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Verruciform Xanthoma Of The Penis In A Young Male Masquerading As Squamous Cell Carcinoma: Case Report

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Abstract

Verruciform xanthoma (VX) is a relatively rare benign lesion of unknown etiology with a predilection for oral mucosa. Penis is an uncommon location and to date only 17 cases of VX of penis have been reported. The lesion assumes importance as it can be easily mistaken for verrucous carcinoma or squamous cell carcinoma. We present herein a case of penile VX in a 25-year-old man who was referred to our institute with a histopathologic diagnosis of squamous cell carcinoma reported elsewhere. On microscopy the lesion demonstrated florid verruciform hyperplasia of the epidermis with hyperkeratosis, parakeratosis and acanthosis. There was a dense infiltrate of foam cells in the papillary dermis (a hallmark of VX), which were positive for CD68 and negative for S-100 protein. We present this case to highlight the significance of recognizing VX in extra oral location. An awareness of this entity is crucial to prevent misdiagnosis and halt inappropriate therapeutic intervention.

Keywords

Penis, squamous cell carcinoma, verruciform xanthoma, verrucous carcinoma

Introduction

Verruciform xanthoma (VX) is a rare benign lesion of the adults that was first described by Shafer in 1971 in the oral mucosa as a distinct pathological entity(1). Extra oral cases of VX have been reported and common locations include vulva, penis and scrotum(2,3,4,5). To our knowledge only 17 cases of penile VX have been previously reported in literature(3,4,6,7,8) since it was first described in the penis by Kramer et al(4) in 1981. Majority of the cases have been reported in white adults and usually present as verruciform, polypoid or flat lesion up to 2.0cm in diameter. Clinically, genital VX may simulate squamous papilloma, verrucous carcinoma and squamous cell carcinoma. As the name indicates VX irrespective of its location is characterized by the presence of foamy histiocytes (xanthoma cells) in the submucosa or the elongated dermal papillae between the proliferating squamous epithelial ridges. Despite its distinctive histological features, the lesion can be mistaken for several benign and malignant lesions such as condyloma acuminatum, verrucous carcinoma and squamous cell carcinoma. We present this case of penile VX in a 25-year-old man to highlight the common diagnostic pitfalls in light of information available from literature.

Case Report

A 25- year- old male was referred to our institute who underwent circumcision in another hospital for a penile lesion. The clinical characteristics during the circumcision procedure were not available. The excised specimen was sent to a local pathology laboratory where it was diagnosed as squamous cell carcinoma. In our department we received one haematoxylin and eosin stained slide and the corresponding paraffin block for review. The original slide and slides made from the block showed a lesion exhibiting acanthosis, florid
papillomatosis, hyperkeratosis and parakeratosis of the epidermis (Fig. 1A). The epidermal cells did not show any atypia or koilocytotic change. The rete ridges appeared elongated. Between the rete ridges, the papillary dermis had diffuse sheets of uninucleated foamy histiocytes (Fig. 1B). The histiocytes were confined to the papillary dermis. Immunohistochemical stains were done using monoclonal antibodies against pancytokeratin (Dako, 1:100), S-100 (Dako, 1:300), CD1a (Dako1:150) and CD68 (Dako, 1:100). The xanthomatous cells were positive for CD68 (Fig. 1C) and negative for cytokeratin, CD1a and S-100 protein. Cytokeratin however decorated the epidermal squamous cells (Fig. 1D). These histomorphologic features were diagnostic of VX. However, the patient did not come for further follow-up.

Discussion

Penile verruciform xanthoma is a relatively rare entity that might simulate a malignant lesion\(^4,6\). The precise etiology and pathogenesis is however unknown. The lesion possibly results from local irritation or viral infection\(^9,10\). VX have been reported in patients in association with HPV type 6\(^10\). However, attempts to detect the association between oral VX and HPV DNA by PCR and insitu hybridization have not been successful so far\(^11\). It has been proposed that degenerating keratinocytes may be the source of these lipid droplets in the foam cells.

VX has been described with several oral and skin diseases including oral discoid lupus erythematosus, lichen planus, epidermal nevi, leukoplakia, focal acantholytic dyskeratosis,
pemphigus vulgaris and chronic GVHD\cite{12, 13, 14, 15}. Most of these lesions exhibit basal membrane destruction and epithelial cell damage. Therefore it is likely that the presence of concomitant VX lesions in such patients is due to secondary reactive phenomenon. Majority of the cases of VX of penis has been described in adults (mean age 48 years). In the present case, the patient was a young male.

Clinically and histologically, VX may simulate several benign and malignant lesions including squamous papilloma, verruca vulgaris, condyloma acuminatum, verrucous carcinoma, Bowen disease and squamous cell carcinoma. Irrespective of the location, the morphologic hallmark of VX is the presence of xanthoma cells (foamy macrophages) in the submucosa or papillary dermis. The entities like squamous papilloma, verruca vulgaris and condyloma acuminatum do not contain foam cells in the dermal papillae. Moreover these lesions frequently exhibit koilocytic change in the epithelial cells which is known to be absent in VX. Histologically VX can be easily mistaken for a well differentiated squamous cell carcinoma especially when the pathologist is unaware of this particular entity. The present case was referred to our institute with a histologic diagnosis of squamous cell carcinoma. Squamous cell carcinoma can be distinguished from VX by its cellular atypia, invasion and lack of xanthoma cells. On a small superficial biopsy, the differentiation between VX and verrucous carcinoma may be extremely difficult. Both lesions have verrucous configuration and lack significant atypia. However, verrucous carcinoma does not show the characteristic foamy histiocytes of VX and the rete pegs appear elongated and bulbous. In one of the study VX has been reported to occur in association with squamous cell carcinoma\cite{16}. However there is no evidence that VX is a precancerous lesion. It is important that the accurate diagnosis is rendered in order not to misdiagnose VX as verrucous carcinoma or squamous cell carcinoma to avoid unnecessary excessive surgical intervention. Other conditions like histiocytosis X may have some foam cells, but these are usually a component of a polymorphic cell population that includes S-100 positive Langerhans cells. Granular cell myoblastoma may be associated with pseudoepitheliomatous hyperplasia of the overlying epithelium. But the epithelial proliferation in this condition is non-verrucous and the granular cells extend into the deep dermis which is usually S-100 positive. VX is usually treated by a conservative excision. To date only three cases of recurrence have been reported\cite{17}. Metastasis or death from the disease has not been reported for both oral and extra-oral VX.

**Conclusion**

Verruciform xanthoma is a rare benign penile lesion. Although its pathogenesis remains unclear, it is likely to be the result of inflammatory reaction rather than viral infection. Awareness of the lesion is important as it may mimic a malignant lesion leading to an overdiagnosis of squamous cell carcinoma. Recognition of this entity in extraoral location can prevent unnecessary radical surgery as simple excision is curative.
References

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