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Case Report

A Case of Transparent Cornea Maintained after Removal of Dislocated Graft from Descemet's Stripping Automated Endothelial Keratoplasty

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Keywords
Corneal transplantation · Keratoplasty · Corneal endothelium · Visual acuity

Abstract

Introduction: Corneal graft detachment is a major postoperative complication of Descemet's stripping automated endothelial keratoplasty (DSAEK). When a corneal graft becomes detached, corneal endothelial function generally fails, and repeat corneal transplantation is required. Herein, we report a rare case in which a transparent cornea was maintained after the removal of a dislocated DSAEK graft. ***Case Presentation:*** A 79-year-old woman with a residual lens cortex who had undergone cataract surgery was referred to our hospital. The cortex was removed, and bullous keratopathy progressed. Six months after the initial surgery, DSAEK was performed under topical anesthesia without any complications. Although the corneal graft had attached fairly well, it detached from the host cornea 3 weeks later. Two months after DSAEK, an air tamponade was used to treat the anterior chamber with single interrupted suturing; however, the graft detached again, except for the suture site. Because the detached cornea became cloudy in the anterior chamber, it was surgically removed 8 months after DSAEK. Accordingly, the host cornea transparency improved to a best-corrected visual acuity of 0.8 with a rigid gas permeable lens and a central corneal thickness of 580 µm. The corneal endothelial cell density was 995 cells/mm^2. ***Conclusion:*** Removal of the corneal graft from the dislocated cloudy graft improved the visual acuity of this patient after DSAEK. The condition of the cornea should be carefully monitored after corneal endothelial transplantation, even after the graft has been dislocated.

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Introduction

Descemet's stripping automated endothelial keratoplasty (DSAEK) is a surgical technique that transplants the corneal endothelium using Descemet's membrane [1]. DSAEK is favored over penetrating keratoplasty for corneal endothelial cell loss because it results in less astigmatism and better visual acuity outcomes and poses relatively fewer risks [2].

Graft detachment is a major DSAEK complication. The frequency of graft detachment has been reported to be 0–43%, showing a wide range depending on the reports' sample size [3, 4]. Suturing or air tamponade is necessary when the graft is detached [3, 5]. However, for patients for whom those additional treatments are ineffective, the corneal graft generally fails, and secondary keratoplasties are required to maintain corneal transparency [5]. Herein, we report a rare case of graft detachment after DSAEK; graft reattachment was not achieved by air tamponade with suturing, and graft removal alone restored corneal transparency and visual acuity.

Case Report

A 79-year-old woman was referred to our hospital 3 months after cataract surgery for treatment of corneal edema with iridodialysis; the lens cortex remained in the anterior chamber of her left eye. At initial presentation, the patient was conscious and in good general condition. She had previous treatment histories of colon and cecum cancer, and she had medication for hypertension. Slit-lamp examination revealed residual lens cortex in the anterior chamber, iridodialysis, and corneal edema in the left eye (Fig. 1a). Her best-corrected visual acuity (BCVA) was 0.2, and intraocular pressure was 11 mm Hg in her left eye. Corneal endothelial cell density (ECD) could not be evaluated using noncontact specular microscopy, suggesting that it was below the minimum threshold for detection (Fig. 1b). Her central corneal thickness (CCT) was 573 μm as measured by anterior-segment optical coherence tomography (CASIA2, Tomey, Japan) (Fig. 1c, d). Post cataract surgery, the patient was prescribed a regimen of 0.1% betamethasone four times daily, 0.5% tropicamide/0.5% phenylephrine once daily, and 2% rebamipide four times daily to stabilize intraocular inflammation. Despite these measures, corneal edema persisted, prompting iris suturing and cortex aspiration 4.5 months later as a preparation for DSAEK surgery.

Postoperatively, the patient's BCVA in the left eye was recorded at 0.1, with a notable increase in the corneal thickness to 722 μm (as shown in Fig. 1e–g). Given the lack of improvement in corneal edema, we identified the condition as bullous keratopathy. Six months after the initial cataract surgery, DSAEK with 8-mm Descemet's stripping was performed under topical anesthesia without complications. The ECD of the donor graft was 2,646/mm^2. Although the corneal graft attached well without any space, it started to detach from the host cornea 3 weeks after surgery (Fig. 2a, b). The patient was instructed to remain in a prone position; however, graft adhesion was not achieved. Two months after the DSAEK procedure, graft adhesion was attempted using air tamponade. However, 2 days after the initial air tamponade, the lower part of the graft detached from the host cornea. To address this issue, a single interrupted suture was placed using 10-0 nylon with air tamponade. The suture was inserted into the anterior chamber from the area below the graft where it was absent and passed through the graft and host from the endothelial side. However, despite this intervention, graft adhesion still failed to occur. The sutured part of the graft was attached to the host cornea, although the other areas remained detached, shrunken, and opacified. Eight

