



ORIGINAL ARTICLE

Sialolithiasis of minor salivary glands: A review of 17 cases



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Abstract *Background/purpose:* To our knowledge, sialolithiasis in minor salivary glands is very rare, and information about the disease is limited. The current study aimed to provide updated data regarding the disease in Taiwan. The data were compared with those of previous case series studies.

Materials and methods: The features of 17 cases of histopathologically confirmed sialolithiasis in minor salivary glands between 1991 and 2015 in our institution were retrospectively analyzed. *Results:* Most of the patients were male ($n = 14$; 82.35%), with only three female patients (17.65%). The mean age of the 17 patients was 62.93 years (range, 35–82 years). Fifteen cases (~88%) were found within the 6th–9th decades. Seven cases (~41%) were identified in patients aged ≥ 70 years, six of which had been diagnosed in the most recent 5 years (2011–2015). The most common site was the buccal mucosa ($n = 7$; 41.18%), followed by the upper lip ($n = 5$; 29.41%), lower lip ($n = 3$; 17.65%), and vestibule and retromolar area (each $n = 1$; 5.88%). Only one case (5.88%) was clinically diagnosed as sialolithiasis prior to biopsy examination.

Conclusion: The current study demonstrated an aging tendency and a male predilection of sialolithiasis in minor salivary glands in Taiwan when compared with published case series studies. Copyright © 2015, Association for Dental Sciences of the Republic of China. Published by Elsevier Taiwan LLC. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

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Introduction

Sialolithiasis, which usually causes duct obstruction, inflammation, or a painful infection, is a common disease of the major salivary glands (SGs). On the other hand, the symptoms of this disease in minor SGs may be painless or may manifest as mild tenderness, which is easily ignored. To the best of our knowledge, since the first case reported by Papin¹ in 1865, there have been approximately 270 cases of sialolithiasis of minor SGs reported in English-language literature^{1–30}; however, only five case series studies on sialolithiasis of minor SGs are available.^{8,11–13,29}

To provide updated findings regarding sialolithiasis of minor SGs in Taiwan, the current study aimed to evaluate the characteristics of sialolithiasis of minor SGs histopathologically diagnosed by the oral pathology and maxillofacial radiology department of a teaching hospital in southern Taiwan. Additionally, the findings were compared with those of the five previous case series reports in the literature.^{8,11–13,29}

Materials and methods

A total of 17 cases of sialolithiasis of minor SGs confirmed by histopathological examination, as indicated in Figure 1, were retrieved from the database of the oral pathology and maxillofacial radiology department between January 1991 and January 2015. The age, year of diagnosis, sex, clinical impression, and treatment of the 17 cases were

retrospectively analyzed and compared with those in the published case series studies.^{8,11–13,29}

Results

A total of 72 cases of sialolithiasis were identified in the database of the oral pathology and maxillofacial radiology department, 17 of which occurred in minor SGs (23.61%). The features of these 17 cases, including age, year of diagnosis, gender, clinical impression, and treatment, are summarized in Table 1.

Briefly, the mean age of the 17 patients was 62.93 years (range, 35–82 years). Most cases ($n = 15$; 88.23%) were identified within the 6th–9th decades; seven patients (41.18%) were aged ≥ 70 years, six of whom had received a diagnosis in the most recent 5 years (2011–2015). The patients were predominantly male ($n = 14$; 82.35%), with only three female patients (17.65%). The most common location was the buccal mucosa ($n = 7$; 41.18%), followed by the upper lip ($n = 5$; 29.41%), lower lip ($n = 3$; 17.65%), and vestibule and retromolar area (each $n = 1$; 5.88%). Only 1 of the 17 cases (5.88%) was clinically diagnosed as sialolithiasis prior to biopsy; the clinical impressions of the remaining cases were fibroma ($n = 4$; 23.53%), oral cancer or oral precancerous lesions ($n = 4$; 23.53%), mucocele ($n = 3$; 17.65%), minor salivary gland tumor ($n = 3$; 17.65%), and inflammation ($n = 2$; 11.76%). Fourteen of the 17 patients (82.35%) had undergone surgical excision.

