

CASE REPORT / OLGU SUNUMU

A herpetic keratitis determined immunosuppressed patient with atypical presentation

İmmunsuprese hastada herpetik keratit ile atipik prezentasyon

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ABSTRACT

We aimed to report a case of atypical herpetic keratitis that developed after renal transplantation. A 48-year-old woman presented with redness, photophobia, tearing, foreign body, sensation and decreased vision in the left eye. The patient had undergone renal transplantation 6 months ago, was being treated by systemic immunosuppressants. (Cyclosporine A, mycophenolate mofetil and prednisolone acetate). Examination revealed Dellen ulcers and dendriform lesions which were parallel to the limbus, extending from the temporal area upper-quadrant to the nasal area. Based on the clinical findings, atypical herpes simplex virus keratitis (HSVK) was suspected. The subsequent corneal scraping and real-time polymerase chain reaction (PCR) analysis yielded a positive result for Herpes Simplex Virus Type 1 (HSV-1) genome. Topical and systemic acyclovir therapies were prescribed. One

week later, the dendriform lesions showed significant improvement and healed completely within 3 weeks. In immunocompromised patients, herpetic keratitis can appear in unusual presentations. Patient on immunosuppressive treatment with suspected herpetic keratitis, early topical and systemic antiviral therapy may prevent permanent vision loss and sequelae.

Keywords: Atypical herpetic keratitis, immunosuppression, renal transplant patients

ÖZ

Olgumuz, böbrek nakli sonrası gelişen bir atipik keratit vakası olup oldukça nadir görülmektedir. 48 yaşında kadın hasta, sol gözde kızarıklık, fotofobi, yabancı cisim hissi ve görme azalması şikâyeti ile tarafımıza başvurdu. Hastaya 6 ay önce böbrek nakli yapılmıştı, sistemik immün baskılayıcılar ile tedavi edilmekteydi. (siklosporin A, mikofenolat mofetil ve prednizolon asetat) Muayenede, korneanın üst temporal kısmından nazal kısma kadar uzanan, limbusa paralel Dellen ülseri ve dendriform lezyonlar tespit edildi. Klinik bulgulara göre, atipik herpes simpleks virüs keratitinden (HSVK) şüphelenildi. Kornea kazıma ve gerçek zamanlı polimeraz zincir reaksiyonu (PCR) analizi, Herpes Simplex Virus Tip 1 (HSV-1) genomu için pozitif bir sonuç verdi. Hastaya topikal ve sistemik asiklovir tedavisi verildi. Bir hafta sonra, dendriform lezyonlarda belirgin iyileşme başladı ve 3 hafta içinde kornea tamamen iyileşti.

Bağışıklık sistemi baskılanmış hastalarda, atipik herpetik keratit görülebilir. Erken dönemde topikal ve sistemik antiviral tedavi ile kalıcı görme kaybını önlenir.

Anahtar Sözcükler: Atipik herpetik keratit, immünoşüpresyon, böbrek nakli

INTRODUCTION

The herpetic keratitis is well appreciated that the major clinical problem in adults is recurrence of the infection, which increases the likelihood of severe stromal reactions with secondary vascularization and permanent structural alteration of the cornea.¹⁻³ HSV ocular infection may present in various clinical forms. Acute blepharoconjunctivitis with or without keratitis, recurrent corneal ulcers and recurrent stromal keratitis are common forms of the disease.

Herein, we present the unusual case of HSV keratitis with peripherally located dendritic ulcers and Dellen formation, caused by HSV-1 in a renal transplanted patient.

CASE REPORT

A 48-year-old woman presented with redness, photophobia, tearing, foreign body sensation, some mucoid discharge of left eye and decreased vision. The patient had undergone renal transplantation 6 months ago, was being treated by systemic immunosuppressants (Cyclosporine A, mycophenolate mofetil and prednisolone acetate). The patient had a previous history of oral herpes simplex infection. On ocular examination, the best-corrected visual acuity (VA) was 10/10 in the right eye and 5\10 in the left eye. Intraocular pressure (IOP) was 15 mmHg in the right eye and 14 mmHg in the left eye. Slit-lamp examination revealed conjunctival and ciliary injection, minimal corneal epithelial edema, rare punctuate epitheliopathy, dendriform lesions parallel to the limbus, extending from the temporal upper quadrant to the nasal area and Dellen ulcers in the left eye (Figure 1-2). There was no evidence of uveitis in the left eye. Slit-lamp examination of the right eye was normal. HSV infection was suspected in left eye and acyclovir treatment, both oral (200 mg, 5 times daily) and topical (3% ointment, 5 times daily) treatment was started. HSV DNA was isolated from the cornea swap sample using the spin colon technology (QIAamp MinEluteVirus Spin kit, Qiagen GmbH, Hilden, Germany). Afterwards, HSV DNA was determined by a commercial PCR assay (Artus HSV-1/2 RG PCR kit, Qiagen GmbH, Hilden, Germany) on the real-time platform (Rotor-Gene Q, Qiagen GmbH, Hilden, Germany) (Figure 3). One week later, the dendriform lesion showed significant improvement and dexamethasone (1mg/ml, 6 times daily) drop was administered in addition to antiviral therapy. 3 weeks later, the dendriform lesion and corneal epithelial edema were completely resolved and the spectacle-corrected VA was 10/10 in the left eye (Figure 4).

