

Bilateral *Bipolaris* Keratomycosis in a Borderline Lepromatous Patient¹

Kala Gopalakrishnan, Ebenezer Daniel, Renu Jacob,
Gigi Ebenezer, and Mary Mathews²

ABSTRACT

This is a report of an unusual case of *Bipolaris* mycotic keratitis infecting the corneas of both eyes in a cured, immunocompetent patient with previous borderline lepromatous disease. *Bipolaris* keratomycosis is probably more common than is generally appreciated, and is probably often overlooked in patients with Hansen's Disease.

RÉSUMÉ

Un cas inhabituel de kératite mycotique due à *Bipolaris* est rapporté. Cet agent mycotique a infecté les cornées des 2 yeux chez un patient guéris et immunocompétent présentant des commémoratifs de lèpre lépromateuse borderline. La kératomycose causée par *Bipolaris* est probablement plus commune que l'on pense, et est peut-être sous-diagnostiquée chez les patients souffrant de Maladie de Hansen.

RESUMEN

Se presenta un caso, raro, de queratitis micótica *Bipolaris* en la córnea de ambos ojos de un paciente inmunocompetente, curado de lepra lepromatosa subpolar. La queratomycosis *Bipolaris* es probablemente más común de lo que se piensa y seguramente pasa desapercibida en los pacientes con la enfermedad de Hansen.

India has the largest number of cured and newly diagnosed leprosy patients in the world. The eyes of many of these patients are particularly vulnerable to injury. There have been several reports on fungal infections involving the cornea of leprosy patients^(1, 5, 7, 11, 17). Fungal keratitis is usually caused by saprophytic organisms in the setting of trauma and altered host resistance. Although bilateral corneal ulcers are not uncommon in leprosy, bilateral mycotic keratitis, to our knowledge, has never been reported in a leprosy patient. *Bipolaris* (pre-

viously known as *Dreschlera*) is a cosmopolitan organism in nature and is isolated from plant debris and soil. It has been described to cause allergic fungal sinusitis, disseminated infections, keratitis, and skin infections in both immunocompromised and immunocompetent host^(1, 12, 13, 16, 18). We report the occurrence of *Bipolaris* mycotic keratitis infecting both the corneas of a cured leprosy patient over a period of 2 yrs.

CASE REPORT

In January 1999, a 45-year-old borderline lepromatous patient who had been on irregular anti-leprosy treatment for 5 yrs was re-evaluated and put on multi-drug (MDT) therapy. Six months after starting MDT, in July 1999, he presented with an ulcer in the left eye but gave no history of trauma to the eye. Ocular examination revealed the presence of old bilateral lagophthalmos for which lateral tarsorrhaphy had been done. Superficial punctate keratitis was also present, as well as exposure keratitis, decreased corneal sensation, irregular pupils with posterior synechia, iris atrophy and immature cataracts in both the eyes. Corneal

¹ Received for publication on 28 October 2002. Accepted for publication on 10 January 2003.

² K. Gopalakrishnan, M.B.B.S., M.S., D.O., Specialist; E. Daniel, M.B.B.S., M.S., D.O., M.P.H., Head; R. Jacob, M.B.B.S., M.S., Specialist, Department of Ophthalmology; G. Ebenezer, M.B.B.S., M.D., Head, Department of Histopathology and Experimental Pathology, Schieffelin Leprosy Research and Training Center; M. Mathews, M.B.B.S., M.D., Professor, Microbiology Department, Christian Medical College and Hospital, Vellore, India.

Reprint requests to: Ebenezer Daniel, Head, Department of Ophthalmology, Schieffelin Leprosy Research and Training Center, Karigiri, Vellore District, Tamilnadu, India - 632106.



FIG. 1. Photomicrograph showing erect, sympodial, geniculate conidiophore and ellipsoidal conidia of *Bipolaris* spp. (LPCB \times 400)

scrapings taken from the ulcer and plated on Sabouraud's dextrose agar showed a cottony white growth which turned into a mouse grey color with dark pigmentation on the reverse side of the plate. Microscopic examination revealed this to be a colony of fungus belonging to the species *Bipolaris*. The patient improved slowly over a period of 4 wks after rigorous treatment with ketoconazole eye drops. This was reported earlier⁽¹⁶⁾.

