

A Very Rare Lesion of the Nipple with Preliminary Diagnosis of Malignancy: Verruciform Xanthoma

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Abstract

Verruciform xanthoma (VX), a significantly rare benign skin lesion, is often confused with other malignant and benign lesions due to its macroscopic clinical appearance. A 35-year-old female patient was admitted to the general surgery clinic with a VX lesion on her nipple. The lesion was subject to excision due to suspected malignancy. As far as we are aware, this is the only case in which VX was found on the nipple of a patient. Multiple skin lesions with similar macroscopic appearances were accompanied by a clinical history of lactation, mastitis and breast lesion. We aimed to discuss this unique case due to its rarity, aetiology and clinical location history in light of the literature.

Keywords: The nipple, verruciform xanthoma, Papet's disease, verruca vulgaris, skin malignancies

INTRODUCTION

Verruciform xanthoma (VX) is a benign, rare tumour of unknown aetiology usually found in the oral mucosa.¹⁻³ Most of the extra-oral lesions are located in the genital area.³ It is mentioned in the literature as occurring in mucosal areas, such as the oesophagus, larynx, glottis, nose and lips and the extremities, such as the hands, feet and legs.^{2,4} A single case with a breast location was described as a lesion of cystic structure on the retro-areolar region of the breast of one patient.⁴ As for the aetiology, immunological factors, long-term local irritation and viral agents have been suspected, but no definitive cause has been identified.^{2,3,5} The microscopic appearance of VX is typical and diagnostic. Excision is the most appropriate method for its diagnosis and treatment. In haematoxylin sections of the dermis, hypertrophic epidermis, irregular acanthosis in rete ridges and lipid-rich macrophages (xanthomas) are typical.

CASE PRESENTATION

A 35-year-old woman, who had given birth eight months previous to her clinical admission and was still breastfeeding, applied to the general

surgery polyclinic with complaints of pain, tenderness and swelling in the upper external quadrant of her right breast. No ultra-sonographic findings were detected in either breast. The case was diagnosed with a breast infection due to lactation, and drug treatment was given. After four months, the patient was admitted to the polyclinic with a mass in her right breast. Additionally, a papillomatous lesion of about 3 mm in diameter was detected on the nipple. According to the information provided on the pathology requisition form, the papillomatous lesion on the nipple was excised with suspicion of Papet's disease, nipple adenoma, and skin malignancies. On macroscopic examination, skin with an irregular surface measuring 0.6x0.5x0.3 cm was observed. In haematoxylin sections, verrucous hyperplasia of epidermis, acanthosis, occasionally parakeratosis and neutrophil leukocyte infiltration were observed in some areas (Figure 1). Many foamy cytoplasmic cells which occasionally entered the epidermis and completely filled the papillary dermis and elevated capillary vessels were observed (Figure 2). P-16 and human papillomavirus were also administered immunohistochemically. No staining was observed in either marker. Accordingly, the case was diagnosed as VX. Three months after this diagnosis, the patient was admitted to the dermatology polyclinic with papillomatosis lesions

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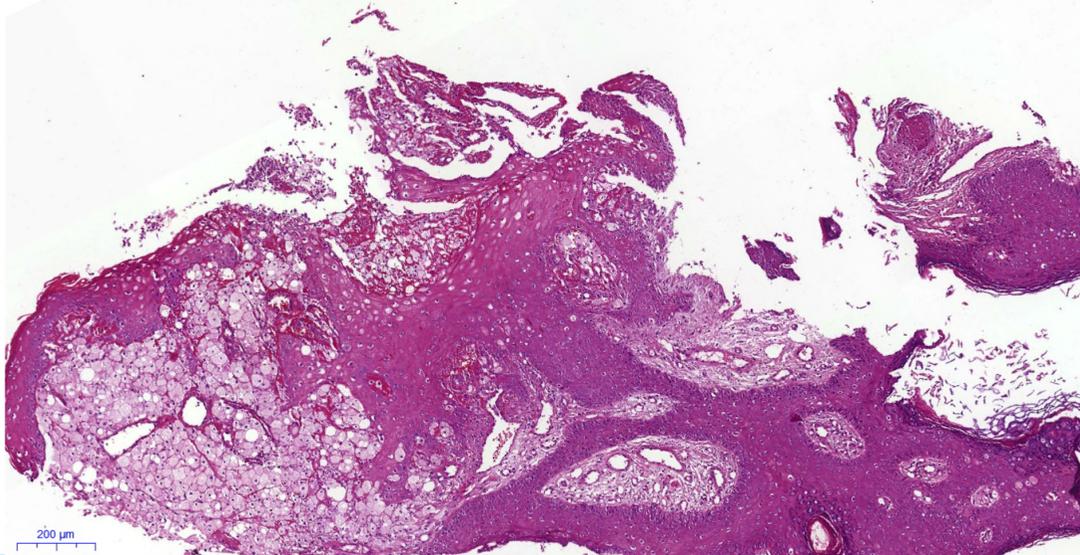


Figure 1. Verrucous hyperplasia of epidermis (H&E)

