

## Necrotizing fungal infection due to *Saksenaea erythrospora*: A case report and review of literature

Bipasha Mukherjee, Debi Kundu

Mucormycosis caused by *Saksenaea erythrospora* is an emerging infection seen with soil contamination, burns and/or nosocomial infections. PCR amplification and internal transcribed spacer sequencing is gold standard for its identification. Here, we report a case of necrotizing fungal orbital infection by *S. erythrospora* in an immunocompetent child.

Access this article online	
Quick Response Code:	Website: www.ijo.in
	DOI: 10.4103/ijo.IJO_389_18

Department of Orbit, Oculoplasty, Reconstructive and Aesthetics, Medical Research Foundation, Chennai, Tamil Nadu, India

**Correspondence to:** Dr. Bipasha Mukherjee, Department of Orbit, Oculoplasty, Reconstructive and Aesthetics, Medical Research Foundation, Chennai - 600 006, Tamil Nadu, India. E-mail: beas003@yahoo.co.uk

Manuscript received: 28.03.18; Revision accepted: 04.06.18

**Key words:** Mucormycosis, necrotizing, *Saksenaea*

*Saksenaea erythrospora*, a species of the order mucorales, has recently reported to cause human mucormycosis.

### Case Report

A 2-year-old female child presented with rapidly progressive swelling in the medial canthal region of her right eye for past 1 month. Her parents gave history of watering with discharge from the right eye since birth. She was diagnosed to have congenital nasolacrimal duct obstruction elsewhere. She was treated with oral antibiotics without any symptomatic relief. There was no history of weight loss, trauma, nasal blockage, or epistaxis.

On examination the child was febrile and irritable. Her right eye was proptosed with tender, erythematous, periorbital swelling with conjunctival congestion and purulent discharge [Fig. 1a]. Ocular movements were restricted, with pupil reacting normally. Rest of the ophthalmic examination was within normal limits. Magnetic resonance imaging (MRI) of the orbit revealed an ill-defined soft tissue mass in the right

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

**For reprints contact:** reprints@medknow.com

**Cite this article as:** Mukherjee B, Kundu D. Necrotizing fungal infection due to *Saksenaea erythrospora*: A case report and review of literature. Indian J Ophthalmol 2018;66:1513-6.

medial preseptal area, medial extraconal space, erosion of the medial wall with extension into the ethmoid sinus. The mass was iso-intense in T1 and T2, showing postcontrast heterogeneous enhancement [Fig. 2]. Blood picture showed hypochromic microcytic anemia with absolute neutrophilic leucocytosis; and increased erythrocyte sedimentation rate (ESR). Blast cells were not detected on peripheral blood smear. She was advised incisional biopsy under general anesthesia. Preoperatively, the skin over the mass was friable with an underlying ill-defined firm mass replacing normal architecture [Fig. 1b]. Cutaneous abscesses were located and drained and sent for microbiological examination, which was negative for gram stain, KOH stain, bacterial culture, and fungal culture at 48 h and 12 days. Broad-width, aseptate hyphae consistent with mucorales infection (mucormycosis) with surrounding granulomatous inflammation were noted on histopathological examination [Fig. 3]. PCR-based DNA sequencing by targeting ITSr (interspace region) of fungus genome identified the species as *S. erythrospora*. The child

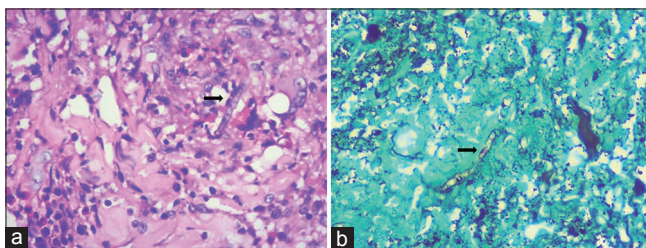
was transferred to a multispecialty childrens' hospital 5 days after presentation for intravenous amphotericin B, which was given as 1-h infusion once daily with regular monitoring of her renal function. Daily dressing of the local area with gauze soaked in amphotericin B was done. After a week the child was taken up for extensive debridement under general anesthesia [Fig. 1c]. The child was discharged on oral posaconazole after receiving injection amphotericin B for 42 days. Healthy granulation tissue was seen in the local area at the time. She was reviewed after a month when her right eye fixation was central, steady, and maintained; ocular motility was free, full, and painless; right medial periocular area showed healthy scar tissue with lid retraction due to anterior lamellar shortening, but no lagophthalmos [Fig. 1d]. MRI showed near complete resolution of the mass [Fig. 4]. At her last follow-up 8 months from presentation, she had central, steady fixation both eyes with lower lid retraction, and mild lagophthalmos [Fig. 1e and f]. Rest of her ocular examination was within normal limits. She was advised to patch left eye for 2 h per day by the pediatric ophthalmologist. She has also been advised right eye medial canthoplasty during her next visit.

