Congenital erosive and vesicular dermatosis healing with reticulated supple scaring. A case report and review of the literature

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ABSTRACT

Congenital erosive and vesicular dermatosis healing with reticulated, supple scarring is a rare cutaneous condition of unknown etiology. It presents patchy or generalized erosion and vesicles recognizable at birth that heal with reticulated scarring. We report the case of a female child with this condition, the first one recognized in Argentina, together with a review of the literature (Dermatol. Argent., 2011, 17(3): 214-220).

Keywords:

congenital erosive and vesicular dermatosis, reticulated supple scarring

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Introduction

Congenital erosive and vesicular dermatosis healing with reticulated supple scarring (CEVDHRSS) is a rare disease, first described by Cohen *et á.l.*[1] in 1985. Its etiology is unknown and, in the literature, we have found thirteen publications with a total of 18 cases (Table 1). This disorder frequently occurs in premature neonates who at birth present erosive lesions which produce reticulated supple scarring sequelae. The present case is the first one described in Argentina.

Clinical case

We report the case of a 4-year-old girl born after a 39-week gestation to non-consanguineous parents. The delivery was normal with 9/10 Apgar score. At birth, the skin resembled a collodion baby. At 9 days of age, she was submitted to our Department. The examination rendered the following result: neonate in good health with erythema, erosions, vesicles, crusts, scaling and fissures affecting 75% of the skin surface. Her face presented small linear erosions, crusts and dry, fissured lips. Her eyebrows and eyelashes were barely visible and her hair scarce. The morphology and implantation of her ears were normal but their surface presented small erosions. The fissures were located over big folds and, on the trunk, over the fetal position folding sites. The neck and diaper area presented vesicles on erythematous, macerated skin. On the thorax, the linear fissures took precedence over the vesicles and surrounded square areas of healthy skin of the pectoral region (Photo 1). There was a strip of parallel linear fissures which marked the boundary between the thorax and abdomen areas. On the flexural areas of upper limbs, the vesicles and erosions adopted a predominantly linear pattern (Photo 2).

Her fingers and toes were flexed and the soles presented scaling. The nails were normal. At the age of 2 months her skin was dry and presented two types of scarring lesions, hypertrophic and atrophic. The hypertrophic lesions followed a reticular pattern on the neck and thorax (Photo 3). The atrophic lesions were linear and depressed and were located on the folds of thighs and upper limbs (Photo 4). Sweating was normal and the ophthalmological examination did not show any alterations.

At the age of 2 years and a half, the patient presented the same scarring lesions, though attenuated, and fine scaling on the pre-sternal region (Photo 5) and diaper areas. A skin biopsy at the neonatal stage evidenced three distinct areas: at one end there was acanthosis with moderated hypergranulosis and hyperkeratosis; the sample from the central region revealed more acanthosis and significant parakeratotic scale; at the opposite end of the sample, foci

Photo 1: 9-day newborn. Linear erosions and crusts on face. Dry, fissured lips. Linear, sinuous fissures on trunk around healthy skin areas. On neck, vesicles sit on erythematous, macerated skin.

Photo 2: 9-day newborn. Upper limb with vesicles and erosions on erythematous skin.

Photo 3: 6-month infant with slightly hypertrophic scarring sequelae with reticular pattern on thorax.

Photo 4: 6-month infant with slightly atrophic, depressed scars with linear pattern on skin on left upper limb.

