

Nur Syareena Musa<sup>1</sup>, Jeyasakthy Saniasiaya<sup>2</sup>, Norhaslinda Abdul Gani<sup>1</sup>

## A case of unexpected cheek swelling

### Obrzęk policzka związany z torbielą zastoinową zatoki szczękowej – opis przypadku

<sup>1</sup> Department of Otorhinolaryngology, Hospital Tuanku Ja'afar, Jalan Rasah, Seremban, Negeri Sembilan, Malaysia

<sup>2</sup> Department of Otorhinolaryngology, Faculty of Medicine, University of Malaya, Jalan Universiti, Kuala Lumpur, Malaysia

Correspondence: Jeyasakthy Saniasiaya, Department of Otorhinolaryngology, Faculty of Medicine, University of Malaya, Jalan Universiti, 50603 Kuala Lumpur, Malaysia, tel.: +6012-6841223, e-mail: shakthy\_18@yahoo.com

#### ORCID iDs

1. Nur Syareena Musa <https://orcid.org/0000-0001-5688-4835>
2. Jeyasakthy Saniasiaya <https://orcid.org/0000-0003-1974-4379>
3. Norhaslinda Abdul Gani <https://orcid.org/0000-0002-4725-6666>

#### Abstract

**Aim of the study:** The aim of this case presentation is to emphasise the importance of meticulous history-taking and physical examination, and increase the awareness of various possibilities for the diagnosis and management of patients with unilateral cheek swelling. Maxillary mucocele is a rare benign cystic lesion of paranasal sinuses. It may be associated with varying symptoms, and patients commonly present with compression symptoms during their first visit, as the maxillary sinus is adjacent to vital structures, notably the orbit and skull base. **Case report:** Herein, we present the case of a middle-aged man who presented with painless unilateral cheek swelling that turned out to be maxillary mucocele. **Conclusions:** Presentation of maxillary mucocele often mimics cancerous lesion types. Thorough assessment and proper imaging help to guide otorhinolaryngologists towards making an accurate diagnosis and initiating prompt management.

**Keywords:** maxillary mucocele, cheek swelling, benign mass

#### Streszczenie

**Cel pracy:** Celem przedstawionego opisu przypadku jest zwrócenie uwagi na konieczność dokładnego badania podmiotowego i przedmiotowego u pacjentów zgłaszających się z jednostronnym obrzękiem policzka oraz zwiększenie świadomości w zakresie różnych możliwości diagnostycznych i metod postępowania w takich przypadkach. Torbiel zastoinowa zatoki szczękowej jest rzadko występującą, łagodną zmianą torbielowatą, która rozwija się w obrębie zatok przynosowych. Może wywoływać rozmaite objawy, a pacjenci najczęściej zgłaszają się do poradni z objawami uciskowymi, co wynika z umiejscowienia zatoki szczękowej w pobliżu kluczowych struktur anatomicznych, zwłaszcza oczodołu i podstawy czaszki. **Opis przypadku:** W pracy przedstawiono przypadek mężczyzny w średnim wieku, który zgłosił się do poradni z powodu bezbolesnego jednostronnego obrzęku policzka. W toku diagnostyki ustalono, że jest to torbiel zastoinowa zatoki szczękowej. **Wnioski:** Objawy torbieli zastoinowej zatoki szczękowej często przypominają objawy towarzyszące zmianom nowotworowym. Dokładna ocena i odpowiednie badania obrazowe pomagają otorynolaryngologom w ustaleniu prawidłowego rozpoznania i szybkim wdrożeniu leczenia.

**Słowa kluczowe:** torbiel zastoinowa zatoki szczękowej, obrzęk policzka, łagodny guz

## INTRODUCTION

**M**ucocele of the paranasal sinus is a slow-growing, benign, mucous-containing sac with an expansive nature. It can expand locally, whilst destroying adjacent structures, with ensuing compression pressure from the overgrowth of the mass<sup>(1-3)</sup>. Its locally aggressive nature often mimics a cancerous type of lesion, making clinical diagnosis difficult. The most common site of mucocele is the frontal sinus, which accounts for about 65% of cases, followed by the ethmoid-sphenoid sinus region, accounting for 30% of cases, and finally, the maxillary sinus. Maxillary sinus mucoceles represent about 3 to 10% of all mucocele types<sup>(1-3)</sup>. A progressively enlarging cheek swelling in an elderly man points towards a malignancy, however a benign entity, such as mucocele, also needs to be kept in mind as a diagnostic possibility.

