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Case Report

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Sinocutaneous Fistula Secondary to Chronic Maxillary Rhinosinusitis

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Abstract

Sinocutaneous fistula is a rare complication of chronic rhinosinusitis (CRS), most commonly involving the frontal sinus, with maxillary sinus involvement being uncommon and typically associated with trauma, surgery, or odontogenic infection. We report a rare case of a maxillary sinocutaneous fistula secondary to chronic maxillary rhinosinusitis in the absence of trauma, prior surgical intervention, fungal infection, or neoplasia. The condition resulted from longstanding inflammation with associated osteitis and cortical erosion of the maxillary sinus wall, leading to abnormal communication with the overlying facial skin. Diagnosis was established through clinical evaluation and contrast-enhanced computed tomography of the paranasal sinuses. Definitive management consisted of functional endoscopic sinus surgery with complete excision of the fistulous tract and primary multilayered closure. This case underscores the potential for chronic inflammatory sinonasal disease to cause rare bony complications and highlights the importance of considering CRS in the differential diagnosis of persistent facial cutaneous fistulae.

Keywords: Chronic rhinosinusitis; Maxillary sinus; Osteitis; Sinocutaneous fistula

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INTRODUCTION

Chronic rhinosinusitis (CRS) is defined as inflammation of the nasal cavity and paranasal sinuses lasting for more than 12 weeks. Diagnosis requires the presence of at least two of the following symptoms: nasal obstruction or congestion, anterior or posterior nasal drainage, hyposmia or anosmia, and facial pressure, pain, or fullness supported by endoscopic or radiographic findings. When inadequately treated, CRS can be complicated by secondary infection, with contiguous spread beyond the paranasal sinuses to adjacent structures. Complications are traditionally classified as orbital, bony, and intracranial, with orbital involvement being the

most frequently reported.[1]

Sinocutaneous fistula is a rare manifestation of rhinosinusitis, characterised by abnormal communication between a paranasal sinus and the overlying skin. It typically develops as a result of osteitis of the sinus walls, a bony complication that may progress to cortical erosion. The frontal sinus accounts for the majority of reported cases, whereas involvement of the maxillary sinus is uncommon.[2,3] Maxillary sinocutaneous fistula formation is usually associated with facial trauma or previous surgical intervention rather than primary inflammatory disease.[4,5]

We report a rare case of a sinocutaneous fistula arising from chronic maxillary rhinosinusitis in the absence of trauma, prior surgery, fungal infection, or neoplasia. This case highlights an unusual bony complication of CRS and underscores the importance of considering chronic inflammatory disease in the differential diagnosis of facial cutaneous fistulae.

CASE REPORT

A 44-year-old man with no known medical comorbidities presented with a persistent discharging dimple over the right cheek for one year. At symptom onset, he experienced right facial swelling, which subsided following a course of oral antibiotics. However, purulent discharge persisted, accompanied by a dull ache during mastication. He also reported symptoms consistent with CRS, including nasal obstruction, facial discomfort, and purulent nasal discharge. There was no history of facial trauma or previous surgery. The patient initially sought treatment from the Oral and Maxillofacial Surgery (OMFS) service, where extractions of teeth 15 and 16 were performed due to suspected odontogenic pathology. Despite this intervention, his symptoms failed to resolve. A cone beam computed tomography (CBCT) scan demonstrated soft-tissue density in the bilateral maxillary sinuses, prompting referral to the Otorhinolaryngology service for further evaluation.

On examination, there was a skin dimple with an external opening discharging pus over the right buccal cheek region (**Figure 1a**). Intraoral examination revealed a fully edentulous right upper gingiva with no visible swelling or defect. Multiple retained teeth were also noted in the upper left gingiva. Nasal endoscopy showed medialisation of the lateral nasal wall, oedema of the middle meatus, and thick mucopurulent discharge bilaterally (**Figure 1b**). No nasal masses or polyps were identified.

Based on clinical findings, a diagnosis of CRS with suspected right maxillary sinocutaneous fistula was made. Contrast-enhanced computed tomography of the paranasal sinuses (CECT PNS) revealed heterogeneous soft-tissue density involving all paranasal sinuses, with erosion of the inferolateral wall of the right maxillary sinus and right maxillary process (**Figure 2**). The lesion extended anterolaterally into the right cheek, associated with skin thickening and dimpling, confirming the diagnosis of a right maxillary sinocutaneous fistula. The left maxillary sinus walls appeared sclerotic.

The patient was started on mometasone nasal spray (two puffs once daily), alkaline nasal douching, and a one-week course of oral amoxicillin-clavulanate (625 mg three times daily). He was subsequently scheduled for bilateral functional endoscopic sinus surgery (FESS) with excision of the right sinocutaneous fistula via a Caldwell–Luc approach. Intraoperatively, the maxillary antrum was found to be lined with thickened, inflamed mucosa, and mucopurulent material was drained. A bony defect in the anterolateral wall of the right maxillary sinus was palpated, covered only by thickened mucosa, with adjacent granulation tissue (**Figure 3a**).

