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RESEARCH ARTICLE

AN UNUSUAL CASE REPORT OF TWO COLLODION BABIES ON SUCCESSIVE DELIVERIES OF A MOTHER

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ABSTRACT

Background: Collodion Baby refers to clinical presentation in neonates present within a thick taut shiny translucent membrane resembling a collodion, usually this membrane sheds off within 2-3 weeks after birth
Case Characteristics: Both the infants were full term and had abnormal skin with ectropion and eclabium. Apgar was normal with no gross systemic anomalies.
Intervention: The infants were placed in an incubator and temperature monitoring, IV antibiotics, fluid and electrolyte replacement was done.
Outcome: Both the infants died 1 in hospital due to sepsis and second one during transportation so we cannot ascertain the etiology.
Message: Early diagnosis and intervention can reduce morbidity and mortality in neonates.

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INTRODUCTION

The term Collodion baby is a disease entity which may lead to development of one of a variety of congenital ichthyosis or may present as a self-limiting disease condition. In 1880 first clinical description was given by Perez continues to be valid: "The baby's skin is replaced by a cornified substance of uniform texture, which gives the body a varnished appearance" (Cortina *et al*., 1975). The pressure exerted by the collodion membrane distorts the feature of the face and fingers. Even after membrane sheds off it does not leads to normal integument because of varying grades of erythema (Monteagudo *et al*., 2005). The type of ichthyosis depends on mode of inheritance usually lamellar ichthyosis and NBCIE commonest one are Autosomal recessive type (Arenas *et al*., 1996). Harlequin ichthyosis most severe and extremely rare form is inherited as AR disease. ABCA 12 gene (adenosine triphosphate binding cassette A 12), located at chromosome 2q33-q35, is found to be cause of lamellar ichthyosis and mutation being responsible for harlequin ichthyosis (Akiyama, 2006). CB is an extremely rare dermatological condition with an estimated incidence of 1 in 300,000 of childbirth worldwide (O'Connell, 1977; Zapalowicz *et al*., 2006). We report these cases as to highlight the rarity and early intervention can improve the prognosis.

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Case Report

Case 1

In a community based hospital a term female neonate was delivered by a primigravida mother with parents being non-consanguineous. At birth abnormal skin is noted. On examination the weight of neonate is 2.85 Kg, length and head circumference being 51 and 32 cm, respectively. Apgar score was normal 6/9/9 at 1,5 and 10 minutes respectively. There was ectropion and eclabium present which gave an appearance of fish-mouth. Systemic examination reveals no anomalies (Figure-1). Generalized erythema and edema was present on the body and on this basis diagnosed as Collodion baby and was managed in incubator. The monitoring of body temperature, adequate fluid and electrolyte replacement was done. Prophylactic antibiotics and tube feeding was done, but unfortunately the neonate died of sepsis in the hospital and so we were not able to ascertain the etiology of Collodion baby.

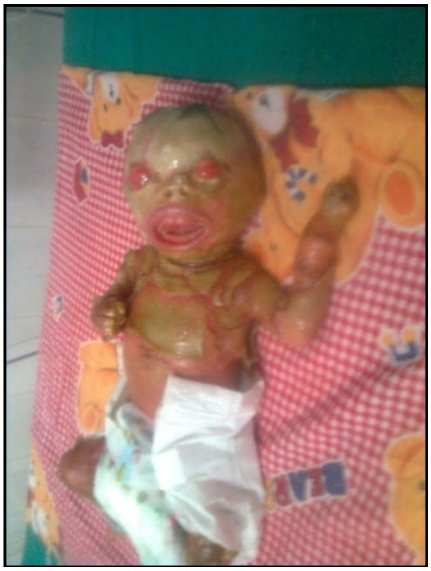

Figure 1. Reported case 1 on day 1

Case 2

After the birth of first collodion baby the mother of this child again conceived in another 6 months of period. The antenatal period was uneventful and at 36 weeks of gestational period underwent LSCS due to neonatal distress and previous caesarian section and delivered a male neonate with similar characteristics as that of first born i.e., abnormal skin, ectropion and a 'O' shaped mouth due to tension produce by collodion membrane (Figure-2).

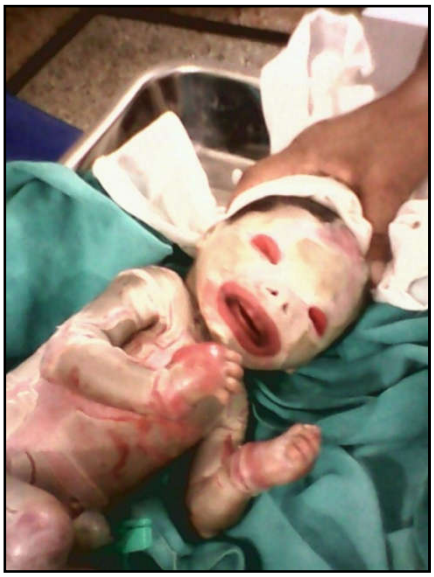

Figure 2. Reported case 2 on day 1

The birth weight was 2.6kgs length and head circumference was 52 and 34 cm respectively. Apgar was 8/9 at birth and no gross systemic anomalies were seen. The neonate was managed in an incubator and closed temperature monitoring was done. Dermatological and ophthalmological reviews were sought and following which he received keratolytics, moisturisers and artificial tears was applied. Following all the preliminary treatment the baby was referred to higher centre for further management but however baby died during transportation.

DISCUSSION

The term collodion baby is given to newborns who present with an extra layer of skin that has a collodion like property. It is a descriptive term for a characteristic clinical presentation at birth, not a specific diagnosis as such it is a syndrome. This clinical presentation was exactly noted in our patients who had extra layers of transparent skin. The previous studies reported various types of ichthyosis: Congenital ichthyosisform erythoderma (43%), Lamellar ichthyosis (19%) dominant ichthyosis vulgaris (12%) and normal skin (25%) (Van Gysel *et al*., 2003). The course of the disease is intriguing, approximately 75% of CB will develop AR congenital ichthyosis either lamellar or NBCIE (Dermatology at the Millennium, 1999) and in 10% cases baby sheds off the skin layers and had normal skin (Van Gysel *et al*., 2002) this is known as self-healing collodion disease. Rest 15% of cases can be due to variety of keratinizing disorders and it includes ichthyosis vulgaris and trichodystrophy. The exact cause of CB syndrome is not well known but it is inherited as an AR trait. The insufficiency of placenta and post maturity has also been documented in some studies. This could be due to defective DNA repair and transcription gene anomalies in prenatal period. A rare recessive disorder Trichodystrophy (TTD) cause by mutation in genes involving NER pathway nucleotide excision repair has also been documented (Moslehi *et al*., 2010). However our patients were all term born and with no placental abnormalities were seen. These babies are at increased risk of developing hypovolemia due to high loss of transcutaneous fluid, dehydration, hyponatremia or hypernatremia and cutaneous infections gram-positive and Candida species. Both the cases were managed in a humified incubator and fluid and electrolyte replacements were given. Tube feeding was done and managed with antibiotics but the first died of sepsis in hospital only and the second baby was referred to higher center but lost during transportation so we cannot follow up the patient and cannot ascertain the cause of collodion baby whether it was lamellar or congenital ichthyosiform erythema.

Conclusion

It is a rare syndrome so we need a protocol for management of these patients, proper instructions to follow in treatment and managing the unforeseen complications. Only supportive treatment is given to these patients. The parents must be reassured that the condition will get better. The long term follow up is required to ascertain the etiology of the disease.

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