

Case Report

A Rare Cause of Fungal Rhinosinusitis: *Exophiala jeanselmae*

V Sha Kri Eh Dam¹, Ramiza Ramza Ramli²

Abstract

Fungal rhinosinusitis (FRS) is a rarely encountered disease; however, the incidence is increasing. It has a strong association with the impaired immune system of the host as well as a suitable environment for fungal growth. Fungal spores are abundant in the surroundings and can enter the nasal airway and the paranasal sinuses (PNS) through inhalation; however, only a small number of patients develop FRS. The primary organisms responsible for FRS are *Aspergillus* sp., *Rhizopus* sp., and some of the dematiaceous fungi. We present a case of FRS in an immunosuppressed patient caused by scarce fungal species, *Exophiala jeanselmae*. This rare fungus has never been reported to be associated with FRS before. They are dematiaceous fungi of low virulence and infection are usually restricted to skin and soft tissue.

Keywords: Fungal, rhinosinusitis, *Exophiala jeanselmae*

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Introduction

Fungal infection of paranasal sinuses (PNS) is assumed as uncommon previously; however, the incidence has been reported to be increasing.¹ A recent study conducted in Singapore showed prevalence of 8.4%,² while another studies in India showed higher prevalence of 30% to 44%.³ Fungal rhinosinusitis (FRS) can be classified into invasive and non-invasive type, depending on the potential of the fungal hyphae to invade the tissues through the epithelium and also in terms of chronicity, either less or more than four weeks.⁵ Non-invasive FRS is further categorized into two subgroups, which are fungal ball and allergic FRS. In comparison, invasive type FRS can be divided into three subgroups, namely acute invasive FRS, chronic invasive FRS, and chronic granulomatous invasive FRS.

Invasiveness of the fungal infection is dependent on the host and environment factors, in which

the majority of cases are non-invasive type.⁴ Lately, invasive FRS has been identified as a significant cause of morbidity and mortality. This is believed due to the extensive use of broad-spectrum antibiotics, immunosuppressive therapy, cancer chemotherapy and increased incidence of immunodeficiency diseases that lead to the increase in incidence of FRS.⁴ Usage of broad-spectrum antibiotics will suppress the growth of normal bacterial flora, thus increases the burden of fungal colonization and the risk of disseminated fungal infection.⁶

Approximately 300 documented fungal species that cause diseases in humans and animals. More than 50 fungal pathogens have been isolated as causative organism of FRS.⁷ Generally, *Aspergillus* sp. is the most common organism in FRS, however *Rhizopus* sp. is identified as the main organism responsible for specific invasive type of FRS.^{8,9}

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Other less commonly encountered fungi are dematiaceous fungi, including *Curvularia*, *Bipolaris*, and *Exserohilum*. Fungal spores are abundant in the atmosphere and are part of healthy sinonasal flora. The fungus is prevented from proliferating by the immunological cascades in a normal condition. The pathological potential developed when the environment is suitable for it and if there is a disruption of the normal immunological pathways.

Exophiala jeanselmei is a dematiaceous fungus and classically reported to be associated with subcutaneous infection due to trauma, which resulted in eumycetoma.¹⁰ This fungus also has been reported as causative organism for chromoblastomycosis, disseminated infections, endocarditis, and arthritis.¹⁰ Other rare systemic presentation of *Exophiala jeanselmei* infection reported in the lung, brain, peritoneum, and esophagus.¹¹ However, to the best of our knowledge, there is no reported case of FRS caused by this organism.

Case report

A 40-year-old man with underlying diabetes mellitus type 2 and chronic kidney disease stage 5, presented with right nasal blockage and foul-smelling yellowish nasal discharge for one-week duration. It was associated with right facial pain, which worsened on bending forward. There was no fever, headache, blurring of vision, or diplopia.

Nasoendoscopic examination revealed polypoidal mass at right middle meatus with mucopus (Figure 1). The sensation was intact upon probing. Otherwise, the surrounding mucosa looked healthy with no eschar seen. Endoscopic directed pus swab was taken for culture and sensitivity (C&S). The left nasoendoscopic examination showed normal finding. Initially, the patient was treated as right maxillary sinusitis and was given a week of oral antibiotic (amoxicillin-clavulanate), nasal decongestant, steroid nasal spray, and alkaline nasal douching.

