Original Article: Case Series

Lip pleomorphic adenomas: case series and literature review

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Background: Pleomorphic adenoma (PA) is the most frequent benign salivary gland tumor, but a lip PA is rare. Although this tumor may be definitively diagnosed by imaging or a tissue biopsy if it is reasonably large, PAs on the lip are relatively small, and they present findings that are similar to those of other lip lesions, which can make a preoperative diagnosis difficult.

Methods: We analyzed all PAs in the oral region and lesions on the lips treated in our department over the past 20 years, and we discuss them together with the relevant literature.

Results: We found that 11.8% (n=6) of the PAs occurred on a lip (upper lip: 9.8%, lower lip: 2.0%), and ~1% of all mass lesions of the lips were PAs. The average size of the lip PAs was 1.5±0.7 cm (range, 0.7–2.2 cm). For preoperative diagnostic assistance, ultrasonography (US) (n=4), magnetic resonance (MR) (n=3), or no imaging (n=2) was used. An excisional biopsy was performed in all cases, and to date, no recurrence or malignant transformation has been observed.

Conclusions: Lip PA is relatively rare. Because almost all of these lesions are small, a preoperative diagnosis is more difficult compared to palatal lesions. This tumor is also prone to long-term neglect and has the potential for recurrence and malignant transformation. It is thus necessary to perform an excision that includes the capsule and surrounding tissues, and careful postoperative follow-up should be continued.

Keywords: Pleomorphic adenoma (PA); upper lip; lower lip; minor salivary gland tumor; case series

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Introduction

Minor salivary glands are independent tissues of salivary cells and are present in the mucosa of the oral cavity and throat (1,2). Unlike the major salivary glands, the minor salivary glands secrete saliva directly from the salivary gland tissue into the oral cavity without passing through a duct. Minor salivary glands are not well visualized on computed tomography (CT) or magnetic resonance (MR) imaging, but in some cases, they can be the source of neoplastic lesions (3). Most of these lesions are malignant, but benign tumors occasionally develop. The hard and soft palates are the most common sites of minor salivary gland tumors,

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Table 1 Characteristics of the patients with a PA occurring in the lip, treated at our institution over a 20-year period

Pt. no.	Age, years	Sex	Site	Size, cm	Disease period*	Imaging method	Clinical diagnosis
1	35	F	U	0.7	7 yrs	None	Benign tumor
2	68	M	U	1.3×1.3	10 yrs	US, MR	Benign tumor
3	69	M	L	1.5×2.0	3 yrs	US, MR	Minor salivary gland tumor
4	54	F	U	0.8	4 yrs	US	Minor salivary gland tumor
5	37	M	U	2.2×1.2	6 mo	US, MR	Minor salivary gland tumor
6	33	F	U	2.0	–	None	Benign tumor

*, from the time of self-awareness or suggestion by others to the time of the first visit; –, unclear. PA, pleiomorphic adenoma; Pt, patient; F, female; U, upper; yrs, years; M, male; US, ultrasonography; MR, magnetic resonance; L, lower; mo, months.

whereas the lip is a relatively rare site.

Pleomorphic adenoma (PA) is the most frequent benign salivary gland neoplasm, occurring most frequently in the parotid gland among the major salivary glands and in the palatine gland among the minor salivary glands (4,5). The occurrence of a PA in a lip is rare. This tumor is described as pleomorphic or mixed because it shows a variety of histopathologies and is composed of a mixture of glandular epithelium, myoepithelial cells, and connective tissue elements (5). PAs appear as asymptomatic firm masses with a slow growth rate, and they tend to be left untreated for a relatively long time before medical attention is sought.

In this article, we describe three rare cases of PA occurring in the lip treated at our department, and we present the results of our analysis of the relevant previous cases with a literature review. We present the following article in accordance with the AME Case Series reporting checklist (available at https://gs.amegroups.com/article/view/10.21037/gs-22-308/rc).

Methods

Study participants

The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013). The study was approved by Ethics Committee of the Okayama University Graduate School of Medicine, Dentistry and Pharmaceutical Sciences and Okayama University Hospital (approval No. 1804-015). The specimens examined in this study had been used for pathological diagnosis for treatment determination. No extra tissue was collected for this study. We obtained comprehensive informed consent from the patients to use the post-diagnosis specimens in this study.

Patients were included when they had: (I) a PA confirmed by a pathological examination, (II) high-quality images, and (III) complete clinical data. Patients without pathological results and patients whose images were poor-quality were excluded. Based on these criteria, we identified and report herein the cases of three of the total of six patients with a lip PA who visited our Department of Oral and Maxillofacial Surgery, Okayama University Hospital (Okayama, Japan) during the 20-year period from 2001 to 2021. We also analyzed the PAs in the oral region and lesions on the lips treated at our department during the same 20-year period and compared them with the existing literature regarding lip PAs.

