CASE REPORT

Anterior segment optical coherence tomography of intrastromal corneal donor lenticule for recurrent intrastromal epithelial inclusion cyst in a corneal graft

Rebika Dhiman, Ganesh Pillay, Seema Kashyap, Murugesan Vanathi

Dr R P Centre for Ophthalmic Sciences, All India Institute of Medical Sciences, New Delhi, India

Correspondence to
Dr Murugesan Vanathi,
vanathi_g@yahoo.com

Accepted 14 May 2015

SUMMARY

Intracorneal cysts are uncommon lesions that occur either due to congenital or traumatic pathology. Treatments described include cyst excision, drainage or intracyst injection of chemical solutions, such as ethanol. Recurrences commonly occur with these modalities and there is no clear consensus on the best approach to take. In this article, the authors present a rare case of a recurrent intrastromal epithelial corneal cyst following suture removal in an 11-year-old boy who had undergone optical penetrating keratoplasty in his left eye. It was managed effectively with an innovative technique of lamellar excision and donor corneal stromal lenticule lamellar placement to prevent recurrence. Serial postoperative anterior segment optical coherence tomography (ASOCT) was performed to monitor for cyst recurrence. No recurrence was observed for one and a half years with clear corneal graft. ASOCT is an effective tool in the diagnosis and follow-up of a recurrent intrastromal epithelial inclusion cyst.

Treatments described include cyst excision or drainage. Recurrence rates can be high and the surgery itself may have negative visual consequences.⁷ Intracyst injection of chemical solutions, such as ethanol, has been effective in destroying the epithelial cell wall, resulting in a lower recurrence rate.⁸

We report a rare case of a recurrent intrastromal epithelial inclusion cyst of the cornea at the graft host junction in a case of penetrating keratoplasty following suture removal which was managed with an innovative technique of focal anterior stromal flap lift, deep lamellar dissection of the residual corneal stroma containing the epithelial cyst and intrastromal lamellar donor corneal stromal lenticule placement. To the best of our knowledge, this rare case report is the first to describe the occurrence of an intrastromal corneal epithelial inclusion cyst in a full thickness graft at the previous suture site. Anterior segment optical coherence tomography (ASOCT) proved to be a useful aid in the diagnosis and serial follow-up.

BACKGROUND

The first description of corneal cysts was by Appia¹ in 1853. Intracorneal cysts commonly occur due to sequestration of corneal limbal cells in the stroma either due to congenital or traumatic pathology.² They have been reported to occur following cataract surgery,³ lamellar keratoplasty⁴ and eye muscle surgery.^{5, 6} Histologically, corneal cysts comprise a cavity filled with necrotic epithelial cells.²⁻⁴ They are slow-growing, painless unilateral lesions that can result in visual disturbance and cosmetic deformity. Though surgical intervention is mandated in vision-threatening cases, there is no common consensus on the best approach to take.

CASE PRESENTATION

An 11-year-old boy, who had earlier undergone an optical penetrating keratoplasty in his left eye for a postinfectious keratitis corneal scar, presented to our keratoplasty follow-up clinic with a history of a slow-growing whitish lesion in the cornea for 18 months. The patient's history revealed that suture removal had been carried out one and a half years following penetrating keratoplasty in the left eye. A retrospective review of clinical records revealed the presence of a small intrastromal cyst in the nasal aspect of the corneal graft close to the graft host junction at the 9 o'clock position with a visual acuity of 20/30 not improving further with

Figure 1 (A) Slit lamp biomicroscopy photograph of the left eye of a patient showing an intrastromal cyst with fluid level. (B) Anterior segment optical coherence tomography photograph of the left eye of a patient showing the mid to posterior stromal location of the lesion.