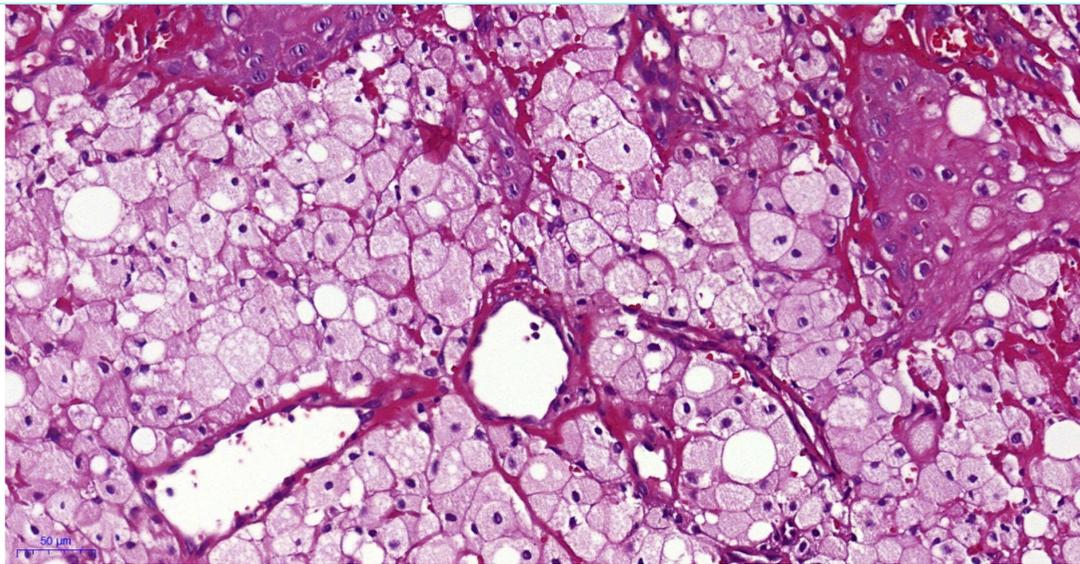


Figure 2. Lipid-rich macrophages (xanthomas) (H&E)

in the abdomen, midline of the back, lumbar region and neck. These skin lesions started about three months prior to her admission, and their numbers were increasing. In the derma-pathology polyclinic, cryotherapy was applied with the preliminary diagnosis of seborrheic keratosis and verruca vulgaris. Unfortunately, the pathology of these lesions could not be evaluated, as no samples were sent to the laboratory for examination. Informed consent was obtained.

DISCUSSION

Chronic irritation may be emphasized as one of the factors of VX aetiology.²⁻⁵ In our case, the patient was lactating, which exposed her nipple to chronic irritation. Also, four months prior to the appearance of the lesion, mastitis developed secondarily to lactation. Chronic trauma and infection are considered triggering factors in the development of this lesion. No other cases of VX located on the nipples of patients

were revealed by a literature review. Our case is the first reported VX case located on the nipple of a patient. Although some similarities exist between our case and a case reported in 2014, the morphological patterns and locations of the lesions are different.⁴ The lesion reported in 2014 was defined as a cystic VX of the breast originating from the squamous epithelium of the lactiferous ducts in the retro-areolar region,⁴ and both cases had a history of mastitis. However, the VX in our case was located on the nipple.

In both cases, chronic inflammation predisposition to VX is suggested by the presence of a mastitis history, but in our case, due to lactation, the nipple was also exposed to continuous irritation. In our case, all brown-coloured papillomatous lesions in the clinical history had the same onset. However, the lesion on the nipple was surgically removed due to suspicion of malignant lesions of the breast and skin (Paget's disease, nipple adenoma, Bowen's disease, squamous cell carcinoma

etc.). The lesions at other locations were examined in the dermatology clinic three months after the excision of the nipple. In the dermatology polyclinic, cryotherapy was applied to 20 other papillomatous lesions, but no samples were taken for pathological evaluation. Although we did not have pathological specimens of the other lesions, the lesions on the breast and other locations were concurrent and had the same macroscopic appearance. VX can be macroscopically confused with verruca vulgaris, condyloma acuminatum, seborrheic keratosis, squamous cell carcinoma, verrucous carcinoma, or Bowen's disease and biopsy materials may be sent with these preliminary clinical diagnoses.^{5,6} It is reported that VX can be diagnosed only by histopathological examination, and excision is sufficient as treatment.² Therefore, in our case, cryotherapy was considered the appropriate treatment. However, due to the absence of histopathological findings of the other lesions, we only claim that the other lesions might have been VX. There are skin lesions and syndromes observed with VX in the literature, but we could not find a case of verruca vulgaris or seborrheic keratosis skin lesions coexisting with VX.⁶⁻⁸

In the studied case, chronic trauma and inflammation are considered triggering mechanisms or critical causes of VX aetiology.²⁻⁵ VX on the nipple was observed for the first time, and malignancy was suspected based on its macroscopic appearance. We recommend considering VX in such lesions of the nipple and obtaining a biopsy for differential diagnosis. If the pathology of the other concurrent skin lesions could have been examined, more detailed and precise answers could have been obtained to some of the questions about VX. Nevertheless, in this case, the positive side was that excision or cryotherapy was sufficient for VX treatment. In our case, no other skin lesions developed during a 5-year clinical follow-up.

MAIN POINTS

- This is the first VX case reported on the nipple in the literature.
- Due to its macroscopic appearance, VX can be mistaken for nipple malignancies by clinicians.
- Microscopic appearance is typical and diagnostic.

ETHICS

Informed Consent: It was obtained.

Peer-review: Internally peer-reviewed.

Authorship Contributions

Concept: S.E., Design: S.E., Fundings: A.H.K., Materials: S.E., A.H.K., Data Collection and/or Processing: S.E., A.H.K., Analysis and/or Interpretation: S.E., Literature Search: S.E., Writing: S.E.

DISCLOSURES

Conflict of Interest: No conflict of interest was declared by the authors.

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