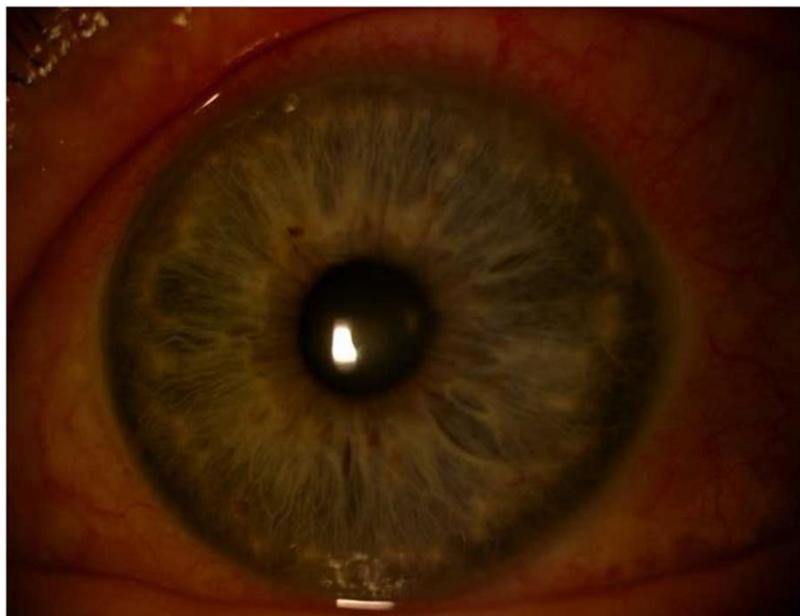


Figure 1: Pretreatment anterior segment photograph of the immunosuppressed patient shows ciliary injection and minimal corneal epithelial edema.

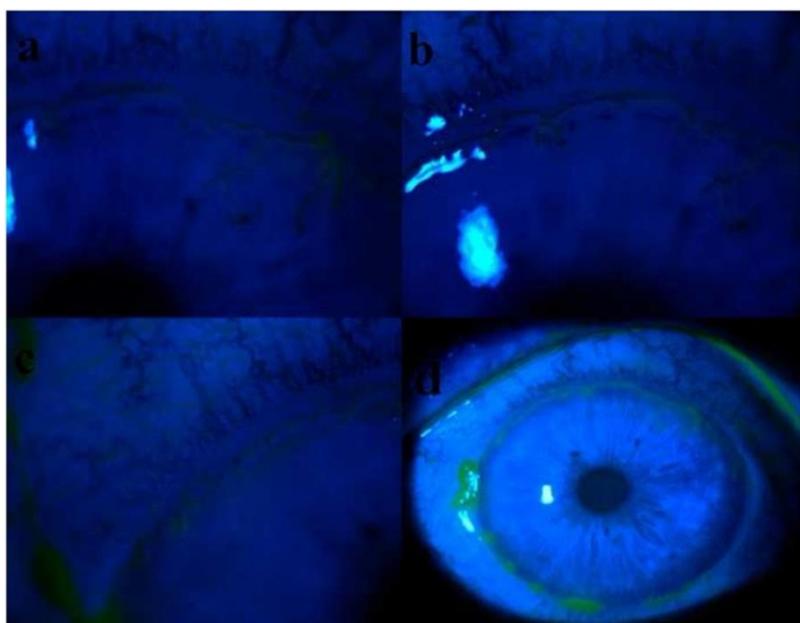


Figure 2: Pretreatment anterior segment photographs of the immunosuppressed patient a-c. Fluorescein staining revealed dendriform lesions running parallel to the limbus extending from the temporal upper quadrant to the nasal area. d. Dellen ulcers.

