CASE REPORT

Fungus ball in the nasal cavity mimicking a rhinolith

Ajay M Bhandarkar,¹ Ranjini Kudva,² Keshwar Damry,¹ Balaji Radhakrishnan²

¹Department of Otolaryngology-Head and Neck Surgery, Kasturba Medical College, Manipal University, Manipal, Karnataka, India
²Department of Pathology, Kasturba Medical College, Manipal University, Manipal, Karnataka, India

Correspondence to
 Dr Ajay M Bhandarkar,
 bm.ajay@gmail.com

Accepted 13 June 2016

SUMMARY

A 63-year-old man presented to the outpatient department of our tertiary care hospital, with unilateral left nasal obstruction, foul smelling nasal discharge and occasional mild epistaxis. On examination, a hard greyish black mass was noted in the anteroinferior portion of the inferior turbinate, with extension posteriorly into the inferior meatus with surrounding unhealthy granulation tissue. Provisional diagnosis of rhinolith was surprisingly reversed by histopathology, which suggested the presence of an *Aspergillus* fungus ball with *Actinomyces* colonisation.

BACKGROUND

A fungus ball in the paranasal sinuses is a relatively common entity. However, the presence of a fungus ball in the nasal cavity is extremely rare. It can be misdiagnosed as a rhinolith due to its appearance, which more or less resembles a rhinolith. Although the management is similar, there are subtle differences in imaging that can help in preoperative diagnosis of a fungus ball in the nasal cavity. We present the case of a 63-year-old man who presented with features suggestive of a rhinolith and, astonishingly, histopathological examination suggested an *Aspergillus* fungus ball with *Actinomyces* colonisation.

CASE PRESENTATION

A 63-year-old man presented to the outpatient department of our tertiary care hospital, with unilateral left nasal obstruction, foul smelling nasal discharge and occasional mild epistaxis of 4 years duration. There was no history of headache, facial pain, postnasal drip or fever. There was no history of diabetes mellitus, trauma or prior odontogenic procedures. The patient did not smoke, and neither consumed alcohol nor inhaled snuff. However, he had a history of a prior nasal procedure carried out 5 years prior to this presentation, the details of which were not known.

Nose and paranasal sinus examination revealed a deviated nasal septum to the right side and thick mucopurulent discharge in the left nasal cavity. On suction clearance of the discharge, a hard greyish black mass was noted in the anteroinferior portion of the inferior turbinate, with extension posteriorly into the inferior meatus with surrounding unhealthy granulation tissue (figure 1). There was no paranasal sinus tenderness. Ear, oral cavity and oropharynx examination was normal. Systemic examination was within normal limits. Clinically, a provisional diagnosis of rhinolith was considered.

INVESTIGATIONS

A swab from the nasal cavity, sent for culture and sensitivity, was suggestive of the presence of

Actinomyces. Contrast-enhanced CT scan revealed a calcific dense lesion in the left inferior turbinate, suggestive of a rhinolith with right frontal sinusitis (figure 2).

DIFFERENTIAL DIAGNOSIS

Owing to the presence of prior nasal procedure and colonisation of the nasal cavity with *Actinomyces*, we settled for a primary diagnosis of rhinolith with secondary *Actinomyces* infection. Osteoma and chondroma were the other remotely considered diagnoses, but our primary diagnosis was rhinolith. We did not suspect the possibility of a fungus ball because of its rarity.

TREATMENT

The patient underwent endoscopic removal of the nasal lesion under general anaesthesia. During the procedure, it was noted that the mass was attached to the inferior turbinate with no erosion of mucosa. It was removed in toto. Postoperatively, the specimen was dissected to find any evidence of a nidus for the mass formation from the previous nasal procedure, without any success. The mass was sent for histopathological examination. Haemostasis was achieved and the nasal pack placed in situ. The pack was removed 2 days postoperatively following which the patient was discharged.

