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Pediatric invasive fungal rhinosinusitis: An investigation of 17 patients

Daniel Vinh, MD [a, *], Michael Yim, MD [a], Ankhi Dutta, MBBS, MPH [a, b], John K. Jones, MBBS [a, b], Wei Zhang, PhD [c], Matthew Sitton, MD [a, b]

[a] Baylor College of Medicine Department of Otolaryngology-Head & Neck Surgery, Houston, TX, USA
[b] Texas Children's Hospital Department of Otolaryngology-Head & Neck Surgery, Houston, TX, USA
[c] Texas Children's Hospital Department of Statistics, Houston, TX, USA

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ABSTRACT

Purpose: To investigate outcomes of pediatric patients at a single institution with invasive fungal rhinosinusitis (IFRS) and to determine variables that impact overall survival.
Methods: All pediatric patients at a large tertiary children's hospital diagnosed with IFRS confirmed by surgical pathology from 2009 to 2015 were retrospectively reviewed. Demographics, underlying diseases, symptoms, antifungal therapy, absolute neutrophil count (ANC), surgical management,and outcomes were analyzed.
Results: Seventeen patients were identified with IFRS with an average age of 8.7 years and 53% male. Hematologic malignancy was the most common (n = 13) underlying disease. The most common presenting symptoms were fever (82%) and congestion (41%). 15 patients had severe neutropenia (Absolute Neutrophil Count (ANC) < 500) within 2 weeks prior to diagnosis. The average ANC at time of diagnosis was 1420 cells/uL. 16 patients were treated with serial nasal endoscopy and debridement, while 1 patient was treated with an open approach. 16 received combination antifungals while 1 was treated with amphotericin monotherapy. The most common genus cultured was Fusarium (n = 6). The average number of surgical interventions was 3.4, with the average interval between interventions 6.2 days. 13 of 17 (76%) were cleared of IFRS. Overall survival at 6 months was 41%.
Conclusion: Pediatric IFRS is a life-threatening disease that requires a coordinated surgical and medical approach. Despite a relatively high local control rate, overall mortality remains disappointingly high, reflecting the disease's underlying pathogenesis - lack of host defense and risk of disseminated fungal infection. Further investigation is necessary to reveal optimal management with regards to antifungal therapy, surgery, and utility of labs.

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1. Introduction

Acute invasive fungal rhinosinusitis (IFRS) is an aggressive, often fatal submucosal infiltration of fungal organisms in the nasal cavity and paranasal sinuses. If left untreated it will spread to involve orbital or intracranial structures. The disease occurs in the immunocompromised host, such as those patients with hematologic malignancies undergoing chemotherapy, chronic immunosuppressive drug therapies [1], or poorly controlled diabetes mellitus [2]. Epidemiological data within the pediatric population is lacking. Among the adult population, however, incidence is estimated at 1.77 per 1000 patients who have underlying hematological disorders [3]. The initial presentation is non-specific, typically with persistent fevers, congestion, and rhinorrhea in an immunocompromised child.

When IFRS is suspected, prompt workup must be initiated to prevent the disease's aggressive and potentially lethal dissemination [4]. Rigid nasal endoscopy may reveal crust formation, granulation tissue, or frank necrosis of mucosa with a black, grey, white, or green discoloration, most commonly identified on the head of the middle turbinate [5]. An example nasal endoscopy is shown in Fig. 1. While this examination may be useful for diagnosis, its utility is limited as it can only visualize a portion of the entire nasal cavity and such a procedure may be technically difficult in an uncooperative child. CT and MRI imaging of the sinuses may also aid in the diagnosis, with some data indicating that MRI may be more sensitive than CT for the diagnosis of IFRS (85% and 86% among two

* Corresponding author.
E-mail address: daniel.vinh@bcm.edu (D. Vinh).

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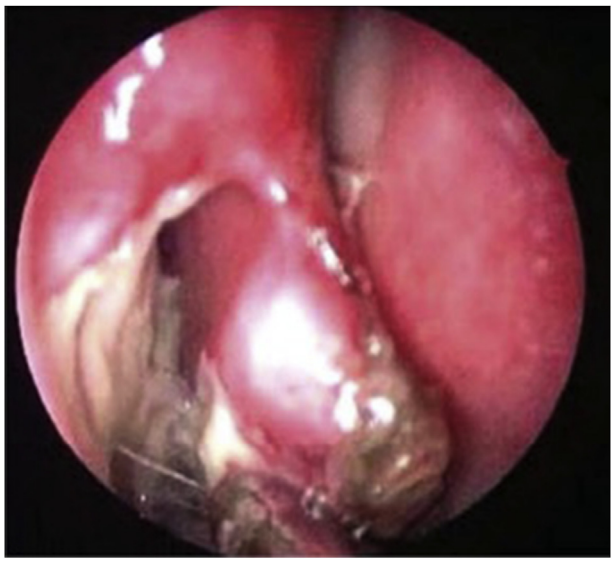

