

# Oral Bowenoid Papulosis

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## PRACTICE POINTS

- Bowenoid papulosis is triggered by human papillomavirus infection and manifests clinically as solitary or multiple verrucous papules and plaques that usually are located on the genitalia.
- Oral bowenoid papulosis is rare, and recognition of this unusual presentation is important for the diagnosis and management of this disease.

## To the Editor:

A 22-year-old Somali woman presented to our institution with oral lesions of 2 years' duration. The lesions started as small papules in the corners of the mouth that gradually continued to spread to the mucosal lips and gums. The lesions did not drain any material. The patient reported that they were not painful and had not regressed. She was concerned about the cosmetic appearance of the lesions. The patient believed the lesions had developed from working in a chicken factory and was concerned that they appeared possibly due to contact with a substance in the factory. Additionally, she noted that her voice had become hoarse. She was otherwise healthy and denied any sexual contact or ever having a blood transfusion.

Physical examination revealed 10 to 15 flesh-colored papules measuring 2 to 3 mm in diameter on the vermilion, mucosal surfaces of the lips, and upper and lower gingivae (Figure 1). No lesions were seen on the hard and soft palate, tongue, buccal mucosa, or posterior pharynx.

Skin biopsy of the left lower mucosal lip revealed parakeratosis, acanthosis, superficial koilocytes, and atypical keratinocytes with frequent mitoses (Figures 2A–2C).



**FIGURE 1.** Flesh-colored papules (2–3 mm in diameter) on the vermilion, mucosal surfaces of the lips, and upper and lower gingivae.

In situ hybridization testing for human papillomavirus (HPV) was negative for low-risk types 6 and 11 but positive for high-risk types 16 and 18 (Figure 2D). Laboratory investigations including complete blood cell count, electrolyte panel, and liver function studies were normal, and serum was negative for syphilis and human immunodeficiency virus antibodies.

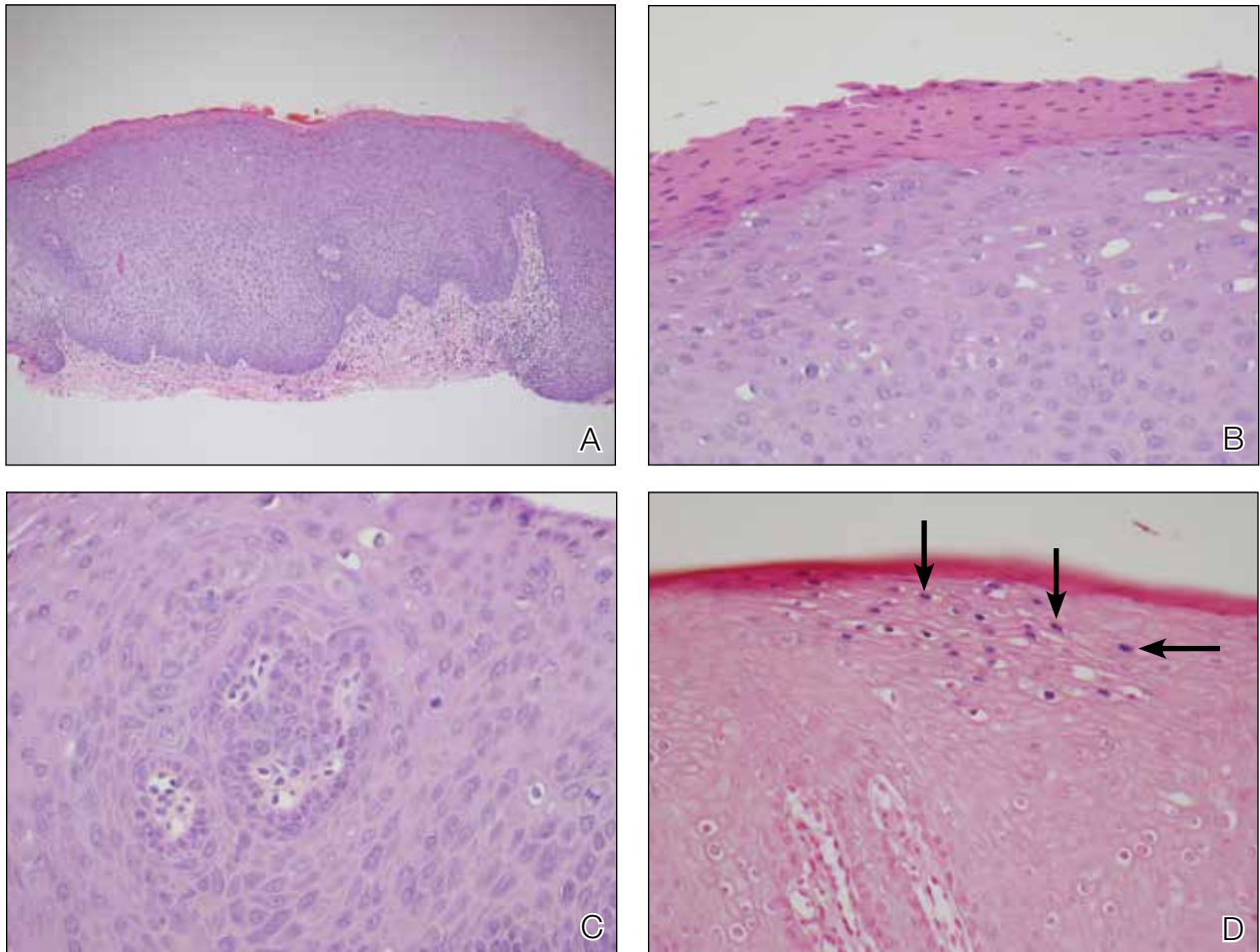
The combined clinical and histologic findings were diagnostic of oral bowenoid papulosis. Gynecologic evaluation showed that the patient had undergone female circumcision, and she had a normal Papanicolaou test. The patient was referred to both the ear, nose, and throat clinic

