J Afric Neonatal 2025;2: 57 - 60

CASE REPORT

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A rare case of genodermatosis treated at Bugando Medical Centre, North-Western Tanzania

https://dx.doi.org/10.4314/jan.v3i2.6

Received: 23rd April 2025
Accepted: 16th June 2025

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Abstract: Collodion Baby (CB) is a rare condition with cutaneous, articular and systemic manifestations which may cause deformity and psychological trauma. Early diagnosis and multidisciplinary approach are important to predict probable course, prognosis and to provide supportive management to patients, parents or caregivers. A full-term female baby was brought to our hospital, 10 hours post-delivery, with complaints of a shiny membranous covering which was noted at birth. There was no history of consanguinity and no family history of skin disorders or congenital abnormalities was reported. She was noted to have bilateral ectropion, eclabium, generalised membranous sheet covering the whole body and some peeling of the skin. She had mild hypernatraemia and was treated with antibiotics, intravenous dextrose normal saline, Vaseline petroleum jelly and eye care. Multidisciplinary management plan, genetic counselling and pre-natal diagnosis should be considered in affected families in order to inform patients about the risk of recurrence in the subsequent pregnancies.

Key words: Collodion Baby, Ectropium, Eclabium and Ichthyosis

Résumé: Le bébé collodion (BC) est une affection rare qui se manifeste par des symptômes cutanés, articulaires et systémiques pouvant entraîner des déformations et des traumatismes psychologiques. Un diagnostic précoce et une prise en charge multidisciplinaire sont essentiels pour évaluer le pronostic, orienter l'évolution clinique et offrir un soutien approprié aux patients, et aux familles. Nous rapportons le cas d'un nouveau-né de sexe féminin, admis 10 heures après un accouchement à terme, présentant une membrane brillante recouvrant l'ensemble du corps. Il n'y avait aucun antécédent de consanguinité et ni de trouble cutané familial ou d'anomalies congénitales. Elle a présenté un ectropion bilatéral, un éclabium, une membrane généralisée recouvrant tout le corps et une desquamation de la peau. Elle a présenté une légère hypernatrémie et a été traitée par antibiotiques, solution de glucose-sérum sale en intraveineuse, la vaseline et des soins oculaires. Une prise en charge multidisciplinaire, un conseil génétique et un diagnostic prénatal doivent être envisagés dans les familles affectées afin d'informer les patientes du risque de récidive lors des grossesses ultérieure.

Mots clés: Bébé collodion, ectropion, éclabium et ichtyose

Introduction

Collodion Baby (CB) is a rare condition worldwide and has a reported incidence of 1 in 300,000 live births[1]. At birth, these babies are covered by a collodion which is a translucent skin covering resembling a parchment paper that peels off within the first 2 to 4 weeks of life[2], to reveal the true nature of the underlying condition which may either be lamellar ichthyosis, recessive x-linked ichthyosis, congenital ichthyosis form erythroderma, harlequin ichthyosis or a normal variant, which is also known as self-healing CB. Conditions associated with CB such as cutaneous, articular and systemic manifestations may cause deformity and psychological trauma. Hence, early diagnosis is helpful to predict probable course, prognosis and to provide supportive management to patients, parents or caregivers. A multidisciplinary approach is therefore important to optimize the likelihood of long-term survival of these babies.

Case Presentation

A full-term female baby was brought to Bugando Medical Centre (BMC), as a referral case from Geita regional referral hospital, 10 hours post-delivery. She was born by spontaneous vertex delivery (SVD) at gestational age of 37 weeks and 5 days. Her birth weight was 2.7kgs and had an Apgar score of 9 and 10 at first and fifth minutes respectively.

She was referred to our tertiary hospital due to a shiny membranous covering which was noted at birth. Her mother attended antenatal clinic regularly at the near by dispensary and had an uneventful pregnancy throughout. There was no history of use of alcohol, cigarrete smoking, drug use or exposure to radiations during pregnancy. This baby was the first-born child to a 23-year old mother and 25-year old father who were both peasants. There was no history of consanguinity and no family history of skin disorders or congenital abnormalities was reported. Both HIV and syphilis screening tests were non-reactive. Pre-natal ultrasound scan was not done. She was given Ferrous Sulphate and malaria chemoprophylaxis as part of routine care during pregnancy.

On examination, she was alert and had stable vital signs. She was noted to have bilateral ectropion and a widely opened mouth like a fish (eclabium). Her whole body was covered by a generalised membranous sheet covering the whole body and peeling of the skin was also noted (Fig 1). Systemic examination was normal. Complete blood count (CBC) revealed a hemoglobin level of 11.2g/dl, white blood cell (WBC) count of 9,500/mm^3 with normal differentials and platelet count of 256 x 10^3mm^3. Serum sodium level was 146.85 mmol/l indicating mild hypernatraemia while normal serum creatinine (99 mmol/l) and potassium (4.9 mmol/l) were recorded. Blood culture and swab culture reported no bacteria growth. She was therefore diagnosed to be a collodion baby (CB) with hypernatraemia. Due to financial constraints, parents of this baby were not able to pay for histological evaluation of the skin biopsy.

