**CASE REPORT**

**Nodulisporium: An Extremely Rare Cause of Allergic Fungal Rhinosinusitis**

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**ABSTRACT**

The world of rhinology has smelt and dealt with a steadily increasing number of allergic fungal rhinosinusitis cases that were initially mistaken for chronic rhinosinusitis. Although both group of patients present similarly, the allergic fungal counterpart does not respond to conventional medical therapy. We report a case of a young gentleman who combated chronic rhinosinusitis for years before undergoing functional endoscopic sinus surgery which yielded an extremely rare fungal pathogen, Nodulisporium species, clinching the diagnosis of allergic fungal rhinosinusitis. Operative intervention was key to definitive diagnosis and treatment for this case. We discuss the challenges and methods used in identifying Nodulisporium, the surgical approach, and procedures performed in treating this condition.

**Keywords:** Chronic rhinosinusitis, Endoscopic sinus surgery, Fungal sinusitis, Mycosis, Paranasal sinuses.

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**INTRODUCTION**

Allergic fungal rhinosinusitis (AFRS) has been commonly described as an allergic reaction to environmental fungi, namely, dematiaceous fungi or Aspergillus species, in an otherwise immunocompetent host.¹ Fungal pathogens commonly implicated in AFRS includes Aspergillus, Rhizopus, Alternaria, Bipolaris, and Curvularia.² The emergence of Nodulisporium as an agent of phaeohyphomycosis has been reported in isolated case reports as the pathogen responsible in AFRS and even in a case of central nervous system infections.³⁴ Although specific fungal species identification carries little weight in the current treatment, it is pivotal to understand its nature and disease process in humans for advancement and development of future treatment. We discuss the challenges encountered in the identification and diagnosis of this extremely rare fungal species, Nodulisporium causing AFRS in our patient. Functional endoscopic sinus surgery (FESS) was aimed at obtaining the specimen, correction of anatomical deformity which impeded sinus drainage, and ample widening of the maxillary ostium. By adequately widening the maxillary ostium, we created a window for future endoscopic access to the diseased sinus. This window was crucial for further antral lavages and monitoring of the disease until complete resolution.

**CASE DESCRIPTION**

A 36-year-old gentleman presented with complaints of rightsided nasal blockage, facial pain, and purulent foul-smelling nasal discharge for the past 2 years. He works as a police officer but had history of working in a wood factory 17 years ago. Rigid nasoendoscopy showed high deviated septum to the right, small grade I polyp, and grossly congested right osteomeatal complex (OMC) that was filled with secretion. Computer tomography (CT) of the paranasal sinuses revealed complete opacification of the right maxillary sinus, blocking the OMC (Figs 1 and 2). There was sclerosis of the maxillary sinus wall suggestive of disease chronicity. Ipsilateral anterior ethmoidal air cells and frontal sinus had mucosal thickening. The impression was chronic rhinosinusitis (CRS) with nasal polyps of right ethmoidal and maxillary sinuses. The patient subsequently underwent septoplasty with right FESS under general anesthesia. We corrected the high deviated nasal septum, performed uncinctomy, polypectomy, and large middle meatal antrostomy (MMA) for ample widening of the maxillary ostium to drain the thick and brownish secretion that resemble allergic mucin from maxillary antrum. The overlying mucosa was severely hypertrophied, polypoidal, and unhealthy. Tissue biopsies of the antrum and polyps were obtained, and they were consistent with AFRS. Nodulisporium species was identified in the biopsied tissue sent for fungal culture. The patient recovered well without the use of steroids or immunotherapy. However, he relapsed after 3 months with similar but milder symptoms. Rigid nasoendoscopy revealed congested nasal mucosa with thick discharge from the maxillary antrum. Antral washout was performed under in the clinic. We took advantage of the widened maxillary ostium and inserted a J-sucker Eicken antrum cannula into the antrum and performed saline lavage. Purulent discharge and debris was suctioned out, giving the patient symptomatic relieve. The patient recovered well giving the patient symptomatic relieve. The patient recovered well

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following the antral lavage with a complete resolution of nasal symptoms. Subsequent reviews revealed a satisfied asymptomatic patient with clear nasoendoscopy findings. This patient does not require antifungal, oral steroids, and intranasal steroid spray since the operation and has been disease free for more than 1 year.

