



Chronic Invasive Fungal Sinusitis Associated with Intranasal Drug Use

Kelly Pekala M.S., Matthew Clavenna M.D., Ross Shockley M.D., Justin H. Turner M.D. Ph.D.
Department of Otolaryngology-Head and Neck Surgery; Vanderbilt University School of Medicine; Nashville, TN

Abstract

Chronic invasive fungal sinusitis (CIFS) is a rare but potentially aggressive form of invasive fungal disease that occurs in immunocompetent patients. We report a case of CIFS in an otherwise healthy young adult associated with intranasal illicit drug abuse. Tissue necrosis and ulceration related to intranasal drug should be recognized as a potential risk factor for invasive fungal sinusitis.

Introduction

Invasive fungal sinusitis is an increasingly prevalent, but still rare infectious disease heralded by progressive tissue destruction, angioinvasion, and generally poor outcomes¹. Unlike acute invasive fungal sinusitis (AIFS), which usually occurs in immunocompromised hosts, CIFS typically presents in otherwise healthy individuals. The disease often progresses slowly over weeks or months, and a lack of obvious predisposing factors can present a diagnostic dilemma and delay needed surgical or medical interventions. Left untreated, CIFS, like its acute counterpart, can result in proptosis, altered mental status, seizures, and intracranial complications^{2,3}. The causative organisms in CIFS are saprophytic fungi such as the Zygomycetes (*Mucor*, *Rhizopus*, *Rhizomucor*) and multiple species of *Aspergillus*. Herein, we describe the case of an otherwise healthy, young male who developed CIFS related to a nasal septal and palatal perforation and prior intranasal cocaine use.

Case Report

A 24-year-old Caucasian male presented with intractable nasal and palate pain, and a history of progressive nasal congestion and rhinorrhea for the past three months. He also complained of worsening dysphagia, dysarthria, and an approximately thirty-pound weight loss. He reported a history of two small palatal erosions that initially developed two years earlier in the setting of intranasal cocaine abuse. Examination revealed a saddle nose deformity, nasal crusting and rhinorrhea. Nasal endoscopy demonstrated heavy crusting, necrotic areas of mucosa, absent inferior turbinates, and erosion of the middle turbinates and maxillary sinus cavities. The erosive process extended through the hard palate with almost complete deficiency of the soft palate. Computed tomography (CT) imaging mirrored the physical exam findings, showing an extensive erosive process involving the palate, nasal cavity, and maxillary sinus without obvious intracranial pathology (Figure 1). An MRI confirmed a lack of intracranial disease, and again showed extensive tissue loss involving the nasal septum, palate, and paranasal sinuses (Figure 2).

A bedside biopsy of the palate and nasal septum was performed due to the patient's worsening symptoms and physical exam findings. Initial pathology was concerning for fungal invasion and the patient was subsequently taken to the operating room for a formal endoscopic exam and surgical debridement. This revealed partial absence of the nasal floor due to the large palatal perforation with the endotracheal tube easily visualized intranasally (Figure 3A) Nonviable tissue and diseased mucosa was removed along the nasal floor, nasal septum, and lateral nasal walls until viable, bleeding tissue was encountered (Figure 3B). Histopathology from the intraoperative debridement and tissue biopsy revealed numerous fungal hyphae within the mucosa, submucosa, and vascular lumen of bone (Figure 4). Subsequent culture and speciation identified the fungus as *Aspergillus flavus*. Postoperatively, intravenous amphotericin B was initiated, but this was subsequently changed to voriconazole after fungal speciation.

