Congenital Bilateral Eversion of the Eyelids

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ABSTRACT
The authors report a case of congenital bilateral eversion of the eyelids with secondary conjunctival prolapse in a 4-hour-old neonate. Conservative treatment with hypertonic normal saline, lubricants, antibiotics, and padding was instituted. No surgery was performed. Complete eye opening was achieved by the 10th day after birth. Medical management is advantageous in that it is inexpensive, noninvasive, and can be performed even in a center with poor surgical facilities, as is commonly encountered in developing countries. The sociopsychological aspect of the disease, possible pathophysiology, and treatment are discussed. [J Pediatr Ophthalmol Strabismus 2008;45:371-373.]

INTRODUCTION

Congenital eyelid eversion may be described as a condition in which there is a complete out-turning of the eyelids associated with swelling, severe conjunctival prolapse, and chemosis. It could be congenital, acquired from infections, inflammation, or trauma through the birth canal, or associated with systemic anomalies such as Down’s syndrome. It could also occur following oculoplastic surgeries such as repair of congenital ptosis.

This means that eyelid eversion could be present at birth. Some authors have proposed that it is presumably brought about mechanically as the head traverses the birth canal. Prolonged and difficult labor have been incriminated in some cases, whereas no known cause has been found in others. The condition is usually bilateral, but unilateral cases have been documented. The incidence is unknown because it is a rare condition that could occur in an otherwise normal infant.

Treatment depends on whether the condition is congenital or acquired. For acquired cases, treatment is aimed at removing or preventing the underlying causes. Congenital cases could be treated medically with antibiotics and patching or surgically by applying temporary tarsorrhaphy with mattress sutures. Reversion of the eyelid may be difficult in some cases due to massive inflammation and chemosis. The condition usually resolves within 2 to 3 weeks.

We describe a 4-hour-old neonate presenting with this condition, which is the first case we have encountered in 10 to 15 years of practicing ophthalmology in Nigeria. The aim is to highlight the peculiarity of the use of 5% hypertonic saline as an inexpensive, noninvasive method of treatment and the sociopsychological aspect of the disease, particularly in a developing country with relatively limited resources such as Nigeria. We also discuss the possible etiopathogenesis of the condition and treatment.

CASE REPORT

A 4-hour-old male neonate presented in the emergency department of our teaching hospital with fleshy protrusion of the eyelids and the inability to open both eyes since birth. He had been referred from a private hospital immediately after birth following an uneventful full-term pregnancy and labor and spontaneous vaginal delivery. The perinatal period was also uneventful. There was no history or symptoms suggestive of genital infections in the mother. There was no eye discharge in the neonate, nor was any traditional eye medication applied. He was the fourth child of a polygamous family and the first child of the mother.

Examination revealed clean and non-discharging eyes with bilateral exposed chemotic prolapsed conjunctiva covering the eyeballs (Fig. 1). Attempts at retraction of the eyelids revealed tense eyelids,
clear corneas, and normal anterior chamber depths. Further examination was difficult. The parents of the neonate refused admission on financial and social grounds. He was therefore seen on a daily basis at the outpatient clinic for management.

Treatment consisted of daily eye toileting with 5% hypertonic saline, padding, and review. On the first day, chloramphenicol ointment was applied to serve as an antibiotic lubricant and the eye was padded. The neonate was then referred to a pediatrician, who confirmed his overall health as normal after a comprehensive examination. On the second day, a mild serous discharge was noticed and gauze soaked in 5% hypertonic saline was placed over the chemotic conjunctiva before an eye pad was applied. No repositioning of the eyelid was done.

On the third day, a 50% reduction of the prolapsed conjunctiva and complete mechanical ptosis were noticed. On the fourth day, there was a near total reduction of the prolapsed conjunctiva, improving ptosis with 1- to 2-mm opening of the palpebral aperture, and clear visible cornea in the right eye. The patient could blink to light on the fifth day. On day 8, the right eye had mild ptosis, mild chemosis, clear cornea, and complete eye opening, whereas the left eye had a 3- to 4-mm palpebral aperture opening and mild chemosis. On the 10th day, visual acuity testing revealed that the neonate could follow light and complete eye opening was achieved in both eyes, with some residual ptosis on the left eye. Dilated funduscopy with 0.5% guttae mydriacyl showed normal fundi (Fig. 2).

