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Case Report

Perianal warty dyskeratoma

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ABSTRACT

Warty dyskeratoma is a rare keratinization disorder marked by acantholytic dyskeratosis and belongs to the spectrum of acantholytic dyskeratosis in dermatology. It was first documented in 1954 and typically manifests in the head, neck, oral mucosa, and trunk, with several cases reported since its discovery. A 35-year-old unmarried gentleman presented in April 2022 with a 5-year history of perineal itching and discomfort. His physical examination revealed a 7-8 centimeter perianal grouped papular plaque. His medical history was unremarkable, and he has no family history of similar conditions or other dermatological disorders. Lab results showed non-reactive rapid plasma reagin and human immunodeficiency antigen/antibody combo. Liver and renal function tests were within normal range as well. The microscopic findings revealed features including prominent hyperkeratosis, invagination filled with keratin, and the surrounding area exhibiting acantholysis and intercellular edema. In conjunction with the patient's clinical presentation, this histological profile led to the diagnosis of warty dyskeratoma. After treatment with calcipotriene and other topical agents, the last presentation showed very good improvement; the physical examination indicated remarkable thin, flat, soft, grayish plaque in the perianal area. This case report presents a unique instance of warty dyskeratoma affecting the perianal area, shedding light on differential diagnosis and clinical management in this atypical context, which significantly broadens the understanding and emphasizing that the Warty dyskeratoma can manifest in diverse body regions, urging its consideration in the differential diagnosis of perianal papular lesions.

Keywords: Warty dyskeratoma, Perineum, Perianal dermatitis, Focal acantholytic dyskeratosis

INTRODUCTION

Warty dyskeratoma (WD) constitutes an uncommon keratinization disorder characterized by a histological pattern known as acantholytic dyskeratosis if it falls within the broader spectrum of focal acantholytic dyskeratoses, alongside conditions such as Darier disease, Haiely-Hailey disease, papular acantholytic dyskeratosis (PAD), and Grover disease.[1] The origins of WD trace back to its initial description by Helwing in 1954, where he documented a case resembling isolated Darier's disease. Over time, it has been referred to by various names, including isolated dyskeratosis follicularis.[2] WD typically manifests as papulonodular benign tumors, predominantly appearing on the head and neck region, with a tendency to present as solitary lesions; however, although infrequent, there have been documented instances of multiple lesions.[3,4]

CASE REPORT

We present a case of a 35 years old unmarried gentleman who presented to us in April 2022 with a 5-years history of perineal itching and discomfort. His physical examination revealed 7-8 centimeters perianal grouped papular plaque. His medical history was unremarkable and he has no family history of similar condition or other

dermatological disorders. Relevant lab results showed non-reactive rapid plasma reagin and human immunodeficiency antigen/antibody combo. Liver and renal function tests were within normal range as well.

The first histopathology biopsy was done in the country of origin and showed epidermolytic acanthoma. Five milligrams of daily Levocetirizine oral tablets were prescribed without benefit, and cryotherapy was advised. The patient was scheduled for a follow-up visit after seven days, in which the clinical examination showed no improvement. Based on that, the skincare was instructed, and the patient was educated about the illness. Topical steroids (salicylic acid 5% in mometasone 0.1%) and urea 10% cream were prescribed. Also, cryotherapy was discussed with the patient. After this treatment, the lesion showed a response, but recurrence was documented 15 days later.

On the next visit, the physical examination revealed a perianal gray-white papular plaque. Hydrocortisone cream and imiquimod cream 3x/w for six weeks were prescribed, leading to an improvement documented in the next follow-up visit (The lesion changed into a thin grayish-white plaque around the anal region). In the same visit, Fexofenadine tablets (180 mg) were added to the course of treatment, and the case was planned for follow-up in two months. Following this treatment, his condition improved, but the patient started to develop itchiness with a circinate rash in the groin area and scratch marks over the trunk and the limbs. Cryotherapy was applied, and topical medications were prescribed to him (tacrolimus ointment 0.1%, isoconazole/diflucortolone cream, and moisturizer cream).

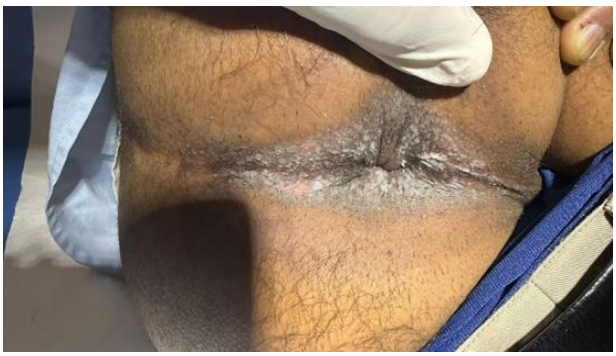

Figure 1: Thin flat soft grayish plaque in the perianal area, perianal warty dyskeratoma case report, Qatar University Health Center, Doha, Qatar, 2022.

