

Case Study

Non-surgical management of bilateral ectropion in a 5 hours old Collodion baby- a case report

Abstract

Aim: To report the effectiveness of non-surgical treatment of bilateral congenital ectropion in a collodion baby.

Case report: A successful non surgical management of five-hour-old collodion boy with congenital bilateral upper lids ectropion delivered by 21-year-old woman is presented.

Conclusion: Prompt and early commencement of non surgical treatment is effective in congenital ectropion.

Key words: Collodion baby; congenital ectropion; lamellar ichthyosis, non surgical treatment

Introduction

Lamellar ichthyosis is a congenital, disfiguring, non life threatening disorder. It is mostly autosomal recessive, occasional autosomal dominant^{1,2} and rare phenotype due to homozygous missense mutation in TGM1^{3,4} has been described. Genetic studies of lamellar ichthyosis 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. 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 scales⁵. 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, subconjunctiva injection of hyaluronic acid, fornix sutures and full thickness skin graft of the upper lid.⁶

We reported a case of successful non surgical management of a five-hour-old collodion boy with congenital bilateral upper lids ectropion.

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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 shining scaly yellowish body. The mother attended antenatal clinics however, the boy was delivered at home, at term through spontaneous vaginal delivery. 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.

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

48 Ocular examination revealed bilateral upper eye lids eversion, bilateral conjunctiva
49 hyperaemia and chemosis [Figure 1]. However; the globes were essentially normal. The full
50 blood count was within normal limits and there was no microbial growth on blood culture.

51 A diagnosis of collodion boy with bilateral congenital ectropion was made. A consult was
52 sent to paediatric team to co-manage the patient with ophthalmic team. Meanwhile, the
53 management of congenital ectropion commenced including application of hypertonic saline
54 solution 4 times daily, chloramphenicol eye ointment 5 times daily and gauze patching
55 [Figure 2]. The patient was admitted to special care baby unit and managed in an incubator.

56 The boy had intravenous 10% Dextrose/Water 65ml 12 hourly, Unasyn
57 (Ampicillin/Sulbactam) 162 mg 12 hourly for 2 days, gentamycin 5.4 mg 12 hourly for 2 days
58 and intramuscular vitamin k 1mg stat dose (AquaMephyton). Oral antibiotic was commenced
59 two days on admission; syrubb cefuroxime suspension 125mg 12 hourly for one week. The
60 child was placed on graded/graduated mother's expressed breast milk and had topical bodily
61 olive oil (Goya extra virgin) two hourly.

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

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69 Discussion

70 Collodion baby as a term was first used by Hallopeau^{7, 8, 9} in 1884 since then many cases
71 has been reported in literature. However; this was the first case of bilateral congenital
72 ectropion in a collodion baby managed at our centre. Though, a variety of collodion baby was
73 inherited as autosomal recessive¹⁰ disorder with positive history of consanguinity marriage in
74 8% but in our reported case the parents are not blood relations. This case of collodion boy
75 might be the occasional non-inherited form. However, it may be an autosomal dominant form
76 of lamellar ichthyosis which has been described.⁵

77 It is possible that the ectropion in the collodion boy was produced by the skin tightening
78 resulting from contracture of the collodion membrane and subsequent oedema of the
79 subcutaneous tissue. The only apparent ocular condition found in this collodion boy was
80 upper eyelid ectropion. However, other ocular manifestation have been reported including
81 exposure keratitis secondary to ectropion, unilateral megalocornea, enlarged corneal nerve,
82 blepharitis, absence of the meibomian gland, trichiasis, madarosis, and absence of lacrimal
83 puncta. The ectropion of both upper and lower eyelids have also been reported.¹¹ The early
84 presentation and appropriate intervention prevented possible complications such
85 asconjunctival keratinisation, purulent conjunctivitis and, keratitis.. The collodion poses
86 challenges, the skin is compromised leading to infection and dehydration. The boy had
87 empirical antibiotics in view of envisaged skin contamination leading to sepsis. The topical
88 olive oil not only mitigates evaporation but also lubricates the skin preventing dryness.
89 Moreover, the chloramphenicol ointment lubricated the exposed Conjunctival preventing
90 desiccation/keratinisation and served as prophylaxis against eye infection. The topical
91 hypertonic saline is believed to relief lid oedema through osmosis as it has high osmolarity

92 compared to the lid tissue fluid, the successful used of this solution has been documented by
93 previous study.¹²

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95 Conclusion

96 Prompt presentation and appropriate intervention can ensure early resolution of congenital
97 ectropion and prevent complications in collodion disease. Interdisciplinary approach can also
98 ensure good management outcome. The case confirms the effectiveness of non-surgical
99 management of congenital ectropion.

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161 Figure 1: The collodion boy at presentation

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181 Figure 2: The collodion boy with eye patch management

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188 Figure 3: The collodion boy at day ten into management

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