Int J Clin Exp Med 2018;11(5):5241-5244
www.ijcem.com /ISSN:1940-5901/IJCEM0066521

Case Report

Multiple warty dyskeratoma: a case report and review of the literature

Songting Wang[1*], Juan Bai[2*], Zixiang Si[1], Jianjun Qiao[1,2]

[1]Department of Dermatology, Beilun People's Hospital, The First Affiliated Hospital Beilun Branch of Zhejiang University, Ningbo, China; [2]Department of Dermatology, The First Affiliated Hospital, College of Medicine, Zhejiang University, Hanghzou 310003, Zhejiang, China. [*]Equal contributors.

Received September 30, 2017; Accepted February 8, 2018; Epub May 15, 2018; Published May 30, 2018

Abstract: Warty dyskeratoma is an uncommon entity usually characterized by a single solitary papule or nodule on the head or neck with a comedo-like plug. Multiple warty dyskeratoma is an extremely rare variant. To our knowledge, there have been only 8 cases reported in English. We report a 62-year-old Chinese male presented with a 2-year history of multiple papules on the scalp and forehead. Physical examination revealed numerous firm flesh-colored or grayish hyperkeratotic papules and nodules with an umbilicated center located on the right parietal, forehead, and right temporal region. Histopathology showed cup-shaped invagination of the epidermis with acanthosis, suprabasal clefting, zones of acantholysis, and dyskeratosis with corps ronds and corps grains extending into the papillary dermis. Multiple warty dyskeratoma should be differentiated with verruca, seborrheic keratosis, sebaceous hyperplasia, and actinic keratosis. The histopathologic differential diagnoses of warty dyskeratoma include Darier's disease, Grover's disease, Hailey-Hailey disease, acantholytic acanthoma, familial dyskeratotic comedones, acantholytic actinic keratosis, and acantholytic squamous-cell carcinoma.

Keywords: Warty dyskeratoma, multiple

Introduction

Warty dyskeratoma, first described as 'isolated Darier disease' in 1954 [1, 2], is an uncommon benign skin lesion usually characterized by a single solitary papule or nodule on the head or neck with a comedo-like plug [1]. Multiple warty dyskeratoma is an extremely rare condition that, so far as we know, has been previously reported in just a few cases [3-8]. Herein, we present a new case of multiple warty dyskeratoma on the scalp and forehead.

Case report

A 62-year-old Chinese male presented with a 2-year history of asymptomatic papules on the scalp and forehead. Recently, the number of the cutaneous lesions increased quickly. The patient had no history of systemic diseases. He had neither a drinking nor smoking history. Physical examination revealed numerous firm flesh-colored or grayish hyperkeratotic papules and nodules with an umbilicated center located on the right parietal and forehead. Brownish-black verrucous papules and nodules were found on the right temporal region (**Figure 1**). The lesions were discrete or grouped. The sizes of the lesions ranged from 0.5 to 2 cm in diameter. Dermoscopic image of a lesion on the forehead showed a pale to gray area with center depression (**Figure 2**). The histopathology of two specimens from the forehead and the right parietal lesions, respectively, were the same. The histopathological images showed cup-shaped invagination of the epidermis with acanthosis, suprabasal clefting, zones of acantholysis, and dyskeratosis with corps ronds and corps grains extending into the papillary dermis (**Figure 3**). The central crater was filled with cornified debris. A mild to moderate inflammatory infiltrate was found in the superficial dermis beneath the tumor.

Discussion

Warty dyskeratoma is a rare entity histopathologically characterized by acantholytic dyskera-

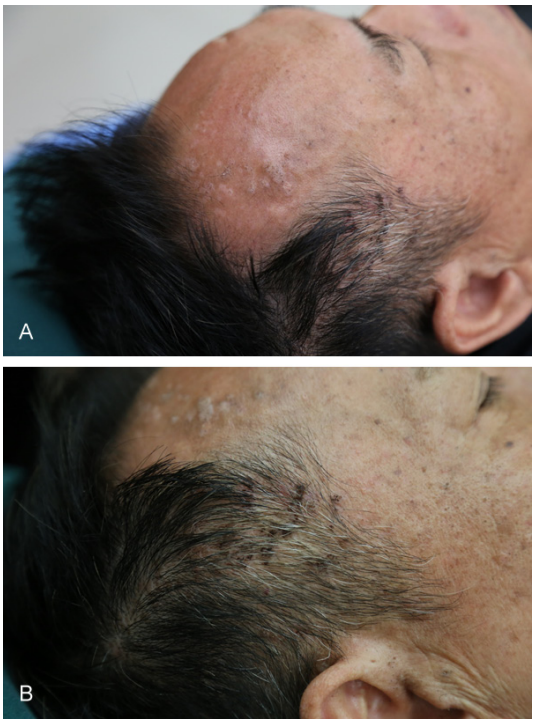

Figure 1. Clinical presentation of multiple warty dyskeratoma. A, B. Multiple hyperkeratotic papules on his right scalp and forehead.

