

Case Report

***Sphingomonas paucimobilis* keratitis post cataract surgery: First case report from India**

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Abstract

Sphingomonas paucimobilis is a rare, aerobic Gram-negative soil bacillus rarely associated with intraocular infections. With only 3 cases of ophthalmic manifestations reported so far, we are presenting the first case of *S. Paucimobilis*, causing keratitis after cataract surgery, from India. The organism, which was resistant to initial medical treatment, eventually responded to a patch graft and the patient improved to a final visual acuity of 20/40.

Key words: *Sphingomonas Paucimobilis*, corneal ulcer, keratitis, patch graft

Introduction

Sphingomonas paucimobilis is a rare, aerobic Gram-negative soil bacillus commonly detected in hospital equipment such as temperature probes, humidifiers, bedside water containers, and sinks (Homes *et al* 1977). It is rarely associated with intraocular infections, with only 3 cases of ophthalmic manifestations reported so far (Adams *et al* 2006, Seo SW *et al* 2008, Ratnalingam V *et al* 2013).

We present a unique case of *Sphingomonas paucimobilis* keratitis, not responding to medical therapy, which developed 6 weeks post uneventful cataract surgery.

Case Report

A 46-year-old Indian woman underwent uneventful cataract surgery for her left eye 6 weeks prior to presenting to our hospital. She was referred to the cornea subspecialty of our hospital for worsening pain and diminution of vision in the operated eye three weeks after her cataract surgery. After cataract surgery, routine post operative medicines (moxifloxacin 0.5% four times a day and prednisolone acetate 1% eye drop suspension six times a day in tapering doses) were prescribed. She was diagnosed with infective keratitis by the operating surgeon at her third week follow up. Topical steroids were stopped and moxifloxacin was increased to one-hourly frequency. In addition, she was started empirically one hourly tobramycin 0.3% eye drops and Natamycin 5% ophthalmic solution. There was no mention of any corneal scrapping which was done. The operating surgeon did not provide detailed records of patient examination. However, by six weeks post operatively, when the patient didn't respond to treatment, she was referred to us with a diagnosis of refractory

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keratitis. The patient gave no history of trauma or contact lens use after cataract surgery, and no previous systemic illness was noted.

On initial examination, her unaided distant visual acuity in the right eye was 20/80 and in the left eye was counting fingers at 1 metre (with accurate light perception in all quadrants). Slit lamp examination for the right eye was normal except for a posterior subcapsular cataract. The left eye, however, revealed prominent eyelid edema and moderate circumcilliary congestion. Within the central corneal stroma, a 6mm x 4 mm grey infiltrate with feathery margins was noted which extended peripherally in the 9 o'clock direction. Minimal stromal oedema surrounded the infiltrate. An associated 4mm x 2 mm epithelial defect and 1mm convex hypopyon were also present. The lesion was covering the surgical incision site (side port incision) with sparing of adjacent sclera (Figure 1). The patient seemed to have undergone a superior phacoemulsification. No satellite lesions were evident. Corneal sensations were normal. A well-centred posterior chamber IOL in the bag was present. Posterior segment examination was normal bilaterally. Left nasolacrimal duct was patent on syringing, done by a 23-gauge irrigating cannula. Systemic work up did not reveal any systemic illness and blood sugar values were within normal limits.

Microbiological Evaluation

Under topical anesthesia (4% lignocaine hydrochloride) and slit-lamp magnification, corneal scrapings were obtained from the base and edge of the ulcer using a sterile surgical blade (no. 15 on a Bard Parker handle). Corneal scrapings were smeared on a glass slide for Gram stain, Giemsa stain and 10% potassium hydroxide (KOH) mount. The samples were also inoculated onto blood agar, chocolate agar, McKonkey and nutrient agar for 7 days Gram staining did not reveal any organism, and KOH was negative for fungus. In the absence of any conclusive laboratory evidence, a provisional

diagnosis of indolent bacterial keratitis with corneal ulcer was made. The patient was started on combination of one hourly fortified ophthalmic solutions of cefazolin sodium, 50 mg/mL, and tobramycin sulfate, 14 mg/mL. She was also advised 2% homatropine eye drops three times a day, and kept on close follow up. On day 4, therapy was changed to one hourly fortified amikacin 20 mg/mL and moxifloxacin 0.5% along with 1% atropine sulphate three times a day; due to lack of clinical progress. Despite this change, both infiltrate size and epithelial defect were found to have increased on review at day 6. The original cultures finally demonstrated growth at day 6 (Figure 2a, 2b). Cultures were sent for identification of the microbe using Biomeriue's VITEK®2 system. VITEK®2 identified the organism as *Sphingomonas paucimobilis*. Sensitivities to amikacin, cefazoline, ceftazidime, cephalixin, ciprofloxacin, gatifloxacin, moxifloxacin and tobramycin were demonstrated. Review of available literature suggested *S. paucimobilis* to be a highly atypical organism associated with limited inflammatory potential. B-scan was performed ruling out intraocular spread. Whilst this information was reassuring, the fact that the ulcer had progressed despite best available topical therapy led to a consensus team decision for immediate patch grafting (Figure 3). For the patch graft, donor tissue was prepared by trephination of the donor corneal rim. A smaller trephine measuring 5 mm was used to punch out the donor tissue. The donor tissue was then placed over the recipient, and its linear dimensions were fashioned manually in accordance with the dimensions of the involved cornea. The donor button margins were noted to be overriding the margins of the involved cornea. 15 10-0 monofilament nylon sutures were used to suture the donor cornea. The diseased corneal button was cut in to half, one half of which was sent for histopathological work up, and the other half for microbiological evaluation. This correlated with the microorganism, which was found in

