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Case report

Spontaneous corneal perforation complicating ocular rosacea: Case report

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ABSTRACT

Introduction: Ocular rosacea is a multifactorial disease. Its pathophysiology remains unclear.
The ocular manifestations of rosacea are not specific and can range from simple blepharoconjunctivitis to sight-threatening such as corneal perforation.
Case report: We report the case of a 10-year-old child who presented with a red painful right eye. Based on the clinical findings, we concluded that she had a corneal perforation on ocular rosacea. She benefited from an ipsilateral lamellar autokeratoplasty by lamellar autograft.
The evolution was marked by a good healing and a good visual recovery despite a corneal scar.
Discussion: Ocular rosacea is a multifactorial disease, with an unclear physiopathology. Corneal involvement remains the least common, but the most challenging since serious complications can occur. Corneal perforation is the most severe.
Several techniques have been reported and used in the management of corneal perforations such as conjunctival flap, amniotic membrane grafting, and the use of a corneal patch. The later, corneal autografting, remains a simple and effective technique with satisfactory anatomical results.
Conclusion: Ocular rosacea is a pathology that is still poorly understood and of delayed diagnosis.
It can lead to serious vision-threatening complications such as corneal perforation. The corneal patch is a simple, effective and efficient technique that has given good results in our case.

1. Introduction

Rosacea is a chronic inflammatory skin condition with a fluctuating course. It is characterized by flushing, transient erythema, papules, pustules and telangiectasias [1]. The physiopathology of this disease remains unclear at this date. The available treatments mostly act on the symptoms and leave the unknown underlying mechanism untouched [2]. The ocular manifestations of rosacea are not specific and can range from simple blepharoconjunctivitis to sight-threatening corneal neovascularization, with thinning and corneal perforation [3]. It must be diagnosed early, since severe functional sequelae are possible.

This study has been reported in accordance with the SCARE criteria [4].

2. Observation

This case involved a 10-year-old child with a history of herpetic keratitis at the age of 8 years without sequelae. The child presented a painful red right eye with decreased visual acuity, without any notion of trauma one day prior to her consultation. The ophthalmological examination of the right eye revealed an uncorrected visual acuity of counting fingers, the slit lamp examination revealed conjunctival hyperemia, blepharitis, phlyctenes, a corneal perforation of about 2 mm/2 mm in the infero-nasal area, partially sealed by the iris with a spontaneous seidel, superficial punctate keratitis in the inferior area with corneal infiltrates and a very shallow anterior chamber (Fig. 1).

Examination of the opposite eye revealed an uncorrected visual acuity of 6/10 and a meibomitis with follicular conjunctivitis. The rest of the examination was unremarkable (Fig. 2).

The parents were informed about the situation and gave their consent for the surgery.

The patient was then hospitalized. She underwent an ipsilateral lamellar autokeratoplasty by lamellar autograft.

After local disinfection and careful trimming of the bottom and edges of the perforation, a corneal lamellar graft was carefully dissected on the upper temporal quadrant and then transposed into the ulcerated area where it was fixed with single 10/0 filament stitches (Fig. 3).

