

Oral warty dyskeratoma of the retromolar trigone: An unusual presentation of a rare lesion



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INTRODUCTION

Warty dyskeratoma (WD), also known as *isolated Darier disease* or *focal acantholytic dyskeratosis*,¹ is a rare mucocutaneous lesion. It usually presents as a solitary, asymptomatic umbilicated papule, most often in the head and neck region of middle- to older-age adults. A slight male predilection has been observed. When it occurs in the oral cavity, WD is most commonly seen on the hard palate and alveolar ridge.² Fewer than 50 cases of oral warty dyskeratoma are reported. Here we present a case of WD occurring on the left retromolar trigone in a 78-year-old man.

CASE REPORT

A 78-year-old man was referred to an oral and maxillofacial surgeon for evaluation of an ulcerated lesion of the left retromolar trigone. The patient described feeling “a hole in his gums” behind his lower left teeth. The duration of the lesion was unknown. The patient’s medical history was significant for glaucoma and chronic obstructive pulmonary disease but was otherwise unremarkable.

Clinical examination found a 0.4-cm round mixed red and white papule with indurated borders. Portions of the lesion had a vaguely warty or verruciform appearance; these areas appeared white in color (Fig 1). The area exhibited mild tenderness on palpation but was otherwise asymptomatic. The remainder of the intraoral examination was unremarkable. In addition, a panoramic radiograph showed no bony involvement in the affected area. Clinical differential diagnosis included chronic ulcer, exuberant granulation tissue, irritated papilloma,

Abbreviations used:

SCC: squamous cell carcinoma
WD: warty dyskeratoma

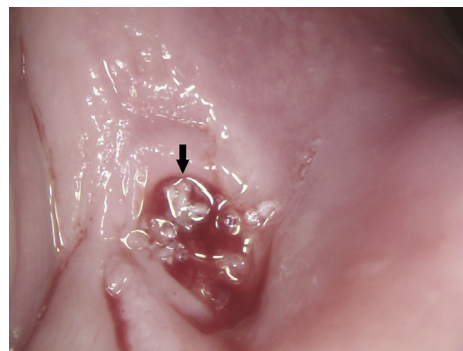


Fig 1. Oral warty dyskeratoma. A 0.4-cm round, mixed red-white papule with indurated borders was observed on the left retromolar pad on clinical examination. Portions of the lesion had a vaguely verruciform appearance (arrow).

and squamous cell carcinoma (SCC). A portion of the lesion and surrounding normal mucosa was biopsied and sent for histologic analysis.

Histologic examination found soft tissue covered by benign-appearing stratified squamous epithelium. Overlying the epithelium, keratotic material was present. Within the surface epithelium, intraepithelial (suprabasilar) clefting and elongated, test-tube shaped, rete ridges were identified. In addition, the lesion displayed features of basilar hyperplasia and dyskeratosis. A slender, keratin-filled crevice was seen extending from the epithelial surface to the

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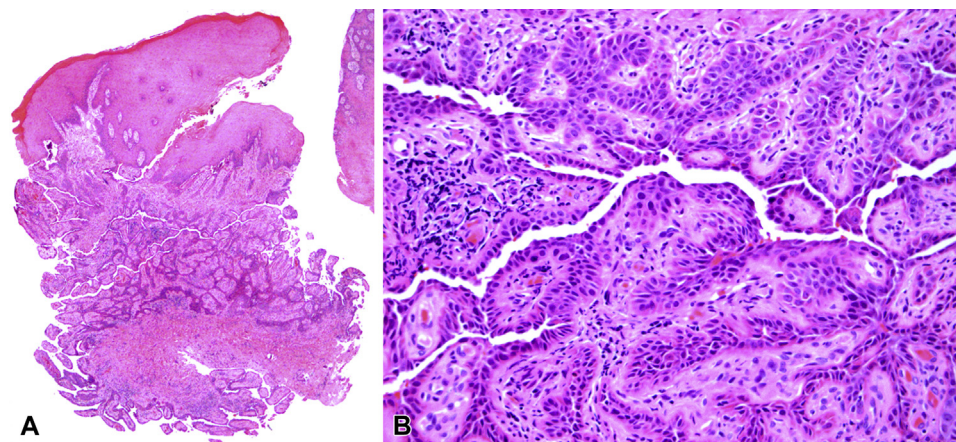


