Candida kefyr in Invasive Paranasal Sinusitis

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Abstract Fungi are being increasingly implicated in etiology of rhino sinusitis. Invasive fungal sinusitis occurring in diabetics and immunocompromised patients is notorious for its insidious onset, rapid intra cranial spread and tissue destruction. *Candida* as a cause of this invasive infection is infrequently reported in India. We report a rare species of *Candida*, *Candida kefyr* in a female diabetic patient presenting with invasive fungal sinusitis.

Keywords Invasive fungal sinusitis · Diabetic · *Candida kefyr*

Introduction

Fungal sinusitis is the inflammation of sinus mucosa caused by a wide variety of fungi. *Aspergillus* is the most common and *Rhizopus*, *Mucor*, *Cladosporium*, *Candida* are amongst the others [1, 2].

According to WHO, there will be epidemic of diabetes world over and India is going to be 'A diabetic capital of world' by 2025. With the increased incidence of diabetes, the associated complications are also bound to increase and opportunistic fulminant fungal infection is one of them.

Incidence of mycotic infection and diversity of pathogenic fungi have increased dramatically in recent years. Fungal disease of paranasal sinuses are assigned in invasive and non invasive forms. There are three forms of non

invasive fungal rhino sinusitis; superficial sinonasal mycosis, allergic fungal rhinosinusitis and fungal ball.

Invasive fungal rhino sinusitis (IFRS) has following sub groups. Acute or fulminant, invasive and chronic invasive. Patient symptoms with IFRS include nasal stuffiness, nasal discharge, facial pain, fever and headache. Since mortality rates are very high with intra cranial disease an early diagnosis and treatment is the best option. With improved diagnostic tools and effective treatment strategies the prognosis has improved in this otherwise fatal infection [3].

The diagnosis is made on basis of histological ground which shows invasion of surrounding tissues and or vessels. This often occurs in patients with diabetes having keto acidosis. Culture is required to identify the fungal species. Treatment preferred in such invasive disease is the surgical debridement of non viable tissue along with systemic administration of anti fungal drugs [4, 5].

Case Report

A 47 year old female patient resident of Hyderabad presented with headache for 6 months, pain and mild protrusion of left eye for 20 days and mucopurulent discharge left upper eye-lid for 5 days. There was no diplopia, or diminished vision. Patient first went to ophthalmology OPD and was referred to ENT OPD. There was no history of prolonged antibiotic treatment, allergy, steroids or asthma. Patient was a known diabetic and hypertensive for 6 years on irregular treatment (Figs. 1).

On examination, swelling over left eye-lid, preseptal cellulitis with mucopurulent discharging sinus was seen. Local examination of nose revealed polypoidal mass in left middle turbinate with mucopurulent discharge. Provisional diagnosis of left sided fungal frontal sinusitis extending

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into left orbit was made AND investigated further after admission. Patient underwent routine hematological investigations and except for blood glucose levels (FBS—250 mg/dl), all other investigations were within normal limits. She was HIV and HBsAg seronegative. X-ray of paranasal sinuses showed haziness of left frontal, ethmoidal and bilateral maxillary sinuses. CT Scan revealed complete opacification of left frontal sinus with air spaces characteristic of fungal etiology, bilateral anterior ethmoiditis, and bilat maxillary sinusitis. Also resorption of medial wall of orbit with intra cranial and intra orbital extension was noted (Figs. 2, 3, 4).

Pus from discharging sinus of left upper eyelid was collected with sterile swab. It was non foul smelling, mucopurulent discharge. On gram stain there were plenty of pus cells. 10% KOH mount was negative for fungal elements. Culture for bacteria on Blood agar and Mac-Conkey agar showed no growth but on plain Sabauraud's Dextrose Agar, yeast like colony was observed after 48 h of incubation (Figs. 5, 6).

Yeast was subjected to germ tube test, chlamyospore formation and identified using fungifast—a commercial

preparation of sugar assimilation test for yeast. Fungifast showed positive assimilation of cellobiose and raffinose and negative for trehalose and maltose. *Candida kefyr* was confirmed (Fig. 7). Patient was put on Amphotericin B and showed remarkable improvement with reduction in mucopurulent discharge and proptosis.

Discussion

Paranasal sinus mycosis is being recognized more frequently in different parts of the world because of increased awareness (the use of unwarranted antibiotics, antihistamines, and corticosteroids in treatment of nasal allergy and sinusitis may play role in pathogenesis of disease by suppressing bacterial flora and supporting fungal overgrowth).

Patients with non invasive and invasive disease present differently. The non invasive form presents with chronic sinusitis unresponsive to conservative medical treatment

Fig. 1 Patient presenting with proptosis and swollen eye lid

Fig. 3 Bilateral anterior ethmoiditis and bilateral maxillary sinusitis

Fig. 2 Complete opacification of left frontal sinus with air spaces characteristic of fungal etiology

Fig. 4 Resorption of medial wall of orbit with intra cranial and intra orbital extension

and repeated sinus irrigation, and symptoms of rhinorrhoea and nasal obstruction. In contrast, patients with invasive disease had a firm enlarging mass in the cheek, the orbit and paranasal sinuses. It is unclear why a particular fungus behaves in different ways in non-immunocompromised patients resulting in non invasive and invasive types of fungal sinusitis [6].

Non invasive: these generally don't invade bone or tissues and more often are a result of hypersensitivity skin reactions. But a long-term disease can eventually erode the bone by pressure necrosis and hence cause an intracranial or intra orbital complication. It occurs in immunocompetent host and is characterized by presence of allergic mucin, charcot leyden crystals, eosinophils etc.

