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Case Report

Punctate Palmoplantar Keratoderma Associated with Atopic Dermatitis: A Case Report

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Abstract

Observations: Punctate palmoplantar keratodermas are rare autosomal dominant cutaneous disorders characterized by numerous hyperkeratotic papules that are irregularly distributed on the palms and soles. The differential diagnosis of punctate palmoplantar keratoderma include warts, *Darier*'s disease, basal cell carcinoma, arsenical keratoses and pachyonychia congenita.

Symptomatic therapeutic approach is essential, and for this goal keratolytics are preferred. Both topical and systemic retinoids have been used with variable success. We report a 6 year-old boy with punctate palmoplantar keratoderma associated with atopic dermatitis.

Introduction

Palmoplantar keratodermas (PPK) are characterized by excessive formation of keratin on the palms and soles. PPKs are classified as acquired and hereditary forms. Acquired forms are usually associated with paraneoplastic syndrome, AIDS, psoriasis and lichen. Hereditary forms include diffuse palmoplantar keratodermas (*Unna-Thost*), punctate palmoplantar keratoses, *Papillon- Lefèvre* syndrome, mal de Meleda, *Howel-Evans* syndrome, acrokeratoelastoides [1].

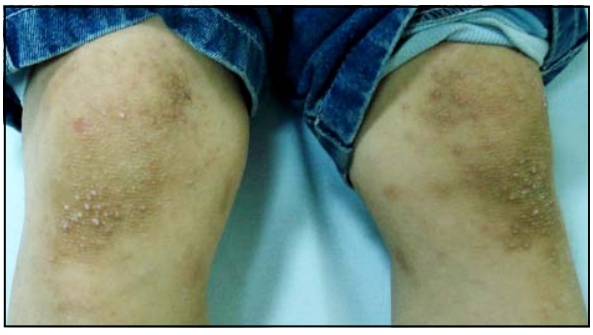

Figure 1. Knees of the patient with keratosis pilaris

Punctate palmoplantar keratodermas (PPPK) are also known as keratosis punctate palmaris et plantaris, *Buschke–Fischer–Brauer* type. It is clinically characterized by multiple keratoses, 2-8 mm in diameter, which may vary in size and tend to be more prominent, verrucous over time [2].

Case Report

A 6-year-old boy presented to our clinic with diffuse pruritus, xerosis and warty like lesions on palms, soles, elbow and knees. Xerosis and pruritus began when he was 6 months old. He also complained of recurrent herpes like symptoms on lips and pruritic scaling erythema on the face, and extremities. Warty like yellowish lesions on his palms and soles started to develop in the last few years. There was no family history of atopy. Dermatologic examination revealed *Dennie-Mor-*

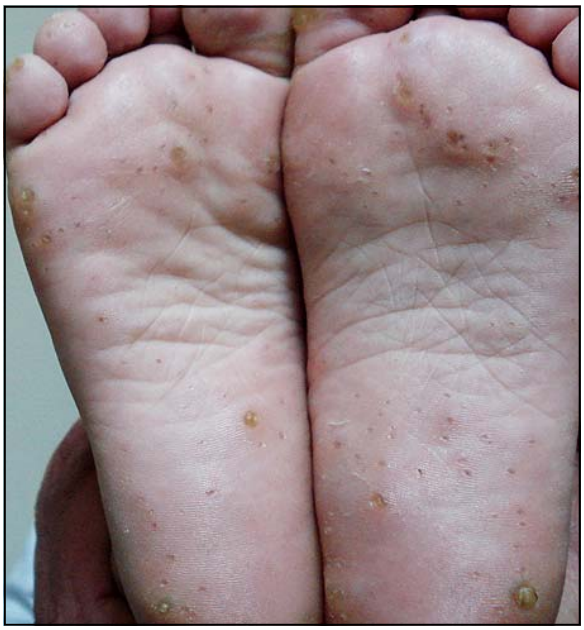

Figure 2. Soles of the patient with multiple hyperkeratotic papules

gan lines, diffuse xerosis, angular cheilitis, dull pink keratotic papules topped by keratotic pits on elbows and knees (**Figure 1**). Also hyperkeratotic yellow-grey papules were seen over the palms, soles, distal interphalangeal and metacarpophalangeal joints (**Figure 2**). He had remarkable half moon distal leukonychia with pink arc on all fingernails. Oral mucosa was not involved. Histologic examination of a punch biopsy specimen taken from the hyper keratotic papules on palms showed an acanthotic epidermis with prominent granular layer, hyperkeratosis with focal parakeratosis, dilated vessels in the papillary dermis.

