

# Rhinosinusitis Caused by Unusual Fungus *Fonsecaea pedrosoi* - A Rare Case Report from South India

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**Abstract** Phaeohyphomycosis causing rhinosinusitis is not very frequent. We here report a case of rhinosinusitis caused by *Fonsecaea pedrosoi* in a 53 year old diabetic male, with deviated nasal septum. With surgical correction and antifungal treatment with itraconazole, the patient's condition improved and he was discharged. *Fonsecaea* exists in the environment as a saprophyte and its infection is usually not lethal. However in immunocompromised individuals, when there is systemic invasion the mortality rate is high. With limited treatment options available for this fungus, early and prompt identification plays a major role not only in treatment, but also prevents further invasion.

**Keywords:** phaeohyphomycosis, *Fonsecaea pedrosoi*, fungal rhinosinusitis

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## 1. Introduction

Fungi are uncommon causes of rhinosinusitis, but the incidence of these infections is increasing [1]. At 2009, Chakrabarti A et al [2] had given the most accepted system of classification of Fungal rhinosinusitis. It is mainly divided into invasive and noninvasive type based on tissue invasion by fungi. The invasive diseases include acute, granulomatous and chronic invasive fungal rhinosinusitis. The noninvasive diseases include fungal ball and allergic fungal rhinosinusitis.

Rhinosinusitis implicated by uncommon species of fungi is also an increasingly recognized entity, in immunocompromised as well as immunocompetent individuals. The most common etiological agents of fungal sinusitis is found to be *Aspergillus* species followed by *Candida* and *Mucor* [3]. Phaeohyphomycosis belong to the group of fungi that have a brown pigment due to the presence of melanin (dematiaceous fungi). Most of them belong to the order; *Pleosporales* (*Alternaria*, *Bipolaris*, *Curvularia*, *Exserohilum*) and *Chaetothyriales* (*Cladophialophora*, *Fonsecaea*, *Exophiala* and *Ramichloridium*) [4].

*Fonsecaea* is one of the agents causing phaeohyphomycosis. The genus was revised by de Hoog et al. [5] and Najafzadeh et al. [6] based on the ribosomal DNA internal transcribed spacer (ITS) sequence data recognizing two species; *F. pedrosoi* and *F. monophora*.

*Fonsecaea* spp. is one of the causative agents of the important agents of chromoblastomycosis, paranasal sinusitis, keratitis, and fatal brain abscesses following hematogenous dissemination [7].

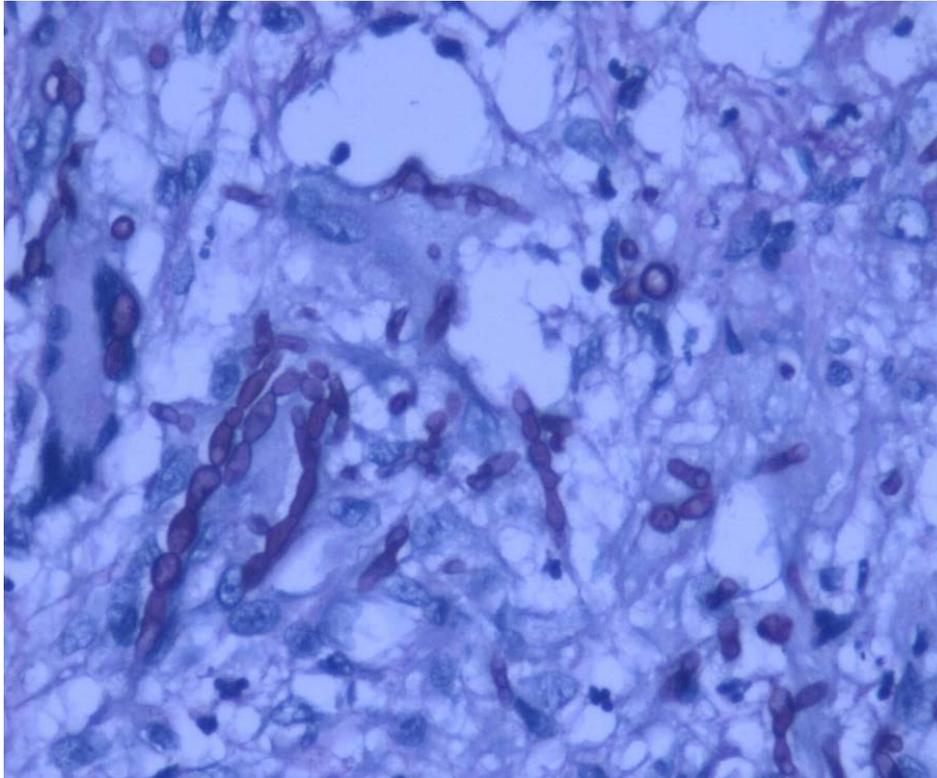
Phaeohyphomycosis causing rhinosinusitis is very rare, and phaeohyphomycosis caused by *Fonsecaea* has hardly been reported. Our case is one such due to *Fonsecaea pedrosoi* occurring in a diabetic male in whom early diagnosis, prompt treatment with the appropriate antifungal agent and surgical correction of the anomaly cured the patient.

