

Bowen's disease: epidemioclinical study of 9 Tunisian cases

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Maladie de Bowen: étude épidémioclinique de 9 cas tunisiens

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R É S U M É

Prérequis : La maladie de Bowen (MB) est une forme de CE in situ, caractérisée par une évolution chronique et progressive, avec un faible potentiel de malignité.

But : Etudier les aspects épidémiologiques et cliniques de MB dans une série tunisienne.

Méthodes : Etude rétrospective de neuf cas de MB suivis au service de dermatologie de l'hôpital Habib Thameur de Tunis.

Résultats : Il y avait 7 hommes (77,8%) et 2 femmes (22,2%). L'âge moyen était de 68,8 années (46-89). Les lésions étaient solitaires dans 7 cas. La MB siégeait au visage (1 patient), au tronc (2 patients), aux membres (6 patients). Le diamètre moyen de la tumeur était de 3,4 cm. L'aspect clinique était celui d'une plaque érythémateuse bien limitée avec des bords irréguliers et une surface croûteuse ou recouverte de squames. L'examen histologique a montré dans tous les cas des kératinocytes anormaux avec des troubles de la maturation et perte de la polarité, sur toute la hauteur de l'épiderme. Le principal traitement était la chirurgie (n = 5). Seul un patient a eu une radiothérapie (cas 1). Deux patients ont été suivis au bout de 1 année et 12 ans, sans récurrence.

Conclusion : Notre série souligne les caractéristiques épidémiologiques et cliniques de MB en Tunisie à travers un échantillon restreint mais représentatif. Comme dans la littérature, cette condition a prévalu surtout chez les sujets âgés de plus de 60 ans. Dans notre étude, la MB a affecté principalement l'homme et a touché les régions non exposées dans 8 cas. Ce profil est rare dans un pays ensoleillé telle que la Tunisie, en l'absence d'autres agents étiologiques.

S U M M A R Y

Background: Bowen's disease (BD) is a form of in situ SCC, characterized by chronic and progressive course, with low potential for invasive malignancy.

Aim: To assess epidemiology and clinical features of BD in a Tunisian cohort.

Methods: A retrospective study of 9 cases of BD managed in a Tunisian dermatology department.

Results: There were 7 males (77.8%) and 2 females (22.2%). The mean age of patients was about 68.8 years (46-89). Lesions were solitary in 7 cases and occurred in various sites: face (1 patient), trunk (2 patients), limbs (6 patients). The mean diameter of the tumour was about 3.4 cm. Lesions presented clinically as an enlarging well demarcated erythematous plaque with irregular borders and crusted or scaling surface. Histological examination showed in all cases abnormal keratinocytes with disordered maturation and loss of polarity replacing the epidermis in its whole thickness. The main treatment was surgery (N=5). Only one patient had radiotherapy (case 1). Outcome was mentioned in 2 patients who remained free from recurrence respectively after a follow-up of 1 and 12 years.

Conclusion: Our series outlines epidemiological and clinical features of BD in Tunisia through a small but representative sample. As in the literature, this condition prevailed mainly over 60 years. In our study, BD occurred predominantly in men and affected non-exposed sites in 8 cases. This profile is uncommon in a sunny country in Tunisia, in the absence of other aetiological agents.

Mots-clés

Maladie de Bowen, Carcinome épidermoïde

Key- words

Bowen's disease, squamous cell carcinoma

Cutaneous squamous cell carcinoma (SCC) includes many subtypes with widely varying clinical behaviours, ranging from indolent to aggressive tumors. Bowen's disease (BD) is a form of in situ SCC, first described by Bowen in 1912 (1). It is characterized by chronic and progressive course, with low potential for invasive malignancy. We report here in 9 cases of BD managed in a tunisian dermatology department.

PATIENTS AND METHODS

All cases of BD confirmed histologically, observed in a tunisian dermatology department from 1990 to 2008 (19 years) were retrospectively included in this study. We noticed for each patient: epidemiological features, clinical and histological presentation, treatment modalities and outcome.

RESULTS

Nine patients were diagnosed to have BD with an estimated prevalence of 0.47 cases per year. BD represents 9.5 % of all SCC. Epidemioclinical features of our patients are summarized in table I.