Laboratory investigation revealed Hb: 11.7 g/dl (13.5-17.5), Htc: %36.2 (41-53), MCV: %69.8 (80-100), ferritin: 11.57 ng/ml (30-400), total IgE: 197 IU/ml (14.4-52) with negative VDRL. The blood chemistry and thyroid function tests were within normal limits.

Discussion

PPPK is a rare form of keratoderma. The prevalence is 1 case per 100,000 population. This condition shows an autosomal dominant inheritance, however, sporadic cases are encountered. The age of onset is variable [**1, 2**]. Although to date no specific genes have been identified, *Zhang* et al. identified the chromosome 8q24.13–8q24.21 as the responsible gene in two Chinese families [**3**].

Atopic dermatitis (AD) is a common skin disorder in childhood and is characterized by recurrent severe pruritic eczema. AD was diagnosed by the criteria of *Hanifin-Rajka* [**4**]. With these criteria, the patient must exhibit three of the major and three of the minor criteria. Our patient had 3 major criteria (pruritus, typical morphology, chronic recurrent dermatitis) and 5 minor criteria (xerosis, early age onset, elevated serum IgE, cheilitis and *Dennie-Morgan* infraorbital fold). Variable nail changes in the form of half moon distal nail plate dystrophy, subungual hyperkeratosis, longitudinal ridge, thickened nail plate, medial canaliformis, lunular changes, onychogryphosis, transverse ridge and koilonychia have been reported in PPPK [**5**]. Leukonychia may be present in in PPKs [**2**]. Histopathological examination is not diagnostic for PPPK which shows hyperkeratosis and parakeratosis, basal layer spongiosis and dilated, occluded sweat ducts, blood vessels, and lymph vessels [**1**]. Differential diagnosis of PPPK include warts, callus, punctate porokeratosis, *Darier* disease, arsenical keratoses. Verruca vulgaris demostrate multiple bleeding points clinically and histopathologically characteristic foci of vacuolated cells and koilocytotic cells.

Punctate porokeratosis is very similar to punctate keratosis and is distinguished by presence of a cornoid lamella histologically. *Darier* disease may show multiple punctate palmoplantar keratoderma. However, *Darier* disease is characterized by the presence of warty papules in the seborrheic areas and mucosal lesions as well as nail involvement. Arsenical keratosis distinguished by their histopathological examination.

Treatment of keratodermas is difficult and symptomatic. On the basis of the literature, lactic acid or urea containing preparation and topical retinoids have been used with variable success [**1, 2**]. Successful treatment of PPPK with oral retinoids has been reported [**6, 7**]. We started treatment with 20% salicylic acid in vaseline and 5% salicylic acid in 10% urea cream once daily that was subsequently discontinued because of lack of efficacy after 1 month. He was treated topically with 0.1% retinoic acid ointment with a good clinical response.

PPK associated with AD has been reported uncommonly.

Anderson et al and *Mitteral* et al have shown that there is a significant association

between atopy and keratoses [**5, 8**]. Keratosis punctata of the palmar creases (KPPC) was considered a morphological variant of PPPK was reported to be associated with AD [**1**]. *Loh* et al reported six cases of palmar-plantar keratoderma of *Unna-Thost* associated with AD [**9**]. Recently *Kumari* et al reported a 8 year old male with the diagnosis of keratosis palmoplantaris in association with keratosis pilaris. They declared this as the first case report of punctate keratoderma with an early age of onset unusually appearing with keratosis pilaris on elbows and knees [**10**].

In conclusion, we think that PPPK associated with AD are not uncommon conditions, but may be underreported.

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