## 2. Case Report

A 53 year old male came with complaints of nasal block on and off for the past 2 months, associated with headache. Nasal block more on left side associated with foul smelling nasal discharge. The patient was treated medically for the same symptoms one month back. Diagnostic nasal endoscopy done 10 days back showed deviated nasal septum towards the left with chronic rhinosinusitis. The patient is a known case of diabetes mellitus diagnosed two years back and on treatment. The patient was admitted and functional endoscopic sinus surgery with septoplasty was done. Polypoidal mass from maxillary antrum was sent for histopathological examination (HPE) and fungal culture. HPE showed chronic inflammatory reaction predominantly with lymphocytes, plasma cells and histiocytes. The special

stain done with Periodic Schiff Stain (PAS). showed darkly stained fungal hyphae which was branched and septate (Figure 1). Fungal elements suggested the possibility of phaeohyphomycosis. Inoculation of the material was done on Sabouraud's dextrose agar with chloramphenicol (50 mg/mL) into two tubes and each tube and one was incubated at 35°C and the other at 25°C. Macroscopically the colonies were floccose, velvety and olivaceous black in colour (Figure 2) Slide culture preparation was made to study the undisturbed

morphology. Lactophenol cotton blue mount showed olivaceous conidigenous cells, arranged in loosely branched systems, with bunch of prominent denticles. The shape of the conidia was clavate to ellipsoidal, in short chains, subhyaline, smooth & thin walled (Figure 3). The fungus was identified as *Fonsecaea pedrosoi*. The patient was treated with oral itraconazole 200 mg twice daily for 3 months. The patient's condition improved clinically and was discharged.



**Figure 1.** PAS stain showing darkly stained branched and septate hyphae (MagX40)



**Figure 2.** Colonies on SDA were floccose, velvety and olivaceous black in colour



**Figure 3.** Lactophenol cotton blue mount showing pale, olivaceous conidigenous cells, loosely arranged branches with cladosporium type of conidiation

### 3. Discussion

Fungal sinusitis can present in two forms: non invasive and invasive type. Non invasive includes *first*, allergic fungal sinusitis in which the histopathological examination shows inflammatory mucin containing hyphal elements and eosinophils within the mucin. *Second*, a fungal ball occupying the sinus cavity, which manifests as obstruction (there is no apparent fungal invasion into local soft tissue or bone in either of these two forms).-In case of invasive variety it would be extending into the host tissue such as bone and even the brain [2]. Although phaeohyphomycosis of the sinus is uncommon mainly implicated in allergic fungal sinusitis, its incidence seems to be on the rise, with reported invasive fungal infection rates of 1 per million persons per year [8].

There are quite a few reports on *F.pedrosoi* causing chromoblastomycosis and cerebral phaeohyphomycosis, however there has been a rarity of reports on sinusitis caused by *Fonsecaea pedrosoi* from India. *Fonsecaea pedrosoi* causing infection of the paranasal sinuses simulating a neoplasm has been reported in 1993 in India [9].

Structural abnormalities and chronic sinus conditions can predispose a person to be colonized by fungus. There could be a possibility of extension of this growth leading to the formation of a fungal ball. Rarely this may also become invasive after substantial immunosuppression [10]. Our patient is a case of chronic non invasive fungal maxillary sinusitis caused by *F.pedrosoi*. The patient had a deviated nasal septum and a known case of diabetes mellitus. These were the only apparent underlying disease conditions. He responded well to surgical correction and oral dose of itraconazole.

The genus *Fonsecaea* was first described by Negrini in 1936 to include all fungi that have two common types of

sporulation: 1)The *Cladosporium* type, where there are long chains of conidia. 2) The *Rhinochadiella* type where spore is formed at the end of the conidiophores or along its sides [11]. Additionally there is a third type which is known as the *Phialophora* type, where the conidia is formed at the end of the flask shaped conidiophore which is named as *Phialide*. This type is not considered as a major and stable characteristic of the genus *Fonsecaea*, since it is rarely produced by this fungi [7,8]. It is primarily based on all the above characteristics that *Fonsecaea* is differentiated from the genus *Rhinochadiella* and its closely related genus *Ramichloridium*. The latter two genera produce conidia sympodially and no blastoconidia are formed from the primaries. In addition, colonies of *Ramichloridium* are brown to olive green with shades of pink or orange color on reverse, and the conidia are hyaline to sub hyaline to pale brown. The colonies of the present isolate was black to grey-black with black reverse, and the fungi produced dark brown with spore heads loosely arranged with clavate conidia arranged in chains, similar to *Cladosporium* type of sporulation) [7,8] (Figure 3).

The isolate was identified as belonging to the species *Fonsecaea pedrosoi*. Confirmation of the isolate was done by gene sequencing. The internal transcribed region (ITS) was amplified and sequenced using ABI PRISM 3100 Genetic Analyzer (Applied Biosystems, USA). The sequence was then used for a nucleotide-nucleotide search using the BLAST algorithm at the NCBI website (<http://www.ncbi.nlm.nih.gov/BLAST/>). The BLAST hits more than 98% were considered. The identity was with *Fonsecaea pedrosoi*. The sequence was deposited in the GenBank database. The accession number is **KJ623701**. The culture was deposited in the culture collection at the CBS, Utrecht, Netherland. The accession number is **CBS 137216**.

## 4. Conclusion

It is important to keep in mind that fungal rhinosinusitis can be caused by a varied group of fungi from the environment. It is not only *Aspergillus* species which can cause this type of clinical infection. The microbiologists and the clinicians should be aware of such unusual fungi which can cause infection of the sinuses and should not be neglected as contaminants.

## Conflict of Interest

None.

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