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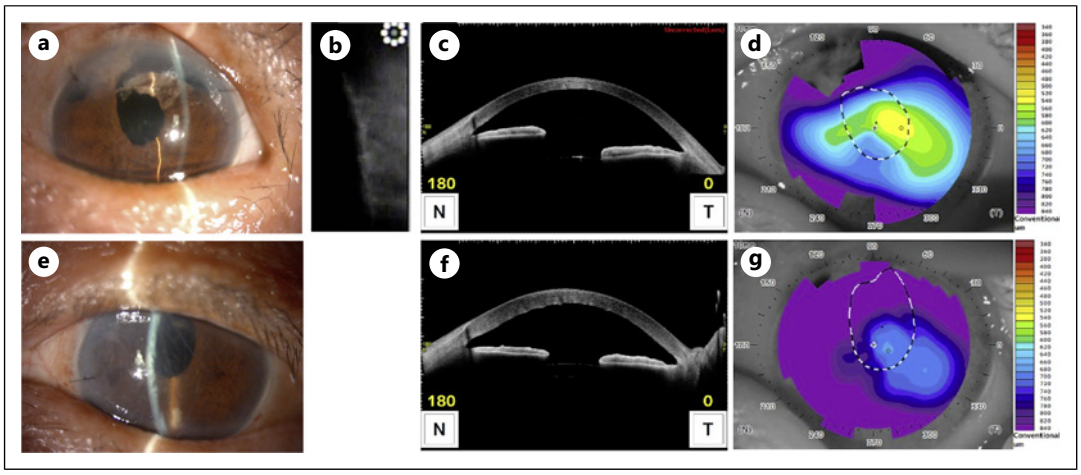

Fig. 1. Examinations of the left eye at the first visit and after iris suturing and cortex removal. **a** Slit-lamp findings of the left eye showing residual cortex, iridodialysis, and corneal edema. **b** Specular microscopy image taken during the initial visit, showing the cornea. Due to significant corneal edema, endothelial cells are not discernible in this image. **c**, **d** Corneal edema is observed in the left eye by anterior-segment optical coherence tomography. CCT is 573 μm (**c**) with asymmetrical corneal edema (**d**). **e** Slit-lamp photo of the anterior segment 6 days after the procedure. The cornea is thickened and edematous. **f**, **g** Corneal edema is observed in the left eye after iris suturing and cortex removal. CCT measured by anterior-segment optical coherence tomography is 722 μm.

months after DSAEK, although the transplanted graft was highly opacified, the host corneal edema and transparency improved (Fig. 2c, d). The CCT of the host cornea improved to 529 μm.

Therefore, we removed the graft 8 months after DSAEK surgery. The surgery was performed without any complications. In the pathological examination, the removed corneal graft included scarce corneal endothelium cells (Fig. 3a). One month after graft removal, corneal graft clarity was maintained (Fig. 3b). The BCVA improved to 0.7, and the CCT remained at 580 μm (Fig. 3c, d). Correction for the corneal irregular astigmatism with a rigid gas permeable lens resulted in a visual acuity of 0.8 10 weeks after graft removal. Subsequently, good corneal transparency and visual acuity were maintained for 4 months postoperatively, with an ECD of 995/mm^2 (Fig. 3e).

Discussion

Graft detachment is a major postoperative complication of DSAEK, and several treatments have been reported [3, 4]. In addition to surgical interventions, the noninvasive option of maintaining a prone position has been previously reported as effective, although there is insufficient evidence [6]. Therefore, we first attempted to treat the patient by asking her to remain in a prone position; however, this was not effective. Air tamponade is also a useful technique for treating graft detachment in DSAEK [3, 5]; however, 31% of patients' grafts fail after this technique [5]. In such patients, repeat keratoplasty is required for graft survival; however, this procedure is invasive and requires multiple corticosteroid doses. Therefore, instead of performing repeat keratoplasty after air tamponade failed in our patient, we performed graft removal from the anterior segment; this clarified the transparent cornea and unexpectedly improved the BCVA.

