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PHOTOQUIZ

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Glistening film on the dorsal hands and feet of a newborn

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CASE PRESENTATION

The patient is a male infant born at 37 weeks and 2 days gestation by spontaneous vaginal delivery at a weight of 2860 grams. The pregnancy and delivery were uncomplicated. During his newborn examination, desquamation of the dorsal surface of his hands and feet was noted (Figures 1-3). The rest of his body was unaffected, including the palms and soles. The parents were advised to apply petroleum jelly to the area twice daily. The parents reported that within a week, the tight membrane on his hands and feet began to crack and that by his third week of life, the dorsal surfaces of his hands and feet had smooth, normal skin. He has since remained without health concerns. No significant dermatological family history was found.

FIGURE 1

FIGURE 2

FIGURE 3

WHAT IS THE DIAGNOSIS?

Diagnosis: Acral self-healing collodion baby

DISCUSSION

Acral self-healing collodion baby (ASHCB) is a rare variant of collodion baby, in which the collodion membrane is confined only to the dorsum of the hands and feet. Collodion baby is the term used to describe the temporary appearance of an infant encased in a glistening, transparent, tight film at birth. Most commonly, the collodion membrane is generalized and involves the majority of the skin. Approximately 90% of the infants born in a generalized collodion membrane develop autosomal recessive congenital ichthyosis, and 10% of the cases are self-healing with no subsequent skin problems.[1] Both clinical forms appear to be a result of an autosomal recessive mutation. The genes that have been implicated include *TGM1*, *ABCA12*, *ALOXE3*, *ALOX12B*, *NIPAL4*, and *CYP4F22*.[2,3]

Acral self-healing collodion baby was first described in 1952 by Finlay and Bound. They reported a continuous shining, yellowish film located only on the dorsum of the hands and feet of a newborn female. They biopsied the skin, which revealed parakeratosis, hypergranulosis, and a line of separation between the granular and corneal layer. The membrane began to crack in the first week of life and completely resolved within 3 weeks.[4]

In 2009, ASHCB was associated with a *TGM1* mutation resulting in a transglutaminase-1 (TGase-1) deficiency.[2] This paper analyzed the TGase-1 activity of two siblings. One of the siblings was born in a generalized collodion membrane and subsequently developed lamellar ichthyosis (LI), while the other sibling was born with a collodion membrane only covering the dorsal surfaces of the hands and feet, which resolved within a few weeks. Histochemical TGase-1 assay in the skin showed absent activity in the case of LI and reduced activity in the case of ASHCB. TGase-1 is involved with forming the cornified cell envelope, a structure that surrounds skin cells and helps form a protective barrier.[5]

A case series in 2016, a case report in 2017, and an abstract in 2018 add seven more analogous cases of ASHCB to the literature.[6-8] All of the cases of ASHCB have a similar progression. The infant is born with a shiny, tight, translucent membrane covering the dorsum of the hands and feet, which begins to crack and peel during the first week of life, and resolves within a few weeks, revealing normal skin.

Acral self-healing collodion baby appears to affect genders equally, representing six female and four male cases.[2,4,6-8] The majority of the cases involved both the hands and feet; however, one case only involved the hands, and one also involved the periumbilical region.[6,8] The acral collodion membranes resolved between 6 days and 6 weeks, with a mean resolution time of approximately 3 weeks. Eight of the infants subsequently had normal skin, but one developed dry skin, and another developed mild ichthyotic skin.

At presentation, the differential diagnoses to consider are peeling skin syndrome, post maturity desquamation, congenital syphilis, and mild generalized collodion. Additional workup or genetic analysis typically is not necessary for diagnosis due to ASHCB's easily recognizable clinical picture. However, in two cases of ASHCB, their sibling had ichthyosis.[2,8] Genetic counseling should be considered due to the increased risk of having a child with a more severe variant of ichthyosis. Additionally, genetic analysis should be considered to help further our understanding of this disease process and its relation to ichthyosis.

We report this rare case of ASHCB to enhance awareness regarding the clinical course of this variant of collodion baby. This phenotype has consistently proven to be a benign entity that resolves within a few weeks. This information can help relieve the anxiety of new parents.

DATA AVAILABILITY STATEMENT

Data sharing not applicable to this article as no datasets were generated or analysed during the current study.

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