

## A Case of Adenomatous Hyperplasia of the Minor Salivary Glands

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Adenomatoid hyperplasia of the minor salivary glands is a rare clinicopathologic entity with an unknown etiology. The clinical features of the lesion are nodular, non-painful swellings, and the histologic features are aggregates of normal-appearing, salivary gland tissue, in excess of what would be anticipated for the anatomic site. This tumor is of significance because of its clinical resemblance to salivary gland tumors. Although this lesion may occur at any site on the oral mucosa, the labial mucosa is known to be the least common site. We herein report a rare case of adenomatous hyperplasia of the minor salivary glands located on the lower labial mucosa. (*Ann Dermatol (Seoul)* 18(1) 5~8, 2006)

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*Key Words:* Adenomatous hyperplasia, Minor salivary glands, Labial mucosa

### INTRODUCTION

Adenomatous hyperplasia of the minor salivary glands is a rare clinicopathologic entity, first described in 1971 by Giansanti et al.<sup>1</sup> Clinically, the lesion appears as a localized, non-painful swelling that mimics a salivary gland neoplasm or a fibroma, but histologically, it is composed entirely of normal-appearing salivary gland tissue. To date, more than 90 cases have been reported in English literature, with three large series<sup>2-4</sup> being reported. Almost all the cases occurred on the palate, although the aforementioned reports and others<sup>2,5</sup> have shown that other sites may occasionally be affected. Hence, while this lesion has attracted a great deal of interest in the dental and otolaryngology journals, no case has been reported in the dermatology journals. In addition, there have been no reported cases in Korea. We herein report on a case of adenomatous

hyperplasia of the minor salivary glands, which occurred on the lower labial mucosa of a 15-year-old Korean man.

### CASE REPORT

A healthy 15-year-old man presented with asymptomatic, multiple nodules on the lower labial musosa. The lesion had first appeared as a solitary papule two years ago, and it had slowly increased in size and number. On admission, five well-demarcated, 0.5-1.5 cm sized, round, flesh-colored, soft nodules were distributed on the lower labial mucosa (Fig. 1). On physical examination, no other remarkable finding were discovered. His past medical and family history were non-contributory. Microscopically, the lesion was composed entirely of multiple clusters of normal-appearing mucous acini surrounded by a fibrous connective tissue stroma (Fig. 2). The acini appeared hypertrophic and they were filled with excessive mucus so that nuclei were pushed to the basal aspect of the cells. The architecture and cytology of the glandular tissues were essentially normal. In contrast to the hyperplastic acini, the ductal elements were inconspicuous. The overlying epithelium was intact, but it exhibited psuedoepitheliomatous hyperplasia (Fig. 3). Inflammatory

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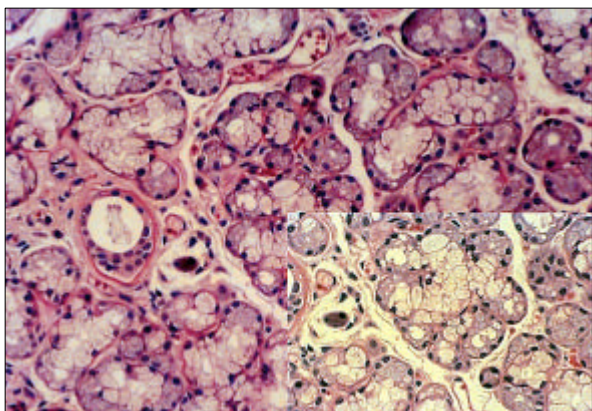
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**Fig. 1.** Multiple, well-demarcated, 0.5-1.5 cm sized, round, flesh-colored, soft nodules on the lower labial mucosa (H & E,  $\times 100$ ).

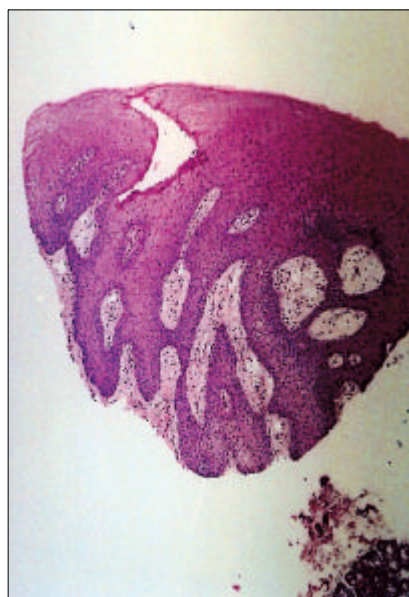


**Fig. 2.** Normal-appearing mucous acini and ductal structures from a lower lip lesion (H & E,  $\times 200$ ; inset,  $\times 400$ ).

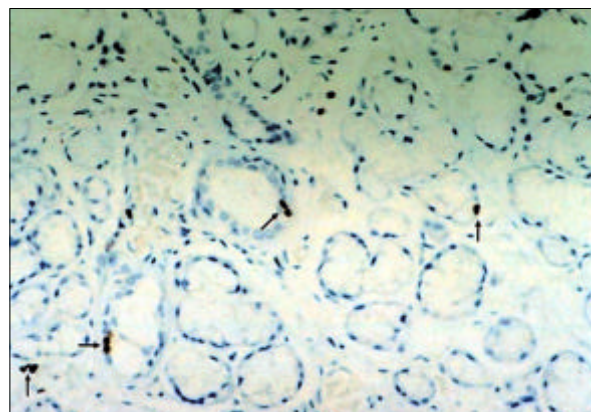
infiltrates were minimal, and mucous spillage or fibrosis was not observed. In order to analyze the cell proliferative activity of the adenomatous hyperplasia, Ki-67 immunochemistry was performed. The numbers of immunopositive nuclei were counted and the percentage was calculated. The labeling index of AH was 1.4% (Fig. 4). The lesion was excised and there have been no signs of recurrence 1 year after excision.

