Pleomorphic Adenoma of the Lower Lip: A Case and Review of the Japanese Literature

by

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Introduction

Pleomorphic adenoma is the most common tumor of the salivary glands and mainly occurs in the major salivary glands, the parotid gland in particular^[1,2], and it is relatively rare in the minor salivary glands. Among the latter the palate is the most common site for pleomorphic adenomas^[3,4], and pleomorphic adenomas in the lip are rare, particularly in the lower lip^[5,6].

We encountered a case of pleomorphic adenoma in the lower lip and reviewed the Japanese literature on labial pleomorphic adenomas.

Report of Case

On November 7, 1980, a 53-year-old female visited our University Dental Hospital at Matsudo with the chief complaint of swelling in the lower lip. She noticed a mass 6 months ago, but she did not seek medical care because there was no pain. However, the mass gradually grew bigger and she went to a private dentist who referred her to our hospital. On her first visit, a soy-bean sized tumor was observed in the lower right lip on oral examination (Fig. 1). The tumor was hard and elastic and the overlying mucosa appeared to be quite movable against the underlying tissue. Her general condition was not remarkable. Her past and family histories showed nothing unusual. The clinical diagnosis was adenoma. The tumor was excised under local anesthesia. She had no recurrence for 4 years after that.

Grossly, the excised material was an oval and slightly hard tumor measuring 1.0 by 0.8 by 0.6 cm. On the cut surface, it appeared solid and grayish-white with a thin capsule (Fig. 2).

Microscopically, the tumor consisted of the proliferation of polyhedral or cuboidal epithelial cells with oval or vesicular nuclei (Figs. 3 and 4). These cells formed small glands or cystic spaces containing PAS or alcian blue positive materials and revealed solid alveoli or trabeculae in some areas (Fig. 4). A few squamous metaplasia of the epithelial cells were also seen. The stroma contained about 30 % stromal content, consisting of alcian blue positive myxoid tissues

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Fig. 1 A soy-bean sized tumor is observed in the lower right lip on the first visit.

Fig. 2 The excised material is an oval and slightly hard tumor that is grayish-white on the cut surface.

Fig. 3 .Histopathology of the tumor showing epithelial proliferations surrounded by fibrous connective tissue, including normal labial glands. (Hematoxylin and eosin, $\times 40$)

associated with fibrous and hyalinized connective tissues (Fig. 4). The tumor was surrounded by thin fibrous connective tissue with normal labial glands (Fig. 3). The histopathology of the tumor was classified as pleomorphic adenoma corresponding to subtype I using the subclassification of pleomorphic adenomas by SEIFERT et al^[2].

Fig. 4. The tumor cells are composed of polyhedral and cuboidal epithelial cells which occasionally form small glands and cystic spaces. The stroma reveals myxomatous tissues. (Hematoxylin and eosin, $\times 100$)

Discussion

The occurrence rate of pleomorphic adenomas is the highest among salivary gland tumors. According to RAUCH et al.^[7], they are located 92.5 % of the time in the major salivary glands, 6.5 % in the minor salivary glands and in the others, 1 %. Among the minor salivary glands, the palatal gland is the most affected site, 67.5 %, then the lip 10 %, cheek 5 %, tongue 2.5 % and others 15 %^[4]. Thus the occurrence of minor salivary and labial gland pleomorphic adenomas is generally rare. The occurrence rate of labial pleomorphic adenomas in the minor salivary glands ranged from 2.3 to 30.4 %^[5,8-12]. There have been 33 cases of labial pleomorphic adenoma reported in Japanese literature. The Japanese cases of labial pleomorphic adenomas, including our own case, are shown in Table 1. As for the location, the upper lip was more affected than the lower lip (29 upper and 4 lower). BERNIER^[20] and KROLLS et al.^[5] stated the same tendency, ranging from 85 to 92 %. The occurrence of lower lip pleomorphic adenomas is rare, as mentioned above. For the reason for predominance of the upper lip, BERNIER^[20] gave the explanation that the developmental process was more complex in the upper than in the lower lip and resulted in an aberration of embryonal epithelium. In addition, KROLLS et al.^[5] suspected that the lower lip, being exposed to trauma such as biting and absorption, induces inflammatory changes and destroys tumor potential cells. Twelve cases were seen on the left and right sides, respectively, and 5 in the median. BERNIER^[20] reported a slight predominance of the left side. The ratio

