Unilateral Congenital Upper Eyelid Eversion in a Neonate: A case report
Éversion congénitale unipatérale de la paupière supérieur chez un nouveau-né : à propos d'un cas

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ABSTRACT

Congenital upper eyelid eversion (CUEE) is a rare ocular condition, typically presenting bilaterally, with few documented cases of unilateral involvement. We report a case of unilateral CUEE in a 3-hour-old male neonate, also presenting with bilateral clubfoot. Conservative management, including saline irrigation and antibiotic eye drops, led to full resolution of the eyelid eversion within five days. This case highlights the successful management of unilateral CUEE and its rare association with other congenital anomalies, suggesting a potential genetic origin.

Key words : Eyelid eversion, Congenital, Neonate, clubfoot

RÉSUMÉ

L'éversion congénitale de la paupière supérieure (ECPS) est une affection oculaire rare, se présentant généralement de manière bilatérale, avec peu de cas documentés d'atteinte unilatérale. Nous rapportons un cas d'ECPS unilatérale chez un nouveau-né de 3 heures, présentant également des pieds bots. Le traitement conservateur, incluant des irrigations salines et des collyres antibiotiques, a permis une disparition complète de l'éversion en cinq jours. Ce cas met en lumière la gestion réussie de l'ECPS unilatérale et son association rare avec d'autres anomalies congénitales, suggérant une origine génétique potentielle.

Mots clés : Éversion palpébrale, Anomalie congénitale, Nouveau-né, Pied bot

Introduction

Congenital upper eyelid eversion (CUEE) is a rare but benign ocular condition characterized by the outward turning of the upper eyelid, often resulting in conjunctival exposure and potential visual impairment [1]. First described in 1896 as a case of double congenital ectropion [2], CUEE has been associated with several factors, including birth trauma, reduced orbicularis muscle tone, and incomplete fusion of the orbital septum with the levator aponeurosis. Conservative management approaches, such as topical hypertonic therapy, lubricants, and association of antibiotics and corticosteroids, have been suggested as effective treatments for CUEE [1,3]. Early intervention is crucial to restore both the anatomical and functional position of the eyelid, and to prevent corneal complications [4]. In this report, we present a case of unilateral CUEE, highlighting its clinical presentation and successful management.

Case report

A three-hour-old male neonate of Ivorian origin, born to non-consanguineous parents and with no significant family history, was admitted for polymorphic syndrome. The pregnancy was irregularly monitored and complicated by gestational diabetes mellitus. Viral serologies were negative throughout the pregnancy. The newborn was delivered post-term via cesarean section due to a bicornuate uterus,with no signs of fetal distress at birth. On examination, the neonate was vitally stable. His height, weight and head circumference were within normal ranges. Additional findings included caput succedaneum and bilateral clubfoot. Ophthalmic examination revealed severe chemosis of the left eye, with complete eversion of the upper eyelid and exposure of the tarsal plate, while the right eye appeared normal (Figure 1). The eyelids were carefully separated to inspect the globes, which were of normal size in both eyes.

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Figure 1: Clinical photograph upon presentation showing (A) pronounced chemosis with full eversion of the upper eyelid and (B) caput succedaneum

The infectious workup results were normal, and both orbital and transfontanellar ultrasounds showed no abnormalities. Unfortunately, a genetic study could not be performed due to socio-economic issues.Management of the eyelid eversion was conservative. Treatment included regular eye irrigation with saline solution, antibiotic and antiseptic eye drops, and covering the eyelid with a sterile compress. Chemosis gradually resolved over the course of five days, enabling manual repositioning of the eyelid (Figure 2).

Figure 2: Clinical photograph on the fifth day from the presentation showing resolution of the chemosis

Discussion

In this paper, we report a rare case of unilateral CUEE in a neonate, associated with bilateral clubfoot. While CUEE is an uncommon condition, its co-presentation with other congenital anomalies has been rarely reported [1], making this case notable. The absence of any significant family history and the patient's birth to non-consanguineous parents suggest that the condition is likely sporadic. However, a genetic predisposition cannot be excluded, especially considering the association with other developmental anomalies. The pathogenesis of CUEE is not fully understood, though several anatomical factors have been implicated, including orbicularis oculi muscle hypotonia, lack of lateral canthal ligament support, and abnormalities in the fusion of the orbital septum with the levator aponeurosis [5]. Unilateral cases of CUEE, like the one presented here, are exceedingly rare [6,7]. Most reported cases involve bilateral eyelid eversion [1,2,8]. This raises important questions about whether unilateral CUEE might represent a distinct clinical subtype or simply a variation in the severity of the condition. While bilateral CUEE has often been linked to systemic syndromes or birth trauma [9], the absence of such factors in our case supports a congenital origin. However, the presence of caput succedaneum introduces the possibility of a traumatic contribution. Although caput succedaneum is generally benign and resolves spontaneously, its association with CUEE and bilateral clubffot in this case raises the question of whether mild birth trauma could have exacerbated an underlying congenital predisposition. The association of CUEE with other congenital anomalies like clubfoot, may offer important insights into the etiology of these conditions. Clubfoot, reported in several syndromes such as Down syndrome [9,10], was present in this case without other dysmorphic features suggestive of chromosomal abnormalities. These findings could suggest an underlying genetic disorder, potentially affecting multiple developmental pathways. Unfortunately, due to socio-economic constraints, genetic testing could not be performed in our case, leaving the exact etiology unresolved. However, the absence of significant family history makes a de novo genetic mutation a plausible explanation. Previous reports have documented cases of CUEE associated with systemic anomalies, such as umbilical hernia or other musculoskeletal malformations [1,7]. Additionally, CUEE can manifest a few days after birth, supporting the importance of close monitoring of newborns with congenital anomalies [11]. Such measures are crucial to ensure timely intervention and to avoid complications that could arise from delayed treatment, such as keratopathy and corneal perforation [9]. Conservative management, as employed in this case, remains the standard treatment for CUEE. It aims to to reduce conjunctival chemosis, prevent infection, and allow spontaneous correction of the eyelid position [5,12]. This typically includes moist dressings, pressure patching, topical antibiotics, and lubricants [1,4]. Our case supports the evidence that conservative treatment remains the cornerstone of management for uncomplicated CUEE, with a favorable prognosis. In more severe cases, where conjunctival chemosis persists, corticosteroids may be used to reduce inflammatory, either via subconjunctival injection or systemic route [4,12]. However, surgical intervention is typically reserved for refractory cases or when complications arise. It includes invasive approaches such as temporary tarsorrhaphy or hyaluronic acid injections [4,9,12]. Fortunately, our patient responded well to conservative treament, with resolution of chemosis and successful manual repositioning of the eyelid within five days, avoiding the need for surgery.

Conclusion

This case highlights a unique presentation of unilateral CUEE in association with clubfoot, adding to the limited literature on this rare ocular condition. Imme-

diate diagnosis and treatment are vital to restore the anatomical and functional alignment of the eyelid and to prevent corneal complications. While conservative management remains effective, further research is needed to elucidate the genetic or developmental factors that may contribute to the pathogenesis of CUEE and its associated anomalies. Genetic studies in similar cases could provide valuable insights into the mechanisms of this rare congenital condition.

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