sciendo

CASE REPORT Serbian Journal of Dermatology and Venereology 2019; 11 (2): 61-64

DOI: 10.2478/sjdv-2019-0009

Unilateral Mediothoracic Exanthem: Case Reports of two Indian Female Children

Shruti SHARMA, Rajesh L. PATHI, Anjali YADAV, Kamal AGGARWAL

Department and institution: Department of Dermatology, Venereology & Leprology, University of Health Sciences, Rohtak (Haryana)

*Correspondence: Dr Shruti Sharma, E-mail: kaush0211shruti@gmail.com

UDC 616.5-002.1-031.4-053.2(540)

Abstract

Unilateral mediothoracic exanthem is a variant of a common entity in Western literature, namely Asymmetric Periflexural Exanthem of Childhood (APEC). Less than ten cases of unilateral mediothoracic exanthem have been described worldwide, and only two are reported in Indian literature. Herein, we report two childhood cases of unilateral mediothoracic exanthem from India. Dermatologists must be aware of this self- limiting condition, in order to avoid invasive and exhaustive investigations and misdiagnosis.

Key words: Exanthema; Skin Diseases; Child; Thoracic Wall; Diagnosis; Treatment Outcome; India; Histamine Antagonists

Introduction

Asymmetric periflexural exanthem of childhood (APEC), a distinctive clinical entity usually seen in Caucasian children, is characterised by the acute onset of unilateral prurigenous erythematous rash, which may be maculopapular, micropapular, scarlatiniform or eczematous. It classically involves the flexures (axilla or groin) initially and later on, progresses centrifugally to involve trunk and extremities. The rash may become bilateral in later course of the disease (1, 2).

APEC is usually preceded by a prodrome of low-grade fever and involving upper respiratory tract or gastrointestinal tract. The general health of the child remains unaffected. It may be associated with regional lymphadenopathy. Spontaneous remission occurs in about 3 to 6 weeks (1–3).

A variant of APEC, namely unilateral mediothoracic exanthem (UME), is not periflexural in onset and usually touches the midline in contrast to APEC, where the rash being periflexural does not touch the midline (4). Only a handful of cases of UME have been described so far in worldwide literature (3, 5). To the best of our knowledge, the only cases described from India were in a 16-year-old adolescent boy and a 35-year-old female (5). Herein, we are reporting two childhood cases of UME from North India. Ours are the only childhood cases of UME in Indian literature.

Case studies

Case 1

A six-year-old female child presented with slightly itchy, skin coloured to reddish eruption affecting the right side of her body for five days duration. The rash began suddenly on her right chest and then progressed down to involve the same side of abdomen and right thigh.

The rash was not associated with pain, fever, constitutional symptoms, upper respiratory or gastrointestinal infection. There was no history of similar rash in siblings or parents. Any exposure to chemicals, sand, topical preparation, or pets was absent. Drug history was unremarkable. No triggering or precipitating factor was identified. The child was otherwise in good general health.

On cutaneous examination, micropapular skin coloured to erythematous exanthem was noted on the right half of chest and abdomen, which was strictly unilateral and stopped abruptly at the midline **(Figure 1 A)**. It extended to the right lower limb as well; however, axilla and groin remained uninvolved **(Figure 1 B)**. The rash was confluent and ec-

© 2019 The Serbian Association of Dermatovenereologists

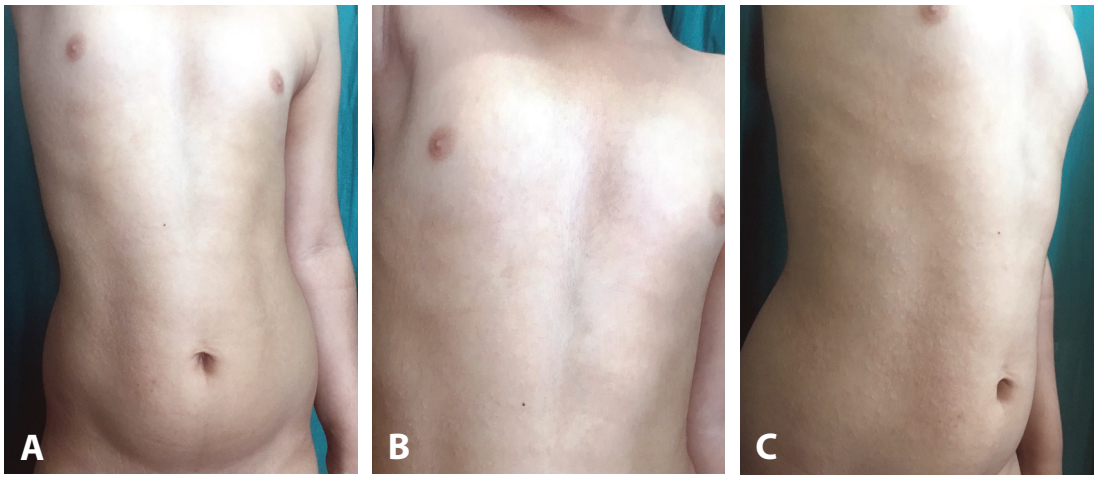

Figure 1. A). Skin coloured to erythematous rash on the right half of trunk, abruptly stopping at the midline. B). Unilateral mediothoracic exanthem sparing the axilla. C). The right half of trunk; unilateral exanthem is confluent and eczematous on the abdomen

zematous on the abdominal area **(Figure 1 C).** Vesicles, purpura and petechiae were absent. It was not associated with regional lymphadenopathy. Systemic examination revealed no abnormality.

