

## Case report

# Adenomatoid hyperplasia of minor salivary glands on buccal mucosa: a case report

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### Abstract:

*Adenomatoid hyperplasia (AH) of minor salivary glands is a non-neoplastic focal enlargement of the intraoral mucous glands. The pathogenesis is uncertain. However, it has been speculated that local trauma may play a role. This lesion most often occurs on the hard or soft palate. The lesion clinically presents as a sessile tumor-like nodule that mimics a neoplasm. The histopathologic findings include benign hyperplasia and hypertrophy of the minor mucous glands.*

*This article reports a rare AH case of buccal mucosa found in a 23-year-old Thai female patient. Relevant literature of its etiology is reviewed, together with the overall clinical and histopathologic features of AH of minor salivary glands.*

**Keywords:** ● Adenomatoid hyperplasia ● Minor salivary glands ● Buccal mucosa

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## รายงานผู้ป่วย

# การรอกเกินคล้ายเนื้องอกของต่อมน้ำลายย่อยบริเวณกระพุ้งแก้ม: รายงานผู้ป่วย

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### บทคัดย่อ

การรอกเกินคล้ายเนื้องอกของต่อมน้ำลายย่อย เป็นการเจริญเกินหรือการรอกเกินของต่อมน้ำลายย่อยในช่องปากชนิดเมื่อกรวยโรคนี้ไม่ใช่เนื้องอก ไม่ทราบสาเหตุในการเกิดอย่างแน่ชัด อย่างไรก็ตาม การบาดเจ็บเฉพาะที่อาจเป็นสาเหตุหนึ่งในการเกิด รอยโรคส่วนใหญ่พบที่เพดานแข็งและเพดานอ่อน ลักษณะทางคลินิกมักจะเป็นก้อนเนื้อคล้ายเนื้องอก ลักษณะทางจุลพยาธิวิทยาจะพบการขยายขนาดและเพิ่มจำนวนของต่อมน้ำลายย่อยในช่องปากชนิดเมื่อกรวย

รายงานผู้ป่วยนี้ จะนำเสนอผู้ป่วยหญิงไทยอายุ 23 ปี ที่มีการรอกเกินคล้ายเนื้องอกของต่อมน้ำลายย่อยบริเวณกระพุ้งแก้ม พร้อมทั้งบททวนวรรณกรรมที่เกี่ยวข้อง

**คำสำคัญ:** ● การเจริญเกินของต่อมน้ำลาย ● ต่อมน้ำลายย่อย ● กระพุ้งแก้ม

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### Introduction

Adenomatoid hyperplasia (AH) is a non-neoplastic enlargement of minor salivary glands. It was first described in 1971 by Giansanti et al<sup>1</sup>. AH is considered a benign pseudotumoral lesion. Clinically, the lesion presents as an asymptomatic, firm, sessile, nontender, and nodular mass that is not ulcerated<sup>2</sup>. This case occurs mostly on the hard or soft palate, but it was also reported in other oral minor salivary glands. Previously, the lesion had been reported as benign minor salivary gland hypertrophy, salivary glandular hyperplasia, adenomatous hyperplasia of minor salivary glands, acinar AH, and AH of minor salivary glands<sup>3</sup>. This condition has been regarded as idiopathic. However, the role of chronic local trauma has been suggested.

This article reports an AH case of minor salivary glands on the buccal mucosa, which is a rare localization for this entity. The etiology, clinical, and histopathological aspects, and also differential diagnosis will be discussed.

### Case report

A 23-year-old Thai female patient was presented with a complaint of a soft tissue mass at her right buccal mucosa, of which the size had gradually increased for a year. The patient gave a history of long-term chronic cheek biting, considered as a chronic local trauma. She denied smoking or any drug use. Her medical history was non-contributory. No regional lymphadenopathy was observed. Regarding intraoral examination, the lesion had a normal overlying mucosa, exophytic nodular appearance, smooth surface with 1 cm in size (Figure 1). There was no pain or tenderness. Panoramic radiography showed no abnormal finding. The clinical diagnosis of an irritation fibroma was rendered. The treatment plan consisted of a surgical removal under local anesthesia. The excised lesion was sent for histopathologic examination.

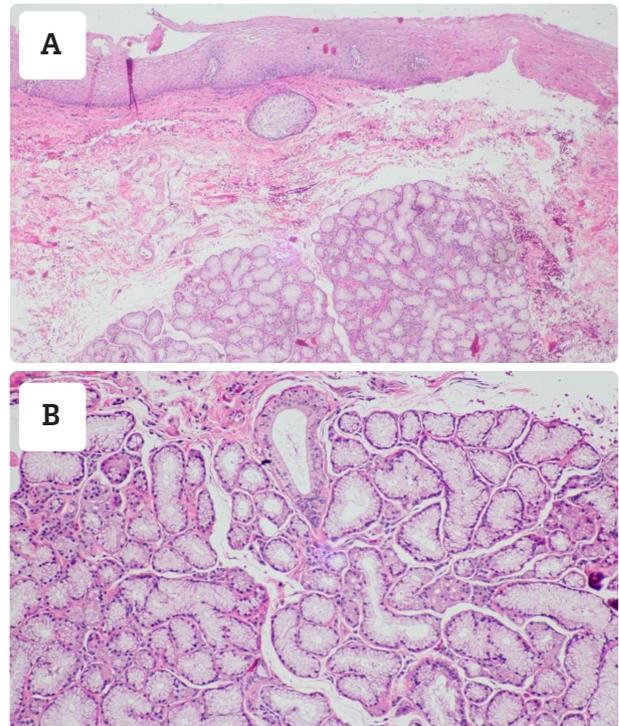
Histopathologically, the section revealed a soft tissue nodule covered by parakeratinized stratified squamous epithelium. Numerous salivary gland lobules with normal-

appearing mucous-acini were seen within subjacent fibrous connective tissue (Figure 2). Inflammatory reaction is rarely seen in the lesion. The definitive diagnosis obtained from these findings as an AH of minor salivary glands.