Results

Table 1 summarizes the detailed characteristics of the six patients with a lip PA treated at our department. The average age of the patients (three males, three females) was 49.3±16.6 years (range, 33–69 years). The upper to lower lip ratio of the lesion location was 5:1, revealing a predominance of upper-lip PAs (upper: Patients 1, 2, and 4–6; lower: Patient 3). The average size of the tumors was 1.5±0.7 cm (range, 0.7–2.2 cm). The average disease period of PA was 4.9 years (range, 0.5–10 years). As an imaging method for diagnostic assistance, ultrasonography (US) was used in four cases (Patients 2–5), MR was used in three cases (Patients 2, 3, and 5), and none was used in two cases (Patients 1 and 6). As differential clinical diagnoses, benign tumor and minor salivary gland tumor were candidates. We next describe three typical cases.

Patient 1

A 35-year-old Japanese woman was referred to our

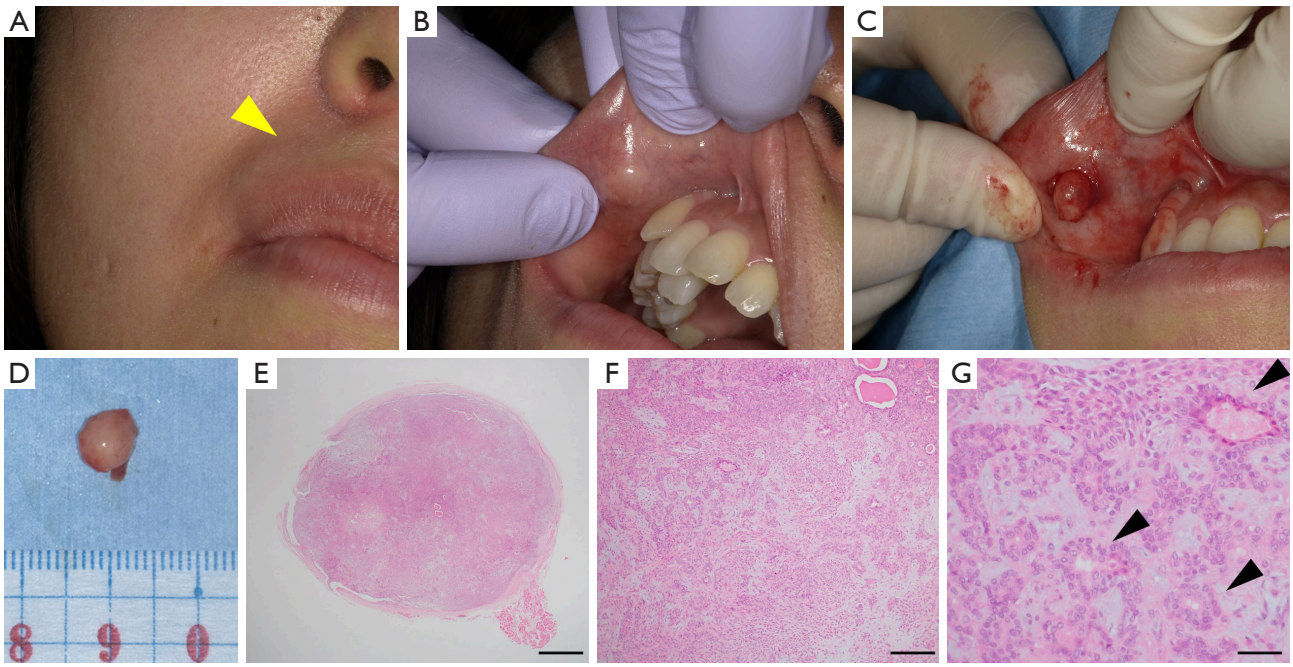

Figure 1 Clinical and histological findings of Patient 1. (A) Extra-oral findings at the first physical examination. Localized swelling above the upper lip on the right side (arrowhead). (B) Intra-oral findings at the first physical examination. A spherical mass with a smooth surface was recognized inside the right upper lip. (C) Intraoperative photograph at the excisional biopsy. (D) Macroscopic image of the resected specimen. A mass with a capsule measuring 5×7 mm. (E-G) H&E staining showing histopathologic images of the resected specimen. Both epithelial cells and myoepithelial cells were observed in the tumor area. The epithelial cells formed ductal structures, and myoepithelial cells were observed surrounding the outside of the duct. There were two layers of epithelium and myoepithelium, with myoepithelial cells proliferating to migrate into the stroma (arrowheads). (E) Gross image. The tumor was well encapsulated. Scale bar: 1 mm. (F) Low-power magnification. Scale bar: 200 µm. (G) High-power magnification. Scale bar: 50 µm. The images are published with the patient's consent.