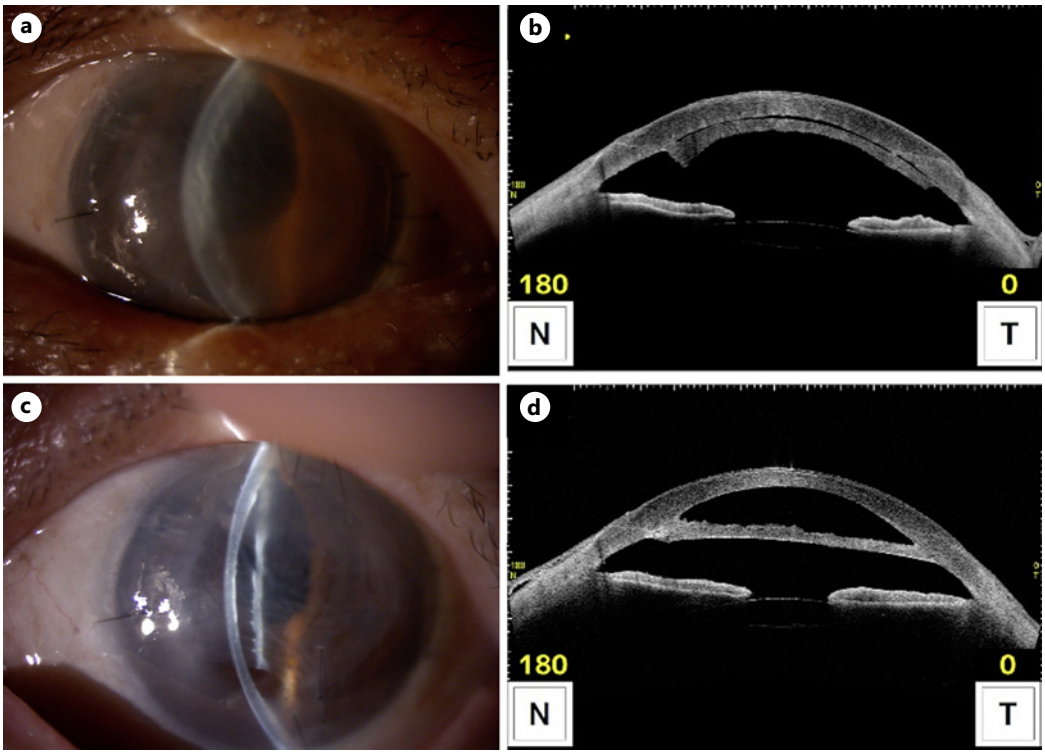

Fig. 2. Clinical images after Descemet's stripping automated endothelial keratoplasty (DSAEK). **a** Photo of the anterior segment 3 weeks after DSAEK. The graft is detached from the host cornea. **b** Anterior segment optical coherence tomography 3 weeks after DSAEK. The graft is detached from the host cornea. **c** Anterior segment after the second tamponade. The graft suture is observed in the inferior part. The transparency of the host cornea is improved. **d** Anterior-segment optical coherence tomography 7 months after DSAEK. The graft is detached and shrunken, although attached at the sutured part. The CCT of the host cornea is 529 μm.

Corneal endothelial cells migrate both in vitro and in vivo [7–9], and migration from the graft to the host cornea has been reported [7]. In this case, endothelial cells from the corneal graft possibly migrated to the host cornea, which may have restored the corneal endothelial pump function and maintained corneal transparency even after graft removal. This is supported by the results for the corneal endothelial cells, which could not be measured for detection during the initial examination because it was below the minimum threshold and became measurable after surgery. Scaffolds are required for cell migration. In this case, the graft was sutured to the host cornea at a second air tamponade. Although the graft was dissected, the graft suture site remained attached to the host cornea; the partially adherent sites may have functioned as scaffolds. Rho kinase inhibitors have been reported to promote the migration of corneal endothelial cells [9]. Although a Rho kinase inhibitor was not administered in this case, it may be possible to further induce the migration of corneal endothelial cells through this means in similar cases.

Jacobi et al. [7] have documented that endothelial cells can migrate from a DMEK graft to the area of Descemet's stripping after DMEK surgery. Daravagka et al. [10] have reported spontaneous clearance of central corneal edema after DMEK surgery with central graft detachment. Additionally, Ponce de Los Rios et al. [11] have reported endothelial cell repopulation in a 9-mm diameter descemetorhexis area in a DMEK case for non-Fuchs bullous keratopathy with a partially attached folded graft measuring 5.5 × 3.3 mm. Based on these

