Epithelial Inclusion Cyst of the Tongue Clinically Mimicking Tongue Cancer

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Abstract

Epithelial inclusion cysts in the tongue are extremely rare. We describe a case of an epithelial inclusion cyst occurring in the tongue in which it was necessary to differentiate the lesion from a malignant tumor. Clinical observation of the lesion and histopathological findings of biopsy tissue suggested the possibility of a malignant tumor. However, observation after immunohistochemical staining for Ki–67 and p53 ruled out malignancy. The lesion with a margin for safety was excised completely and a follow-up clinical examination at 3 years 4 months revealed no evidence of recurrence.

Introduction

Epithelial inclusion cyst is an uncommon lesion in mucosal sites such as the tongue. Even dermoid and epidermoid cysts, which are generally common lesions, represent less than 0.01% of all cysts in the oral cavity (1). Excluding dermoid and epidermoid cysts in the middle portion of the floor of the mouth, pathogenesis with inclusion of epithelial cells in the submucosal region is not well–known. Nonetheless, there is concern that these epithelial cells may have malignant potential.

We report a case of an epithelial inclusion cyst located at the tongue margin that initially appeared to be cancerous.

Case Report

A 71–year–old man with a history of chronic smoking reported swelling and continuous pain of the left tongue margin and was referred to the Oral and Maxillofacial Surgery Department of Wakayama Medical University. These symptoms first appeared 1 month earlier. There was no obvious related history of trauma. A localized swelling with whitening of the surface and a small erosive lesion on the left tongue margin was observed. The mass measured 12 mm×12 mm×14 mm. Submucosal induration in the dorsal portion of the tongue was noted on palpation (Fig. 1). Abnormal findings such as lymphadenopathy at the left submandibular and lateral cervical regions were not recognized on palpation. The clinical impression was tongue cancer and a subsequent incision biopsy was performed. Histopathological study of the biopsy tissue revealed many squamous cell islands with nuclear enlargement and mitosis in the submucosal muscle layer (Fig. 2). However, immunohistochemical staining revealed Ki–67– and P53–positive cells with preservation of polarity localized only in the basal layer of the epithelial islands (Fig. 3). Due to these histological findings, a malignant tumor was ruled out. The lesion, including the overlying

Fig. 1. Localized swelling at the tongue margin with white change of the mucosal surface and small area of erosion (arrow).
mucosa with a margin for safety, was excised completely under local anesthesia (Fig. 4). Histopathological examination of the excised tissue showed several cystic cavities lined with stratified squamous
epithelium in the muscle layer of the tongue. The lining epithelium showed orthokeratosis and a significant amount of keratin was seen in the cystic cavities. In the tissue surrounding the cysts, fibrous changes with inflammatory cell infiltration were observed (Fig. 5). No direct connection between the overlying mucosal epithelium or salivary glands and the cystic epithelium was observed. The definitive diagnosis was epithelial inclusion cyst of the tongue.

The patient was followed for 40 months with no evidence of recurrence (Fig. 6).

Discussion

Epithelial inclusion cysts in oral soft tissue are rare. Although dermoid and epidermoid cysts located on the floor of the mouth occur more frequently, they represent less than 0.01% of all oral cavity cysts (1). Only one case of naturally occurring epithelial inclusion cyst of the base of the tongue has been reported (2). Other reports concerned secondary development of cystic lesions after surgical procedures such as gingival grafting and uvulopalatopharyngoplasty (3, 4). There are several hypotheses concerning the etiology of epithelial inclusion cysts. Ahn et al. and other authors believe that in the absence of physical trauma, the cysts develop by implantation of epithelium into the lamina propria (5, 6). Several authors have reported cases of epithelial inclusion cyst occurring after surgical procedures (3, 4, 7, 8). Zappia et al. reported a case of a palatal inclusion cyst developing after uvulopalatopharyngoplasty. In that case, the cyst was intimately adherent to the previous palatal suture line (4), suggesting that the occurrence of these lesions may have been caused by implantation of epithelium into the incised wound during surgery. A hypothesis put forth by Nalini et al. states that epithelial inclusion cysts could arise due to squamous metaplasia of the glands (2). Cutaneous epidermal cysts, which are common lesions comprising approximately 85% to 90% of all excised cysts, originate from heterotopic epithelial cells misplaced in the neural tube during the embryo stage (9). Dermoid and epidermoid cysts of the floor of the mouth appear to be derived from the inclusion of epithelial cells on the median line at the fusion point between the first and second brachial arches (10). In the present case study, although the exact etiology of the cyst formation is unknown, one possibility is that chronic biting of the tongue margin caused epithelial implantation into the submucosal muscle layer. This is suggested because the covering mucosal color change and thickening are evidence of chronic irritation and also because there is no direct connection between the cystic lesions and the salivary glands.

Differential diagnosis of an epithelial inclusion cyst in the oral cavity includes a lymphoepithelial cyst. A lymphoepithelial cyst of the oral cavity presents as a small, asymptomatic, well-circumscribed, yellowish, elevated nodule, usually on the floor of the mouth or the ventral or posterolateral surface of the tongue (11-13). The cystic cavity is lined with thin layers of stratified squamous epithelium embedded within a well-circumscribed mass of lymphoid tissue, which often has well-developed follicles and/or germinal centers (13, 14). In the present case, spotted lymphoid cell infiltrations around cysts and epithelial islands were observed, but they were not dense and were without lymphoid structure having follicles and germinal centers.

In additional, lingual tonsils and minor salivary glands were not observed near the cysts microscopically. Therefore, the lesion can be distinguished from a lymphoepithelial cyst.

Because there have been reported cases of
squamous cell carcinoma developing in a mucosal epithelial inclusion cyst (2) and most surface epithelial-stromal tumors of the ovary are thought to arise from epithelial inclusion cysts (15), the potential for malignancy in epithelial cells in an inclusion cyst must also be considered. McDonald has reported that in a series of 637 epidermal inclusion cysts and sebaceous cysts of the skin, 7 carcinomas (1.1%) were found arising in the wall of these cysts (16). In this reported case, although the lesion of the tongue initially appeared to be a carcinoma, immunohistochemistry showed that Ki-67- and p53-positive cells were localized only in the basal cell layer of the epithelial islands. Therefore, malignancy was able to be ruled out. Forty months after the surgical removal of this epithelial inclusion cyst, no evidence of recurrence has been observed. However, careful follow-up of the patient was mandated due to the possibility of recurrence and subsequent malignant changes.

References