

Linear Verruciform Xanthoma of Lower Limb

Dear Editor,

A 23-year-old female presented to our out patient department (OPD) with a moderately itchy, linearly arranged verrucous plaque extending from left great toe to the knee since early childhood. Two years ago, she developed a verrucous growth medial to the existing linear verrucous plaque. There was no significant family history.

On clinical examination, there were multiple yellow to skin-colored verrucous papules coalescing to form a well-defined linear plaque, extending from the medial side of the left great toe to the lateral border of the knee with surface crusting and surrounding inflammation. A similar plaque was seen medial to it as shown in Figure 1.

On dermoscopy with DermLite DL4, contact polarized view at 10× magnification, we could appreciate, white structureless areas, pigment globules, red dotted vessels in a patchy pattern, and yellow areas as shown in Figure 2.

After removing the superficial crusts, a 4 mm punch biopsy was done from the plaque, which showed an exophytic growth with significant papillomatosis, changes of crusting in stratum corneum, acanthosis, and infiltration of papillary dermis with foamy macrophages^[1] as shown in Figure 3.

Based on the clinical, dermoscopic, and histopathological correlation, the diagnosis of verruciform xanthoma (VX) was made.

There were no limb defects or bony abnormalities on clinical and radiological examination.

As VX is known to be associated with inflammatory linear verrucous epidermal nevus (ILVEN)^[1] and as the clinical picture of our patient is similar to ILVEN, three more punch biopsies were taken from multiple sites of the linear verrucous plaques. All of them were consistent with VX, and a diagnosis of linear VX was made.

Though this case can be considered as VX of the lower extremity, the clinical picture of linear plaque, and the history since childhood is in favor of VX arising in a background of ILVEN (Linear Verruciform xanthoma).

The patient was counseled regarding the benign nature of the disease.

VX was first described by Shafer in 1971 as an oral entity.^[2] It is an uncommon lesion with an incidence of 0.025%-0.05% of all the pathology cases and is usually misdiagnosed clinically as papilloma.^[1] Extraoral cases tend to involve the scrotum or anogenital mucosa. Extramucosal cases are rarely described in the literature^[1] and are seen in association with lymphedema,^[2-4] epidermolysis



Figure 1: A well-defined linear plaque extending from medial aspect of left great toe to lateral aspect of left knee with surface showing crusting, and a similar plaque medial to it

bullosa,^[3,4] pemphigus,^[3] discoid lupus erythematosus,^[3,4] epidermal nevi and graft-versus-host disease,^[3] as well as within cutaneous lesions of the X-linked dominant disorder CHILD syndrome.^[4] Other associations include squamous cell carcinoma,^[1] lichen planus,^[5] bone marrow transplantation, and other immunocompromised states.

The etiology and pathogenesis remain unknown. Histopathological findings are pathognomonic of these lesions. These foam cells stain positively with CD68 antibody (a macrophage marker) suggesting origin from monocyte/macrophage lineage.^[1] Excision is the treatment of choice^[1] and is rarely followed by recurrence. However, no potential for malignancy has been reported till date.^[1]