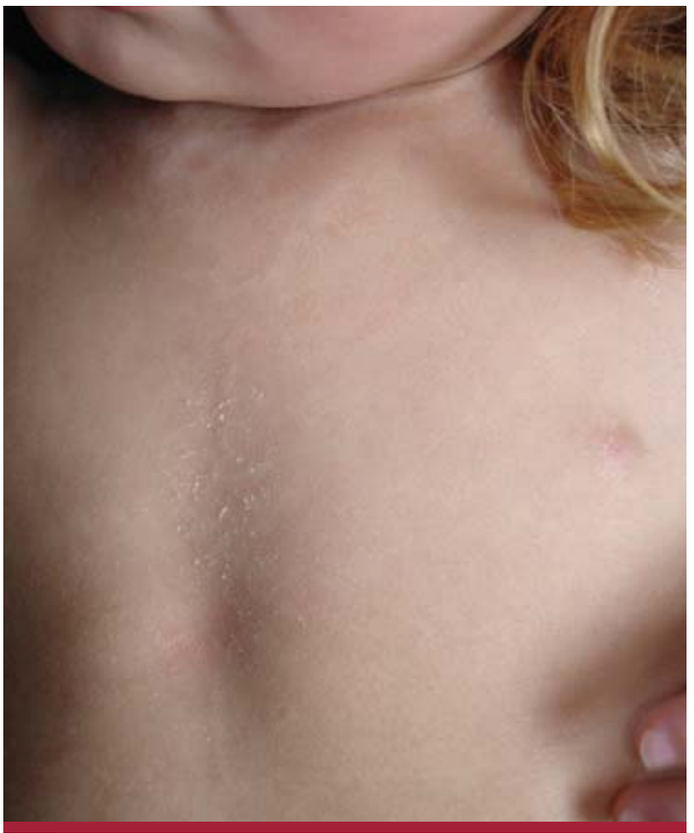

Photo 5: 2-year-old girl with soft reticulated scars and scaling on presternal region.

of necrotizing purulent acute infundibular folliculitis were found (Photo 6).

At 6 months of age, the prominent scarring sequelae on the trunk and depressed scarring sequelae of the thigh were examined through a biopsy. Both histologies rendered similar results: light acanthosis and mild hyperkeratosis with retention of the granular layer.

Discussion

Out of the 18 published cases of congenital erosive and vesicular dermatosis healing with reticulated, supple scarring, 4 corresponded to term neonates (TN) and 14 to pre-term neonates (PTN). None was a case of consanguinity. Gestational age ranged between 24 and 40 weeks. One of the cases was a gemelar pregnancy and, oddly enough, only one of the newborn girls was affected.[1] As regards the sex, 11 were boys and 7 girls. Except for the case in Vun *et ál.*,[2] in which it was reported that the lesions appeared at three days of age, in all others the skin was already affected at the time of birth.

Initial lesions included superficial erosions, crusts and some vesicles.[1] One of the patients was born a collodion baby and was diagnosed with ichthyosis.[3] Our patient resembled a collodion baby in its late stage, when the membrane starts to come loose. In most cases, the involvement was generalized with 75 % of the body surface affected,[1, 3-7] as was the case with our patient.

The lesions were most usually located on the scalp, trunk and limbs. Palms and soles were spared in most cases. In ours, as well as in others in the literature,[1-6, 8-9] the face was affected. The trunk and limbs were affected in seventeen patients but in the remaining case the localization was not reported.[5]

Metz *et ál.*[10] described a patient in whom the vesicles reappeared in the trauma area, whereas Sidhu-Malik et al.[3] reported the case of a 4-year-old girl, whose vesicles reoccurred in fingers and ankles, mainly during the summer. Stein et al.[9] and Fernandez *et ál.*[11] also reported cases of re-occurrences. Our patient did not present recurring vesicles. The lesions healed quickly with reticulated supple scarring.[5,7-8] Our patient also presented linear scars similar to the ones described by other authors. [9,10,12]

Another characteristic of this type of dermatosis was heat intolerance and risk of hyperthermia due to the absence of sweating in the affected areas. The histopathological analysis of the scarring areas in some of the cases [1,5,7] did not reveal any eccrine glands, which could explain the sweating alteration. Compensating hyperhydrosis of healthy skin was frequently observed.[1,3,5,10] The nails were affected in 6 cases [1,3,5,9-10] and the tongue, in 5.[1,4,5,7] Optical microscopy was not conclusive. Decrease or absence of eccrine glands [1,5,6,7,12] and increase of collagen fibers [1,4,10-13]

were observed. Neurological alterations were detected in 4 patients. Three of them were pre-term neonates,[1,3,5] and these disorders could not be determined to be directly associated to CEVDHRSS or to their premature births.