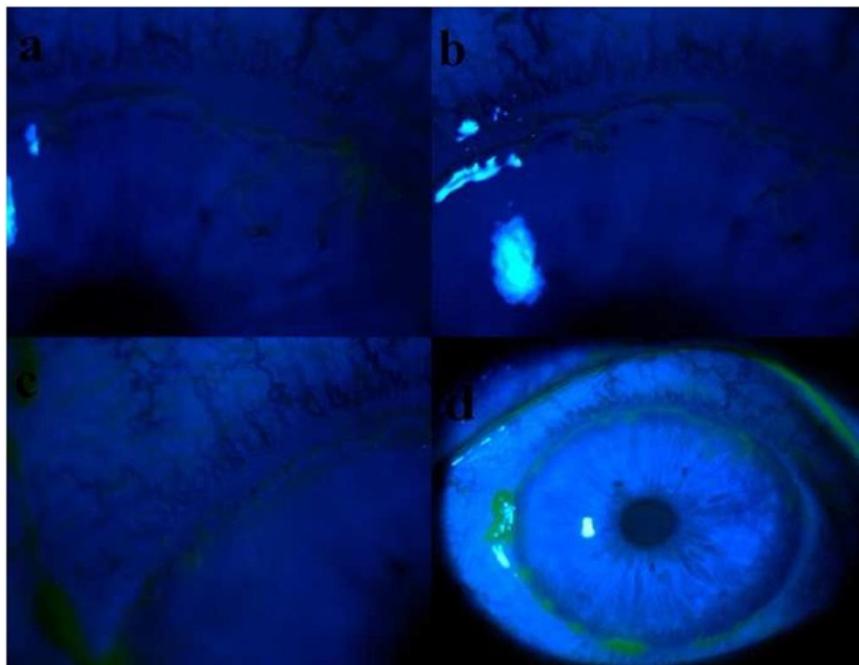


Figure 3: HSV-1 genome was detected by Real-time PCR

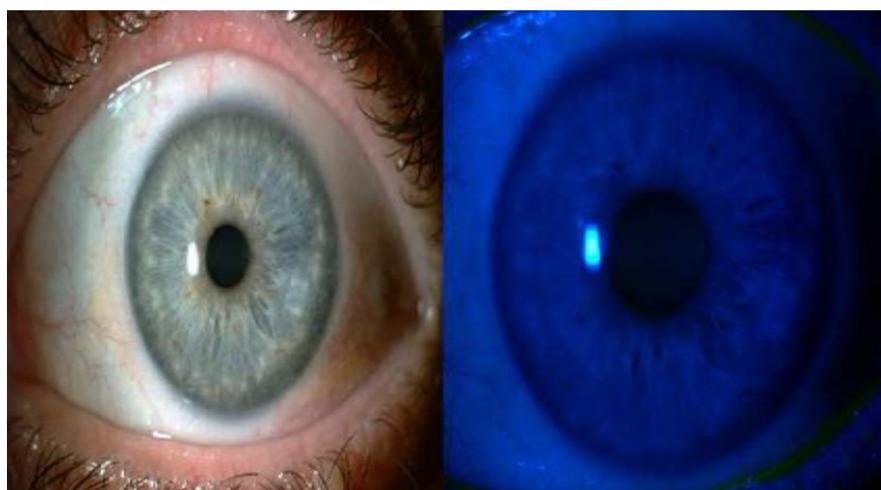


Figure 4: After three weeks of systemic and topical antiviral treatment, our patient's left eye completely healed, corneal epithelial edema had resolved.

DISCUSSION

Atypical presentations of HSVK have been reported.⁴⁻⁷ Clinical diagnosis of atypical HSVK can often be difficult. It may be seen in infections caused by HSV-1, HSV-2, VZV, bacteria and fungi. In our case, we suspected herpetic keratitis clinically. Subsequent virological investigations (PCR) confirmed the etiology (HSV-1) in this case. PCR is a technique for amplifying short regions of DNA between regions of known sequences.⁸ With PCR, HSV can be identified, and the efficacy of this method in the diagnosis of herpetic keratitis has been

described.^{9, 10} Also, PCR for HSV has been shown to be most useful method for the clinician in atypical presentations of herpetic ocular disease.^{4, 9, 11}

While Langerhans cells are abundant in the skin, conjunctiva and corneal limbus, they are absent in the normal central cornea of the adult human, mouse, rabbit and guinea pig.¹²⁻¹⁵ Following HSVK, Langerhans cells have been demonstrated in higher numbers in all regions of human and animal corneal epithelium including the central cornea.¹⁴⁻²⁰ Localization of dendritic keratitis in the central and paracentral cornea is thought to be occurred as a result of migration of Langerhans cells to the central cornea and other complex immunoreactions. Being immunosuppressed of our case may have caused atypical presentation of dendriform lesion and disruption in this pathological process.

It is interesting to note that it is very unusual for a herpes infection alone to present with peripheral ulcers and Dellen formation. In our case unlike other examples of literature peripheral localization of herpes lesions were accompanied by Dellen formation.^{6, 7} Due to the severe atypical HSVK in our case, destruction of the tear film layer was thought to cause Dellen formation. Such atypical findings might be related to the underlying systemic condition of our patient, as patients with a compromised immune system such as atopy, congenital immune deficiencies, or recipients of organ transplants, are apt to show unusual features during disease course.

This case highlights the following: Herpetic keratitis with is more often seen in patients who are immunocompromised and unusual features may develop.^{6, 7} These patients should have periodic and thorough ocular examinations especially if they are on immunosuppressive treatment, and clinicians should also be aware of such atypical features. PCR assay described in this report appears to be a useful tool for the rapid, sensitive, and specific detection of herpes virus. We believe that in immunosuppressed cases with suspected herpes keratitis, prompt institution of prophylactic topical and systemic antivirals can prevent permanent vision loss and sequelae.

Conflict of Interests: The authors declare no conflict of interests.

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