department for the examination of a mass on her upper lip. At her first visit, the extraoral evaluation revealed a slight swelling change in her right upper lip (*Figure 1A*). She had been aware of the mass for 7 years and had taken no action because it was painless. A palpation examination showed a spherical mass measuring 7 mm in diameter with a smooth surface inside the right upper lip (*Figure 1B*). The mass was rigid, mobile, and not tender. The patient had no significant medical history or medication. We suspected a benign tumor (e.g., an irritation fibroma) and performed an excisional biopsy to establish a definite diagnosis.

Tumor resection was performed under local anesthesia (*Figure 1C*). The tumor in the upper lip was surrounded by a capsule, and the tumor was removed as a single mass including the capsule (*Figure 1D*). The histopathological examination revealed many cells with round nuclei and eosinophilic cytoplasm inside the tumor by hematoxylin and eosin (H&E) staining (*Figure 1E-1G*). The resected tumor was encapsulated by a thin fibrous coating (*Figure 1E*). The tumor was composed of variable epithelial and myoepithelial/stromal components in a mixture of patterns (*Figure 1F,1G*). The epithelial cells formed ductal structures, and myoepithelial cells were observed surrounding the outside of the duct (*Figure 1G*). Based on these histopathological findings, we finally diagnosed the PA in the upper lip. We have been conducting a regular follow-up for 6 months, and there has been no sign of local recurrence.

Patient 2

A 68-year-old Japanese man was referred to our department with an enlarged mass on his upper lip. He had been aware of the mass for ~10 years and had taken no action because it was painless. At his first visit, the physical examination showed a movable mass measuring approximately 10 mm in diameter with a smooth surface inside the left upper lip. The mass was rigid and not tender. The patient had no

