Verruciform Xanthoma Occurring after Surgery for Genital Paget’s Disease

Dear Editor,

We herein describe a case of verruciform xanthoma (VX) on the scrotum, which occurred after surgery for genital Paget’s disease 3 years postoperatively.

A 75-year-old man visited our hospital, complaining of genital erythematous lesions which he first noticed 3 years previously. Physical examination showed ill-defined erythemas with slight erosions on the proximal region of the penis shaft [Figure 1]. Skin biopsy revealed a number of Paget’s cells, which were positive for PAS and alcian blue, in the epidermis without invasion to the dermis [Figure 2a]. Immunohistochemistry revealed that the tumor cells were strongly positive-stained with CEA and cytokeratin 7 (CK7), while partially positive-stained with GCDFP-15 [Figure 2b-d]. The lesions were completely removed with a 1-cm margin, and split thickness skin graft from the thigh was performed. Inguinal lymph node metastasis was not observed. There was no local recurrence or metastasis during the follow-up period. However, 3 years postoperatively, a nodule appeared on the scrotum. Physical examination revealed a reddish asymptomatic, firm, well-circumscribed, slightly elevated dermal nodule on the left scrotum [Figure 3a]. The nodule was sized 6-mm in diameter, and not adhered to the subcutis. Histological examination of the excised nodule showed papillomatous acanthosis with parakeratosis and xanthomatous cells in the papillary dermis between acanthotic rete ridges [Figure 3b]. Foam cells were immunoreactive for CD68. A diagnosis of VX was made. No recurrence was observed at the 1-year follow-up period.

VX is a relatively rare benign tumor that is histologically characterized by the presence

Figure 1: Erythematous lesions with slight erosions on the genitalia

Figure 2: (a) Histological features showed Paget’s cells in the epidermis (hematoxylin-eosin stain, original magnification ×200). (b) CEA staining revealed intense expression in the epidermal Paget’s cells (×200). (c) CK7 staining revealed intense expression (×200). (d) GCDFP-15 staining revealed partial expression (×200)

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of a number of foamy histiocytes within the dermal papillae. There are several disorders in which xanthomatous changes are seen in the dermis, including plane xanthoma, xanthogranuloma, mycosis fungoides, epithelial nevi, and dystrophic epidermolysis bullosa, all of which were histologically excluded. VX most frequently occurs in the oral mucosa; however, in the dermatological field, there are a number of reports on VX arising on the genitalia, predominantly in male patients. Although the etiology is still uncertain, it has been suggested that VX is a unique epidermal reaction to a traumatic event or an inflammatory process. Repeated epidermal and/or dermal damages, chronic local minor trauma, preexisting dermatoses with increased epidermal turnover and/or degeneration, lymphedema, ultraviolet or viral etiology such as human papillomavirus, have been suggested to trigger entrapment of epithelial cells in the papillary dermis. Subsequent degeneration of these cells and lipid formation, engulfment of released lipids by macrophages cause accumulation of foam cells between the rete ridges.[1,2] A few cases of VX in relation with chronic lymphedema have been observed, in which VX occurred on the toes of lymphedematous lower extremities.[3,4] Although lymph node dissection was not carried out in the present case, the patient developed VX 6 months after undergoing surgery for genital Paget's disease, which may suggest that VX was induced by lymphedema or circulation impairment. To date, there has been only one reported case of VX occurring in a patient with genital Paget's disease,[5] which showed vulvar VX in an 80-year-old female. To the best of our knowledge, this is the first case of VX that occurred after operation of genital Paget’s disease. In our case, VX was not associated with Paget’s disease and it was not the underlying disease-causing VX. In conclusion, VX should be listed as one of the nodules that can occur after genital surgery, especially in male patients.

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 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.

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Conflicts of interest
There are no conflicts of interest.

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