Novel Tape-Splint Tarsorrhaphy Technique to Aid in the Diagnosis of Exposure Keratopathy from Floppy Eyelid Syndrome Masquerading as Recurrent Unilateral Keratoconjunctivitis

Soha Noorani[1] and Dooho Kim[2]

[1]Philadelphia College of Osteopathic Medicine - Georgia Campus
[2]Professional Eye Associates

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Introduction:

Floppy Eyelid Syndrome (FES) describes the disease process whereby extreme laxity of the eyelid can lead to recurrent irritation of the ocular surface and corresponding palpebral keratoconjunctivitis[1,2]. Clinical exam characteristically reveals an eyelid that is easy to evert with traction superiorly[2]. However, FES can masquerade as conditions such as dry eye syndrome, keratitis, conjunctivitis, and dermatochalasis[1-3]. Incidence in the general population has been estimated to range from 2.3% to 15.8%, though notably it may not always be correctly diagnosed and may be more common[1,2].

We describe a case of unilateral and chronic keratoconjunctivitis unresponsive to standard medical therapy. Applying upward traction on the upper eyelid confirmed the diagnosis, as it revealed extensive laxity of the tarsal plate with resultant lagophthalmos and exposure keratopathy. Tape splint tarsorrhaphy (TST) has been recently described as a novel and non-invasive treatment modality for persistent corneal epithelial defects[4]. In this case, the TST was used as a novel and non-invasive diagnostic and temporizing tool to splint the eyelid down to address exposure keratopathy from lagophthalmos. Resolution of signs and symptoms was diagnostic and therapeutic confirmation that the keratoconjunctivitis resulted from the lagophthalmos and FES. This is the first such report to our knowledge discussing the specific use of this technique to more effectively diagnose refractory keratoconjunctivitis due to lagophthalmos and FES.

Case Presentation:

A 63-year-old female initially presented to her primary care doctor for "chronic red eye" and was started on over-the-counter olopatadine (Pataday, Alcon) eye drops for allergies twice a day (BID) in the left eye (OS) and was additionally prescribed topical polymyxin B/trimethoprim antibiotic drops four times a day (QID) OS. The patient did not improve and was referred to an optometrist for further evaluation.

The optometrist determined that the left eye had been erythematous, watery, and accompanied by burning sensations and blurry vision for three months, failing to improve with the above treatment. Past medical history was significant for developmental cognitive delay with a history for childhood sleep apnea and use of continuous positive airway pressure (CPAP). The patient is currently on bilevel positive airway pressure (BiPAP). Pertinent social history is relevant for requiring constant care by her mother due to her multiple chronic medical problems. Past ocular and family history were otherwise non-contributory.

Slit lamp examination revealed 1-2+ inferior bulbar conjunctival injection OS as well as bilateral 2-3+ inferior superficial punctate keratopathy (SPK). The tear volume was also found to be reduced in both eyes. The patient was diagnosed with punctate keratitis bilaterally (OU). She was advised to stop the above

eyedrops and begin aggressive lubrication with artificial tears up to six times a day along with erythromycin ophthalmic ointment at bedtime. Additional recommendations included incorporating supplemental oral fish oil 1000 mg daily and increasing fluid intake. Despite therapeutic compliance, she returned three weeks later without subjective improvement and an ocular examination essentially unchanged. Tear break up time (TBUT) was noted to be three minutes in the right eye (OD) and two minutes OS. The patient was encouraged to continue conservative therapy and return for a recheck for dry eye syndrome. She was then prescribed a three-month supply of cyclosporine 0.05% (Restasis, Allergan) for use BID OU.

At the third optometry visit nearly one month later, the patient was frustrated and expressed only mild subjective improvement in her symptoms despite compliance with treatment. Although the tear film was noted to be normal on anterior segment examination, there was persistent SPK in the left eye. The patient was then referred to ophthalmology (D.B.K.) for a second opinion.

The patient presented to our clinic with persistent 2+ punctate keratopathy and 2+ conjunctival injection in the left eye. There was also peripheral corneal vascularization and subepithelial haze along the inferior one-fourth of the cornea in the left eye. Retraction of the upper eyelids revealed a fairly normal tarsal plate in the right eye (Figure 1A); however, there was profound flaccidity of the left upper eyelid, such that the upper eyelid margin was retractable 15 millimeters (mm) above the superior orbital rim (Figure 1B). There was also 1.5 mm of lagophthalmos in the left eye only (Figure 2). Schirmer 2 testing revealed 21 mm in the right and 22 mm in the left.

Despite the atypical presentation with extreme asymmetry in eyelid flaccidity, in light of the longstanding history for OSA, the diagnosis of floppy eyelid syndrome as a cause of lagophthalmos and exposure keratopathy of the left eye was made. The patient was counseled on these conditions and their association with obstructive sleep apnea (OSA). Temporizing treatment options were discussed, including continued aggressive lubrication, taping of the eyelids, tape splint tarsorrhaphy (TST), and moist chamber goggles. However, it was reiterated that surgical intervention would be necessary for definitive treatment.