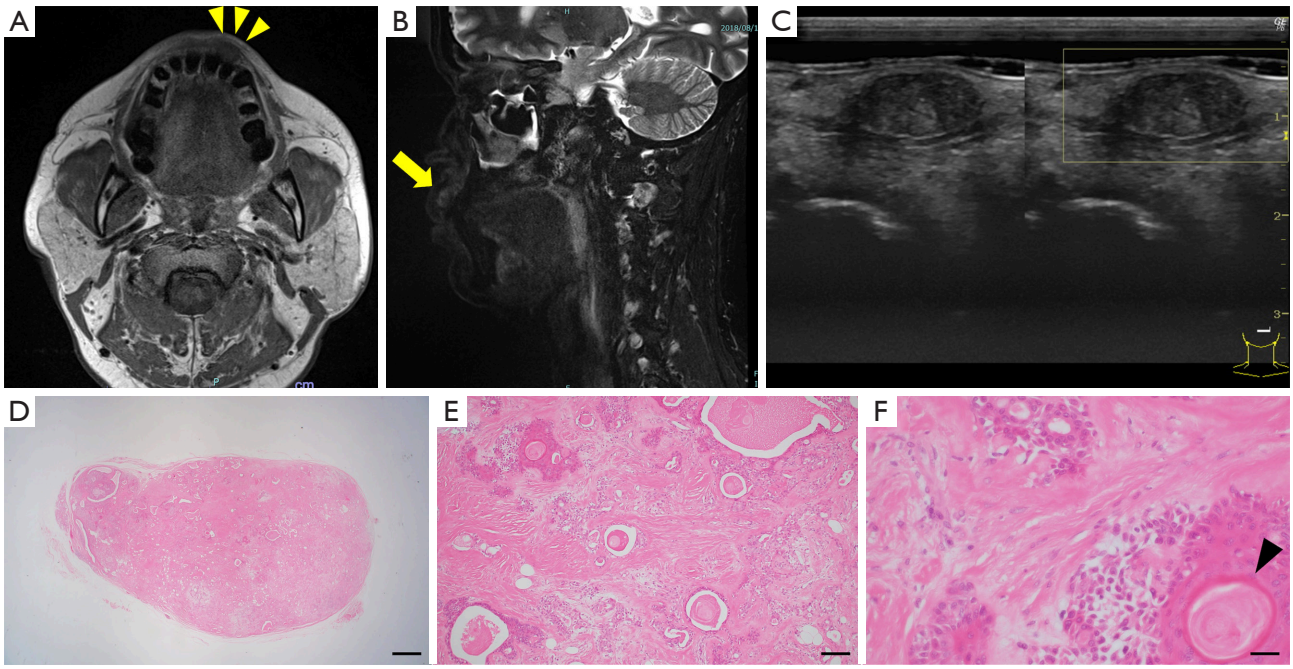

Figure 2 Imaging and histological findings of Patient 2. (A) Axial T1-weighted MR image showing the mass with mid-level signal intensity (arrowheads). (B) Sagittal STIR MR image showing the mass with low-to-high signal intensity (arrow). (C) US showing the circular mass with no internal blood flow. (D-F) H&E staining showing histopathologic images of the resected specimen. Both epithelial cells and myoepithelial cells were observed in the tumor area, and they had a bilayer structure. Fibrous stroma was found. Some of the epithelial cells had squamous metaplasia (arrowhead). (D) Gross image. Scale bar: 1 mm. (E) Low-power magnification. Scale bar: 200 μm. (F) High-power magnification. Scale bar: 50 μm. MR, magnetic resonance; STIR, short T1 inversion recovery; US, ultrasonography.

significant medical history.

MR imaging revealed mid-level signal intensity in the left upper lip on the T1-weighted image (*Figure 2A*) and low-to-high signal intensity on the short T1 inversion recovery (STIR) image and fat-suppressed T1-weighted image (*Figure 2B*). The border between the tumor and the surrounding area was almost clear, but partially uneven. US revealed a well-defined circular area with no internal blood flow (*Figure 2C*). Based on these results, we suspected a clinically benign salivary gland tumor and planned an excisional biopsy to establish a definite diagnosis at the patient's request.

Tumor resection was performed under local anesthesia. The histopathological examination of the resected tumor demonstrated that the tumor was not encapsulated completely by a thin fibrous tissue (*Figure 2D*). The tumor was composed of variable epithelial and myoepithelial/stromal components in a mixture of patterns. There were ductal structures inside the tumor and myoepithelial component proliferations around the structures. Spindled myoepithelial cells streamed from ductal elements into the fibrous stroma. Some of the epithelial cells had squamous metaplasia (*Figure 2E*,*2F*). Based on these histopathological findings, we diagnosed a PA in the upper lip. There has been no sign of local recurrence to date, 3 years after the tumor's resection.

Patient 3

A 69-year-old Japanese man was referred to our department with an enlarged mass on his lower lip. He had been aware of the mass for ~3 years, and had taken no action because he felt that it was small and inconsequential. At the patient's first visit, the physical examination showed a movable mass measuring approx.; 20 mm in diameter with a smooth surface inside the left lower lip (*Figure 3A*). The patient had hepatitis B virus and was being treated for it.

MR imaging revealed uneven signal intensity on the T1-weighted image and high signal intensity on the STIR image in the left lower lip (*Figure 3B*,*3C*). US revealed that the border between the tumor and the surrounding area was clear, and part of the border was lobular-shaped (*Figure 3D*). We suspected a minor salivary gland tumor and performed