Berk et al. included this condition in the group of pediatric neutrophilic dermatoses,[3,8] provided the acute lesions presented a diffuse interstitial neutrophilic infiltrate in the dermis.[4,6,8] Direct immunofluorescence revealed unspecified deposits of IgG, IgA, IgM, C3 and fibrin in the erosive areas.[3,4,9-10]

The etiology of congenital erosive and vesicular dermatosis healing with reticulated supple scarring is unknown. Sadick et al. suggested that an intrinsic intrauterine event would be responsible for this acute, self-limited process. 8 Cohen et al. reported the case of 28-week-gestation identical twins with monochorial placenta with two umbilical cords.[1] One of the twins presented generalized scarring and erythematous crusted eruption on the scalp, trunk and limbs whose diagnosis was retrospective. The other twin had no lesions. Clinical discordance and absence of family history would suggest an acquired intrauterine cause.

Plantin et al. mentioned the adherence of the amniotic membrane to the fetus's skin during pregnancy as a possible cause. The laboratory tests could not confirm a viral infection in any of the cases.[3,8] The different types and stages of the lesions were observed in our patient, which led us to an interpretation of the evolution sequencing, from the erythema to the formation of linear reticulated scars. At the beginning, the rounded or oval whitish vesicles appeared over erythematous skin, then became eroded and converged, originating a linear, reticulated pattern. The limbs presented depressed linear scars, whereas on the neck and thorax they were raised, with reticulated patterns, which characterize this entity. Differential diagnosis was reached with bullous genetic disorders and infectious diseases with vesicles and blisters.[8,14] Epidermolysis bullosa [1,6,9-10,12,15] was dismissed due to the absence of new blisters after trauma and the reticulated appearance of the scars. Stein et al.[9] reported the case of a male who was diagnosed with epidermolysis bullosa for 9 years due to his recurrent vesicular-blistering condition. In focal dermal hypoplasia[1,2,6,10,12] the patient may be born presenting eroded lesions, but the circumscribed atrophic lesions which followed Blaschko lines, the subcutaneous cell tissue hernias and the periorificial papilloma are the keys to the diagnosis. Incontinentia pigmenti[10] in its initial phase can present vesicles and blisters with linear distribution which follow Blaschko lines.

Shidu-Malik et al. described a neonate who had the appearance of a collodion baby with ectropion.[3] In our case, we have pointed out a resemblance to collodion baby at the neonatal stage, as well as further ichthyosiform scaling on the scalp, pre-sternal and diaper areas. From the histopathological point of view, hypergranulosis and hyperkeratosis were similar, both in the neonatal and infancy biopsies.