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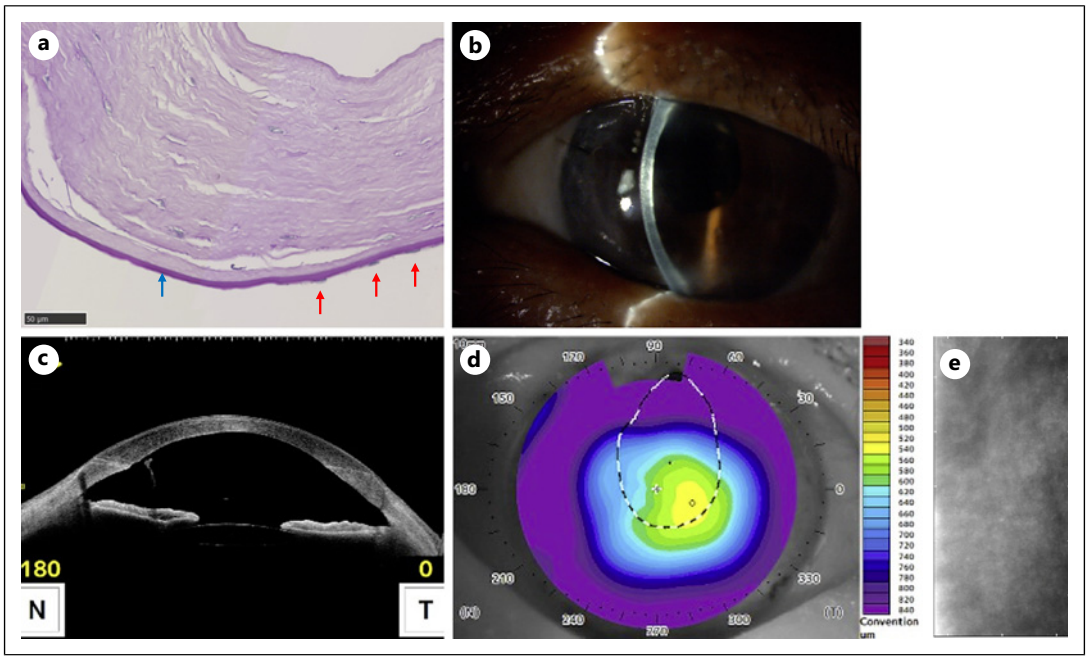

Fig. 3. Clinical images after corneal graft removal from the anterior chamber. **a** Excisional specimen histology (Periodic acid-Schiff [PAS], ×400) demonstrating preserved Descemet's membrane (blue arrow) and only a few remaining endothelial cells (red arrows). **b** One month post graft removal. Corneal transparency is maintained. **c**, **d** Anterior segment optical coherence tomography images of the patient 1 month after graft removal. Corneal thickness has improved to 580 μm, and corneal clarity is maintained. **e** Specular microscopy image captured after the removal of the graft. This image shows the corneal endothelial cells in the central area of the cornea with a density of 995 cells/mm^2, where Descemet's membrane stripping was performed.

findings, along with the observation of endothelial cells in the central area following an 8-mm Descemet's stripping in our case, it seems likely that endothelial migration from the graft to the central area via peripheral area could occur following graft detachment. This is particularly true if the peripheral cornea is incapable of contributing endothelial cells to the central area due to decompensation.

The report by Kinoshita et al. on cell injection therapy highlights the potential of using injected cultured allogeneic corneal endothelium to engraft and restore corneal ECD [12]. The therapy is effective when the patient assumes a prone position for some time. This finding underscores the possibility of corneal recovery facilitated by endothelial cells present in the anterior chamber. Moreover, it suggests that their attachment to the cornea could promote repopulation in areas devoid of endothelial cells.

In our case, we noticed that the transplanted tissue underwent shrinkage and lost elasticity, which caused it to straighten out, sometime after graft suturing. Given that elastin's role in tissue elasticity has been associated with the phenomenon of DMEK graft rolling [13], this observation prompts further inquiry. While reports of grafts straightening and shrinking post-DSAEK, as in our case, are uncommon, it raises questions about the involvement of factors like elastin in the migration or detachment of corneal endothelial cells from the graft and the resultant morphological changes.

Although there are no previous reports of graft removal, and the evidence is insufficient, our case suggests that in some patients who experience graft failure after DSAEK, re-transplantation can be avoided by following graft sutures with graft removal. When

rebubbling and suturing fail in DSAEK graft detachment, it might be beneficial to keep the patient under observation for a period, anticipating potential endothelial cell migration from the graft to the host cornea.

In conclusion, we reported a patient who had graft failure after DSAEK in which the cloudy graft was removed, and visual acuity improved. This case suggests that corneal graft removal from a dislocated cloudy graft can improve patient visual function for patients with graft failure after DSAEK. The CARE Checklist has been completed by the authors for this case report, attached as online supplementary material at https://doi.org/10.1159/000539392.

Statement of Ethics

This study was conducted in accordance with the guidelines of the World Medical Association's Declaration of Helsinki. This retrospective review of patient data did not require ethical approval in accordance with the local and national guidelines. Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

Y.N. and T.O. reviewed the medical records and wrote the manuscript. T.M. performed the surgical treatments, examined the patient, and wrote the manuscript. Y.A. and A.K. performed the pathological examination. Y.T., T.T., M.K., and M.A. reviewed and edited the manuscript. All the authors have approved the final version of the manuscript for publication.

Data Availability Statement

All data supporting the findings of this study have been included in this article. Further inquiries can be directed to the corresponding author.

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