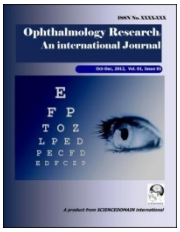

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Non-surgical Management of Bilateral Ectropion in a 5 hours Old Collodion Baby: A Case Report

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Authors' contributions

This work was carried out in collaboration between all authors. Author KFM designed the study, wrote the protocol, and wrote the first draft of the manuscript. Author AAA managed the literature searches, analyses of the study performed the spectroscopy analysis and author TOL managed the literature searches. All authors read and approved the final manuscript.

Case Study

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ABSTRACT

Congenital bilateral upper eyelids eversion is rare clinical condition. However it is known to be associated with collodion baby, Down syndrome and children of black race. This condition responds well to early active conservative management. We reported a case of congenital bilateral upper eyelids eversion in a collodion neonate delivered by 21 year old lady at home through a spontaneous vaginal delivery. This case was successful managed with lid reposition, topical antibiotic and eye patching.

Keywords: Collodion baby; congenital ectropion; lamellar icthyosis; non-surgical treatment.

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1. INTRODUCTION

Collodion baby is a clinical entity in which the baby skin is replaced by a confined substance of uniform texture, which gives the body a shinning appearance. The most significant concern in this baby is the appearance of disseminated or generalised ichthyosiform genodermatosis characterized by dry skin, scaling, generalized erythroderma and hyperkeratosis, reminiscent of fish scales an entity know as ichthyosis [1].

Types of ichthyosis depend on the mode of inheritance, clinical and anatomic/pathological. Ichthyosis is classified into 3 groups: true ichthyosis, ichthyosiform states and epidemolytic hyperkeratosis [1].

Lamellar ichthyosis is a congenital, disfiguring, non life threatening disorder. It is mostly autosomal recessive, occasional autosomal dominant [1-3] and rare phenotype due to homozygous missense mutation in TGM1 [4,5] has been described. Genetic studies of lamellar icthyosis have shown markers on band 14q11 in the region of the TGM1 gene locus. The mutation in the gene for transglutaminase 1 (TGM1) produces accelerated epidermal turnover with proliferative hyperkeratosis [4,5]. At birth the affected child is covered by collodion membrane, a tough, film-like membrane that fissures when stretched. The membrane is shed within two weeks, revealing diffuse erythema and fish like skin scales [6]. Other features may include nail and scalp abnormalities, eye (ectropion, eclabium, conjunctivitis), deformed ears and inflexible digits. Though not life threatening disorder, the aforementioned features make lamellar ichthyosis presents with medical challenges that can compromise life and vision.

The skin functions including protection from infection, heat and fluid loss are compromised, so also eyelid ectropion predisposes the adnexial/eyeball to among others infection and dryness. Therefore, in the management of lamellar ichthyosis one should consider infection, fluid and heat balance. The management of congenital eyelid eversion may be non surgical or surgical. The non-surgical treatment includes application of 5% hypertonic saline, eye lubrication with antibiotic (chloramphenicol eye ointment), injection of hyaluronic acid and eye patching. While surgical treatment includes temporal tarsorrhaphy, subconjunctival injection of hyaluronic acid, fornix sutures and full thickness skin graft of the upper lid [7,8]. We reported a case of successful non surgical management of a five-hour-old collodion boy with congenital bilateral upper lids ectropion.

2. CASE REPORT

A five-hour-old boy delivered by 21-year-old para 2 woman, presented at our eye clinic on account of bilateral inability to close both eyes properly since birth. There was associated generalised shinning scaly yellowish body. The mother was regular with her antenatal clinics, labour was spontaneous at term but the delivery was at home unsupervised.

There was no history of premature rupture of membrane, prolong labour and maternal infection during the pregnancy. There was no history of consanguinity. There was no similar family history of lid eversion and there was no history suggestive of collodion baby syndrome in the other siblings.

The general examination revealed active baby boy, with the whole body cover with transparent, shiny, yellowish membrane and areas of scaly/peeling skin Fig. 1. The boy had vital signs within normal limits and weighed 2.2kg at presentation.

Ocular examination revealed bilateral upper eye lids eversion, bilateral conjunctiva hyperaemia and chemosis Fig. 1. However; the globes were essentially normal. The full blood count was within normal limits and there was no microbial growth on blood culture. Skin biopsy was not done because there was no histopathology service in our centre.

A diagnosis of collodion boy with bilateral congenital ectropion was made. A consult was sent to paediatric team to co-manage the patient with ophthalmic team. Meanwhile, the management of congenital ectropion commenced including application of hypertonic saline solution 4 times daily, chloramphenicol eye ointment 5 times daily and gauze patching Fig. 2. The patient was admitted to special care baby unit and managed in an incubator. The boy had intravenous 10% Dextrose/Water 65ml 12 hourly, Unasyn (Ampicillin/Sulbactan) 162 mg 12 hourly for 2 days, gentamycin 5.4mg 12 hourly for 2 days and intramuscular vitamin k 1mg stat dose (AquaMephyton). Oral antibiotic was commenced two days on admission; syrub cefuroxime suspension 125mg 12 hourly for one week. The child was placed on graded/graduated mother's expressed breast milk and had topical bodily olive oil (Goya extra virgin) two hourly.

At one week on admission he was immunised with BCG and oral polio while on treatment. The boy did well and he was discharged two weeks after, with normal positions of both upper lids and apparent normal skin Fig. 3. Patient last follow up was 2 months ago.

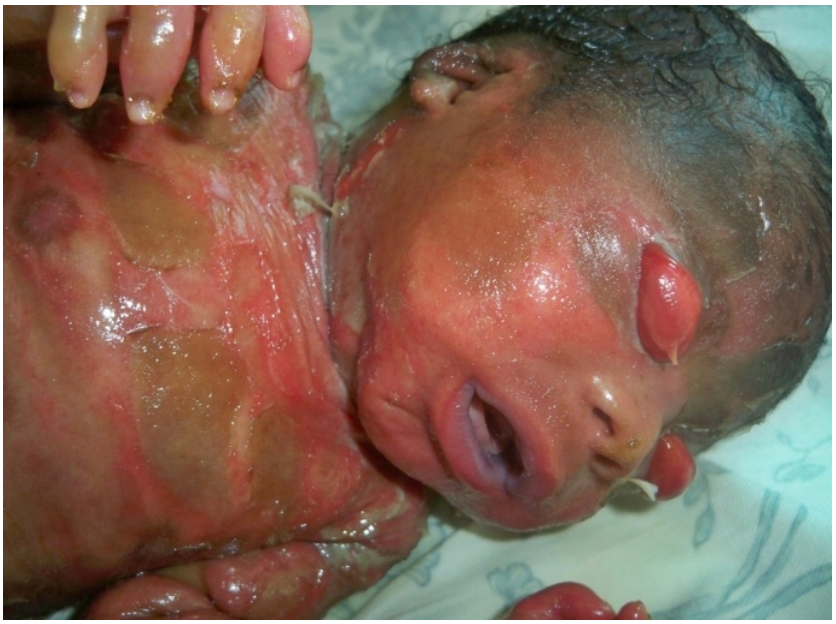

Fig. 1. The collodion boy at presentation

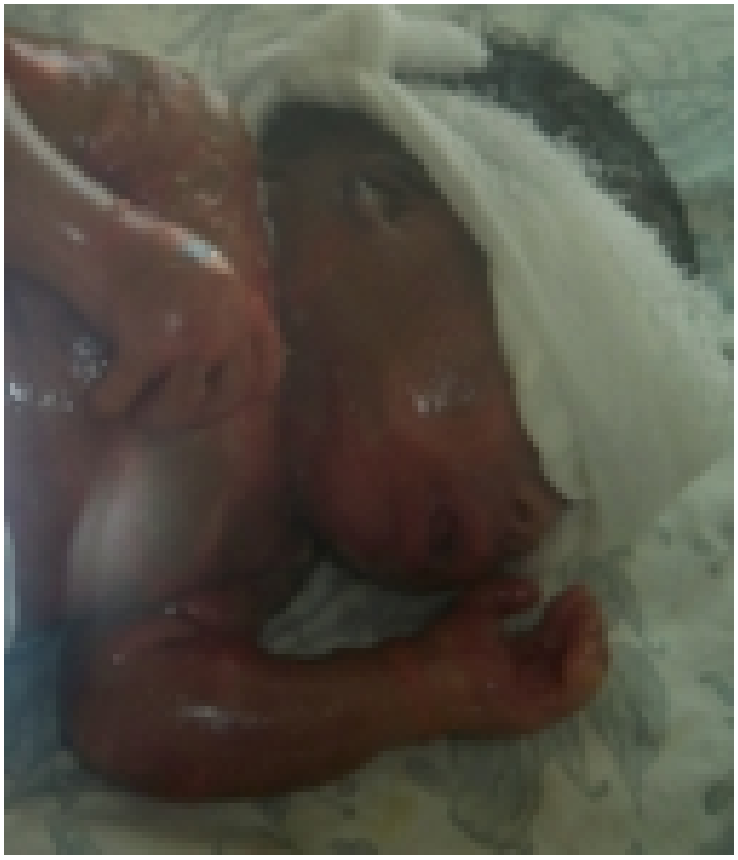

Fig. 2. The collodion boy with eye patch management

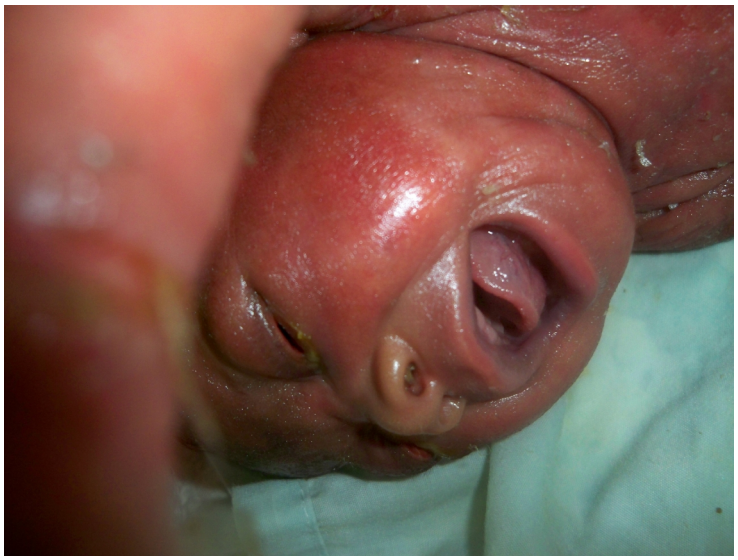

Fig. 3. The collodion boy at day ten into management

3. DISCUSSION

Collodion baby as a term was first used by Hallopeau [8-11] in 1884 since then many cases has been reported in literature. However; this was the first case of bilateral congenital ectropion in a collodion baby managed at our centre. Though, a variety of collodion baby was inherited as autosomal recessive [12] disorder with positive history of consanguinity marriage in 8% but in our reported case the parents are not blood relations. This case of collodion boy might be the occasional non-inherited form. However, it may be an autosomal dominant form of lamellar ichthyosis which has been described [6].

It is possible that the ectropion in the collodion boy was produced by the skin tightening resulting from contracture of the collodion membrane and subsequent oedema of the subcutaneous tissue. The only apparent ocular condition found in this collodion boy was upper eyelid ectropion. However, other ocular manifestation have been reported including exposure keratitis secondary to ectropion, unilateral megalocornea, enlarged corneal nerve, blepharitis, absence of the meibomian gland, trichiasis, madarosis, and absence of lacrimal puncta. The ectropion of both upper and lower eyelids have also been reported [13]. The early presentation and appropriate intervention prevented possible complications such as conjunctival keratinisation, purulent conjunctivitis and, keratitis. The collodion poses challenges, the skin is compromised leading to infection and dehydration. The boy had empirical antibiotics in view of envisaged skin contamination leading to sepsis. The olive oil not only mitigates evaporation but also lubricates the skin preventing dryness. Moreover, the chloramphenicol ointment lubricated the exposed Conjunctival preventing desiccation/keratinisation and served as prophylaxis against eye infection. The topical hypertonic saline is believed to relief lid oedema through osmosis as it has high osmolarity compared to the lid tissue fluid, the successful used of this solution has been documented by previous study [14]. Our case responded well to conservative management however, not all case respond to non surgical management. Blechman et al. [15] reported a case of a one-day-old baby who underwent surgery following an unsuccessful conservative treatment. Furthermore Alvarez et al. [16] reported a case which failed to resolve with two months of conservative management.

4. CONCLUSION

Prompt presentation and appropriate intervention can ensure early resolution of congenital ectropion and prevent complications in collodion disease. Interdisciplinary approach can also ensure good management outcome.

CONSENT

All authors declare that ‘written informed consent was obtained from the patient parents for publication of this case report and accompanying images.

ETHICAL APPROVAL

The ethical approval for this case report was obtained from Ethical Committee of Federal Medical Centre, Birnin Kebbi, Kebbi state, Nigeria.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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