The condition was stable for two months, after which the patient developed a few small fissures in the same area (perianal). A punch biopsy was done, and no change in the treatment was made while waiting for the biopsy report. Two months later, the condition improved (thin perianal white grayish plaque), and the pathological report showed a warty dyskeratoma and no malignancy. Calcipotriene topical was prescribed to the patient for 60 days, and education about the diseases and skin care was confirmed.

The last presentation showed very good improvement; the physical examination indicated remarkable thin flat soft grayish plaque in the perianal area. The patient continued on the calcipotriene in addition to clindamycin–benzoyl peroxide gel and clotrimazole topical cream for 60 days.

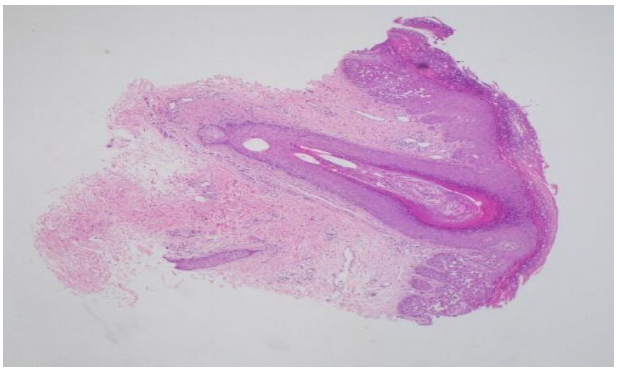

Figure 2: Skin punch biopsy shows prominent hyperkeratosis with an invagination filled with keratin. The area around the invagination show my degree of acantholysis and intracellular edema (4x).

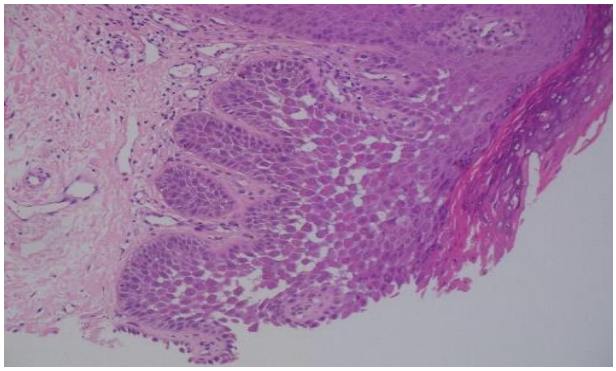

Figure 3: Acantholytic epidermis and acantholytic dyskeratotic cells (20x).

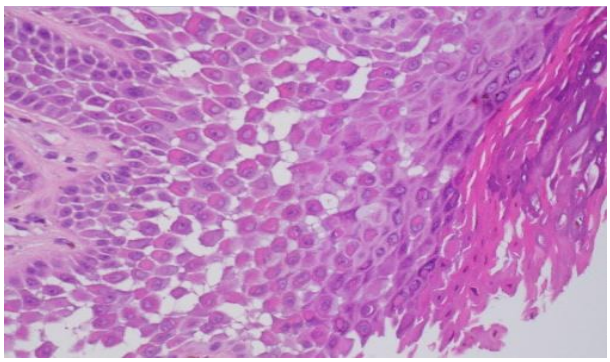

Figure 4: Acantholytic epidermis and acantholytic dyskeratotic cells (40x).

DISCUSSION

In our case, microscopic findings revealed features including prominent hyperkeratosis, invagination filled with keratin, and surrounding area exhibiting acantholysis

and intercellular edema. In conjunction with the patient's clinical presentation, this histological profile led to warty dyskeratoma (WD) diagnosis. We have obtained both written and verbal consent from the patient to ensure the ethical publication of the case details.

To the best of our knowledge and following a review of the published literature, our case marks the first reported instance of WD occurring in the perianal area. While exceedingly rare, cases of vulvar WD have been reported, including three cases involving female patients described by Duray and colleagues.[5] Moreover, within the same spectrum of conditions, papular acantholytic dyskeratosis (PAD) has been reported in a female patient, affecting both the perianal and vulvar areas.[6]

CONCLUSION

In conclusion, our case expands the clinical understanding of warty dyskeratoma by presenting a unique occurrence in the perianal region. It highlights the importance of considering WD within the differential diagnosis when confronted with papular perineal lesions. Such consideration not only helps the diagnostic process but also helps avoid unnecessary treatments.

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