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Case Report

Pan-sinusitis in an Immunocompetent Pediatric Patient with a History of Intranasal Cocaine Abuse

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Abstract

Pan-sinusitis due to dematiaceous (darkly pigmented) fungi appears to be increasing in frequency. The following report describes an immunocompetent pediatric female with a history of nasal congestion, rhinorrhea, severe headaches, and proptosis. Her social history included cocaine snorting and chronic use of intranasal steroid-containing sprays. A purulent, foul-smelling yellowish-green mucus and right ethmoid sinus tissue were submitted for bacterial and fungal cultures and pathology studies. The stains were positive for black-pigmented, septate hyphae and fungal cultures grew dematiaceous, furry-like colonies. Macroscopic and microscopic appearance of growth was consistent with the mold, *Curvularia*. Her sinuses contained sheets of eosinophils along with numerous Charcot leyden crystals. These findings along with the isolation and identification of *Curvularia,* were diagnostic for allergic fungal pan-sinusitis. Fungal sinusitis should be considered in the differential diagnosis of patients presenting with a similar history.

Keywords: Pediatric; Immunocompetent; Pansinusitis; Cocaine-Abuse; Dematiaceous Fungus

Introduction

Sinusitis, also called rhinosinusitis, is an inflammation of the mucosal lining of the nasal passages and paranasal sinuses. There are four sinuses connected to the nasal passages, i.e. frontal (behind the forehead), ethmoid (behind the nose), maxillary (behind the cheekbones), and sphenoid (behind the eyes). Symptom duration has been used to classify sinusitis: acute (< 4 weeks of symptoms) and subacute (between 4 and 12 weeks) that generally resolve completely; recurrent acute with infections that last < 30 days each, but recur after = > 10 days of resolution of each bout; and chronic (> = 12 weeks) [1,2]. Although most acute infections are caused by a virus, a small number are caused by bacteria often preceded by a viral infection. Various fungi cause chronic rhinosinusitis, most commonly due to *Aspergillus*. Allergic fungal sinusitis appears to be increasing in frequency secondary to infection with a dematiaceous or darkly pigmented fungus, notably *Bipolaris, Curvularia,* and *Exserohilum spp*. [3,4]. Although most cases of sinusitis are found in immunocompromised patients, e.g. diabetics, we cannot dismiss the possibility that immunocompetent patients also may develop fungal rhinosinusitis.

This report reviews the findings of a 15-year-old, immunocompetent female presented to a pediatric ENT service with a two-year history of nasal congestion, rhinorrhea, snoring, and a six-month history of severe headaches. Her social history included cocaine snorting and chronic use of decongestant sprays, some containing steroids. A physical examination was performed that revealed hypertelorism (abnormally increased distance between the eyes) and bilateral proptosis (protruding eyes). Examination with a nasal endoscope revealed numerous nasal polyps that can result from previous viral infections. Computed tomography (CT) and magnetic resonance imaging (MRI) scans are particularly useful for viewing the paranasal sinuses, diagnosing chronic fungal sinusitis, and determining the extent of infection [5]. Cultures, performed on foul-smelling, yellowish-green mucus and right ethmoid sinus tissue collected during surgery, grew a dematiaceous mold identified as *Curvularia* sp. and the patient was placed on antifungal treatment.

The aforementioned history and diagnosis in this pediatric patient represent a relatively uncommon cause of pan-sinusitis that appeared to be related to the use of intranasal cocaine. It is important to consider fungal sinusitis in the differential diagnosis of immunocompetent patients of any age with similar findings.

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Vital signs were taken, followed by a chest x-ray, CT scan and MRI. Hematology and chemistry tests included a complete blood count and liver function test, respectively, HIV screen and CSF tests. For anergy testing, a T and B lymphocyte functional assay was performed using peripheral blood mononuclear cells (PBMC) stimulated using specific Purified Protein Derivative (PPD) and *Candida albicans* and non-specific phytohemagglutinin, concanavalin A and pokeweed mitogens. Serum concentrations of IgG, IgM, IgA, IgE and IgG subclasses were also determined. Flow cytometry was performed to identify any abnormalities related to the quantity of lymphocytes, B-cells, T-helper and T-suppressor cells. A purulent, foul-smelling, yellowish-green mucus and right ethmoid sinus tissue were submitted to the Microbiology Laboratory for bacterial and fungal cultures and inoculated onto Columbia blood agar (BA), chocolate, CNA, MacConkey, and PEA media incubated aerobically at 35°C in 5% CO_2 and PRAS thioglycolate broth, BA, chocolate, KV, and ANA media incubated anaerobically in BBL GasPak jars. Fungal media included Sabouraud dextrose agar (SDA) and Sabhi agar slants with blood and chloramphenicol incubated at 30°C. Dematiaceous colonies were sub-cultured to potato dextrose agar (PDA) and PDA plus cornmeal agar (CMA) plates. Gram and Calcofluor white stains of the ethmoid sinus mucus, PAS, Gomori methenamine silver stains of the tissue, and lactophenol cotton blue (LPCB) stain of growth from BA and PDA subcultures from Sabhi medium were performed for identification of the isolate. The patient was initially treated with amphotericin B, but due to toxicity was changed to miconazole for 8 weeks and discharged on oral itraconazole for 71 days. CT scans were used to monitor improvement.

