Warty and clear-cell Bowen's disease successfully treated with photodynamic treatment

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None declared.

Summary

Clear cell Bowen's disease (BD) is a rare histopathological subtype of BD, characterized by epidermal atypical keratinocytes with clear-cell changes exceeding 80% of the tumor population. Warty and clear cell Bowen's disease (WCCBD) is a recently described variant of clear cell BD, with verrucous and hyperkeratotic surface changes in addition to pathological features consisting of an extremely acanthotic epidermis, hyperkeratosis and clear-cell changes. Herein we present a 72-year-old man with a 1-year history of tumoral lesion located on the left cheek with a diagnosis of WCCBD, in whom we achieved excellent result with photodynamic treatment. To the best of our knowledge, this is the second reported case of this entity.

Warty and clear-cell Bowen's disease (WCCBD) represents a recently described proliferative form of a cutaneous squamous cell carcinoma *in situ*, characterized by a verrucous, hyperkeratotic nodule or plaque and histopathological features consisting of an extremely acanthotic epidermis, hyperkeratosis and clear-cell changes exceeding 80% of the neoplastic cell population (1). The prognosis of WCCBD is not known because of the paucity of clinical trials. Herein, we report a patient with WCCBD, in whom we achieved resolution of the lesion with an excellent cosmetical result after photodynamic treatment (PDT) and no recurrence despite a 2-year follow-up period. As far as we are aware, PDT of this rare entity has not been reported in the literature.

Case report

A 72-year-old man presented with a 1-year history of a tumoral lesion located on the left cheek. The lesion had enlarged slowly over time and and occasional bleeding occurred with minor trauma. There was a history of chronic sun exposure. On physical examination, a solitary, fragile, verrucous, hyperkeratotic nodule was noted on the left cheek (Fig. 1a). The histopathological examination of the skin-biopsy specimen revealed neoplastic epidermal proliferation with pronounced acanthosis, papillomatosis, hyperkeratosis and lymphohistiocytic infiltration of the upper dermis (Fig. 2). The neoplastic proliferation consisted of large, atypical keratinocytes with hyperchromatic and pleomorphic nuclei. A clear-cell population of atypical keratinocytes exceeded 80% of the epidermis. No dermal invasion was detected. Periodic acid-Schiff staining with or without diastase digestion was negative. A diagnosis of WCCBD was established with clinical and histopathological findings. Based on the recent reports indicating high cure rates of Bowen's disease (BD) with PDT, we planned PDT for our patient (2). Surface crusts were gently removed with a scalpel blade but no invasive curettage or tumor debulking was performed before the PDT. Twenty percent 5-aminolevulinic acid in an oil-in-water emulsion was applied under an occlusive dressing (Tegaderm; 3M Health Care, St Paul, MN, USA) for 5 h, and covered with a gauze to prevent sunlight exposure. Illumination was performed with a noncoherent red light source (wavelength of 580–740 nm, Waldmann PDT 1200, Villingen-Schwenningen, Germany), delivering 150 J/cm^2 at a light intensity of 100 mW/cm^2. Four weeks after the onset of treatment, marked regression of the lesion was achieved. A second session of PDT using the same technique resulted in clearance of the lesion with a slight postinflammatory hypopigmentation 1 month later (Fig. 1b).

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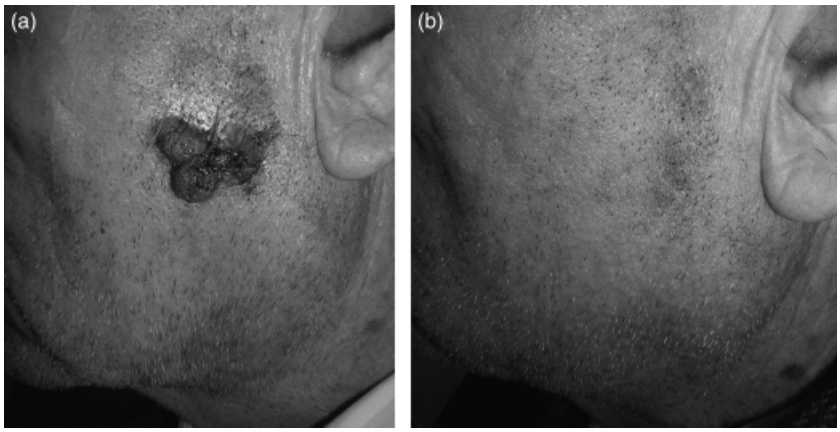

Fig. 1. (a) Verrucous, hyperkeratotic and fragile nodule on the left cheek. (b) Clearance of the lesion without scarring after two sessions of photodynamic treatment.

