Contact-lens related keratitis caused by an atypical organism

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Abstract: This is a case report of contact-lens related, infectious keratitis with a subsequent perforation. A tectonic penetrating keratoplasty was performed and Sphingomonas paucimobilis was cultured from the corneal button. One week post surgery, the patient developed a re-infection involving the donor button. This was treated based on culture and sensitivity of the corneal button and the infection resolved. Two months post surgery, the patient's vision was counting fingers in the affected eye. The corneal infection had completely resolved, but the graft had failed despite the use of topical steroids.

Keywords: Sphingomonas paucimobilis, corneal ulcer, contact lens, keratitis, atypical organism

Case report
A 41-year-old Malaysian woman was referred to our centre for a left eye contact-lens related corneal ulcer. She was initially treated at a district hospital, where Stenotrophomonas maltophilia was cultured from her contact lens casing and solution.

At presentation, her vision was hand movement in the affected eye. There was a central corneal ulcer measuring 4 x 3.4 mm with an area of thinning centrally. There were satellite lesions within the corneal stroma and a hypopyon measuring less than 1 mm. Corneal sensation was reduced.

She was treated with ceftazidime and fortified gentamicin drops. However, as her condition continued to deteriorate, topical antifungals, amphotericin B drops and fluconazole drops were commenced. Six weeks after presentation, she developed a central corneal perforation.

A tectonic penetrating keratoplasty was carried out after which intensive topical antibiotics and antifungals were continued. The excised corneal button culture was positive for Sphingomonas paucimobilis which was sensitive to ciprofloxacin, gentamicin and augmentin. The histopathological examination showed no fungal elements.

One week post-operatively, the patient developed a recurrent corneal ulcer involving the donor button at 6 o'clock position (Fig. 1). Based on the sensitivity of the previously excised corneal button,

Fig. 1. Recurrence of infection day 7 post penetrating keratoplasty.

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the patient was treated with ciprofloxacin and fortified gentamicin drops. The infection responded and resolved completely (Fig. 2). At her last visit there was no infection, but her corneal graft had failed despite the administration of topical steroids.

**Discussion**

*Sphingomonas paucimobilis* keratitis

*Sphingomonas paucimobilis* is an aerobic, non-fermenting, oxidase and catalase-positive, gram-negative bacillus. It was first reported in 1977 and named *Pseudomonas paucimobilis*. However, based on phylogenetic data, it was later renamed under the genus *Sphingomonas*.2

Unlike *Pseudomonas aeruginosa*, which is more virulent and a notorious cause of contact-lens keratitis,3,4 *Sphingomonas paucimobilis* has low virulence and is rarely isolated from human materials, just like many non-fermentative bacilli. This is due to the absence of a lipopolysaccharide outer core, which has been replaced by glycosphingolipids.5 A pubmed search revealed only two case reports of ocular infections with *Sphingomonas paucimobilis*. Both were cases of postoperative endophthalmitis.6,7 There have been no reported cases of *Sphingomonas paucimobilis* keratitis.

Following surgery in our patient, cultures of the infected corneal button grew *Sphingomonas paucimobilis*. One week postoperatively, the patient developed a recurrence of the infection along the inferior rim of the donor button which was successfully treated with ciprofloxacin and gentamicin based on the disc susceptibility. Apart from our culture and sensitivity findings, the referring district hospital reported indirect evidence of a possible infection with *Stenotrophomonas maltophilia* based on cultures of the patient’s contact lens solution and contact lens casing. While we cannot rule out co-infections with two atypical organisms, we were unable to re-confirm results from the first culture as the specimens were no longer available.

Both these organisms are very similar and known to colonize fluids including irrigation solutions. *Sphingomonas paucimobilis* is a non-fermentating, oxidase- and catalase-positive bacillus, unlike *Stenotrophomonas maltophilia* which is a catalase-positive, oxidase-negative bacillus and has positive reaction for extracellular DNase. Our findings were confirmed by these tests.
Conclusion
This case reports a corneal infection with Sphingomonas paucimobilis which to the best of our knowledge is the first to be reported in literature. It also illustrates the difference between two similar organisms, Stenotrophomonas maltophilia and Shigomonas paucimobilis, which were both previously classified under the same genus pseudomonas. This is important in order to identify the micro-organism correctly and to ensure effective treatment.

References