

Case Report

Submandibular gland mucocele: a case report and literature review

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Abstract: Mucoceles occur most commonly in the oral mucosa and minor salivary glands. The lower lip is the most common site for the development of a mucocele, and occurrence in one of the major salivary glands is rare. A review of literature written in English revealed 13 cases of submandibular gland mucocele. The etiology of mucoceles is thought to involve partial obstruction or disruption of the salivary gland duct. Mucoceles are categorized as retention or extravasation mucoceles based on their histopathological appearance. Surgical removal is the preferable treatment. Excision of a mucocele in continuity with the submandibular gland reduces the recurrence rate. A rare case of a submandibular gland mucocele is reported.

Keywords: Mucocele, salivary gland, submandibular gland, retention, extravasation

Introduction

Mucoceles are common benign cystic lesions that occur in the minor salivary glands. About 60 to 70% of mucoceles occur in the lower lip, 6 to 15% occur on the floor of the mouth, and only a few cases have been reported to occur in the submandibular gland [1]. Factors such as trauma or obstruction of the salivary gland ducts are thought to contribute to the development of mucoceles [2]. Total excision of mucoceles reduces the recurrence rate. A case of a 24-year-old man with a submandibular gland mucocele presenting as right-sided lateral neck swelling is reported.

Case report

The patient was a 24-year-old man with Down syndrome. He was sent to the otolaryngology outpatient department for treatment, and painless swelling of the right lateral neck for one week was reported. There was no sore throat, toothache, dysphagia, otalgia, weight loss, or fever. There was no family history of disease, and there was no history of habitual cigarette smoking or alcohol consumption.

The initial physical examination showed one palpable 5.0×4.0-centimeter mass over the

right submandibular triangle of the neck (**Figure 1**). The neck mass was smooth and mobile with no tenderness, and there was no additional lymphadenopathy or palpable mass of the neck. The nose, ears, oral cavity, pharynx, and larynx were within normal limits in a series of examinations. The laboratory findings showed a white blood cell count of $10.1 \times 10^3/\mu\text{L}$, neutrophil count of 75.5%, lymphocyte count of 14.0%, and C-reactive protein level of 0.9 mg/dL. Computed tomography of the nasopharynx to the neck with contrast showed one 4.2×2.7-centimeter cystic-like, hypo-dense, and peripherally enhanced lesion involving the submandibular gland in the right submandibular space (**Figure 2**). Fine needle aspiration cytology of the cyst was performed, and the yellowish, serous, aspirated fluid was negative for malignancy. After physical, laboratory, radiological, and cytological examinations, as well as consideration of the patient's history, a branchial cleft cyst was the tentative diagnosis.

Excisional surgery under general analgesia was performed. The incision was made about two fingerbreadths below the jawline. The cystic-like tumor closely adhered to the submandibular gland, and no tract connecting the tumor to the pharynx or hyoid bone could be identified. The



Figure 1. One palpable 5.0×4.0 centimeter mass over the right submandibular triangle of neck.



Figure 2. Computed tomography of the nasopharynx to the neck shows one hypo-dense lesion of about 1.2×2.7 centimeters with peripheral enhancement involving the submandibular gland in the right submandibular space.

tumor was completely excised along with the submandibular gland (**Figure 3**). The operative procedure went smoothly with no immediate complications. On gross examination of the specimen, the cyst had a thick wall with yellowish serous fluid contents. Microscopic examination revealed mucus covered with granulation tissue without a lining epithelium (**Figure 4**).

The patient then received timely follow-up in the otolaryngology outpatient department for six months. The patient had no evidence of recurrence or additional problems.

Discussion

Mucoceleles are benign, cystic-like lesions that can present at various sites of the oral mucosa and most commonly develop in minor salivary

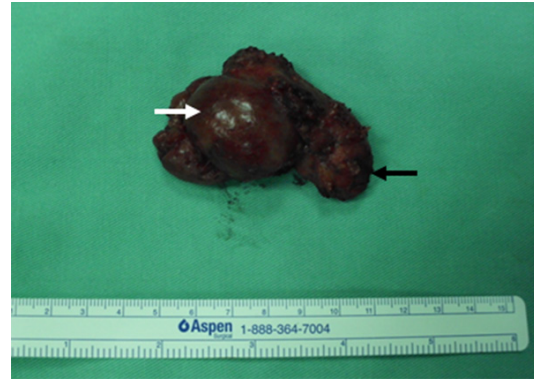


Figure 3. Macroscopic appearance of the specimen shows the excised cyst (white arrow) and submandibular gland (black arrow).

