Otolaryngology Case Reports 9 (2018) 4–7

Contents lists available at ScienceDirect

Otolaryngology Case Reports

journal homepage: www.elsevier.com/locate/xocr

Developmental anomaly – A duplicate fronto-ethmoidal sinus complex

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ARTICLE INFO

Keywords:
Paranasal sinus
Ethmoid sinus
Development anomaly
Tumor

ABSTRACT

Background: Fronto-ethmoidal sinus tumors can be benign or malignant.
Case presentation: This case reports a patient with a long-standing frontal sinus lesion who presented with seizures. Imaging findings suggested malignancy. The patient underwent craniofacial resection – intraoperative findings revealed a lesion centered in the anterior ethmoid sinuses with extension into the frontal sinus, characterized by multiple bony septations. Histology was negative for malignancy. These findings were similar to that of ethmoid sinus morphology, which are of endochondral bony origin. As the lesion has been long-standing, this was postulated to be a duplicate ethmoid sinus.
Conclusion: This is the first reported case of a possible duplication of the ipsilateral ethmoid sinus. This aids clinicians in diagnosing patients who present similarly, avoiding extensive surgical procedures with significant morbidity risk.

Introduction

Embryologically, the ethmoid is of endochondral bony origin, whereas the other sinuses are extensions from the ethmoid as membranous bone. Thus the ethmoid sinus has paper-thin bony lamellae, which can take different extramural migratory paths, whilst other sinuses form septations that are more rigid and robust [1].

In our clinical practice, we encountered a patient with what we postulated to be a developmental anomaly of the fronto-ethmoidal sinuses – a duplication of the ipsilateral ethmoidal sinus. The patient presented with a seemingly aggressive frontal sinus mass, but was worked up to have a benign development anomaly.

Case Presentation

Our patient was a 58-year-old Chinese male who presented to the emergency department with a generalized tonic-clonic seizure. He reported prior mild frontal headaches and intermittent nasal congestion, and his family reported that he had facial asymmetry and right eye proptosis for many years. On examination the patient was alert, extraocular eye movements, visual acuity, and neurological examination was normal. Nasoendoscopy showed a fleshy, bosselated tumor obstructing the right anterior nasal space (ANS) (Fig. 1). There was no cervical lymphadenopathy. Blood investigations showed an elevated total white cell count (TWC) of 13 with left shift. Electrolytes were normal.

Magnetic resonance imaging (MRI) of the brain showed a 8cm multi-lobulated cystic lesion in the right ANS, encroaching onto bilateral frontal sinuses (right more than left), right anterior ethmoid and maxillary sinuses (Fig. 2). Superiorly there was right frontal lobe invasion with vasogenic edema and dural enhancement, causing a 5mm leftward midline shift. Inferolaterally there was right orbit extension with associated proptosis, abutting the superior rectus muscle complex but sparing the optic nerve. This mass was hyperintense on T2. Radiological features were suspicious for malignancy.

Computed Tomography (CT) scan of the sinuses showed multiple calcified septations present within the frontal, ethmoid and maxillary sinuses, as well as a portion of the tumour within the right orbital apex and anterior cranial fossa. CT scan of the thorax, abdomen, pelvis and bone scan done showed no distant metastases (Fig. 3).

In view of the clinical history and radiological features that suggested an aggressive pathology, multiple biopsies were done. Endoscopic biopsy showed sinonasal mucosa without malignancy. A repeat deeper biopsy showed ulceration with reactive changes and benign respiratory epithelial lined cysts. The third open biopsy of the supraorbital component via a right anterior orbitotomy, showed benign mucocele(s).

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https://doi.org/10.1016/j.xocr.2018.07.004
Received 15 February 2018; Accepted 16 July 2018
Available online 17 July 2018

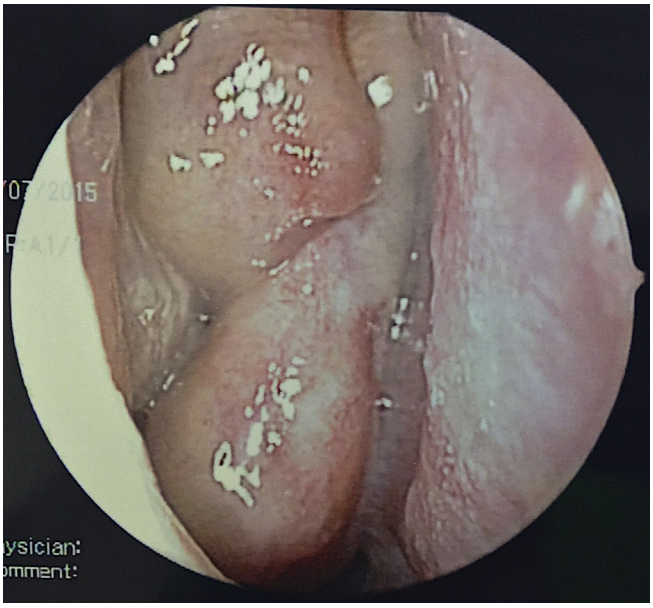

Fig. 1. Nasoendoscopy showing right anterior nasal space entirely obscured by frontoethmoidal sinus lesion.