The patient refused to have any surgical intervention, so tape splint tarsorrhaphy was chosen as a temporizing measure. The TST was placed over the left eyelid in the office (Figure 3) and the mother was instructed on how to apply it at home. She was counseled to maintain the TST during the day as much as possible as well as at bedtime.

The patient returned one month later with dramatic improvement in both signs and symptoms. The punctate keratopathy and conjunctival injection were resolved in the left eye; however, the inferior corneal vascularization and haze persisted. The patient and her mother were counseled extensively about the limitations of TST and that definitive treatment in the form of surgery to address the eyelid laxity would likely be needed if the problem recurred.

The patient missed the 3-month follow-up but returned in six months and was found to have recurrent keratoconjunctivitis due to poor compliance. Surgical correction of the FES with a lateral tarsal strip was reiterated and encouraged as a more definitive treatment toward resolution.

Discussion:

When managing a case of keratoconjunctivitis, the differential diagnosis can be broad; it may include dry eye syndrome, blepharitis, a multitude of inflammatory processes, and infectious etiologies. It is imperative to perform a detailed examination that includes a thorough slit lamp examination. A unilateral case of keratoconjunctivitis recalcitrant to medical therapy is highly unusual and requires a high index of suspicion to look beyond the typical causes. Upper eyelid eversion is required to identify conjunctival foreign bodies, evaluate the palpebral conjunctiva for any papillary or follicular reactions, and rule out lagophthalmos. Moreover, it is essential to screen for eyelid laxity through retraction of both upper and lower eyelids. Findings of easy eversion, supple tarsal plates, or abnormal retraction suggests FES[2]. Lid retraction exceeding six millimeters may be flagged as abnormal[5].

Floppy Eyelid Syndrome (FES) is an incompletely understood condition which was initially described in 1981

as papillary conjunctivitis and conjunctival keratinization resulting from eyelids that easily and spontaneously evert during sleep[6]. More recent variations in nomenclature have been developed such as "lax eyelid condition (LAC)" emphasizing the eyelid laxity and "lax eyelid syndrome (LES)" when ocular surface disease is involved[1]. FES is typically bilateral and often associated with obesity, middle-age onset, higher predilection in males, and obstructive sleep apnea[1,2]. This case is rather unusual in that there is significant asymmetry as well as extreme left upper eyelid laxity—15 mm of retraction beyond the superior orbital rim.

Chan et al. describe Tape splint tarsorrhaphy (TST) as a novel, non-surgical technique to help splint the eyelid closed in order to treat persistent corneal epithelial defects[4]. The benefits of this method include accessibility, reversibility, and non-invasiveness. The patient is asked to close the eyelids, and while the examiner retracts the patient's eyebrow superiorly, the eyelid is placed on stretch to eliminate the natural lid crease. A two-inch piece of medical grade plastic tape is applied such that the tape covers the entire eyelid. In other words, the bottom edge of the tape is placed on the upper eyelid from the level of the lash-line up toward the brow. The rigidity of the tape effectively creates a splint, preventing the eyelid from opening, which in turn, results in an effective, non-surgical tarsorrhaphy.

We describe a new use of the TST as a diagnostic and therapeutic maneuver to determine the cause for unilateral keratoconjunctivitis recalcitrant to medical therapy. Our patient was found to have extreme left eyelid laxity and lagophthalmos, and application of the TST resulted in dramatic improvement in signs and symptoms of keratoconjunctivitis. Accordingly, clinicians may consider use of TST as a helpful diagnostic and temporizing tool for patients presenting with similar findings of keratoconjunctivitis non-responsive to standard treatment. If TST application does not relieve symptoms, then exposure keratopathy may be a less likely differential diagnosis, and the treatment can easily be reversed without damage to the patient as compared to a traditional tarsorrhaphy. Conversely, a positive response suggests there could be a component of nocturnal lagophthalmos and exposure.

Furthermore, all eye-care specialists can teach patients how to apply the TST independently. Patients or caregivers can reapply the tape at home without having to reschedule an appointment to the clinic. Limitations of the TST include patient dependence to properly apply the tape, potential for allergy to the tape adhesive, and instability of the tape throughout the day or night. It is important to also recognize that the TST is not a long-term solution to the problem; rather, surgical procedures aimed at correcting lid laxity such as lateral tarsal strip, full-thickness wedge incision, canthal tendon plication, medial lid shortening, and lateral tarsorrhaphy are more definitive options[1,2,7].

TST is an excellent diagnostic and therapeutic maneuver to determine if nocturnal corneal exposure is the potential source for a patient's corneal pathology[4].

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Figure Captions:

Figure 1A: Upward retraction of the right upper eyelid shows normal stretch of the eyelid margin to the orbital rim.

Figure 1B: Upward retraction of the left upper eyelid shows significant eyelid laxity with the eyelid margin being stretched 15mm above the orbital rim.

Figure 2: Upon eyelid closure, there is no lagophthalmos in the right eye and ˜1.5 mm lagophthalmos in the left eye.

Figure 3: Application of the tape-splint tarsorrhaphy reveals complete closure of the left eyelid with no evidence for corneal exposure.