

Diagnosis and treatment of 64 cases of sublingual gland cysts

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Research Article

Keywords: Sublingual gland cyst, Intraoral type, Extraoral type, Sublingual gland excision

Posted Date: July 16th, 2026

DOI: <https://doi.org/10.21203/rs.3.rs-9844065/v1>

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Additional Declarations: No competing interests reported.

Version of Record: A version of this preprint was published at BMC Oral Health on October 17th, 2026.
See the published version at <https://doi.org/10.1186/s12903-026-10224-6>.

Abstract

Objective: Sublingual gland cysts are caused by the leakage of mucus within the sublingual gland. It is a retention cyst. The author collected cases admitted in the past four years for a retrospective study, analyzing its diagnostic methods and therapeutic effects.

Method: Collect patients who were diagnosed with sublingual gland cysts and underwent inpatient surgery at the Department of Oral and Maxillofacial Surgery of Nantong First People's Hospital from January 2022 to December 2025. Conduct statistical analysis on their gender, age, diagnosis and treatment outcomes.

Result: A total of 64 patients met the criteria, including 22 males and 42 females. There were 57 cases of intraoral type and 7 cases of extraoral type. All the intraoral cases were diagnosed preoperatively through clinical examination, while the extraoral cases were diagnosed by CT (1 case), MR (5 cases), and ultrasound (1 case). Sixty-two patients underwent intraoral approach sublingual gland and cyst excision, with an average operation time of (51.84 ± 20.27) minutes. One extraoral case underwent submandibular approach sublingual gland, submandibular gland, and cyst excision, and one case underwent bag-type suture. The postoperative follow-up period ranged from 6 months to 4 years. Postoperative complications included 5 cases of tip of tongue numbness, 2 of which persisted for 1 year after surgery, and 3 cases recovered within 3 months. One patient had significant bleeding from the floor of the mouth 3 hours after surgery and recovered normally after a second operation. There was no local infection, and one intraoral case recurred 3 months after surgery.

Conclusion: Intraoral type of sublingual gland cyst is more common, and the diagnosis method is clear. The diagnosis and treatment of extraoral type pose challenges to clinical practice. Although complete excision of the sublingual gland is currently the preferred treatment option, there is still a certain probability of complications.

Introduction

Sublingual gland cysts are caused by the leakage of mucus from the sublingual gland. They are a type of true obstructive cyst, with an incidence rate of less than 1-10%, accounting for 6% of all oral salivary gland cysts. This lesion is commonly seen in children and adolescents, and reaches its peak in the second decade. However, a large pediatric study conducted in China in 2016 indicated that this condition can occur at any age [1]. Based on clinical symptoms, it is classified into three types: oral type (superficial type), extraoral type (neck type, deep type, drooping type), and mixed type (a combination of the first two types) [2]. Among them, the extraoral type of sublingual gland cysts is relatively rare. Excessive saliva leakage can extend to the base of the skull and spread downward through the soft tissues of the neck to the supraclavicular area, even entering the mediastinum [3], and sometimes it may spread towards the parapharyngeal space [4]. In order to understand the basic situation of sublingual gland cysts admitted to our hospital, the authors collected data, conducted case follow-ups, and performed statistical analysis, and summarized to find better diagnostic and treatment plans.

Clinical data

Collect hospitalized patients who were diagnosed with sublingual gland cysts at the Department of Oral and Maxillofacial Surgery of Nantong First People's Hospital within the past 4 years (from January 2022 to December 2025). Collect their age, gender, medical history, preoperative examinations and diagnoses, as well as the surgical and postoperative recovery situations. Conduct telephone follow-ups to exclude patients who were lost to follow-up. Perform statistical analysis on the collected valid data. This study was approved by the Ethics Committee of Affiliated Nantong Clinical College of Nantong University (Nantong First People's Hospital) (2025-KT242-01), and this study obtained informed consent from all participants. The author has obtained informed consent from the parents or legal guardians of participants under the age of 16.