Fig. 1. Representative gross appearance of invasive fungal disease involving the middle turbinate.

reviewer's results compared to 57% and 69%) [6]. CT imaging may show bony invasion or unilateral sinus opacification and soft tissue thickening of the mucosa along the nasal floor and lateral wall, while MRI findings may include obliteration of periantral fat planes and lack of mucosal Gadolinum contrast enhancement [6–8]. These features, however, are typically only seen in the late stages of the disease. The gold standard for diagnosis is biopsy, demonstrating invasion of fungal organisms into the submucosa with or without angioinvasion. The most commonly reported organisms reported are Aspergillus, Zygomycetes (mucor), and Fusarium [2,9,10] (see Table 1).

Once diagnosed, prompt treatment must be started which includes a combination of surgical debridement, systemic antifungals, and restoration of host immune function [5,9]. Despite these aggressive interventions, the reported overall mortality remains disappointingly high at 50–80% [5,9,10]. Due to lack of IFRS data in the pediatric population, more studies are needed to evaluate factors in management that lead to improved survival. In this study, we investigated the demographics, presentation, surgical management, and outcomes of pediatric patients treated at a large tertiary children's hospital diagnosed with invasive fungal rhinosinusitis (IFRS) with the goal of identifying variables that may impact survival.

2. Methods

After institutional review board approval was obtained, an internal database of all inpatient otolaryngology consult patients treated at Texas Children's Hospital in Houston, TX was reviewed from 2009 to 2015 using the search terms “invasive fungal sinusitis” and “IFRS.” Inclusion criteria for the study included patients treated at Texas Children's Hospital, having an underlying hematologic disorder, and having biopsy-proven invasive fungal disease by surgical pathology. A total of 18 patients met inclusion criteria. Exclusion criteria included those with invasive fungal disease of non-hematologic disorder etiology such as diabetic ketoacidosis in order to use absolute neutrophil count (ANC) as a quantitative proxy for overall innate immune function. A total of 17 patients met inclusion and exclusion criteria. Data collected included patient demographics, presenting symptoms, imaging studies, underlying disease, treatment course, ANC, survival status after diagnosis, and cause of death. Overall survival was calculated from death of any cause following IFRS diagnosis at 6 months and 12 months. Kaplan-Meier survival curves were generated and stratified by variables of interest and compared using a Wilcox log-rank test.

Table 1
Demographics, underlying hematologic disorders, and presenting symptoms of pediatric IFRS patients.

Patient Demographics	
Age	9.66 ± 6.19 years
No. F (percentage)	9 (53)
Underlying Hematologic Disorder	
AML	n = 4
ALL	n = 6
Aplastic anemia	n = 3
Histiocytosis	n = 2
Hodgkin's Lymphoma	n = 1
Infantile leukemia	n = 1
Fungi isolated	
Fusarium	n = 6
Zygomycetes (Mucor)	n = 4
Curvularia	n = 4
Exherosilum	n = 3
Aspergillus	n = 2
Bipolaris	n = 2
Curvularia	n = 2
Candida	n = 2
Cladosporium	n = 1
Alternaria	n = 1
Most common presenting symptom	
Fever	82%
Headache	41%
Congestion	41%
Rhinorrhea	35%
Facial Pain	18%
Epistaxis	12%
Vision Changes	6%
Ophthalmoplegia	6%
Proptosis	6%

3. Results

Patient demographics and presenting symptoms collected from 2009 to 2015 are shown in Table 1. The average age of 17 patients was 9.7 years (median 11, range 0.58–19). There were 9 females and 8 males. The most common underlying hematologic disorder was acute lymphocytic leukemia (ALL, n = 6), followed by acute myelogenous leukemia (AML, n = 4). Other disorders included aplastic anemia (n = 3), langerhan's histiocytosis (n = 2), Hodgkin's lymphoma (n = 1), and infantile leukemia (n = 1). Seven (41%) patients had received a bone marrow transplant prior to diagnosis. The most common presenting symptoms were fever (82%), headache (41%), and congestion (41%). 8 patients had a CT scan with contrast of the nasal cavity and paranasal sinuses prior to surgical intervention with 3 of 8 (37.5%) reporting findings suggestive of invasive fungal sinusitis. 4 patients had MRI with contrast prior to surgical evaluation with 3 of 4 (75%) reporting findings suggestive of invasive fungal disease. 5 patients did not have pre-surgical imaging or did not have accessible imaging results. 16 of 17 patients underwent serial endoscopic debridement while 1 patient was debrided with an open approach. The average total number of surgeries was 3.35 (range 1–7), while the average interval between surgeries was 6.21 days (range 0–25.7). The most common fungi

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