Extra-anogenital HPV16-related bowenoid papulosis

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Abstract

In this report we describe a case of bowenoid papulosis (BP) on the face of a 44 year old Algerian man. To our knowledge this is the first reported case of isolated extragenital BP. The presence of type 16 human papilloma virus was demonstrated by in situ hybridisation. This case underscores that multiple HPV-induced severe intraepithelial neoplasia, the so-called BP, is not restricted to the anogenital area.

Bowenoid papulosis (BP) is a particular form of multiple anogenital warts. In most cases lesions appear as pigmented papules and plaques but they may also form erythematous macules or leukoplakia. Histologic features are those of in situ carcinoma or severe intraepithelial neoplasia, i.e. acanthotic epidermis with loss of stratification, atypical cells, and mitosis, over an intact basement membrane. Signs of viral infection, i.e. koilocytes, are inconsistent findings.¹⁻³ The course may be chronic or regressive but, despite the threatening histologic features, malignant transformation seems rare, although this issue is not well-documented. The involvement of human papilloma virus (HPV) in these lesions has been shown by DNA hybridisation techniques. Different HPV types including 34, 35, 39, and 42 have been identified but the most common is type 16.⁴

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Figure 1 Pigmented papules on the chin.

Figure 2 Severe intraepithelial neoplasia with koilocytes.

Case report

In 1988 a 44 year old male north African Moslem consulted our department for coalescing pigmented papular lesions on his chin (fig 1). These lesions which were diagnosed as flat warts had been developing since 1986.

Clinical examination was normal. There was no history of sexually transmitted disease. No lesions were found in the genital region. Peniscopy with an acetic acid test was negative. Histological examination revealed in situ carcinoma with koilocytes (fig 2) identical to that described for BP. In situ hybridisation using sulfonated probes was performed with five separate probes (HPV6, HPV11, HPV16, HPV18, HPV33).⁵ Results were only positive for HPV16, particularly in superficial cells from the epidermis. Treatment with solid carbon dioxide followed by topical 5-fluorouracil was undertaken. The lesions regressed but the patient was lost from view. An epidemiologic study could not be done since sex partners were multiple and occasional.

Discussion

Our case was histologically and virologically typical of HPV16-induced BP in all respects except the

unusual location. BP or similar lesions may exceptionally occur outside the anogenital area. In a retrospective study, wart-like extragenital lesions have been noted in patients with BP,¹² but this finding was not confirmed by histologic or virologic study. A case of "gingival BP" has been reported but this claim was not supported by virologic evidence and clinical features were not consistent with the diagnosis.⁶ Probably the first documented case of extra-anogenital BP was reported by Lookingbill *et al*⁷ who described multiple papules on the buccal mucosa in a patient with genital BP and in situ carcinoma of the tongue. The histologic features of these lesions were compatible with BP and dot blot hybridisation of cell DNA was positive for HPV16. To our knowledge, our case represents the first time that BP has been reported in an isolated extragenital location.

HPV16 is usually detected in lesions of the anogenital area: common anogenital warts, bowenoid papulosis, Bowen's disease, or invasive cancer.⁸⁻¹¹ HPV16-related lesions have only exceptionally been reported in extragenital locations: invasive carcinomas of the fingers,¹² tongue,¹³ lung¹⁴ and larynx¹⁵ and non-invasive lesions of the buccal mucosa,⁷ thumb,¹⁶ foot¹⁷ and face (as in our case). This

underscores the fact that HPV16 is not specific for anogenital area.

Since an epidemiologic study was not performed the source of contamination was not determined in our case. The absence of associated anogenital lesions makes sexual transmission unlikely. Given the location of the lesions in the beard area, a possible explanation would be shaving with a razor that had been used by a sexually infected partner to remove pubic hair. This is a traditional practice among Moslems.

This case shows that primary development of multiple HPV-induced severe intraepithelial neoplasia, the so-called bowenoid papulosis, may occur everywhere.

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