CASE REPORT

Fungal keratitis presenting as radial keratoneuritis

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SUMMARY

A 44-year-old man presented with severe pain, redness, watering and photophobia for 10 days in the right eye without any history of trauma. Diagnosis of herpes simplex disciform keratitis was made and he was prescribed topical steroids. The patient showed clinical worsening and presented with ring infiltrate, diffuse stromal oedema and radial keratoneuritis, a finding pathognomonic of acanthamoeba keratitis. With two inconclusive corneal scrapings and the patient showing clinical worsening, an urgent therapeutic penetrating keratoplasty was carried out. Histopathological and microbiological examination of the excised corneal button revealed the presence of fungus. At 5 weeks follow-up, the patient has best-corrected visual acuity 20/40 with no recurrence of infection.

BACKGROUND

We present a case which initially presented as herpetic stromal keratitis and later developed features of acanthamoeba keratitis including radial keratoneuritis. The final diagnosis of fungal keratitis could be established only on histopathological confirmation of the excised corneal button. This case is the first report of fungal keratitis presenting as radial keratoneuritis. It also highlights the importance of a meticulous microbiological and histopathological work-up and aggressive treatment in a case of keratitis presenting with unusual clinical features.

CASE PRESENTATION

A 44-year-old non-diabetic, male journalist presented with severe pain, redness, watering and photophobia in the right eye (oculus dexter; OD) for 10 days. There was no history of trauma and no history of similar symptoms in the past or in the other eye. He had received a local ophthalmic consultation 1 week earlier where he was prescribed topical fluorometholone six times a day, cyclosporine 0.1% four times a day, acyclovir eye ointment five times a day and preservative free carboxymethyl cellulose eye drops four times a day. As he did not get relief on these medications, he was referred to our institute for further management.

At presentation, he had a best-corrected visual acuity (BCVA) of 20/50 in OD and 20/20 in oculus sinister. Clinical examination revealed oedematous eyelids and diffusely congested conjunctiva. Slit lamp biomicroscopy of the cornea showed an intact epithelium, stromal oedema (2.5×4 mm) involving posterior two-third stroma and endothelium, Descemet's membrane folds and fresh keratic precipitates on endothelium. Anterior chamber showed the presence of cells 2+ and flare 1+. Lens was clear

and fundus examination was normal (figure 1A). Based on the above findings, a clinical diagnosis of herpes simplex disciform keratitis was made. He was prescribed topical fluorometholone 0.1% eight times a day and atropine sulfate 1% eye drops three times a day. After 3 days, he had improved symptomatically but worsened clinically (figure 1B). Conjunctival congestion had reduced but the cornea showed infiltrate (6×6.5 mm) involving full stromal thickness with an intact corneal epithelium. As the patient had worsened on steroids, diagnosis of herpes simplex virus (HSV) keratitis was thought to be unlikely. The topical steroids were stopped and the patient was advised a diagnostic corneal biopsy.

The patient turned up for corneal biopsy after another 3 days. At this visit, he had a corneal epithelial defect, 7×6.5 mm ring infiltrate, radial keratoneuritis, 0.5 mm hypopyon and stromal oedema extending beyond the margin of ring infiltrate, findings described classically for acanthamoeba keratitis^{1 2} (figure 1C). Since there was an epithelial defect, corneal scraping was carried out which did not reveal any organism on Gram stain or potassium hydroxide wet mount with calcofluor white. As per the institution protocol, the patient was started on topical gatifloxacin 0.3% every 1 h and asked to review after 3 days.

After 4 days, he presented with severe pain, redness and watering with a markedly congested conjunctiva and full thickness dense corneal infiltrate measuring 8×8.5 mm (figure 1D). Corneal scraping was repeated which again showed no organisms. Culture report including non-nutrient agar sent 3 days ago also did not reveal any growth. As two diagnostic scrapings did not make a diagnosis and the patient showed severe clinical deterioration, therapeutic penetrating keratoplasty was carried out on the same day. Central 8 mm cornea was trephined taking care to include all the infiltrate and a clear corneal rim of 1 mm all around. This was replaced with 8.5 mm donor tissue secured with 16 interrupted 10-0 monofilament nylon sutures. The excised host corneal tissue was sectioned into two and sent for histopathology and microbiology.