To cite: Dhiman R, Pillay G, Kashyap S, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2014-209273

Figure 2 Histopathological examination of an eosin-stained specimen showing a cavity filled with necrotic debris lined by a non-keratinised stratified squamous epithelium. (H & E stain X100)

refraction. As it was not affecting the visual axis or interfering with vision, no intervention had been performed and a decision to observe the lesion had been made. Over the following one and a half years, the lesion gradually increased in size. Clinical evaluation showed an uncorrected distance visual acuity (UCDVA) of 20/60 in his left eye and 20/20 in his right eye. The best corrected distance visual acuity (BCDVA) was 20/30 with a refractive correction of +1.5 dioptre sphere and -2.25 dioptre cylinder at 180° axis. Slit lamp biomicroscopy revealed an intrastromal cystic lesion in the nasal aspect of the graft, involving the mid-stroma and posterior stroma, measuring 4×4.2 mm, extending into the host at the site of the previous 9 o'clock suture (figure 1A). Fluid level was seen in the cyst, 2 mm in height, with a turbid appearance. The corneal graft size

was 8 mm, with the graft remaining clear in the other uninvolved regions. Two retained 10-0 monofilament nylon sutures were seen at the 2 o'clock and 7 o'clock positions. On closer examination, the lesion was noted to have a crystal-like appearance at the previous suture site. ASOCT of the cyst was performed using a high-resolution Fourier-domain optical coherence tomography system with an axial resolution of 18 µm and broadband light of 1310 nm (Visante, Carl Zeiss Meditec, Inc, Dublin, California, USA). ASOCT confirmed the presence of a cystic lesion involving the mid-stroma and posterior stroma of the corneal graft in the left eye (figure 1B).

Considering the slow-growing nature of the intrastromal cystic lesion involving the corneal graft at the graft host junction, along with an area of crystalline appearance, a differential diagnosis of a slow-growing infective pathology was also considered, apart from an intrastromal corneal cyst that may have developed following suture removal for penetrating keratoplasty. Subsequent to a progressive increase in size of the intrastromal cystic lesion with impending encroachment into the pupillary area and a cosmetic blemish, a surgical intervention was conducted. Cyst aspiration with chemo-cytodestruction of the cyst cavity was performed with povidone iodine 1%.

A small biopsy was taken from the anterior wall of the cyst. Samples of the fluid and biopsy were sent for bacterial culture and BACTEC and histopathology examination. Smear and culture were negative for micro-organisms. Histopathology revealed a cavity lined by a non-keratinised stratified squamous epithelium confirming the diagnosis of an intracorneal epithelial inclusion cyst (figure 2).

Figure 3 Postcyst aspiration anterior segment optical coherence tomography showing the collapsed cyst with the clear graft.

Figure 4 Surgical steps of intrastromal donor lenticule placement. Step 1: lamellar dissection anterior to the cyst. Step 2: trephine marking (5 mm) for lifting the anterior stromal flap after dissection. Step 3: anterior stromal flap reflected, deep stromal dissection performed, cyst excised followed by the Betadine treatment of the stroma. Step 4: stromal lenticule (4 mm) fashioned and positioned in the host bed. Step 5: anterior stromal flap repositioned over the lenticule. Step 6: anterior stromal flap sutured with a 10-0 monofilament suture.

Figure 5 Diagrammatic representation of the surgical steps of intrastromal donor lenticule placement. Step 1: 5 mm trephine marking made over the cyst region. Step 2: 5 mm anterior stromal flap raised. Step 3: lamellar posterior stroma containing the cyst dissected and excised. Step 4: 4 mm donor lamellar lenticule fashioned. Step 5: donor lenticule placed in the host bed and flap repositioned and sutured with a 10-0 monofilament suture.

The corneal graft was clear in the immediate postoperative period with the same being confirmed on ASOCT ([figure 3](#)). However, after an uneventful postoperative follow-up for 7 months, recurrence of the cyst with fluid was noted.

TREATMENT

As the initial cyst aspiration with cytodestruction had failed to prevent recurrence, a repeat aspiration was not planned. Instead, we planned to remove the recurrent epithelial inclusion cyst by an innovative technique of focal anterior stromal flap lift, deep lamellar dissection to excise the stroma containing the recurrent cyst and donor corneal stromal lenticule lamellar placement beneath the flap followed by suturing of the flap.