Results

A total of 64 patients met the criteria, including 22 males and 42 females, with a male/female ratio of 0.52. The average age was (40.61 ± 19.81) years, with the youngest being 5 years old and the oldest 81 years old. The average duration of medical history was (16.05 ± 37.98) weeks, the shortest being 1 week and the longest 260 weeks. There were 57 cases of intraoral type and 7 cases of extraoral type, with 29 cases on the right side and 35 cases on the left side. The size of the lesion was (1.88 ± 2.05) cm^3 , with the largest being 9 cm^3 and the smallest 0.09 cm^3 . 49 patients had not received any treatment before the visit, and 15 patients had a varying duration of self-administered antibiotic treatment. The intraoral type were diagnosed preoperatively through clinical examination, while the extraoral type were diagnosed through CT (1 case), MR (5 cases), ultrasound (1 case), or aspiration of viscous fluid, resulting in a clear diagnosis. 62 patients underwent intraoral approach sublingual gland + cyst excision surgery for treatment. The operation duration was (51.84 ± 20.27) minutes. One extraoral type underwent mandibular approach sublingual gland + submandibular gland + cyst excision surgery, and one underwent bag-type suture. The postoperative follow-up lasted for up to 4 years, with the shortest being 5 months. Postoperative complications included numbness of the tongue in 5 cases, with 3 cases recovering tongue sensation within 3 months after the operation, and 2 cases not fully recovered during the 1-year follow-up. One patient had massive bleeding in the incision 3 hour after the operation, which was successfully treated again. There were no cases of local infection. One intraoral type relapsed 3 months after the operation and did not receive further treatment.

The basic data were analyzed using SPSS. 25 software. For the measurement data that followed a normal distribution, the mean and standard deviation were used to describe them, and the two independent sample t-test was employed for comparisons between groups; for the measurement data that did not follow a normal distribution, the median and quartiles were used for description, and the Mann-Whitney U test was used for comparisons between groups. The count data were described using frequencies and percentages, and the chi-square test was used for comparisons between groups. A P value < 0.05 was considered to indicate statistically significant differences. After conducting a chi-square test for comparison, there were no significant differences between different genders and sides (P =

0.109 > 0.05) (Table 1). Using the Mann-Whitney U test for comparison, there was no significant difference in lesion size between whether or not treatment was received ($P = 0.207 > 0.05$) (Table 2).

Table 1

A chi-square test was used for comparison, and no significant differences were found between different genders and sides (P = 0.109 > 0.05).

	Side		χ^2	P
	Left			
sex			2.568	0.109
Male	9(40.9%)	13(59.1%)		
Female	26(61.9%)	16(38.1%)		

Table 2
, The Mann-Whitney U test was used for comparison. There was no significant difference in lesion size between the two treatment groups ($P = 0.207 > 0.05$).

	treatment yes/no		Z	P
	yes	no		
Size of lesion(cm3)	1.00(1.00, 4.00)	1.00(0.28, 2.13)	1.263	0.207

Discussion

3.1 Epidemiological characteristics of sublingual gland cysts

Sublingual gland cysts usually occur due to traumatic rupture of the salivary gland ducts, followed by the leakage of mucus, resulting in local mucus accumulation within the submucosal connective tissue of the oral floor. This manifests as a pale blue, transparent or semi-transparent mass in the sublingual area of the oral floor. In some cases, the mass may rupture and discharge egg-white-like viscous fluid, leading to fluctuating symptoms. Generally, there are no obvious symptoms. When the mass is large, there may be a sense of foreign body in the mouth or it may affect speech and eating. The external type, also known as the drooping type, the cervical type or the deep type, is relatively rare. It presents as swelling in the submandibular or cervical area, even the parapharyngeal region, with a relatively firm texture. The basic formation mechanism includes the presence of ectopic sublingual glands at the posterior part of the neck of the genioglossus muscle; perforation or rupture of the genioglossus muscle allowing mucus to pass through and accumulate in the submandibular area; and iatrogenic factors, such as the recurrence of oral cysts, complications of surgeries on the oral floor (such as stone removal), displacement of the Walton tube, and dental implantation, etc. [5–8]. The results of Lee's study [9] indicate that most of the drooping-type sublingual gland cysts are formed by the extension of the defect in the mandibulo-glossal

muscle forward. Recently, duct variations have been found to be an inducing factor for cyst formation [10]. If there are bilateral cysts or cysts occur in siblings, genetic abnormalities may also play a role in the pathogenesis of these cases [11]. There are also reports indicating that oral external type salivary gland cysts are numerous in cases related to HIV infection, and they are associated with human immunodeficiency virus (HIV) infection, although the exact pathogenesis of this association is still unclear [12–15]. A study conducted in the New Zealand region [16] shows that the overall annual incidence rate is 24 cases per 100,000 person-years. The specific gender incidence rate for men is 3.2 cases per 100,000, for women it is 20 cases per 100,000, and the age-adjusted incidence rate is the highest among the Maori population (6.7 cases per 100,000), followed by Pacific Islanders (44 cases per 100,000), then Asians (0.7 cases per 100,000), and finally Europeans (0.6 cases per 100,000).

