

Submandibular Gland Surgery: Our Clinical Experience

Selim Sermed Erbek¹, Alper Köycü², Özgül Topal³, Hatice Seyra Erbek¹, Levent Naci Özlüoğlu¹

¹Department of Otorhinolaryngology, Başkent University Ankara Hospital, Ankara, Turkey

²Clinic of Otorhinolaryngology, Siirt State Hospital, Siirt, Turkey

³Department of Otorhinolaryngology, Başkent University Konya Hospital, Konya, Turkey

Original Investigation

Abstract

Objective: The aim of this study was to assess the demographic findings and surgical results of patients who underwent submandibular gland excision at a tertiary care center.

Methods: The clinical characteristics and histopathological results of 45 patients who had undergone submandibular gland excision between 1997 and 2014 were evaluated in detail.

Results: Twenty-eight (62.2%) and 17 (37.8%) patients presented with a complaint of a painful mass and painless mass, respectively. Histopathologic investigation of the surgical specimens revealed sialolithiasis in 14 patients (31.1%), chronic sialadenitis in 16 (35.6%), benign tumor in 12 (26.7%), malignant tumor in two (4.4%), and mucocele extravasation in one. As complications, permanent paralysis of the

marginal mandibular branch of the facial nerve was seen in one patient (2.2%), temporary paralysis of the marginal mandibular branch of the facial nerve was seen in seven (15.6%), orocutaneous fistula was seen in one (2.2%), and temporary paralysis of the hypoglossal nerve was seen in one (2.2%).

Conclusion: This study revealed that in patients presenting with complaints of a submandibular gland mass, sialolithiasis, sialadenitis, and benign masses were the mostly diagnosed disorders. Transcervical submandibular gland excision is a satisfactory procedure with low complication and recurrence rates when it is performed on selected patients and obeyed to surgical techniques.

Keywords: Salivary gland, submandibular gland, surgery, complication

Introduction

Primary masses of the submandibular gland are extremely rare in comparison to the parotid. Therefore, the cause of a significant portion of surgeries of this gland is the stones in the salivary gland and the chronic infections of the salivary gland usually developing secondary to sialolithiasis. Although the incidence of sialolithiasis is reported as 1% in autopsy studies, symptomatic stones are clinically much rarer (1). Of the symptomatic stone cases, 80-92% arises from the submandibular gland (1). A decrease in the production of saliva in the long term on the basis of chronic obstruction and chronic inflammation reflecting to the clinic as tightening and stiffening that give the impression of a focal mass in the gland with palpation may develop in these patients. No surgery is needed in these patients who are asymptomatic most of the time. However, recurrent episodes of acute inflammation require this treatment.

Only 10-15% of the neoplasms arising from salivary gland are seen in the submandibular gland (2). Of these masses, 50-57% are benign (pleomorphic adenoma most commonly) and 43-50% are malignant (adenoid cystic carcinoma most commonly) (3).

Transcervical submandibular gland excision has been used for many years in chronic sialadenitis, sialolithiasis and benign or malignant gland tumors for diagnosis or treatment. However, only limited numbers of reports related to long-term large series of cases of submandibular gland excision are available in the literature (4, 5). The aim of this study is to share the symptoms, demographic features, clinical and radiological diagnostic methods, surgical specimen histopathological findings, surgical complications and follow-up information of the patients who underwent submandibular gland excision in our clinic.



Address for Correspondence:
Alper Köycü,
E-mail: alperkoycu@gmail.com

Received Date: 27.12.2015
Accepted Date: 26.04.2016

© Copyright 2016 by Official Journal of the Turkish Society of Otorhinolaryngology and Head and Neck Surgery Available online at www.turkarchotorhinolaryngol.org
DOI: 10.5152/tao.2016.1467

Methods

This study has been retrospectively conducted by using the file information belonging to 45 patients who underwent submandibular gland excision between the years of 1997 and 2014 in a tertiary hospital. The symptoms, demographic features, clinical and radiological diagnostic methods, histopathological findings of surgical specimen and surgical complications were obtained by retrospectively scanning the files of patients who underwent excision of the submandibular gland in our clinic. The study has been approved by the local Ethics Committee and conducted with the permission number KA14/108.

