Oral mucosa for reconstructive surgery in a case of severe inflammatory necrotizing sclero-uveitis

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The purpose of this case is to show the efficacy of buccal mucosa as an alternative to treat a case of severe necrotizing sclero-uveitis (NSU) associated with ocular perforation. We show a severe inflammatory NSU case that did not improve with topical treatment and scleral patch. We performed a buccal mucosa graft taken from the lower lip with excellent functional and anatomical result, with no signs of relapse of the NSU after 2 years of follow-up. Buccal mucosa can be a safe, useful, and effective alternative for the reconstruction of the scleral wall.

Key words: Amniotic membrane transplantation, buccal mucosa, necrotizing scleritis, necrotizing sclero-uveitis, oral mucosa

Scleritis is an inflammatory disease that presents with edema and infiltrates that affect the entire thickness of the sclera. It is frequently associated with systemic inflammatory diseases (80%).[1,2] Necrotizing scleritis constitutes the most aggressive form of scleritis. It typically appears in the elderly ages, bilaterally (50%), and can severely affect visual acuity (VA) (>50%).[3] The risks of associated systemic disease and visual loss in necrotizing scleritis is high (80% and 50%, respectively).[4] Symptoms include pain and redness. On examination, we can find distorted or occluded episcleral blood vessels, scleral necrosis, and sometimes anterior uveitis (sclero-uveitis).[5] The prevalence of sclero-uveitis is low, unless associated with a systemic disease.[5] Among the most common causes of sclero-uveitis, we can include herpes virus infection, rheumatoid arthritis, tuberculosis, sarcoidosis, or other rheumatologic diseases.[5] Treatment of necrotizing sclero-uveitis (NSU) consists of systemic corticosteroids, combined with immunosuppressants and/or immunomodulators.[6,7] Surgery is reserved only to treat complications, mainly ocular perforation. In these cases, a conjunctival flap or other autologous tissue grafts may be an option.[8]

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Case Report

An 86-year-old male presented to our hospital for a second opinion with a history of NSU in the right eye (RE) refractory to treatment with ofloxacin drops three times a day, brinzolamide twice a day, and latanoprost every night. His past ocular history included evisceration of the left eye 1 year before, secondary to a corneal perforation. Uncorrected VA was 0.3 decimal (20/60) and best-corrected VA was 0.6 (20/32). On examination, RE presented conjunctival hyperemia, very engorged scleral vessels, and an area of de-epithelialization with scleral thinning with perforation [Fig. 1]. Anterior chamber was deep, with moderate Tyndall flare, abundant cells, and endothelial keratic precipitates. Intraocular pressure (IOP) was 8 mmHg. Fundus examination confirmed that the vitreous cavity was uninflamed. Thus, a diagnosis of NSU was suspected.

Although clinically the lesion did not look infectious, corneal scrapes were taken to rule out infective etiology, which came back negative. In addition, blood samples for immunological (p-ANCA, c-ANCA, ANAs, rheumatoid factor, ECA, B27) and infectious (hepatitis, HIV, syphilis) markers, Mantoux/quantiferon-TB Gold test, and radiodiagnosis tests were performed to rule out systemic diseases. General examination did not show any abnormalities or signs of systemic disease, such as rheumatoid arthritis. Given the seriousness

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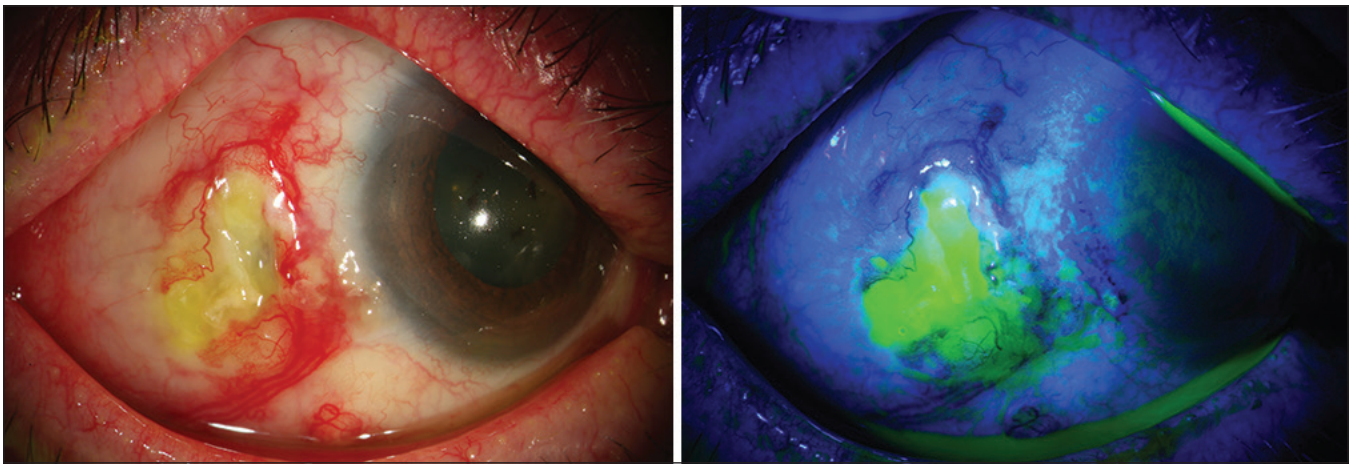