Routine blood investigations, C-reactive protein and ASO titres were within normal limits. Serology for parvovirus B19, Epstein- Barr virus, cytomegalovirus, adenovirus, hepatitis B and C viruses were negative. The parents of this child did not give consent for skin biopsy. Based on history, distinctive distribution of rash, and investigations, the diagnosis of unilateral mediothoracic exanthem (UME) was made. Oral antihistamines and bland emollients were advised. Complete remission was noted within three weeks of the onset of rash.

Case 2

A five-year-old female came with complaint of asymptomatic skin coloured papular eruption on the left half of chest and abdomen for about a week. The rash also progressed to the left thigh, but never involved axilla, groin or contralateral half of body **(Figure 2 A and B)**. Vesicles and purpura were not found.

There was no history of fever, upper respiratory or gastrointestinal infection. Family history was unremarkable. No triggering factors, for instance, chemicals, sand, topical or oral drugs were identified. General health was good and systemic examination revealed no abnormality. Lymphadenopathy was absent.

Blood chemistry, C-reactive protein, urine examination, ASO titres were within normal limits. Viral markers were negative. Consent for skin biopsy was not given by her parents. The clinical diagnosis of UME was made and oral antihistamines and emollients were prescribed. The rash subsided within four weeks of its onset.

Discussion

In 1962, Brunner et al. (6) described a distinct eruptive disorder with unilateral periflexural distribution, under the heading “A new papular exanthem of childhood”. Similar exanthem was called as “Localised erythema with regional lymphadenopathy” by Taieb et al. in 1986, which they renamed as APEC (Asymmetric periflexural exanthem of childhood) in 1993. (7). In 1992, Bodeman and deProst proposed the term “unilateral laterothoracic exanthem (ULM)”, based on the most common site of this rash (8). Since then, numerous cases of ULM/ APEC have been described in Western literature, mainly from France and Italy (1). However, its variant, unilateral mediothoracic exanthem (UME), is a rare disease, with only a couple cases reported from India (5).

Unilateral exanthem usually occurs in Caucasian children of age group 1-5 years, with male to female ratio 1:2. There is a seasonal predilection from February to September. Human transmission is not noted. Al-

© 2019 The Serbian Association of Dermatovenereologists

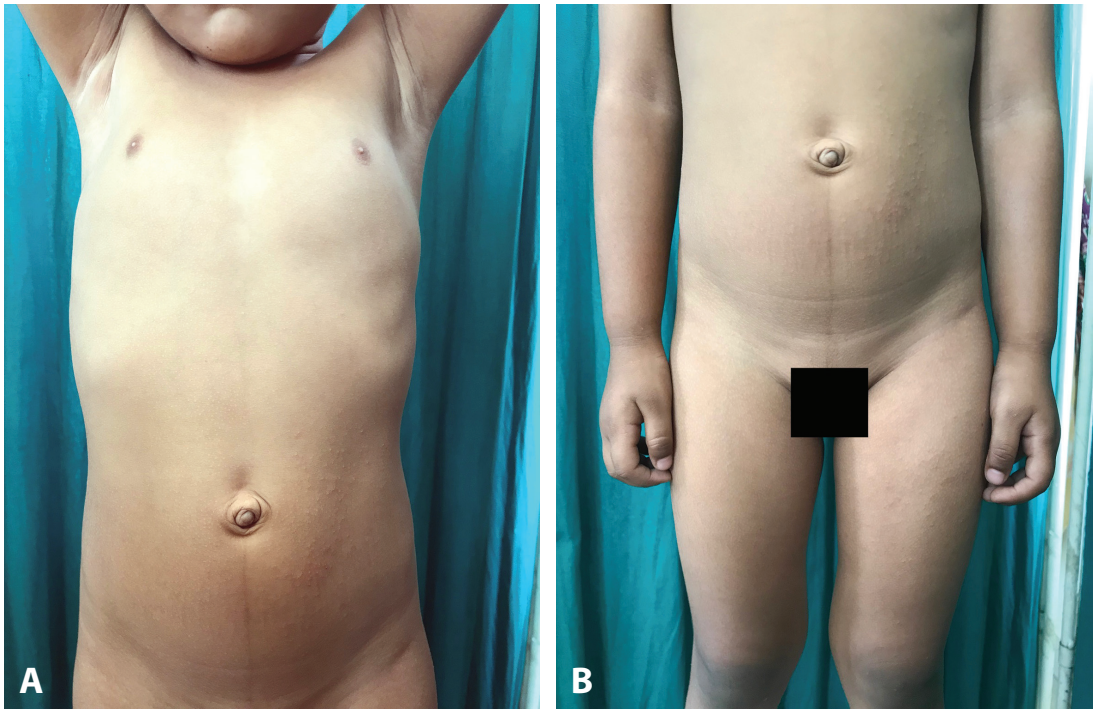

Figure 2. A). Unilateral mediothoracic exanthem on the left half of the trunk without axillary involvement. B). Unilateral exanthem extending to the left thigh without involving groin

