Case Reports

Extragenital Bowenoid Papulosis Associated with Atypical Human Papillomavirus Genotypes


Abstract

Background: Bowenoid papulosis typically appears as grouped violaceous or red-brown papules in the genital or perianal regions and clinically resembles condylomata acuminata. Isolated extragenital Bowenoid papulosis is rare and has been reported in only a few case reports.

Objectives: A 51-year-old immunocompetent, healthy woman had two solitary papules on the elbow; a 41-year-old HIV-positive man had a solitary cutaneous plaque on the abdomen. No genital, periungual, or other extragenital sites of involvement were noted in either patient. The diagnosis was confirmed histologically in both cases. Lesional skin from the female patient was tested with the Digenehybrid HPV DNA assay and was positive for a mixture of low-risk HPV subtypes (6, 11, 42, 43, 44). Lesional skin from the male patient was tested with polymerase chain reaction (PCR). Consensus primers targeted for the HPV L1 region, which is a highly conserved sequence common to more than 20 HPV subtypes encoding a viral capsid protein, were used. PCR using the consensus primers was positive, but type-specific probes for HPV types 6, 11, 16, 18, 45, 31, 33, 35, and 39 were negative.

Conclusions: To our knowledge, our male patient represents the first case of isolated Bowenoid papulosis of the abdominal skin. Isolated upper-extremity Bowenoid papulosis in our female patient is also a unique case in both location and involvement of low-risk HPV types (6, 11, 42, 43, 44), which have not been previously associated with extragenital Bowenoid papulosis.

Sommaire

Antécédents: La papulose bowénoíde se manifeste généralement par l'apparition de groupes de papules voilacées ou rouge-brunâtres dans les régions génitales ou périanales ressemblant cliniquement aux condylomes acuminés. Les papuloses bowénoïdes extragénitales sont rares et n'ont été mentionnées que dans de rares exposés de cas.

Objectif: Une femme immunocomptente et en bonne santé, âgée de 51 ans, présentait deux papules solitaires sur le coude; un homme séropositif de 41 ans présentait une plaque cutanée solitaire sur l'abdomen. Chez aucun de ces deux sujets on n'a relevé la présence de sites de lésion génitaux ou périunguéaux ni autres sites extra-génitaux. Dans les deux cas, une histologie a confirmé le diagnostic. Un test d'ADN a été pratiqué sur les prélèvements de peau lésionnelle de la patiente. Ces prélèvements présentaient une combinaison de sous-types de HPV (6, 11, 42, 43, 44) à faible risque. Quant au patient mâle, ses tissus cutanés lésionnels ont été testés par réaction en chaîne de la
Bowenoid papulosis (BP) is caused by human papilloma virus (HPV) infection and tends to affect young, sexually active individuals. Most patients have multiple violaceous or red-brown papules in the genital or perianal regions that clinically resemble condylomata acuminata. The majority of cases are asymptomatic, although pruritus and tenderness may be evident. Several case reports have described isolated extragenital BP, with the lesions congregated in the cervical and mental regions. Our male patient represents the first case of isolated BP of the abdominal skin; abdominal involvement with concomitant genital/perianal involvement was described once previously. Our female patient is the first report of isolated BP of the upper extremity and the first documentation of low-risk HPV types (6, 11, 42, 43, 44) with extragenital BP.

Case One

A 41-year-old black man was seen with a complaint of a mildly pruritic plaque on the inferior abdominal skin which had been gradually enlarging for 1 1/2 years. The patient was known to be HIV positive but denied a history of any AIDS-defining illness. His CD4+ cell count was 282 mm$^3$ (normal 365–1388 mm$^3$). On physical examination there was a solitary, well-demarcated 1 x 2-cm violaceous, elliptical, verrucous plaque in the lower-left quadrant of the abdomen with some lichenification and grayish scale. The genital examination was normal. The patient denied any past medical history of condylomata or exposure to a sexual partner who had them.

A biopsy of the lesion showed an acanthotic epidermis interspersed with large, atypical keratinocytes. Hyperchromatic, pleomorphic nuclei were noted. A few multinucleated keratinocytes and atypical mitoses were also seen. Keratinocytic atypia was noted only in focal areas; a background of organized cell maturation was preserved in the epidermis. The histological picture was most consistent with bowenoid papulosis (Figs. 1 and 2).

The entire lesion was subsequently excised and sent to LabCorp Corporation for PCR analysis and typing. The degenerate consensus primer pair MY09/MY11 of the HPV L1 region was used for amplification; probes with type-specific primers were subsequently used for HPV subtyping. The highly conserved HPV L1 region was detected using the consensus primers; the viral capsid protein encoded by this region is common to more than 20 HPV subtypes. Analysis for specific subgroups 6, 11, 16, 18, 45, 31, 33, 35, and 39 was negative using specific probes.

The clinical picture of this patient was not classical for BP. Our major concern was to rule out a squamous cell carcinoma in this immunosuppressed patient. Bowenoid papulosis was convincingly diagnosed histologically. The abdominal location, without concomitant genital lesions, is the first report of isolated BP in this location.

Case Two

A 51-year-old woman complained of two 5-mm brown, slightly hyperkeratotic plaques on her right elbow (Fig.