Results

In the study, the files of 54 patients who underwent submandibular salivary gland excision were initially scanned, but nine patients were excluded because their pathology did not primarily originate from submandibular glands (four patients with lymphadenitis, three patients with lipoma, two patients with metastases) and 45 patients 21 of whom were women and 24 were male were included in the study. The average age was 44.8 (14-82). Right submandibular salivary gland excision was performed in 23 (51.1%) of the patients and left submandibular salivary gland excision in 22 (48.9%). The average follow-up duration is 38.4 months (3-60 months) (Table 1).

Twenty-eight (62.2%) of the patients applied with the complaint of swelling and 17 (37.8%) of them with the complaint of painless swelling. According to the findings obtained from the physical examination of submandibular triangle, there were palpable mass in 15 of the patients (33.3%), pain-sensitivity of glands on palpation in 21 (46.7%) of the patients, hard gland in 6 (13.3%) of the patients and palpable stone in 3 (6.7%) of the patients (Table 1).

Table 1. Demographic characteristics of the patients

Average age		44.8
Gender, n (%)	Male	24 (%53.3)
	Female	21 (%46.7)
Side, n (%)	Right	23 (%51.1)
	Left	22 (%48.9)
Follow-up period (months)		38.4
Complaint	Painful swelling, n (%)	28 (%62.2)
	Painless swelling, n (%)	17 (%37.8)
Findings	Palpable mass, n (%)	15 (%33.3)
	Painful gland, n (%)	21 (%46.7)
	Firm gland, n (%)	6 (%13.3)
	Palpable stone, n (%)	3 (%6.7)
Complication	Permanent marginal nerve paralysis, n (%)	1 (%2.2)
	Temporary marginal nerve paralysis, n (%)	7 (%15.6)
	Orocutaneous fistula	1 (%2.2)
	Temporary paralysis of the Hypoglossal N.	1 (%2.2)

N: nerve

Most frequently, ultrasonography (USG) was used as the diagnostic method (31 patients, 68.8%). Four patients (8.8%) were diagnosed with computed tomography (CT) and three patients (6.6%) with magnetic resonance imaging (MRI). Both ultrasonography (USG) and magnetic resonance imaging (MRI) were preferred as a diagnostic method in one patient (2.2%). While fine needle aspiration (FNA) was performed in six (13.3%) patients who applied to the clinic with the complaint of painless swelling and who was considered to have tumor in preoperative evaluation, the accurate diagnosis could be made only in three (50%) of them.

In the histological examination of the surgical specimen, sialolithiasis was found in 14 (31.1%) patients, chronic sialadenitis in 16 (35.6%) patients, benign tumor in 12 (26.7%) patients, malignant tumor in two (4.4%) patients and extravasation mucocele in one (2.2%) patient (Table 2).

Pleomorphic adenoma was found the most often among the benign tumors at a ratio of 24.4% (11 patients) (Figure 1). The remaining one patient (2.2%) had cavernous hemangioma arising from submandibular gland. The diagnoses of two patients with malignant tumor were lymphoma and adenoid cystic carcinoma. When these diagnoses were compared with the preoperative FNA results, while accurate diagnosis was made in three pleomorphic adenomas, the lymphoma case was found to be reported as lymphoid cells.

As postsurgical complications, it was observed that permanent paralysis of facial nerve marginal mandibular branch developed in one patient (2.2%), temporary paralysis of the marginal mandibular branch in seven (15.6%) patients, oro-cutaneous fistula in one patient (2.2%) and hypoglossal nerve temporary paresis in one patient (2.2%). No additional treatment was applied to the patients with temporary paralysis of marginal mandibular branch of the facial nerve and a complete spontaneous remission was observed in approximately 3 weeks. Revision surgery was not implemented due to postsurgical infections, stones or recurrent mass.

