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Michelin Baby Associated With Facial Abnormalities☆

Bebé Michelin asociado a anomalías faciales

To the Editor:

The term Michelin tire baby (MTB) was first used in 1969 by Ross[1] to refer to the case of a baby with multiple, widespread skin folds associated with hemihypertrophy and a broad bridge of the nose. We report the case of a girl with MTB and mild associated dysmorphic alterations.

A 3-month-old girl of Romanian origin, born at 34 weeks, with no history of consanguinity, was seen for excessive skin folds. She had no personal or family history of interest. Physical examination revealed abundant circumferential folds of skin of soft but solid consistency around the limbs and neck (Fig. 1). In addition, we observed microcephaly with frontotemporal flattening and hypotelorism, small, low-set ears, and teletelia. Skin biopsy showed an excessive accumulation of adipose tissue in the superficial dermis (Fig. 2). Cardiology study and transfontanellar ultrasound were normal. Ophthalmologic examination revealed convergent strabismus, microphthalmos, and blepharophimosis. The karyotype was normal, 46 XX.

A proposal has been made to replace the original term Michelin tire baby by the term circumferential skin folds syndrome or phenotype when the condition is associated with extracutaneous alterations.[2] These associated manifestations, detected in up to 74% of MTB patients, include facial abnormalities, neurologic disturbances (mental retardation, delayed psychomotor development, hypotonia, convulsions), ocular and cardiac alterations, genital hypoplasia or malformation, scoliosis, and pectus excavatum (Table 1).[2–5] Our patient presented teletelia, not previously reported in association with MTB. To date, no consistent pattern of inheritance has been identified.[6]

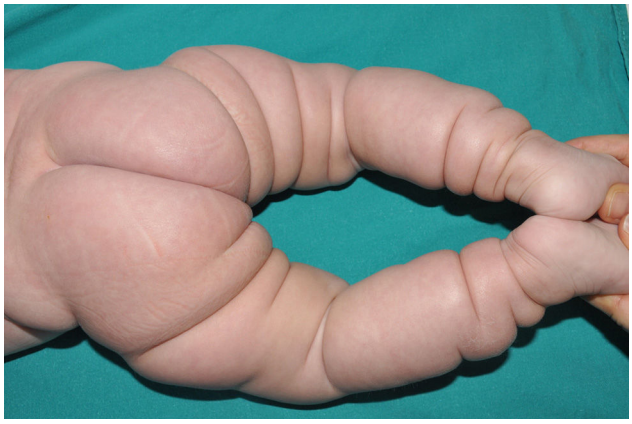

Figure 1 Abundant circumferential skin folds of soft but solid consistency around the limbs and neck.

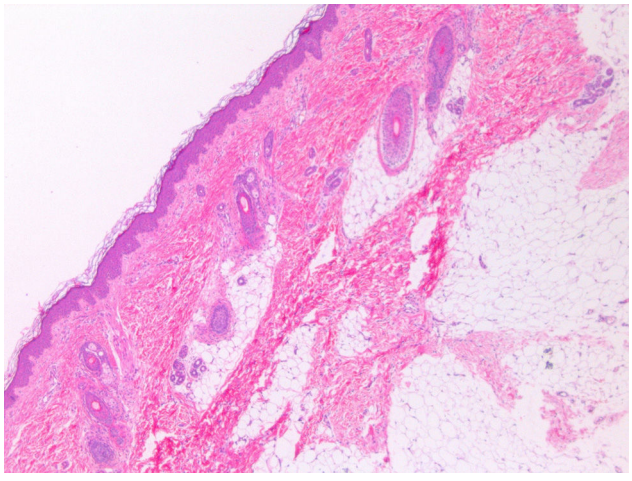

Figure 2 Skin biopsy showed an excessive accumulation of adipose tissue in the superficial dermis.

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Table 1 Summary of the Number and Percentage of Cases of Circumferential Skin Folds Syndrome Reported in the Literature up to the Time of Writing This Review Article, Including Our Case.

Description	Number	Percentage
All cases	45	100
Lipomatous nevus	8	17
Smooth muscle hamartoma	10	22
Facial abnormalities[a]	27	60
Neurological alterations[b]	20	44
Eye alterations[c]	10	22

[a] Most frequent facial abnormalities, reported in 3 or more patients: malformations of the auricles of the ear, broad or sunken bridge of the nose, epicanthic folds, hypertelorism, cleft palate, microphthalmos, micrognathia, and central facial flattening.
[b] Most frequent neurologic abnormalities, reported in 3 or more patients: delayed psychomotor development, mental retardation, dilatation of the ventricles, hypotonia, convulsions, and microcephaly.
[c] Most frequent eye abnormalities, reported in 3 or more patients: blepharophimosis and strabismus or esotropia.
Source: Adapted and updated from Rothman[2] 2014.

Histology in MTB can reveal signs of smooth muscle hamartoma, lipomatous nevus, degenerated collagen, scarring, or even normal skin, although biopsy was not performed in 59% of reported cases.[1,3]

This new case of MTB with lipomatous nevus associated with facial dysmorphism, ocular alterations, and teletelia, highlights the need to perform complete clinical evaluation in these children, including neurologic and ophthalmologic examination. When associated malformations are present, the term circumferential skin folds syndrome should be used.

Conflicts of Interest

The authors declare that they have no conflicts of interest.

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Annular Elastolytic Giant Cell Granuloma in Sun-Protected Sites Responds to Dapsone☆

Granuloma elastolítico anular de células gigantes en área no fotoexpuesta con respuesta a dapsona

To the Editor:

We present the case of a woman aged 59 years with a history of left renal agenesis. She came to outpatients with a 2-month history of persistent painful lesions on both hands. She had been treated with oral corticosteroids (deflazacort, 30 mg, in a tapering regimen) and topical methylprednisolone aceponate with no improvement. On examination, indurated red-violaceous papulonodular lesions were observed on the dorsum of the fingers and on the palms of both hands (Fig. 1A).

Histology revealed a dermal granulomatous lesion with no areas of necrobiosis and with numerous multinucleated giant cells showing elastophagocytosis, suggestive of annular elastolytic giant cell granuloma (AEGCG) (Fig. 2, A and B).

After making the diagnosis and because of the lack of response to corticosteroid therapy, treatment was started with dapsone, 100 mg/d, leading to complete resolution of the lesions after 4 months of treatment, with no recurrence after a year of follow-up (Fig. 1B).

AEGCG is a rare granulomatous disease characterized by the appearance of annular lesions with elevated borders formed of erythematous papules that spread centrifugally to leave an atrophic central area. Histologically, dermal granulomas formed of histiocytes and multinucleated giant cells are observed in close association with degenerated elastic fibers and elastophagocytosis. The lesions are usually asymptomatic, although mild pruritus or pain can develop, and they tend to persist for months or years, after which spontaneous remission typically occurs. The first case was described in 1975 by O'Brien,[1] who coined the term actinic granuloma as the lesions arose in sun-exposed areas. In 1979,

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