She was admitted to our neonatal intensive care unit (NICU) and empirical intravenous Ampicillin (135 mg 12 hourly for 7 days), Cloxacillin(135 mg 12 hourly for 7 days) and Gentamycin (13.5mg once a day for 7 days) were initiated while waiting for culture results. Intravenous Dextrose Normal Saline,20mls 3 hourly, was given for the first 24 hours. This was followed by expressed breast milk from the mother, 27 mls 3 hourly starting on the second day of life, via an orogastric tube. The eyes of this child were examined by an ophthalmologist. He suggested to cover them with a gauze soaked with normal saline in the first three days of life and later on chloramphenicol eye ointment was applied on both eyes twice daily. This baby was also reviewed by the dermatologist on admission and her plan was to manage the skin conservatively by applying pure petroleum vaseline jelly every 2 hours in order to moisturize the skin.

We observed a significant shedding of collodion skin on the 14th day of life (Fig 2). We discharged her from our hospital 6 weeks post admission after a significant clinical improvement was observed and instructed to continue caring for the baby's skin by moisturizing it frequently with pure petroleum vaseline jelly. We also counselled her parents to return to our pediatric outpatient department (POPD) four weeks post discharge. However, they were unable to attend her follow up clinic visit due to financial constraints. At 13th month of life, her skin had significant recovery (Fig 3).

Fig 1: Day 1 of life

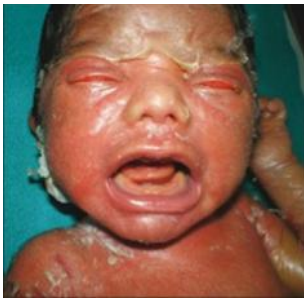

Fig 2: Day 14th of life

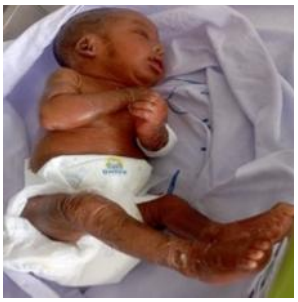

Fig 3: Thirteenth month of life

Discussion

Collodion baby is a rare genodermatosis which was first described in 1884 by Hallopeu and Watelet[3]. Since then, several cases have been reported worldwide and in African children[4,5,6]. These newborn babies are covered with a thick tight membrane all over the body which subsequently detaches in 2 to 4 weeks, usually revealing a permanent ichthyosis phenotype[7]. Different phenotypes can be differentiated based on their clinical manifestations, genetic presentation and histologic findings[8].

CB are often born prematurely encased within a shiny tight membrane. The tight skin around the eyes and mouth often leads to ectropion and eclabium respectively[9, 10] which were present in our case. The tight shiny membrane sheds off, revealing generalised scales which are arranged in a mosaic pattern resembling fish skin, involving the entire body and is increased in the flexural

surfaces causing constrictures/ fissures [9, 11]. In this current case, the baby was full term similarly to cases reported elsewhere[4,6]. CB are prone to excessive dryness of the skin leading to problems with temperature regulation, fluid loss and secondary infections[9]. If they are not kept in a humidified room, fluid loss through bare skin and loss to surroundings may lead to dehydration and subsequent hypernatraemia in these babies. Additionally, it was reported that, transepidermal fluid loss is six to seven times higher in collodion babies as compared to normal skin [12]. Our patient had mild hypernatremia which was corrected by intravenous dextrose normal saline similarly to the case which was managed at Muhimbili National Hospital in Tanzania [4].

Due to resource limitations, our patient was not cared for in a high humidity incubator, as recommended, in order to prevent hypernatremic dehydration and hypothermia [13]. However, we closely monitored our patient in NICU and adhered to infection prevention practices in order to reduce the risk of transmission of infections. Furthermore, we empirically initiated our baby on intravenous Ampicillin, Cloxacillin and Gentamycin while waiting for culture results even though there were no signs of sepsis. Previous reports revealed that CBs may die within few days of life due to sepsis and septic shock [14,15]. Therefore, intravenous antibiotics were prescribed due to the potential risk of infections in this baby because of impaired skin barrier function.

Eye care is another important aspect of care of CB due to the fact that ectropion may further complicate to exposure keratitis, hyperopia and anisometropia[16]. Surgical management may be needed in case of persistence of ectropion beyond 6 months [13]. The non-surgical treatment includes application of 5% hypertonic saline, eye lubrication with antibiotic ointment like chloramphenicol eye ointment, injection of hyaluronic acid and eye patching[6]. Our case was conservatively treated with eye patching with normal saline and chloramphenicol eye ointment similarly to other cases [4,6].

Our multidisciplinary approach to this case, faced some challenges. We were unable to do genetic testing to determine the phenotype since it is not available in Tanzania. Genetic counselling which helps in informing patients about the risk of recurrence in the subsequent pregnancies was also not done. Furthermore, pre-natal ultrasound scan could have assisted in early intrauterine diagnosis and counselling could have been done before birth. Regular clinic follow up was not done in our patient. However, at 13th month of life, this child was seen at our POPD with good outcome of the treatment she received indicating good prognosis and survival of CB beyond the first year of life in our setting.

Conclusion

Collodion baby is a rare congenital disorder and its prognosis depends on the type, presentation and initial neonatal management. Early diagnosis and multidisciplinary approach are crucial in order to prevent long term morbidity and mortality. Genetic counselling and pre-natal diagnosis should be considered in affected families.

Acknowledgement

We acknowledge both parents for agreeing to consent for academic purpose and publication. Special thanks goes to doctors and nurses who comprehensively managed this patient.

Author's contributions

RK: Corresponding author, management of the case, preparation and review of the case report
JK: Management of the case, preparation and review of the case report
NM: Management of the case and review of the case report
NC: Management of the case and review of the case report

Conflict of Interest: None
Funding: None

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