Results and Discussion

Exploring this young female's past medical history, she was physically abused two years prior to this current admission and sustained injury to her right eye with temporary loss of vision. She received a medical evaluation that included a CT scan of the face to rule out fractures. Although the scan was negative for fractures, it did reveal maxillary and paranasal sinusitis. The patient was asked to follow up with her physician, although she was not compliant. During this current visit, vital signs, chest x-ray, and routine chemistry, hematology, CSF tests, and all concentrations of serum immunoglobulins and IgG subclasses were within normal limits. The PPD and *Candida* control were non-reactive, and the patient had good responses to all mitogens in the lymphocyte functional assay. Although there was a mild absolute lymphopenia with relative granulocytosis and increase in B cells, there were no abnormalities in the T-helper and T-suppressor cells, or evidence of immunodeficiency noted by flow cytometry. Her HIV screen was negative.

A CT scan was consistent with pan-sinusitis with extension of a hyperdense mass in the ethmoid, maxillary, and frontal sinuses. Opacification was observed in the sphenoid sinus. Purulent mucus and sinus tissue submitted to the Microbiology Laboratory for cultures were positive at 48 hours. Mold colonies were observed on routine BA and chocolate media incubated aerobically and on the fungal media. All other inoculated media were negative for mold.

Gram and the Calcofluor white stains of the mucus and tissue showed moderate leukocytes and hyphal elements. PAS and Gomori methenamine silver stains of the tissue were also positive for septate hyphae. Colonies on SDA were described as mousey grey, black-pigmented, with a furry appearance. LPCB stained preparations of growth on BA and the PDA subcultures demonstrated dark, septate hyphae with large 3 - 4 cell, dark poroconidia borne on conidiophores bent at points where the poroconidia originated. This microscopic morphology is referred to as geniculate conidiophores. The central cells of the conidia were darker and enlarged compared to the end cells, giving the conidia a curved appearance. Based on the macroscopic appearance of the colonies and microscopic description, the mold was identified as *Curvularia* species.

Both sinuses contained laminated masses of pale eosinophilic mucus containing sheets of eosinophils. Many of these were degranulated, and there were numerous Charcot-Leyden crystals present. These findings along with the observation of fungal hyphae were diagnostic for allergic fungal sinusitis. The patient received a total of three endoscopic debridements. She was started on amphotericin B, but on the third day the patient developed signs of nephrotoxicity with an increased BUN, creatinine and decreased potassium and amphotericin B was discontinued for several days to see if the serum chemistries would normalize. Since the BUN and creatinine remained elevated, intravenous miconazole was given for a total of eight weeks. During treatment, the patient was fol-

lowed by CT scans which showed improvement of all sinuses and a decrease in the size of the mass. She was then given oral itraconazole for a total of 71 days and was completely asymptomatic.

Curvularia is a dematiaceous fungus with worldwide distribution. This fungal hyphomycete characteristically demonstrates branched, septate mycelium and is commonly found on submerged decaying leaves and other organic matter. There is great diversity of species within the genus *Curvularia* that can be delineated by microscopic morphology and sequence analysis [6]. Most infections are acquired by direct inoculation or inhalation [7]. In the past, fungal sinusitis was thought to be primarily a disease found in immunocompromised patients, but more recently there have been reports of dematiaceous molds including *Curvularia* that can cause invasive subcutaneous or systemic disease [8,9] or localized infections in young healthy immunocompetent patients as described in this case [10]. Sinusitis due to dematiaceous melanin-containing molds has been referred to as phaeohyphomycosis [11]. Melanin is likely a virulence factor for these fungi [12]. Katzenstein., *et al*. [13] reported in 1983, seven cases of what was then a newly recognized form of chronic sinusitis in young adults described as allergic *Aspergillus* sinusitis. These patients presented with nasal polyps and radiographic evidence of pan-sinusitis. Histologic findings showed mucin containing eosinophils, Charcot-Leyden crystals derived from necrotic eosinophils, and fungal hyphae. That said, Rinaldi., *et al*. in 1987 documented *Curvularia* with similar pathophysiology of allergic sinusitis [14], similar to the findings in this report.

In this report, the patient had a history of cocaine and steroid decongestant use which may have contributed to paranasal sinusitis. The street form of cocaine leads to vasoconstriction and local irritation of the thin respiratory epithelium of the nasal airway. Repeated snorting can cause ischemia, inflammation, micronecrosis, infection and possibly micronecrosis leading to perforation [15-17]. Unlike some patients with chronic cocaine snorting that develop nasal septum perforations of cartilage and bone tissues, deformities of the nose and erosion of the facial anatomy with recurrent sinus infections, the case described here was fortunate in having a localized infection, treated successfully with debridement and long term antifungal drugs.

Conclusion

This case highlights the importance of considering fungal sinusitis in immunocompetent pediatric patients related to risk factors. The patient herein described was a young, immunocompetent female with a past history of physical abuse to the head, use of intranasal cocaine and steroid-containing decongestant sprays, with an incidental finding of maxillary and paranasal sinusitis by CT scan that was used to rule out fractures of the face. Her current visit confirmed the presence of a sinus mass by CT scan, followed by tests to evaluate both humoral and cellular Immunity that confirmed immunological competence. Microbiology and pathology studies discovered a black-pigmented mold with septate hyphae that was isolated in cultures and identified as *Curvularia*. Eosinophils and Charcot-Leyden crystals were seen in both sinuses, showing evidence of an allergic response to infection. This finding together with the pathology and culture results supported a diagnosis of allergic fungal sinusitis.

Conflict of Interest

The author has no financial or conflicts of interest to disclose.

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