DOI: 10.7860/JCDR/2023/66190.18768

Lingua Plicata: A Precursor of Pemphigus Vegetans

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Keywords: Cerebriform tongue, Fissured tongue, Oral mucosal lesions

A 45-year-old female presented to the Dermatology outpatient department with complaints of painful oral erosions on the lips and a burning sensation on the tongue for two weeks. Oral cavity examination showed a tongue with multiple fissures (resembling sulci) with a few erosions on the lips. Regarding the erosions on the lips, cheilitis and nutritional deficiencies were considered. She was treated conservatively with multivitamin supplements, topical analgesics, and emollients. Complete dermatological examination was normal, and she was kept under regular monthly follow-up. Four months after the onset of oral lesions, she presented with raw areas over the trunk, bilateral upper and lower limbs, raised lesions over the bilateral axillae, and gluteal region [Table/Fig-1a]. On examination, vegetative plaques with oozing and crusting were present over the bilateral axillae and gluteal region. Multiple erosions were present on the trunk, groin, bilateral upper and lower limbs. The classical oral presentation of a fissured tongue [Table/Fig-1b] with cutaneous erosions and vegetative plaques pointed towards a provisional diagnosis of pemphigus vegetans with drug reaction and other bullous dermatoses like pemphigus vulgaris, pemphigus foliaceous, bullous pemphigoid, and paraneoplastic pemphigus as differentials. A skin biopsy was done and sent for histopathological examination, which showed suprabasal clefts containing abundant eosinophils and acantholytic cells [Table/Fig-1c]. Based on the clinical findings of vegetative plaques and cerebriform tongue (Premalatha's Sign) [1], along with the histopathological findings, the diagnosis of pemphigus vegetans was confirmed.

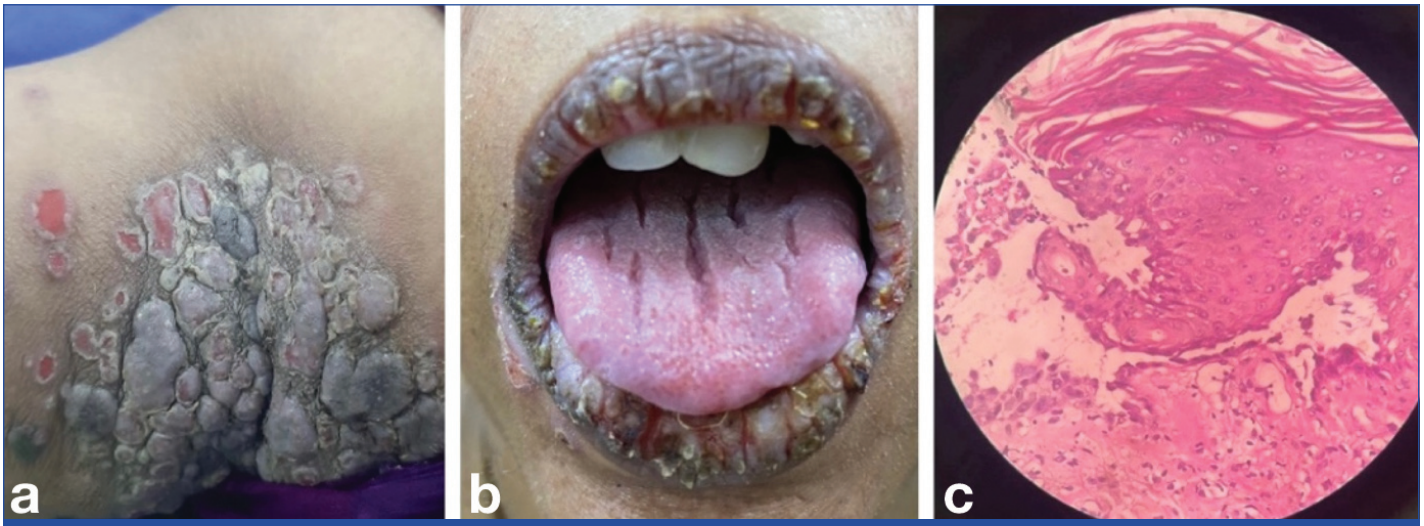
The tongue can serve as an indicator of systemic disease and, as such, can herald the onset of several systemic and cutaneous diseases. A benign disorder known as lingua plicata (also referred to as scrotal tongue, fissured tongue, furrowed tongue, plicated tongue, cerebriform tongue, or grooved tongue) affects the tongue, with deep fissures appearing on the lateral and dorsal surfaces as its defining feature. Fissured tongue usually remains asymptomatic, but in certain cases, it can be associated with a constant burning sensation that prompts the patient to seek medical attention. While the exact aetiology of the condition is unknown, there is evidence that lingua plicata may be an inherited condition and may represent a polygenic trait or an autosomal-dominant trait with partial penetrance [2].

The most frequently observed fissuring pattern in the above-mentioned study was the central longitudinal pattern, while the lateral longitudinal pattern was infrequently observed [3].

There are various disorders associated with lingua plicata, including pemphigus vegetans, pustular psoriasis, geographic tongue, Melkersson-Rosenthal syndrome, Sjogren's syndrome, Down syndrome, chronic granulomatous disease, pachyonychia congenita, Cowden syndrome, and acromegaly, to name a few. A study by Vörös-Balog T et al., showed evidence of the coexistence of diabetes mellitus and hypertension with fissured tongue, while studies by Jahanbani J et al., and Ebrahimi H et al., suggested a relationship between fissured tongue and geographic tongue [4-6].

While there is no effective treatment for the complete clearance of fissured tongue, implementing general measures such as avoiding hot and acidic foods, chewing betel nuts, smoking, and using tobacco can provide symptomatic relief. Utilising a specific tongue scraper and practicing good dental hygiene may also help prevent issues [7].

Pemphigus vegetans belongs to a group of immunobullous disorders and is characterised by vegetative plaques over the flexures and involvement of the oral mucosa. This variant of pemphigus vulgaris accounts for about 1 to 2% of all cases of pemphigus [8]. The two major subtypes are the Hallopeau and Neumann types, and of the two, the Neumann type is considered a severe form and



[Table/Fig-1]: a) Vegetative plaques over the axilla; b) Fissured tongue with erosions over the lips; c) High power view (H&E 40x) of section of the skin showing a suprabasal split with acantholytic cells and eosinophils.

more commonly associated with oral mucosal involvement, such as the cerebriform tongue described by Premalatha S et al., [1]. Premalatha S et al., reported that this eponymous sign can precede, occur simultaneously, or follow the cutaneous lesions of pemphigus vegetans and is usually refractory to treatment [1].

The present paper aimed to shed light on oral manifestations such as fissured tongue, which can herald the onset of several cutaneous and systemic conditions, as seen in the current case where oral lesions manifested months before the onset of the cutaneous lesions of pemphigus vegetans.

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PLAGIARISM CHECKING METHODS: [Jain H et al.]
- Plagiarism X-checker: Jun 24, 2023
- Manual Googling: Aug 30, 2023
- iThenticate Software: Oct 05, 2023 (5%)

ETYMOLOGY: Author Origin

EMENDATIONS: 6

AUTHOR DECLARATION:
- Financial or Other Competing Interests: None
- Was informed consent obtained from the subjects involved in the study? NA
- For any images presented appropriate consent has been obtained from the subjects. Yes

Date of Submission: **Jun 22, 2023**
Date of Peer Review: **Aug 26, 2023**
Date of Acceptance: **Oct 07, 2023**
Date of Publishing: **Dec 01, 2023**