## Discussion

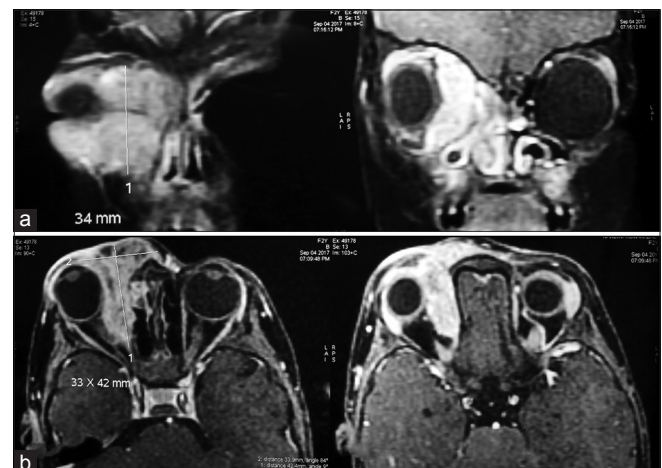
*Saksenaea*, a genus of mucorales fungus, was discovered in 1953.<sup>[1]</sup> Mucormycosis caused by *Saksenaea* species most often occurs after traumatic injury with soil contamination, but can also be due to nosocomial infections.



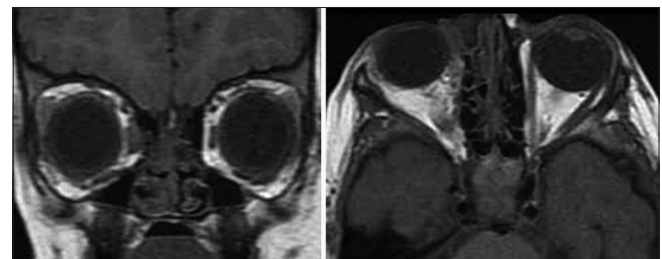
**Figure 1:** (a) The patient at presentation showing proptosis of the right eye with periocular mass and pus pointing over medial canthal area. (b) Intraoperative picture after drainage and incisional biopsy. (c) Intraoperative picture during debulking of the mass. (d) Postoperative appearance of the patient showing complete resolution of proptosis; right periocular healthy wound with eyelid retraction. (e) Late postoperative appearance of the patient showing right lower lid retraction. (f) Mild medial lagophthalmos with no corneal exposure in the right eye



**Figure 3:** Microphotographs showing aseptate hyphae (arrow) with surround inflammatory cells. (a) Hematoxylin and eosin; (b) Gomori methamine silver (GMS) stain. (40x)



**Figure 2:** (a and b) Coronal and axial T2-weighted MRI scan pictures showing diffuse isointense soft tissue mass lesion showing heterogeneous contrast enhancement



**Figure 4:** Coronal and axial T1-weighted MRI scans showing mild residual thickening of RMR. Rest findings are within normal limits

**Table 1: Case reports of human infection with *Saksena* *erythrospora***

Author	Year	Number of patients	Mechanism of injury	Risk factors	Country	Management	Final Outcome
Hospenthal <i>et al.</i> <sup>[6]</sup>	2011	1	Combat injury	Burn	Iraq	Debridement	Died
Relloso <i>et al.</i> <sup>[2]</sup>	2014	1	Sailing injury	Wound exposure to soil, debris	Argentina	Hyperbaric oxygen Oral posaconazole INF gama	Improved
Tendolkar <i>et al.</i> <sup>[5]</sup>	2015	1	Indeterminate	Indeterminate	India	Debridement Exenteration IV Amphotericin B	Improved
Akers <i>et al.</i> <sup>[7]</sup>	2015	1	Combat injury	Burns	Afghanistan	Debridement L-Amb (5 mg/kg IV/24h) Voriconazole (4 mg/kg IV every 12 h)	Improved
Rodriguez <i>et al.</i> <sup>[8]</sup>	2016	1	Aesthetic breast augmentation surgery	Iatrogenic trauma	Colombia	Surgical debridement IV Amphotericin B Caspofungin Hyperbaric oxygen	Improved
Chander <i>et al.</i> <sup>[4]</sup>	2016	5	Intramuscular injection	Iatrogenic trauma	India	Debridement Liposomal Amphotericin B	4 improved; 1 died
Present report	2017	1	Indeterminate	CNLDO?	India	Debridement + IV Amphotericin B + Oral Posaconazole	Improved

L-Amb: Liposomal Amphotericin B, CNLDO: Congenital NasoLacrimal Duct Obstruction

Most of *S. erythrospora* infections occurred in immunocompetent individuals. Mucorales tend to invade arteries, leading to thrombosis and gradually generating wide necrotic areas; this favors progression and invasion of deep tissues.<sup>[2]</sup>

For over half a century, cutaneous infections by *Saksena* had been attributed to *S. vasiformis*. However, a recent taxonomic revision of *Saksena* based on morphological as well as on molecular characteristics revealed two new species, *S. oblongispora* and *S. erythrospora*.<sup>[3]</sup> *S. erythrospora* is characterized by flask-shaped sporangia and ellipsoid, biconcave sporangiospores, resembling erythrocytes.<sup>[3]</sup>

