

Management of Bilateral Congenital Upper Eyelid Eversion in a Neonate

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Abstract

Congenital upper eyelid eversion is a rare condition characterized by the prolapse of chemosed palpebral conjunctiva from everted eyelids. Initial conservative management option commonly used in the treatment was unsuccessful in resolving the condition in our patient. However, temporary tarsorrhaphy resulted in satisfactory resolution. This case gives further credence to the effectiveness of temporary tarsorrhaphy as a surgical option in the management of congenital upper eyelid eversion.

Keywords: Congenital, eyelid eversion, tarsorrhaphy

INTRODUCTION

Congenital upper eyelid eversion is an uncommon abnormality of uncertain etiology.^[1] It was first reported by Adams in 1896 as a case of double congenital ectropion.^[2] The disorder is characterized by the prolapse of edematous conjunctiva from everted eyelids.^[3] Most cases are bilateral and present at birth; however, unilateral and late-onset cases have been reported in the literature.^[1,4] Although the incidence of congenital upper eyelid eversion is not known due to its rarity, there appears to be high preponderance of the disease in neonates with Down's syndrome, collodion babies, and black infants.^[3] Treatment in most cases is conservative, while surgery is often reserved for unresponsive cases.^[5]

In this report, we present a case of bilateral total upper eyelid eversion that responded satisfactorily to surgical treatment after initial unsuccessful conservative approach to management.

CASE REPORT

A seven-hour-old male neonate was brought to our center by his parents who complained of an abnormal and strange protrusion of fleshy masses from their baby's eyes noticed from birth. Pregnancy had been uneventful, and delivery which was at home was spontaneous by vaginal route. There was no history of premature rupture of membrane, prolonged labour,

maternal genital infection, or birth trauma. The child was the first of the parents and the mother was 19 years old.

Systemic assessment and evaluation by a neonatologist revealed no abnormality. Ocular examination revealed bilateral fully everted, tense upper eyelids, exposed and severely edematous tarsal conjunctiva totally covering the eyeballs [Figure 1]. After application of topical anesthetic agent (proparacaine hydrochloride 0.5%), the everted eyelids were retracted using Desmarres lid retractors to expose the eyeball, which was found to be grossly normal.

Attempts at manual repositioning of the everted eyelids were unsuccessful because of the marked chemosis. The neonate was admitted to the hospital and commenced on empirical antibiotics (ointment chloramphenicol eight hourly and eye drops ciprofloxacin six hourly), 5% hypertonic saline-soaked gauze dressing over the chemosed conjunctiva, and six-hourly application of topical methylcellulose gel. By the fourth day

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of hospital admission, there was no significant reduction in the chemosis and a second attempt at manual repositioning of the everted upper eyelids failed again. A decision for surgical intervention in the form of a temporary tarsorrhaphy was made.

Topical anaesthetic ointment (lidocaine 5%) was applied on the eyelids of both eyes. After 10 min of waiting for it to take effect on the skin, central, medial, and lateral tarsorrhaphy sutures were applied to the right eyelids using 5/0 vicryl to achieve closure of the lids without visible prolapsed conjunctival tissue. A single central tarsorrhaphy suture applied on the left eyelids was adequate [Figure 2].

After 48 hours, all tarsorrhaphy sutures were removed. Both right and left eyelids remained reverted and normal with mild residual chemosis. He was placed on twice-daily ciprofloxacin eye drops (as prophylaxis against infection) and six-hourly methylcellulose gel to keep the ocular surface adequately lubricated. At one and six weeks of follow-up, the neonate's condition had improved remarkably with sustained resolution of the eversion and chemosis. All other ocular examination findings were normal [Figure 3].

