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CASE REPORT



Bony orbital changes in allergic fungal sinusitis are reversible after treatment

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ABSTRACT

Allergic fungal sinusitis (AFS) arises from a host hypersensitivity reaction to fungi residing within the sino-nasal tract. Computed tomography imaging may show heterogenous sinus opacification with bony erosion and expansion into the orbits. With advanced orbital involvement there is a risk of optic neuropathy and irreversible vision loss. We present a patient with AFS who presented with bilateral proptosis and early optic neuropathy. Radiologically, there was evidence of bony erosion and orbital wall compression. Following oral corticosteroids and full-house endoscopic sinus surgery, these changes reversed considerably. This case shows that bony and anatomical orbital changes from AFS are reversible with adequate surgical treatment.

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Allergic fungal sinusitis;
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Allergic fungal sinusitis (AFS) is categorised as a type of non-invasive fungal disease, usually affecting patients who are young and immunocompetent with a history of atopy. The disease arises from a host hypersensitivity reaction to fungi within the sinuses, leading to chronic inflammation. Implicated fungi include *Aspergillus* and dematiaceous fungi such as *Bipolaris*, *Culvularia* and *Exserohilum*.¹ Expansion of affected ethmoid sinuses can compress the orbits and such patients may present with proptosis, epiphora and decreased vision secondary to compressive optic neuropathy.² Computed tomography (CT) imaging may show heterogenous sinus opacification with bony erosion and expansion into the orbits.^{2,3} We present the first reported case of AFS where bony orbital erosion and deformity have been demonstrated to reverse considerably after treatment.

Case report

A 25-year-old black African female presented to otolaryngology outpatient clinic with a 6-month history of severe nasal congestion and loss of both taste and smell. In addition, she described worsening bilateral proptosis over the past 3 months with blurred vision and epiphora of the left eye. Her medical history included mild asthma and previous functional endoscopic sinus surgery 2 years prior. Nasal endoscopy revealed grade 2 nasal polyposis. CT scan showed signs of AFS with bony erosion and lateral displacement of the lamina papyracea and

complete “serpiginous” opacification within the sinuses (Figure 1).

On ophthalmic assessment her vision was 6/5 OU with no relative afferent pupillary defect (RAPD). Ishihara plates were 14/15 OD and 10/15 OS. Subjective luminance and red saturation testing were both 6/10 OS compared to 10/10 OD. On fundoscopy, optic discs were healthy with a cup-disc ratio of 0.3 bilaterally. Hertel exophthalmometry revealed bilateral proptosis worse on the left, reading 24 mm OD and 28 mm OS (Figure 2). Extraocular movements were full however with pain on extremes of gaze. There was no lagophthalmos. Visual field testing with standard automated perimetry was normal. A diagnosis of early left compressive optic neuropathy was made.

The patient was prescribed oral prednisolone 25 mg once daily and underwent bilateral full house endoscopic sinus surgery (FESS). Intraoperative findings included diffuse fungal debris occupying all paranasal sinuses with dehiscent laminae papyracea. All debris was removed endoscopically from all sinuses including the ethmoid sinuses bilaterally resulting in a cavity on each side with the lamina papyracea laterally, the skull base superiorly and middle turbinates medially. Histopathology of removed tissue showed inflammatory oedematous mucosa with eosinophils and plasma cells. Periodic acid-Schiff staining was negative for fungal hyphae. However, tissue culture grew *Aspergillus fumigatus* complex.

The patient's post-operative course was uncomplicated and her oral prednisolone was weaned. At



Figure 1. Coronal and axial computed tomography scans of sinuses and orbits on presentation (top) and after treatment (bottom). Reversal of bony changes of skull base and lamina papyracea shown by arrows.



Figure 2. Clinical patient photograph on initial review (top) and at 3-month follow-up post endoscopic sinus surgery (bottom).

3-month follow-up at the orbital clinic her proptosis had improved, reading 21 mm OU on exophthalmometry (Figure 2). Visual acuity remained 6/5 OU with normal visual fields on standard automated perimetry and no RAPD. In addition, Ishihara testing was normal at 12-month follow-up indicating likely reversed optic

neuropathy. Her symptoms of nasal congestion, loss of taste and smell, epiphora and blurred vision had resolved. She no longer experienced pain with extremes of gaze. Repeat CT (performed 3 months post-operatively due to suspected residual right maxillary sinus disease) showed post-operative changes with

remarkable, almost complete, radiological resolution of disease, including reversal of previous bony erosion and sinus expansion into the medial orbits (Figure 1).

The patient continues to be reviewed annually at the orbital clinic.

Comment

Up to one quarter of patients with AFS may experience ophthalmic consequences from orbital involvement of the disease.² In our case, the patient had developed early optic neuropathy signified by loss of colour vision and red discrimination. Therefore, any patient with suspected orbital involvement should be referred promptly to an orbital specialist as signs of optic neuropathy may be subtle.

CT is the imaging of choice for AFS. Typical appearance includes unilateral or bilateral nonhomogeneous, classically “serpiginous” opacification of the paranasal sinuses.^{2,3} Bony erosion of the lamina papyracea or skull base may be evident with associated expansion of disease into the orbits or intracranial cavity respectively.⁴ Bony erosion is more commonly found in patients of younger age and of African ethnicity.³

Management choices include oral or intranasal corticosteroids followed by FESS. The aim of surgery is to remove the fungal debris, excise involved ethmoid septae/cells and inflammatory polyps, and facilitate drainage of the thick allergic-mucin typically found within the sinonasal tract of affected patients.⁵ Histopathology of removed content demonstrates inflammatory debris with eosinophils and plasma cells and occasionally sparse fungal hyphae without features of invasion.⁵ Culture may isolate a particular pathogen, however may take several weeks and is often negative. Orbital decompression or reconstruction is rarely required for AFS. Decreased orbital volumes in affected patients with proptosis have been

shown to improve after successful endoscopic sinus surgery.⁶

To our knowledge this is the first reported case of severe AFS where bony erosion, deformity and lateral displacement of the lamina papyracea are shown to reverse considerably after FESS. This was paralleled with post-operative clinical resolution of our patient’s proptosis and optic neuropathy at 12-month follow-up. Therefore, even in cases of severe AFS with extensive orbital changes, marked clinical and radiological improvement of disease is possible with adequate treatment.

Disclosure Statement

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the article.

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