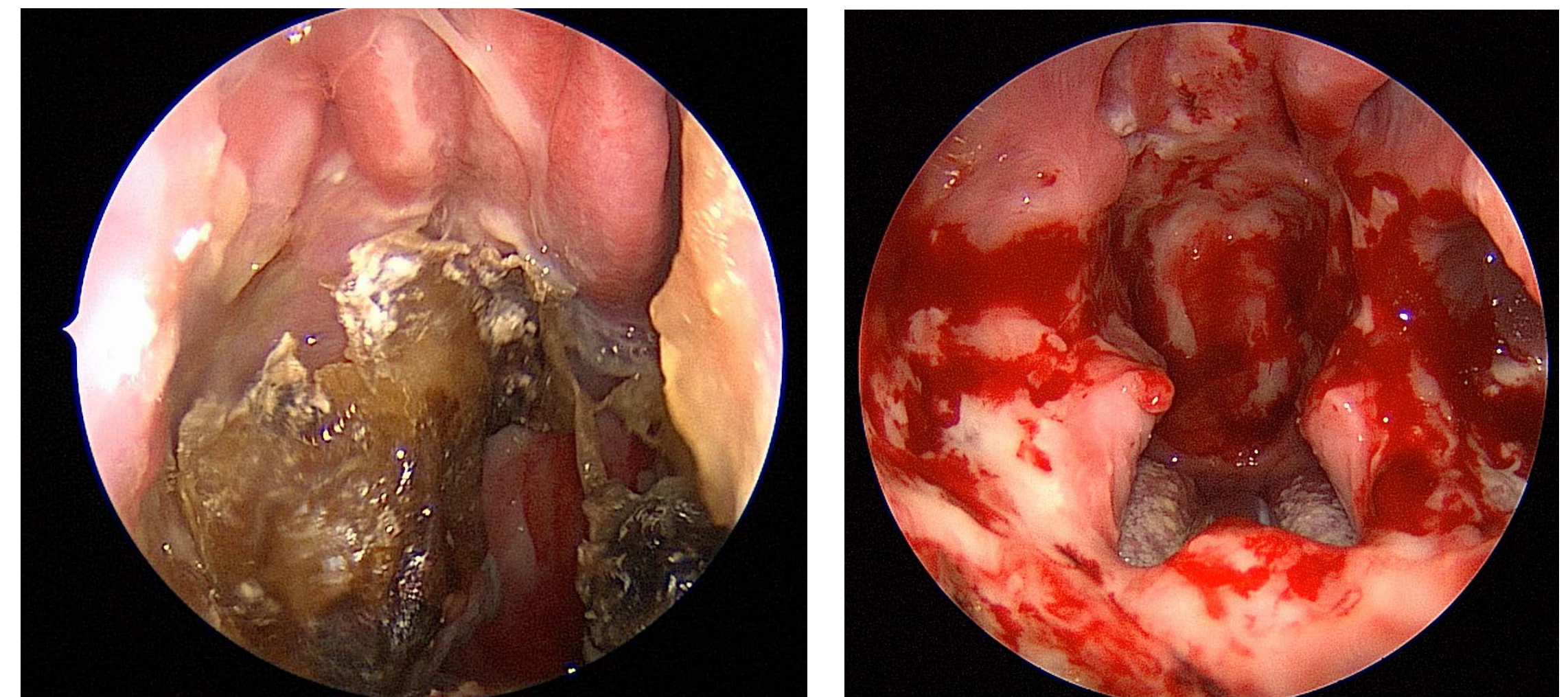


Figure 3. Intraoperative and postoperative endoscopy

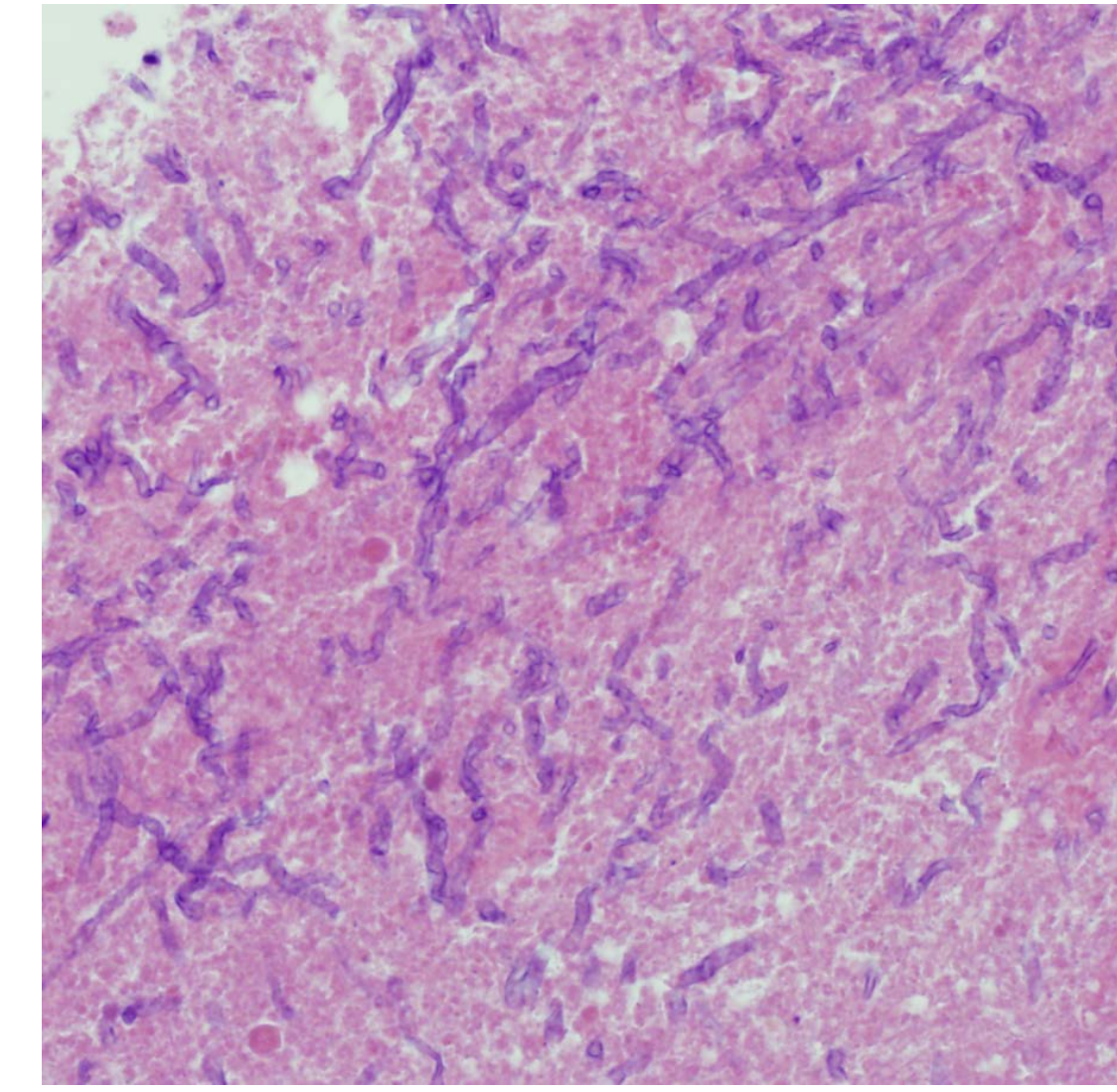


Figure 4. H&E stain of palatal biopsy



Figure 1. Coronal Face CT



Figure 2. Sagittal MRI Head

Discussion

CIFS is a rare clinical entity that primarily affects immunocompetent individuals and which presents challenges both in diagnosis and treatment. Previous case reports have associated CIFS with use of topical nasal steroids and chronic sinonasal disease^{2,4}, however, there is a lack of clear consensus regarding putative predisposing factors. Our patient presented with a large septal and palatal perforation and adjacent CIFS due to cocaine abuse. To our knowledge, this represents the first report of invasive fungal sinusitis associated with the intranasal use of illicit drugs.

Intranasal use of cocaine is associated with sinonasal tissue ischemia, palatal and septal perforation, and midline destructive lesions^{7,8}. Chronic use can result in progressive sinonasal symptoms and major complications that include orbital neuropathies, osteomyelitis, and intracranial abscess⁹⁻¹¹. The exact etiology of such complications is unclear but may be secondary to cocaine-induced vasoconstriction and severe mucosal inflammation that ultimately results in ulceration and tissue necrosis.

