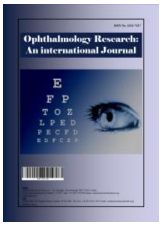

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Amniotic Membrane Graft in Ocular Cicatricial Pemphigus: A More Conservative Approach

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Author's contribution

The sole author designed, analysed, interpreted and prepared the manuscript.

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Case Study

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ABSTRACT

Purpose: To report more conservative management of ocular cicatricial pemphigus (OCP) lesions by performing an initial corneal partial amniotic membrane graft without disturbance of the inflamed conjunctiva.
Methods: A case report of a patient having a stage 3 OCP in his left eye with a corneal ulcer. He was managed by partial corneal amniotic membrane (AM) graft.
Results: The course was favorable with complete corneal ulcer healing and incidentally there was an important regression of associated ocular surface inflammatory signs with a less prominent inferior symblepharon strand.
Conclusion: We propose to manage newly diagnosed patients having advanced ocular cicatricial pemphigus disease with partial corneal amniotic membrane graft before performing radical adnexal surgery with prior immunosuppressive treatment.

Keywords: Ocular cicatricial pemphigus; corneal ulcer; symblepharon; amniotic membrane graft.

1. INTRODUCTION

OCP is a subset of mucous membranes pemphigoid characterized by a predominant conjunctival involvement secondary to an autoimmune disease where the basement membrane and epithelium separation results in blistering and pathologic cicatrization [1].

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We report a case of a patient with a persistent corneal defect in the context of OCP treated surgically with an AM graft.

There still a debate about the importance of direct immunofluorescence to establish the diagnosis,[1] but the clinical examination is of paramount importance.

Preoperative immunosuppressive agents to prevent exacerbation of inflammation [2] is the classic approach; but in this article, we report a new approach to improve ocular surface without prior immunosuppressive treatment.

2. CASE REPORT

We present the case of a male patient aged 71 years-old, who reports as being diagnosed for an OCP 10 years ago.

We didn't perform direct immunofluorescence tests thanks to recent evidence-based studies that proved direct immunofluorescence as not essential to diagnose OCP [3].

The patient presented a red painful left eye 2 weeks earlier. Examination pointed out a visual acuity limited to light perception, advanced blepharo ankylosis, dystrophic cornea and inferior fornix vertical shortening (less than 2 mm) in the right eye. While in the left eye, visual acuity was hand movements perception, conjunctival injection, a central corneal ulcer of 6 mm diameter involving the deep stroma and an inferior symblepharon with a reduced inferior fornix depth (5 mm) (Fig. 1).

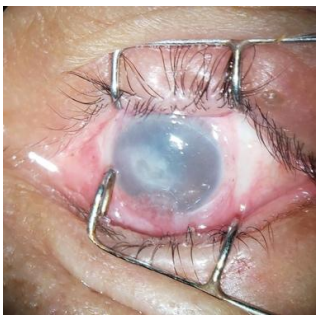

Fig. 1. Photography of the left eye at presentation: An extensive deep corneal ulcer, inferior symblepharon, and important conjunctival hyperemia

Initial management consisted of performing smears for bacterial and fungi examination which proved negative. Immunosuppressive treatment was proposed but declined by the patient. Using a cryopreserved AM with glycerol additive; we performed AM graft in three layers; the first layer being tucked into an incision created alongside the ulcer border, then the two other layers were fixed to the cornea exclusively avoiding any surgical trauma to the adjacent conjunctiva. The conjunctiva was spared to avoid triggering further ocular surface inflammation in the setting of OCP (Fig. 2). The patient received hyaluronic acid based artificial tear eye drops only during two weeks. The course was favorable with complete cicatrization of the corneal ulcer, but also and to our surprise a manifest regression of the inferior symblepharon after three weeks of follow up (Fig. 3).

