

Congenital upper lid eversion and severe chemosis in a new born

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Abstract

The purpose of this study is to report a case of bilateral congenital eversion of the upper eyelid in an 8 h- old male baby. JD is an 8 h-old male baby born by normal vaginal delivery after prolonged labor, to a 24-year-old primigravida. Pregnancy was said to be term but was complicated by pregnancy-induced hypertension. A midwife delivered the baby and it was by spontaneous vertex delivery. He was said to have cried immediately after birth, but shortly afterward parents noticed a reddish fleshy swelling over the eyes and this made it difficult for them to see the eyeballs. The reddish swelling steadily increased in size necessitating their prompt referral to our center. On examination, a full term baby was seen with complete eversion of both upper eyelids and marked conjunctival chemosis. Ocular examination revealed normal globes with healthy cornea and briskly reactive pupils. The child was admitted and started on 3-hourly chloramphenicol ointment, ciloxan eye drop (ciprofloxacin hydrochloride United States Pharmacopeia equivalent to ciprofloxacin 0.3% w/v) - 8 times daily and 5% hypertonic saline patch over the chemosed conjunctiva. The pediatrician was also invited to co-manage the patient. After 9-days of treatment, the conjunctival chemosis fully resolved and lids reverted back to normal position. At 4-week follow-up, the lids continued to maintain their normal position and child could now open eyelids spontaneously. Congenital upper eyelid eversion is a rare clinical entity even though it is said to be commoner in blacks. Knowledge of its complete resolution with conservative management will help in future management of such cases thereby preventing complications that may arise from poorly treated cases.

Key words: Congenital, eversion, upper eyelids

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Introduction

Congenital upper eyelid eversion is a rare disorder of unknown etiology. The condition is typically bilateral^[1] but unilateral cases have also been reported^[2], with a higher incidence observed in black newborns, infants with trisomy 21, and children of multiparous mothers.^[3] Congenital lid eversion can be due to infections, inflammation and birth trauma; or may be associated with systemic anomalies such as Down syndrome.^[4] It has also been associated with prolonged and difficult labor^[5] though in some case, no known cause has been found.^[6] Congenital eversion of the upper eyelid was first described by Adams^[7] in 1896, and he called it “double congenital ectropion”. Since the first reported case, many more have appeared in the literature.

In Nigeria, several authors have reported cases of congenital upper eyelid eversion. In South-South Nigeria, Osahon and Ibadin^[8] in Benin City, and Dawodu^[9] in the Delta State each reported cases of bilateral congenital upper eyelid eversion in male neonates. In the same vein, in Western Nigeria, Adeoti *et al.*^[10] in Ibadan reported three cases of bilateral congenital lid eversion in males who presented at between 4 h and 4 days of birth. Similarly, Omolase *et al.*^[11] in Owo reported a case in a male neonate who presented within 3 h of birth. However, no such case/cases have been reported in Rivers state or other neighboring states in the South-South.

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We hereby report a case of bilateral congenital upper lid eversion with severe conjunctival chemosis in an 8- h old male baby.

Case Report

JD is an 8- h old male baby born by normal vaginal delivery to a 24- year- old primigravida after a full term pregnancy complicated by pregnancy-induced hypertension. Mother was offered caesarean section but declined and eventually delivered in a private clinic after labor that lasted over 17-h. Baby was delivered by spontaneous vertex delivery and was said to have cried immediately after birth. Shortly after birth, parents noticed reddish swelling over the eyes, which had been steadily increasing in size thereby necessitating their prompt referral.

