CORRESPONDENCE

Plaque-type warty dyskeratoma of the scalp with multiple follicular involvements

Case report

A 47-year-old woman presented with an asymptomatic brown plaque on her scalp for many years (Figure 1). The lesion was a well-demarcated, 3 × 2-cm, hyperkeratotic brown plaque with multiple yellow follicular plugs on the vertex. There was no discharge or bleeding. Oral mucosa, vulva mucosa, and nails were all normal. Laboratory examinations, including hemogram and biochemical studies, were within normal limits. Histopathological examination revealed multiple cup-shaped epidermal invaginations filled with keratin (Figure 2A). In the bottom portion, acantholysis with suprabasal clefting and villi formation was seen (Figure 2B). These cup-shaped lesions seemed to be arising from the hair follicles (Figure 2C). There were grains and corpus ronds (Figure 2D). The individual lesion appeared to be warty dyskeratoma (WD), but this case had an aggregate of multiple lesions. Laser ablation therapy was suggested, but the patient refused further treatment.

Discussion

WD is a rare benign epidermal tumor and often presents as a hyperkeratotic and umbilicated single nodule. In 1954, Helwig¹ first discovered a solitary lesion resembling Darier’s disease and named it as “isolated Darier’s disease.” Three years later, Szymbanski² first coined the term “warty dyskeratoma,” when he found seven cases of benign cutaneous tumors that were similar to Darier’s disease microscopically. In 1969, Tanay and Mehregan³ reviewed 112 cases and noted that all WDs were solitary and occurred on sun-exposed sites during middle age and were distributed on head and neck (79%) and on trunk (15%).

Although multiple WDs were rare, cases have been reported in the Japanese.⁴ Azuma and Matsukawa⁵ reported multiple WDs distributed over scalp, neck, cheek, and hand in one patient, and they suggested that these lesions derived from the hair follicle. Griffiths et al⁶ also presented two more extensive cases with approximately 25 discrete hyperkeratotic papules with central plugs on the scalp, and those lesions were proved to be WD. Subsequently, WDs of oral mucosa,⁷ vulva mucosa,⁸ and subungual⁹ area have also been reported.

Recently, plaque-form WD was first reported by Omulecki et al⁹ in a case with a plaque-like lesion on lower back. Clinically, squamous cell carcinoma was suspected, but histological features showed the coexistence of acantholysis and dyskeratosis. The lesion seems to be a new type of acanthoma related to WD because typical cup-shaped character was not seen, and the authors believe that it may be a result of a local concentration of several WDs. In our case, clinical appearance resembled seborrheic keratosis with a stuck-on plaque-type pattern; however, the presence of multiple yellow follicular plugs of our case was unusual. In addition, an aggregate of multiple cup-shaped epidermal invaginations in histopathological study was different from Omulecki’s report and was rare in medical literature.

The etiology of WD is still controversial. Some authors proposed a range of exogenous factors, but none has been substantiated. Viral origin has been suggested as well. However, according to Kaddu et al,¹⁰ 13 lesions of WD were all negative for Human papillomavirus-DNA (HPV-DNA) analysis with polymerase chain reaction. At present, follicular derivation of WD has been considered most possible, and many cases indicated close association between WD and pilosebaceous structures.⁴,⁵,¹⁰,¹¹ Kaddu et al¹⁰ retrieved 45 patients and discovered that most cases showed variable signs of differentiation toward the infundibular portion of a normal hair follicle. Griffiths et al¹¹ human keratin monoclonal antibodies HKN-6 and HKN-7, specific for human hair cortex and inner root sheath, respectively. Diallo et al¹² discovered that cytokeratin 1 (CK1) and CK10 were expressed in the suprabasal levels of the WD, whereas CK5 and CK17 were noted in the basal layer, further supporting a follicular origin. In our case, clinical appearance showed multiple yellow follicular plugs, and histologically, there was also an association between WD and hair follicle. However, WD arising from subungual area and mucosa where hair follicles are absent cannot explain the follicular hypothesis. Therefore, subungual and mucosal lesions might represent a different derivation rather than true WDs.

Figure 1 The lesion is a well-demarcated, 3 × 2-cm, brown hyperkeratotic plaque with multiple yellow plugs on the vertex. These plugs are around the hair follicles.
The clinical differential diagnosis of WD includes skin appendage tumors, seborrheic keratosis, verruca vulgaris, actinic keratosis, or other skin tumors. The characteristic pathological pattern of WD can easily differentiate it from other skin lesions. Acantholytic dyskeratosis is also noted in several diseases, such as Darier's disease, Grover's disease, and Hailey–Hailey disease. In our case, she had no family history, and a solitary plaque appearance would not favor the aforementioned diseases.

In summary, we presented with a rare plaque-type WD with multiple yellow follicular plugs, and clinically or histopathologically, our case revealed a close relationship with hair follicle, which was consistent with the presumption that WD derived from pilosebaceous units.

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