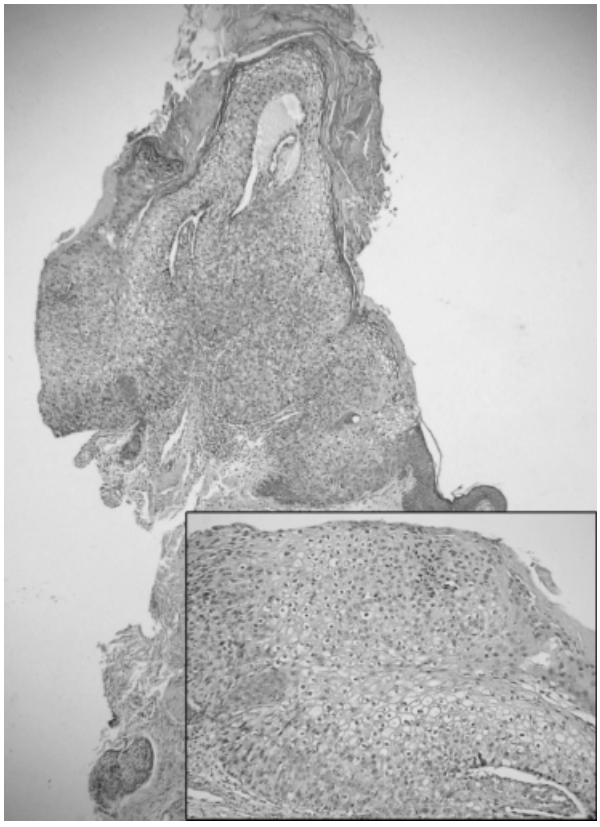

Fig. 2. Histopathological examination revealed a pronounced acanthosis, papillomatosis and hyperkeratosis with epidermal proliferation showing clear-cell changes (hematoxylin and eosin stain; original magnification, × 10); inset shows crowded nuclei through the epidermis and keratinocytic atypia with clear-cell changes.

No scarring was detected. A control punch biopsy performed at this time showed senile skin features including effacement of rete ridges and solar elastosis; however, residual neoplastic proliferation and clear-cell changes were absent. No recurrence was noted during 2 years of follow up.

Discussion

Clear-cell BD is a rare histopathological subtype of BD, characterized by epidermal atypical keratinocytes with clear-cell changes exceeding 80% of the tumor population (3). Clear-cell BD is rare, with 14 cases reported in the English literature. WCCBD is a recently described variant of clear-cell BD, with verrucous and hyperkeratotic surface changes in addition to pathological features consisting of an extremely acanthotic epidermis, hyperkeratosis and clear-cell changes (1). The case reported herein is the second reported case of WCCBD. The lesion of the previously reported patient was localized on the dorsum of the left foot in association with multiple BDs on the legs, abdomen and buttocks. By way of comparison, additional BD lesions and peripheral inflammation of the surrounding skin were not observed in the patient described herein, and the tumor was located on a sun-exposed area (i.e., on the face). The pathological differential diagnosis included sebaceous carcinoma, clear-cell porocarcinoma *in situ* and ectopic extramammary Paget's disease. Sebaceous carcinoma has a lobulated histological pattern with undifferentiated basaloid cells, which were not detected in our patient. Although intraepidermal neoplastic proliferation with clear cells and hyperchromatic and pleomorphic nuclei may also be observed in clear-cell porocarcinoma *in situ*, the absence of ductal differentiation helped us to exclude the diagnosis (4). Ectopic extramammary Paget's disease presents as epidermal neoplastic cells with a large nucleus and a pale, ample cytoplasm located on areas other than the inguinal or the axillary region, such as the face or the chest (5). The lack of a cytoplasmic bluish hue and compression of the basal cell layer was against a diagnosis of ectopic extramammary Paget's disease in this instance. The prognosis of WCCBD is unknown. The preferred treatment of the previously reported case was surgical excision, although there was no mention of a follow-up period for recurrence. The standard duration for topical application of a photosensitizing agent for the treatment of BD with PDT is 4 h. However,

application durations of as much as 8 h have been described in the literature. We favored a 5-h occlusion time to allow complete penetration of the photosensitizing agent throughout the markedly acanthotic epidermis of this rare variant of BD. The clinical evaluation of BD and additional treatments with PDT, when indicated, have been reported to be undertaken with a 4–8-week interval. At 4-week follow-up, we detected a marked regression of WCCBD. Because there was no inflammation or crusting related to previous PDT at the end of this time, another treatment session was undertaken, which resulted in clearance of the lesion by the next 4-week follow-up. We suggest that the course of WCCBD may not be any worse than classical BD, and PDT may be used as an effective treatment modality with additional advantages over surgical excision, such as a lack of scarring.

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