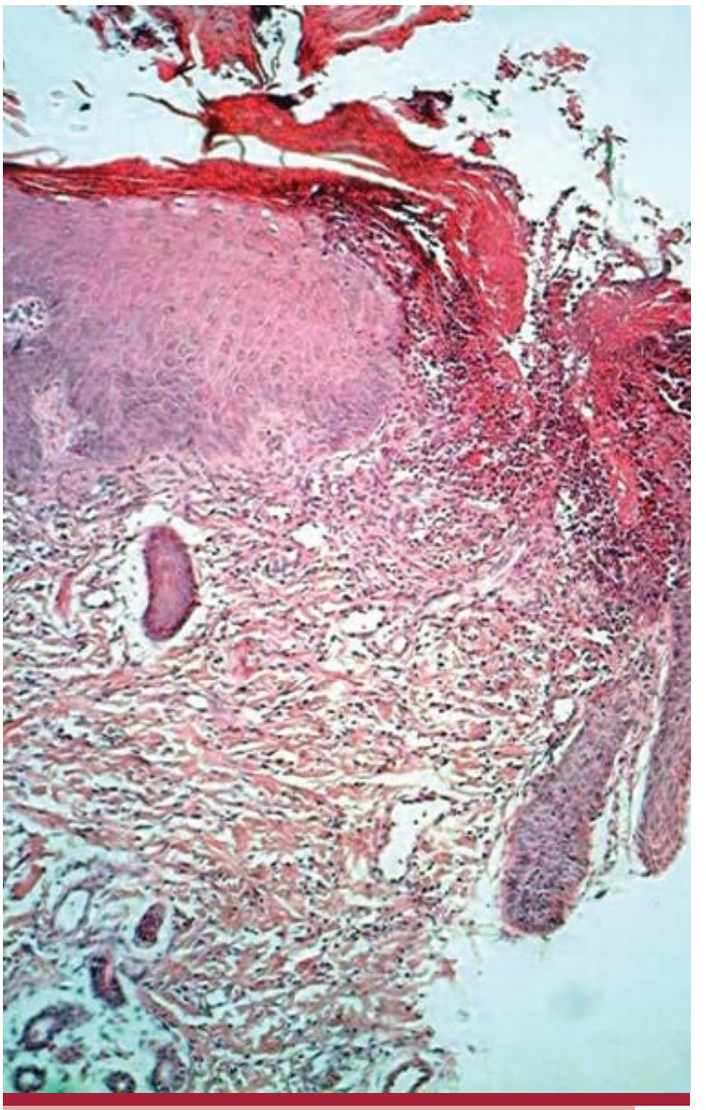

Photo 6: On left end, epidermal acanthosis, mild hypergranulosis and hyperkeratosis. The middle region reveals more acanthosis and relevant parakeratosis. The opposite end shows necrotic, purulent acute infundibular folliculitis focus (Hematoxylin-Eosin stain, original magnification: 100x.

It is rare for the staphylococcal scalded skin syndrome [5,6,7,10] to be congenital. It is caused by epidermolytic toxins produced by Staphylococcus aureus and heals without any scarring sequelae.

In the case of our patient, CEVDHRSS diagnosis resulted from clinical findings and dismissal of other skin pathologies presenting erosions, blisters or vesicles at birth. Reticulated scarring, characteristic of this disease, supported diagnosis. The etiopathogenesis of this benign, infrequent congenital skin disease is still obscure.

TABLE 1: Congenital erosive and vesicular dermatosis healing with reticulated supple scarring. Summary of findings in each clinical case