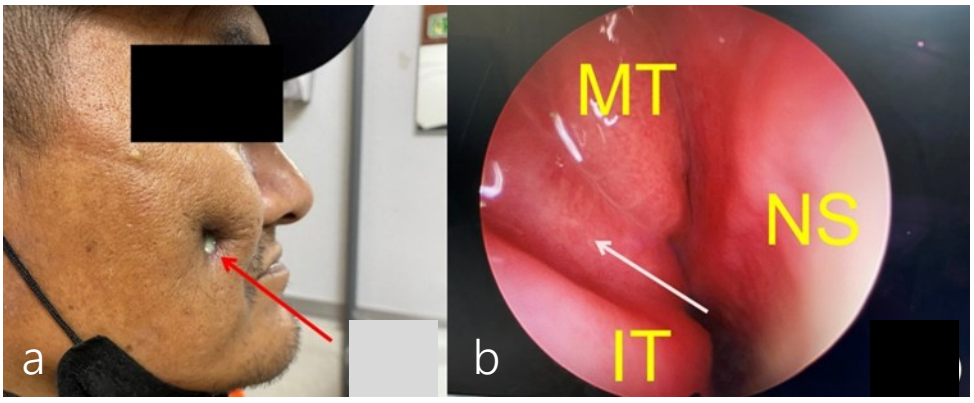

Fig. 1: a) Skin dimple with discharging pus over right buccal cheek (red arrow: skin dimple). 1b} Nasal endoscopy of right nose showed medialisation of the lateral nasal wall, oedema of the middle meatus, and thick mucopurulent discharge (white arrow: middle meatus; MT: middle turbinate; IT: inferior turbinate; NS: nasal septum).

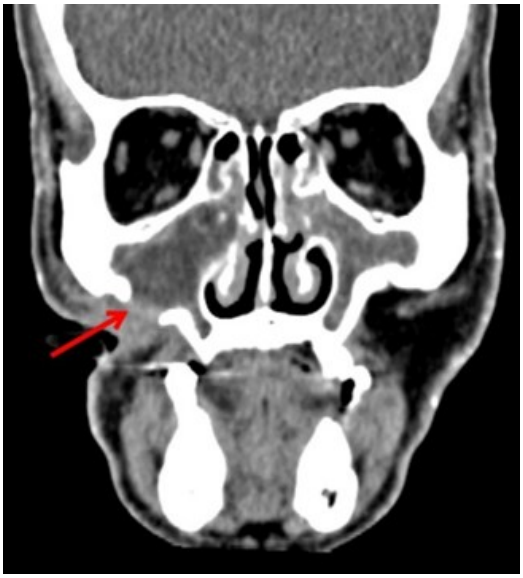

Fig. 2. Contrast-enhanced computed tomography of the paranasal sinuses (CECT PNS) revealed heterogeneous soft-tissue density involving the maxillary sinuses, with erosion of the inferolateral wall of the right maxillary sinus and right maxillary process (red arrow: site of bony erosion)

The fistulous tract was identified extending from the external cheek to the maxillary antral wall defect and was inspected through a sublabial incision (**Figure 3b**). The tract measured approximately 0.5 cm in length. An elliptical incision was made around the skin dimple, and the fistulous tract was carefully dissected and excised in its entirety. The wound was closed in layers, involving subcutaneous tissue and skin.

At one-week postoperative review, the surgical wound was well healed, and sutures were removed. Histopathological examination of the excised tract revealed inflamed tissue with no evidence of malignancy. Unfortunately, the patient did not attend subsequent follow-up visits.

DISCUSSION

CRS is not limited to mucosal inflammation and may extend to the underlying bone, resulting in osteitis. Histopathologically, CRS-related osteitis is characterised by periosteal thickening, increased osteoblastic and osteoclastic activity, fibrosis, and predominant formation of immature woven bone. This disorganised bone is structurally weaker than mature lamellar bone, predisposing it to structural failure. Osteitis in CRS has been associated with bacterial biofilms, persistent inflammatory cytokines, dysregulation of RUNX2-mediated osteogenesis, and altered transforming growth factor–β (TGF-β) signaling, leading to ongoing bone remodeling and fragility.[6]

In our case, longstanding untreated CRS likely led to progressive maxillary osteitis, erosion of the sinus wall, and subsequent sinocutaneous fistula formation, presenting as persistent purulent discharge from the right cheek. Chronic inflammation, immature bone formation, and increased intramaxillary pressure from infection likely acted together to promote cortical erosion and fistulation. Erosion may have been further influenced by an odontogenic source, as the floor of the maxillary sinus lies close to the roots of the first and second molars, facilitating direct spread of dental