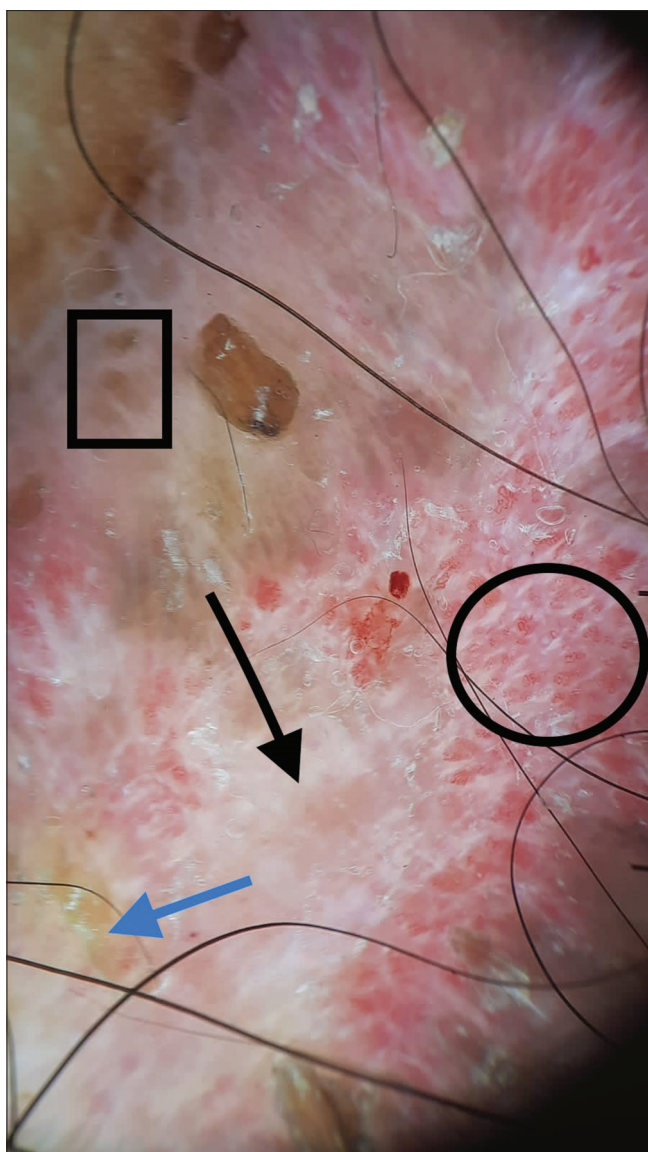


Figure 2: Dermoscopy (Dermlite™ DL4, 3Gen Inc; 10× magnification; polarized mode) of the lesions after removal of crusts shows white structureless areas (black arrow), pigment globules (black square), dotted vessels in a patchy pattern (black circle) and yellow areas (blue arrow)

We report this case to increase awareness regarding this rather rare but distinctive condition that should be considered in the differential diagnoses of linear lesions.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The

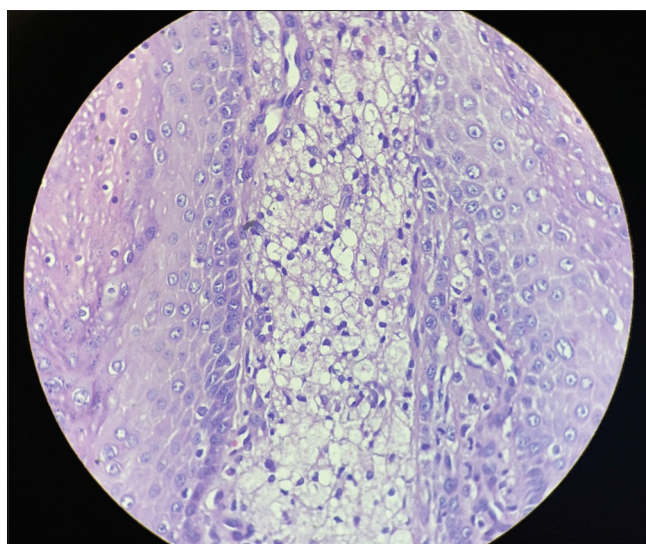


Figure 3: Numerous foamy macrophages are noted within the papillary dermis, (H & E, 40x)

patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

**Penmetsa Swathi, Patnala Guruprasad,
Sudhir B. Karri¹, Kethireddi S. Divya**

*Department of D.V.L., Andhra Medical College, ¹Consultant Pathologist,
Vijaya Diagnostic Centre, Visakhapatnam, Andhra Pradesh, India*

Address for correspondence:

*Dr. Kethireddi S. Divya,
Department of D.V.L., Andhra Medical College,
Visakhapatnam - 500 020, Andhra Pradesh, India.
E-mail: drksdivya89@gmail.com*


References

1. Singh S, Khandpur S, Agarwal S. Verruciform xanthoma overlying inflammatory linear verrucous epidermal nevus and in broad segmental distribution. *BMJ Case Rep* 2018;:bcr2018225964. doi: 10.1136/bcr-2018-225964.
2. Scully C. *Oral and Maxillofacial Medicine*. 3rd edn. Edinburgh: Churchill Livingstone, 2013.
3. William trent messengale. xanthoma. in: Jean l.bolognia, Julie v.schaffer, lorenzo cerroni (eds). *dermatology*. fourth edition. usa: Elsevier; 2018.p1634-43.
4. Vasanth sathiyakumar, steven R.jones, Seth s.martin. Xanthomas

and lipoprotein disorder-in: Sewonkang, masayuki amagai, anna L bruckner, Alexander H.euk, david j.margolis, amy j.mcmichael, jeffrey s.orringer(eds). Fitzpatrick's dermatology. Ninth edition. Uk: Mc graw hill education; 2019.p2273-91.

5. Li Z, Wang Y. Verruciform xanthoma of the thumb. Indian J Dermatol Venereol Leprol 2018;84:67-9.

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

Access this article online	
Website: https://journals.lww.com/idoj	Quick Response Code 
DOI: 10.4103/idoj.idoj_630_22	

How to cite this article: Swathi P, Guruprasad P, Karri SB, Divya KS. Linear verruciform xanthoma of lower limb. Indian Dermatol Online J 2023;14:876-8.

Received: 21-Nov-2022. **Revised:** 03-Jan-2023.

Accepted: 07-Jan-2023. **Published:** 05-Oct-2023.

© 2023 Indian Dermatology Online Journal | Published by Wolters Kluwer - Medknow