Author/Year/ Case Nª	Sex/ GA/Weight/ Perinatal antecedents	Location	Type of lesion at birth	Evolution	Mucosa/Oral cavity/ Sweating	Optical microscopy/ Electronic microscopy	Intellectual and motor development
Cohen et al. 1 1985 Case 1	F. 28 weeks. 1,000 gr. Gemelar. Placenta previa. Pelvic presentation. No consanguinity.	Scalp, trunk and limbs.	Crusting erythematous eruption with generalized scaling. Isolated blisters. 75 % skin involvement.	Age 11: cobble-stone scars on scalp and forehead. Reticular scars on back, chest and limbs. Longitudinal, parallel scars on limbs. Scarce hair, absence of toenails on both halluces and on fourth right toe. No recurrence.	Yellowish scarring patch on tongue. No sweating in scarring areas. Compensatory hyperhidrosis. Hyperthermia Skin fragility.	Biopsies: ages 1,12 and 14. Scarring area of back. Decrease of hair follicles and absence of eccrine glands.	Normal.
Cohen et al. 1 1985 Case 2	M. 34 weeks. 1,630 gr. Placenta with ischemic signs. No consanguinity.	Trunk and limbs.	Fissures which produced crusting reticular ulcerations. 75% skin involvement.	Age 5: symmetric, extensive reticulated scars on arms, back, chest. No recurrence.	Altered sweating in affected areas. Heat intolerance.	Left buttock scarring area biopsy: dermis thickening, no skin appendages and slight decrease of elastic fibers.	Normal.
Cohen et al. 1 1985 Case 3	F. 29 weeks. 1,190 gr. No consanguinity.	Scalp, trunk and limbs.	Erosion patches on limbs. Blisters on upper limbs. Alopecia with scales on occiput. Scales on remaining skin. 75% skin involvement.	Age 5: generalized reticulated scars on trunk and limbs. 2-cm alopecia patch on scalp with cobble-stone scar. Hypoplastic fifth left fingernail.	Absence of papilla on some areas of tongue. No sweating in affected areas. Compensatory hyperhidrosis.	Scarring area biopsy: slight increase of collagen fibers and hypocellularity. No eccrine glands.	Hemiparesis.
Gupta et al. 5 1987 Case 4	M. 35 weeks. 2,720 gr. Apgar 9/10. No consanguinity.	Scalp, trunk and limbs.	Erythematous patches, deep erosions, vesicles. Over 75% skin involvement.	Age 3 months: scar healing of lesions. Age 8: symmetric, extensive reticulated scars. Several hypoplastic or absent hand nails. No recurrence.	Affected tongue. No sweating in affected areas. Hyperthermia. Compensatory hyperhidrosis.	Left forearm scar biopsy (age 8 months): loss of rete ridges pattern. No eccrine glands. Decreased elastic fibers.	Brain paralysis. Macular scar.
Gupta et al. 5 1987 Case 5	M. Term neonate.	-	Vesicles, su perficial erosions.	Age 2 months: reticulated scars. Age 4: scarring lesions. No recurrence.	-	-	Brain paralysis.