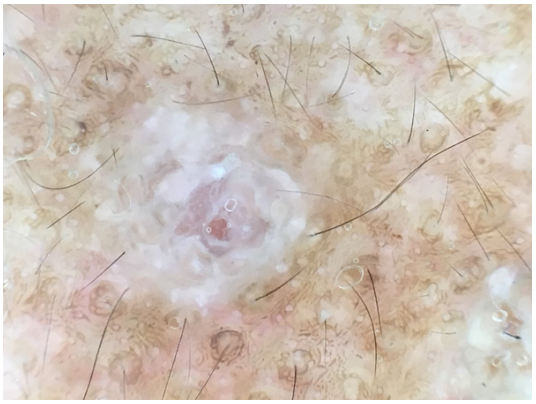

Figure 2. Dermoscopy image of multiple warty dyskeratoma. Pale to gray area with center depression.

tosis [1]. Although a male predisposition has been reported previously, while Kaddu reported a series presenting male/female (M/F) ratio is 1:1.8 [5]. The pathogenesis of warty dyskeratoma has not been elucidated. Studies have suggested that it is associated with ultraviolet light, autoimmunity, chemical carcinogens, and tobacco [8]. The most common clinical presentation is a circumscribed isolated cup-shaped papule or nodule located on the head or neck

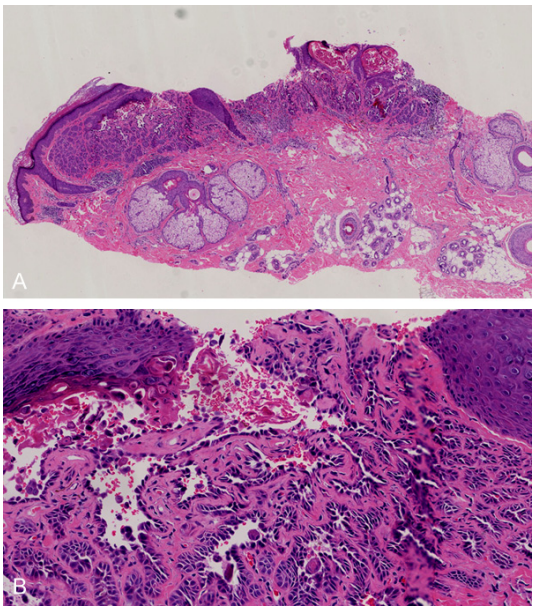

Figure 3. Histopathology of warty dyskeratoma. A. Cup-shaped invagination of epidermis and keratin-filled plug (H&E, original magnification × 25); and B. Acantholysis and dyskeratosis of the epidermis (H&E, original magnification × 200).

and in lesser frequency, on the trunk and extremities. There have been occasional reports of warty dyskeratoma arising at the mons pubis, subungual, and oral or genital mucosa [9, 10].

Multiple warty dyskeratoma is usually solitary. To date, only a few cases of multiple warty dyskeratoma have been reported. We reviewed previously reported cases and summarized them in **Table 1**. Consistent with earlier descriptions, multiple warty dyskeratoma in our study presented mostly as solitary papules or nodules located on the scalp and forehead, with histopathologically cup-shaped pattern. Multiple warty dyskeratoma presented no sexual difference (M/F ratio 1:1) and involved the scalp, face, neck, hand, or trunk. The scalp was the most commonly involved site (6/8, 75%), followed by the face (4/8, 50%). No correlation with underlying diseases were shown. Histologically, five cases showed cup-shaped invagination with acantholysis and dyskeratosis. One case showed a mixture of two kinds of histological features. The clinical differential diagnoses of multiple warty dyskeratoma are verruca, seborrheic keratosis, sebaceous cyst, and hypertrophic actinic keratosis. The histopathologic differential diagnoses are Darier's disease,