The overlying uninvolved anterior stroma was dissected to lift an anterior stromal lamellar flap. A partial thickness trephination mark was placed over the region of the intrastromal cyst using a 5 mm trephine. The underlying mid-stroma and deep stroma containing the cyst was excised by careful deep lamellar dissection up to the level of Descemet's membrane and the host bed was treated with povidone iodine. A 4mm donor corneal stromal lenticule was fashioned by free hand dissection and the underlying Descemet's membrane and overlying epithelium were removed. This was placed in the host bed beneath the flap. The anterior stromal lamellar flap was then repositioned over the donor lenticule followed by closure with 10-0 monofilament nylon sutures ([figure 4](#)). The diagrammatic representation of the surgery is depicted in [figure 5](#).

Figure 6 (A) First postoperative day slit lamp biomicroscopy picture of the left eye showing the well positioned intrastromal donor stromal lenticule. (B) Anterior segment optical coherence tomography showing the donor lenticule with the overlying repositioned flap.

Figure 7 (A) Slit lamp biomicroscopy of the left eye at one and a half years follow-up showing the recurrence of a cyst. (B) Anterior segment optical coherence tomography showing the recurrence of the intrastromal cyst, deeper to the donor lenticule.

OUTCOME AND FOLLOW-UP

Postoperatively, UCDVA was 20/30 with good graft clarity. Slit lamp examination and ASOCT at a subsequent serial three-monthly follow-up showed no evidence of cyst recurrence. The slit lamp biomicroscopic photograph at the 6-month postoperative period showed a clear intrastromal lenticule (figure 6A) which was corroborated by ASOCT (figure 6B).

After one and a half years, a hyperlucent space was noted in the posterior stroma deeper to the site of donor lenticule placement which was confirmed on ASOCT (figure 7A, B). As the patient was asymptomatic with good visual acuity and the cyst was small in size away from the visual axis, serial follow-up with observation was planned.

DISCUSSION

Intrastromal corneal epithelial cysts are uncommon lesions. Undetected trauma may force surface epithelial cells into the corneal stroma where they become isolated after closure of the wound. The sequestration of these cells in the corneal stroma and their subsequent slow proliferation account for the continued growth of the cyst. Metabolic activity of the corneal epithelium is greater in children than in adults, which explains the predilection for traumatic corneal cysts in younger individuals.^{3 9 10} In our case, the removal of the suture at the 9 o'clock position of the graft-host junction may have served as the source of introduction of limbal epithelial cells into the corneal stroma. The sequestration of the epithelial cells in the posterior stroma resulted in cyst formation, with the difficulty in lamellar separation of the stromal tissue being probably responsible for the slow-growing nature of the intrastromal cyst.

Various surgical treatments of intrastromal epithelial cysts have been reported. Treatment has been based on the depth, extent and location of the lesions. Simple incision and drainage of cysts has almost universally been recognised as a prelude to recurrence.^{3 9 11} Excision of the anterior cyst wall combined with cytoreductive treatment of the cyst bed has been associated with decreased cyst recurrence. If destruction of the epithelial cells is incomplete, the remaining epithelial cells proliferate and the cyst recurs.^{10–14} Injection of povidone iodine into the collapsed cyst cavity allows effective intracellular penetration. Maintenance of a closed system is important because diffusion of povidone iodine into the adjacent corneal stroma or anterior chamber may induce keratocyte and corneal endothelial cell injury or lead to inflammation. However, the exact concentration and duration of the various cytoreductive agents that may cause complete destruction of the epithelial cells in the cyst cavity has not been established conclusively. Thus, cyst recurrence is seen in a large proportion of these cases, as was also seen in our patient. This might be due to the proliferation of the remnant epithelial cells that were not effectively destroyed during the primary procedure.

Recurrence entails a more invasive approach for management. In our case, this was carried out by placing an intrastromal donor lenticule in the region of the pathology that was excised. In contrast to a penetrating/lamellar keratoplasty, this technique permits retention of the host endothelial as well as epithelial surfaces, thereby preventing various complications. However, recurrence of the cyst seems to be inevitable. After an initial good postoperative period for one and a half years, on serial postoperative ASOCT, a recurrent slow-growing space is noticeable distal to the implanted lenticule, thus emphasising the possibility of incomplete removal of the epithelial cell-containing cyst wall. Another possibility of repeated recurrences could be excessive metabolic activity of epithelial cells seen in the younger age group.