## CASE REPORT

A 59-year-old man presented to our clinic with a 6-month history of painless left cheek swelling. According to the patient, the left cheek swelling had been progressively increasing in size. The patient denied any history of nasal obstruction and eye problems. There was no significant history of sinusitis, allergy or trauma, and no history of nasal, sinus or dental surgery, either. Clinical examination revealed the presence of left cheek swelling measuring 3 × 4 cm, fixed to the underlying structure, with firm inconsistency. Nasal endoscopy only showed congestion of the left ostiomeatal complex (OMC), whereas oral examination revealed the presence of a cystic lesion in the upper left third molar region, which was tender on palpation. Urgent computed

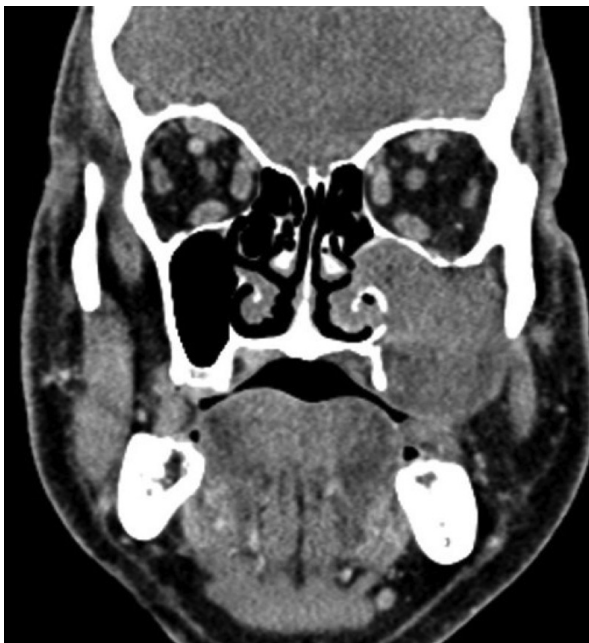


Fig. 1. Coronal view of expansile left maxillary sinus mass eroding the floor of sinus

tomography of the paranasal sinuses (CT PNS) was done and showed heterogeneously a left maxillary sinus mucocele measuring 4.4 × 4.2 × 4.1 cm. The mass abutted the infratemporal fossa and the masseter space (Figs. 1, 2). The outgrowth of the mass compressed and destroyed the left maxillary anterior, inferior, medial, and lateral wall, with no evidence of enhancing lesion that was suggestive of malignancy.

We proceeded with bedside intraoral incision and drainage (I&D) under local anaesthesia (LA), and 50cc of thick brownish pus was aspirated. A sample of pus and tissues was sent for analysis, and the result of the sample was suggestive of chronic inflammation, with no evidence of malignancy, while cultures of pus revealed no evidence of organism growth. Imaging of CT PNS post I&D showed no residue of the left maxillary sinus mucocele with minimal mucosa thickening in the left maxillary sinus and left ethmoid air cells. The presence of bony destruction of the anterior, inferior, medial, and lateral walls of the left maxillary sinus remained the same as compared with the previous imaging examination. Functional endoscopic sinus surgery was scheduled for the patient in six months. The importance of preoperative sinus care with nasal irrigation and long-term use of a nasal corticosteroid spray was emphasized to the patient before the surgery.

## DISCUSSION

Mucoceleles are basically lined by non-neoplastic epithelium which contains a thick mucoid secretion<sup>(4)</sup>. The pathophysiology of mucocele is linked to an obstruction of the sinus ostium as well as its drainage system. Due to the ongoing mucous production, erosion and remodelling of adjacent bony wall structures follow as the mucocele slowly enlarges<sup>(1)</sup>. As for the aetiology of mucocele, it is mainly due to either allergic sinonasal disease, chronic infection



Fig. 2. Homogenous mass in the left maxillary sinus, eroding the left floor and lateral wall

with inflammation, post-trauma scarring, or previous nasal surgery<sup>(1,4,5)</sup>.