3.2 Diagnosis of Sublingual Gland Cysts

The diagnosis of oral-type and mixed-type sublingual gland cysts is relatively straightforward. Clinical examination alone can usually make the diagnosis. In cases where a definitive diagnosis is needed, a puncture of the cyst fluid can reveal a thick, egg-white-like consistency, thereby confirming the diagnosis. All the oral-type cases in this study were diagnosed through clinical examination. However, the diagnosis of the extratemporal type has always been a clinical challenge; its clinical manifestations are diverse, and sometimes it is similar to other lesions in the neck. The differential diagnosis includes submandibular gland mucinous cysts, abscesses, lymphatic malformations, dermoid cysts, cystic hygroma, benign teratomas, and branchial cleft cysts [17]. The 7 cases of extratemporal type in this study underwent CT, MR or ultrasound examinations before surgery, and the diagnosis was confirmed after extracting egg-white-like thick liquid from the submandibular puncture.

The external oral type of diagnostic methods mainly include CT, MR or ultrasound examinations. In CT scans, the external oral cyst appears as an approximately elliptical cyst, with a uniform central attenuation area inside, and the cyst wall is almost invisible [9] (Fig. 1).

On MR, the sublingual gland cysts are all clear in boundary and homogeneous in texture, presenting as a mass on DWI with high signal intensity. Its unique diffusion-weighted imaging (DWI) and apparent diffusion coefficient (ADC) features can be accurately identified (Fig. 2) [18]. Due to its high protein concentration, it forms specific signal intensity, so imaging examinations are helpful for differential diagnosis and can measure the range of the cyst. It is formed by the partial absence of the posterior margin or part of the anterior part of the geniohyoid muscle, and the tail sign (a smooth and gradually shrinking liquid mass) may be a significant feature of magnetic resonance imaging [19].

Ultrasound (US) has become the preferred imaging method for evaluating salivary gland cysts (especially recurrent salivary gland cysts). This is because it is simple to operate, non-radiating, capable of achieving dynamic real-time visualization of anatomical structures, and has a lower cost. A study [20] suggests that the majority of exodontial sublingual gland cysts are accompanied by submandibular mylohyoid aponeurosis defects during ultrasound examination, and the two have a close correlation in surgery. The cystic mandibular swelling, mentohyoid muscle fissure, and submandibular gland

prolapsing hernia (collectively known as the "Jain triad") shown in the "real-time" state (Fig. 3) is helpful for the clear diagnosis of exodontial sublingual gland cysts. The author believes that ultrasound can accurately display the entire range of the lesion. In addition, ultrasound has no radiation risk, is easy to obtain, has low cost, and is suitable for all age groups, so it should be used as the preferred diagnostic method.

The ultrasound standards established by research indicate that all cysts have a close anatomical relationship with the sublingual gland (SLG) and the genioglossus muscle (MM). 97.6% of the lesions have hypoechoic to anechoic echoes. Compared with oral-type cysts, deep cysts are larger and have more irregular shapes. Ultrasound is an excellent diagnostic tool for differentiating oral-type (ER) and extraoral-type (PR) cysts [21]. Some authors [22] believe that ultrasound combined with palpation can effectively display the anatomical abnormalities consistent with cystic adenomatous sublingual gland cysts. Palpation helps to clarify the relationship between the sublingual gland, the cyst fluid, and the often accompanying genioglossus muscle fissure.

Some authors believe that in addition to the above-mentioned examination methods, fine needle aspiration and analysis of the liquid within it is crucial for a clear diagnosis. Since cystic cysts are essentially caused by the exudation of mucus, performing a puncture on the neck mass can extract a substance similar to egg white (Fig. 4). Through biopsy and biochemical analysis, inflammatory cells, dominant tissue cells, high amylase content, and protein content can be detected from the cyst fluid. At the same time, differential diagnosis can be made based on the puncture fluid. If dark red blood appears during fine needle aspiration, it may indicate the basis for the diagnosis of venous malformation. Lymphatic malformations and cystic hygroma present clear yellow or brown liquid, and lymphocytes can be detected through microscopic examination of smears. For branchial cysts, the puncture can extract clear yellow or brown liquid, accompanied or not by cholesterol crystals; teratoma cysts also have their own specific secretions. However, for thyroid thyroglossal duct cysts, the puncture can also extract transparent, slightly cloudy yellow, thin or viscous liquid. Through negative staining with amylase and clinical examination of the movement of the mass when swallowed, it can be differentiated.

