Zosteriform lymphangitis carcinomatosis: A dermatologist's enigma

Dear Editor,

A 46-year-old man presented with tender well-defined reddish plaques over the neck and upper back for the past 3-months. His medical history was unremarkable, except for a history of mucoepidermoid carcinoma of the left parotid gland. He had undergone left radical parotidectomy with pectoralis major myo-cutaneous flap reconstruction of the neck along with chemotherapy and radiotherapy 3 years back. The patient was declared free of malignancy after the treatment.

On examination, there was a well-demarcated, indurated, erythematous plaque extending from the posterior to the front of the neck on the left side and the upper back [Figure 1a]. It also involved the left side of the cheek and ear with overlying haemorrhagic and translucent papulovesicular lesions and erosions in the root of the neck [Figure 1b]. Tzanck smear did not show any multinucleated giant cells. Histopathology of the lesion showed keratinised stratified squamous epithelium. The dermis showed tumour cells arranged in an acinar pattern, cord, nest and comedo pattern [Figure 2a]. The tumour cells were round to oval with moderate cytoplasm and hyperchromatic nuclei [Figure 2b]. This suggested metastatic invasion from the salivary gland as the primary site. On immunohistochemical staining, tumour cells were strongly and diffusely positive for Androgen receptor (AR) [Figure 3a] and epithelial membrane antigen (EMA) [Figure 3b] and negative for estrogen receptor (ER) [Figure 3c]. On further investigation, a positron emission tomography (PET) scan revealed recurrence of malignancy in the parotid gland. A diagnosis of cutaneous zosteriform lymphangitis carcinomatosis was made based on the clinical, histopathological and imaging features.

Figure 1a: Shows well-demarcated, indurated erythematous plaque with papulovesicles and few erosions over the neck and upper back.

Figure 1b: Shows well-demarcated,indurated erythematous plaque with papulovesicles and few erosions over neck, cheek and ear.

How to cite this article: Singh BSTP, Nayak MK, Biswal R, Singh S, Biswal A. Zosteriform lymphangitis carcinomatosis: A dermatologist's enigma. Indian J Dermatol Venereol Leprol. doi: 10.25259/IJDVL_417_2023

Received: April, 2023 **Accepted:** November, 2023 **EPub Ahead of Print:** April, 2024

DOI: 10.25259/IJDVL_417_2023

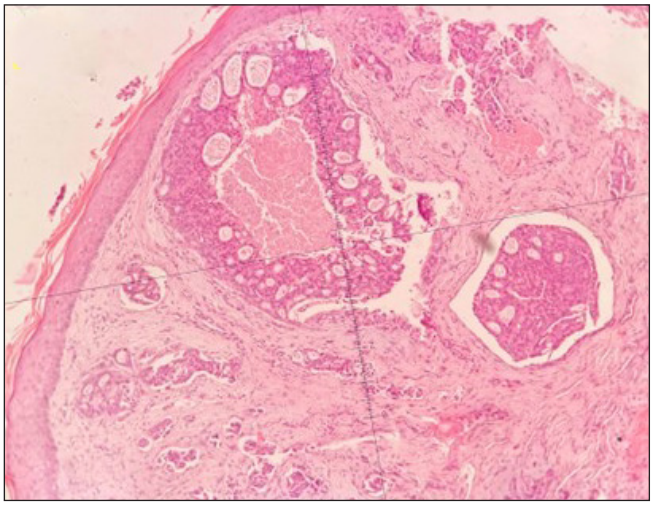

Figure 2a: Histopathology showing the source of tumour cells was discovered in later nests inside lymphatic channels. (Haematoxylin and Eosin, 100x).

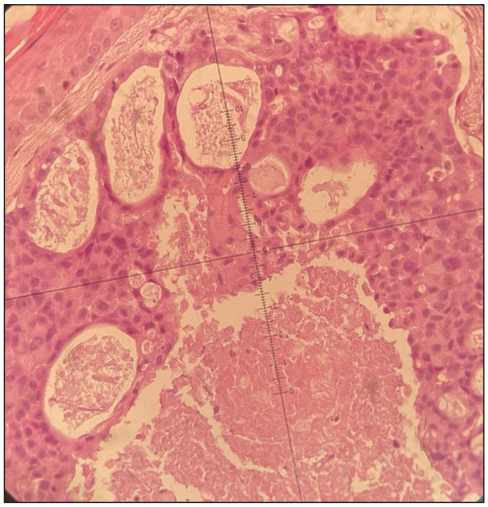

Figure 2b: Histopathology showing tumour cell nests with hyperchromatic nuclei with a high N:C ratio inside lymphatic channels (Haematoxylin and Eosin, 400x).

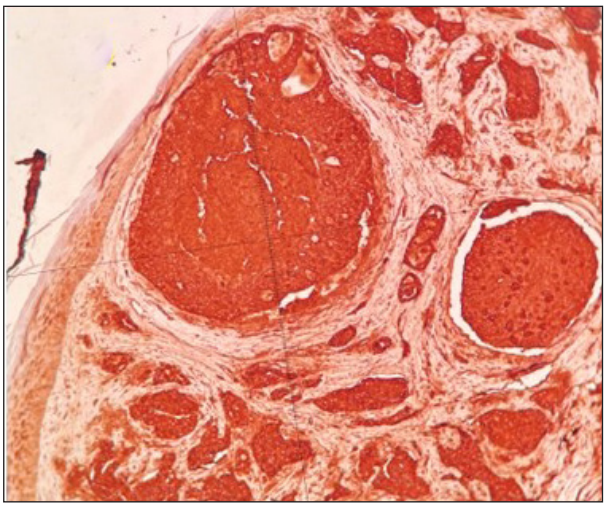

Figure 3a: Show immunohistochemistry results for androgen receptor (AR).