Till date, there have been 10 reports of human infections caused by *S. erythrospora* [Table 1].<sup>[4,5]</sup> Hospenthal *et al.* reported the first case of human *S. erythrospora* infection in 2011 in a soldier following combat injury.<sup>[6]</sup> This patient had a fatal outcome. Another patient survived *S. erythrospora* infection following blast injury.<sup>[7]</sup> Relloso *et al.* reported infection with *S. erythrospora* following a sailing injury.<sup>[2]</sup> Rodríguez *et al.* reported a patient who developed *S. erythrospora* infection after breast augmentation surgery.<sup>[8]</sup> The first report of human *S. erythrospora* infection from India was described by Tendolkar *et al.*, in an immunocompetent patient, presenting with orbital cellulitis and pansinusitis.<sup>[5]</sup> Chander *et al.* reported five cases of *S. erythrospora* infection from the same hospital in North India. The authors postulated high rate of this rare infection from the same geographical area due to the peculiar local practice of administering injections through (possibly soil contaminated) clothes. Moreover, the possibility of the needles, or the injections being contaminated, could not be ruled out.<sup>[4]</sup>

In all probability, our patient had a secondary infection with soil contamination of her congenitally obstructed right nasolacrimal duct as she was from a rural farming community. As is often the case in children, there was no history of trauma in this patient.

The microbiological identification of agents of mucormycosis is difficult to obtain. Isolates may fail to grow on subculture or sporulate. Therefore, persistent attempts to induce sporulation on nutritionally deficient media should be made. The use of molecular methods based on PCR amplification and internal transcribed spacer (ITS) sequencing should be utilized when faced with a nonresolving atypical infection of the skin and the soft tissues.<sup>[9]</sup>

In a suspected case of mucormycosis, PCR result can be obtained in a few hours, whereas histopathology need days and cultures need weeks for growth and identification.<sup>[10]</sup>

Surgical debridement along with antifungal regime remains the mainstay of treatment.<sup>[10]</sup> Members of the family *Saksena* are considered to respond better to posaconazole and itraconazole, than to amphotericin B, a fact to be considered when selecting empirical therapy.<sup>[9]</sup>

## Conclusion

The rapid progression of the disease highlights the importance of correct and early diagnosis, identification of the etiological agent and appropriate treatment.

### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

### Financial support and sponsorship

Nil.

### Conflicts of interest

There are no conflicts of interest.

## References

1. Saksena SB. A new genus of the mucorales. *Mycologia*. 1953;45:426-36.
  2. Relloso S, Romano V, Landaburu MF, Herrera F, Smayevsky J, Veciño C, *et al.* *Saksenaea erythrospora* infection following a serious sailing accident. *J Med Microbiol* 2014;63:317-21.
  3. AlvarezE, Garcia-Hermoso D, Sutton DA, Cano JF, Stchigel AM, Hoinard D, *et al.* Molecular phylogeny and proposal of two new species of the emerging pathogenic fungus *Saksenaea*. *J Clin Microbiol* 2010;48:4410-6.
  4. Chander J, Singla N, Kaur M, Punia RS, Attri A, Alastruey-Izquierdo A, *et al.* *Saksenaea erythrospora*, an emerging mucoralean fungus causing severe necrotizing skin and soft tissue infections – a study from a tertiary care hospital in north India. *Infect Dis* 2017;49:170-77.
  5. Tendolkar U, Diepeningen AV, Joshi A, Koomen J, Bradoo R, Baveja S, *et al.* Rhinosinusitis caused by *Saksenaea erythrospora* in an immunocompetent patient in India: A first report. *JMM Case Rep* 2015;2:1-4.
  6. Hospenhal DR, Chung KK, Laird K, Thompson EH, Guarro J, Renz EM, *et al.* *Saksenaea erythrospora* infection following combat trauma. *J Clin Microbiol* 2011;49:3707-9.
  7. Akers KS, Rowan MP, Niece KL, Graybill JC, Mende K, Chung KK, *et al.* Antifungal wound penetration of amphotericin and voriconazole in combat-related injuries: Case report. *BMC Infect Dis* 2015;15:184.
  8. Rodríguez JY, Rodríguez GJ, Morales-López SE, Cantillo CE, Le Pape P, Álvarez-Moreno CA. *Saksenaea erythrospora* infection after medical tourism for esthetic breast augmentation surgery. *Int J Infect Dis* 2016;49:107-10.
  9. Muszewska A, Pawlowska J, Krzysciak P. Biology, systematics, and clinical manifestations of Zygomycota infections. *Eur J Clin Microbiol Infect Dis* 2014;33:1273-87.
  10. Mukherjee B, Raichura ND, Alam MS. Fungal infections of the orbit. *Indian J Ophthalmol* 2016;64:337-45.
-