The child was prescribed a 15-day course of oral antibiotics with

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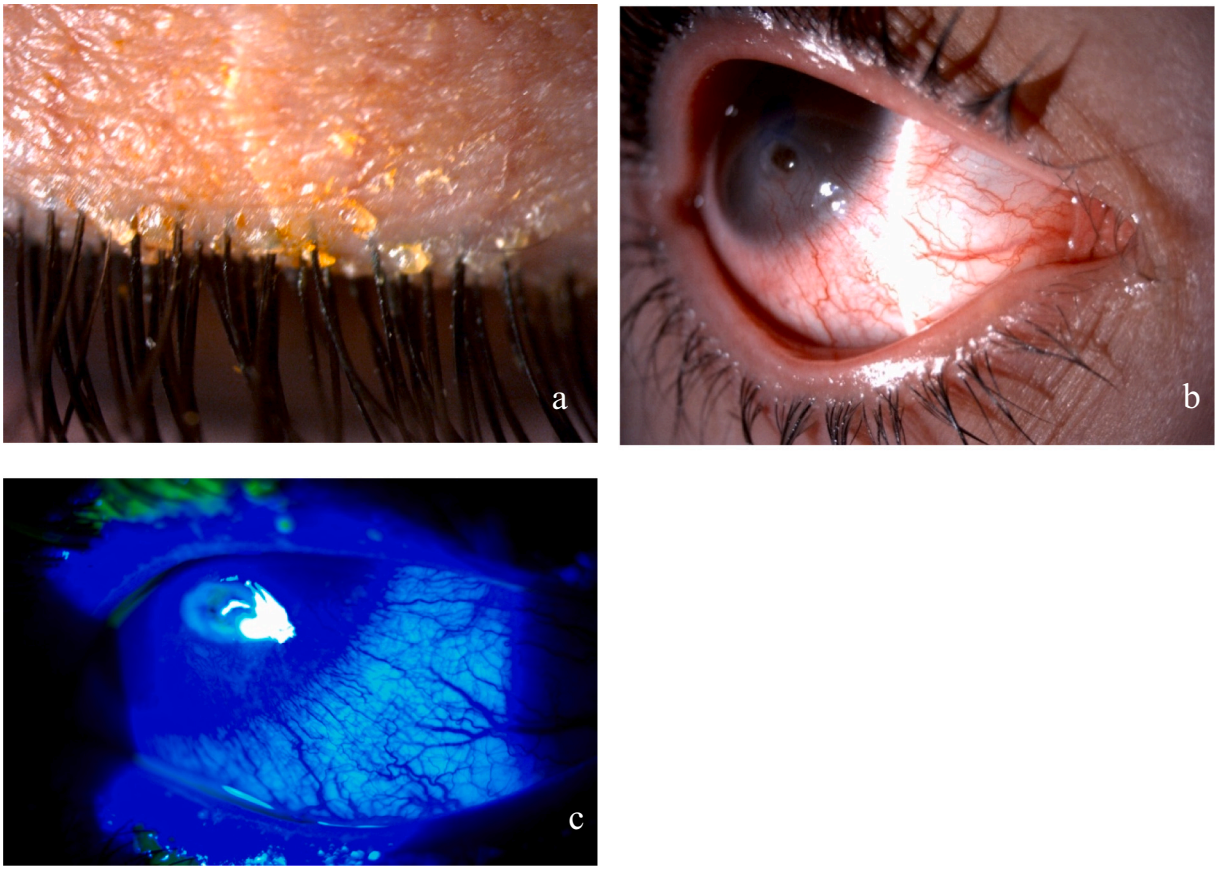

Fig. 1. (a): Slit lamp pictures showing anterior palpebral blepharitis with scaling sticking to the lashes.
(b): Slit lamp image showing an inferonasal corneal perforation sealed by the iris with neovascular appeal in front.
(c): Slit lamp image with cobalt blue light showing an inferonasal corneal perforation sealed by the iris with a spontaneous seidel.

Josamycin, a short course of local corticosteroids, 6 months of local antibiotics with azythromicin, artificial tears and 2 months of cycloplegia. We also insisted on the necessity of a good palpebral hygiene.

The outcome was satisfactory: healing of the graft, anterior chamber reformation and regression of inflammatory signs.

The ophthalmological examination of the right eye at 2 months postoperatively found a corrected visual acuity of 6/10, regression of blepharitis and a clear cornea apart from a slight scar at the site of the perforation (Fig. 4).

3. Discussion

Rosacea is a chronic inflammatory skin disorder with an unclear pathogenesis. The diagnosis of rosacea is usually clinically defined. Flushing, centro-facial erythema and papules are the most frequently observed. In 2002, the expert committee of the National Rosacea Society developed a classification of rosacea, describing 4 different subtypes: erythematotelangiectatic, papulopustular, phymatous and ocular rosacea [5].

Regarding ocular rosacea, 20% of patients will develop ocular manifestations initially, 53% have pre-existing skin involvement and 27% have combined ocular and skin involvement simultaneously [6]. Palpebral involvement remains the most frequent [7,8]. E. K. Akpek et al. reported that 81% of patients presented with telangiectasias of the free eyelid margin, 78% with meibomus gland dysfunction, and 65% with blepharitis [9]. Corneal involvement remains the least common, but the most challenging as shown in P. A. Starr and A. Macdonald study where 33% of rosacea patients had corneal involvement [10]. Superficial punctate keratitis is the most common corneal involvement. It can also lead to ulcerations, perforations, corneal spotting and corneal neovascularization. Its frequency ranges from 5% to 33%. Wise et al. reported that 67% of patients presenting with corneal complaints had corneal neovessels and infiltrates [8,1].

In children's cases, clinical signs are often misleading and may be limited to an isolated red eye without skin involvement. Symptoms may include blinking, pruritus, tearing and/or secretions. Ocular rosacea in children initially presents as blepharoconjunctivitis and as blepharokeratoconjunctivitis in more advanced forms.

