This also opens the possibility of hormonal manipulation in treating such tumours.

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Sir,

Scedosporium apiospermum keratomycosis with secondary endophthalmitis

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Scedosporium apiospermum, the asexual form of *Pseudallescheria boydii*, is a saprophytic filamentous fungus found in soil, manure, and polluted water. In the tropics, it is a common cause of mycetoma and systemic infections have been reported in immunocompromised patients.[1] Ocular infection is frequently associated with trauma and generally the prognosis is poor. We report the treatment of a case of keratomycosis caused by *S. apiospermum* that progressed to endophthalmitis. Although infection was controlled, vision was ultimately lost as a result of secondary glaucoma.

Case report

A 56-year-old female contact lens wearer became aware of a foreign body sensation in her right eye while walking in a wooded area of southern England. A corneal abrasion was treated in A&E with topical chloramphenicol 0.5% drops. After 1 week, the patient presented with a central corneal abscess and hypopyon. A sample of corneal tissue was taken for microscopy and culture. Treatment was modified to topical ofloxacin 0.3% hourly. There was a rapid clinical improvement and the patient was discharged on topical ofloxacin and prednisolone (0.3% q.i.d.). On review, 1 week later, there was an increased corneal infiltrate and the hypopyon had recurred. *S. apiospermum* was cultured from the initial corneal scrape material and, therefore, the topical steroid was withdrawn and treatment changed to topical clotrimazole (1%, hourly), oral fluconazole (200 mg/daily) and intravenous liposomal amphotericin B (1 mg/kg/day). This was later modified to hourly topical econazole (1%) and amphotericin B (0.15%) with oral itraconazole (200 mg/daily) when the results of *in vitro* susceptibility testing became available. No donor corneal material was available and despite continued intensive topical therapy, the cornea perforated after 4 days (Figure 1a).

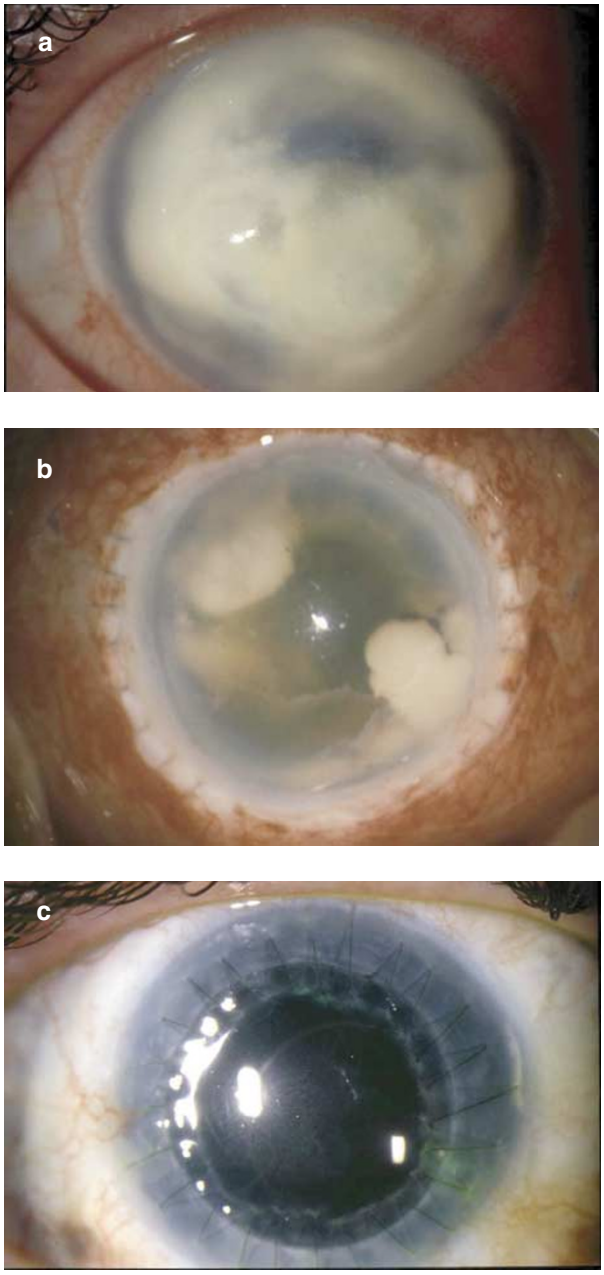

Figure 1 (a) Perforated cornea prior to first corneo-scleral graft. (b) Spherical deposits rapidly appeared in the anterior chamber after the first graft. These were aspirated and *S. apiospermum* was isolated from this material. (c) Final appearance two years after the third graft and intraocular lens insertion.

A 15.5-mm diameter corneoscleral graft was required to provide a 2-mm clearance of the infiltrated cornea and amphotericin B (5 μg in 0.1 ml) was injected into the anterior chamber at the end of surgery. Histology of the excised cornea showed that the graft margins were clear, but fungal hyphae had permeated the anterior chamber. *S. apiospermum* was grown from the excised cornea and from samples taken from the surface of the iris.