Plantin et al. 7 1990 Case 6	M. 30 weeks. 1,000 gr. C-section. Apgar 3/6. Normal placenta. No consanguinity.	Face, limbs. Face, trunk, soles.	Diffuse eczematous dermatosis. Adherent membranes, possible remains of amniotic membrane. They cause erosions when removed.	Age 1: reticulated scars. Scarring alopecia.	Age 1: scars on tongue. No hyperthermia.	Scarring area biopsy: slight epidermal atrophy. No eccrine glands.	Normal.
Sadick et al. 8 1995 Case 7	M. Term neonate. 40 weeks.	Trunk and limbs.	Multiple erosions and crusts. On the face, pustules and erosions. Flaccid blister on right eyelid. Cribriform scar on right thigh. Linear lesions. Less than 75% skin involvement.	Postnatal blistering occurrence. Age 7 days: healed skin lesions. Age 2 months: depressed reticulated scars. No further recurrence. Age 10 months: scars on face, trunk, soles.	Spared tongue. No hyperthermia.	Dorsal area erosion biopsy: epidermal erosions, fibrin and neutrophils on ulcer bed. Parakeratotic scales on adjacent skin. Papillar and reticular dermis edema. Diffuse interstitial infiltrate with predominant neutophils, few eosinophils, histiocytes and multinucleated giant cells. Left leg scar biopsy (age 8 weeks): flattened rete ridges. Minimal perivascular lymphocytic infiltrates. Anagen hair follicles.	Normal.
Fernández et al.11 1997 Case 8	M. 29 weeks.	Scalp, forehead, trunk and limbs.	Generalized erosions and vesicles.	New vesicles after perinatal period.	No hyperthermia.	After 10 days, ulcerations, subcorneal pustules. Age 8 months: adipose tissue nests on reticular dermis, connective tissue increase, normal eccrine glands.	Normal.
Sidhu et al. 3 1998 Case 9	F. 24-25 weeks. 650 gr. Apgar 3/6 PMR. No consanguinity. Postransfusion TORCH + for herpes. Sepsis.	Head, neck, intertriginous areas. Back, shoulders, buttocks.	Neonate: generalized erosions on scalp, trunk and limbs. Spared palms and soles. Lesions heal in 2 weeks with scars. 75% skin involvement.	Age 3: reticulated scars. Scarring alopecia. Scars on palms, back and lateral edges of fingers. Anonychia on fifth finger of right hand. Recurrence on fingers and ankles. Skin fragility.	Compensatory sweating. Heat intolerance.	-	Normal. Bilateral partial hearing loss. Recurrent papilloma.
Sidhu et al. 3 1998 Case 10	M. 28 weeks. 871 gr. Apgar 3/ 4-6. PMR. No consanguinity.		Collodion baby-like membrane with scaling, fissures, erosions on head, neck, intertriginous areas, bilateral ectropion.	Age 8 months: generalized reticulated scars. Palms, soles and parts of face unaffected. Anonychia on three left fingers and left foot. Scarring alopecia.	-	-	Corneal scar. Cognitive and motor retardation. Low weight and height.
Sidhu et al. 3 1998 Case 11	F. 28 weeks 980 gr. Apgar 6/8. Breech presentation.		Age 3 days: symmetric erosions on upper back, back of shoulders, upper part of arms. Small, whitish scars on back and buttocks..	Age 7 months: whitish reticulated scars on back part of shoulders and upper back. Scars on trunk. No recurrence.	Normal sweating.	EM: granular deposits in keratinocytes.	Normal.

