Case Report

A very rare case of adenomatoid hyperplasia of minor salivary gland in a 55 year old male patient

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ABSTRACT

Adenomatoid hyperplasia of minor salivary glands (AHMSG) is an extremely rare non-neoplastic enlargement of minor salivary glands with uncertain pathogenesis. It is most commonly seen in males around 40–50 years of age. The most common sites are hard or soft palate. Clinically, the lesion presents as a sessile tumor-like nodule which mimics neoplasm with histological findings of benign hyperplasia and hypertrophy of mucous glands. Herein, we report a very rare case of AHMSG in a 55-year-old male patient who presented with asymptomatic swelling over the floor of the mouth since 8 months.

Key words: Adenomatoid hyperplasia, Minor salivary glands, Oral cavity swelling

Minor salivary glands are found on the walls of the oral cavity which secrete a minimal amount of saliva. They are named depending on their location such as labial, buccal, palatal, lingual, minor sublingual, palatoglossal, and Ebner glands. These glands are mixed mucous and serous in nature. Adenomatoid hyperplasia of the minor salivary glands (AHMSG) is an extremely rare hyperplastic oral cavity lesion. It was first described by Hendricks and Tyldesley, but first reported by Giansant and Waldron in 1971 [1]. Adenomatoid hyperplasia typically presents as a solitary, painless, well-circumscribed, firm sessile mass, or an elevated nodule. It can be of two types, either acinar type or ductal type. Occasionally, it may be painful, rarely penduculated or ulcerated. It is a pseudotumorous condition often mistaken for pleomorphic adenoma of the minor salivary glands. Only 102 cases were published worldwide. In Indian literature, only four cases have been reported till now [2].

The purpose of this report is to present a case of adenomatoid hyperplasia within the oral mucosa and to familiarize clinicians with this uncommon pathology of minor salivary glands, which should be differentiated from other non-neoplastic and neoplastic lesions of minor salivary glands. Herein, we report a very rare case of a 55-year-old male patient who presented with asymptomatic swelling in the retromolar area.

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CASE REPORT

A 55 year old male patient presented to the Department of Otorhinolaryngology in our institute with asymptomatic swelling over the lateral surface of the tongue on the right side behind the second premolar since 8 months. It started as a small peanut-sized swelling initially and then progressed to the present size of 2 cm. There was a history of one episode of bleeding from the site of swelling. The patient has a history of tobacco chewing and alcohol consumption but stopped 4 years ago (Fig. 1). Swelling was excised and sent to the department of Pathology for histopathological examination.

In the department of pathology, we received a single soft-tissue mass measuring 2×2×1 cm. The external surface was grey-brown to grey-white and congested. The cut section is grey-brown (Fig. 2). Bits were taken from representative areas and routine processing was done. The paraffin-embedded blocks were used for the preparation of sections and then stained with hematoxylin and eosin stain.

Histologically, it consists of aggregates of predominantly normal-appearing mucinous glands intermixed with a few serous glands separated by fibroconnective and adipose tissue. In our case, sections studied show mucosa lined by stratified squamous epithelium with focal epithelial hyperplasia. Submucosa shows fibrocollagenous tissue along with hyperplasia of normal-appearing mucinous acini arranged in lobules. Also seen are chronic inflammatory infiltrate, congested, and dilated blood

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vessels. Sections studied are negative for any dysplasia or malignancy (Fig. 3). Based on the distinctive histological findings, it was diagnosed as adenomatoid hyperplasia of the minor salivary gland.

DISCUSSION

AHMSG is an uncommon and extremely rare lesion of minor salivary glands. It is a localized hyperplastic nodular mass appearing as a soft or firm swelling on the oral mucosa [3]. Adenomatoid hyperplasia typically presents as solitary, painless, well-circumscribed, firm sessile mass or an elevated nodule. The lesion size ranges from 1 to 1.5 cm. Occasionally, it may be painful, rarely pedunculated or ulcerated. The most common sites are hard or soft palate and other sites such as retromolar trigone, tongue, buccal mucosa, and the lip can also be involved [4].

The etiology is mostly idiopathic but common possible factors are local trauma, chronic irritation, ill-fitting dentures, systemic diseases, drugs, bulimia, and also ingestion of tobacco products. It is also unlikely that it represents a hamartomatous process as it presents classically in the older age group [5]. Our case had a previous history of tobacco consumption. Typically men and women in the 40 to 50 years of age group are affected, but it may occur in any group from 9 to 79 years. The male-to-female ratio is 2:1. Previous case reports showed a male predominance [6]. Our case is also a male patient aged 55 years.

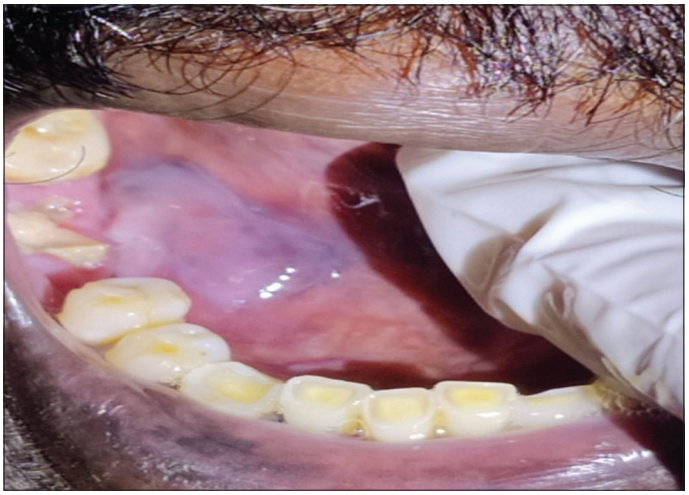

Figure 1: Swelling located at the floor of the mouth

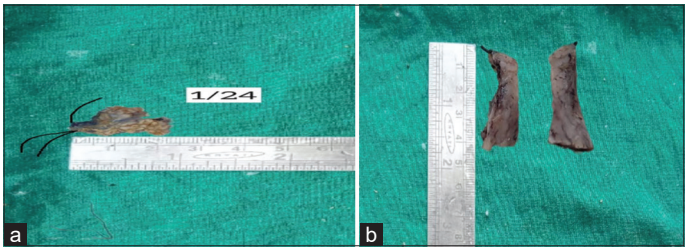

Figure 2: (a) Received single soft-tissue mass measuring 2×2×1 cm. The external surface is grey-white to grey-brown and congested; (b) Cut section is grey-white with focal grey-brown areas

The differential diagnosis must include fibroma and other benign and malignant salivary gland neoplasms such as mucocele, pleomorphic adenoma, and other palatal lesions. Since it clinically mimics malignancy, it is described as "sheep in wolf's clothing" [7]. It is distinguished from salivary neoplastic pathology based on architectural features. The interest in this entity arises from the fact that although it is a benign pseudotumoral lesion, it can be clinically confused with benign or malignant tumors.

Previous studies reported a very low Ki67 Index in these lesions which is similar to that of normal salivary glands indicating its benign nature [8]. If due care is not taken, mistaken diagnosis could result in inappropriate treatment. Hence, histopathology plays an important role in distinguishing adenomatoid hyperplasia from other lesions. These lesions may rarely continue to enlarge if not excised. The treatment is total excision as it is a benign condition [9]. On post-operative follow-up, the patient is fine without any symptoms.

However, it is reported that one palatal case has developed into mucoepidermoid carcinoma several years after diagnosis. In addition, there was one palatal case report, in which the cytogenetic finding showed translocation t(2;14)(q21;q22). This translocation t(2;14) has been reported in malignant tumors, such as chronic lymphocytic leukemia, chronic myeloid leukemia, B-cell precursor acute lymphoblastic leukemia, or in Toriello–Carey syndrome but occurred at different breakpoints. It is not known whether this chromosomal aberration may be a risk for malignancy of adenomatoid hyperplasia, and further studies are required to completely understand the possible roles of t(2:14) in these conditions [10].

In 1981, Arafat *et al.* [8] gave this entity clinical recognition by describing 10 such cases and adopting the term adenomatoid hyperplasia. Buchner *et al.* [5] and Barret and Speight [7] studied 40 and 20 cases, each, of adenomatoid hyperplasia in 1991 and 1995, respectively, based on clinical and histological features. Only 102 cases were published worldwide. In Indian literature, only four cases were reported till now (Table 1) [2,11-14].

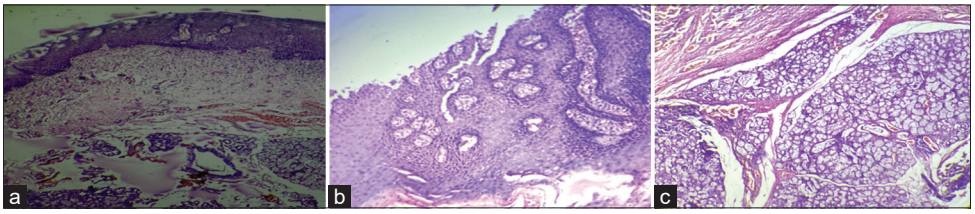

Figure 3: (a) Stratified squamous epithelium with mucinous acini (H&E ×40); (b) Focal hyperplasia of epithelium without atypia (H&E ×400); (c) Hyperplasia of normal-appearing mucinous acini (H&E ×400)

Table 1: Adenomatoid hyperplasia of minor salivary glands – Indian patients data including this case

Serial no	Presenter/year	No. of. cases	Sex	Site	Country
1	Sharma *et al*./2011	1	Male	Left lower lip	India
2	Ingle *et al*./2015	1	Female	Hard palate	India
3	Devera *et al*./2021	1	Female	Left side of tongue	India
4	Raju *et al*./2022	1	Male	Left side of lingual vestibule	India
5	Present case				

CONCLUSION

Adenomatoid hyperplasia of the minor salivary gland is an extremely rare, benign hyperplastic lesion, and the floor of the mouth is an unusual site. Histopathological examination plays an important role in the definitive diagnosis. Complete excision is preferable for both curative diagnostic and therapeutic purposes. The nature of this lesion has to be further explored with the aid of immunohistochemistry, cytogenetics, and molecular studies.

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