

Adenomatous hyperplasia of the minor salivary glands presenting as “tongue bump:” A rare case report

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ABSTRACT

The tongue remains the most common intraoral site for oral cancer worldwide, and in several countries, it is still considered a serious public health problem. Any lesion presenting as swelling on the tongue can mimic malignancy on clinical grounds and conclusive opinion requires histopathological examination. Adenomatoid hyperplasia of minor salivary glands is a rare hyperplastic oral lesion that can present anywhere in the oral cavity as an asymptomatic mass or nodule. It mainly involves soft and hard palate but its occurrence in the tongue is very rare with only one case cited in the literature. Here, we report the case of adenomatous hyperplasia of minor salivary glands in a 55-year-old female. The purpose of this case report is to highlight one such case which presented as a nodule on the tongue to make the clinicians and dentists aware of this unusual entity, as at this location, it becomes highly significant to distinguish adenomatous hyperplasia of minor salivary glands from relatively common epithelial and salivary gland neoplasms.

Key words: Adenomatoid hyperplasia, Malignancy, Minor salivary gland, Tongue

The tongue remains the most common intraoral site for oral cancer worldwide. In several countries, cancer of the tongue became a serious public health problem with significant morbidity and mortality. In India, the incidence rate of tongue cancer among males is 6.5/100,000 per annum [1]. Early detection and management play an important role in reducing morbidity and mortality. Any pathological condition presenting as swelling on the tongue can mimic malignancy on clinical grounds and conclusive opinion requires histopathological examination. Any error on the part of the clinician, dentist, and pathologist can result in radical surgical procedures, wherein patients may have to pay a heavy price in terms of lifelong morbidity.


Adenomatoid hyperplasia of minor salivary glands is one such rare hyperplastic oral lesion that can present anywhere in the oral cavity as an asymptomatic mass or nodule. It is a non-neoplastic condition, but its presence at a site like a tongue becomes a matter of concern. This unusual lesion was not well delineated in the literature until described by Giansanti *et al.* in 1971 [2], he described two such lesions on the palate which microscopically showed focal hyperplasia of minor salivary glands. To the best of our knowledge, only one case has been reported till now by Buchner *et al.* [3] on the tongue. This makes our case probably

second to be reported worldwide. The purpose of this case report is to highlight one such case which presented as a nodule on the tongue and to make the clinician and dentists aware of this unusual entity.

CASE REPORT

A 55-year-old female presented in the ENT outpatient department with a chief complaint of painless soft-tissue swelling on the left side of the tongue for the past 6 months. General examination was within normal limits. On local examination, the swelling was present on the lateral aspect of the left side of the anterior 2/3rd of the tongue. It was soft polypoidal swelling measuring 2 cm × 1 cm in size. The outer surface was smooth with no ulceration and well-defined margins. There were no cervical or submandibular lymph nodes on palpation. Routine hematological tests were within normal limits.

The swelling was excised under local anesthesia and sent for histopathological examination in 10% formalin. Grossly, the specimen consisted of a grayish-white soft-tissue piece measuring 2 cm × 1 cm × 1 cm. Microscopically, histopathological examination of stained sections revealed polypoidal lesion lined by stratified squamous epithelium with mildly edematous subepithelial tissue (Fig. 1) and nodules of seromucinous glands

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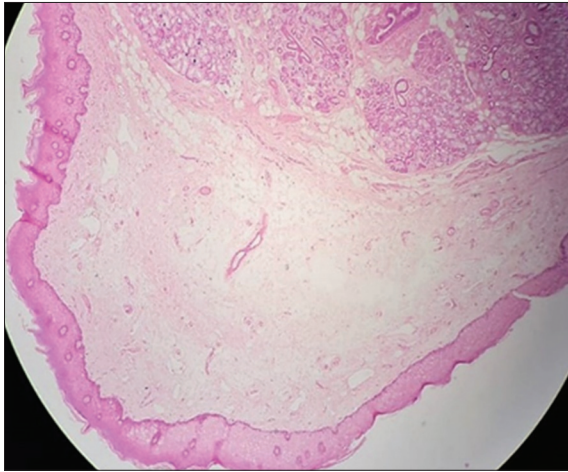


Figure 1: Scanner view showing polypoidal lesion with normal stratified squamous epithelium covering mildly edematous subepithelial connective tissue and aggregates of salivary glandular tissue (H and E 40×)

separated by adipose tissue along with accompanying large ducts (Fig. 2). There was no evidence of inflammation or epithelial dysplasia. Based on the distinctive histological findings, a diagnosis of adenomatoid hyperplasia of the minor salivary gland was rendered.