During the follow-up one week after, there was no more facial pain, nasal blockage and nasal discharge, but the polypoidal mass with mucopus still presence in the right middle meatus on nasoendoscopy. The patient was instructed to continue the same antibiotic for another two weeks

as C&S showed so far no growth. Unfortunately, he defaulted follow up and presented again five months later with intermittent right nasal blockage, post-nasal drip, and halitosis. Nasoendoscopic examination showed mucopus at the right middle meatus with surrounding inflamed polypoidal mucosa. C&S taken on the first visit showed the growth of *Exophiala jeanselmei*.

Computed tomography (CT) of PNS showed a soft tissue density occupying almost the entire right maxillary sinus cavity, extending to the middle meatus, ethmoid sinus and frontal sinus. There was presence of bony erosion at the medial wall of the right maxillary sinus, ethmoid sinus and uncinate process (Figure 2 and 3). No intralesional calcification was seen. The patient was scheduled for right functional endoscopic sinus surgery; however, he was not keen for surgery at the moment due to his current medical comorbidity. He was kept on closed follow-up and was advised on strict compliance on medical treatment, i.e., continuation usage of steroid nasal spray and alkaline nasal douching regularly.

On subsequent follow-ups, the patient's disease remained stable, whereby he had no new complaint, and the examination revealed no mucopus at the right middle meatus, and the surrounding mucosa looked healthier. Thus, this case was categorized into non-invasive FRS with characteristic features more suggestive of allergic type.

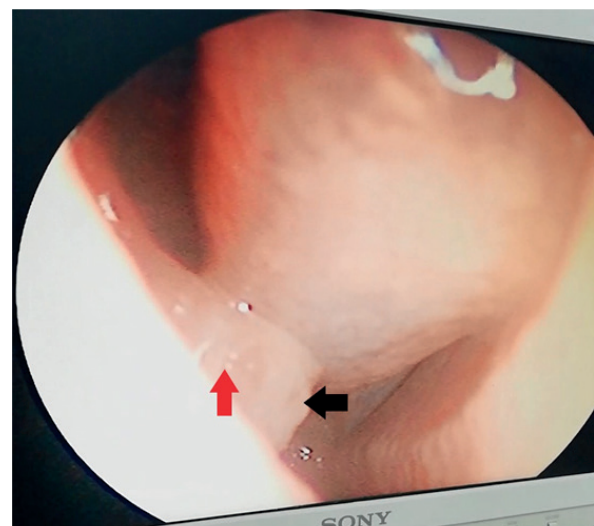


Figure 1. Endoscopic view of right nasal cavity shows polypoidal mass (red arrow) at the right middle meatus with overlying mucopus (black arrow). Surrounding mucosa is healthy.



Figure 2. CT PNS, coronal view shows soft tissue density in the right maxillary sinus, extending to middle meatus, ethmoid sinus and frontal sinus. There is presence of erosion at the medial wall of right maxillary sinus, ethmoid sinus and uncinate process. No intralesional calcification seen.

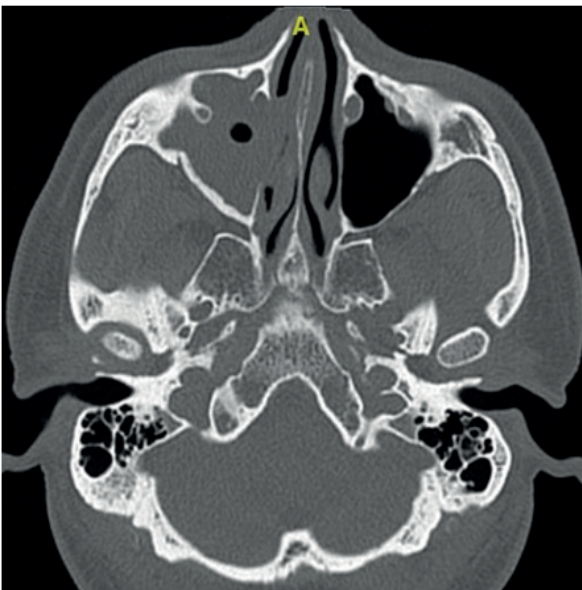


Figure 3. CT PNS, axial view shows soft tissue density in the right maxillary sinus with presence of erosion at the medial wall. No intralesional calcification seen.