Learning points

- ▶ An epithelial intrastromal corneal cyst may occur as a rare, often remote complication following suture removal in a penetrating keratoplasty. Consideration of intrastromal epithelial cysts in the differential diagnosis of patients with corneal cystic lesions may allow for early surgical intervention.
- ▶ Anterior segment optical coherence tomography is an effective imaging tool to detect intrastromal epithelial inclusion cysts of the cornea. Imaging on followup visits can help to detect early recurrence to facilitate timely intervention.
- ▶ Our technique of placing an intrastromal donor lenticule may be a useful and less radical approach as against lamellar or penetrating keratoplasty. This technique, by preserving the host epithelium and endothelium, prevents the related complications and may offer an effective alternative.
- ▶ Care must, however, be taken to ensure complete cyst wall removal if a permanent cure is desirable.

Acknowledgements The authors would like to acknowledge the assistance provided by Sana Tinwala, MD, Department of Ophthalmology, RP Centre, AIIMS, India.

Contributors RD assisted in the writing of the manuscript. GP conducted the imaging required for the case documentation. SK carried out the histopathology for the case. MV is the treating surgeon following up the case; and was responsible for the drafting and conceptualisation of the manuscript.

Competing interests None declared.

Patient consent Guardian consent obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Appia A. Arch Ophtalmol Paris: 1853. Cited by Bischler V. Un cas de kyste congenital sclerocorneen. *Ophthalmologica* 1947;114:371–6.

- 2 Bloomfield SE, Jakobiec FA, Iwamoto T. Traumatic intrastromal corneal cysts. *Ophthalmology* 1980;87:951–5.
- 3 Reed JW, Dohlman CH. Corneal cysts. A report of eight cases. *Arch Ophthalmol* 1971;86:648–52.
- 4 Sanders N. Corneal intrastromal cyst. *Am J Ophthalmol* 1971;71:1138–9.
- 5 Miffin MD, Byers TL, Elliot R, *et al.* Surgical treatment of an intrastromal epithelial corneal cyst. *Cornea* 2001;20:222–5.
- 6 Bhatt PR, Ramaesh K. Intrastromal corneal limbal epithelial implantation cyst. *Eye* 2007;21:133–5.
- 7 Al-Towerki AE, Binali G. Traumatic epithelial inclusion cyst of the anterior corneal stroma. *Cornea* 2008;27:368–9.
- 8 Javadi AM, Sharifi A, Hashemian SJ, *et al.* Management of intracorneal epithelial cysts with ethanol irrigation and cyst wall excision: a clinicopathologic report. *Cornea* 2006;25:479–81.
- 9 Liakos GM. Intracorneal and sclerocorneal cysts. *Br J Ophthalmol* 1978;62:155–8.
- 10 Rao SK, Fogla R, Biswas J, *et al.* Corneoscleral epithelial cysts: evidence of developmental etiology. *Cornea* 1998;17:446–50.
- 11 Chan MY, Liao HR, Cheng FJ. Traumatic intracorneal cyst. *Ann Ophthalmol* 1989;21:303–5.
- 12 Mahmood MA, Awad A. Congenital sclerocorneal cyst. *Am J Ophthalmol* 1998;126:740–1.
- 13 Avni I, Cahane M, Blumonthal M, *et al.* Transformation of corneal epithelial cyst into anterior chamber implantation cyst and scleral cyst: a rare occurrence. *J Pediatr Ophthalmol Strabismus* 1989;89:303–6.
- 14 Gokhale NS. Management of recurrent intracorneal epithelial cyst with ethanol irrigation and vacuum assisted cyst wall excision. *Int Ophthalmol* 2013;33:429–33.

Copyright 2015 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- Submit as many cases as you like
- Enjoy fast sympathetic peer review and rapid publication of accepted articles
- Access all the published articles
- Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow