Case Report

A Case Report of Bilateral Congenital Ectropion in a Rare Collodion Baby

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Introduction

Collodion baby syndrome also known as lamellar ichthyosis, is a rare genetic disorder with an incidence of 1 in 3 lacs live birth. It is characterized by tight shiny membrane covering the body like a plastic wrap.[1] On shedding of membrane dry rough scaly skin like that of fish is visible. The ocular feature typical of this syndrome is ectropion which is mainly bilateral, however cases with unilateral ectropion have also been reported.[2] The ectropion present in these children makes them prone to exposure keratopathy which could be sight threatening. It is a rare clinical condition in which timely intervention and conservative management can prevent permanent vision loss because of exposure to keratopathy.[3-5] So, we are reporting a rare case of bilateral congenital ectropion.

Case Report

A preterm female born with normal vaginal delivery presented one day after birth, to our OPD with bilateral ectropion (Figure 1). There was no evidence of discharge, corneal opacities or keratitis. No other ocular abnormalities were found. A general examination of the child revealed the presence of a thick parchment-like membrane covering the body with defects at some places. Other findings were eversion of lips, flattening of nose and ears and claw-like hand. On ocular examination, right eye, severe ectropion of both lids was observed, while in the left eye moderate and mild ectropion was noted in the upper and lower lid, respectively, while rest of the anterior segment was within normal limits in both eyes. A cardinal test for ectropion couldn't be elicited due to the thick membrane restricting the movement of lid. She was managed conservatively in a humidified incubator with fluids and antibiotics. She was advised lubricants and antibiotic eye drops with patching at night for ocular involvement. The child was kept under close follow-up. After 1 week, her condition improved and ectropion also started to resolve as the membrane started shedding off (Figure 2). After one month of follow-up, the ectropion was resolved completely (Figure 3). Since she was a preterm baby, ROP screening was also done which showed no clinical evidence of ROP. However, the temporal retina was avascular in both eyes. The child was followed up regularly, and at 1 year old, the membrane formed again (Figure 4). She is presently two years of age and during the last one year, she has had two cycles of membrane formation and peeling. With each cycle of membrane formation, the child developed ectropion, which resolved completely when the membrane had shed off. The parents were advised to take proper care of the eyes with regular use of lubricants and antibiotics to prevent exposure to keratopathy.

Pedigree Analysis

The parents had a history of two children born with similar findings. One was born seven years ago but died at the age of 6 months, while the other was born four years ago and died at the age of 6 days. However, they have two normal children- one is 11-years-old and the other is 3-years-old- both female.

Discussion

Ichthyosis is a skin condition that predominantly affects male babies. It is characterized by excessive drying of the skin and scales over the body. The characteristic features of the disease include a parchment-like membrane covering the skin, which is usually shed away over a period of a month. The basic pathology is dehydration so proper thermoregulation and water and electrolyte balance should be maintained in these neonates. Bhardwaj *et al.*[1] in 2011 reported a case of

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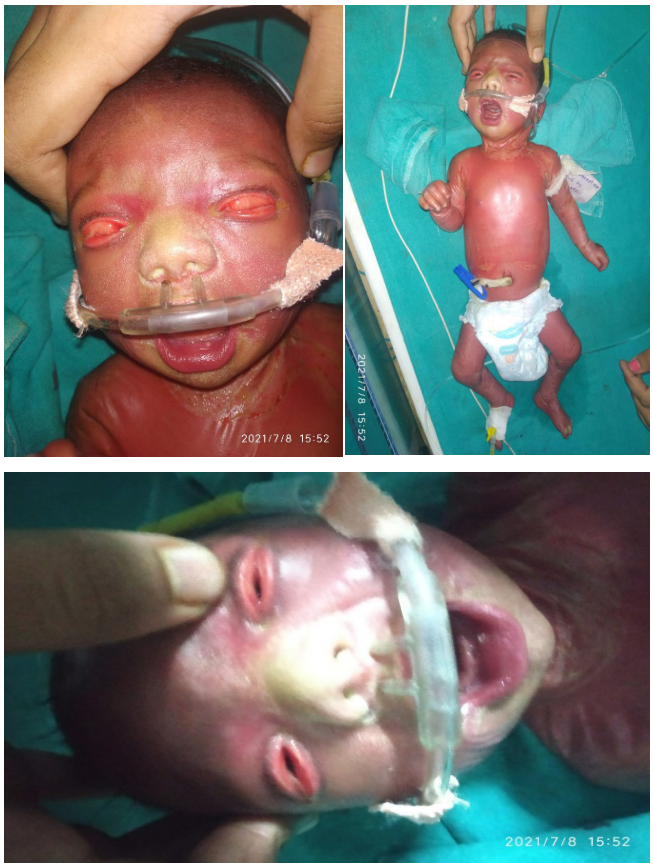

