Asymmetric Periflexural Exanthem in an Adult

Sir,

Asymmetric periflexural exanthem of childhood (APEC) or unilateral latero-thoracic exanthem of childhood is a "new" disease, which has never been reported affecting adults. We here present a sporadic case observed in an adult male.

CASE REPORT

In winter, a 33-year-old man was seen for a papular eruption affecting the left axillary fold and left lateral surface of his chest. Papules were pink, small (2–3 mm in diameter), strictly grouped but never coalescent; peripherically papules were more sparsely arranged. Left axillary lymphadenopathy was found by palpation (Fig. 1). Dermatitis had not been preceded or accompanied by the herald patch of pityriasis rosea.

The patient was in general good health and complained of only mild pruritus. He had had no fever, no insect bites, and no intake of drugs.

Oral antihistamines and a short-term topical therapy with crotamiton and a moderate potency corticosteroid failed to improve dermatitis, which, after 15 days, centrifugally spread to both sides. Results of routine blood examination and complete blood cell count were normal except for a mild eosinophilia (8%). Sedimentation rate, antistreptolysinic and antistaphylo lysinic titres were negative. Serologic examination for Borrelia burgdorferi, HIV, hepatitis A, B, C viruses, Epstein Barr virus, cytomegalovirus and enteroviruses was negative. Only toxoplasma IgG and rubella IgG showed a weak positivity. Skin biopsy showed a non-specific superficial dermatitis characterized by a moderate perivascular lympho-monocytic infiltrate, exocytosis and spongiosis.

One month later dermatitis was resolved, with weak brownish hyperpigmentation, dryness and turgiduous desquamation. No recurrence was observed after 6 months.

DISCUSSION

APEC is an exanthem of unknown aetiology, recently described by Taieb et al. in 1986 (1), but a very similar clinical and pathological disease was previously reported by Brunner et al. in 1962 (2).

The exanthematic eruption, the possible association with upper respiratory tract infections, the occurrence within families sometimes observed, the age of children and the seasonal preference of onset (spring, summer) seem to support the hypothesis of an infectious aetiopathogenesis (3, 4). Nevertheless a true epidemic course could be suspected only in the series of Laur (5). While the aetiologic role of Spiroplasma was not confirmed (1, 3), a viral agent is now supposed (3). Gelmetti et al. (6) found similarities between Giannotti-Crosti syndrome and APEC, as both may be considered a localized form of a systemic disease.

APEC is always clinically described as a maculo-papular scarlatiniform eruption or an eczematous dermatitis which involves the skin folds (axilla, groin) with a granular texture on palpation (1–10). It is moderately pruriginous, and a mild local lymphadenopathy is found in about 50% of cases. Exanthem after 1–2 weeks centrifugally spreads on thorax, and sometimes a widespread diffusion of sparse papules is observed. Resolution with mild hyperpigmentation or pityriasis desquamation is noted in about 1 month.

Skin biopsy usually shows the features of an aspecific inflammatory dermatitis.

In the absence of positive serological and laboratory tests, it is necessary to exclude other more common pathologies such as miliaria, acariasis, allergic contact dermatitis, drug eruptions, Giannotti-Crosti syndrome and atypical unilateral pityriasis rosea. The most difficult differential diagnosis to carry out is with atypical pityriasis rosea. The absence of the herald patch or a papular aspect characterizes both atypical pityriasis rosea and APEC. Nevertheless, in APEC asymmetry is predominant and the clinical course lacks subintranat typical eruptions of pityriasis rosea. Furthermore, on histological examination dyskeratotic cells of pityriasis rosea have never been found in APEC.

Since 1986 there have been numerous cases of APEC documented in literature (1, 3, 4, 6–10). In the years 1950–60 Brunner et al. and Laur refer to about two hundred cases of a disease which shares some morphological aspects with APEC (2, 5). The authors of both recent and past papers agree with the idea that APEC is not a rare disease, even if uncommon.

The exanthem is observed almost exclusively in children under 5 years (3, 4, 6) but one case aged 15 was reported in the series of Laur (5). To our knowledge no case of APEC in adults has been reported up to now. Our case suggests that asymmetric periflexural exanthem may occur also in adults, as

Fig. 1. Papular dermatitis involving the left axillary fold and left side of thorax (scar of the biopsy under the left nipple).
could be a reaction pattern rather than a rash related to a specific cause, even if an infective origin is probable.

REFERENCES


Accepted April 15, 1996.

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Palmar-plantar Erythrodysesthesia Syndrome and Other Cutaneous Side-effects after Treatment with Tegafur

Sir,

Tegafur is a fluorinated pyrimidine analogous to 5-fluorouracil, used in the treatment of digestive neoplasms with low myelosuppression and absence of immunosuppression. Neuologic, gastrointestinal and mucocutaneous side-effects are common (up to 2–8% of cases). Lesions are usually shown by slight stomatitis, skin dryness, disseminated rash, alopecia, diffuse or nail-restricted hyperpigmentation (1), Stevens-Johnson syndrome (2) and rarely palmar-plantar erythrodysesthesia syndrome (PPES) (3) or palmar-plantar keratoderma secondary to acral erythema (4).

PPES is characterized by a burning-dysesthetic erythema and oedema of palms and soles followed by desquamation. This syndrome has been described after usage of several chemotherapy drugs but until now we have found only one report of palmar-plantar keratoderma secondary to acral erythema related with Tegafur in the English literature (4).

We present 2 patients with digestive neoplasia and hepatic metastasis who developed a typical PPES (case 1) and with palmar-plantar keratoderma secondary to PPES associated with cutaneous diffuse, tongue and nails hyperpigmentation (case 2) after treatment with Tegafur.

CASE REPORTS

Case 1

A 57-year-old woman underwent a subtotal gastrectomy in 1998 for an adenocarcinoma. In March 1992, hepatic metastasis was detected and treatment with Tegafur 500 mg/day and Folinic acid 120 mg/day was started. Three months later, she developed a burning dysesthetic erythema and oedema of palms and soles (Fig. 1a), followed by fissuration and desquamation. On cutaneous biopsy, oedema, vascular proliferation and a discrete lymphohistiocytic infiltrate were seen in the dermis. In the epidermis, scattered necrotic keratinocytes, mild vacuolar degeneration of the basal layer and mild spongiosis were found. No changes were seen at the eccrine sweat glands. With the diagnosis of drug reaction, Tegafur was stopped and the lesions disappeared in 10 days. No other drugs were involved.

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Fig. 1. (a) Case 1. Symmetric, well-defined swelling and erythema of palms. (b) Case 2. Palmar-plantar keratoderma secondary to PPES.