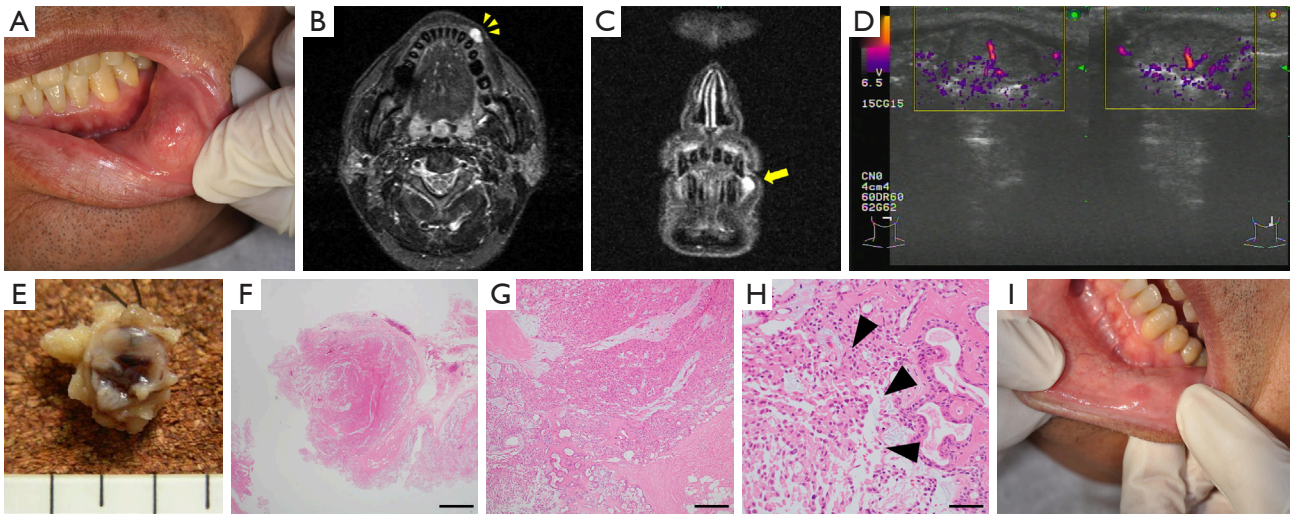

Figure 3 Clinical, histological, and imaging findings of Patient 3. (A) Intra-oral findings at the first physical examination. A mass with irregularities and a smooth surface was recognized inside the left lower lip. Axial (B) and coronal (C) STIR MR images showing the mass with high signal intensity (arrowheads and arrow). (D) US showing a well-defined but partly lobulated mass with poor internal blood flow. (E) Macroscopic image of the resected specimen. (F-H) H&E staining showing histopathologic images of the resected specimen. Both epithelial cells and myoepithelial cells were observed in the tumor area, and they had a bilayer structure. Plasmacytoid cells were observed (arrowheads). (F) Gross image. The surgical margin was unclear. Scale bar: 1 mm. (G) Low-power magnification. Scale bar: 200 µm. (H) High-power magnification. Scale bar: 50 µm. (I) Intra-oral findings at a follow-up after the tumor resection. There were no signs of recurrence, and good healing progress was observed. The images are published with the patient's consent. STIR, short T1 inversion recovery; MR, magnetic resonance; US, ultrasonography.

an excisional biopsy to establish the definite diagnosis.

Tumor resection was performed under local anesthesia. The resected tumor in the lower lip was resected as a single mass, but its morphology was irregular (*Figure 3E*). The histopathological examination of the resected tumor revealed that the tumor was surrounded by a thin fibrous coating, but the surgical margin was unclear (*Figure 3F*). The tumor was composed of variable epithelial and myoepithelial/stromal components, and plasmacytoid structures were also observed (*Figure 3G,3H*). Based on these histopathological findings, we diagnosed a lower-lip PA. At the time of follow-up 2 weeks after the excisional biopsy, the healing process of the wound was good (*Figure 3I*). There has been no sign of local recurrence to date.

PAs and lip lesions experienced in our department

Over the past 20 years, our department has experienced a total of 51 cases of PA, of which six (11.8%) were on the lips (*Figure 4*). Of the six cases of lip PAs, five (9.8%) were in the upper lip and the other (2.0%) was in the lower lip (*Table 1*, *Figure 4*). Our department has encountered a total of 590 cases of mass lesions of the lips in the past 20 years, of which only approx.; 1% were resected as PAs (*Figure 4*).

The present investigation is limited by two factors: (I) the patient series included only individuals who underwent oral surgery (otolaryngology and plastic surgery patients were not included), and (II) the target disease was only resected lesions (untreated lesions and lesions that were treated only medically were excluded). The patient series being drawn from a single institution also necessitates further research involving a larger number of patients.

Discussion

Among all salivary gland tumors, PA is the most common, occurring in 60–70% of cases, with 84% in the parotid gland, 8% in the submandibular gland, 0.5% in the sublingual line, 6.5% in minor salivary glands, and 1% in others (6). PA of the minor salivary glands occurs in the palate (67.5%), lips (10%), cheeks (5%), tongue (2.5%), and other sites (15%); PA in a lip is relatively rare (7).

Although the cases of lip PA at our department accounted for a relatively large proportion compared to