Some authors advocate for the detection of amylase levels in the aspirate fluid to assess the possibility of saliva origin [23–25]. For instance, if the cytological examination shows a high amylase content, an increase in tissue cells, and no epithelial cells, this helps to distinguish sublingual gland cysts from other oral or cervical masses (such as thyroid lingual cysts, branchial cleft cysts, cystic hygroma/lymphatic malformation, intramuscular hemangioma, lymphangioma, abscess, or dermoid cysts) [17, 26–27]. A study [28] described a technique for measuring the amylase content in cervical cysts, and it was found that the amylase levels in the contents of all cases were ≥ 3 U/L. There was a significant variation in amylase concentration (range: 5–560 U/L), and 70% of the cases had an amylase concentration lower than 200 U/L. The aspirate usually presented as viscous (87.5%) and yellow or pale yellow.

3.3 Treatment of Sublingual Gland Cysts

The author believes that the treatment of sublingual gland cysts mainly involves two concepts: one is to remove the source, and the other is to ensure smooth drainage. Ostomy surgery, micro-ostomy surgery, laser ablation, cryotherapy, intracystic steroid injection, and sclerosing agent therapy, etc. [29] are used to establish a good channel for cyst fluid discharge, preventing the accumulation of cyst fluid. However, the best treatment concept is to eliminate the exudation of sublingual gland secretions from the source, which can be achieved by surgically removing the sublingual gland through the oral route.

For external-type treatment methods, they include cystectomy; combined cyst and sublingual gland resection; removal of the cyst, sublingual gland, and submandibular gland; OK-432 intracystic injection; and cyst aspiration. Studies have shown [30–31], that using OK-432 for sclerosing therapy to treat sublingual gland cysts is a very effective treatment method, but it may not be effective for external-type sublingual gland cysts.

Currently, orally removing the sublingual gland is the gold standard for the treatment of extracapsular sublingual gland cysts [32–34]. However, there is controversy over whether the cysts located in the neck or under the jaw need to be removed together. These cysts have a thin wall and are prone to rupture, making them easily ruptured. Additionally, extracapsular cysts are usually closely related to the submandibular duct, lingual nerve, and submandibular gland. These anatomical factors make surgical treatment challenging and may lead to adverse consequences, especially in hands of inexperienced doctors. Studies [35–38] suggest that whether it is a cyst confined within the oral cavity or an extracapsular cyst extending to the neck, the most reliable and successful treatment method is to remove the related sublingual gland through the oral cavity, and removing the cyst itself is not necessary. Suresh et al. [39] cited the research results of Patel et al [40], regarding the most effective treatment method, the conclusion was "neck surgery for drooping cysts is unnecessary because cyst removal is not necessary, and there is no causal relationship between the submandibular gland and cyst formation." Moreover, a recent textbook on salivary gland surgery has detailed the best surgical management method for drooping cysts by separately removing the ipsilateral submandibular gland and then performing oral drainage [41], which is expected to enable the best treatment method for cysts to be widely understood, accepted, and practiced [42]. In this study, 6 out of 7 patients with extracapsular cysts underwent oral approach sublingual gland + cyst excision, and 1 patient underwent submandibular approach submandibular gland + sublingual gland + cyst excision surgery due to tight adhesion between the cyst and the submandibular gland. All patients with extracapsular cysts had no recurrence or complications such as tongue nerve injury after follow-up.

Conclusion

Based on the experience summarized in this article, the author believes that the diagnosis and treatment of intraoral sublingual gland cysts do not present significant difficulties. However, the diagnosis of extratemporal type requires detailed clinical examination, combined with CT or MR and ultrasound examinations, as well as fine needle aspiration of the fluid for analysis of its nature, appearance, and in some cases, cytological and amylase tests, as any single diagnostic method has certain limitations.

Although complete removal of the sublingual gland is currently the preferred treatment option for both intraoral and extratemporal sublingual gland cysts, there are risks of tongue nerve injury and incision hematoma.