Figure 1: Image of the scleral ulcer in the right eye showing moderate conjunctival hyperemia, engorged superficial vessels, and marked scleral thinning with calcification

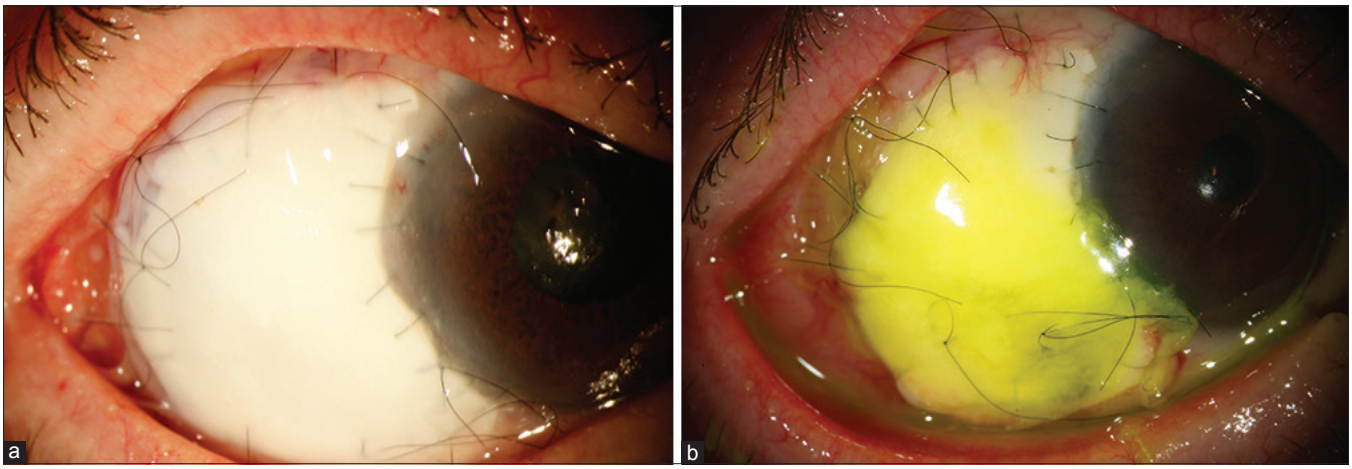

Figure 2: (a) Immediate postoperative image of the semilunar scleral patch (8 × 8 mm), with amniotic membrane covering the entire surface of the graft. (b) Image of the eye 2 months later showing signs of advanced necrosis on the scleral graft