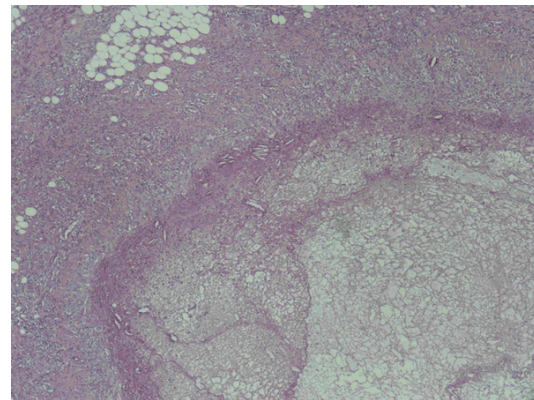


Figure 4. Histological examination of the cyst reveals mucinous material covered by granulation tissue (hematoxylin and eosin stain, ×40).

glands. The occurrence of mucoceles in major salivary glands is uncommon, and mucoceles in the submandibular gland are rare. Searching published literature written in English, we found 13 cases of submandibular gland mucocele were reported since the first case published by Surkin et al at 1985 (**Table 1**). This literature review reveals uneven distribution between genders that the incidence was higher in males compared to females (11 males and 2 females). Moreover, submandibular gland mucocele occurred in patients ranged from 16 months to 39 years of age (mean age 20.7 ± 11.4 years). The lesion was observed on the right side in 8 patients, on the left side for 4 patients and 1 patient had bilateral lesions.

Mucoceleles are categorized as retention or extravasation mucoceles based on their histopathological appearance. A retention mucocele

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Table 1. Demographic characteristics of 13 published cases of submandibular gland mucocele

No.	Author	Gender	Age	Duration of symptoms	Side	Treatment*	Follow-up
1	Anastassov et al [1]	Male	18 years	3 months	Left	Excision with SMG, SLG	16 months
2	Anastassov et al [1]	Male	30 years	10 years	Right	Excision with SMG, SLG	12 months
3	Surkin et al [4]	Male	39 years	6 weeks	Right	Excision with SMG	No evidence of disease
4	Choi et al [6]	Male	16 months	Several days	Bilateral	Excision with SMG, SLG	24 months
5	Van der Goten et al [7]	Male	7 years	3 weeks	Right	Excision with SMG	Not available
6	Van der Goten et al [7]	Female	18 years	8 weeks	Left	Excision with SMG, SLG	Not available
7	Boneu-Bonet et al [9]	Male	25 years	6 months	Right	Excision with SMG	No evidence of disease
8	Hze-Khoong et al [10]	Male	21 years	1 week	Right	Excision with SMG	No evidence of disease
9	Okumura et al [11]	Male	7 years	2 years	Right	Excision with SMG, SLG	No evidence of disease
10	Ozturk et al [12]	Female	11 years	6 months	Right	Excision with SMG, SLG	8 months
11	Ozturk et al [12]	Male	38 years	8 years	Left	Excision with SMG, SLG	34 months
12	Stranc et al [13]	Male	29 years	1 month	Right	Excision	Recurrence
13	Cholankeril et al [14]	Male	25 years	6 months	Left	Excision with SMG	6 months

*SMG: submandibular gland; SLG: sublingual gland.

is a mucus-containing cyst with an epithelial wall covering, while an extravasation mucocele is also a mucus-containing cyst but is covered by granulation tissue without an epithelium [3].

The mechanism of development is thought to be partial obstruction of the salivary gland duct in the case of retention mucoceles or disruption of the salivary gland duct in the case of extravasation mucoceles [4]. Retention mucoceles are more common at advanced age, while extravasation mucoceles are commonly found among young people [5].

The clinical symptoms of submandibular gland mucoceles include a painless, slow-growing mass in the submandibular region. They usually present as a soft, well-circumscribed, mobile masses [6]. The differential diagnosis of a submandibular cystic-like mass includes congenital lesions such as branchial cleft cysts, dermoid cysts, cystic hygroma, and thyroglossal duct cysts, as well as acquired lesions such as ranula, abscesses, and cystic degeneration of neoplasms [7].

Computed tomography or magnetic resonance imaging examinations are important for determining the accurate location of the lesion and for distinguishing between benign and malignant lesions. In addition, fine needle aspiration cytology is useful for diagnosing cervical masses. The sensitivity rate for such masses is about 90 to 100%, but the false negative rate for cervical cysts is as high as 50% [8].

Surgical intervention is the preferable treatment for a submandibular gland mucocele. Currently, the surgical methods include marsupialization, cystectomy, injection of sclerosing agents, incision and drainage, excision of the mucocele in continuity with the sublingual gland, and/or the submandibular gland [1]. Compared to other surgical methods that maintain the submandibular gland, excision of a mucocele in continuity with the submandibular gland has the advantage of a lower recurrence rate. If the submandibular gland mucocele affecting or closely next to the sublingual gland, it is recommended to excise the sublingual gland as well [9].

In summary, even though mucoceles are rare and benign lesions that occur in the submandibular gland, they should always be included in the differential diagnosis of submandibular cystic-like masses. The clinical symptoms include a painless, slow-growing mass in the submandibular region. Surgical intervention with excision of the mucocele in continuity with the submandibular gland is the preferred treatment.

Disclosure of conflict of interest

None.

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