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**FIGURE 2.** Results from a skin biopsy of left lower mucosal lip showed parakeratosis and acanthosis (A), as well as superficial koilocytes and atypical keratinocytes with frequent mitoses (B)(H&E, original magnifications  $\times 10$  and  $\times 20$ ). Higher magnification of superficial koilocytes and atypical keratinocytes with mitoses (C)(H&E, original magnification  $\times 40$ ). In situ hybridization testing for human papillomavirus was negative for low-risk types 6 and 11 but positive for high-risk types 16 and 18 (arrows)(D)(H&E, original magnification  $\times 40$ ).

as well as the dermatologic surgery department to discuss treatment options, but she was lost to follow-up.

Bowenoid papulosis is triggered by HPV infection and manifests clinically as solitary or multiple verrucous papules and plaques that are usually located on the genitalia.<sup>1</sup> Only a few cases of bowenoid papulosis have been reported in the oral cavity.<sup>1-5</sup> Because this disease is sexually transmitted, the mean age of onset of bowenoid papulosis is 31 years.<sup>2</sup> There is a small risk (2%–3%) of developing invasive carcinoma in bowenoid papulosis.<sup>1-3,6</sup> Most lesions are associated with HPV type 16; however, bowenoid papulosis also has been associated with HPV types 18, 31, 32, 35, and 39.<sup>2</sup>

Some investigators consider bowenoid papulosis and Bowen disease (a type of squamous cell carcinoma [SCC] in situ) to be histologically identical<sup>1,6</sup>; however, some histologic differences have been reported.<sup>1-3,6</sup> Bowenoid papulosis has more dilated and tortuous dermal capillaries and less atypia and dyskeratosis than Bowen disease.<sup>1,6</sup>

In contrast to bowenoid papulosis, Bowen disease is characterized clinically as well-defined scaly plaques on sun-exposed areas of the skin in older adults. Invasive SCC can be seen in 5% of skin lesions and 30% of penile lesions associated with Bowen disease.<sup>2</sup> Risk factors for Bowen disease include sun exposure; arsenic poisoning; and infection with HPV types 2, 16, 18, 31, 33, 52, and 67.<sup>1,6</sup>

Oral bowenoid papulosis is rare. A PubMed search of articles indexed for MEDLINE using the term *oral bowenoid papulosis* yielded 7 additional cases, which are summarized in the Table. In 1987 Lookingbill et al<sup>2</sup> described one of the first reported cases of oral disease in a 33-year-old immunosuppressed man receiving prednisone therapy for systemic lupus erythematosus who had both mouth and genital lesions. All lesions were positive for HPV type 16. The patient subsequently developed SCC of the tongue.<sup>2</sup> Since then, 7 other cases of oral bowenoid papulosis have been described including the

Reported Cases of Oral Bowenoid Papulosis

Reference (Year)	Age, y	Location of Papules	Immunosuppression Status	Genital Involvement	Outcome	HPV Type
Lookingbill et al <sup>2</sup> (1987)	33	Hard palate, buccal mucosa	Prednisone therapy for SLE	Yes	SCC of the tongue	16
Kratochvil et al <sup>3</sup> (1989)	21	Hard and soft palate, buccal mucosa	Chemotherapy for Hodgkin lymphoma	Yes	Lost to follow-up	16
Daley et al <sup>1</sup> (2000)	29	Lower lip (solitary)	None	No	Lost to follow-up	NA
	34	Tongue (solitary)	None	No	Surgical excision, no recurrence	NA
	54	Lower lip (solitary)	None	No	Surgical excision, no recurrence	16, 18
Degener et al <sup>4</sup> (2004)	39	Lower lip	HIV	Yes	No follow-up	32
Rinaggio et al <sup>5</sup> (2006)	42	Lips	HIV infection	Yes	Failed treatment with imiquimod and interferon alfa	32
Current case	22	Vermilion, mucosal surfaces of the lips, upper and lower gingivae	None	No	Lost to follow-up	16, 18

Abbreviations: HPV, human papillomavirus; SLE, systemic lupus erythematosus; SCC, squamous cell carcinoma; NA, not available; HIV, human immunodeficiency virus.

current case.<sup>1,3-5</sup> Four (57%) cases in total were associated with immunosuppression and genital lesions.<sup>2-5</sup>

The risk for progression of oral bowenoid papulosis to invasive SCC is not known. Our search yielded only 1 case of this occurrence.<sup>2</sup>

Two of 3 cases of solitary lip lesions in oral bowenoid papulosis were treated with surgical excision.<sup>1</sup> Other treatment options include CO<sub>2</sub> laser therapy, cryotherapy, 5-fluorouracil, bleomycin, intralesional interferon alfa, and imiquimod.<sup>1-3,5,6</sup>

Our case represents a rare report of oral bowenoid papulosis. Recognition of this unusual presentation is important for the diagnosis and management of this disease.

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