The same patient presented with watering, pain, and redness in the right eye in August 2001. He had completed only 14 doses of MDT because of the irregularity of his visits. His skin smears done in 1999 and 2000 were negative for acid fast bacilli. There was history of trauma to the right eye with paddy. On examination, the right eye had a visual acuity of 3/60 and there was a corneal ulcer with approximate dimensions of 6 mm \times 4 mm in the lower-middle portion of the cornea with attending hypopion of 2 mm. Lagophthalmos with tarsorrhaphy was present. The naso-lacrimal ducts were patent but corneal hypo-aesthesia was present in both eyes. Scrapings were taken from the edge of the ulcer and sent for Gram's stain, KOH stain, and plating on both blood agar and Sabouraud dextrose agar (SDA). Direct microscopy with Gram's stain showed many pus cells, epithelial cells, and gram-positive septate hyphae. The SDA plate showed the growth of a mouse grey colony. Microscopic examination using a slide culture on SDA revealed numerous conidiophores which were dark, branched, septate, and geniculate (Fig. 1). The macroconidia showed 2 to 5 septa, most often 3.



FIG. 2. Right Eye showing healed corneal opacity with vascularization.

The conidia were straight, oblong or rounded at both ends, with thick septal walls and truncate, dark hilum. This was consistent with *Bipolaris* spp. The patient's blood sugar levels, which were estimated several times, were within normal limits. Both the VDRL test and the HIV anti-body tests were negative. The patient was treated with regular ketoconazole drops and the ulcer healed slowly over a period of 2 months. The patient was seen at regular intervals following the eye treatment. In May 2002, 8 months after stopping all anti-mycotic eye drops and the eyes had only thick healed opacities with vascularization (Fig. 2), swabs were taken from the lower conjunctival fornices of both eyes and plated onto an SDA medium. No growth of any organism was found.

DISCUSSION

India has the largest number of both cured and newly diagnosed leprosy patients in the world. Most patients living in the Indian sub-continent, if they are able to work, do so in agrarian jobs where injury to the eyes with vegetable matter is not a rare event^(4, 10). Such ocular injury and the subsequent development of an infectious fungal corneal ulcer would not be too unexpected in leprosy patients who have decreased corneal sensation and lagophthalmos. Although several cases of *Bipolaris* mycotic keratitis have been reported from India^(1, 9, 16) and among leprosy patients^(1, 16, 18), it is likely that *Bipolaris* keratomycosis in leprosy is greatly underreported because in most leprosy endemic regions there is a lack of laboratory facilities or facilities are too expensive.

The leprosy patient who developed *Bipolaris* keratomycosis presented with two interesting features. Bilateral keratitis by infectious agents is not common and bilateral fungal keratitis is extremely rare. There have been reports of bilateral fungal corneal ulcers occurring in patients who had been immuno-compromised by HIV infection^(2, 15) or had diabetes⁽¹⁴⁾. This patient was negative for HIV infection. Systemic fungal infections have been known to occur in leprosy⁽³⁾, but this patient did not have any systemic fungal infection. The mycotic keratitis also occurred in the right eye 2 yrs after it had occurred in the left eye. What was interesting was the fact that the infecting agent in both the eyes was *Bipolaris*, a fungus that is not a common pathogen. If there were no systemic causes that would promote the growth of such a fungus in the cornea, the local ocular environment (lagophthalmos with lateral tarsorrhaphy and impaired corneal sensation), which was similar in both eyes, could have supported the growth of the fungus. However, long after the ulcers had healed and no anti-fungal therapy was used, conjunctival swabs were taken from both eyes and these did not grow the fungus in culture media. This does not disallow the fact that the fungus could have been a resident in the outer surface of the eye at the time of injury, as it has been shown that fungi occur in the outer eye generally as transients⁽⁶⁾. It could be that the causative agent of the injury was the same and carried the fungus on to the cornea, although a definite history of injury could not be obtained from the patient at the time of the first ulcer. Whatever the etiopathogenesis, this report is possibly the first documentation of a bilateral *Bipolaris* infection of the cornea.

Lateral tarsorrhaphy had been done in both of the patient's eyes. Although this surgery reduced the width of the palpebral fissure, it was not good enough to prevent the occurrence of corneal ulcers (Fig. 3). Patients with impaired corneal sensation and concurrent lagophthalmos constitute a group that requires regular check-ups and intensive health education; this is important in a climate of integration and reduced duration of MDT. The combination of an agrarian oc-



FIG. 3. Right eye on gentle closure showing an exposed palpebral fissure but with the cornea fully covered by the upper lid.

cupation, a vegetable injury, an illiterate and non-compliant patient, lagophthalmos with coexisting impaired corneal sensation, and a health system not alert to the occurrence of fungal corneal ulcers can quite easily lead to blindness or severe visual impairment.

