11,14].

At later stages of development, MMS destroys bone. If this is the case, malignant conditions must also be considered in the differential diagnosis; these include adenoid cystic carcinoma, squamous cell carcinoma, undifferentiated carcinoma, plasmocytoma, and lymphoma[15]. Some authors also claim that MMS in infant patients is associated with cystic fibrosis[16,17].

Chindasombatjaroen et al.[8] suggest the use of MRIs to better identify cystic boundaries. Risk factors for recurrence include multiple mucoceles and extension outside the sinus wall[17].

In Case 3, the patient was referred shortly after the initial symptoms. MRI showed initial invasion into the nasal cavity and erosion of the maxillary anterior wall. Although MRIs delineate the expansion clearly, CT or CBCT alone are adequate examinations for diagnosis and surgical treatment plan.

Treatment

On the whole, APs are usually inoffensive, self-limiting, and require no specific treatment[18]. However, periapical or periodontal inflammatory conditions that are commonly associated with APs must be investigated and treated appropriately[1]. With time, APs tend to decrease or disappear on their own[19].

In the present case, for example, 10 months after the initial evaluation, no radiographic changes were observed and the lesion seemed to have regressed completely.

However, because of their invasive nature, MMS and SCC, must be surgically removed[3,9,14,20]. Surgical access depends on the size of the lesion and the structures involved by it. In cases in which the SCC is separated from the maxillary sinus, a bony window on the maxillary alveolar crest is necessary for surgical removal of the cyst.

In instances where the cyst communicates with, or is inside the maxillary sinus, a Caldwell–Luc approach is necessary. Recurrence is possible if the cyst is incompletely removed[21]. However, if properly enucleated, recurrences are rare.
For MMS, several approaches are extensively cited in literature, including marsupialisation, external approaches, Caldwell–Luc procedures, and endoscopic sinus or trans-nasal surgery[9,11,15,22,23,24].

The recurrence rate of mucoceles of paranasal sinuses can vary from 0.9% to 23%, and usually is more common in patients with chronic paranasal sinus inflammation or with multiple surgery history[15,25]. In Case 3, a Caldwell–Luc approach was used, which offered appropriate visualisation of the MMS, thereby assuring removal of the entire lesion. Long-term follow-up appointments are mandatory.

Conclusion

Owing to their similar radiographic features and asymptomatic presentations, AP, SCC, and MMS can be misdiagnosed. Clinical signs and symptoms, patient history, and adequate radiographic evaluation are necessary to make accurate decisions regarding their initial diagnosis and subsequent treatment plans.

Consent

Because of the retrospective and descriptive nature of this study, institutional ethical review board approval was not needed. The authors state that the Declaration of Helsinki guidelines were followed. Written patient consent has been obtained to publish clinical photographs.

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