**Discussion**

AFRS is essentially a hypersensitivity reaction by an immunocompetent host against fungi in the paranasal sinus. It is both a form of noninvasive fungal sinusitis and a hypersensitivity disease. The causative fungi are usually dermatiaceous fungi or *Aspergillus* species. This is the second case report implicating *Nodulisporium* species, a genus that contains both dermatiaceous and non-dermatiaceous members, as the pathogen responsible for a case of AFRS. *Nodulisporium* is a naturally occurring fungi and tends to accompany conidial anamorphs of certain wood decay ascomycetes. Interestingly, the occupational history of our patient revealed a significant exposure to wood dust while working in a wood factory previously.

The identification process of *Nodulisporium* species was an arduous undertaking. The specimen was cultured onto Sabouraud’s dextrose agar plate and sent at ambient temperature to the Mycology Laboratory, Institute for Medical Research Malaysia. Upon retrieval, a spreading whitish fungal colony was observed. The colony was subsequently re-subcultured onto a potato dextrose agar (PDA) plate, pH 5.6 ± 0.2, to enhance the sporulation and onto corn-meal agar at pH 6.0 ± 0.2, for slide culture preparation. The plates were incubated at 30°C, and the colony maturity was checked every alternate day. After 1 week, the colony on PDA which was initially white turned to grayish color. Lactophenol Cotton Blue (LPCB) staining technique was adopted. A drop of LPCB stain was placed onto a clean glass slide. Using aseptic technique, a small portion of the fungal colony was picked up using a tease needle and placed onto the stain to suspend the cells. Then, a cover slip was attached and pressed down gently to spread out the fungal filaments and cells. The slide was then observed under light microscopy, and the morphology of the fungal elements was noted as septate and slightly roughened hyphae (Figs 3 to 5). *Nodulisporium* species can be wrongly identified as *Daldinia* and *Hypoxylon* species, which have the same nodulisporium-like structure. Accurate identification was carried out using polymerase chain reaction sequencing method.

In order to accurately diagnose AFRS, we utilized modified Bent and Kuhn diagnostic criteria by DeShazo and Swain. The classical Bent and Kuhn criteria includes type I (Immunoglobulin
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E mediated) hypersensitivity which may be absent in more than one-third of patients with AFRS.4 Other criteria such as evidence of nasal polyps, characteristic CT findings, allergic mucin, and positive fungal stain were positive in this patient. As our patient presented like most CRS with nasal polyps patients, we initially performed a FESS and septoplasty with intention of restoring the ventilation and enhancing the sinus drainage. The postoperative result was satisfactory until 3 months later.

Incomplete clearance of fungal debris and allergic mucin through nasal douching will lead to relapse of symptoms as seen in our patient. The optimally widened maxillary ostium from previous large MMA proved beneficial in providing a window to the diseased maxillary antrum. Copious antral lavages through the ostium was performed in the clinic. This procedure abolished the need to subject patient to general anesthesia, hence eliminating operative risk, time, and cost. The result was excellent with complete resolution of symptoms and nasendoscopic finding.

However, we note the possibility of recurrence of the disease and have reserved the role of steroids therapy, immunotherapy, and antifungal therapy as an adjunct to surgical debridement in future.

CONCLUSION

We conclude that there is an emerging identification of extremely rare fungal pathogens such as Nodulisporium species in AFRS due to operative advancements that permit accessibility in obtaining proper sinus specimens. The use of FESS has facilitated the positive yield of sinus culture, allowing accurate identification of the pathogen and therefore targeted therapy in managing AFRS.

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