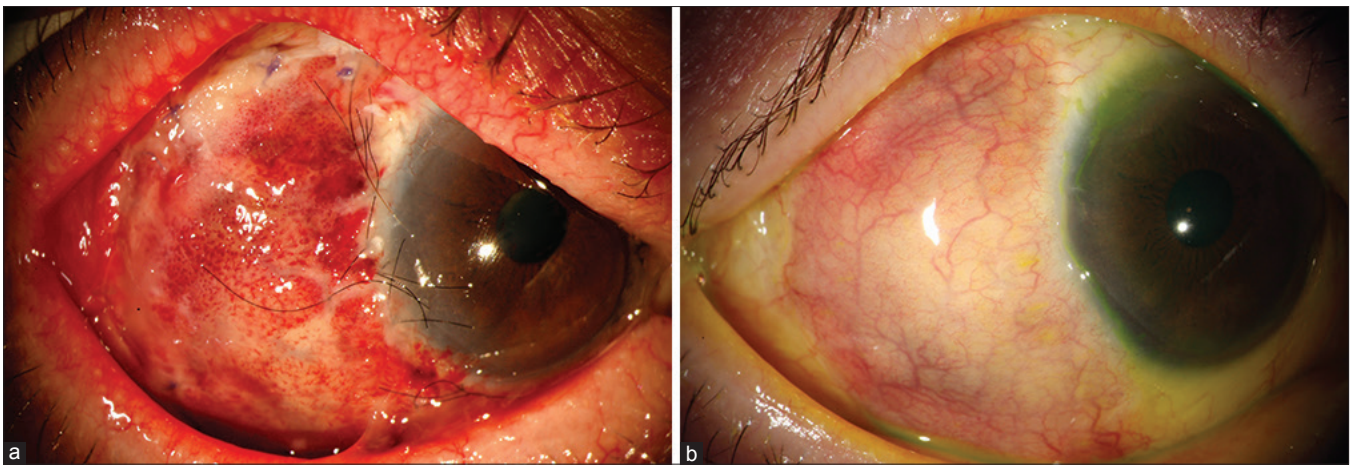

Figure 3: (a) Image of the buccal mucosa graft (18 × 12 mm) the day after surgery. (b) At last follow-up, 2 years later, the buccal mucosa graft looks healthy, with no signs of recurrence of the sclero-uveitis

of the case, and being an "only eye", oral corticosteroids (1 mg/Kg/day) and dexamethasone drops three times a day were given. Ten days later, and based on the minimal response to the treatment, we performed an 8 × 8 mm scleral patch that was fixed with the help of a tissue adhesive (Tissucol Duo®, Baxter AG, Vienna, Austria) and 10/0 nylon sutures (Ethilon® nylon suture, ©Ethicon US, LLC), with an amniotic membrane over, to cover the area of perforation [Fig. 2a]. Blood tests, Mantoux/QuantiFERON-TB Gold test, chest x-ray, thoracic CT scan, and cerebral MRI were unremarkable.

Two months later, the graft presented signs of advanced necrosis [Fig. 2b], and a resection of the necrotic scleral patch was performed, combined with a buccal mucosa graft from the lower lip (18 × 12 mm) [Fig. 3a]. Postoperative treatment consisted of growth-factor-enriched plasma four times a day, gentamycin/retinol/methionine ointment (Epithelizing ointment®; 3 mg gentamycin, 5.5 mg retinol, and 5 mg methionine; Laboratories Thea, Clermont-Ferrand, France) three times a day, prednisolone acetate 10 mg/mL (Pred-forte®; Allergan S.A, Madrid, Spain) five times a day, artificial tears (Thealoz duo®; Laboratories Thea, Clermont-Ferrand, France) every 1–2 h, Combigan® two times a day, and oral prednisolone 60 mg/day, tapered down 10 mg/7 days with a final maintenance dose of 10 mg/day.

At the last follow-up, 2 years later, and after cataract extraction and intraocular lens implantation, VA with correction was 0.1 decimal (20/200). The buccal mucosa graft looked healthy, and the cornea showed swirling epitheliopathy [Fig. 3b]. IOP was 14 mmHg. The patient is currently on Combigan® two times a day, Pred-forte® two times a day, and Thealoz duo® as required, with no signs of recurrence of the sclero-uveitis.

Discussion

The high risk of perforation in NSU can have fatal consequences.[3,9] Thus, it is essential to carry out a rapid diagnosis of suspicion in order to identify the etiology and apply the necessary treatment.[10] Sometimes, despite a correct diagnosis and treatment, complications may occur, given the great aggressiveness of the disease. In our case, there is previous history of evisceration in the contralateral eye due to a similar episode 1 year before. Thus, the management of NSU must be aggressive, focused on controlling the underlying disease.[7,11] In cases, refractory to treatment with imminent risk of perforation, an urgent surgical approach is necessary. Several options have been suggested in the past.[8] Amniotic membrane is usually used as a complement thanks to its anti-inflammatory and epithelium proliferation stimulating effect.[12] It is important to note that these options do not solve the underlying pathology and may not be a definitive solution, as shown in our patient who presented scleral graft necrosis, which required replacement within a few days. In these cases, the oral mucosa of autologous origin can be an effective and lasting alternative, mainly due to its immunological privileges.[13-15] They have been used previously for cases of scleral melt in chemical burns.[16] This situation makes the adaptation to the surrounding tissue more favorable in the buccal mucosa grafts than in scleral ones.

Another positive aspect of the oral mucosa is that it is easily adaptable to practically any surface since it has elastic fibers that provide distensibility, unlike the sclera. In addition, it allows more extensive and easier to manipulate autografts with respect to the conjunctival tissue. In fact, it is the tissue used in the osteo-odonto- and osteo-keratoprosthesis.[17] As a disadvantage, it is worth noting that in cases of evident uveal exposure that require a significant physical resistance, buccal mucosa does not have enough strength with respect to the graft of scleral origin or other synthetic patches.[8]

Conclusion

In conclusion, the application of buccal mucosa can be safe, useful, and effective for the reconstruction of the scleral wall in cases of marked thinning, with minimal uveal exposure, in the context of NSU refractory to medical treatment, preferably after a scleral patch has been attempted.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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