Figure 1: Photographs - Day 1

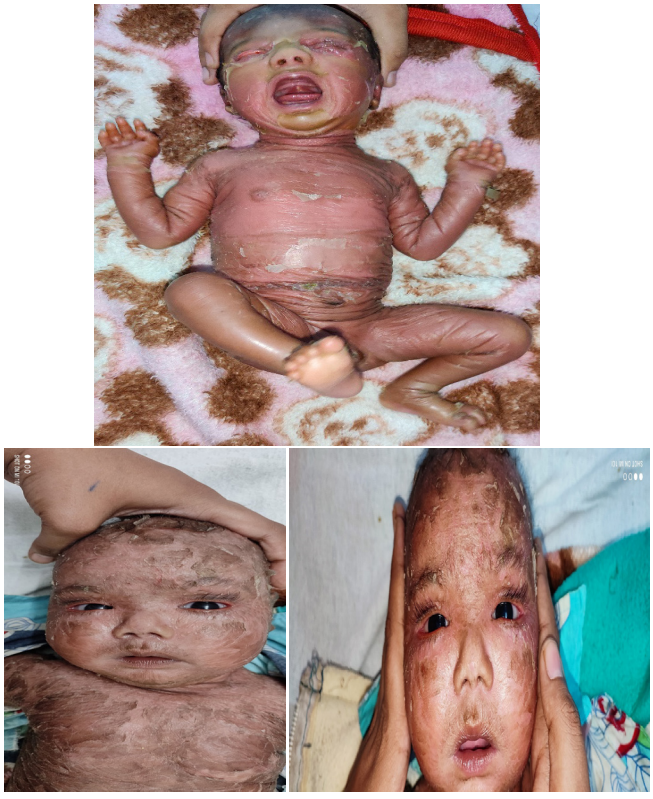

Figure 2: At one week

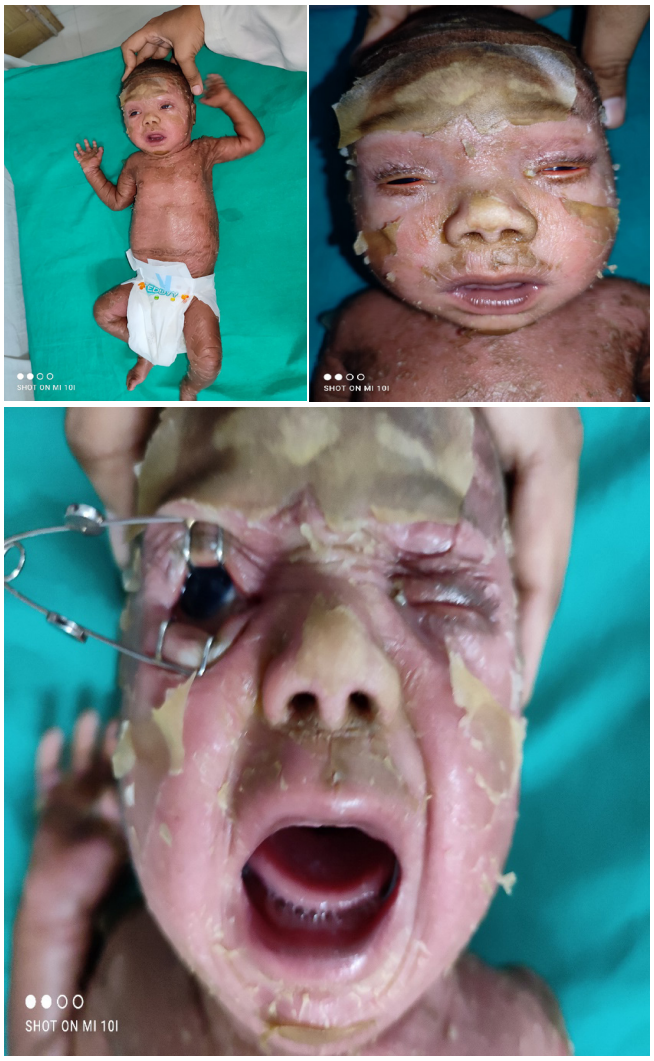

Figure 3: At one month

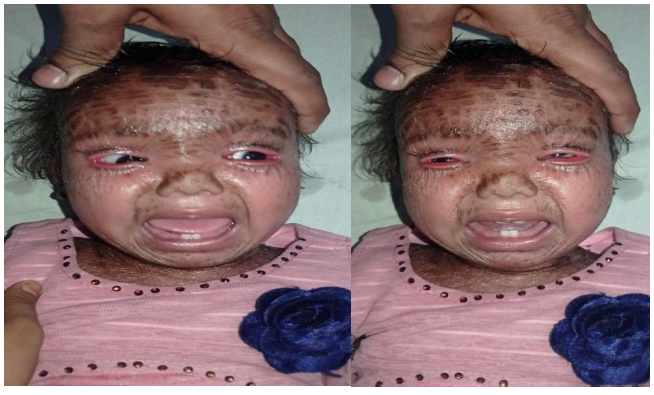

Figure 4: At one year

a collodion baby having similar features as in our case but that child was lost to follow up. With the advent of better NICU facilities and childcare facilities, the overall mortality of these children has reduced. Also, timely ophthalmic management can save vision by preventing exposure to keratopathy.

Conclusion

The ectropion present in the collodion baby, if not managed timely, can lead to severe eye-threatening complications such as keratitis and xerophthalmia, which ultimately lead to corneal blindness. Hence, prompt diagnosis and immediate multidisciplinary management can salvage the vision and life of the baby.

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