though several authors have hypothesised the role of viral aetiology, inoculation with *spiroplasma species*, or a link to pityriasis rosea in the pathogenesis of this exanthem, the real aetiology remains unproven (1). The unilateral predominance of the eruption may be explained by a post zygotic mutation at an early stage of embryogenesis, which renders the keratinocytes of one side of body more reactive to infective agents (9).

During the evaluation of the present cases, other differential diagnoses were ruled out on the basis of history and clinical examination. Herpes zoster was unlikely as there were no vesicles throughout the course of the disease and the rash was not painful. Atypical pityriasis rosea and Gianotti Crosti syndrome were unlikely. Gianotti Crosti syndrome causes a brownish-red rash that is symmetrical and typically involves the cheeks, buttocks, forearms, and legs. It generally does not affect the trunk. Irritant and allergic contact dermatitis were ruled out as detailed history did not reveal any potential irritants or allergens. History also excluded drug related rash, including over-the-counter topical or systemic drugs, and topical and systemic remedies.

Histopathology of unilateral exanthem reveals mild to moderate mononuclear interface dermatitis and mononuclear infiltrate in dermis, with clear predominance around sweat glands (perisudoral infiltrate) (10). This is quite typical of unilateral exanthem, and not seen in any of the above differential diagnoses. In our cases, skin biopsies were not performed due to lack of parents' consent.

Our case reports warrant attention due to paucity of cases of unilateral exanthem in Indian literature. Lack of reports also suggests possibilities of misdiagnosis and underreporting by Indian clinicians. Inability to clinically diagnose unilateral exanthem leads to exhaustive, expensive and invasive workup. Therefore, we urge the dermatologists to become aware of this self-limiting entity in order to avoid a wrong diagnosis and unnecessary investigations.

References

1. Coustou D, Leaute-Labreze C, Bioulac-Sage P, Labbe L, Taieb A. Asymmetric periflexural exanthem of childhood: a clinical, pathologic, and epidemiologic prospective study. Arch Dermatol. 1999;135(7):799-803.

© 2019 The Serbian Association of Dermatovenereologists

2. Zawar VP. Asymmetric periflexural exanthema: a report in an adult patient. Indian J Dermatol Venereol Leprol. 2003;69(6):401-4.
3. Chuh AA, Chan HH. Unilateral mediothoracic exanthem – a variant of unilateral laterothoracic exanthem. Cutis. 2006;77(1):29-32.
4. Arun B, Salim A. Transient linear eruption: asymmetric periflexural exanthem or blaschkitis. Pediatr Dermatol. 2010;27(3):301-2.
5. George A, Chaudhary PR, Kanish B. Unilateral exanthem. Indian J Dermatol. 2015;60(3):311.
6. Brunner MJ, Rubin L, Dunlap F. A new papular erythema of childhood. Arch Dermatol. 1962;85:539-40.
7. Taieb A, Megraud F, Legrain V, Mortureux P, Maleville J. Asymmetric periflexural exanthem of childhood. J Am Acad Dermatol. 1993;29(3):391-3.
8. Bodemer C, de Prost Y. Unilateral laterothoracic exanthem in children: a new disease? J Am Acad Dermatol. 1992;27(5 Pt 1):693-6.
9. Leung AK, Barankin B. Unilateral laterothoracic exanthem. J Pediatr. 2015;167(3):775.
10. McCuaig CC, Russo P, Powell J, Pedneault L, Lebel P, Marcoux D. Unilateral laterothoracic exanthem. A clinicopathologic study of forty-eight patients. J Am Acad Dermatol. 1996;34(6):979-84.

Unilateralni mediotorakalni egzantem: prikaz dva slučaja indijskih devojčica

Sažetak

Unilateralni mediotorakalni egzantem je jedna varijanta čestog entiteta u zapadnjačkoj literaturi, naime asimetričnog perifleksuralnog egzantema u detinjstvu. U svetu je opisano manje od deset slučajeva unilateralnog mediotorakalnog egzantema, a samo dva su objavljena u indijskoj literaturi. Ovde prikazujemo dva slučaja unilateralnog mediotorakalnog egzantema u detinjstvu iz Indije. Dermatolozi moraju biti svesni ovog oboljenja kako bi izbegliinvazivna i isrpljujuća ispitivanja i pogrešnu dijagnozu

Ključne reči: Egzantem; Kožne bolesti; Dete; Torakalni zid; Dijagnoza; Ishod terapije; Indija; Histaminski antagonisti

Received 12.07.2019.
Accepted 25.07.2019.

© 2019 The Serbian Association of Dermatovenereologists