The patient was presented at our institution's multidisciplinary tumor board, and in view of its worrisome clinical profile with possible intracranial/intraorbital involvement, decision was made for craniofacial resection with possible orbitotomy/orbital exenteration.

After comprehensive pre-operative counseling and informed consent taken, the patient underwent endoscopic-assisted craniofacial resection. Intraoperative findings showed a multi-septated lesion centered in the ethmoidal sinus, extending into the frontal sinus. Each of the multiple cells within the lesion were lined with normal appearing respiratory epithelium. The middle and inferior turbinates were not eroded but pushed apart by the lesion. The lesion was endoscopically evacuated piecemeal with a combination of debriding with a powered shaver and curetting. Craniotomy was performed to approach the supero-lateral tumor. There was a significant component of the tumor that remained over the supraorbital ridge extending posteriorly – thinning the posterior table of the frontal sinus. However, there was a clear surgical plane of dissection during the excision of the lesion from the frontal lobe with no breach of dura. Final histology showed benign nasal mucosa with mucin-filled cysts. Gram stain and cultures were all negative.

Post-operatively the patient had an uncomplicated recovery, and was discharged well on the ninth post-operative day. His best-corrected visual acuity was 6/15 for his right and 6/12 for his left. Intraocular pressure, pupillary reflexes, cornea reflex and Bell's reflex were normal.

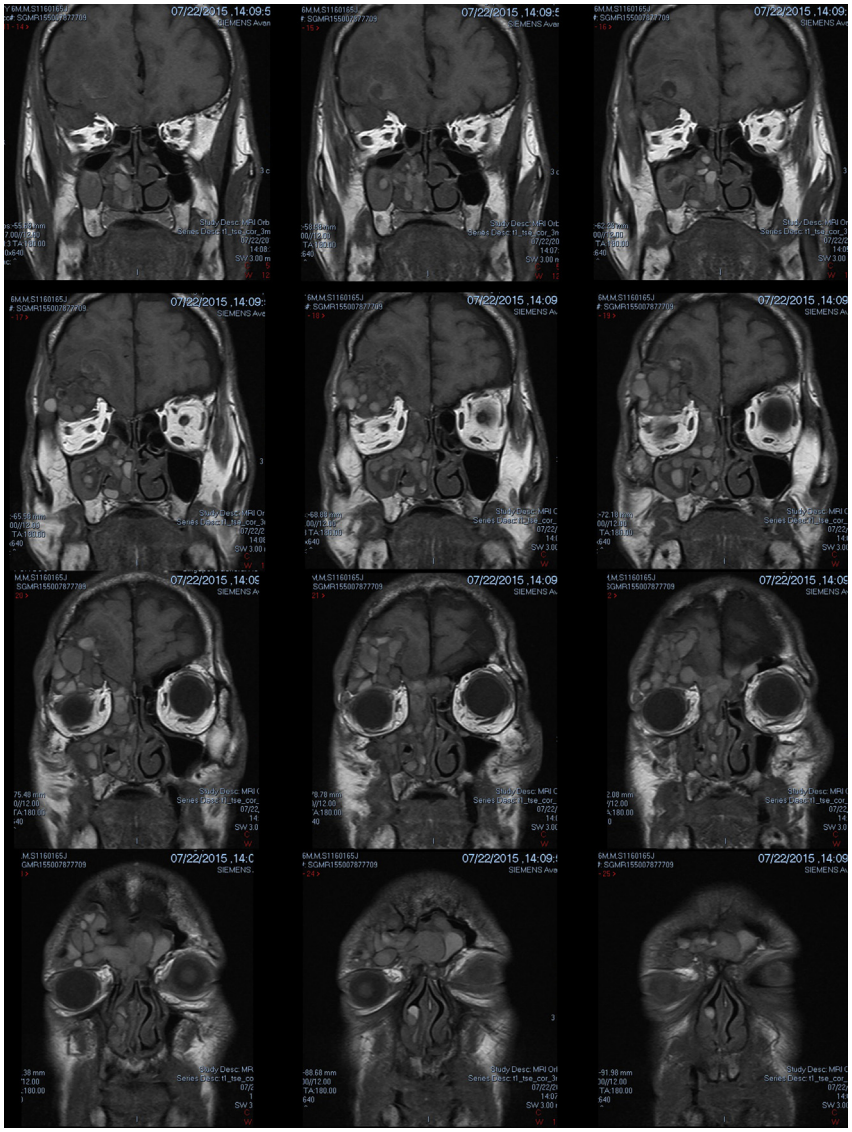

Fig. 2. MRI (T1 sequence) showing consecutive images of frontal sinus mass from posterior to anterior.