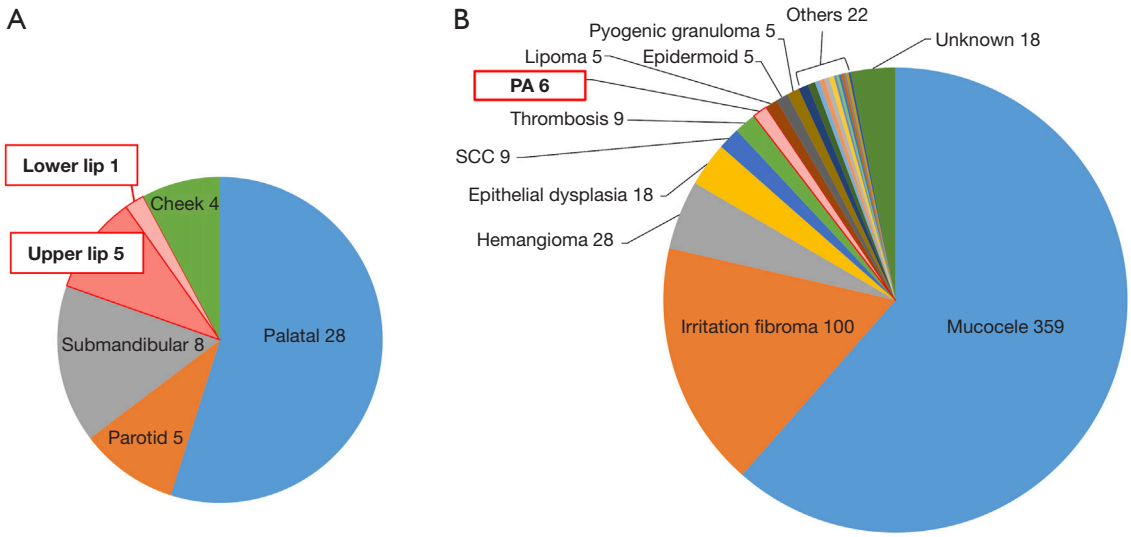

Figure 4 Proportion of lip PAs encountered in our department. (A) Pie chart of all PA cases by site. The number next to the site's name = the number of cases. (B) Pie chart of all mass lesions in lips by disease. The number next to the disease's name = the number of cases. PA, pleiomorphic adenoma; SCC, squamous cell carcinoma.

the above reports, it should be noted that many cases of PA in a parotid gland were referred to otolaryngology (only five cases of PAs of the parotid gland were treated at our department). In a comparison of the frequency of PA occurrence in the upper and lower lips, Bernier reported that of 38 cases, 35 (92.1%) were in the upper lip and three (7.9%) were in the lower lip (8). Krolls and Hicks described that of 4,042 cases of PA, 445 originated from minor salivary glands, of which 16.9% were located in the upper lip and 2.9% in the lower lip (9). The peak incidence of PA in lips was in the third and fourth decades of life, with most of the cases occurring on the upper lip (8-13).

We conducted a search of the literature on lip PAs (in Japan and worldwide) and found 35 cases described in 31 reports since 2000 (*Table 2*) (10,11,14-17,19-22,26-39,41). Of the 35 past cases of lip PA, 30 (85.7%) were in the upper lip (10,11,13,15,17,19-21,23-26,28-30,32-41) and only five (14.3%) were in the lower lip (14,16,22,27,31). The male-to-female ratio of the lip PAs was 4:3 (a nonsignificant difference), and the age range was from 10 to 72 years (*Table 2*).

Krolls and Hicks suggested that the difference in the frequency of PA between the upper and lower lips is due to the fact that, from an embryological point of view, the upper lip is formed by the fusion of three protuberances, whereas the lower lip is formed by the fusion of two protuberances, and thus embryonic cells are more likely to stray into the upper lip than the lower lip (9). Another potential reason is the difference in the number and distribution of labial glands between the upper and lower lips (3,42). Specifically, the upper labial glands are densely located between and scattered outside the corners of the mouth, while the lower labial glands are scattered between and densely located outside the corners of the mouth (3,42). The upper lip thus has a large number of well-developed labial glands, whereas the lower lip has only a few small labial glands, which may also contribute to the difference in the frequency of upper- versus lower-lip PAs (3,42).

PAs, especially of the lips, tend to be left untreated for a relatively long period of time before medical attention is sought, because they develop slowly and are rarely accompanied by ulceration or pain (8). One of our patients had been aware of the mass for ~10 years prior to the first visit, but it had been left untreated (*Figure 1*). One of the reasons why PAs of the lips are left untreated is that they tend to be smaller than those at other sites (43). For example, the PA of Patient 1 was 5×7 mm in dia., which is smaller than the reported PAs (10–160 mm in dia.; mean, 26 mm) (*Figure 1*, *Table 2*).

The clinical differential diagnosis of swelling of the lip includes cystic diseases (e.g., mucocele, dermoid cyst, epidermoid cyst), benign tumors (e.g., fibroma, hemangioma, lipoma, schwannoma), and malignant tumors [e.g., squamous cell carcinoma (SCC), mucoepidermoid carcinoma, adenoid cystic carcinoma], as well as foreign