Table 1. Cases of multiple warty dyskeratoma in the literature

Author	Public year	Age (year)/ gender	Location	Clinical presentation	Number/Size, cm	Medical history	History (year)	Histological feature
Azuma Y, et al. [3]	1993	63/M	Scalp (parietal area, right tempol) Face (cheeks), neck dorsa of hands	Flesh-colored, Brownish verrucous nodules	Several/0.5-0.7	Nephrotic syndrome Pustulosis palmaris et plantaris	12	Cup-shaped
Griffiths TW, et al. [4]	1997	67/F	Scalp	Asymptomatic crusted papule with plugs	25/0.8-1	Cardiac dysrhythmia	1.5	Cup-shaped
Griffiths TW, et al. [4]	1997	84/F	Scalp	Mid pruritic papules	15/0.3-0.5	Hypertension	1	Not reported
Kaddu S, et al. [5]	2002	-	Forehead	-	2/-	-	-	Cup-shaped/cystic Cup-shaped/nodular
Koç M, et al. [6]	2009	52/F	Occipital area	Pruritic verrucous papules	15/0.5-1.3	Health	1.5	Not reported
Martorell-Calatayud A, et al. [7]	2012	76/M	Trunk (chest, back)	Mild pruritus lesions	Numerous/0.5-1.3	Psoriasis	0.5	Cup-shaped
Ugras N, et al. [8]	2014	55/F	Parietal scalp left forehead nose	Brownish papules with smooth surface	8/0.2-0.6	-	3-4	Cup-shaped
Wang S, et al.	Current case	62/M	Right forehead and temporal region		Numerous/0.5-2	Health	2	Cup-shaped

Grover's disease, Hailey-Hailey disease, acantholytic acanthoma, familial dyskeratotic comedones, acantholytic actinic keratosis, and acantholytic squamous-cell carcinoma.

Surgical excision is the most effective method for removing the warty dyskeratoma lesion. Recurrence is rare. Malignant transformation has not been reported. The partial lesions of our patient were surgically excised. The patient is in follow up.

In summary, we diagnosed a case of multiple warty dyskeratoma and reviewed reported cases in the literature. Multiple warty dyskeratoma should be differentiated with verruca, seborrheic keratosis, sebaceous hyperplasia, and actinic keratosis. The histopathologic differential diagnoses of warty dyskeratoma include Darier's disease, Grover's disease, Hailey-Hailey disease, acantholytic acanthoma, familial dyskeratotic comedones, acantholytic actinic keratosis, and acantholytic squamous-cell carcinoma.

Disclosure of conflict of interest

None.

Address correspondence to: Jianjun Qiao, Department of Dermatology, Beilun People's Hospital, No. 1288, Lushandong Road, Ningbo, China; Department of Dermatology, The First Affiliated Hospital, College of Medicine, Zhejiang University, No. 79 Qingchun Road, Hanghzou 310003, Zhejiang, China. Tel: +86-571-87236385; Fax: +86-571-87236385; E-mail: qiaojianjun@zju.edu.cn

References

[1] Lencastre A, Campos S and Cabete J. Warty dyskeratoma. J Am Acad Dermatol 2016; 75: e97-e98.
[2] Panja RK. Warty dyskeratoma. J Cutan Pathol 1977; 4: 194-200.
[3] Azuma Y and Matsukawa A. Warty dyskeratoma with multiple lesions. J Dermatol 1993; 20: 374-377.
[4] Griffiths TW, Hashimoto K, Sharata HH and Ellis CN. Multiple warty dyskeratomas of the scalp. Clin Exp Dermatol 1997; 22: 189-191.
[5] Kaddu S, Dong H, Mayer G, Kerl H and Cerroni L. Warty dyskeratoma--"follicular dyskeratoma": analysis of clinicopathologic features of a distinctive follicular adnexal neoplasm. J Am Acad Dermatol 2002; 47: 423-428.
[6] Koc M, Kavala M, Kocaturk E, Mete O, Can B, Zindanci I and Aktas S. Multiple warty dyskeratomas on the scalp. Dermatol Online J 2009; 15: 8.
[7] Martorell-Calatayud A, Sanmartin-Jimenez O, Traves V and Guillen C. Numerous umbilicated papules on the trunk: multiple warty dyskeratoma. Am J Dermatopathol 2012; 34: 674-675.
[8] Ugras N, Adim SB, Kilicoglu M and Baskan EB. Multiple warty dyskeratomas: case report. Iran J Public Health 2014; 43: 1145-1147.
[9] Vargas-Laguna E, Imbernon-Moya A, Aguilar-Martinez A and Burgos F. An unusual location of subungual warty dyskeratoma: a case report and review of the literature. Case Rep Dermatol Med 2017; 2017: 3613109.
[10] Lora V, Scarabello A and Cota C. Warty dyskeratoma as a cutaneous horn of the mons pubis. Am J Dermatopathol 2015; 37: 802-804.