OUTCOME AND FOLLOW-UP

The patient followed up a week after the primary surgery and, to our surprise, the final diagnosis on histopathology was of a fungus ball with the presence of *Actinomyces* and *Aspergillus* in the specimen. Microscopy revealed the presence of ulcerated stratified squamous epithelium with stroma showing the dense lymphoplasmacytic infiltrate with congested vessels, dead bone spicules, calcification, necrotic debris and actinomycotic colonies with Splendore-Hoeppli material (figures 3 and 4). There was evidence of a fungus ball composed of narrow septate hyphae. Gram staining showed Gram-positive rods (*Actinomyces*), and special staining techniques performed included periodic acid-Schiff (PAS)-positive for an *Aspergillus* fungus ball, and Gomori methenamine silver, which was positive for both, *Aspergillus* fungus ball and *Actinomyces* (figures 5–9). The patient followed up 3 months after the primary surgery and was free from symptoms.

DISCUSSION

Fungus ball was a term introduced by DeShazo. To satisfy the criteria of a fungus ball, the lesion should have the presence of the following findings: (1) a non-invasive nature of the fungus to surrounding tissue; (2) cheesy or mucopurulent

To cite: Bhandarkar AM, Kudva R, Damry K, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2016-215490

Figure 1 Endoscopic image depicting the presence of a hard greyish black mass with surrounding unhealthy granulation tissue.

Figure 2 Coronal CT scan image showing a calcific dense lesion in the left inferior turbinate.

clay-like material; (3) opacification of the lesion with or without calcification; (4) hyphae conglomerate with demarcation from the surrounding mucosa; (5) allergic mucin, granuloma or increased eosinophil levels.¹ Fungus balls are commonly seen in the paranasal sinuses, however, their presence in the nasal cavity is extremely rare. This is presently the fourth case in the world literature of an *Aspergillus* fungus ball in the nasal cavity, and possibly the second case of presence of a mixed infection with *Actinomyces* and *Aspergillus*.

Actinomyces is termed as a masquerader in head and neck infections. In 1877, Harz introduced the term for this filamentous, anaerobic, Gram-positive saprophyte, a name that is synonymous with the Greek for ray fungus, because of its appearance. Israel was the first country to isolate it as a causative agent for clinical disease in humans, and it was termed as *Actinomyces israelii*, which is the commonest *Actinomyces* sub-species. Other subspecies of *Actinomyces* are *Actinomyces*

Figure 3 H&E staining at x10 magnification showing ulcerated stratified squamous epithelium with stroma showing the dense lymphoplasmacytic infiltrate with congested vessels, dead bone spicules, calcification and necrotic debris.

Figure 4 H&E staining at x10 magnification showing the presence of Actinomycotic colonies.

Figure 5 H&E staining at x40 magnification showing the presence of septate fungal hyphae.

odontolyticus, *Actinomyces naeslundii*, *Actinomyces propionica* and *Actinomyces viscosus*. *Actinomyces* causes cervicofacial infections and the main factor is injury to the mucosa, which propagates the disease. The pathognomic sign of Actinomycotic infections is the presence of multiple pus draining fistulae.

Figure 6 Gram staining at $\times 40$ magnification showing the presence of Gram-positive rods (*Actinomyces*).

Figure 7 Magnification at $\times 40$ showing GMS staining positivity of *Actinomyces*. GMS, Gomori methenamine silver

Figure 8 Magnification at $\times 40$ showing PAS positivity of *Aspergillus*. PAS, periodic acid-Schiff.

These infections are usually seen in the age group of 40–70 years, with predominance in males, at a ratio of 1.5–3:1. There is no racial predominance and no predominance seen in immunosuppressive states, although a small percentage of immune depleted patients do demonstrate the presence of this opportunistic saprophytic organism producing a symptomatic infection.^{2–4}

Figure 9 Magnification at $\times 40$ showing GMS positivity of *Aspergillus*. GMS, Gomori methenamine silver.