On postoperative day 1, he was prescribed frequent topical gatifloxacin 0.3% eye drops. Histopathology report was retrieved on emergency basis on the third day and the patient was discharged on topical natamycin 5% every 1 h, atropine sulfate 1% three times a day and systemic ketoconazole 200 mg twice daily. At 2 weeks post-surgery, the graft was clear and there were no signs of residual or recurrence of infection (figure 1E). The antifungal therapy was stopped and the

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Figure 1 The clinical course of the disease. At presentation, the patient had an intact epithelium and stromal oedema involving posterior two-third stroma (A); 3 days later, he had a corneal infiltrate with an intact epithelium (B); subsequently, he presented with corneal epithelial defect, large ring infiltrate, radial keratoneuritis (white arrows), hypopyon and stromal oedema extending beyond the margin of ring infiltrate (C); the patient subsequently showed rapid worsening in next 4 days and presented with full thickness dense corneal infiltrate (D); clinical photograph taken 2 weeks postsurgery showing a clear graft and no signs of residual or recurrence of infection (E).

patient was prescribed topical prednisolone 1% eight times a day and atropine sulfate 1% three times a day.

INVESTIGATIONS

Histopathology of the excised specimen showed an ulcerated epithelium and absent basement membrane. The stroma showed diffuse necrosis, presence of neutrophilic infiltrates, short fungal filaments and fungal cut ends in both H&E staining (figure 2A) and Gomori methanamine silver staining (figure 2B). Culture on Sabaroud dextrose agar showed significant fungal growth after 4 days (figure 2C).

DIFFERENTIAL DIAGNOSIS

Taking into consideration the patient's ocular history and clinical presentation, a clinical diagnosis of herpes simplex disciform keratitis was made. When the patient presented for corneal biopsy, the classical clinical picture suggested a diagnosis of acanthamoeba keratitis. However, fungal keratitis was kept as a distant differential diagnosis since the patient had a corneal ring infiltrate and had worsened on administering topical steroids.

OUTCOME AND FOLLOW-UP

At 3 months follow-up, the patient has BCVA 20/40 with a clear graft, no residual or no recurrence of infection.

DISCUSSION

Patients of fungal keratitis may present with a variety of clinical presentations.³ We present a case who presented with clinical

features of HSV keratitis, progressed to develop features considered pathognomic for acanthamoeba keratitis and was later found to be an infection by a fungus, only after histopathological and microbiological evaluation of excised corneal button after therapeutic penetrating keratoplasty was carried out.

Considerable overlap in the clinical presentation of acanthamoeba keratitis and HSV keratitis has been noted earlier. Qian *et al*⁴ found that in a study of 29 eyes with acanthamoeba keratitis, 42% had been earlier diagnosed to be HSV keratitis. Our patient had been under treatment for HSV keratitis earlier. This factor influenced the decision making towards a diagnosis of acanthamoeba keratitis besides the classical clinical features of a corneal ring infiltrate along with radial keratoneuritis. While a ring infiltrate might be seen in keratitis with Gram negative organism like *Pseudomonas aeruginosa* or *Moraxella*, HSV, fungus and acanthamoeba,¹ the presence of radial keratoneuritis has been considered a pathognomic sign of acanthamoeba keratitis.^{2,5} However, there have been earlier reports of radial keratoneuritis in non-acanthamoeba keratitis by Feist *et al*⁶ and Shinoda *et al*⁷ in *Pseudomonas* keratitis and by Mutoh *et al*⁸ in which the aetiology could not be established. We report the occurrence of radial keratoneuritis in a case of fungal keratitis. To the best of our knowledge, this is the first such report.

This case also re-emphasises the fact that the presence of a characteristic clinical feature can be used as a diagnostic aid for suppurative keratitis; however, microbiological investigations should be performed whenever possible, as stated earlier by Thomas *et al*.⁹ The diagnosis in this case could be reliably established when fungal filaments were seen in the histopathological and microbiology specimen. Hence, an efficient laboratory work-up might be required in a case presenting even with such classical features.

To conclude, fungal keratitis can present in varied presentations including radial keratoneuritis. Hence a meticulous clinical work-up including microbiology, and histopathology when needed, is a must in the management. Therapeutic keratoplasty is a good option for severe, non-responding cases especially when an aetiology has not been established.

Learning points

- ▶ Radial keratoneuritis is suggestive, and not pathognomic, of acanthamoeba keratitis.
- ▶ Fungal keratitis can present with radial keratoneuritis.
- ▶ Meticulous clinical work-up is mandatory and microbiology and histopathology support is essential for efficient management of infectious keratitis.
- ▶ Prompt and aggressive treatment is required in keratitis with unusual presentation.
- ▶ Therapeutic keratoplasty, besides providing visual rehabilitation, helps in establishing an aetiological diagnosis.

Figure 2 The histopathology and microbiology of the excised corneal tissue. Stroma showed short fungal filaments and fungal cut ends (black circles) in H&E staining (A) and Gomori methanamine silver staining (B). Sabaroud dextrose agar showing significant fungal growth (C).

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