Examination revealed a healthy looking baby, not in any obvious distress but with complete eversion of both upper eyelids and marked conjunctival chemosis [Figure 1]. Attempts at repositioning the upper eyelids failed. The lids were then gently parted to examine the eyeball. Both globes appeared to be of normal size with clear corneas and pupils that were briskly reactive to light. The child was admitted into the eye ward and started on 3-hourly ointment chloramphenicol, ciproxin eye drop (ciprofloxacin hydrochloride United States Pharmacopoeia equivalent to ciprofloxacin 0.3% w/v) - 8 times daily and 5% hypertonic saline patching over the chemosed conjunctiva. The pediatrician was also invited to co-manage the patient. At 1- week follow- up, the condition improved and the child could open his eyes partially. The eyelids could be manually repositioned but everted back on releasing. At 9 days of admission the chemosis was completely resolved and baby could fully open both eyes [Figure 2] though they tended to evert on forceful opening while trying to examine the patient. The lids however reverted spontaneously. Baby was then discharged from the hospital to be followed-up on an outpatient basis.

At follow-up, 2 weeks after the baby was first seen, the condition was found to have improved greatly and mother reported that baby could now open his eyes spontaneously though on an intermittent basis. Examination revealed that the chemosis had completely resolved and the eyelids were now well- apposed. The mother was then advised to continue the application of topical lubricants. At 4 weeks follow-up, the eyelids continued to maintain their normal position and child could fixate and follow the light, and the conjunctivae and corneas were normal [Figure 3].

Discussion

Congenital lid eversion is a rare clinical event but since the first description by Adams^[7] several reports have appeared in the literature. The condition is typically bilateral, but



Figure 1: Before treatment



Figure 2: 9 days after treatment



Figure 3: One month after treatment

unilateral cases have been described. The underlying reason for eversion remains obscure, several possible mechanisms have been proposed and associations have been recognized. The incidence appears higher in black infants^[3], trisomy

21^[8] and infants born with collodion skin disease.^[12] Orbicularis hypotonia, birth trauma, vertical shortening of the anterior lamella or vertical elongation of the posterior lamella of the eyelid and failure of the orbital septum to fuse with the levator aponeurosis have all been proposed as possible mechanisms responsible for the eversion.^[13]

Chemosis and prolapse of the conjunctiva may occur secondary to venous stasis during delivery, causing eversion of the eyelids.^[14] This may be the case with our patient who was delivered following prolonged labor in a woman with pregnancy-induced hypertension. On the other hand, Ren and Liu^[2] reported the presence of congenital upper eyelid eversion in baby born by caesarean section. This means that other factors, other than prolonged labor may be responsible for the lid eversion in Ren's case. Once everted, orbicularis spasm may act as sphincter leading to a vicious cycle of conjunctival strangulation and edema secondary to venous stasis. The prolapsed conjunctiva usually protects cornea thereby preventing or reducing corneal complications.^[15] Our baby appeared normal phenotypically even though no deoxyribonucleic acid test was carried out to rule out Down's syndrome.

Several management modalities both conservative and surgical have been proposed for management of congenital eyelid eversion. Conservative treatment involves the use of ointments and lubricants, topical prophylactic antibiotics and patching with 5% hypertonic saline.^[10,16] An attempt to explain the mechanism by which the 5% hypertonic saline soaked gauze dressing worked was put forward by Adeoti *et al.*^[10] as osmosis of fluid from edematous tissues through the semi permeable subconjunctival membrane, encouraged by the hyper tonicity of the patch. This may explain the resolution of the edema and subsequent lid reversion as proposed by Voet *et al.*^[17] Surgical treatment involves lateral tarsorrhaphy with excision of redundant conjunctiva, fornix sutures, full thickness skin graft of the upper lid, and subconjunctival injection with hyaluronic acid.^[2,9,11,18]

Our patient was managed conservatively and despite the marked conjunctival chemosis and lid eversion at presentation, this resolved fully at 9- days post-presentation without any surgical intervention.

Conclusion

Congenital upper eyelid eversion is a rare clinical entity even though it is said to be commoner in blacks. A normal

anatomical outcome with good cosmetic results can be effectively achieved using topical lubricants, antibiotics and application of 5% hypertonic saline patch. Careful ocular examination should however be carried out first to rule out other ocular or systemic complications, in the absence of which conservative management can be instituted. Knowledge of its complete resolution with conservative management will help in future management of such cases thereby preventing complications that may arise from poorly treated cases.

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