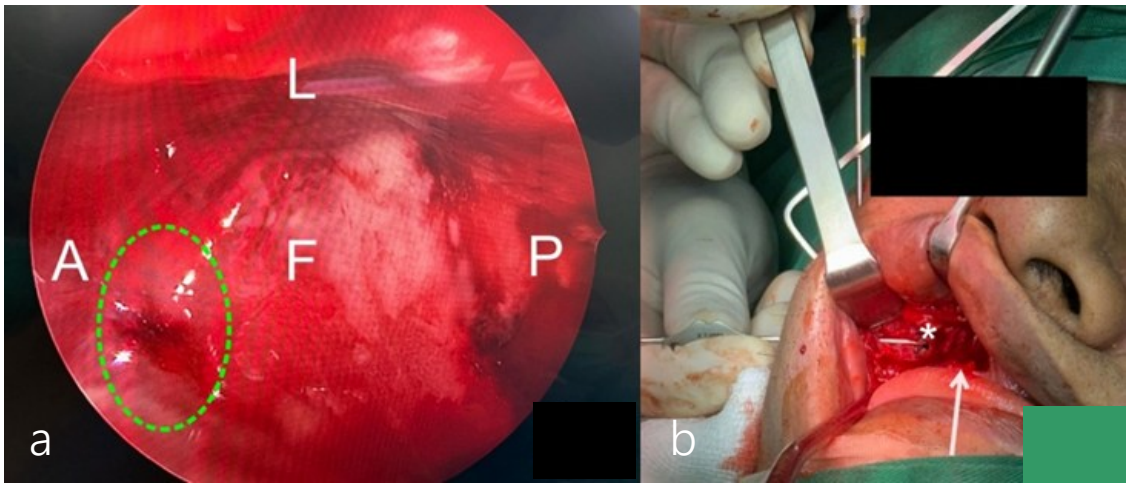

Fig. 3: a) Bony defect in the anterolateral wall of the right maxillary sinus (green circle: bony defect; A: anterior; P: posterior; L: lateral; F: floor). 3b. Fistulous tract extending from the external cheek to the maxillary antral wall defect inspected through a sublabial incision (white star: antral wall defect; white arrow: sublabial incision).

infection. The defect occurred at the anteroinferior maxillary wall, corresponding to the canine fossa, which is the thinnest and structurally weakest region of the maxilla, predisposing it to bone breakdown and fistula formation.[7]

Although sinocutaneous fistula of the maxillary sinus is rarely reported, our case demonstrates its potential occurrence and should be considered in the differential diagnosis of a discharging cheek fistula. Other differentials include tuberculosis infection especially in endemic regions, salivary gland fistula, pyogenic granuloma, and neoplasm.[8] In the present case, the patient was initially referred to the OMFS team, but symptoms persisted despite extraction of the suspected offending teeth. The diagnosis of CRS was only suspected after a CBCT scan was performed.

The primary goal of management is to re-establish effective sinus drainage and to completely excise the fistulous tract. Management strategies vary according to the sinus involved and the associated anatomical challenges. In frontal sinus disease, the narrow and anatomically complex frontal recess presents significant technical difficulty, with an increased risk of injury and recurrence. In such cases, frontal trephination may be required to achieve adequate drainage, and in selected situations, sinus obliteration or flap interposition may be necessary to prevent recurrence.[2,3] In contrast, in our opinion, for spontaneous maxillary sinocutaneous fistulae, FESS combined with complete fistula excision and primary multilayered closure is generally sufficient to achieve disease resolution and prevent recurrence.

CONCLUSION

Maxillary sinus sinocutaneous fistula is a rare complication of CRS. Longstanding inflammation and osteitis can result in cortical erosion and fistula formation, mimicking odontogenic or other cutaneous pathology. Diagnosis requires careful clinical evaluation and imaging, such as CECT PNS. Definitive management involves restoring sinus drainage via FESS and excision of the fistulous tract with primary multilayered closure, which is generally sufficient to achieve disease resolution and prevent recurrence. Awareness of this entity is crucial for timely diagnosis and appropriate surgical management.

Take Home Message

- A persistent discharging lesion over the cheek is not always odontogenic.
- Rarely, CRS can cause a spontaneous sinocutaneous fistula, so it should be part of the differential diagnosis.
- Neglected CRS can extend beyond the mucosa into bone, causing osteitis, weakening the sinus wall and leading to erosion and fistula formation, highlighting the importance of early recognition and adequate treatment of CRS.
- CT imaging of the paranasal sinuses is crucial for identifying sinus disease, as it can demonstrate soft tissue changes as well as bony involvement, including erosion and sclerosis.

Abbreviations

CRS	Chronic rhinosinusitis
OMF	Oromaxillofacial
CBCT	Cone beamed computed tomography
CECT PNS	Contrast enhanced computed tomography of the paranasal sinuses
FESS	Functional endoscopic sinus surgery

Declarations

All the authors declared no competing interests.

Ethical Consideration

Written consent was obtained from all patients for publications of the clinical details and accompanying images.

Acknowledgement

None

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