Acknowledgment. We are grateful to Ms. Esther Reuben for her technical help.

REFERENCES

1. ANANDI, V., SURYAWANSHI, N. B., KOSHI, G., PADHYE, A. A. and AJELLO, L. Corneal ulcer caused by *Bipolaris hawaiiensis*. *J. Med. Vet. Mycol.* **26** (1988) 301–306.
2. ARISTIMUNO, B., NIRANKARI, V. S., HEMADY, R. K. and RODRIGUES, M. M. Spontaneous ulcerative keratitis in immunocompromised patients. *Am. J. Ophthalmol.* **115**(2) (1993) 202–208.
3. ARUNTHATHI, S., EBENEZER, G., DANIEL, E. and SUGUMARAN, S. T. *Nocardia farcinica* pleuritis in a lepromatous patient with severe necrotizing reaction: an unusual presentation. *Int. J. Lepr.* **69** (2001) 104–107.
4. CHANDER, J. and SHARMA, A. Prevalence of fungal corneal ulcers in northern India. *Infection* **22**(3) (1994) 207–209.
5. DANIEL, E., MATHEWS, M. and CHACKO, S. *Alternaria keratomycosis* in a lepromatous leprosy patient. *Int. J. Lepr.* **65** (1997) 492–497.
6. GUGNANI, H. C., GUPTA, S. and TALWAR, R. S. Role of opportunistic fungi in ocular infections in Nigeria. *Mycopathologia* **65**(1–3) (1978) 155–166.
7. HEMASHEHTAR, B. M., VEERAPPA, T. S., VERMA, P. V., HANCHINAMANI, S., PATIL, C. S. and THAMMAYYA, A. Mycotic keratitis caused by *Bipolaris spicifera*. *Indian J. Pathol. Microbiol.* **35** (1992) 274–277.
8. JOHN, D. and DANIEL, E. Infectious keratitis in leprosy. *Br. J. Ophthalmol.* **83** (1999) 173–176.

9. KOSHI, G., ANANDI, V., KURIEN, M., KIRUBAKARAN, M. G., PADHYE, A. A. and AJELLO, L. Nasal phaeohyphomycosis caused by *Bipolaris hawaiiensis*. *J. Med. Vet. Mycol.* **25** (1987) 397–402.
10. LIN, S. H., LIN, C. P., WANG, H. Z., TSAI, R. K. and HO, C. K. Fungal corneal ulcers of onion harvesters in southern Taiwan. *Occup. Environ. Med.* **56**(6) (1999) 423–425.
11. MATHEWS, M. S. and MAHARAJAN, S. V. *Exserohilum rostratum* causing keratitis in India. *Med. Mycol.* **37** (1998) 131–132.
12. McCLAY, J. E., MARPLE, B., KAPADIA, L., BIAVATI, M. J., NUSSENBAUM, B., NEWCOMER, M., MANNING, S., BOOTH, T. and SCHWADE, N. Clinical presentation of allergic fungal sinusitis in children. *Laryngoscope* **112** (2002) 565–569.
13. PAUZNER, R., GOLDSCHMIED-REOUVEN, A., HAY, I., VERED, Z., ZISKIND, Z., HASSIN, N. and FARFEL, Z. Phaeohyphomycosis following cardiac surgery: case report and review of serious infection due to *Bipolaris* and *Exserohilum* species. *Clin. Infect. Dis.* **25** (1997) 921–923.
14. PRAJNA, N. V., RAO, R. A., MATHEN, M. M., PRAJNA, L., GEORGE, C. and SRINIVASAN, M. Simultaneous bilateral fungal keratitis caused by different fungi. *Ind. J. Ophthalmol.* **50** (2002) 213–214.
15. SANTOS, C., PARKER, J., DAWSON, C. and OSTLER, B. Bilateral fungal corneal ulcers in a patient with AIDS-related complex. *Am. J. Ophthalmol.* **102**(1) (1986) 118–119.
16. SHEENA KOSHY and EBENEZER, D. *Bipolaris Keratomycosis* in a leprosy patient: a case report. *Lepr. Rev.* **73** (2002) 76–78.
17. TENDOLKAR, U. M., VARAIYA, A., AHUJA A. S., MOTWANE, S. A., AND GOGATE, A. S. Corneal ulcer caused by *Nocardia asteroides* in a patient with leprosy. *J. Clin. Microbiol.* **37** (1999) 2392.
18. ZAPATER, R. C., ALBESI, E. J. and GARCIA, G. H. Mycotic keratitis by *Drechslera spicifera*. *Sabouraudia* **13** (1975) 295–298.