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Case Report

Visual Recovery and Endothelial Repopulation after DMEK Graft Removal and Vitrectomy for Late Endophthalmitis: A Case Report

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DMEK · Endothelium · Endophthalmitis

Abstract
Introduction: The aim of the study was to report a unique case with excellent clinical outcomes after late endophthalmitis following Descemet's membrane endothelial keratoplasty (DMEK) surgery requiring donor graft removal without replacement. ***Case Presentation:*** A 67-year-old female with a prior ocular history of bilateral cataract surgery, Fuchs endothelial dystrophy, and pseudophakic DMEK in the left eye presented with endophthalmitis 2 months after keratoplasty. DMEK graft removal without replacement with an intracameral washout, pars plana vitrectomy, intracameral, and intravitreal antibiotics resulted in an excellent visual outcome (20/25). ***Conclusion:*** This is a unique case of late endophthalmitis following DMEK surgery requiring graft removal and pars plana vitrectomy with excellent visual recovery without donor replacement.

Introduction

Postoperative endophthalmitis is a rare but potentially devastating infectious or sterile inflammatory complication after intra- and extraocular surgery. Delayed-onset postoperative endophthalmitis is defined as presentation later than 6 weeks after surgery. The rate of

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endophthalmitis after endothelial keratoplasty has been reported to be very low (0.2%) compared to penetrating keratoplasty (0.7%) [1]. Postoperative endophthalmitis and/or any secondary surgical intervention is known to have deleterious consequences on corneal endothelial survival and graft clarity. We report a unique case of late endophthalmitis following DMEK requiring pars plana vitrectomy and donor graft removal without replacement resulting in corneal clarity with excellent visual recovery.

Case Report

A 67-year-old female had a history of type 1 myotonic dystrophy (DM1 [dystrophia myotonica]) [2] and a prior ocular history of bilateral cataract surgery, Fuchs endothelial dystrophy (FED), and pseudophakic Descemet's membrane endothelial keratoplasty (DMEK) surgery in the left eye (OS). Prior to DMEK, the central corneal thickness was 796 microns and visual acuity was 20/200 OS. Preoperative endothelial cell analysis could not be performed due to the degree of edema and corneal clouding. Uneventful DMEK surgery with a Descemetorhexis of 8.0 mm was performed at the same institution. After DMEK surgery OS, the patient experienced prolonged corneal edema with a non-clearing cornea with visual acuity of 20/200 in spite of an attached graft and intensive topical steroid use. Two months after DMEK, she developed redness and purulent discharge of the left eye. The visual acuity was hand motion OS, and intraocular pressure was normal. Slit lamp examination OS showed mild conjunctival injection, a central hazy, and thickened cornea with a rolled Descemet's membrane containing whitish material centrally and 2 mm hypopyon in the anterior chamber (Fig. 1a). Anterior segment optical coherence tomography demonstrated a rolled DMEK graft containing hyperreflective conglomerate centrally (Fig. 1b). Fundus examination OS revealed a hazy view with moderate vitreous opacities and attached retina on B scan ultrasound (Fig. 1c).

The patient was referred to the retina service with a presumptive diagnosis of late endophthalmitis and was subsequently scheduled for pars plana vitrectomy. Removal of the rolled DMEK graft and an anterior chamber washout was performed via a peripheral corneal incision followed immediately by a pars plana vitrectomy. Initial view of the posterior pole was limited but cleared post vitrectomy to reveal no significant pathology other than a thin epiretinal membrane without traction (online suppl. Fig. 1; for all online suppl. material, see https://doi.org/10.1159/000541644). An aqueous and vitreous tap was sent for culture. Intravitreal vancomycin and ceftazidime and intracameral cefuroxime were instilled.

Immediate postoperative examinations of the left eye revealed an edematous cornea, normal anterior chamber and vitreous body, and an attached retina. Both the aqueous and vitreous cultures came back negative for any microorganisms. The patient refused further surgical interventions. Postoperative medical treatment consisted of topical moxifloxacine and dexamethasone. Eight months after DMEK graft removal and vitrectomy (10 months after the initial DMEK), the patient presented with excellent best corrected visual acuity of 20/25 OS. The cornea of the left eye was clear (Fig. 2a) and showed a normal thickness of 534 microns (Fig. 2b) with a central endothelial cell density of 868 cells/mm^2 (Fig. 2c). The CARE Checklist has been completed by the authors for this case report, attached as online supplementary material.

Discussion

The frequency of FED among individuals diagnosed with DM has been documented at a rate of 46% [2]. A shared genetic background and pathophysiologic mechanism of RNA-mediated toxicity between FED and DM1 has been proposed [3, 4].

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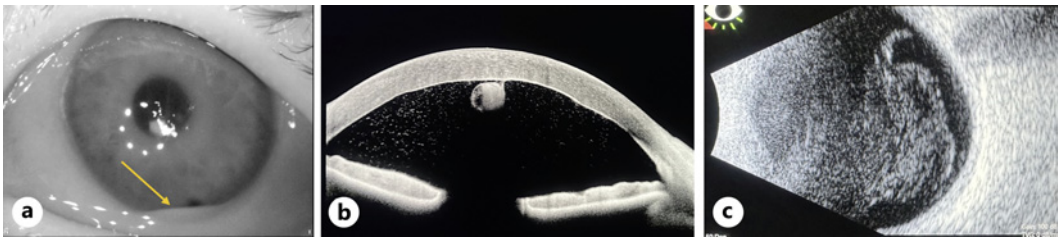

Fig. 1. Clinical findings of the left eye 2 months after DMEK. **a** Anterior segment photo showing semi-transparent cornea and hypopyon (yellow arrow). **b** Swept-source OCT of the thickened cornea with the rolled DMEK graft containing hyperreflective conglomerate and anterior chamber flare. **c** B scan ultrasound of the left eye with moderate vitreous opacities and attached retina.

Graft dislocation and rejection after both DMEK and DSEK is a well-known surgical complication that occurs more commonly in complex cases (e.g., preexisting glaucoma drainage tubes) [5]. The dislocation rate has also been reported to decrease with increasing surgical experience [6]. Spontaneous corneal clearance despite graft detachment after DMEK has been published [7–16]. Spontaneous corneal clearing after a large (standard) Descemetorhexis without endothelial replacement, while extremely rare, has been previously reported. Balachandran et al. [7] initially reported 2 cases of spontaneous clearing after DMEK detachment in 2009. Both cases had a large (>9.0 mm) Descemetorhexis. Visual acuity and corneal clearing improved despite continued graft detachment. Daravagka et al. [15] reported 3 cases of FED with cataract that underwent combined DMEK/phacoemulsification and cleared despite the DMEK graft never adhering. At postoperative month 3, all patients showed a decrease in central corneal thickness and an increase in endothelial cell density accompanied by an improvement in visual acuity. Shah et al. [16] reported a case of bilateral corneal decompensation where the first eye cleared after two unsuccessful DSEK attempts with both grafts detaching. A decision was made to only perform a large Descemetorhexis in the second eye which subsequently cleared. This case, however, is complicated by the fact that the pathology of the removed Descemet's showed a mixed picture of FED and posterior polymorphous dystrophy. Endothelial cells in posterior polymorphous dystrophy are known to have some epithelial-like proliferative capacity. Descemetorhexis without endothelial keratoplasty (DWEK) or Descemet's stripping only was introduced for the management of Fuchs' endothelial dystrophies in patients with guttae limited to the central cornea and a healthy peripheral endothelium [9, 10]. The Descemet's stripping is typically suggested to be no more than 4 mm to preserve less involved peripheral endothelium [12]. It relies on healthy peripheral endothelial cells migrating centrally and restoring normal corneal function. Previous studies demonstrated a mean time of corneal clearance of 3 months after DWEK [10, 11].

Vieira et al. [14] published their long-term results after DWEK and concluded that it is a safe and effective intervention in selected cases of early-stage central FED (central confluent guttae with a peripheral endothelial cell count >1,500 cells/mm^2 and a central corneal thickness <600 microns) [17]. Our case had both a preoperative corneal thickness well in excess of 600 microns (796 microns), and the prior Descemetorhexis (8.0 mm) was significantly larger than the suggested 4.0 mm size leaving a smaller area of intact peripheral endothelial cells behind and a 4 times larger surface area to repopulate.

Armstrong et al. [17] reported a case of *Propionibacterium acnes* endophthalmitis following DMEK surgery [18]. They concluded that re-transplantation may be a viable option

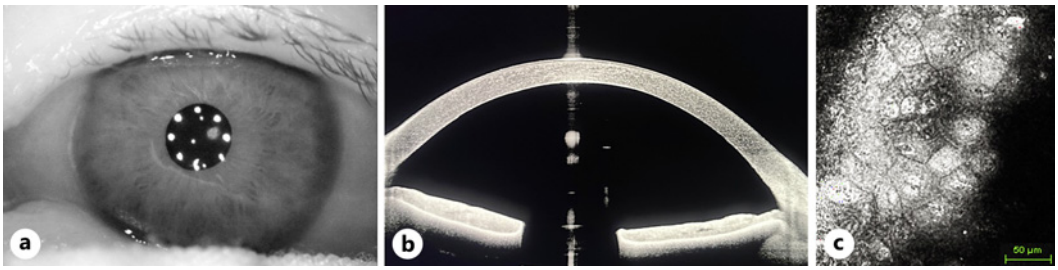

Fig. 2. Clinical findings of the left eye 8 months after DMEK graft removal and vitrectomy. **a** Anterior segment photo of the left eye showing transparent cornea and normal anterior chamber. **b** Swept-source OCT of the left cornea demonstrating normal pachymetry. **c** In vivo confocal microscopy photo of the central endothelial cells with polymegathism and pleomorphism.

with full visual recovery for DMEK patients who experience postoperative endophthalmitis [18]. In our case, spontaneous corneal clearance was observed 8 months after graft removal suggesting that in certain patients re-transplantation may not be needed. It should be highlighted that our patient did not receive a topical Rho kinase inhibitor as part of the medical treatment as this medication is currently not available in Hungary. Our case highlights that surgical intervention of repeat DMEK or DSEK (Descemet's stripping endothelial keratoplasty) in complicated cases where the donor tissue is removed may not be needed and with adequate time corneal clarity can be obtained.

While spontaneous clearing after a failed DMEK or DSEK has been reported, such clearing after a prior large Descemetorhexis is rare and difficult to explain based on current knowledge of endothelial regeneration or migration. While spontaneous corneal clearing and excellent visual acuity after a failed DSEK/DMEK are extremely rare, sporadic cases have been reported. While the normal corneal endothelium has limited regenerative capacity, as with most things in nature, there may be extremes of the bell curve, where some individual's regenerative capacity far exceeds the norm. Alternately, it is possible there was some endothelial transfer from the donor in spite of a detached graft. Patients reluctant to undergo further repeat surgery should be informed that in rare cases, given time, some visual recovery may occur.

Acknowledgment

We thank the patient for granting permission to publish this information.

Statement of Ethics

Ethical approval is not required for this study in accordance with local or national guidelines. Written informed consent was obtained from the patient for publication of the details of their medical case and any accompanying images.

Conflict of Interest Statement

The authors declare no conflict of interests.

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Author Contributions

E.S. and Z.S. are responsible for the design, manuscript preparation, literature search, data acquisition, and data analysis. A.C. and M.W.B. are responsible for the concept, definition of intellectual content, manuscript editing, and manuscript review. All authors read and approved the final manuscript.

Data Availability Statement

The data that support the findings of this study are not publicly available due to their containing information that could compromise the privacy of research participant but are available from the corresponding author (E.S.) upon reasonable request.

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Case Rep Ophthalmol 2024;15:736–741
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