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SHORT COMMUNICATION

Case report

Bilateral *Acanthamoeba* keratitis with late recurrence of the infection in a corneal graft: A case report

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PURPOSE. *To report a case of bilateral* Acanthamoeba *keratitis with late, atypical recurrence after penetrating keratoplasty* à chaud.
METHODS. *A 23-year-old contact lens wearer was treated for bilateral* Acanthamoeba *keratitis and underwent penetrating keratoplasty in the right eye for descemetocele with impending risk of perforation. The postoperative course was uneventful and topical steroids were combined with neomycin and propamidine. Two months after the operation in the right eye the patient presented with active infection in the left eye. One month later recurrence appeared in the right eye, as a central corneal infiltrate in the graft.*
RESULTS. *Recurrences in both eyes were successfully treated with a combination of hexamidine and neomycin, and with polyhexamethylene biguanide respectively. The right eye was regrafted three months after the recurrence and penetrating keratoplasty was done two years later in the left eye. Both grafts were successful and remained clear. There has been no further recurrence in the long-term follow-up.*
CONCLUSIONS. *Recurrence of* Acanthamoeba *keratitis after penetrating keratoplasty à chaud may occur even several months after the operation and the manifestation may be atypical. Current antiamoebal therapy was effective and regrafting in the quiet eye was successful. (Eur J Ophthalmol 2003; 13: 311-14)*

KEY WORDS. Acanthamoeba*, Keratitis, Penetrating keratoplasty*

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INTRODUCTION

Acanthamoeba keratitis is currently treated with the combination of a topical antiseptic biocide (polyhexamethylene biguanide - PHMB, chlorhexidine) and a diamidine (propamidine, hexamidine) (1). In cases with impaired vision due to corneal scarring, penetrating keratoplasty (PK) is usually postponed until resolution of the infection in order to avoid the risk of recurrence of the infection in the graft. However, in advanced cases PK may be necessary during the acute phase of the infection to prevent corneal perforation, and even with appropriate postoperative antiamoebal therapy the risk of recurrence in the graft is high (2). Recurrence in the graft is usually early and aggressive, manifested mostly as peripheral graft stromal infiltrate or as elevated epithelial lines (3,4).

We describe a case of bilateral *Acanthamoeba* keratitis with late recurrence and atypical presentation in the graft three months after PK for impending corneal perforation. Recurrence appeared as a central corneal ring infiltrate with ulcer in the graft. The other eye had shown a recurrence of the *Acanthamoeba* infection one month before.

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Case report

A 23-year-old man, wearing daily soft contact lenses for myopia, complained of pain, photophobia, and tearing in his right eye (RE) in April and one month later in the left eye (LE). The patient was treated by a specialist for suspected herpetic keratitis with topical acyclovir and antibiotics, and later also with topical corticosteroids. As the condition worsened, *Acanthamoeba* was suspected and treatment was started with neomycin and 5% povidone iodine (Betadine).

The patient improved and was subsequently referred to us in July for descemetocele, with a risk of corneal perforation in the RE. He denied rinsing the lenses in tap water but admitted to having taken a shower while wearing the lenses several times. On examination he reported no pain, and visual acuity (VA) was hand motion in both eyes. The RE was mildly irritated with central leucoma and descemetocele; the LE was quiet, with a dense corneal leucoma and central corneal thickness 450 microns (Fig. 1).

One day later penetrating keratoplasty (PK) was done in the RE, with diameter 8.25/8.5 mm. Postoperative treatment was based on topical 0.1% dexamethasone four times daily, a cycloplegic once daily, propamidine (Brolene) and neomycin four times daily and systemic ketoconazole 400 mg daily. Half of the corneal button was sent for histological examination and the other half was cut into small pieces and cultured in non-nutrient agar seeded with *Escherichia coli.* Histology showed cysts in the stroma (Fig. 2); the culture was negative. LE was left untreated and followed.

The postoperative course was uneventful and in September the graft was clear with RE best corrected visual acuity (BCVA) 0.6. Shortly after, however, the patient returned for sudden worsening of the LE with pain, photophobia, inflammation and central corneal ulcer. Corneal scraping was positive for *Acanthamoeba*, so hourly topical hexamidine (Desomedin) and neomycin were immediately initiated. The patient improved quickly and the ulcer healed in a few weeks.

A month later, when the LE was already quiet and the corneal ulcer healed, the patient returned again for sudden impaired vision in the operated RE tearing, pain and photophobia. The eye was inflamed, with a central corneal ulcer accompanied by a ring infiltrate (Fig. 3). Corneal scraping was positive for *Acanthamoeba* and hourly topical treatment was started with propamidine (Brolene) and PHMB. The eye improved quickly and one month later the ulcer was healed, with no more symptoms or inflammation. However, the graft remained opaque and was considered failed.