**DISCUSSION**

This rare ocular condition presented as a congenital case in a male neonate. No known cause could be attributed to it, as similarly observed by previous authors. The mother’s labor was neither prolonged nor difficult and there was no evidence of maternal genital infections. There was also no eye discharge. However, it could have been due to an unknown cause of intrauterine inflammation. This can only be ascertained if the eyelid tissue is examined histologically.

Young found no intrinsic defect after a complete histologic examination of total eyelid eversion in an infant who died on the ninth day after birth. Others have found associations with inclusion conjunctivitis, anterior lamellae inflammation, and Down’s syndrome, whereas Bentsi-Enchill suggested an appreciable overlapping of the lower eyelid margin by the upper eyelid as a predisposing factor. Some have proposed laxity as a cause, whereas others have thought that the occurrence of eyelid laxity is an effect of the eyelid eversion, which usually disappears after the edema and chemosis subsides.

It is suggested that a careful history should be taken to include information on pregnancy and labor, noting any signs or symptoms of vaginal infections in the mother. A pediatrician’s assessment of the child is important to ascertain the normal health of the patient and to rule out any associated congenital anomaly, which might assist in identifying any other associations.

Our case was found to be normal. The mother was a primid and the pregnancy was full term. The roles of grandmultiparity and post-maturity have been questioned in Nigeria and found not to be clear in the occurrence of this condition. However, we believe that the parity of the mother and the maturity of the neonate were not a predisposing factor but may have been coincidental. The mode of delivery was also probably not a predisposition because similar cases have been documented following cesarean section. In essence, the parity, duration of labor, and mode of delivery have not been established to have strong
association. However, interrelated or multifactorial causes need to be considered as possible predisposing factors in future studies.

The daily eye toileting was to keep the external eye clean of any serous discharge, which was thought to come from the edematous tissues of the eyelid. The chloramphenicol eye ointment served as an antibiotic lubricant applied on the chemotic conjunctiva and in the space between the lower and upper eyelid to prevent the cornea from dryness or abrasion from the gauze dressing or other causes. The use of guttane ciprofloxacin prevented secondary bacterial infection.

The mechanism by which the 5% hypertonic saline-soaked gauze dressing applied to the surface of the chemotic conjunctiva worked could be explained as follows. Water is polar due to the presence of hydrogen bonds, which gives it the universal solvent property, thereby separating the ions within the salt. The concentration of water/fluid within the saline-soaked gauze therefore becomes lower than that in the eyelid and conjunctival tissues. Fluid then moved from a region of higher water concentration in the tissues to a region of lower water concentration of the saline-soaked gauze through the semi-permeable conjunctival membrane by the process of osmosis. These processes were thought to have led to the resolution of the eyelid eversion with conjunctival prolapse with subsequent eyelid reversion.

Two authors who reported similar cases in Nigeria performed successful early surgical management using temporary tarsorrhaphy. Another author injected hyaluronidase into the conjunctiva followed by placement of eyelid sutures to revert the eyelid, which led to resolution of the condition within 1 to 2 days. However, Stern et al. reported success with the use of a conservative method to achieve complete resolution, as we have similarly done.

The behavior of the parents of our patient at presentation was striking. The neonate's face was completely wrapped up so that it could only be seen by the physician. They refused admission to avoid stigmatization. Although they claimed they had financial constraints, they continued to travel from the private hospital where he was delivered and pay the bills rather than go home. The grandmother shielded her daughter (the mother) from "disgrace, shame, being labeled a witch with a monster baby, and other embarrassing comments" from the other wives. The neonate was not taken home for the naming ceremony (christening) until the eighth day, after there had been significant resolution of the chemosis and good eye opening. The cosmetic appearance was so unacceptable to the mother and grandmother that the neonate was prevented from being seen by other family members and friends.

This description highlights the sociopsychological aspect of this condition. It is therefore important for physicians to be able to diagnose and treat this condition early. It is also important that primary and secondary healthcare workers be made aware of this condition so that prompt referral for expert management can be offered. Finally, reassuring the parents should be considered the most important part of management.

Conservative management of congenital eyelid eversion, a rare clinical condition, could be effective with the use of 5% hypertonic normal saline to achieve a normal anatomical outlook with good cosmetic result comparable to invasive methods. This inexpensive and noninvasive method is recommended because it would be particularly advantageous to surgically under-equipped hospitals, which are usually commonly encountered in developing countries. This will go a long way to prevent the social stigma and psychological trauma the parents might face.

REFERENCES