DISCUSSION

Adenomatoid hyperplasia of minor salivary glands is a clinicopathological condition resulting from hyperplasia of normal salivary gland tissue at that particular site. In 1981, Arafat *et al.* gave this entity clinical recognition by describing 10 such cases and adopting the term adenomatoid hyperplasia [4]. Buchner *et al.* in 1991 [3] and Barret and Speight in 1995 [5] studied 40 cases and 20 cases of adenomatoid hyperplasia, respectively, on the basis of clinical and histological features. Case series published by all the three listed above concluded that most of such lesions were located in the palate [3-5]. Only a few of them involved buccal mucosa, lips, and retromolar area published as individual case reports [6-8].

The exact pathogenesis of this condition is not clear, earlier studies conducted have suggested chronic inflammatory changes resulting in reactive glandular hyperplasia as the possible etiology [3,5]. Although systemic conditions such as gout, diabetes mellitus, alcoholism, menopause, Sjogren's syndrome, and drugs have been known to cause non-inflammatory and non-neoplastic enlargement of major salivary glands like parotid, none of them have been reported as affecting minor salivary glands [5].

Histologically, it consists of aggregates of predominantly normal-appearing mucinous glands intermixed with few serous glands separated by fibroconnective adipose tissue. The overlying epithelium may be normal or hyperplastic and there may or may not be associated chronic inflammation [3]. Adenomatoid hyperplasia needs to be distinguished histologically from benign and malignant salivary gland neoplasms such as pleomorphic adenoma, mucoepidermoid carcinoma, and adenoid cystic carcinoma [7], and developmental anomalies like hamartomas [5]. Since it clinically

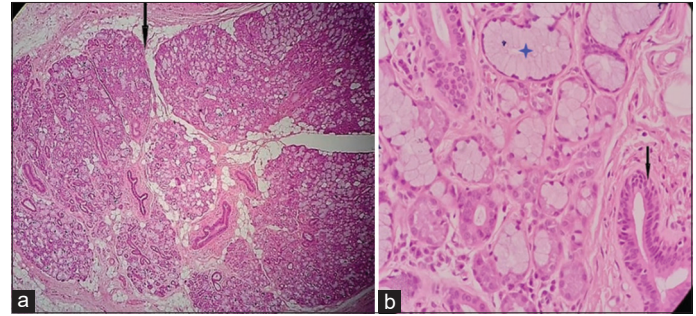


Figure 2: (a) Nodules of seromucinous glands separated by thin fibrous connective tissue (Hand E 100×); (b) higher magnification revealing mucinous glands (star) and ducts (arrow) (H and E 400×)

mimics malignancy, it has earned its description as “sheep in wolf’s clothing” [6]. Differentials can be added with a better understanding mentioning the role of immunohistochemical markers.

It is easily distinguished from salivary neoplastic pathology on the basis of architectural and cytologic features. It is also unlikely that it represents a hamartomatous process as most of the cases present in the fourth and fifth decades of life [3,5]. Adenomatous hyperplasias are more common in males [3,5]; however, in this scenario, it was female in the sixth decade of life. Since the lesion was present on the ventral aspect of the tongue, it created a clinically panicky situation to rule out malignancy which commonly involves this age and site.

Treatment of this condition is total excision. No recurrence has been reported in the literature till now [7]. Cytogenetic analysis of one case of adenomatoid hyperplasia was done by Manor *et al.* [9] to look for its malignant potential, in which he could identify translocation t(2;14) (q21;q22). This translocation t(2;14) has also been reported in malignant tumors where it involves different break points [9]. However, more studies are required to recognize its malignant potential.

CONCLUSION

Adenomatous hyperplasia of the minor salivary gland is a rare benign hyperplastic lesion predominantly involving palatal location. Its occurrence on the tongue is very rare, this being the second such case to be reported. The attending clinician and dentists need to be aware of this entity as this site is notoriously involved in oral malignancy.

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