Discussion

Submandibular gland disorders can be divided into two groups as neoplastic and non-neoplastic. Chronic sialadenitis developing on the basis of sialolithiasis comes into prominence as the most common non-neoplastic pathology in submandibular gland excision. This rate is quite high as 44.8 – 73.5% in some studies in the literature (6, 7). It was found as 18.2% in the study of Dalgiç et al. (4) and as 35.6% in our series.

Neoplastic submandibular gland masses constitute 8-22% of all salivary gland tumors (8). Of these tumors, 50-57% are benign and the rest are malignant tumors (8). In particular, rare incidence of malignant tumors to this extent leads to the lack of large series of cases and thus, inability to reach a consensus on the size of the surgical and nonsurgical oncological treatment. Consistent with the literature, the diagnoses of benign tumors (26.7%) were made the most frequently and among those, di-

Table 2. Histopathological diagnoses of patients

Histopathology		The number of patients	Average age (years)	%
Sialadenitis		16	43.6	35.6%
Sialolithiasis		14	48.8	31.1%
Benign tumors		12		26.7%
	Pleomorphic adenoma	11	43	24.4%
	Cavernous hemangioma	1	36	2.2%
Malignant tumors		2		4.4%
	Lymphoma	1	67	2.2%
	Adenoid cystic carcinoma	1	39	2.2%
Others	Extravasation mucocele	1	19	2.2%

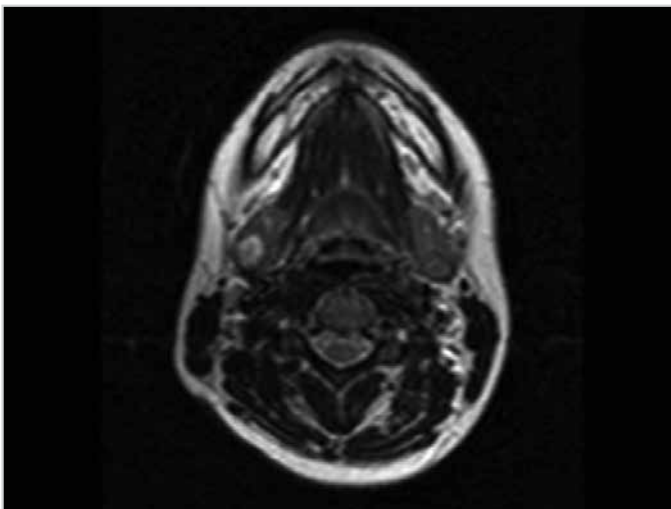


Figure 1. In the right submandibular gland, pleomorphic adenoma that gives hyperintense appearance in Magnetic Resonance Imaging

agnoses of pleomorphic adenoma (11 patients, 24.4%) were the most frequent in our study. Our total rate of malignant tumors is very low (4.4%) and adenoid cystic carcinoma stands out as the primary malignant tumor. The patient with the diagnosis of adenoid cystic carcinoma was postoperatively followed for about 10 years and the patient received a repeated radiotherapy and chemotherapy during this time. The patient in whom widespread metastases in the neck and lungs were found died at the end of the 10th year. The ratio of being benign or malignant of the tumors was found to be 85.7% / 14.3% in our study.

The rate of salivary gland tumors in children is much lower than in adults. It is seen 10 times more often in parotid than in submandibular gland (9). Therefore, pediatric submandibular gland pleomorphic adenomas are sporadically reported rather as case reports. A 14-year-old girl patient has been diagnosed with pleomorphic adenoma in our study as well.

Ultrasonography is an important diagnostic tool in submandibular gland lesions. It is useful especially in submandibular gland superficial lesions (10). Papaspyrou et al. (6) reported the sensitivity and specificity of the USG as respectively 87% and

81.3% in the diagnosis of sialolithiasis. CT and MRI should be selected to investigate the situation of tumor spread, local invasion and perineural invasion in the cases where malignant cells are seen in the disease or cytology extending to deep tissue (10). USG became the examination that the clinicians preferred most commonly in our series of patients with a ratio of 68.8%.