Ocular rosacea is a multifactorial disease. Recent studies have revealed some factors that may be implicated in ocular surface disorders. Barton et al. demonstrated that the level of interleukin (IL)-1 is abnormally high in the tear fluid of patients with ocular rosacea. IL-1 is produced by corneal epithelial cells, lacrimal glands and inflammatory cells of the conjunctiva [11]. IL-1 increases the production of matrix metalloproteinases (MMP) such as gelatinases (MMP 2 and 9), collagenases (MMP 1, 8 and 13). Matrix metalloproteinases (MMPs) are proinflammatory endopeptides involved in corneal epithelial and stromal loss [12]. Rosacea patients with recurrent erosions, peripheral infiltrates and ulcers have high gelatinase B activity. IL-1 also alters the neurosensory threshold and reduces corneal sensitivity. This impaired sensitivity leads to decreased tear production, thus contributing to dry eye. The epithelial cells of the ocular surface, in response to the stress of a dry environment, produce IL-1 and MMP-9 resulting in further inflammatory reactions. All this leads to epithelial and conjunctival degeneration [13]. Interferon (IFN) may also play a role in ocular surface inflammation. This inflammatory factor leads to increased expression of HLA-DR and intercellular adhesion molecule-1 (ICAM-1) on the conjunctival epithelial cells membrane. Leonardi et al. demonstrated that the levels of eosinophilic cationic protein (ECP), eosinophilic neurotoxin (EDN), myeloperoxidase (EPO) and soluble interleukin-2 receptors were

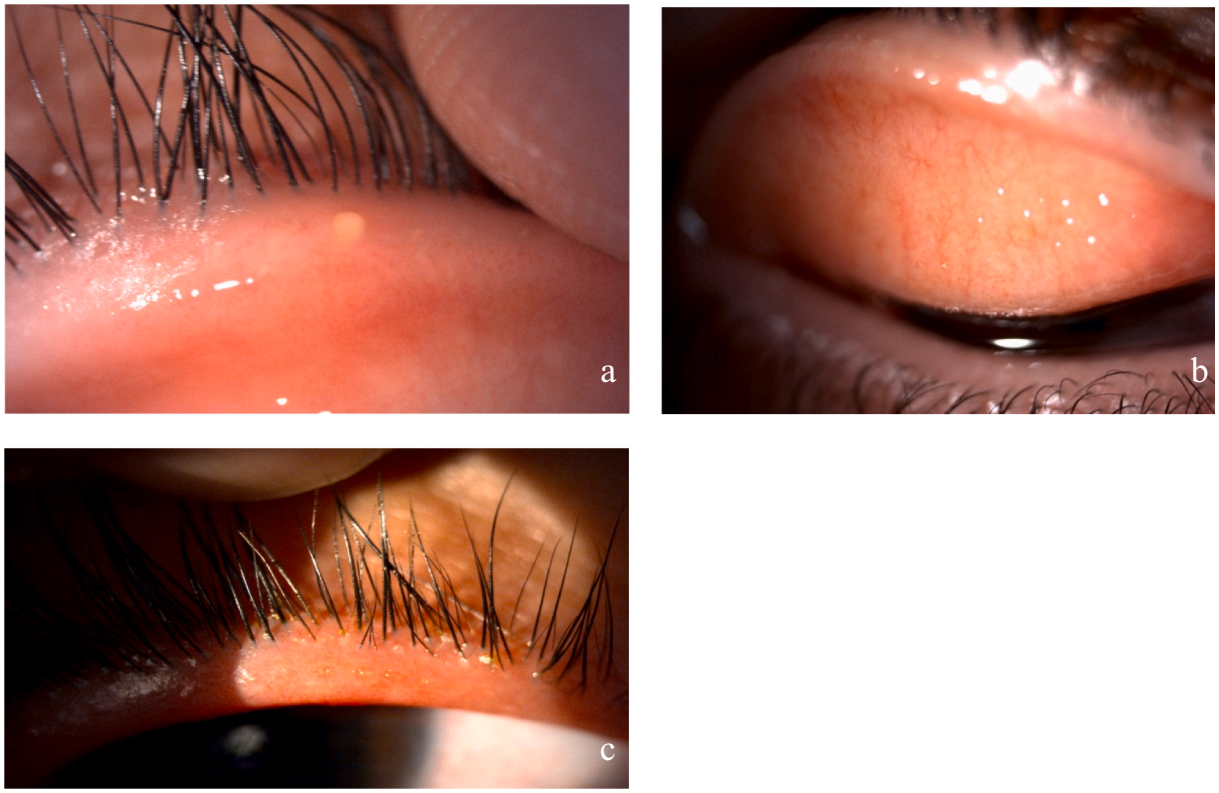

Fig. 2. (a): Slit lamp image showing thick meibom behind the gray line.
(b): Slit lamp image showing follicular conjunctivitis.
(c): Slit lamp image showing mixed blepharitis with some telangiectasias on the lid margin.