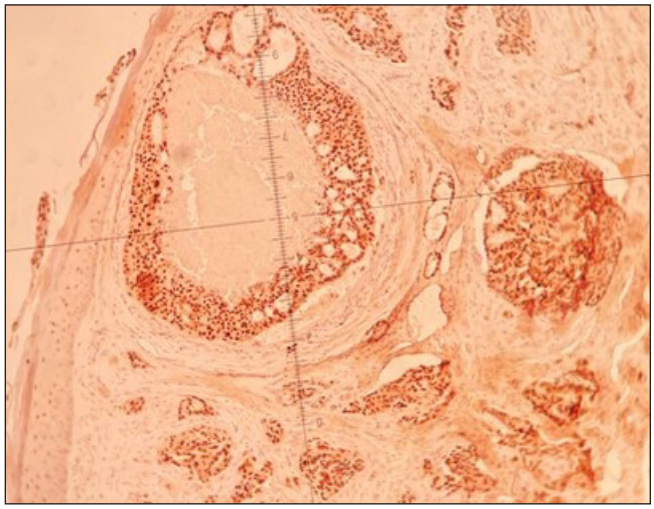

Figure 3b: Show immunohistochemistry results for epithelial membrane antigen (EMA).

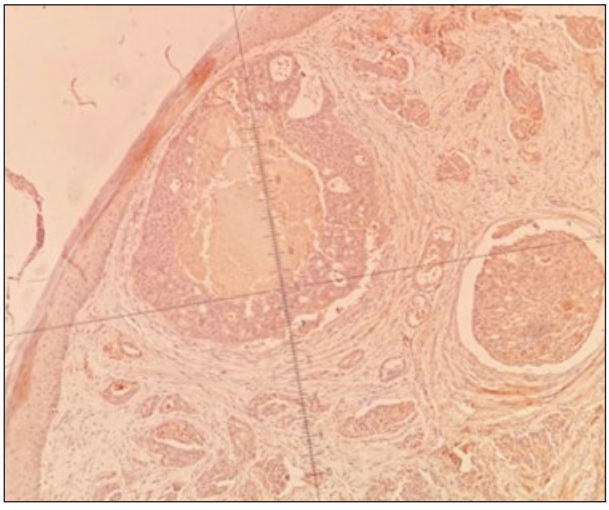

Figure 3c: Show immunohistochemistry results for estrogen receptor (ER).

Cutaneous metastases occur only in 0.7–0.9% of cancer patients.[1] Cutaneous lymphangitis carcinomatosis is a rare presentation of the metastatic spread of cancer to the skin. It is caused by the occlusion of lymphatic channels of the dermis by neoplastic cells.[2] Clinically, it presents as erythematous tender plaques closely resembling lymphangioma circumscriptum, erysipelas, cellulitis, herpes zoster, irritant contact dermatitis or post-irradiation lymphangiectasia. The clinical features may be diagnostic but may mimic several infectious and benign dermatoses. The most common clinical mimicker of this condition would be radiotherapy-induced acquired lymphangiectasia which is a vesicular dilation of the lymphatic channels. The vesicles appear on the normal skin in acquired lymphangiectasia, in contrast to lymphangitic carcinomatosis, where papulovesicles appear on infiltrated, erythematous skin. There was no temporal association of the eruption with the institution of chemotherapy, thereby ruling out the radiation recall phenomenon.

Table 1: Reported cases of lymphangitis carcinomatosis with its features

Author	Presentation	Type of malignancy	Immunohistochemistry	Primary
Hari Kishan *et al*. 2013[1]	Confluent papules, nodules, and on the left side of the neck in a zosteriform distribution	SCC	Not Done	Palate
Lola Prat *et al.* 2013[2]	Erythematous, eczematous, itchy rash localised on the right anterior chest and left shoulder	Adenocarcinoma	Positive for cytokeratin 7 (CK7+) and thyroid transcription factor-1 (TTF-1+) and negative for cytokeratin 20 (CK20-).	Lung
Echeverria-Garcia *et al*. 2013[3]	Erythematous indurated plaque on the right side of neck and jaw, mimicking erysipelas	Lymphoepithelial carcinoma of the parotid	Positive for cytokeratin, lymphocytic expression of CD45	Parotid
Shamsadini *et al*. 2003[4]	Nodules, ulcers, vesicles, inflammatory areas, sclerotic areas, lesions on the left side of the neck	SCC	Not Done	Larynx
Schmitt *et al*.[6] 2011	Violaceous plaque in a collar like distribution over the right side of the neck and trunk	Adenocarcinoma	Positive for TTF-1 and CK7	No primary identified even with exhaustive imaging
Bishnoi A *et al*. 2019[5]	Ill-defined, indurated, erythematous plaque encircling the right side of the patient's neck.	Adenocarcinoma	Positive for CK7 and negative for p63, CK20, PSA, GAT A3 and TTF-1	Pyriform fossa

SCC: squamous cell carcinoma,

Lymphangitic carcinomatosis has been previously reported with mostly the lung and the breast as the site of primary malignancy.[1] A few reported cases of carcinomatosis lymphangitis presenting over the neck have been tabulated in Table 1. These cases presented with metastases from the palate, lungs, pyriform fossa, larynx and the parotid; the primary was unknown in one case.[1–5] Bishnoi *et al*. proposed the term 'carcinoma en gorget' to describe the malignant infiltration of the skin over the neck arising from different malignancies similar to 'carcinoma en cuirasse' for infiltration of the chest wall.[5]

This case is presented to highlight the occurrence of rare zosteriform lymphangitis carcinomatosis metastasizing from the parotid gland. Also, lymphangitis carcinomatosis may be the first clinical marker of recurrence of malignancy after surgical resection as was seen in this case.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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