Table 2 Reported cases of lip PA since 2000

No.	Authors	Year	Age	Sex	Site	Size, cm	Disease period*	Imaging method	Clinical diagnosis
1	To *et al.*, (14)	2002	25	M	L	1.0	3 yrs	None	Benign minor salivary gland tumor
2	Jorge *et al.*, (15)	2002	15	F	U	1.0	2 mo	None	Benign mesenchymal neoplasm, neuroma, neurofibroma
3			18	F	U	1.0	1 yr	None	Benign mesenchymal neoplasm, neuroma, neurofibroma
4	Lotufo *et al.*, (10)	2008	12	M	U	2.0	1 yr	None	Benign minor salivary gland tumor, lipoma
5	Moritani *et al.*, (16)	2008	72	F	U	1.1×1.5×1.5	1 yr	US	Benign tumor
6			60	M	L	1.3	10 mo	US, MR	Benign tumor
7	Asuquo *et al.*, (17)	2009	50	F	U	16	2 yrs	None	–
8			40	F	U	4.0×3.0	10 yrs	None	–
9	McNamara *et al.*, (18)	2009	55	F	U	1.0	30 yrs	None	–
10	Debnath & Adhyapok, (19)	2010	55	F	U	1.5–2.0	1 yr	None	Benign minor salivary gland tumor, lipoma
11	Shrestha *et al.*, (20)	2010	27	F	U	4.5–5.0	3 yrs	None	Benign minor salivary gland tumor
12	Ali *et al.*, (21)	2011	33	M	U	3.0	1 yr, 6–7 mo	CT	Granuloma, benign minor salivary gland tumor
13	Kataria *et al.*, (11)	2011	65	F	U	2.0×1.5	2 yrs	None	PA
14	Sengul *et al.*, (22)	2011	49	M	L	1.5×0.7	–	None	–
15	Dyalram *et al.*, (23)	2012	72	M	U	2.2×2.4	5 yrs	MR	PA
16	Mitate *et al.*, (24)	2013	55	M	U	“little finger”	8 yrs	MR	PA
17	Mariano *et al.*, (25)	2013	69	M	U	2.0×2.0	4 yrs	None	PA, canalicular adenoma
18	Tzermpos *et al.*, (26)	2014	39	F	U	1.0×0.7	3 yrs	None	Periapical granuloma, periapical cyst
19	Sood *et al.*, (27)	2014	46	M	L	1.5×1.2	2 yrs	None	Lipoma, sebaceous cyst
20	Fomete *et al.*, (28)	2015	37	F	U	4.0×3.0×2.0	4 yrs	None	–
21	Singh *et al.*, (29)	2015	55	M	U	2.0×1.5	1 yr	None	Benign minor salivary gland tumor, mesenchymal tumor
22	Khan *et al.*, (30)	2016	60	M	U	3.0×4.0	8 yrs	None	–
23	Taniguchi *et al.*, (31)	2016	72	F	L	1.0×1.5	7 yrs	MR	Benign tumor
24	Metgud *et al.*, (32)	2016	30	M	U	1.0×1.0	5–6 mo	None	Lipoma, sebaceous cyst
25	Fatahzadeh *et al.*, (33)	2017	58	M	U	1.5	“many yrs”	–	–
26	Ahmedi *et al.*, (34)	2017	10	F	U	2.0×1.5	3 yrs	None	Lipoma
27	Alves *et al.*, (35)	2018	18	M	U	3.0	1 yr	None	–
28	Taiwo *et al.*, (36)	2018	33	M	U	4.0	3 yrs	None	Lipoma
29	Yoshimura *et al.*, (37)	2018	52	M	U	1.5	1 yr	CT, US	Benign tumor
30	Bhatia, (13)	2019	23	F	U	1.6×1.8	3 mo	CT	PA

Table 2 (*continued*)

Table 2 (*continued*)

No.	Authors	Year	Age	Sex	Site	Size, cm	Disease period*	Imaging method	Clinical diagnosis
31	Nourwali & Dar-odeh, (38)	2019	26	M	U	2.3×2.2×1.8	2 yrs	US	Mesenchymal tumor (fibroma), PA, sialolithiasis
32	Kazikdas *et al.*, (39)	2020	20	M	U	4.0×3.0	2 yrs	CT	Benign mixed salivary gland tumor
33	Shome *et al.*, (40)	2020	25	F	U	1.5×2.0	2 yrs	None	Benign salivary gland neoplasm
34	Adiyodi *et al.*, (41)	2020	44	M	U	3.0×3.0	5 yrs	None	Peripheral giant cell granuloma, minor salivary gland tumor, lipoma
35			44	M	U	4.0×3.0	5 yrs	None	Peripheral giant-cell granuloma, minor salivary gland tumor, lipoma
36	Umemori *et al.* (present)	2022	35	F	U	0.7	7 yrs	None	Benign tumor
37			68	M	U	1.3×1.3	10 yrs	US, MR	Benign tumor
38			69	M	L	15×20	3 yrs	US, MR	Minor salivary gland tumor

*, from the time of self-awareness or suggestion by others to the time of the first visit; –, unclear. PA, pleiomorphic adenoma; M, male; L, lower; yr(s), year(s); F, female; U, upper; mo, months; US, ultrasonography; MR, magnetic resonance; CT, computed tomography.

body stray, infection, orofacial granulomatosis, Quincke's edema, tuberculosis, and actinomycosis (13,26,29).