DISCUSSION

The exact cause of total eversion of the upper eyelids with conjunctival chemosis and prolapse in otherwise normal neonates remains largely obscure to date. Some factors are thought to play a role in the pathophysiology of this condition. These include hypotonia of orbicularis oculi muscle, birth-related trauma, shortening of the anterior lamella in the vertical plane, or lengthening vertically of the posterior lamella of the eyelid with failure of levator aponeurosis fusion with orbital septum. Other factors include lack of adequately functional lateral canthal ligament and lengthening of the eyelid laterally.^[6] None of these postulated etiologic factors in the pathogenesis of congenital bilateral upper eyelid eversion were found in our patient. This is in keeping with majority of cases reported in the literature. It is noteworthy that histopathological examination by Young of everted eyelids of an infant who died nine days postbirth showed no abnormality.^[7]

It has also been speculated that pressure exerted on the baby while passing through the birth canal during delivery may cause venous stasis within the eyelids, which may also induce marked chemosis and conjunctival prolapse with resultant eversion of the eyelids.^[8] However, method of delivery is unlikely a predisposing factor as cases have been reported following cesarean section.^[1,9]

Following eversion of the eyelids, spasm of the orbicularis oculi muscle could act like a sphincter initiating a cycle of conjunctival strangulation and edema due to venous stasis that further worsens the eyelid eversion and chemosis.^[10] Complications involving the cornea are uncommon due to protection from exposure keratitis by the chemosed conjunctiva.^[6]

Congenital eversion of the upper eyelids can be managed conservatively or surgically. The management goals are to



Figure 1: Bilateral upper eyelid eversion with severe chemosis in a neonate at presentation

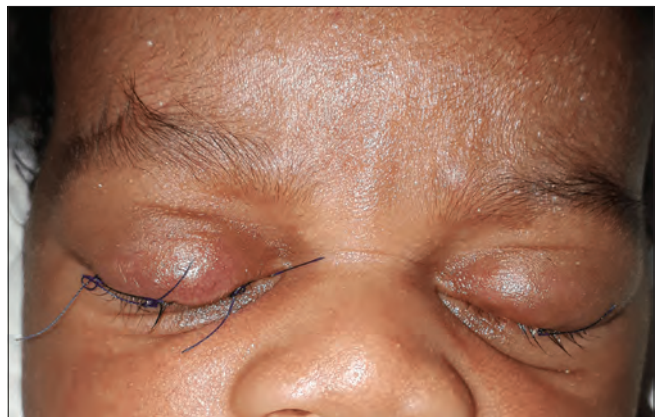


Figure 2: Patient about 15 minutes after bilateral temporary tarsorrhaphy



Figure 3: Picture of the patient at 6 weeks of follow-up with normal eyelids

prevent desiccation and infection of the exposed conjunctiva, reduce conjunctival edema, and achieve reversion of the everted lid to normal.^[8,9]

Conservative treatment entails using ointments and lubricants, topical prophylactic antibiotics, and eye patching with gauze soaked in 5% hypertonic saline.^[3,5,9] The hypertonic saline is believed to induce movement of fluid from edematous

conjunctival tissues through semi-permeable membrane by osmosis, leading to resolution of edema and subsequent reversion of the everted lid to normal.^[9] This treatment modality usually takes days to weeks to resolve chemosis and lid eversion.^[1,8]

Surgical treatment options are often reserved for cases unresponsive to conservative approach or those that present late. These include full-thickness skin graft to upper eyelid, tarsorrhaphy, fornix sutures, and subconjunctival injection of hyaluronic acid.^[6]

Our patient was initially managed conservatively before a switch to surgical approach for a number of reasons. First was the poor response to conservative management. Perhaps, if we had waited longer, the lids could have reverted to normal; however, that would probably entail prolonged hospital stay with its attendant unfavorable economic implications and higher risk of desiccation and infection of the exposed conjunctiva. Second, the reluctance of our patients' parents to continue waiting for few more days or weeks and their desire for prompt resolution of their baby's condition further necessitated the switch to a surgical approach.

CONCLUSION

Congenital upper eyelid eversion is an uncommon condition in a neonate that is often managed conservatively with resolution time of days to weeks. Any safe, affordable, and effective treatment modality that resolves this eyelid disorder in one or two days should be preferred as it reduces the risk of desiccation and infection of the exposed conjunctiva. Such treatment options will also reduce the length of patients' stay in the hospital. Temporary tarsorrhaphy is one such treatment modality; it is affordable, seems safe, and results in quick and satisfactory reversion to normal of the everted eyelids. It is therefore recommended

in cases of congenital total eversion of the upper eyelid with severe chemosis.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the child's father has given consent for images and other clinical information to be reported in the journal. The child's father understands that the names and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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