Treatment continued with topical amphotericin B, econazole, and ofloxacin with oral Itraconazole (increased to 400 mg/daily). Within 24 h of surgery, white spherical deposits began to re-form in the anterior chamber (Figure 1b). These were aspirated on three occasions over the following 6 days and amphotericin B (5 μg in 0.1 ml) injected into the anterior chamber. *S. apiospermum* was isolated from the first aspirate, but not subsequently.

Despite continued antifungal therapy the graft melted peripherally and after 3 weeks, a repeat 19-mm diameter corneoscleral graft was performed combined with cyclodialysis, total iridectomy, intracapsular lensectomy, and a limited anterior vitrectomy. *S. apiospermum* was isolated from the corneal tissue, but not from the iris, trabecular meshwork or anterior ciliary processes. This isolate was reported to be resistant to itraconazole (2.0 mg/l) and oral treatment was changed to fluconazole (400 mg/daily). Oral prednisolone (60 mg/daily) was introduced to reduce the risk of a repeat graft melt. Oral steroid and fluconazole were stopped after 6 weeks, but the topical therapy (amphotericin B and econazole) was continued for a total of 4 months. The graft never cleared and the intraocular pressure was normal. However, the disc could not be visualised because of vitreous haemorrhage.

At 9 months after stopping treatment, a 7.5-mm penetrating keratoplasty was performed combined with insertion of a sutured posterior chamber intraocular lens and an artificial iris (Morcher aniridia lens 67F) to reduce symptoms of glare. The vision returned to 6/18 with refraction of $(+1.5/-4.5 \times 180)$. Unfortunately, the intraocular pressure gradually increased and, despite topical therapy and cyclodiode treatment, the patient developed an afferent pupil defect with optic disc cupping. At 24 months after surgery the third graft remained clear with a stable epithelium (Figure 1c), but vision was reduced to hand movements. The patient was satisfied with the cosmetic result and uses topical fluoromethalone 1% and xalatan once daily.

Comments

Although the outcome of fungal keratitis has improved with the availability of more effective antifungal agents, the prognosis following intraocular spread remains poor and it is often associated with loss of vision. Outside the tropics, *S. apiospermum* is an uncommon cause of keratitis and there are few reported cases of endophthalmitis.[2,3] Indeed, only two other cases of endophthalmitis secondary to *S. apiospermum* keratitis have been reported. Despite treatment with miconazole, both cases resulted in evisceration.[4,5]

The *S. apiospermum* isolate in this case was sensitive *in vitro* to amphoteracin B, econazole and miconazole.

However, the keratitis failed to respond to antifungal therapy and surgical excision was required. Although many isolates of *S. apiospermum* are resistant *in vitro* to amphotericin B, there have been reports of successful treatment of keratitis with amphotericin B as monotherapy, or in combination with nystatin, natamycin and itraconazole.[2,6] Successful treatment of *S. apiospermum* corneal and orbital infections has been achieved using miconazole. However, there are also reports of keratitis and endogenous endophthalmitis which have failed to respond[5,7–11] Although *Scedosporium* spp. are typically resistant *in vitro* to fluconazole, the patient became intolerant to Itraconazole and a clinical decision was made to supplement topical antifungal therapy with oral fluconazole, but this may have been ineffective. The triazole antifungal voriconazole has been used successfully in the treatment of disseminated *Scedosporium* spp. infections.[12] It is clear that an optimum treatment regimen for ocular *Scedosporium* spp. mycoses must still be defined.

This case highlights the dilemma of severe keratomycosis and the continued role of excisional surgery. In any case of atypical keratitis, particularly if there is a history of trauma with vegetative matter, fungal infection must be considered with a high index of suspicion. If this is the case, steroid should be used with great caution due to the risk of enhancing infection. In this case, following the second graft, oral steroid was used successfully to control inflammation and prevent further graft melt. Finally, the risk of secondary glaucoma following large diameter grafts is high. This case illustrates that glaucoma can ultimately limit the visual outcome even after apparently successful surgery.

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Sir,

Primary ductal adenocarcinoma of the lacrimal gland in a patient with neurofibromatosis

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Primary ductal adenocarcinoma of the lacrimal gland is an uncommon tumour and there are only a few published reports.[1,2] We report the third case of primary ductal adenocarcinoma of the lacrimal gland in a patient with neurofibromatosis and discuss the clinical presentation, radiological characteristics, treatment, and histopathological correlation of the tumour.

Case report

A 46-year-old Asian Indian male reported to our institution with complaints of slowly progressive proptosis of the left eye for the past 2 years. Skin examination disclosed numerous subcutaneous soft nodules suggestive of neurofibromas. He also had