Declarations

Acknowledgements

Not applicable.

Author contributions

Conceptualization, Xiaorong Tang, and YunGu; methodology, Xiaorong Tang; validation, Xiaorong Tang, YunGu, and ZhengZheng; formal analysis, Xiaorong Tang, YunGu, and ZhengZheng; investigation, Xiaorong Tang, YunGu, and ZhengZheng; resources, Xiaorong Tang; writing—original draft preparation, YunGu, and ZhengZheng; writing—review and editing, Xiaorong Tang. All authors have read and agreed to the published version of the manuscript.

Funding

Special Research Fund Project of Affiliated Nantong Clinical College of Nantong University (2025LY063)

Data availability

The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

Ethics approval and consent to participate

This study was approved by the Ethics Committee of Affiliated Nantong Clinical College of Nantong University(Nantong First People's Hospital) (2025-KT242-01)and the Helsinki Declaration guidelines were followed. Written informed consent for publication was obtained from all the patients.

Consent for publication

Informed written consent was obtained from all the patients for the publication of this manuscript and the accompanying images.

Competing interests

The authors declare no competing interests

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Figures



Figure 1

Patient Jiang XX, male, 41 years old, presented with a painless mass in the right mandibular region after two years. The disease course showed fluctuations. CT scan: There is a nodular low-density shadow in the right submandibular gland, approximately 2.6×2.4 cm in size, with clear boundaries and uniform density, with a CT value of about 25 HU.



Figure 2

Mr. XX, male, 15 years old. He presented with a painless mass in the left submandibular region for one year. The MRI results indicated: Multiple round-shaped high signal areas with equal signal intensity on T1WI and high signal intensity on DWI were observed in the lateral part of the left submandibular gland, the long cervical muscle, and the masseter muscle muscle space. The size was approximately 26*16mm, with clear boundaries and no abnormal signals in the adjacent bone. It is considered to be a benign lesion in the lateral part of the submandibular gland, the long cervical muscle, and the masseter muscle muscle space, and a lymphatic cyst may be involved.



Figure 3

Wang XX, male, 15 years old. Presented with a painless mass in the left submandibular region for one year. Ultrasound examination: A 36*18mm liquid-dark area was observed in the submandibular region, seemingly connected to the sublingual gland

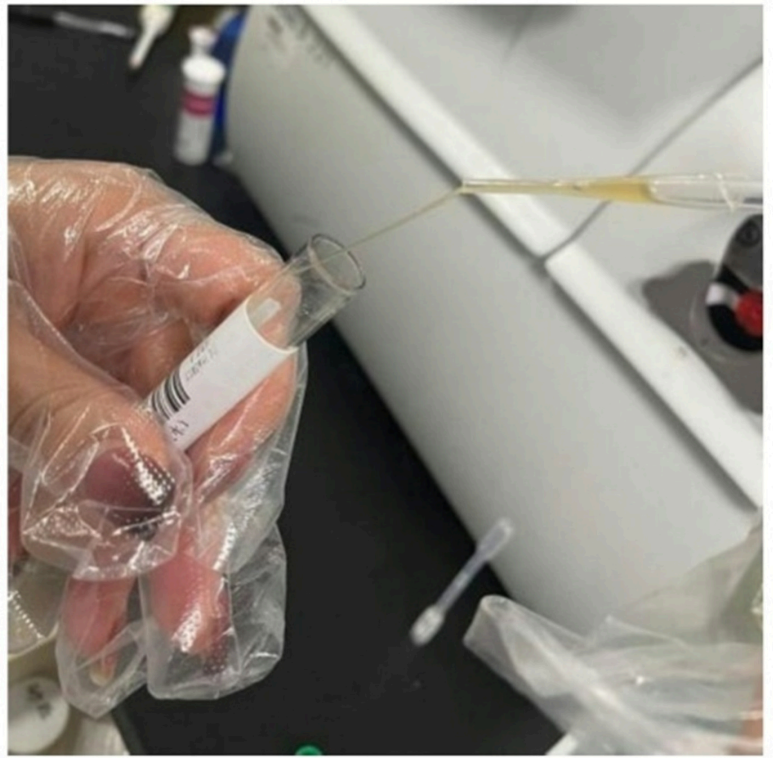


Figure 4

Wang XX, male, 15 years old. Presented with a painless mass in the left submandibular region for one year. A puncture was performed and a milky, viscous, and string-like liquid was extracted.