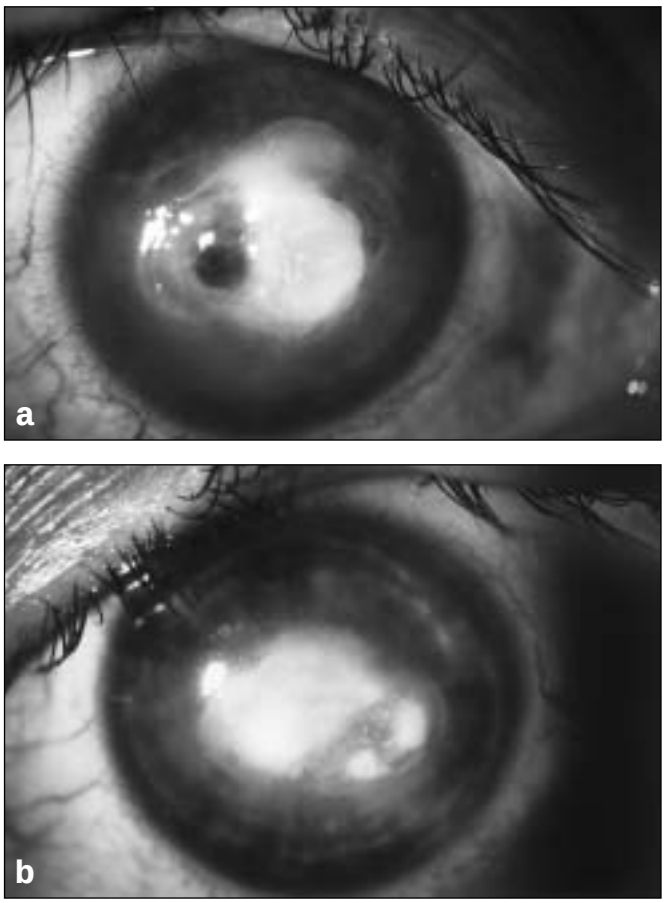

Fig. 1 - *Initial clinical presentation.* ***a****) The RE is mildly irritated with central leucoma and descemetocele.* ***b****) The LE is quiet, with dense corneal leucoma.*

The RE was regrafted in January. Culture of the graft was negative, though some cysts were found in the stroma, but they were considered empty and dead by an expert microbiologist. No postoperative antiamoebal treatment was employed. One year later the suture was removed and BCVA was 1.0, with a clear graft (Fig. 4). The LE underwent PK two years later, with no postoperative antiamoebal treatment. The postoperative course was uneventful and BCVA after suture removal was 1.0.