Invasive: the more fatal variety, known to penetrate the mucosa and cause tissue destruction and lead to intracranial extension. These are known to occur in immunocompro-

Fig. 5 *Candida* on Sabaraud's dextrose agar

Fig. 6 Gram positive budding yeast cells with pseudohyphae

Fig. 7 Fungifast—commercial preparation of sugar assimilation test for yeast

mised hosts in 50% of the cases. Often in diabetic patients having ketoacidosis.

Intracranial extension is one of the most dreaded complications of fungal sinusitis with high mortality rates. The fungus generally spreads through direct extension, hematogenous route, perineural invasion of cranial nerves, cribriform plate of ethmoid bone, rarely—surgery or blood transfusion.

A good history along with symptoms of any systemic ailments should be elicited. A sinusitis patient not responding to conventional antibiotics should arouse suspicion. Diagnostic nasal endoscopy should be performed and one should look for allergic mucin, blackish/brownish discharge, erosion of palate, pale/dark nasal mucosa etc. Fungal culture and biopsy are the mainstay in diagnosis. Culture used is sabaraud's media. Non contrast CT scan in early stages resemble chronic rhino sinusitis showing mucosal thickening which is hypo attenuated. In chronic cases a hyper attenuating soft tissue collection can be seen in one or more sinuses. It often gives a mass like appearance resembling a malignancy with destruction and erosion of bony walls of sinuses and extension into surrounding tissues.

Incidence of mycotic infections and the diversity of pathogenic fungi have increased dramatically in recent years. In most of the series, *Aspergillus* sps. have been found as the commonest fungus causing fungal rhino sinusitis. Apart from that other isolates would be *Candida* sps., *Curvularia*, *Dreschlera* sps., and *Bipolaris* sps.

Candida is an uncommon cause of invasive paranasal sinusitis, when this yeast like fungus is implicated as the causative agent *C. albicans* is the commonest species isolated. In this case we report a very rare species *C. kefyr* as the etiological agent of invasive paranasal sinusitis (Table 1).

Conclusion

Aspergillus species has been the most common isolate in fungal sinusitis. *Candida* species accounts for 1–2% of isolates. Reports of non-albicans *Candida* in fungal sinusitis is rare. Species reported include *C. tropicalis*, and *C. krusei*.

Table 1 Species of *Candida* isolated from paranasal sinusitis

Author/reference	Year	Total isolates	No. of <i>Candida</i>	Hospital/university
Panda et al. [6]	1998	178	<i>C. albicans</i> 1	PGI, Chandigarh
Chakraborti and Sharma [7]	2000	176	<i>C. albicans</i> 1	PGI, Chandigarh
Goh et al. [10]	2005	30	<i>C. albicans</i> 1	National University of Malaysia
Bassiri Jahromi and Khaksar [2]	2006	18	<i>C. albicans</i> 3 <i>Candida</i> sps. 2	Pasteur institute of Iran
Joshi et al. [8]	2007	14	<i>Candida</i> sps. 4	Koirala Institute of Health Sciences, Nepal
Present case	2009	1	<i>C. kefyr</i>	Gandhi Medical College & Hospital, Hyderabad, India.
Soontrapa et al. [9]	2010	87	<i>C. albicans</i> 2 <i>C. tropicalis</i> 1 <i>C. krusei</i> 1	Ramathibodi Hospital, Thailand

To the best of our knowledge this is the first report of *C. kefyr* implicated in invasive fungal sinusitis.

This patient represents an unusual case of chronic invasive fungal pansinusitis with intracranial and intraorbital extension caused by yeast which is rarely been implicated as an etiological agent.

It is a distinct entity in terms of clinical cause and treatment compared with non-invasive fungal sinusitis. It needs to be treated aggressively with surgical debridement and post-op systemic antifungals.

References

- Shah NJ, Rathore A (2009) Intracranial extension of fungal sinusitis. *Otorhinolaryngol Clin Int J* 1(1):55–61
- Bassiri Jahromi S, Khaksar AA (2006) Paranasal sinus mycosis in suspected fungal sinusitis. *Iranian J Clin Infect Dis* 1(1):25–29
- Michael RC, Michael JS et al (2008) Mycological profile of fungal sinusitis: an audit of specimens over a 7-year period in a tertiary care hospital in Tamil Nadu. *Indian J Pathol Microbiol* 51:493–496
- Challa S, Uppin SG et al (2010) Fungal rhino sinusitis: a clinicopathological study from South India. *Eur Arch Otorhinolaryngol* 267:1239–1245
- Venkatachalam VP, Anand N (2007) Paranasal mucormycosis: unusual presentation in otherwise healthy child. *Indian J Otolaryngol Head Neck Surg* 59:254–266
- Panda NK, Sharma SC et al (1998) Paranasal sinus mycoses in north India. *Mycoses* 41:281–286
- Chakraborti A, Sharma SC (2000) Paranasal sinus mycoses. *Indian J Chest Dis Allied Sci* 42:293–304
- Joshi RR, Bhandary S, Khanal B, Singh RK (2007) Fungal maxillary sinusitis: a prospective study in tertiary care hospital of eastern Nepal. *Kathmandu Univ Med J* 5(2):195–198
- Soontrapa P, Larbchaoensub N et al (2010) Fungal rhinosinusitis: a retrospective analysis of clinicopathologic features and treatment outcomes at Ramathibodi hospital. *Southeast Asian J Trop Med Public Health* 41(2):442–449
- Goh B-S, Gendeh BS et al (2005) Prevalence of allergic fungal sinusitis in refractory chronic sinusitis in adult Malaysians. *Otolaryngol-Head Neck Surg* 133:27–31