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A case of atrophoderma vermiculatum responding to systemic isotretinoin

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Abstract

Background: Atrophoderma vermiculatum is a rare, benign follicular disorder that primarily affects children. It is characterized by symmetric reticular or honeycomb atrophy of the cheeks that may extend to the ears and forehead. The defect is believed to be due to abnormal keratinization in the pilosebaceous follicle. Management of atrophoderma vermiculatum is challenging.

Main observations: We report the case of a 10-year-old boy, diagnosed as atrophoderma vermiculatum. The boy underwent two 6-month treatment courses with isotretinoin at a dose of 0.50mg/kg/day, which resulted in partial remission of the lesions. Even though the disease did not completely regressed, isotretinoin stopped atrophy progression and improved cosmetic appearance of the skin.

Conclusions: In this case of a child with atrophoderma vermiculatum isotretinoin stopped atrophy progression and improved cosmetic appearance of the skin. No adverse events were observed. However, always, when applying isotretinoin in children, it has to be considered that, apart from possible common adverse events, this treatment may promote premature epiphyseal plate closure and may stop normal growth in some children.

Introduction

Atrophoderma vermiculatum is a rare, benign follicular disorder that primarily affects children. It is characterized by symmetric reticular or honeycomb atrophy of the cheeks that may extend to the ears and forehead. The defect is believed to be due to abnormal keratinization in the pilosebaceous follicle. Management of atrophoderma vermiculatum is challenging.

Case report

We report the case of a 10-year-old boy who presented with multiple, grouped, pitlike atrophic scars, symmetrically distributed over his cheeks, giving a worm-eaten appearance on the skin (Fig. 1A). The lesions were similar regarding their size and morphology, were oval-shaped, flesh-colored and measured about 1-2mm each. The depth of each individual lesion was about 1mm. The lesions were first observed at the age of 5, and gradually expanded in a centrifugal way, affecting a circular area over both cheeks. Eyebrows and eyelashes had a normal appearance. The boy did not report any subjective symptoms, including pain or pruritus.

According to his medical history, he did not experience any physical trauma or inflammation on the affected area before the disease onset. In addition, the patient did not report any known allergies or history of atopy. Physical examination and routine laboratory test results were unremarkable. Family history was also negative.

The anamnesis, specific clinical features, age of the patient and the negative personal history of the patient led us to the diagnosis of atrophoderma vermiculatum. The boy underwent two 6-month therapeutic courses with isotretinoin at a dose of 0.50mg/kg/day, two weeks apart. Even though the lesions did not completely regress, isotretinoin stopped atrophy progression and improved cosmetic appearance of the skin (Fig. 1B). Other than temporary pruritus, mild exfoliation and dryness of the skin and lips, our patient did not develop any adverse effects. At present, 1 year after treatment cessation, the boy remains stable and free of new lesions.

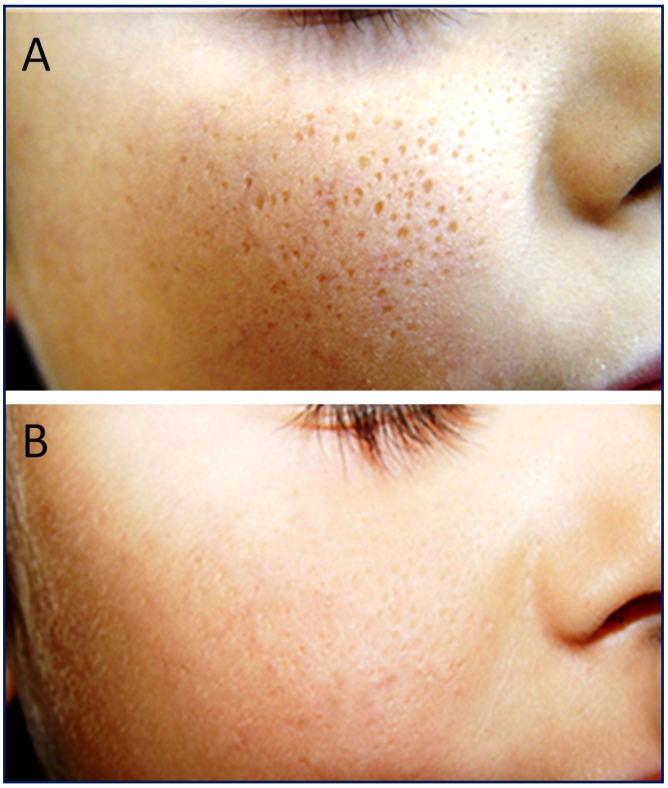

Figure 1

A) Patient before treatment: grouped, pitlike atrophic scars over the cheek, B) Improved cosmetic appearance of the skin after systemic treatment with isotretinoin.

Discussion

Atrophoderma vermiculatum, known also as folliculitis ulerythematosa reticulata, is a rare, benign follicular disorder that primarily affects children. It is characterized by symmetric reticular or honeycomb atrophy of the cheeks that may extend to the ears and forehead.[1,2] Atrophoderma vermiculatum, along with keratosis pilaris atrophicans faciei and keratosis pilaris spinulosa decalvans, represent clinical variants of keratosis pilaris atrophicans.[1,2] Most cases are sporadic, but there have been reports of autosomal dominant inheritance. The defect is believed to be due to abnormal keratinization in the pilosebaceous follicle.[3,5]

Management of atrophoderma vermiculatum is challenging, as all 3 variants of keratosis pilaris atrophicans are exceptionally difficult to treat. Atrophoderma vermiculatum is mainly a cosmetic problem. However, taking under consideration the emotional stress and the permanent atrophy that it provokes, a therapeutic attempt is considered reasonable. Topical treatment options, that have been suggested for this condition include keratolytics, topical and intralesional steroids, and ultraviolet irradiation. When the disease remains stable and well-controlled, dermabration, collagen implants, CO2 and Erbium Yag lasers can be also used.[3,6] The use of topical retionoids was also suggested for keratosis pilaris atrophicans faciei.[7]

Systemic use of retinoids in patients with keratosis pilaris atrophicans, as applied in our patient, is controversial. There have been previous literature data reporting a beneficial effect of prolonged use of oral retinoids.[5] Among oral retinoids, isotretinoin appears to hold the advantage of a shorter half-life, which minimizes possible adverse events.[5] It has to be considered that, apart from possible common adverse events, this treatment may promote premature epiphyseal plate closure and may stop normal growth in some children.[8] In our patient, at 1 year follow-up after treatment cessation, no permanent adverse events were noted.

In conclusion, in this case of a child with atrophoderma vermiculatum isotretinoin stopped atrophy progression and improved cosmetic appearance of the skin. No adverse events were observed.

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