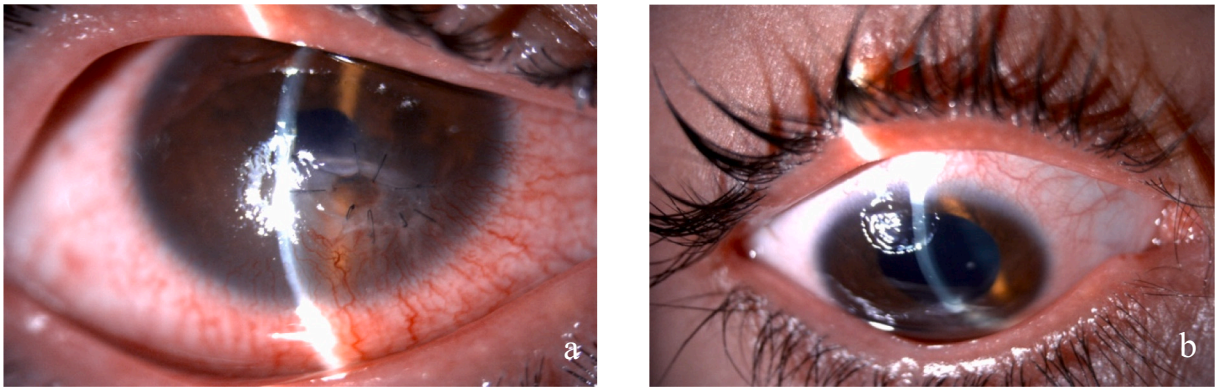

Fig. 3. (a): Slit lamp image showing ipsilateral lamellar autokeratoplasty by lamellar autograft.
(b): Slit lamp image showing the site of corneal sampling in the superior temporal region, larger than the size of the ulceration.

elevated in the tears of rosacea patients with blepharokeratoconjunctivitis [14,15].

Studies of additional pro-inflammatory agents in the skin of rosacea patients showed a production of cathelicidin (LL-37) and kallikrein (KLK5) [16,17]. In fact, LL-37 promotes inflammation, angiogenesis and neovascularization [18]. Although there is no immunohistological study yet on rosacea patients cornea, it can be hypothesized that LL-37 levels would be elevated in their tear film and could contribute to corneal neovascularization. A better understanding of the pathogenesis of this disease may help to find more effective therapeutic modalities [1].

Various surgical techniques have been described [3]. Gracner et al. reported cases of keratoplasty on extensive corneoscleral perforations complicating ocular rosacea [19]. Jain et al. described the use of amniotic membrane grafting for spontaneous corneal perforation in rosacea patient resulting in improved visual acuity at 3 months after surgery [20]. Conjunctival flaps have also been used to manage corneal perforation and pre-perforative ulcers in 2 patients with rosacea [21].

In our case, we used an autografting of a corneal patch. This technique is not widely described in the literature. It is indicated for corneal perforations, especially peripheral ones under 3 mm, when access to corneal grafts and amniotic membranes is difficult.

This technique consists of harvesting a graft of sufficient thickness and of greater diameter than the recipient area from a healthy area of the same cornea. This graft is then transposed and fixed using 10/0 monofilament sutures to the preperforative descemetocele, after a good trimming of the bottom and the edges of the area [22].

Corneal autografting remains a simple and effective technique with satisfactory anatomical results. [23]