Discussion

FRS is a well-known entity; however, diagnosis is often overlooked in patient with rhinosinusitis. The most common presentation of FRS is nasal obstruction, followed by nasal discharge and facial pain or swelling.^{9,11} Our patient also presented with these symptoms; however,

bacteria rhinosinusitis also shared similar presentation, thus making the diagnosis of FRS more challenging. In cases of rapidly worsening of facial swelling, facial numbness, proptosis, diplopia, visual loss, headaches, and neurological deficit especially in an immunosuppressed patient with rhinosinusitis, invasive FRS should be highly suspected. Although the present case also in immunocompromised state, however the disease is non-invasive which could be explained by the low virulence of the causative organism.

The environmental and host factors are believed to be responsible for the development of FRS. The incidence of FRS is reported to be higher in areas of warm and dry climate like north India and Sudan.⁸ Fungus isolated in FRS, particularly allergic FRS, also has a geographic variation. *Aspergillus flavus* is the most common fungus cultured in Asia (India and Saudi Arabia), while dematiaceous fungi are more prevalence in United States.¹² Two retrospective studies conducted in Malaysia showed a consistent result of *Aspergillus* sp. as the leading cause of FRS.^{9,13} A rare organism like *Exophiala jeanselmai* is not previously reported. This could be due to the very rare organism in environment that cause FRS, or low viability and slow growing of the fungus that very difficult to get a positive culture. *Exophiala jeanselmai* is a dematiaceous fungus with low virulence, found in the soil, wood, polluted water, and sewage.¹¹ It can persist in the skin tissue of healthy hosts for months to years without disseminating to other organs.

Exophiala jeanselmai was grow in our specimen and we believed it was due to a high fungal load and proper specimen collection. Endoscopic guided sinonasal culture and immediate transport of the nasal swab to the laboratory probably ensure the fungal viability, therefore, yielding a desirable result. This method has been proven to accurately identify bacterial pathogens in rhinosinusitis patient.¹⁴ Although endoscopic guided pus swab for C&S is not routinely performed in all rhinosinusitis cases, but we think it should be done at least in immunocompromised patients when presence of mucopus. The management will be different if culture is positive for fungus as seen in the present case.

CT scan is another good tool for diagnosis and pre-operative planning. Presence of bony erosion may signify invasive FRS but need to correlate with other physical examinations and

nasoendoscopic findings. There was presence of bony erosion radiologically in the present case, however nasoendoscopic findings showed healthy surrounding mucosa, thus more suggestive of non-invasive type. Prolonged duration of infection may explain the presence of bony erosion in the present case.

The principle of management in all types of FRS is surgically fungal deloading. Antifungal like amphotericin B, liposomal amphotericin and, voriconazole are reserved for the invasive type of FRS.⁵ Our patient warrants a surgical removal and deloading of fungal material under general anesthesia; however, since he was not keen for operation due to his medical problem, we had to reserve to a symptomatic treatment, i.e., regular nasal douching as well as steroid nasal spray and vigilant follow-up. In addition, patient should be advised on immediate follow up if any worsening symptoms or complications arise.

Conclusion

FRS should be suspected in the immunocompromised patient presented with acute or chronic rhinosinusitis. The principle of management is dependent on the type of FRS, the patient's general condition, and the sensitivity of fungi to antifungal therapy. The ability to identify the organism, i.e., fungi, is an ideal for any FRS cases to fine-tune for treatment accordingly,

especially when prescribing antifungal regime. This approach would be more valuable in cases dealing with rare fungi such as this case. Empirical treatment and early surgical deloading showed an impressive outcome but must be supported with sound clinical history and examination, taking into account all the risk factors.

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Conflict of Interest

The authors have disclosed no conflict of interest.

Ethical Approval Issue

There was no ethical approval sought other than getting consent from the patient.

Authors' contribution

Conception and design: VSKED

Critical revision of the article for important intellectual content: RRR

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