CASE REPORT

Recurrent non-tuberculous mycobacterial keratitis after deep anterior lamellar keratoplasty for keratoconus

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SUMMARY

A 26-year-old farmer underwent deep anterior lamellar keratoplasty (DALK) for keratoconus. After 3 months, he presented with interface keratitis. Medical treatment failed and he underwent a repeat DALK. Microbiological scrapings from the interface revealed an infection caused by non-tuberculous mycobacteria. Despite the use of intensive antibiotic therapy and a repeat lamellar keratoplasty, the infiltrates recurred. The patient underwent therapeutic penetrating keratoplasty. Microbiology of the corneal tissue revealed growth of *Mycobacterium chelonae*, and on histopathology, the acid-fast bacilli were noted to be located deep at the pre-Descemet level. There was complete resolution of the infection with no episodes of recurrence and final best-corrected visual acuity was 20/40 at 1 year of follow-up. Medical therapy is unlikely to succeed in post-DALK interface keratitis and penetrating rather than lamellar keratoplasty may be considered the surgery of choice.

BACKGROUND

We reported the first case of postdeep anterior lamellar keratoplasty (DALK) interface keratitis caused by non-tuberculous mycobacteria (NTM), which recurred despite repeat DALK.

In contrast to the management of a post-laser assisted in situ keratomileusis (LASIK) interface keratitis in which a medical therapy or a DALK has been shown to work, the treatment of an interface keratitis post-DALK should be a penetrating keratoplasty. This is because an interface keratitis after LASIK or anterior lamellar keratoplasty (ALK) is located up to the anterior or mid-stroma and an adequate clearance of infection can be expected with DALK. In post-DALK interface keratitis, the infection is already located in the deepest stroma and DALK would not be effective to completely remove the infectious tissue. Hence a complete removal of the infectious tissue in the form of penetrating keratoplasty is required. This hypothesis is ably supported by the histopathological proof of *Mycobacterium* bacilli stuck to the endothelium in our case.

CASE PRESENTATION

A 26-year-old farmer presented with a 2-day history of pain, redness and decreased vision in the eye. He had undergone an uneventful DALK for advanced keratoconus 6 months ago. Three months after the first surgery, he was diagnosed as interface infectious keratitis, which was non-responsive to broad spectrum antibiotics and hence the referring

ophthalmologist resorted to a second DALK. At the time of the second surgery, the bed was irrigated with broad spectrum antibiotics. Cultures grown from the anterior lamellar corneal tissue grew atypical *Mycobacterium*. Despite intensive topical therapy with sensitive antibiotics postoperatively, the keratitis recurred after a month. Forty-five days after the second DALK, the patient was referred to us. On presentation, he was instilling topical amikacin 2.5% 2 hourly and prednisolone acetate 1% eye drops three times a day.

At presentation, his best-corrected visual acuity was 20/40 in both eyes. The right eye showed a large graft with a thick arcus superiorly and few intact sutures. Multiple, round, discrete, whitish opacities, ranging from <0.5 mm to 2 mm in diameter were located at the interface (figure 1A). The largest infiltrate was present superiorly close to the graft–host junction and showed endothelial exudates (figure 1B). The left eye examination showed features of keratoconus. A diagnosis of recurrent interface keratitis post-DALK was made. Confocal microscopy was inconclusive. While amikacin 2.5% eye drops were continued, prednisolone 1% eye drops were stopped. Five days later there was paradoxical worsening, secondary to stopping steroids and the patient presented with severe pain and congestion. His vision had decreased drastically to counting fingers close to the face. The corneal infiltrates, though restricted to the graft, had increased in density, became confluent and the surrounding cornea was now oedematous. The anterior chamber showed a hypopyon (figure 1C). He underwent therapeutic penetrating keratoplasty on the same day. As the graft–host junction was spared all around, the graft was excised along with the host Descemet's membrane using a 9 mm trephine and a 9.5 mm donor tissue was secured with 16 interrupted sutures. The host tissue was sectioned into two and sent for histopathology and microbiology. The excised Descemet's membrane was preferentially sent for microbiology. Postoperatively, he received topical amikacin 2.5% every 1 h and prednisolone acetate 1% every 1 h which were subsequently tapered.

INVESTIGATIONS

Histopathology of the excised specimen, scanner view, showed an intact epithelium and an unremarkable stroma devoid of inflammatory cells in the anterior lamellar tissue. Also seen is the posterior tissue which included the minimal posterior stroma of the graft with the host Descemet's

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Figure 1 The clinical course of the disease: multiple, round, discrete, whitish opacities located at the interface at presentation (A). Largest infiltrate was close to the graft–host junction superiorly and also showed endothelial exudates (B). Worsened clinical course after 5 days with increased corneal infiltration (C). Clear graft without residual infiltrate or recurrence of infection at 1 year of follow-up (D).

membrane (figure 2A). Acid-fast staining (1%) revealed dark pink stained, long, cigar-shaped bacilli within the necrotic tissue (figure 2B), noted under high power. Microbiology of the corneal button showed significant growth of Gram-negative bacilli, sensitive to amikacin, gentamycin and tobramycin by disk diffusion method. These were identified as *Mycobacterium chelonae* by the nitrate reduction test.