For the diagnosis of soft tissue lesions in the oral cavity, MRI and US are effective for confirming the location, size, nature, and morphology of the tumor (44,45). A preoperative diagnosis is particularly important for primary salivary gland tumors of the labial gland, as malignancy is observed in 20–30% of cases (46,47). However, in many cases, it is difficult to preoperatively diagnose minor salivary gland tumors, especially in the lip, as malignant tumors (48). The reasons for this are the difficulty of image evaluation and the uncertainty of obtaining biopsy findings due to the small size of the lip lesion. Although the masses we observed in Patients 1–3 were palpable and had no adhesions to surrounding tissues, and since the long-term courses of the lesions ruled out malignancy, it is useful to conduct imaging and histological examinations in order to establish a differential diagnosis, even when the tumor is clinically diagnosed as benign.

In addition, because an incisional biopsy involves incision into the capsule and the possibility of cell seeding, surgical treatment with the assumption of removal is considered useful. Notably, many PA lesions on the lips are small in size, and if the resection area is larger than necessary, it may cause postoperative scar contracture, deformity of the lips due to tissue loss, and functional impairment. Therefore, if there are no adhesions or infiltrations with surrounding tissues, total removal as the biopsy procedure is better than over-excision including surrounding healthy tissues.

A malignant transformation of a PA may occur when the surgical resection of a benign PA is incomplete (49,50). In addition, failure to remove the entire capsule increases the possibility of local recurrence and requires long-term follow-up (5,18,50). A review of 31 reports since 2000 (*Table 2*) revealed that of a total of 35 resected lip PAs, a histopathologic examination enabled the diagnosis of carcinoma ex pleomorphic adenoma (CXPA) in five patients (14%) (18,23-25,40). In one of these patients, additional resection was performed (18), and the remaining four patients were followed up cautiously, considering that they were completely resected (23-25,40). In all 35 patients, there were no recurrences during the follow-up period (including those not described) (*Table 2*).

In two of our three present patients, we performed only removal with complete inclusion of the capsule, but in the other patient, the capsule was incompletely resected. However, fortunately, to date there has been no recurrence, no deformity of the lips, and no functional impairment in any of the patients. A few reports mentioned a postoperative recurrence of PA, but not much has been published regarding a specific postoperative policy. The recurrence rate of PA is 2–8% (51); some rare recurrences have been identified within a few years after surgery, and some clinicians believe that 5–10 years of observation is not sufficient. For example, in a study by Valstar *et al.* the 20-year overall recurrence rate of PA was 6.7%, with a

median time to first recurrence of 7 years (52).

Few previous PA reports have included a rigorous, long-term follow-up protocol with periodic imaging. PA recurrences rarely cause clinical symptoms such as pain or neurological symptoms, and without periodic imaging during follow-up, the recurrences may remain undetected for a long period of time. Schapher *et al.* showed that US, a routine examination, allowed the early detection of recurrence even before clinical symptoms appeared (53). Although there is no set view on follow-up intervals, we will continue the long-term follow-up of our PA patients while taking into consideration the characteristics of PAs, which may reoccur years after surgery.

Conclusions

PA is one of the most frequently occurring salivary gland tumors, but it rarely occurs in minor salivary glands and is especially rare in the lips. PAs show a wide variety of pathologies and histologies, and it may not be possible to make a diagnosis of benign or malignant status based on imaging findings or preoperative biopsy materials alone; in addition, for lip PAs, imaging studies and a biopsy may be difficult to perform. Clinicians should consider the possibility of malignancy and postoperative recurrence and perform a resection that includes the capsule and surrounding tissue. It is also necessary to continue a careful follow-up for signs of recurrence.

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Footnote

Reporting Checklist: The authors have completed the AME Case Series reporting checklist. Available at https://gs.amegroups.com/article/view/10.21037/gs-22-308/rc

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Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://gs.amegroups.com/article/view/10.21037/gs-22-308/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013) and was approved by the Ethics Committee of the Okayama University Graduate School of Medicine, Dentistry and Pharmaceutical Sciences and Okayama University Hospital (approval No. 1804-015). The specimens examined in this study had been used for pathological diagnosis for treatment determination. No extra tissue was collected for this study. We obtained comprehensive informed consent from the patients to use the post-diagnosis specimens in this study.

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