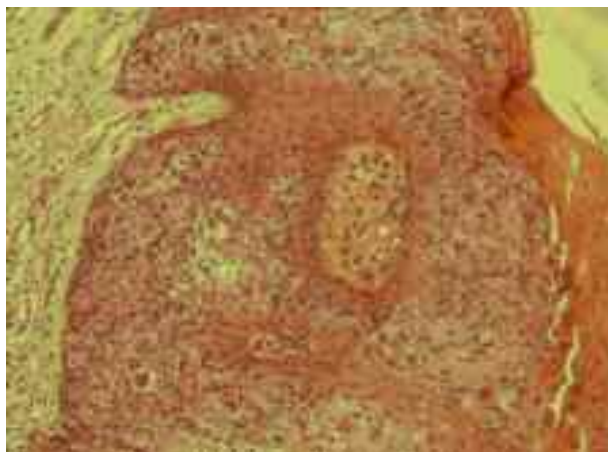
There were 7 males (77.8%) and 2 females (22.2%). The ages of patients at presentation ranged from 46 to 89 years-old (mean age: 68.8 years). The neoplasm evolved for a mean period of 14.4 months (6-36 months), being mainly asymptomatic (N=7). It was painful for 1 patient and itching for another. Lesions were solitary in 7 cases and occurred in various sites: face (N=1), trunk (N=2), limbs (N=6). The tumor presented as a 1 to 6 cm diameter (mean diameter: 3.4 cm) gradually enlarging well demarcated erythematous plaque with irregular borders and crusted or scaling surface (figure 1).

Figure 1: Erythematous plaque with irregular borders and crusted borders



Some particular features were sometimes noticed such as: ulceration (N=1), verrucous surface (N=1), pigmentation (N=2) and bubbling aspect (N=1). In 8 patients, a biopsy was first made while a primary surgical excision was performed in one case. Histological examination showed in all cases abnormal keratinocytes with disordered maturation and loss of polarity replacing the epidermis in its whole thickness. Large and atypical mitotic figures were also seen (figure 2). The main treatment was surgery (N=5). Only one patient had radiotherapy (case 1). The 3 remaining patients left before treatment (patients 2, 3 and 7). Excision was complete in 3 cases (patients 5, 8 and 9). Two patients required further surgery to obtain healthy margins (patients 4 and 6). Outcome was mentioned in 2 patients who remained free from recurrence respectively after a follow-up of 1 and 12 years. The other patients left early after treatment.

Figure 2: Hyperplasic epidermis with disorder of maturation and loss of polarity of keratinocytes with large and atypical mitotic figures.



DISCUSSION

Our series outlines epidemiological and clinical features of BD in Tunisia through a small but representative sample. As in the literature, this condition prevailed mainly over 60 years. However, it may occur at any age in adults but is rare before the age of 30 years (2-9). Symptoms are minor in the absence of ulceration (10). Lesions are usually solitary but can be multiple, as in 2 of our patients, in 10-20% of cases (3-7). Any site may be affected, but some specific sites deserve mention due to the potential for diagnostic confusion including perianal (11-13) and subungual BD (14-16). In these locations, human papilloma virus (HPV) has been implicated in the aetiology of BD. These sites were not found in our patients. BD prevails especially in women (70-85% of cases) on their lower legs (60-85%) (3, 4), and is common on the head and neck of men (5), being suggestive of a relationship with chronic solar damage (6, 8). In our study, BD occurred predominantly in men and affected non-exposed sites in 8 cases (Table 1). This profile is uncommon in a sunny country like Tunisia, in the absence of other

aetiological agents such as arsenic exposure, immunosuppression or pre-existing conditions. Clinical aspect of BD in our patients was typical. Pigmented lesion, noticed in 2 cases, is an uncommon variant, occurring in 1.7% of cases in one series (17). Verrucous BD, observed in one of our patients, is important as it is likely to raise suspicion of invasive carcinoma. In our series, there was no progression to invasive SCC. Diagnosis of BD is primarily on the basis of clinical features. In our patients, histological results were typical in all cases. According to the guidelines published by the British Association of Dermatologists (10), histological confirmation is required for cases with diagnostic doubt, or where invasive transformation is suspected.

Surgery was the main treatment in our study. It is, as in other forms of skin cancer, considered to be the gold standard in the management of BD, making possible the documentation of excisions margins. This major advantage allowed us to

complete excision in 2 patients with initially tumoral margins. This treatment modality was therefore regarded as one of the therapies with the lowest risk of recurrence during a 5-year follow-up (5). Radiotherapy is also an established treatment for BD with cure rates varying between 94% and 100% with follow-up periods of 1-5 years (5). It is a reasonable choice for anal/perianal BD, especially when other options like surgery are difficult. Other treatment modalities, such as cryotherapy, curettage, shave excision and topical 5-fluorouracil were not used in our patients.

CONCLUSION

In Tunisia, BD represents 9.5 % of all SCC. Areas with high sun exposure were not the most affected in our patients. Surgical excision remains the best treatment modality of this condition.

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