Postoperative healing was uneventful. There was no recurrence at a 1-year follow-up.



**Figure 1** Clinical aspect of a well-defined submucosal nodular swelling on right buccal mucosa



**Figure 2** Histopathologically (A) Mucous salivary gland lobules beneath buccal mucosa, which is covered by stratified squamous epithelium. (Hematoxylin and Eosin stain; original magnification  $\times 40$ ) (B) Normal-appearing mucinous acini with ductal structures (Hematoxylin and Eosin stain; original magnification  $\times 100$ )

### Discussion

AH is an uncommon lesion of minor salivary glands. It is a localized hyperplastic nodular mass appearing as a soft or firm swelling on the oral mucosa<sup>4,5</sup>. The lesion has a predilection for the palatal region (more than 50%); other sites such as retromolar area, buccal mucosa, lip, ventral surface of the tongue, and floor mouth are also seen to be affected<sup>4,7</sup>. There is no age predilection<sup>3</sup>. The lesion occurred between the age of 9 and 79 years<sup>4</sup>. The average age of onset is reported to be 39 years<sup>6</sup> and 44.5 years<sup>4</sup> by previous series.

Clinically, most examples present as circumscribed, sessile, and painless masses that may be soft or firm to palpation. They are normal in color, although some lesions are red or bluish<sup>4</sup>. The lesion size is in the range of 0.3-4 cm, with almost half of the lesions are 1-1.5 cm.<sup>3,4</sup> All lesions presented as a solitary mass, except for four multiple lesions<sup>5</sup>. For the palatal lesion, the differential diagnosis must include benign and malignant salivary glands neoplasms<sup>8</sup>. Therefore, the main importance of AH on palate is that it mimics a salivary gland neoplasm (such as pleomorphic adenoma and mucoepidermoid carcinoma), giving it the description of a "sheep in wolf's clothing"<sup>9</sup>.

Etiology of the AH is unknown<sup>4</sup>. It is an idiopathic, focal hypertrophic lesion of the minor salivary glands with limited growth potential. Buchner et al.<sup>4</sup> suggested a probable role of chronic irritations like local trauma, smoking, and dentures in this reactive hyperplasia. On the other hand, it is suggested that AH is not associated with specific factors (drugs, endocrine disorders, nutritional deficits, anorexia nervosa, and neurogenic factors) which may cause sialadenosis or enlarged major salivary glands<sup>4</sup>.

Histopathologically, the lesion contains multiple clusters of normal appearing mucous acini surrounded by fibrous connective tissue<sup>4</sup>. Both glandular hyperplasia and

hypertrophy were presented and sometimes filled with mucous. Focal areas of mucous spillage and fibrosis were occasionally discovered. Chronic inflammation was generally patchy or absent. It is interesting to note that no serous acinar hypertrophy was found in cases of AH arising from the mixed salivary glands<sup>10</sup>. The overlying epithelium was intact, but occasionally exhibiting pseudoepitheliomatous hyperplasia. In the superficial part of the lesion, one case showed features consistent with lichen planus<sup>9</sup> and the other lesion coexisted with pigmented nevus<sup>11</sup>.

The treatment of the lesion is total excision<sup>14</sup>. No recurrence has not yet been reported in the literature. When a lesion is confirmed to be an AH of minor salivary glands on excisional biopsy, no further treatment is required because these lesions do not recur<sup>7</sup>. However, it is reported that one palatal case has developed into mucoepidermoid carcinoma several years after diagnosis<sup>6</sup>. In addition, there was one palatal case report, 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 break points<sup>12</sup>. It is not known whether this chromosomal aberration may be a risk for malignancy of AH, and further studies are required to completely understand possible roles of t(2;14) in these conditions.

This case report occurred at an unusual site. This lesion located on buccal mucosa with the history of long-term cheek biting. Chronic local trauma was also suspected associated factor. Only three cases of buccal mucosa localization were reported in literatures<sup>4,8</sup>. The reason of the rarity of buccal localization may be explained with the low amount of the minor salivary glands in the buccal region, compared to the palate and other

parts of the oral mucosa<sup>8</sup>. Unlike the palatal lesion, the lesions on the buccal mucosa should be differentiated from irritation fibroma and other benign soft tissue tumors. Therefore, it must be excised and examined microscopically. Complete excision is preferable for this case for both curative and diagnostic purposes. Histopathology of this lesion was composed of multiple clusters of otherwise normal-appearing mucous salivary glands. Inflammatory reaction is rarely seen. The overlying epithelium was normal. AH of minor salivary glands was the final diagnosis. And one year after the surgery, the patient has shown no evidence of recurrence.

### Conclusion

AH of minor salivary glands is a rare, benign hyperplastic lesion of the minor salivary glands which is localized mostly on palatal regions. Buccal localization is uncommon and should be considered in the differential diagnosis of fibroma and other benign lesion. The consideration of chronic trauma in the etiology of this entity may be appealing as histology marked of the reactive hyperplasia of minor salivary glands.

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**Conflict of Interest:** None declared.

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