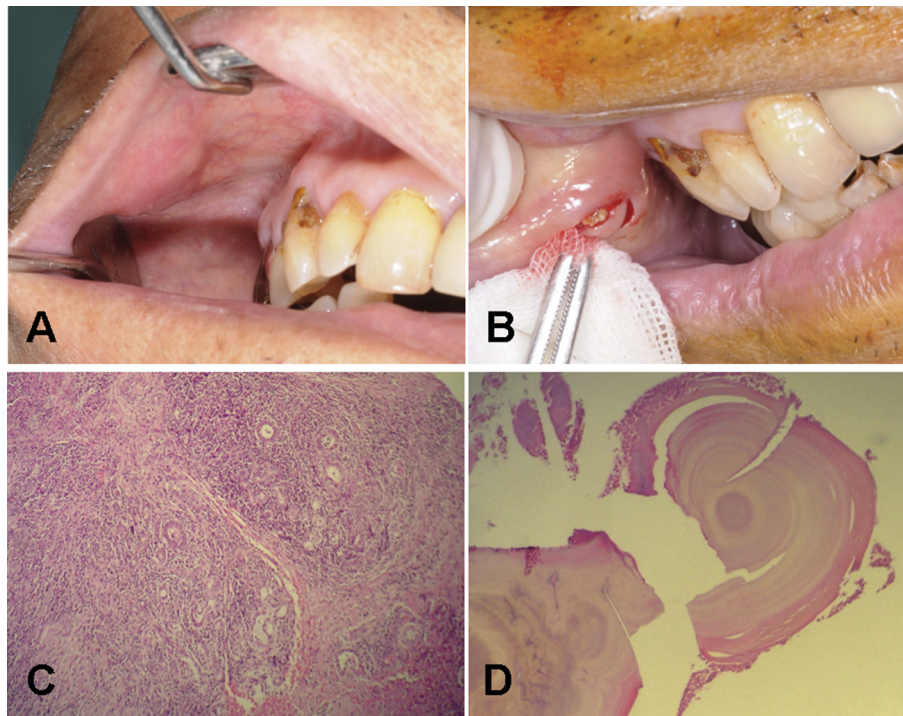


Figure 1 Clinical and pathological presentation of Case 17. (A) An 82-year-old male found a firm mass over his right upper lip. (B) An excisional biopsy was performed with the clinical impression of a salivary gland tumor, and yellowish hard tissue was found in the lesion during the operation. Microscopically ($\times 100$, hematoxylin–eosin stain), it was characterized by (C) inflammatory minor salivary gland tissue and (D) laminated oval-shaped calcified tissue.

Table 1 Summary of the characteristics of 17 cases of sialolithiasis of minor salivary glands.

Case no.	Y of diagnosis	Age (y)	Sex	Site	Clinical impression	Treatment
1	1994	74	Male	Retromolar	Verrucous hyperplasia	Incision
2	1999	35	Male	Upper lip	Fibroma	Excision
3	2000	59	Male	Buccal	Suspected squamous cell carcinoma	Excision
4	2000	55	Female	Vestibule	Fibroma	Excision
5	2004	56	Male	Buccal	Dermoid cyst	Excision
6	2006	55	Male	Lower lip	Mucocele	Excision
7	2007	55	Female	Buccal	Mucocele	Excision
8	2008	59	Male	Lower lip	Sialolithiasis	Curettage
9	2009	54	Female	Buccal	Fibroma	Excision
10	2009	49	Male	Buccal	Leukoplakia	Excision
11	2011	72	Male	Upper lip	Suspected pleomorphic tumor	Excision
12	2012	70	Male	Buccal	Inflammatory fibrous tissue	Excision
13	2013	79	Male	Upper lip	Mucocele	Excision
14	2013	71	Male	Upper lip	Squamous cell carcinoma	Excision
15	2014	73	Male	Lower lip	Mucocele	Excision
16	2014	55	Male	Buccal	Inflammation	Incision
17	2015	82	Male	Upper lip	Salivary gland tumor	Excision