the laboratory work up done pre operatively. Full corneal transplant was reserved in the event patch graft was unsuccessful. Postoperatively, the patient was advised hourly fortified

ophthalmic solutions of cefazolin sodium, 50 mg/mL, and amikacin 20 mg/mL, along with 2% homatropine eye drops three times a day, and frequent lubricating eye drops.



Figure 1 (a,b): The left eye at presentation showing a central corneal ulcer, circumcillary congestion and convex hypopyon.

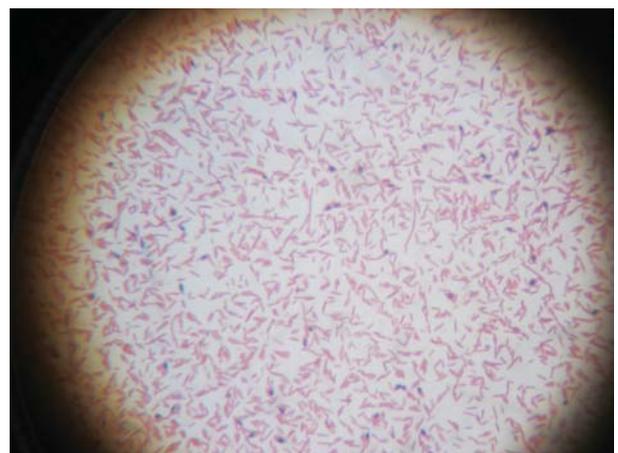


Figure 2 (a,b): Left: Bacterial growth 6 days post inoculation on nutrient, chocolate and blood agars. No growth on McConkey agar seen. Right: Predominantly gram negative bacilli isolated from growth.

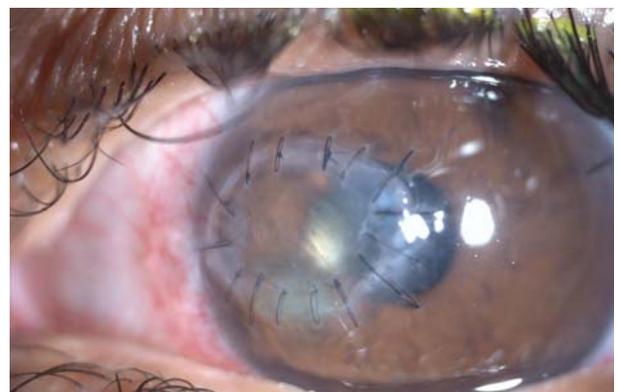


Figure 3 (a,b) Left: Status post patch graft, immediate post operative period; and after 2 months

Patient was regularly followed for two months to ensure continued clinical improvement and that no infiltration of the donor corneal tissue occurred. Follow up intervals were relaxed once unaided distant visual acuity had improved to 20/40 in the left eye.

Discussion

Sphingomonas paucimobilis (formerly known as *Pseudomonas paucimobilis*) is a strictly aerobic, non-fermenting gram-negative bacillus with a single polar flagellum (Homes et al 1977). The natural habitat of this organism has not been fully defined but it is known to be widely distributed in the natural environment. Initial reports of *S. paucimobilis* as a human pathogen date to 1979 when it was implicated in cases of septicaemia, meningitis and leg ulcers (Peel MM et al 1979, Hajiroussou V et al 1979, Slotnick et al 1979).

In context of ophthalmic disease, *S. paucimobilis* appears to be of only limited pathogenic potential. This may be explained by the lack of lipopolysaccharide A (LPS), which is replaced by glycosphingolipid as the major structural component of its gram-negative cell wall (Slotnick et al 1979).

To the best of our knowledge (PubMed search using the terms *Sphingomonas paucimobilis*, keratitis, patch graft), *S. paucimobilis* has not been described as causative agent of infective keratitis in India to date. Its documented role as a pathogen in ophthalmic disease is limited, with only two case reports of post-operative endophthalmitis described to date (Adams WE et al 2006, Seo SW et al 2008). One case presented within 24 hours of cataract surgery and other presented as acute onset delayed endophthalmitis after three months of cataract surgery. In both cases, the patient responded well to treatment. One case has been reported from Malaysia, where keratitis developed after contact lens use (Ratnalingam V et al 2013). In our case, the patient developed infective keratitis after three weeks of uneventful cataract surgery. With prompt surgical intervention by removing the area of infection we were able to

salvage the eye and ensure favourable lasting visual outcomes.

Conclusion

This case is the first known occurrence of *Sphingomonas paucimobilis* as a pathogen causing keratitis post cataract surgery. When managing cases of indolent corneal ulcer we should keep in mind the possibility of *Sphingomonas paucimobilis*. Prompt corneal patch grafting should be considered in such cases.

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