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“A Rare Case of Multiple Warty Dyskeratomas Treated with Off-Label Dupilumab”

Introduction: Warty dyskeratoma (WD) is a rare, benign epidermal neoplasm that typically presents as a solitary keratotic papule or nodule on the scalp, head, or neck. Lesions are generally asymptomatic, and despite its name, WD has no association with verruca vulgaris or human papillomavirus (HPV). Histologically, WD is characterized by focal acantholytic dyskeratosis arranged in a cup-shaped epidermal invagination filled with keratotic debris, often in continuity with the follicular epithelium. Surgical excision is curative for solitary lesions but may be impractical for extensive lesions because of the need for repeated procedures and scarring. Topical medical therapies have shown variable results, and no standardized treatment approach for multiple WD has been established. Dupilumab has shown emerging efficacy in acantholytic dyskeratotic disorders that share the same histopathologic pattern of focal acantholytic dyskeratosis in WD. In the following case, a rare presentation of multiple WD is successfully treated with off-label dupilumab.

Case: A 46-year-old female presented in April 2024 with a several-week history of a pruritic, erythematous, papular rash on the legs. The rash was initially treated as irritant contact dermatitis versus folliculitis with triamcinolone, but the lesions persisted on the posterior thighs without improvement. An outside dermatologist clinically favored verrucae and performed multiple sessions of cryotherapy, also without improvement. She returned in May 2025 reporting pruritus rated 4/10, and interference with grooming and clothing tolerance. Examination revealed multiple lesions on the left rib cage and bilateral thighs. An initial shave biopsy in 2024 showed features consistent with WD, but limited sampling prompted a repeat biopsy. This demonstrated an organized, cup-shaped, crateriform lesion with suprabasilar acantholysis and dyskeratosis, characteristic of WD. By June 2025, approximately 15 pruritic papules and nodules involved the posterior thighs. Because of inadequate response to high-potency topical corticosteroids, and to avoid the adverse effects of prolonged steroid use, dupilumab was initiated. She received a 600 mg loading dose followed by 300 mg subcutaneously every two weeks. At one month, pruritus had improved to 2/10 and the lesion count had decreased from approximately 15 to 4. After four months, near-complete resolution was observed, with only residual post-inflammatory hyperpigmentation, and pruritus had completely resolved.

Discussion: WD most commonly occurs in middle-aged to older adults with a male predominance, and multiple lesions have rarely been reported in the literature. Because acantholytic dyskeratosis overlaps histologically with Grover disease, clinicopathologic correlation was essential. Multiple WD was favored because the lesions were discrete and persisted in the same locations for years rather than appearing as eruptive crops. Histopathology further supported the diagnosis, demonstrating the organized crateriform architecture characteristic of WD. The response to dupilumab was not used to distinguish between these diagnoses, as improvement has also been reported in Grover disease. To our knowledge, this is the first reported use of dupilumab in WD. This case adds to the limited existing literature on multiple WD and supports dupilumab as a potential treatment option for refractory or extensive disease where surgical management is impractical.

