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Case report

Acral self-healing collodion baby: A case series

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ABSTRACT

Collodion baby is a term used to describe a phenotype characterized by the presence of a tight, translucent membrane that covers the entire skin at birth. This membrane usually sheds around 10 to 14 days and reveals the underlying disease (mainly different types of Recessive Ichthyosis or other infrequent disorders). A rare variant of this phenotype is known as acral self-healing collodion baby whereby the patients are born with the typical membrane but limited to the hands and feet only, and after it sheds, the skin appears completely normal.

We report five cases of this very rare subtype of collodion baby. All the patient cases that are presented involved both hands and feet. One of the patients also had the umbilicus embedded in a subtle collodion membrane. None of the patients had a family history of the same entity or any other type of ichthyosis. In all patients, the condition resolved spontaneously within a few weeks and no patients developed any other manifestation. Although no molecular analysis was performed, we contribute to the knowledge of the clinical features of this extremely uncommon and benign entity, since to the best of our knowledge there are only two previous reports available in literature.

Introduction

The term *collodion baby* describes an uncommon and transient disorder of a newborn. Affected children are encased in a glistening membrane, which covers almost their entire body. It represents the initial feature of many diseases, mainly autosomal recessive congenital ichthyosis (ARCI; Frenk and de Techtermann, 1992; Mazereeuw-Hautier et al., 2009). Spontaneous healing distinguishes self-healing collodion baby (SHCB) from other congenital ichthyosis. Only two cases of SHCB that were strictly localized to the extremities have been previously published (Finlay and Bound, 1952; Mazereeuw-Hautier et al., 2009). SHCB has been associated with transglutaminase-1 (TGM1) gene mutations and researchers suggested that the healing of this limited neonatal condition is due to a TGM1 enzymatic activity that is too low in utero, but high enough in later life, to build up a normal epidermis (Mazereeuw-Hautier et al., 2009).

Case series

We present five cases of acral self-healing collodion baby (ASHCB): one boy and four girls (Table 1). The patients were seen at the Dermatology Services department at Hospital Ramos Mejía and Hospital Alemán, Buenos Aires, Argentina during a period of 4.5 years (November 2010–April 2016).

The first case was a girl who was born with a tight glistening film localized only over the dorsum of her hands and feet. The hand involvement extended to the wrists, whereas the feet were covered to a horizontal imaginary line contacting the upper edge of both malleoli (Fig. 1). The patient's palms and soles were spared, as well as the rest of her body (Fig. 2). The girl was born underweight because of a twin pregnancy. Her brother was healthy, and there was no family history of ichthyosis. The membrane began to crack a few days after the birth and its evolution was completed within 4 weeks.

Patient number two was a boy whose dorsum of hands (Fig. 3) and feet and periumbilical area (Fig. 4) were covered at birth by a bright, yellowish membrane with cracks in the flexural areas. The limits of this membrane concurred with the ones presented in the previous case. The glistening film also spared palms and soles. The patient showed a normal appearance after 21 days. There were no other family members affected.

The third case belongs to a newborn female with a healthy elder brother and a no family history of cutaneous diseases. The clinical presentation was identical to case number one. Complete resolution was earlier than the previous cases, with full shedding occurring after the second week after birth.

Last, the fourth and fifth girls presented with a similar phenotype as the girls described before, without any family skin conditions and with complete resolution at 1 month old and 2 weeks, respectively.

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Table 1.
Acral self-healing collodion baby

Patient	Sex	Family history of ichthyosis	Perinatological history	Localization	Time of resolution	Follow up
			Underweight due to twin pregnancy			
1	Female	No	Full term baby	Hands and feet	4 weeks	Normal skin
				Hands and feet		
2	Male	No	Full term baby	Periumbilical area	3 weeks	Normal skin
3	Female	No	Full term baby	Hands and feet	2 weeks	Normal skin
4	Female	No	Full term baby	Hands and feet	4 weeks	Dry skin
5	Female	No	Full term baby	Hands and feet	2 weeks	Normal skin

We have been following these patients since birth. Four patients show a completely normal skin, while the fifth patient shows a very mild phenotype of dry skin.

Discussion

ASHCB is an extremely infrequent subtype of collodion baby that is strictly localized to the extremities (Mazereeuw-Hautier et al., 2009). According to the first ichthyosis consensus, it is a minor variant of ARCI (Oji et al., 2010).

The first description of this entity dates back to 1952, when Finlay and Bound reported a female newborn that presented with a continuous shining, yellowish film, located strictly to the dorsum of her hands and feet. This glistening membrane began to crack at 2 days of life and was completely gone at 21 days of life. The biopsy showed parakeratosis, hypergranulosis and a line of separation between the granular and corneal layer. Despite the complete resolution of the condition, it was considered, at that moment, to be a phenotypic variant of lamellar ichthyosis (Finlay and Bound, 1952).