## DISCUSSION

Adenomatoid hyperplasia of the minor salivary glands is an uncommon clinicopathologic entity



**Fig. 3.** Biopsy specimen from the nodules on the lower lip shows hyperkeratosis and acanthosis in the epidermis (H & E,  $\times 40$ ).



**Fig. 4.** The Ki-67 labeling index is 1.4% (Ki-67,  $\times 200$ ).

which is comprised of a combination of clinical swelling with the histologic findings of aggregates of normal-appearing salivary gland tissue, in excess of what would be anticipated for the anatomic site. The significance of adenomatoid hyperplasia is its clinical resemblance to a salivary gland neoplasm, and this has resulted in the description of the lesion as a "sheep in wolf's clothing"<sup>6</sup>. However, the condition is benign and does not predispose or otherwise relate to a benign or malignant salivary gland neoplasm.

Since the first description of this entity by Giansanti et al.<sup>1</sup> in 1971, 98 cases of adenomatous hyperplasia have been reported in the literature, with the three largest series from Araft et al.<sup>2</sup>, Buchner et al.<sup>3</sup> and Barrett & Speight<sup>4</sup>. This lesion is predominantly found in male patients, with a gender ratio of about 2 : 1. It is found in patients of all ages, but it is more frequently found in patients between the fourth and sixth decades<sup>3,4</sup>. Most of the patients have been Caucasians. This lesion is comparatively uncommon in Asians, with only 17 Asian patients having been reported<sup>7</sup>. To our knowledge, the present lesion appears to be the first reported case in a Korean patient. Although this lesion may occur at any site on the oral mucosa where mucous salivary glands normally exist, the reports in the literature indicate that the palate is the most common site for this lesion. Of the 98 reported cases, 84 lesions were located on the palate, 7 lesions were on the retro-molar area<sup>3-5,8-10</sup>, 2 lesions were on the tongue<sup>3,11</sup>, 2 lesions were on the buccal mucosa<sup>3</sup> and 2 lesions were on the lip<sup>3</sup>. The present lesion is a rare case because of its uncommon location on the labial mucosa. Most of the lesions have presented as a solitary, well-demarcated, asymptomatic, tumor-like mass, except for four cases that reported pain and four cases of multiple lesions. The lesions varied from 2 to 40 mm at the greatest diameter<sup>3,4,12</sup>. Almost all the lesions were 10 to 15 mm in size. With one exception<sup>4</sup>, no recurrence followed excision of the lesions. Therefore, once the diagnosis is established from a biopsy, no further treatment is needed.

Factors such as endocrine disorders, nutritional deficiencies, drugs or neuropathies are known to be causes of the asymptomatic, noninflammatory, non-neoplastic swelling (sialadenosis) of the major salivary glands, and this mainly affects the parotid glands and rarely the submandibular gland<sup>13,14</sup>. However, the etiology of adenomatous hyperplasia of the minor salivary gland is uncertain. Barrett & Speight<sup>4</sup> suggested that chronic local trauma by dental appliances and smoking was a likely cause. However, in most cases, no history of trauma was observed and the histologic evidence of inflammation was absent. The present patient did not reveal a history of chronic irritation or trauma. At least for now, the nature of this lesion is best considered idiopathic.

The clinical differential diagnosis included a pleomorphic adenoma, or other benign tumor, and

a salivary gland or other malignancy. As it has been pointed out previously, the significance of this lesion is derived from its clinical resemblance to a neoplasm of a salivary gland. But histologically, it is easily distinguished from a salivary neoplasm because the latter possess abnormal cytologic and architectural features. Microscopically, this lesion was composed of multiple aggregates of normal-appearing mucous glands surrounded by fibrous connective tissue. The glands appeared hyperplastic, but the acinar morphologic features were generally normal. The acini occasionally appeared hypertrophic and were filled with excessive mucus. In contrast to the hyperplastic acini, the ductal structures were inconspicuous throughout the lesion. Isolated focal areas of mucous spillage and fibrosis were sometimes observed. Some evidence of inflammation was found, but usually this was minimal and localized to one or a few lobules of the gland. The overlying epithelium was intact and occasionally it exhibited pseudoepitheliomatous hyperplasia.

Shinoyama et al.<sup>7</sup> showed that there was no statistical difference in the Ki-67 labelling index between adenomatous hyperplasia (1.7%) and the control salivary glands (1.8%). From that result, they insisted that adenomatous hyperplasia had little or no proliferative activity. In the present study, the Ki-67 labelling index was noted as 1.5%, and this result was consistent with the previous study.

We report here on a rare case of adenomatous hyperplasia which developed at a fairly uncommon location, on the lower labial mucosa, and this case presented with uncommon, multiple lesions. Dermatologic clinicians should be aware of the existence of this type of minor salivary gland lesion which can clinically identified as a salivary gland tumor because of the therapeutic and prognostic implications.

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