Author/Year/ Case Nª	Sex/ GA/Weight/ Perinatal antecedents	Location	Type of lesion at birth	Evolution	Mucosa/Oral cavity/ Sweating	Optical microscopy/ Electronic microscopy	Intellectual and motor development
Stein et al. 9 2001 Case 12	M. 29 weeks. 1,227 gr. Meconium-stained amniotic fluid (AF). MV. Sepsis. Negative cultures.	Trunks, genitalia and limbs.	Neonate: scalded skin. Extensive erosions affecting mucosa. No eyelashes. Blocked tear ducts.	Age 2 months: linear, reticulated, hypo and hyperpigmented scars. Recurrent reticular erosive lesions on trunk, penis and limbs. Anonychia of 20 nails. Eyelashes scarring alopecia. Chronic conjuctivitis. Recurrence. Skin fragility.	Spared tongue. No hyperthermia.	1st week after birth: epidermal necrosis and dermal inflammation. Ages 3 and 4: spongiosis and inflammation on bullous recurrences. Direct Immune Fluorescence of perilesional skin: no evidence of bullous lesion. Mononuclear EM and Ac. Intact epidermal cells with no granular deposits. Normal hemidesmosomes.	Normal.
Vun et al. 2 2005 Case 13	M. 26 weeks. Apgar 6/8. No consanguinity.	Abdomen. Left cheek, forearm and lumbar area.	Erosion areas 3 days after birth with further crusts and scars. Less than 75% skin involvement.	Age 3 months: reticulated scar. Age 8: scar on left cheek. No recurrence. No skin fragility.	Spared tongue. No heat intolerance.	-	Normal.
Metz et al. 10 2005 Case 14	F. 27 weeks. 830 gr. No consanguinity.	Scalp, trunk and limbs.	Blisters.	Age 4 months: diffuse linear and reticulated hypo and hyperpigmented patches on trunk and limbs. Scarring alopecia. Hypoplastic fingernails. Age 3: vesicle recurrence on trauma areas.	Spared tongue and mucosa. No sweating in affected areas. No hyperthermia. Compensatory hyperhidrosis.	Inflammatory skin biopsy: vesicles on superficial papillar dermis. Scar biopsy: slight increase of collagen in dermis.	-
Goncalves et al. 6 2007 Case 15	M. 35 weeks. 2,065 gr. Apgar 8/8. C-section: maternal urinary tract infection (nitrofurantoin) AH. No consanguinity.	Scalp, trunk and limbs.	Erosions and vesicles. Erosions on scalp, trunk and limbs. Eroded blisters with peripheral exfoliation. Spared palms and soles. Over 75% sklin involvement.	Age 45 days: scarring phase. Age 3 months: hypo and hyperpugmented reticulated scars. Age 3: reticulated scars. Thickened, erythematous, scaling skin on palms and soles. No recurrence.	Heat intolerance without compensatory hyperhidrosis. Painful lesions.	Biopsy on 2nd day: epidermal necrosis, basal keratinocytes vacuolization and subepidermal cleavage. Civatte bodies in dermis. Perivascular inflam matory infiltrate with neutrophils near the epidermal necrosis area. Absence of eccrine glands.	Ear dystrophy, low implantation. Bilateral syndactily on 3rd and 4th toes.
Lee et al. 13 2008 Case 16	F. Term neonate. 38 weeks. 3,180 gr. C-section due to previous C-section. Negative TORCH.	Scalp, trunk and flexural areas.	Hemorrhagic, erosive patches on trunk and flexural areas. Reticulated stretch marks. Atrophic strips on trunk and flexural areas of limbs. Alopecia patches. 30% skin involvement.	No recurrence.	-	At birth: erosion edge biopsy: epidermal erosion. Edema on papillar dermis. Collagen fibers increase, adnexal structure decrease, diffuse infiltrate with lymphocytes, plasmatic cells, eosinophils, histiocytes. Biopsies of reticulated atrophic strips and scalp: scarring tissue without hair structures.	-
De Lange et al. 4 2009 Case 17	M. 31 weeks. 1,250 gr. Oligoamnios C-section. Alphaphetoprotein increase. IGR.	Forehead, trunk, limbs.	Erosions on thorax, limbs, forehead. Reticulated scars on back. Frontoparietal alopecia. 80% skin involvement.	Age 7 weeks: no new lesions. Reticulated scars. Age 13 weeks: persistent strips on right iliac fossa.	Small cyst and whitish reticulated scar on tongue.	Biopsy at birth: ulcerated lesion covered with parakeratotic scales with fibrin and neutrophils. Edema, interstitial and perivascular infiltrate with neutrophils, lymphocytes, eosinophils and some multinucleated giant cells. Scar biopsy: connective tissue sclerosis and normal vascular structures.	Normal.
Ma et al. 12 2009 Case 18	F. Term neonate. No consanguinity.	Forehead, face, neck, trunk, limbs.	Vesicular, erosive lesions.	Age 2 weeks: linear reticulated scars. No recurrence.	-	Increase of collagen fibers, decrease of hair follicles and eccrine glands.	-
Rositto et al. 2010 Case 19	F. Term neonate. 3,000 gr. Apgar 9/10. No consanguinity.	Trunk, abdomen, lower limbs.	Age 9 days: linear erosions on trunk and abdomen, inner thighs and popliteal folds. 75% skin involvement.	Age 2 months: scars with reticulated pattern on thorax, neck. Linear scars on folds of thighs and upper limbs. Age 21/2: scars and scaling on presternal region and diaper area.	Normal sweating.	Neonatal biopsy: acanthosis with mild hypergranulosis, hyperkeratosis and parakeratosis. Acute foci of necrotic, purulent infundibularfolliculitis. Scars biopsy: slight acanthosis and mild hyperkeratosis with retention of granular layer.	Normal.

Abbreviations

F: female, **M:** male, **GA:** gestational age, **NB:** newborn, **TN:** term neonate, **PMR:** premature membrane rupture, **MV:** mechanical ventilation, **AH:** artery hypertension, I**GR:** intrauterine growth retardation, **EM:** electronic microscopy, **UL:** upper limbs, **LL:** lower limbs

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