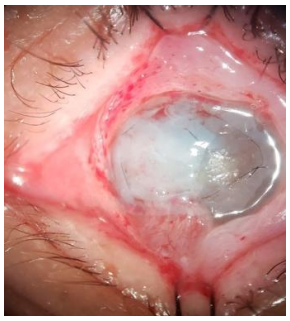

Fig. 2. Photography of the left eye per-operatively: Amniotic membrane sutured in three layers with a running suture; we purposely trimmed the ends of the final knot with long cuts to minimize irritation

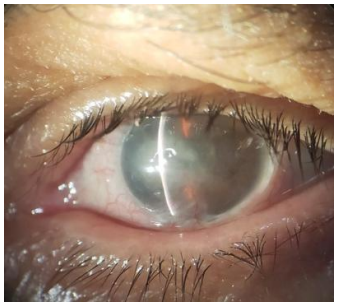

Fig. 3. Photography of the left eye three weeks post-operatively: Complete healing of the corneal ulcer, important regression of inflammatory signs associated with the inferior symblepharon with less conjunctival hyperemia

3. DISCUSSION

The incidence of OCP is estimated to be 1 in 20,000 to 1 in 46,000 of all ophthalmic cases [4].

Typically, more preponderant in female patients with a sex ratio of 3 to 1 [5].

Immunology based diagnosis of OCP is only positive in 40 to 60% of cases [5].

Indeed, the Mucous Membrane Pemphigoid Study Group recommends that patients having clinical signs of OCP with negative biopsy should be considered as direct-immunofluorescence-negative OCP [6].

Different factors can be involved to induce corneal ulcers in the setting of OCP, such as stem cells defects, limbitis, trichiasis, entropion, symblepharon, blepharitis, and conjunctival keratinization [5]. The initial management of a persisting corneal defect after excluding infectious etiologies by appropriate micro-biological assessment could be lash ablation by cryotherapy, laser thermoablation or electrolysis [7], non-preserved ointment as a lubricant; therapeutic contact lens [8] and even autologous serum drops [9].

If all these measures fail then AM graft with a patch over the whole surface is the advocated management [10]. The therapeutic action of AM is drawn from its antiadhesive, antiangiogenic and anti-inflammatory effects. The application of mitomycin topically and subconjunctivelly has shown improvement in postoperative results [11]. Immunosuppressive agents are the treatment of choice to ensure the halt of active disease [10]. Dapsone is the first-line treatment (50–200 mg/day) for mild-to-moderate inflammation, while cyclophosphamide (100-150 mg/day) is reserved for advanced cases. In cases of rapidly progressing disease, systemic prednisolone is used while waiting for the systemic effect of cyclophosphamide to take place.

In an article published by Yesudian, et al. [12]; the authors used AM to treat corneal ulcers and reconstruct fornices successfully with perioperative cyclophosphamide (500 mg) and daily 7,5 mg oral prednisolone.

Another study by Tseng et al showed good results in two cases of OCP by applying mitomycin 0.04% topically and grafting AM to reconstruct the fornices [13]. Others use subconjunctival mitomycin C injection to temporary manage progressive cicatrization of the conjunctiva [14].

Epithelial cells obtained from oral biopsy specimens have been used successfully to be cultured and transplanted onto surgically denuded corneal stroma in 3 cases of OCP by a Japanese team to reconstruct ocular surface and improve corneal transparency [15].

4. CONCLUSION

We propose to use AM graft in the setting of OCP with a two-staged approach; in the first stage before any surgical correction of conjunctival and palpebral anomalies by grafting an AM limited to the cornea to promote a healthier ocular surface with far less inflammation. In a later stage, after immune-suppressive treatment, more extensive surgery to reconstruct the entire ocular surface with an AM may be performed.

CONSENT

As per international standard, patient's consent has been collected and preserved by the authors.

ETHICAL APPROVAL

As per international standard guideline written ethical approval has been collected and preserved by the author(s).

COMPETING INTERESTS

Author has declared that no competing interests exist.

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