There is still no consensus about the best biopsy method to perform because the neoplasms of the submandibular gland are very rare. Olubaniyi et al. (11) reported that diagnosis could not be made in fine-needle aspiration biopsies at a rate of approximately 73% in a study that they compared the biopsy methods in neoplastic pathologies of this area. In addition, because the fine needle biopsy will mostly remain insufficient in the distinction of low grade lymphoma and reactive nodal hyperplasia, it creates excisional biopsy indication in most patients (12). For these reasons, both researchers suggest that “core needle” biopsy be performed in company with ultrasonography in terms of diagnostic efficiency (11, 12). As eighteen gauge and smaller needles are used, it is stated that there isn't a defined tumor seeding. FNAB was used as a preoperative diagnostic tool in 13.3% of the patients in our clinic and a correct diagnosis was able to be reached in 50% of them. However, while the rate of correct diagnosis in FNAB was achieved only in pleomorphic adenoma, a correct diagnosis could not be made in other malignant pathologies. Excisional biopsy is the preferred method when the diagnosis can not be reached through the method of FNA in our clinic.

Temporary marginal mandibular nerve (MMN) damage after transcervical submandibular gland surgeries is reported as 36% (13) and the permanent damage as high a rate as 12% (14) in the literature. Beahm et al. (15) reported these rates between 7.7 and 36% in the study in which they conducted a literature review. Permanent marginal branch paralysis is 2.2% and temporary marginal branch paralysis is 15.6% in our series. The patients with marginal branch paralysis were followed without applying any additional treatment in the postoperative period. While a complete recovery was observed in seven patients within three weeks on average, a permanent paralysis occurred in one patient. Hernando et al. (7) reported two permanent MMN paralysees in a series of

29 patients and while the diagnosis of one of these was malignant tumor of the gland, the other was reported to be sialadenitis. In our study, the patient with permanent MMN paralysis was diagnosed with sialadenitis. Four of the seven patients with temporary paresis were diagnosed with sialadenitis, two patients with sialolithiasis and one patient with pleomorphic adenoma. The presence of temporary or permanent paralysis in our study increases with the inflammation, in other words with sialadenitis. There are three surgical maneuvers to reduce the risk of MMN damage in the transcervical surgical approach for the gland:

1. The technique that is performed with the gland identified lower than hyoid bone and in which the MMN is not sought.
2. The technique in which the MMN is found in the level where it leaves the parotid tail, followed and protected.
3. The technique in which the facial veins are found at a lower level and ligated, and the nerve is suspended by being elevated, thus MMN is kept out of the surgical area.

In our clinic, as Riffat et al. (16) suggests, after the neck incision 2 cm away from the mandibular corpus by bringing the patient's neck into hyperextension, facial veins are ligated at the lower edge of the gland, elevated together with the gland fascia, and thus the MMN is protected. Besides, keeping the nerve in mind while using retractors during surgery, respectful approach to tissue and tissue plans and behaving carefully while using bipolar cautery in hazardous areas during the operation keep the rate of complications at quite low levels. Transoral open and/or endoscopic approaches, the use of which has been increasing recently reduce this risk significantly (15).

Open surgical approaches, endoscope assisted open surgical approaches and total endoscopic approaches are available in submandibular gland surgery. Open surgical approaches are lateral transcervical, submental, retroauricular and transoral ones. Semi endoscopic surgical procedures and total endoscopic surgical methods with a pouch created by giving carbon dioxide gas that are applicable in these regions are also available (15). These surgical procedures have different advantages and disadvantages when compared with each other. They are preferred and can be applied according to the surgeon's experience and capability, accessibility to technology and the characteristics of the patient and disease.

