CASE REPORT

Unilateral cacosmia: a presentation of maxillary fungal infestation

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SUMMARY

We present a case of long-standing unilateral cacosmia in a healthy 67-year-old man due to maxillary fungal infestation. Treatment with septoplasty had been attempted 10 years prior but no further investigation or management undertaken and symptoms continued. Subsequent MRI scan revealed significant opacification of the left maxillary sinus. This was readily amenable to treatment by balloon sinuplasty. This yielded viscous grey mucus which grew *Scedosporium apiospermum*. The case highlights the need for careful investigation of olfactory symptoms, including blood tests to exclude systemic causes, endoscopy and imaging where indicated.

BACKGROUND

Olfactory disturbances are common and distressing. They need to be investigated methodically to exclude serious and treatable underlying pathology. Psychosocial implications of olfactory disorders must also be considered.¹

CASE PRESENTATION

A 67-year-old Caucasian gentleman presented with a 15-year history of left-sided cacosmia; the unilateral nature of the symptom was a key feature. He had presented to his local ENT department on numerous occasions and was diagnosed with a deviated septum, with treatment culminating in a septoplasty 10 years previously. No additional investigations had been performed. He reported no nasal blockage, rhinorrhoea or postnasal drip. There was no known trigger for his olfactory distortion and he had no known allergies. He reported that his sense of taste was intact, had no significant medical history and was a non-smoker (having never smoked). He had had no significant occupational exposure to chemicals.

INVESTIGATIONS

Endoscopic nasal examination revealed residual deviation of the nasal septum with a left-sided septal spur. There was no evidence of any significant pathology in either middle meati and both olfactory clefts were clear. Formal testing of olfaction yielded a Sniffin' Stick Test² score of 13.5/48 indicating functional anosmia (figure 1). Following protocol, further investigations included blood tests (including B₁₂, folate, urea and electrolytes (U&E)), liver function tests, thyroid function tests (TFT), 9.00 cortisol, Venereal Disease Research Laboratory test (VRDL), angiotensin converting enzyme (ACE), antineutrophil cytoplasmic antibodies and

glycated (glycosylated) haemoglobin) and an MRI of the head focused on the olfactory bulbs, tracts and relevant higher centres in the brain. The blood tests were all within normal limits and the MRI was reported to show minimal mucosal thickening in the left maxillary sinus, with no intracranial abnormality. However, further review of the imaging by the senior author, showed opacification of more than 50% of the left maxillary sinus (figure 2).

TREATMENT

The patient was offered surgery to the maxillary sinus and opted to undergo a left maxillary balloon sinuplasty (figure 3). He chose balloon sinuplasty as he wished to have any procedure undertaken with local rather than general anaesthetic. After successful dilation of the maxillary ostium, sinus irrigation produced a large amount of viscous grey mucus. This was sent for microscopy, culture and sensitivity and grew *Scedosporium apiospermum* on fungal culture (figure 4). Topical nasacort (triamcinolone acetonide) and gentamicin were injected into the sinus cavity at the time of surgery. The senior author routinely uses a solution of saline containing baby shampoo and gentamicin for sinus washout; the fungal content was only identified in the lab subsequently.

OUTCOME AND FOLLOW-UP

Postoperatively cacosmia resolved without antifungal agents. Subjective sense of smell also improved and remained improved 4 months later.

DISCUSSION

Fungal spores are known to be an aetiological factor in the development of sinonasal disease, for example allergic fungal rhinosinusitis is increasingly recognised as a specific subtype of chronic rhinosinusitis.³ Patients with fungal sinus disease frequently complain of olfactory disturbance, with 26.7% of patients in a case series of 160 patients reporting cacosmia-hyposmia;⁴ fungal infestation should therefore be considered as part of the differential diagnosis in any patient with smell distortion.

Fungal infestations of the sinuses are most likely to occur in the maxillary sinus and patients with disease in the maxillary or ethmoid sinuses will commonly describe symptoms mimicking those of chronic rhinosinusitis, with rhinorrhoea, blocked nose and facial pain;⁴ clinicians should consider fungal disease as a possible diagnosis in patients with suspected chronic rhinosinusitis.

To cite: Erskine SE, Schelenz S, Philpott CM. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2013-008808

Figure 1 (A and B). Sniffin' sticks scores reveal functional anosmia.

Many different fungal organisms may be implicated in the development of sinus problems.^{3, 4} *S apiospermum* in particular is an environmental filamentous fungus commonly found in soil.⁴ It is the anamorph of *Pseudallescheria boydii* and was first described as a cause of mycetoma over 100 years ago. It is capable of colonising respiratory cavities including the sinuses in otherwise immune competent patients,⁵ but very rarely causes serious infections. *Scedosporium* spp, occasionally cause severe

invasive infections in the immunocompromised host particularly following lung transplantation.⁶

A MEDLINE search did not identify any reported cases of sinusitis caused by *S apiospermum* in immune competent adults although Cooley *et al* reported one case in their review of *Scedosporium* cases. Neither a case series of 160 patients with sinus fungal balls⁴ nor a series of 92 patients with fungal chronic rhinosinusitis³ identified *S apiospermum* as a causative agent.

Figure 2 MRI showing opacification of the left maxillary sinus.

Learning points

- ▶ Olfactory symptoms may indicate serious underlying pathology and can cause distress and impaired quality of life, although many causes are readily amenable to treatment.
- ▶ Olfactory symptoms should be investigated thoroughly and methodically in a timely manner.
- ▶ Imaging should be requested when no clear cause can be identified or when sinonasal surgery is indicated¹ and the requesting clinician should review this personally; in this case the underlying pathology would have been missed if the radiological report was read without assessing the original images.
- ▶ Fungal infestation is one cause for severe cacosmia which can be successfully managed by surgical clearance with topical and/or systemic antifungal treatment as necessary.

Figure 3 Introduction of balloon to maxillary sinus.

Figure 4 *Scedosporium apiospermum*.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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