CASE REPORTS

Keratoacanthoma of the tongue: A diagnostic problem

YUK-KWAN CHEN, DDS, MS, LI-MIN LIN, DDS, MS, PHD, CHENG-CHUNG LIN, DDS, MS, and CHUNG-HO CHEN, DDS, Kaohsiung, Taiwan

Keratoacanthoma (KA) is a benign epithelial tumor characterized by an infiltrating nature, multiple or localized occurrence, and a self-healing course.[1] It commonly occurs on the exposed skin, especially in the head and neck region, and very rarely on the mucosal membrane of the oral cavity.[2] From our review of the English-language literature,[3] this report appears to be the first documented case of KA in the tongue. It also serves to illustrate the problems encountered in establishing such a diagnosis, particularly with respect to the difficulty of distinguishing it from well-differentiated squamous cell carcinoma (SCC).

CASE REPORT

A 23-year-old Chinese woman presented with a 2-week history of a painless ulcer of the left ventral tongue (Fig 1). Her medical history was otherwise noncontributory, with no history of cigarette smoking, alcohol consumption, or betel-quid chewing. Clinical examination revealed an ulcer circumscribed by a raised-rolled margin approximately 1.5 cm in diameter (Fig 1). An incisional biopsy was performed under local anesthesia during the first visit to the ear, nose, and throat department of our institution. The sample was subsequently diagnosed as a well-differentiated SCC by a general pathologist. The patient was referred to the dental department for further evaluation and treatment. The histologic section of the incisional biopsy was then reexamined, with only multiple epithelial islands and prominent keratin bed noted; however, none of the normal adjacent mucosa had been included in the tissue section. Furthermore, because clinical presentation of the lesion was compatible with intraoral KA and in light of the patient's age and lack of precipitating factors, an excisional biopsy, which included the margin of normal mucosa, was performed. Histopathologic examination revealed downward proliferation of the circumscribed lesion with central keratin. A flask-like configuration of cells was revealed, with a lateral collar and abrupt pronounced lipping of the lesion margin (Fig 2). Except for mild hyperchromatism and occasional mitotic activity in the basilar cells, no aberrant cytologic features were determined for the proliferating epithelium. Further, pseudoepitheliomatous hyperplasia and underlying inflammation were noted. Eventually, these histologic findings led to the diagnosis of KA of the tongue. Regular patient follow-up is ongoing. Healing of the area of excision appeared to be uneventful.

DISCUSSION

Intraoral KA, an isolated lesion occurring in the oral cavity, is an extremely rare clinical entity.[3] To the best of our knowledge, this is the twelfth documented case of intraoral KA,[3] and the first occurring in the tongue.

The clinical appearance of intraoral KA may make it extremely difficult to distinguish from the SCC analog, especially in the tongue because this is also a common site for the latter malignancy. Characteristically, KA is a quick-growing nodule with a tendency to lose its central epithelial layer. Thus, it may easily be confused with a neoplastic ulcer, presenting a diagnostic problem to the attending clinician.

To establish the correct histopathologic diagnosis, it is important that the biopsy specimen be fully representative of the lesion. The most characteristic feature of a KA is found at the margins, where the normal adjacent mucous membrane is elevated toward the central portion of the ulcer, with an abrupt change in the normal epithelium at the hyperplastic, acanthotic border. If, as in the present case where

From the Department of Oral Pathology, School of Dentistry, Kaohsiung Medical University.

Reprint requests: Li-Min Lin, DDS, PhD, Oral Pathology Department, School of Dentistry, Kaohsiung Medical University; e-mail, k0285@ms22.hinet.net.

Otolaryngol Head Neck Surg 2003;128:581-2.

0194-5998/2003/$30.00 + 0
doi:10.1016/mhn.2003.116

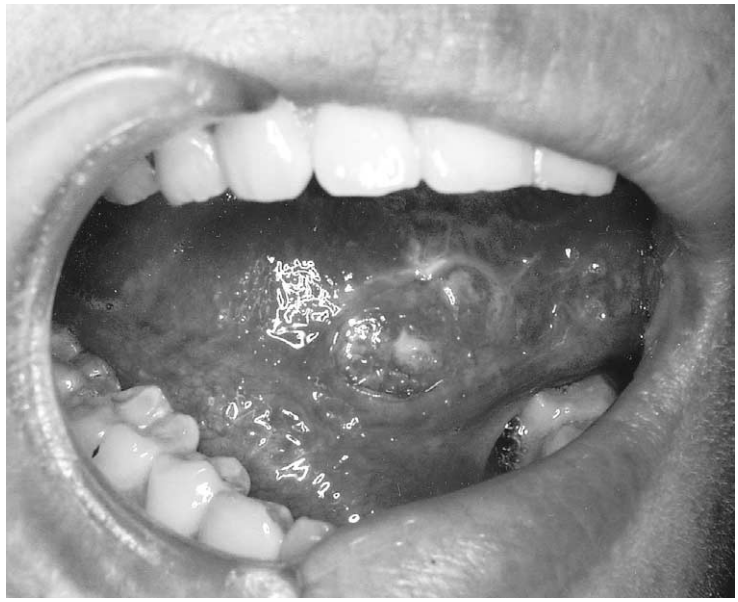

Fig 1. An ulcer circumscribed with a raised rolled margin is visible in the left ventral tongue.

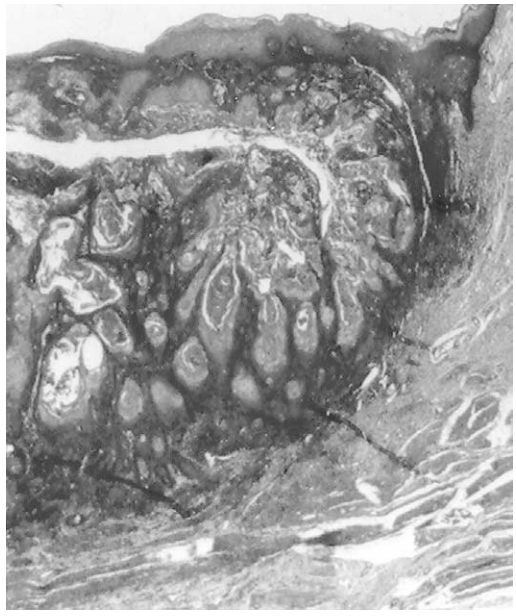

Fig 2. Histopathologic examination revealing downward proliferation of the circumscribed lesion with central keratin. The margin was characterized by flask-like cell configuration with a lateral collar (hematoxylin-eosin stain; original magnification, ×40).

incisional biopsy was performed, the central part of the lesion is sampled for biopsy, the diagnosis may be impossible because the adjacent border of the specimen, which is essential for differential diagnosis, is not included. Clinical data that identify the rapid growth of an ulcerative lesion in a younger individual, without any precipitating factors, may also be of particular diagnostic importance.

Classically, cutaneous KA may involute spontaneously;[1] however, spontaneous regression of the intraoral variant has not been observed because the lesions were completely excised in all of the cases reported.[3] Despite this exclusively surgical treatment, no recurrences were reported,[3] and it seems reasonable to recommend wide excisional biopsy to exclude SCC histologically.[4]

In conclusion, based on our experience in this case, KA should be included in the differential diagnosis where an ulcerated lesion with circumscribed, rolled margins and a keratinized bed are confirmed histopathologically. Careful microscopic analysis should be used for additional validation, in light of the fact that incorrect diagnosis may lead to further unnecessary, aggressive, and invasive surgical intervention.

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