A second case was reported in 2009, when Mazereeuw-Hautier et al. described a newborn girl as an acral collodion baby whose condition completely resolved at the age of 3 weeks. Her elder sister had been born as a classic collodion baby and evolved to lamellar ichthyosis. A mutation in the TGM1 gene p.Val359Met, which translates into decreased protein expression levels with a residual activity between 8% to 12%, was found in both siblings (Mazereeuw-Hautier et al., 2009). TGM1 mutation has also been reported in generalized

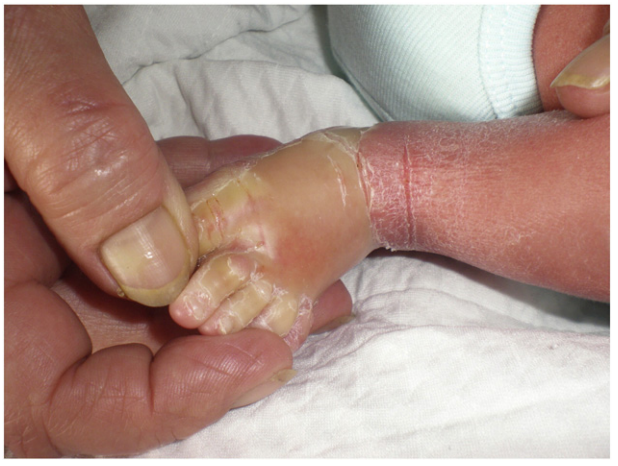

Fig. 1.. Glistening membrane that covers the dorsum of the left foot

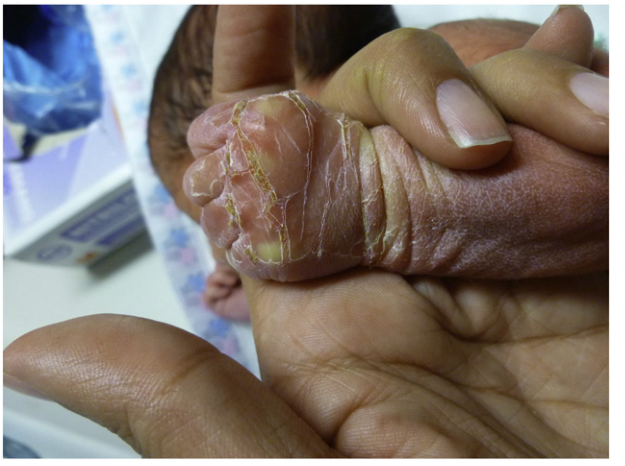

Fig. 3.. Cracked membrane over the left hand

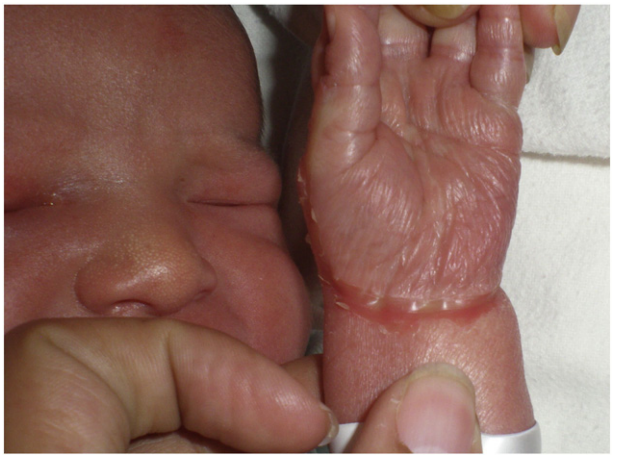

Fig. 2.. Spared palm

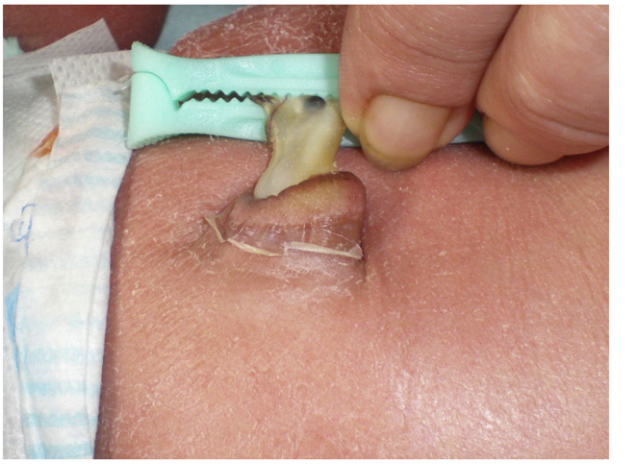

Fig. 4.. Umbilicus embedded in a collodion membrane

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SHCB. Because this mutation affects a cis-trans isomerization property of TGM enzymes, it has been hypothesized that in utero water molecules stabilize the inactive transform with a consequent delayed modification of the stratum corneum, leading to the alteration at birth. Only after birth, when the skin is exposed to air, TGM1 activity is normalized and able to build a more normal cornified layer (Raghunath et al., 2003).

The phenotypic spectrum of alterations because of TGM1 mutations is broad and includes nonbullous congenital ichthyosiform erythroderma, lamellar ichthyosis, SHCB (localized or generalized), and bathing suit ichthyosis (Mazereeuw-Hautier et al., 2009). It has been suggested that regulators of TGM1 could be body site dependent, which would explain the restricted localization to the extremities of ASHCB (Mazereeuw-Hautier et al., 2009; Rodríguez-Pazos et al., 2013). Nevertheless, some cases of SHCB with ALOX12B, ALOXE3 and CYP4F22 gene mutations have also been reported (Noguera-Morel et al., 2016).

Although clinical appearance is very straightforward, some differential diagnoses could be performed, mainly with physiological desquamation of the newborn, generalized SHCB, and congenital syphilis. We emphasize the exact reproduction of the clinical features in all the patients with precise limits: the membrane extends from the hands to the wrists, and on the feet to a horizontal imaginary line contacting with the upper edge of both malleoli. Palms and soles were spared.

Although no molecular analysis was performed, we contribute to the knowledge of the clinical features of this extremely uncommon and benign entity, since there are only two previous reports in the literature. We consider it important that pediatricians, dermatologists, and pediatric dermatologists are aware of this entity so that aggressive procedures and treatments are avoided. Furthermore, knowledge of the self-limiting nature of this condition could allay the anxiety of the parents.

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