Common clinical presentation varies depending on the site of involvement. Usually, patients remain asymptomatic in the early phase and present at a later stage, usually when the mucocele is large enough to cause compression pressure and subsequently lead to serious ocular and intracranial complications<sup>(6)</sup>. The most common presenting complaints of maxillary mucoceles include cheek swelling, pain, nasal obstruction, ophthalmoplegia, and tooth loosening<sup>(7,8)</sup>. Our patient presented with a progressively enlarging unilateral cheek swelling. As the mucocele in our patient expanded in a more inferior direction, the superior and lateral wall was intact; hence there were no ocular symptoms.

CT is the gold standard imaging modality in all mucocele cases. In our patient, the presence of a non-enhancing lesion with smooth bone erosion, which did not invade the surrounding structure, aided in the diagnosis and ruled out a malignancy, which usually appears as an enhanced lesion with irregular erosion of the bony margin and infiltration of the surrounding structure<sup>(3)</sup>. Magnetic resonance imaging aids in delineating the intracranial and orbital extension.

The treatment of choice for maxillary sinus mucoceles is mainly surgery<sup>(5,8,9)</sup>. Numerous surgical approaches exist, including open or endoscopic approaches, depending on the location, extent, and the degree of surgical expertise of the operator. Currently, the most preferred surgical technique is via the endoscopic approach. Endoscopic sinus surgery is deemed effective, as it enables easier tumour resection with minimal morbidity and low recurrence rate<sup>(5,10,11)</sup>. The endoscopic surgical approach enables evacuation of the mucocele content through a wide middle meatal antrostomy<sup>(5,9)</sup>. However, in extensive paranasal sinus mucoceles, the endoscopic technique can have some limitations, thus necessitating a combination approach<sup>(11)</sup>. Additionally, non-surgical treatment such as nasal douching and nasal steroids may aid in improving mucociliary clearance of the paranasal sinus, as well as reducing inflammation and infection that attribute to mucocele formation. As with all mucoceles, long-term follow-up is still necessary to watch for signs of recurrence<sup>(10)</sup>.

## CONCLUSION

Maxillary sinus mucoceles has an excellent prognosis if diagnosed early with prompt surgical intervention. We would like to highlight a more feasible method of drainage of inferiorly extended maxillary sinus mucoceles via an intra-oral approach. Regular and long-term follow-up also helps to detect early lesions and prevent their recurrence.

### Conflict of interest

*Authors do not report any financial or personal connections with other persons or institutions that might claim authorship rights to this publication.*

### References

1. Marks SC, Latoni JD, Mathog RH: Mucoceles of the maxillary sinus. *Otolaryngol Head Neck Surg* 1997; 117: 18–21.
2. Özcan M, Akdoğan Ö, Gün T: Giant mucocele of the maxillary antrum: report of a case. *Türk Arch Otolaryngol* 2002; 40: 150–152.
3. Jayaraj SM, Patel SK, Ghufoor K et al.: Mucoceles of the maxillary sinus. *Int J Clin Pract* 1999; 53: 391–393.
4. Lund VJ, Henderson B, Song Y: Involvement of cytokines and vascular adhesion receptors in the pathology of fronto-ethmoidal mucoceles. *Acta Otolaryngol* 1993; 113: 540–546.
5. Kennedy DW, Josephson JS, Zinreich SJ et al.: Endoscopic sinus surgery for mucoceles: a viable alternative. *Laryngoscope* 1989; 99: 885–895.
6. Morita S, Mizoguchi K, Iizuka K: Paranasal sinus mucoceles with visual disturbance. *Auris Nasus Larynx* 2010; 37: 708–712.
7. Martin RJ, Jackman DS, Philbert RF et al.: Massive proptosis of the globe. *J Oral Maxillofac Surg* 2000; 58: 794–799.
8. Khong JJ, Malhotra R, Selva D et al.: Efficacy of endoscopic sinus surgery for paranasal sinus mucocele including modified endoscopic Lothrop procedure for frontal sinus mucocele. *J Laryngol Otol* 2004; 118: 352–356.
9. Busaba NY, Salman SD: Maxillary sinus mucoceles: clinical presentation and long-term results of endoscopic surgical treatment. *Laryngoscope* 1999; 109: 1446–1449.
10. Devars du Mayne M, Moya-Plana A, Malinvaud D et al.: Sinus mucocele: natural history and long-term recurrence rate. *Eur Ann Otorhinolaryngol Head Neck Dis* 2012; 129: 125–130.
11. Lee TJ, Li SP, Fu CH et al.: Extensive paranasal sinus mucoceles: a 15-year review of 82 cases. *Am J Otolaryngol* 2009; 30: 234–238.