Table 1 Pleomorphic Adenomas of the Lips in Japan

	Reference	Location	Patient's age (years), sex	Duration (years)	Size	Histological characters			
						Gland	Squamous metaplasia	Myxoid	Chondroid
1955	OHUE et al. ^[6]	Upper, Left	41, M	5	1.5×1×1 cm	+	—	+	—
1958	KATADA et al. ^[6]	Upper, Right	60, F	20	Thumb tip	+	—	+	—
1959	KITAMURA et al. ^[6]	Upper, Right	58, M	10	1.2×0.8×0.8 cm	+	—	+	—
	SUGAWARA et al. ^[6]	Upper, Left	65, M						
1961	KOBAYASHI et al. ^[6]	Lower	46, F						
1965	MASUDA et al. ^[6]	Upper, Left	15, F		Small fingertip	+	—	+	+
		Lower	25, M	6/12	Soy bean	+	+	+	—
	ISHIKURA ^[6]	Left	39, F						
1967	YOSHIDA et al. ^[6]	Upper, Right	29, M	2	1.8×1.5×1.1 cm	+	—	+	—
1969	TAKAHASHI et al. ^[6]	Upper, Left	53, M	15	1.7×1.5×1.4 cm	+	+	+	—
		Upper, Right	22, F	4	1.7×1.6×1.2 cm	+	+	—	+
	MURAGISHI et al. ^[6]	Upper, Right	42, F		Thumb tip	+	—	+	+
1971	UCHIDA et al. ^[6]	Upper, Left	66, F	1&8/12	Pigeon's egg	+	—	+	—
	TOMITA et al. ^[6]	Upper, Median	36, M	5	3×2.5×2.5 cm	+	—	+	+
1974	ASAKA et al. ^[6]	Upper, Median	20, F	2	2×1.5×1.5 cm	+	—	+	—
	TAKIGAWA et al. ^[6]	Upper, Left	17, F	6	1.5×1.5 cm	+	+	+	—
		Upper, Right	42, M	20	2.5×2×2 cm	+	—	+	—
1976	ONITSUKA et al. ^[13]	Lower, Median	3/12 F	3/12	Thumb tip	+	—	+	—
1978	YAMAMOTO et al. ^[6]	Upper, Median	29, F	5&6/12	1×0.8×0.7 cm	+	+	+	—
	MARUMO et al. ^[14]	Upper	65, M		1.5×1×1 cm	+	—	+	—
1979	YAMAGUCHI et al. ^[14]	Upper, Left	48, F	1/12	1.8×1.4×1.4 cm	+	—	+	+
	HIRAGA et al. ^[14]	Upper, Median	26, F	10	Small fingertip				
		Upper, Right	18, M	3	Small fingertip				
		Upper, Right	40, M	1	Small fingertip				
1981	MORII et al. ^[14]	Upper, Right	24, F	10	1.8×1.6×1 cm	+	+	+	+
	SUGI et al. ^[15]	Upper	59, F	2	2×2 cm				
		Upper	46, F	20	2×3 cm				
	NAKAJIMA et al. ^[16]	Upper, Left		10	Small fingertip	+	—	+	—
1982	INAGAKI et al. ^[17]	Upper, Right		10	Small fingertip	+	—	+	—
		Upper, Left		3	Index fingertip				
1983	KURODA et al. ^[18]	Upper, Left	45, M						
	FUKAYA et al. ^[19]	Upper, Right	17, F	6	Small fingertip				
1984	YAMAMOTO et al.	Lower, Right	65, F	30	Small fingertip				
			53, F	6/12	1×0.8×0.6 cm	+	+	+	—

Table 2 Differential Points between Pleomorphic Adenomas and Mixed Tumors

	Pleomorphic adenomas	Mixed tumors of the skin
Clinical Findings		
Location	Submucosa	Subcutaneous
Age	20–40 decades	30–50 decades
Sex	Male $\leq$ Female	Male $>$ Female
Duration	7.2 years	7 years
Size	ϕ 1–2 cm	ϕ 1–2 cm
Histological Findings		
Myxoid area	Many	Relatively few
Modification of nests	Many	Few
Glandular wall	Mainly double	Eccrine: single Apocrine; more than double
Solid area	Common	Few
Squamous metaplasia	Few	Many

on males to females was 1:1.7 (12 males and 20 females). However, males were more affected in other countries^[5,20]. The average age of Japanese labial pleomorphic adenomas was 38.6 years, ranging from 3 months to 66 years. Duration of the illness was from 3 months to 30 years. The average was 7.2 years. The size of the tumor was, in general, small with an average diameter of 18 mm.

SEIFERT et al.^[2] classified pleomorphic adenomas into four types; I, II_{a–e}, III and IV. In 20 cases with detailed histologic descriptions, tumors associated with the myxoid area corresponding to subtype I or II_a by SEIFERT's subclassification were more often in labial pleomorphic adenomas. Chondroid stroma was not seen as often in the labial pleomorphic adenomas as compared with that of pleomorphic adenomas in major salivary glands according to FOOTE et al.^[21] The structure of the labial gland is similar to other salivary glands except for the presence of the short duct^[22]. The reason why chondroid stromata occur less in labial pleomorphic adenomas is not very clear.

At the lip, there were not only pleomorphic adenomas in the salivary gland but also mixed tumors in the sweat glands^[6,23–25]. It is necessary to differentiate their origin from that of salivary or sweat glands because of their close anatomic relationship at the lip. The differential points between pleomorphic adenomas in the salivary gland and mixed tumors of the sweat gland are listed in Table 2 based on the reported literature^[6,23–25]. In differential clinical points of view, pleomorphic adenomas are located in the submucosa of the oral cavity, but mixed tumors of the skin are subcutaneous. The patient's age is a little higher in mixed tumors than in pleomorphic adenomas. Histopathologically, the myxoid area and modification of tumor nests are more often met in pleomorphic adenomas. Basically, the glands of pleomorphic adenomas are formed by double layers. Squamous metaplasia is more frequent in mixed tumors.

Summary

Pleomorphic adenomas of the lower lip are rare. We described a case of a pleomorphic adenoma in the lower lip of a 53-year-old female. The cases reported

in Japanese literature were reviewed. The differential points between labial pleomorphic adenomas and mixed tumors of the skin were also discussed.

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