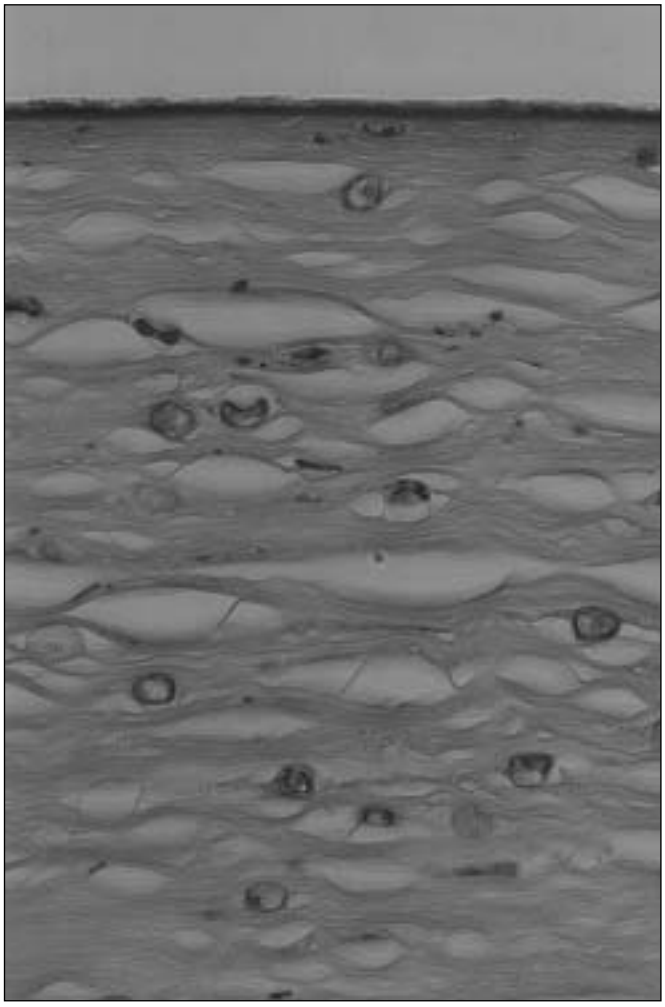

Fig. 2 - *Cysts in the stroma of the corneal button from the RE.*

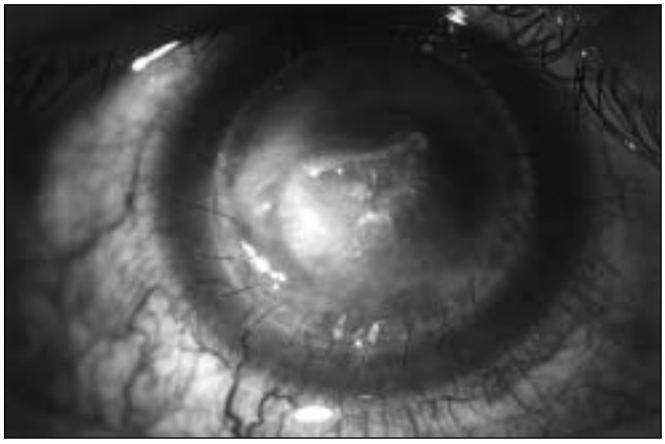

Fig. 3 - *Recurrence of* Acanthamoeba *keratitis in the RE.*

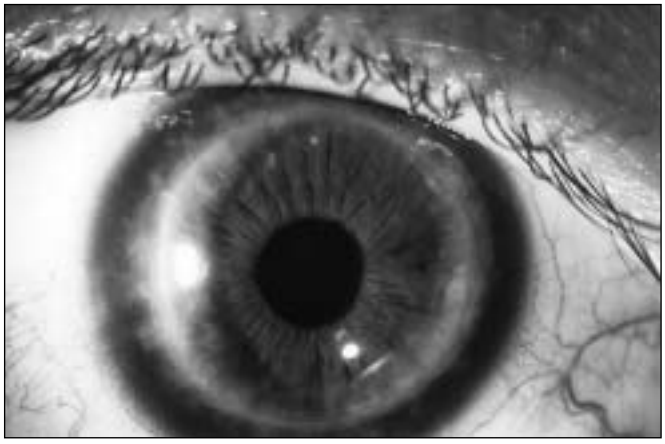

Fig. 4 - *The RE after suture removal, BCVA 1.0.*

DISCUSSION

Several antibiotics and antiseptics, including povidone iodine, have proved effective *in vitro* on *Acanthamoeba* cysts and trophozoites (1, 5). *Acanthamoeba* keratitis is currently treated with a combination of a diamidine and an antiseptic biocide and the prognosis is favourable if diagnosis is made early and appropriate therapy is applied (1). When corneal leucoma persists, PK is recommended several months after discontinuing treatment, when there is complete resolution of clinical signs and symptoms, in order to avoid the risk of recurrence of the infection in the graft (3). We are not aware of any general agreement on the need for postoperative antiamoebal treatment in these cases but these drugs are usually administered as it is impossible to prove there are no residual cysts in the recipient stroma.

In severe and advanced cases PK may be necessary even during the acute phase of infection, to avoid corneal perforation, with considerable risk of complications such as recurrence of *Acanthamoeba* in the graft, rejection, epithelial defects, glaucoma, secondary infections, scleritis and wound dehiscence (3, 6). In these cases antiamoebal treatment is recommended, with topical steroids to prevent inflammation and rejection. However, antiamoebal drugs may have toxic effects and the risk of recurrence in the graft is high even with this postoperative therapy (3).

We describe a case of PK performed for impending corneal perforation, in a patient given moderate postoperative topical antiamoebal drugs (four times dai-

ly) in view of the lack of clinical signs of persistent infection, negative culture of the corneal button, and previous treatment with topical neomycin and povidone iodine, the latter usually being effective on *Acanthamoeba* trophozoites and cysts, at least *in vitro* (5).

Recurrence of *Acanthamoeba* in the graft is usually early and aggressive, manifested mostly as peripheral graft stromal infiltrate or as elevated epithelial lines (4). This patient experienced late recurrence with atypical presentation, the infection manifesting as a central corneal infiltrate, three months after uneventful PK and a few weeks after recurrence of *Acanthamoeba* in the fellow eye. Topical treatment was successful in treating the infection but the graft lost its clarity. Regrafting after several months was successful and the patient regained 1.0 of vision.

This case raises some questions about the most appropriate post-PK antiamoebal treatment and the pathogenesis of recurrence. Usually *Acanthamoeba* recurs in the graft within the first month after the operation, and is considered to be caused by residual amoeba spreading from the recipient stroma into the graft (3). In this case we could not confirm whether recurrence was caused by residual cysts in the recipient stroma which led to an atypical late recurrence, or by reinfection from the fellow eye; as far as we know, transmission from the fellow eye has never been reported.

We conclude that *Acanthamoeba* keratitis in a corneal graft can recur even several months after the operation and may present as a peripheral stromal infiltrate, as well as a central infiltrate. It is not clear what is the most appropriate antiamoebal treatment after PK *à chaud* but regrafting after treatment of the recurrence usually has good chances of success (1-3).

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