Discussion

Ben Lagha et al³⁰ reported that only 2% of sialolithiasis cases develop in minor SGs and the sublingual glands. However, about 24% of all lithiasis cases in our database occurred in minor SGs. This may be due to the fact that most lithiasis cases of the major SGs, especially those of the parotid glands, are treated by ear, nose, and throat specialists, with the excised specimens examined by general pathologists. On the other hand, nearly all intraoral minor SG specimens would be examined by the oral pathology department of the institution. Therefore, the data regarding sialolithiasis of minor SGs presented in the current study are representative of the occurrence of such lesions among the cohort of patients for whom a biopsy procedure is performed within the institution. Nevertheless, the medical pathology department would possibly contribute a very small number of cases of minor SGs. So, the data regarding sialolithiasis of minor SGs

derived from the present study may not represent a precise estimation of the frequency of the disease in minor SGs in the institution.

The most frequent sites of occurrence of sialolithiasis of minor SGs were the buccal mucosa and the upper lip (Table 2), consistent with previous case series studies.^{8,11–13,29} On the other hand, according to a review of English-language literature, with the exception of Jensen et al,¹¹ an almost equal gender distribution or a slight male predominance has been noted in previous case series studies of sialolithiasis of minor SGs,^{8,12,13,29} which contradicts our findings, in which the percentage of male patients was as high as 82%.

The mean age of the patients in the present study was about 10 years older than the mean ages reported in the previous case series studies of Pullon and Miller,⁸ Anneroth and Hansen,¹² and Yamane et al.¹³ Additionally, seven (~41%) of the patients in the current study were aged ≥ 70 years, six of whom had been diagnosed in the most

Table 2 Comparison of case series of sialolithiasis of minor salivary glands.

Case series (y of publication)	No. of cases	Male–female ratio	Mean age (y)	Most common location (no. of cases)	Correct diagnosis rate (%)
Pullon & Miller ⁸ (1972)	55 (45 reviewed; 10 new)	2.6:1	51	Buccal (24), upper lip (22)	0 (for new cases)
Jensen et al ¹¹ (1979)	47	1:1.3	58.4	Upper lip (22), buccal (19)	Not available
Anneroth & Hansen ¹² (1983)	49	1.45:1	54.5	Upper lip (23), buccal (15)	20
Yamane et al ¹³ (1984)	55	1.05:1	49 (male) 48 (female)	Upper lip (37), buccal (26)	Not available
Brazao-Silva et al ²⁹ (2015)	25	1.27:1	58.7	Upper lip (11), buccal (9)	8
Wang et al (present study, 2015)	17	4.6:1	62.9	Buccal (7), upper lip (5)	6

recent 5 years. Taken together, this may indicate that perhaps due to geographic distribution, there has been an aging tendency for sialolithiasis of minor SGs in Taiwan. Moreover, to our knowledge, this is the first case series report on histopathologically confirmed sialolithiasis of minor SGs in Taiwan. Interestingly, the case identified in the present study that occurred in the retromolar area is, to our knowledge, the first case to be noted in that area.

Accurate clinical diagnosis of sialolithiasis of minor SGs is difficult, with studies showing 0%,⁸ 6% (current study), 8%,²⁹ and 20%¹² consistency between clinical impressions and histopathological diagnoses (Table 2). This may be attributed to the very small size of the lesions, which therefore cannot be easily detected by computed tomographic or ultrasonic examinations. So, surgical excision, as demonstrated in the present study, is the preferred method by which to confirm the diagnosis.²⁴ For a clinically palpable mass with a hard consistency within the region of the distribution of minor SGs, low-dose intraoral radiography may be helpful to detect the possibility of a radiopaque image, suggesting that clinical differential diagnoses include sialolith, phlebolith, tonsilolith, osseous choristoma, and pleomorphic adenoma.

In conclusion, most cases of sialolithiasis of minor SGs in the current study were found within the 6th–9th decades, with about 41% of patients aged ≥ 70 years. Moreover, it occurred predominantly in male patients and most frequently in the buccal mucosa and the upper lip.

Conflicts of interest

The authors have no conflicts of interest relevant to this article.

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