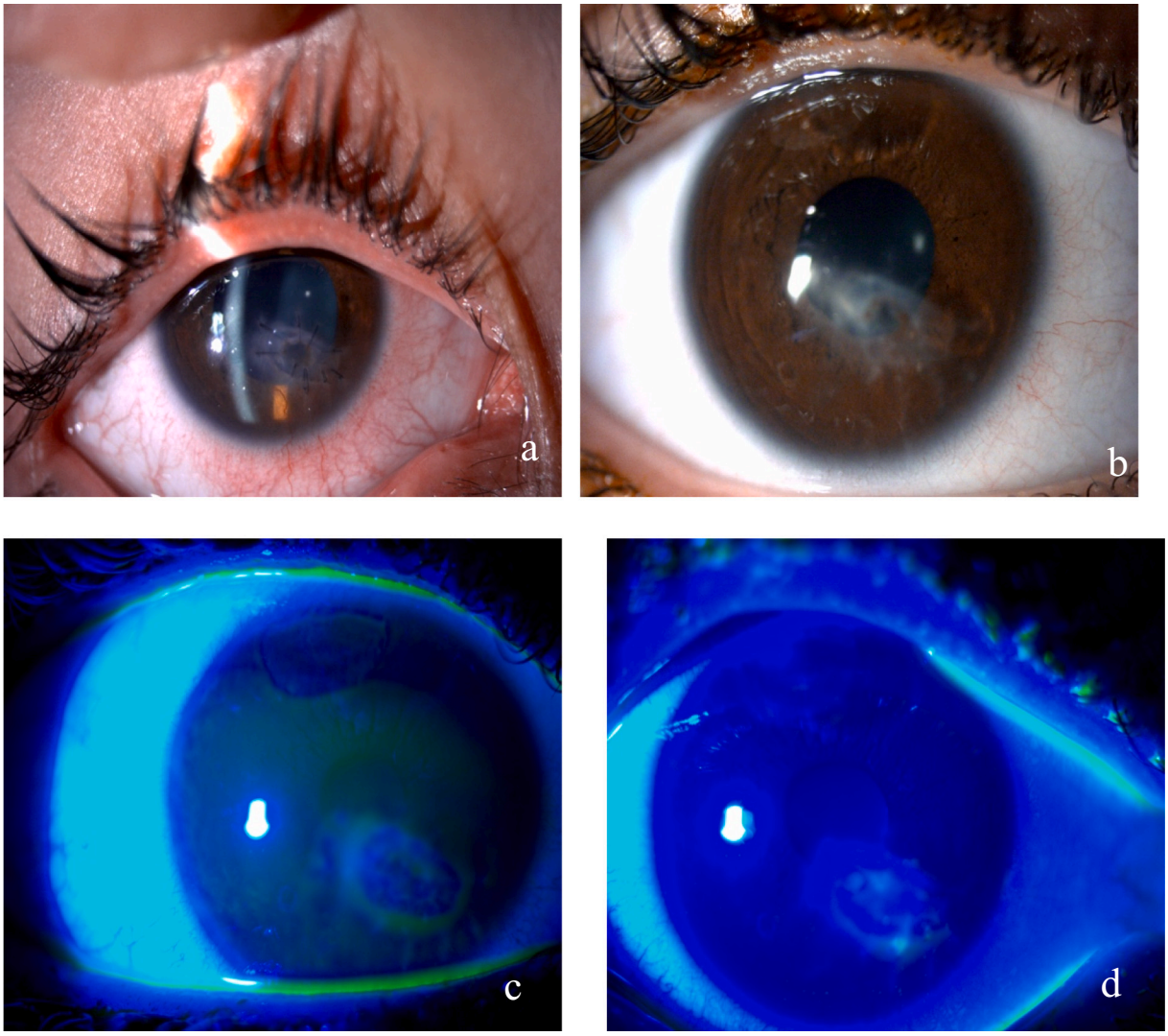

Fig. 4. (a): Slit lamp image showing ipsilateral lamellar autokeratoplasty after 3 months of surgery with the resolution of neovascularization and inflammatory signs.
(b): Slit lamp image showing an ipsilateral lamellar autokeratoplasty after 6 months of surgery and after removal of the sutures.
(c): Cobalt blue slit lamp image showing the location of the autokeratoplasty after removal of the wires.
(d): Cobalt blue slit lamp image showing the autokeratoplasty site without fluorescein intake.

The standard treatment of rosacea is based on palpebral hygiene with the application of heat to the eyelids (heating glasses, wet compresses, etc.). Eyelid care is associated with topical azithromycin-based antibiotic therapy as a first-line treatment. In the most severe forms, oral antibiotic therapy is prescribed: cyclins, suitable for adults, are contraindicated in children under 8 years of age and may in some cases be replaced by erythromycin or metronidazole [24]. Short-term corticosteroid therapy may be prescribed during inflammatory outbreaks. Finally, ciclosporin is used during therapeutic escalation, in case of lack of improvement with previous measures, for cortisone sparing [25].

4. Conclusion

Ocular rosacea is a pathology that is still poorly understood and of delayed diagnosis. Corneal perforation is a serious and challenging complication especially in children.

Ipsilateral lamellar autokeratoplasty is a simple, effective, efficient and non-expensive technique with good anatomical restitution.

Consent

Written informed consent was obtained from the patient for publication of this case report. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Provenance and peer review

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Ethical approval

This type of study does not require any ethical approval by our institution.

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Research registration number

This is a case report that does not require a research registry.

CRediT authorship contribution statement

O. Nabih: drafting and writing the article, acquisition of data
H. Hamdani: revising the article
L. El maaloum: study design
B. Allali: study concept
El kettani: final approval.

Declaration of competing interest

The authors declare that they have no competing interests.

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