DIFFERENTIAL DIAGNOSIS

At presentation, the patient's ocular history included repeat-DALK for interface keratitis. Microbiology of the excised specimen had revealed atypical from the interface, in smears as well as the culture. Therefore, when the patient presented to us with a recurrence of infiltrates with in the interface, the most probable diagnosis was recurrent atypical mycobacteria keratitis. A differential diagnosis at this point would include fungal keratitis, in view of the insidious nature of the infiltrates and the appearance of multiple satellite-like lesions. Other pathogens that have an insidious onset that can be considered would include *Nocardia* keratitis.

TREATMENT

Treatment options included either medical therapy or surgical therapy. In view of the location of the infection, medical therapy alone was unlikely to be effective. Surgical options included interface scrapings and antibiotic irrigation, repeat DALK or removal of the infected corneal stroma in the form of a penetrating keratoplasty. Keratoplasty was considered the best choice as the infection was deep seated and had recurred in spite of adequate medical therapy.

OUTCOME AND FOLLOW-UP

At 1-year follow-up, the patient has a clear graft, no residual infiltrate, no recurrence of infection and a best-corrected visual acuity of 20/40 with minimal cataract (figure 1D).

DISCUSSION

Interface keratitis after lamellar keratoplasty is known. Probably, its first report was published by Panda *et al* who reported a small interfacial infiltrate noted 5 days postoperatively which was identified as bright red colonies of *Rhodotorula*. Following

Figure 2 The histopathological details of the corneal specimen: scanner view showing an intact epithelium and a non-contributory stroma devoid of inflammatory cells in the anterior lamellar tissue. Also seen is the posterior tissue which included the minimal posterior stroma (graft) with host Descemet's membrane. (A) Acid-fast staining (1%) revealed dark pink stained, long, cigar-shaped bacilli within the necrotic tissue noted under high power (B).

removal of the graft, the infiltration regressed with topical anti-fungal therapy.¹ Subsequently, a case series of two patients who underwent DALK for keratoconus was published. They presented with white-coloured to cream-coloured interface deposits about 2 months after DALK, which was found to be *Candida* species.² Reports of recurrent *Acanthamoeba* keratitis³ and recurrent interface keratitis by *Candida orthopsilossi*⁴ have since been published. To the best of our knowledge and literature search in PubMed, there is no report published so far of recurrent interface keratitis caused by *M. chelonae* species.

We presented a case of a young man who developed a recurrent interface keratitis following a repeat DALK after an initial uneventful DALK performed for keratoconus. There are a few interesting aspects to our case: first, what could be the possible source of infection? Newman *et al*⁵ has postulated that *M. chelonae* is resistant to chemical disinfectants and Williams *et al*⁶ have associated outbreaks and pseudo-outbreaks of rapidly growing mycobacteria with contaminated tap water. In the present case, the inoculation could have occurred either due to breach in sterilisation or use of reusable dissectors. However, the referring ophthalmologist did not use any other method of sterilisation apart from full-cycle autoclaving. Therefore the source of infection is speculative. Second, is repeating DALK an option for infectious interface keratitis post-DALK? Susiyanti *et al*⁷ reported a case of bilateral NTM infiltrates following bilateral LASIK, which responded well with no recurrence after bilateral DALK. Lyall *et al*⁸ reported a case of infectious interface keratitis following an anterior lamellar keratoplasty which was managed by DALK. However, we believe that post-LASIK

or post-ALK interface keratitis is located upto the mid-stroma and an adequate clearance can be expected with DALK. In post-DALK interface keratitis, the infection is already located in the deepest stroma and DALK would not be effective to completely remove the infectious tissue. Hence, a complete removal of the infectious tissue in the form of penetrating keratoplasty is required. This hypothesis is ably supported by the histopathological proof of bacilli stuck to the endothelium.

To conclude, *M. chelonae* is a rare cause of post-DALK interface keratitis. Medical therapy is unlikely to succeed and based on our experience, penetrating rather than lamellar keratoplasty may be considered as the first surgery of choice.

Contributors RJ and SIM were involved in the conception and design. RJ, SIM and RS were involved in the analysis and interpretation of the data. RJ was involved in drafting the article. SIM and VSS were involved in revising it critically for important intellectual content. SS and VSS were involved in the final approval of the version to be published.

Competing interests None.

Patient consent Obtained.

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Learning points

- Post-deep anterior lamellar keratoplasty (DALK) interface keratitis may be caused by non-tuberculous *Mycobacterium*.
- Medical therapy in interface keratitis is of limited role.
- Interface in a post-DALK is much deeper than that in post-LASIK, and hence a deeper location of interface keratitis.
- Penetrating keratoplasty and not lamellar keratoplasty should be considered as the first surgery of choice.

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