Fig 2. Oral warty dyskeratoma. **A**, Low-power photomicrograph shows a keratin-filled crevice, intraepithelial clefting, basilar hyperplasia, and elongated rete-ridges. **B**, High-power photomicrograph shows the characteristic intraepithelial clefting. (**A** and **B**, Hematoxylin-eosin stain; original magnifications: **A**, $\times 40$; **B**, $\times 400$.)

region of suprabasilar clefting (Fig 2, A and B). These histologic findings were consistent with a diagnosis of warty dyskeratoma. The classic thick, parakeratin plug of WD was not identified; however, this was attributed to the manner in which the specimen was embedded. Deeper levels of the specimen were reviewed; however, the tissue was exhausted because of small specimen size.

DISCUSSION

Oral WD is a distinctly uncommon lesion, with less than 50 reported cases in the literature. Allon and Buchner¹ published a comprehensive review of oral WD, in which they catalogue 41 total cases, including a case of their own occurring on the buccal mucosa of an 81-year-old woman. Since their publication in 2012, only 1 additional case of oral WD has been reported (excluding the current case); this was by Steele et al³ in 2014 who described a case of WD occurring on the buccal mucosa of a 60-year-old white man. Ugras et al⁴ reported a case of warty dyskeratoma-like lesions occurring on the skin of the scalp and face of a 55-year-old woman; however, these lesions were multiple, and a diagnosis of Darier disease was made.

When it presents intraorally, WD most commonly appears as a white nodule, papule, or plaque, which may be corrugated and contain an umbilicated center.² This appearance contrasts with cases of cutaneous WD, which are more often skin colored and exhibit a rolled, smooth edge and hyperkeratotic central plug.⁵ In both cutaneous and mucosal WD, lesions are typically asymptomatic, although there may be a central area of drainage or bleeding.⁶ Cutaneous WD is most common on the skin of the

head, neck, trunk, and extremities.⁵ Intraorally, WD is usually seen on keratinized mucosa such as the maxillary and mandibular alveolar ridges and hard palate.¹ In our case, the lesion of WD presented on the left retromolar trigone, which is an area of nonkeratinized tissue containing attachments of the buccinator and superior constrictor muscles and the tendon of the temporalis muscle. Allon and Buchner¹ reported no lesions occurring in this location in their meta-analysis.

The etiologies of both oral and cutaneous WD are unknown. Cutaneous WD is derived from pilosebaceous units; however, this is not the case with oral WD, as pilosebaceous units are not normally found intraorally.⁷ Although ectopic sebaceous glands (Fordyce spots) can be seen on the buccal mucosa, these do not correlate with the intraoral distribution of WD.¹ Local trauma caused by cheek biting or ill-fitting dentures have been implicated in some cases of oral WD. Some investigators suggest a link between tobacco use and oral WD.¹

Histologic differential diagnosis of oral WD may include SCC, particularly the acantholytic forms, keratoacanthoma, and Darier disease.¹ Oral WD may mimic SCC clinically because of the presence of ulceration and verruciform features, as was the case with our patient. However, WD often presents as a cup-shaped configuration not typically seen in SCC. Furthermore, the more common intraoral locations for WD are less frequently involved in SCC. Histologically, SCC shows features of cellular atypism and increased mitotic activity not seen in WD.⁸ Keratoacanthoma may show clinical and histologic similarities because of its shared cup-shaped configuration; however, it lacks features of

dyskeratosis and is unlikely to occur intraorally aside from the outer lip.¹ Darier disease will present with multiple lesions and is caused by mutations in the *ATP2A2* gene on chromosome 12q23-24, which encodes the sarcoplasmic/endoplasmic reticulum calcium-adenosine triphosphate isoform 2.⁹ This mutation is only described in Darier disease and is not seen in WD. Intraoral manifestations of Darier disease may also cause a cobblestone appearance of the oral mucosa absent in cases of WD.² Complete surgical excision is the preferred treatment for WD. WD does not recur after removal, and no malignant potential has been reported.²

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