Fungus balls in the nasal cavity have unknown aetiology. Presence of a contracted sinus and a wide meatus, previous dental treatment, poor orodental hygiene and previous surgery causing mucosal trauma, have been suggested as possible predisposing factors, however, if a fungus ball has to develop in any cavity, the pathogenetic mechanism should be the presence of an ostial closure, which will trap fungal spores in an anaerobic environment favouring fungal growth and further accumulation of fungal salts would give it a bony hard consistency. The literature suggests the role of exogenous and endogenous niduses as being responsible for concretion formation.^{2 4–6} The presence of an endogenous nidus such as inflammation leading to pus in the nasal cavity due to entrapped fungus can be hypothesised as the possible nidus in the pathogenesis of our case. The presence of *Actinomyces* colonisation may be secondary to the long-standing infestation of the fungus ball with minimal injury to the mucosa, which may be the predisposing factor to the presence of *Actinomyces* in the nose.

The symptomatology is typical of a foreign body or rhinolith, with unilateral nasal obstruction, headache or facial pain due to sinus inflammation caused by blocked ostia, postnasal drip, and nocturnal cough and cacosmia. Endoscopic examination usually reveals the presence of a mass in the nasal cavity, associated with foul smelling discharge.^{2 4 5} Imaging and culture of the discharge usually clinches the diagnosis in most cases. The fungal concretum phenomenon suggests that the increased density of fungus balls is due to the presence of deposition of phosphate and sulfate salts of calcium in the necrotic centre of the fungal mass. The incidence of focal and well-defined hyperdense foci on CT scan is 50% and 25%, respectively. This can be particularly differentiated from a rhinolith, which shows increased peripheral density on CT scan. MRI, though complementary in diagnosis, may show isointense or hypointense signals to the brain on T2-weighted images.^{5 7} Fungal culture for *Actinomyces* in brain-heart or blood agar reveals the presence of filamentous Gram-positive rods rich in sulfur granules, which not only confirms the presence of *Actinomyces* but also differentiates it from nocardiosis, which has sulfur granules without Gram-positive rods. Histology shows a typical fungal granuloma with *Actinomyces* colonies with reactive inflammation and necrotic areas with scar formation.² Gomori methenamine silver and PAS are the special staining techniques employed in the diagnosis of *Aspergillus* and *Actinomyces*.

The treatment of fungus balls is endoscopic sinus surgery and debridement. Antibiotic therapy with penicillin or tetracycline/clindamycin/erythromycin for 6 weeks is the ideal therapy for *Actinomyces*. The role of antifungals is reserved only in cases of bone invasion by the fungus. Hyperbaric oxygen has been used as adjunct therapy.^{2 5} There are no incidents of recurrence of fungus balls in the nasal cavity in the literature.

Learning points

- ▶ Fungus ball is an important differential diagnosis for rhinolith.
- ▶ CT scan may be able to differentiate between a rhinolith and fungus ball.
- ▶ Special staining techniques such as Gomori methenamine silver and periodic acid-Schiff are useful in diagnosing fungus balls.
- ▶ Functional endoscopic sinus surgery is the treatment of choice for a fungus ball in the nasal cavity.

Acknowledgements The authors thank the ENT and pathology departments for their support.

Contributors AMB prepared the manuscript and is the corresponding author. RK was the histopathologist and reviewer. KD performed the review of the literature and prepared the images and case history. BR was the histopathologist and reviewer.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 deShazo RD. Fungal sinusitis. *Am J Med Sci* 1998;316:39–45.
- 2 Ozcan C, Talas D, Görür K, *et al.* Actinomycosis of the middle turbinate: an unusual cause of nasal obstruction. *Eur Arch Otorhinolaryngol* 2005;262:412–15.
- 3 Baliga S, Shenoy S, Wilson G, *et al.* An unusual case of actinomycosis. *Ear Nose Throat J* 2002;81:44–5.
- 4 Kim KS, Kim HJ. A case of the nasal cavity fungus ball. *Mycoses* 2011;54:e244–7.
- 5 Lee GH, Yang HS, Kim KS. A case of inferior meatus fungus ball. *Br J Oral Maxillofac Surg* 2008;46:681–2.
- 6 Nayak DR, Bhandarkar AM, Valiathan M, *et al.* Incidental 'ethmoid sinolith'—an unusual cause of frontal recess obstruction. *BMJ Case Rep* 2014;2014:pii: bcr2014204157.
- 7 Palacios E, Jones W, Alvernia J. Sinonasal mycetoma. *Ear Nose Throat* 87:606–8.

Copyright 2016 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow