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Tectonic keratoplasty with limbal allograft transplantation in non-traumatic, non-infectious corneal perforation associated with vernal keratoconjunctivitis

Vernal keratokonjonktivitte non-travmatik non-enfeksiyöz spontan kornea perforasyonu nedeniyle limbal allogreft ve tektonik keratoplasti uygulaması

İlayda Korkmaz Özlem Barut Selver Melis Palamar Onay

Ege University, Faculty of Medicine, Department of Surgical Medical Sciences, Department of Ophthalmology, Izmir, Turkey

ABSTRACT

To report the management of non-traumatic, non-infectious corneal perforation with limbal allograft transplantation and tectonic keratoplasty in a patient with bilateral vernal keratoconjunctivitis.

A 27-year-old male with bilateral vernal keratoconjunctivitis with accompanying limbal stem cell deficiency presented with redness, photophobia and discharge in the right eye. Best corrected visual acuity (BCVA) was 20/640 OD and 20/33 OS. Slit-lamp examination revealed bilateral corneal vascularization due to limbal stem cell deficiency and a 1.5 mm perforation at the paracentral lower temporal cornea of the right eye. Conservative treatment was unsuccessful. Therefore, tectonic keratoplasty with limbal allograft transplantation was performed. Two months later, the patient admitted to the hospital with redness, pain and blurry visual deterioration in the affected eye. Corneal graft was edematous and keratic precipitates on the corneal graft were evident. The patient was diagnosed as corneal allograft rejection and subconjunctival and topical intensive steroid treatment were initiated. One month after treatment, allograft rejection regressed, BCVA was measured as 20/125 OD. He is still stable for 18 months.

Keratoplasty combined with limbal allograft transplantation is one of the treatment options in patients with corneal perforation secondary to vernal keratoconjunctivitis accompanying with limbal stem cell deficiency. In these cases, the possibility of allograft rejection should be kept in mind. Patients and their relatives should be informed about the procedure and encourage to admit in any case of redness, pain or visual impairment in order to manage the rejection and provide a prolonged graft survival.

Keywords: Vernal keratoconjunctivitis, non-traumatic, non-infectious corneal perforation, limbal allograft transplantation, tectonic keratoplasty.

ÖZ

Bilateral vernal keratokonjonktivitli bir hastada, non-travmatik ve non-enfeksiyöz spontan kornea perforasyonu nedeniyle tektonik keratoplasti ve limbal allogreft uygulamasını ortaya koymaktır.

Bilateral vernal keratokonjonktivit ve eşlik eden limbal kök hücre yetmezliği olan 27 yaşındaki erkek hasta; sağ gözde kızarıklık, fotofobi ve sekresyon artışı şikayeti ile kliniğimize başvurdu. En iyi düzeltilmiş görme keskinliği (EİDGK) sağ gözde 20/640 ve sol gözde 20/33 düzeyindeydi. Biyomikroskobik muayenede limbal kök hücre yetmezliğine bağlı bilateral korneal vaskülarizasyon ve sağ gözde parasantral inferior temporal yerleşimli 1,5 mm'lik perforasyon alanı mevcuttu. Konservatif tedavinin başarısız olması nedeniyle hastaya limbal allogreft ile eş zamanlı tektonik amaçlı keratoplasti cerrahisi uygulandı.

Corresponding author: Özlem Barut Selver
Ege University, Faculty of Medicine, Department of Surgical Medical Sciences, Department of Ophthalmology, Izmir, Turkey
E-mail: *ozlembarutselver@yahoo.com*
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Postoperatif 2. ayda hasta kızarıklık, ağrı ve bulanık görme şikayeti ile başvurdu. Korneal greftte ödem ve keratik prespitatlar tespit edildi. Allogreft reddi gelişen hastaya subkonjonktival ve topikal yüksek doz steroid tedavisi başlandı. Tedaviden 1 ay sonra, allogreft red reaksiyonunun gerilediği ve sağ gözde EİDGK'nin 20/125 düzeyinde olduğu görüldü. Postoperatif 18 aylık takip süresince hastanın klinik durumunun stabil olduğu izlendi.

Vernal keratokonjonktivite bağlı gelişen limbal kök hücre yetmezliği zemininde ortaya çıkan non-travmatik ve non-enfeksiyöz kornea perforasyonunda limbal allogreft ile eş zamanlı tektonik keratoplasti etkin ve güvenli bir tedavi seçeneğidir. Bununla birlikte bu olgularda red reaksiyonu ihtimalinin yüksek olabileceği akılda tutulmalıdır. Hasta ve yakınlarının bilgilendirilmesi ile kızarıklık, ağrı, bulanık görme gibi olası red reaksiyonu semptomları varlığında erken başvuru yapmalarının sağlanması, red reaksiyonlarının tedavi edilebilmesinde faydalı olacaktır.

Anahtar Sözcükler: *Vernal keratokonjonktivit, non-travmatik ve non-enfeksiyöz korneal perforasyon, limbal allogreft, tektonik keratoplasti.*

INTRODUCTION

Vernal Keratoconjunctivitis (VKC) is a chronic bilateral allergic disease, which commonly occurs in young males (1). An atopy and associated allergic disease history are present in most of the VKC patients. VKC symptoms are characterized by photophobia, pruritus, ocular irritation, mucous secretion and blepharospasm (2). Usual clinical signs of VKC are; giant papillary reaction, limbal hypertrophy and upper tarsal conjunctival nodules (1). The mechanical effect of giant papillae causes corneal epithelial defects followed by scarring and neovascularization. Chronic inflammation because of eosinophil and mast cell activation also worsen the damage (3). Complications such as shield ulcer, keratoconus, and limbal stem cell deficiency (LSCD) may occur (4). Recently, non-traumatic, non-infectious corneal perforation has been recognized as a rare complication of VKC (5).

It was aimed to report the management of non-traumatic, non-infectious corneal perforation with limbal allograft transplantation and tectonic keratoplasty in a patient with bilateral VKC accompanying with LSCD.

CASE REPORT

A 27 year-old male with bilateral VKC accompanying with LSCD presented with redness, photophobia and discharge in the right eye. Best corrected visual acuity (BCVA) was 20/640 OD and 20/33 OS. Slit-lamp examination revealed bilateral corneal vascularization due to LSCD and a 1.5 mm perforation at the paracentral lower temporal cornea of the right eye with no infectious sign (Figure-1a and b).

Conservative treatment (patching, bandage contact lens and medical treatment) was ineffective. Therefore, tectonic keratoplasty with living related (mother) limbal allograft transplantation was performed two weeks after presentation. Interrupted 10-0 monofilament nylon suturation technique and 7.5-7.75 mm vacuum-punch trephines were used for the keratoplasty (Figure-1c). There were not any intraoperative complications. The patient received topical antibiotics, steroids, cyclosporine 0.05% and artificial tear drops postoperatively and was discharged from the hospital 5 days after the surgery. He did not attend the clinic visits postoperatively. Two months later, the patient admitted to hospital with redness, pain and blurry visual deterioration in the affected eye. Corneal graft edema and keratic precipitates on the corneal graft were present (Figure-1d). The patient was diagnosed as corneal allograft rejection, so subconjunctival and topical intensive steroid treatment was initiated. After treatment, allograft rejection findings regressed. BCVA was 20/125 OD one month after the allograft rejection and remained stable during the follow up (18 months) (Figure-1e).

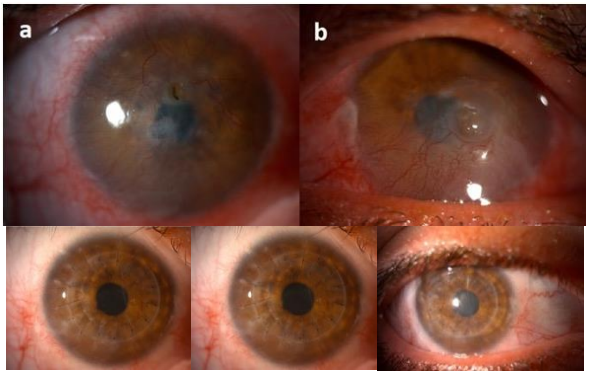

Figure-1. Anterior segment photograph of the right (a) and left (b) eyes of the patient at first examination. (c) Right eye after tectonic keratoplasty with limbal allograft transplantation. (d) One month after surgery; graft edema and keratic precipitates on corneal graft were present. (e) One month after the allograft rejection graft was translucent.

DISCUSSION

Non-traumatic, non-infectious corneal perforation is a rare clinical entity that may occur secondary to rheumatologic systemic diseases or ocular pathologies such as dry eye.

According to the literature non-traumatic, non-infectious corneal perforation secondary to VKC and associated LSCD is not a common condition (6). Matrix-metallo-proteinase (MMP) 2 and 9 have been shown in tear fluid in VKC (7). Also a correlation between MMP-9 and corneal involvement in VKC has been demonstrated (8). These mediators could be responsible for the non-traumatic, non-infectious corneal perforation secondary to VKC. There is only one report especially addressing cooccurrence of VKC and corneal perforation in the literature (5). In this case series, three cases were diagnosed as non-traumatic, non-infectious corneal perforation secondary to VKC. Two of the patients had keratoconus and the third patient had corneal scarring and astigmatism. First case was on steroid ointment for eyelid eczema, second case was on topical cyclosporine drops, while third case was not receiving any topical medication at the time of perforation, and the perforations were sudden at all cases. The treatment of the perforations was varied (bandage contact lens, amniotic membrane transplantation, keratoplasty) and successful healing of the cornea was achieved in all cases.

In the present case report, patient was not receiving any medication at the time of presentation and perforation occurred suddenly. Despite the conservative treatment applications, perforation persisted and required tectonic keratoplasty. When conservative treatment is insufficient or corneal perforation area is wide; penetrating keratoplasty is indicated in non-traumatic, non-infectious corneal perforations (9). In addition to providing anatomical integrity, it is also important to control the etiological factor that causes corneal perforation in order to provide graft survival (10). As in this case, penetrating keratoplasty combined with limbal allograft is an effective and safe treatment option in non-traumatic, non-infectious corneal perforations which may develop in the presence of VKC and accompanying LSCD. However, it should be kept in mind that the possibility of rejection is higher in these cases. Informing the patients and their relatives, and early treatment in a possible rejection reaction will be useful in order to control rejection reactions.

Conflict of interest

There is no conflict of interest.

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