Diagnosis of CIFS, as outlined by deShazo et. al., requires 1) radiologic evidence of chronic rhinosinusitis, and 2) histopathological evidence of fungal hyphae within the sinonasal mucosa, submucosa, blood vessels or bone¹². However, findings on CT or MRI are often nonspecific and timely diagnosis of CIFS is consequently quite difficult. This is evident in the current report, as our patient was evaluated at multiple medical institutions over a three month period without a diagnosis. The slowly progressive symptoms and destructive process identified on endoscopy ultimately heightened clinical suspicion and led to an accurate diagnosis and prompt treatment.

The current report suggests that patients presenting with non-healing sinonasal ulcerations or necrotic septal or palatal perforations may be at increased risk of developing CIFS. A high index of suspicion for invasive fungal disease in this patient population may lead to earlier diagnosis and more prompt surgical and medical management.

Conclusions

Chronic invasive fungal sinusitis is an indolent, but potentially aggressive infection that typically occurs in immunocompetent individuals. Causative fungal organisms, such as *Aspergillus*, preferentially colonize decomposing or necrotic tissue, and in this case inhabited an ulcerated palatal and septal defect related to long-term intranasal cocaine abuse. Patients with mucosal ulceration or necrosis due to intranasal drug use may be at an increased risk of invasive fungal sinusitis.

Contact

Justin H. Turner, MD, PhD
Vanderbilt University School of Medicine
Department of Otolaryngology-Head and Neck Surgery
Justin.h.turner@vanderbilt.edu

References

1. Turner, J. H., Soudry, E., Nayak, J. V. & Hwang, P. H. Survival outcomes in acute invasive fungal sinusitis: a systematic review and quantitative synthesis of published evidence. *Laryngoscope* 2013; 123:1112-1118.
2. Vakharia, K. T., Durand, M. L., Hamilos, D. L., Geyer, J. T. & Holbrook, E. H. An atypical case of chronic invasive fungal sinusitis and its management. *Otolaryngol Head Neck Surg* 2010; 142:150-151.
3. Thurtell, M. J. et al. Neuro-ophthalmology of invasive fungal sinusitis: 14 consecutive patients and a review of the literature. *Clin. Experiment. Ophthalmol.* 2013; 41:567-576.
4. Stringer, S. P. & Ryan, M. W. Chronic invasive fungal rhinosinusitis. *Otolaryngol. Clin. North Am.* 2000; 33:375-387.
5. Deshazo, R. D. Syndromes of invasive fungal sinusitis. *Med. Mycol.* 2009; 47:Suppl 1, S309-314.
6. Thompson, G. R. & Patterson, T. F. Fungal disease of the nose and paranasal sinuses. *J. Allergy Clin. Immunol.* 2012; 129:321-326.
7. Trimarchi, M. et al. Sinonasal osteocartilaginous necrosis in cocaine abusers: experience in 25 patients. *Am J Rhinol* 2003; 17:33-43.
8. Sercarz, J. A., Strasnick, B., Newman, A. & Dodd, L. G. Midline nasal destruction in cocaine abusers. *Otolaryngol Head Neck Surg* 1991; 105:694-701.
9. Goldberg, R. A. et al. Orbital inflammation and optic neuropathies associated with chronic sinusitis of intranasal cocaine abuse. Possible role of contiguous inflammation. *Arch. Ophthalmol.* 1989; 107:831-835.
10. Kurloff, D. B. & Kimmelman, C. P. Osteocartilaginous necrosis of the sinonasal tract following cocaine abuse. *Laryngoscope* 1989; 99:918-924.
11. Rao, A. N. Brain abscess: a complication of cocaine inhalation. *N Y State J Med* 1988; 88:548-550.
12. DeShazo, R. D. et al. A new classification and diagnostic criteria for invasive fungal sinusitis. *Arch. Otolaryngol. Head Neck Surg.* 1997; 123:1181-1188.