Transoral approach first entered the literature in 1960, but gained popularity with the study that Hong et al. (17) conducted in 2000 (18). Hong et al. (17) reported the advantages of transoral approach to lateral transcervical surgery as the reduction in MMN paralysis and the risk of mucocele formation and lack of skin scars in their two series of 31 and 77 patients. However, in transoral surgery, they detected an abnormal sensation in the tongue at a rate of 74% and limitations in tongue movements at a rate of 70% in the early postoperative period. The limitation of the tongue movements declined to about 10% (17, 19) during the follow-up. Weber et al. (20) identified the abnormal sensation in the tongue at a rate of 43%, but they didn't observe any limitations in the tongue movements and wound scar.

They reported that they managed this by staying a little away from the lingual gingiva to make the incision and thus reducing the tension. They also mentioned that abnormal sensation could be prevented by avoiding stretching the lingual nerve. A history of gland surgery and a small oral cavity were mentioned as relative contraindications for transoral surgery. In the study that Çukurova et al. (21) conducted, 10 patients who underwent transoral surgery and 10 patients who underwent transcervical surgery were compared. They found that transoral surgical approach significantly shortened the duration of hospital stay in comparison to the transcervical approach. However, sialadenitis repeated due to the remaining glandular tissue in a patient who underwent transoral surgery (21).

Springborg et al. (5) reported postoperative infections at a rate of 9.4% in their series of 139 patients. They reported that they used preoperative antibiotic prophylaxis in order to prevent this complication. We don't use preoperative prophylaxis in our clinic. However, prophylactic antibiotics are administered for 7 days postoperatively.

Among the surgical complications, hypoglossal nerve damage has been reported very rarely (0-1.4%) (5). This situation is caused by the nerve's settlement partially sheltered by the posterior belly of the digastric muscle that is in the inferomedial of the lower 1/3 of the gland. In the patient with cavernous hemangioma who was operated in our clinic, paresis of the 12th nerve that lasted for 5 months postoperatively developed due to heavy bleeding during the surgery and frequent and uncontrolled use of clamp/bipolar cauterization. In the follow-up, the patient recovered without sequelae.

Conclusion

The most common pathology in submandibular gland is the formation of stones in the gland or ducts. In particular, extremely rare incidence of malignant tumors leads to the lack of large series of cases and thus, inability to reach a consensus on the type of the surgical and nonsurgical oncological treatment. Transcervical submandibular gland excision is an applicable surgery with acceptable rates of complications and sequelae. The risk for neurological complications which may occur due to the nerves close to surgical area can be reduced with approaches that are loyal to the surgical principles and respect to tissue planes.

Ethics Committee Approval: Ethics committee approval was received for this study from the ethics committee of Başkent University Hospital (KA 14-108).

Informed Consent: Written informed consent was obtained from patients or parents of the patients who participated in this study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept - S.S.E., H.S.E.; Design - A.K., S.S.E., L.N.Ö.; Supervision - S.S.E., H.S.E., L.N.Ö.; Resources - A.K., Ö.T.; Materials - A.K., Ö.T.; Data Collection and/or Processing - A.K., Ö.T., H.S.E.; Analysis and/or Interpretation - A.K., H.S.E., L.N.Ö.; Literature Search - A.K., Ö.T.; Writing Manuscript - A.K., Ö.T.; Critical Review - S.S.E., L.N.Ö.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study has received no financial support.

References

- Williams MF. Sialolithiasis. *Otolaryngol Clin North Am* 1999; 32: 819-34. [\[CrossRef\]](#)
- Stenner M, Klussmann JP. Current update on established and novel biomarkers in salivary gland carcinoma pathology and the molecular pathways involved. *Eur Arch Otorhinolaryngol* 2009; 266: 333-41. [\[CrossRef\]](#)
- Rinaldo A, Ferlito A, Pellitteri PK, Robbins KT, Shaha AR, Bradley PJ, et al. Management of malignant submandibular gland tumors. *Acta Otolaryngol* 2003; 123: 896-904. [\[CrossRef\]](#)
- Dalgic A, Karakoc O, Karahatay S, Hidir Y, Gamsizkan M, Birkent H, et al. Submandibular triangle masses. *J Craniofac Surg* 2013; 24: e529-31.
- Springborg LK, Møller MN. Submandibular gland excision: long-term clinical outcome in 139 patients operated in a single institution. *Eur Arch Otorhinolaryngol* 2013; 270: 1441-6. [\[CrossRef\]](#)
- Papaspyrou G, Werner JA, Sesterhenn AM. Transcervical extirpation of the submandibular gland: the University of Marburg experience. *Eur Arch Otorhinolaryngol* 2014; 271: 2009-12. [\[CrossRef\]](#)
- Hernando M, Echarri RM, Taha M, Fragueiro LM, Hernando A, Mayor GP. Surgical complications of submandibular gland excision. *Acta Otorrinolaringol Esp* 2012; 63: 42-6. [\[CrossRef\]](#)
- Spiro RH. Salivary neoplasms: overview of a 35-year experience with 2807 patients. *Head Neck Surg* 1986; 8: 177-84. [\[CrossRef\]](#)
- Bentz BG, Hughes CA, Lüdemann JP, Maddalozzo J. Masses of the salivary gland region in children. *Arch Otolaryngol Head Neck Surg* 2000; 126: 1435-9. [\[CrossRef\]](#)
- Lee YY, Wong KT, King AD, Ahuja AT. Imaging of salivary gland tumours. *Eur J Radiol* 2008; 66: 419-36. [\[CrossRef\]](#)
- Olubaniyi BO, Chow V, Mandalia U, Haldar S, Gok G, Michl P, et al. Evaluation of biopsy methods in the diagnosis of submandibular space pathology. *Int J Oral Maxillofac Surg* 2014; 43: 281-5. [\[CrossRef\]](#)
- Sellon E, Moody A, Howlett D. Ultrasound guided core biopsy is the diagnostic tool of choice in salivary gland swellings. *BMJ* 2012; 345: e7782.
- Smith WP, Peters WJ, Markus AF. Submandibular gland surgery: an audit of clinical findings, pathology and postoperative morbidity. *Ann R Coll Surg Eng* 1993; 75: 164-7.
- De M, Kumar Singh P, Johnson AP. Morbidity associated with submandibular gland excision: a retrospective analysis. *Internat J Head Neck Surg* 2006; 1: 1.
- Beahm DD, Peleaz L, Nuss DW, Schaitkin B, Sedlmayr JC, Rivera-Serrano CM, et al. Surgical approaches to the submandibular gland: A review of literature. *Int J Surg* 2009; 7: 503-9. [\[CrossRef\]](#)
- Riffat F, Buchanan MA, Mahrous AK, Fish BM, Jani P. Oncological safety of the Hayes-Martin manoeuvre in neck dissections for node-positive oropharyngeal squamous cell carcinoma. *J Laryngol Otol* 2012; 126: 1045-8. [\[CrossRef\]](#)
- Hong KH, Kim YK. Intraoral removal of the submandibular gland: a new surgical approach. *Otolaryngol Head Neck Surg* 2000; 122: 798-802. [\[CrossRef\]](#)
- Downton D, Qvist G. Intra-oral excision of the submandibular gland. *Proc R Soc Med* 1960; 53: 543-4.
- Hong SM, Yang YS. Surgical results of the intraoral removal of the submandibular gland. *Otolaryngol Head Neck Surg* 2008; 139: 530-4. [\[CrossRef\]](#)
- Weber SM, Wax MK, Kim JH. Transoral excision of the submandibular gland. *Otolaryngol Head Neck Surg* 2007; 137: 343-5. [\[CrossRef\]](#)
- Çukurova İ, Arslan İB, Bulgurcu S, Demirhan E. Transoral versus transcervical approach to submandibular gland: techniques and outcomes. *Kulak Burun Bogaz Ihtis Derg* 2015; 25: 319-23. [\[CrossRef\]](#)