

# Surgery for Submandibular Gland Diseases: 14-years of Experience of a Single Tertiary Center

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## ABSTRACT

**Objective:** Surgical removal is a valid option in the treatment of submandibular gland diseases but there are still operative and diagnostic pitfalls. We aimed to review and investigate the long-term clinical, surgical, and pathologic outcomes of our cases that had undergone submandibular gland removal.

**Materials and Methods:** Retrospective analysis was conducted on patients who underwent submandibular gland excision via the transcervical approach with minimal skin incision between January 2007 and April 2020. Demographic data, preoperative FNA cytology results, clinical findings, surgical procedure, postoperative histopathological diagnosis, and postoperative morbidity were reviewed and analyzed.

**Results:** 66 patients (41.7%) had sialolithiasis, 47 patients (29.7%) had chronic sialadenitis. Pleomorphic adenoma was found in 30 of the 44 patients that been operated on for submandibular mass. Gland derived carcinoma was found in 9 patients. The sensitivity of FNA biopsy in our case series was 83.3% (10/12), specificity was 78.1% (25/32), positive predictive value was 71.4% (10/14), and the negative predictive value was 96.1% (25/26). Permanent facial nerve marginal mandibular branch palsy was found in two patients (1.2%).

**Conclusion:** The transcervical approach to the submandibular gland offers safe and reliable removal of the submandibular gland with a minimal risk of complications. FNA cytology was found to be useful in the preoperative evaluation of the submandibular masses.

**Keywords:** Sialolithiasis, submandibular gland, submandibular gland excision, submandibular gland neoplasm

## INTRODUCTION

Surgical excision of the submandibular gland is indicated in patients with submandibular neoplasm or inflammatory diseases. Ten percent of the salivary gland neoplasms are located in the submandibular gland, and there is a higher malignancy rate than other major salivary glands (1, 2). Fine needle aspiration (FNA) biopsy is a minimally invasive procedure and provides diagnostic information before the operation. Since high-grade and locally advanced malignant submandibular masses may require more extensive surgery or a multi-modality treatment, FNA biopsy may affect the type or the timing of the surgical treatment (3). However, many studies reported different outcomes in the diagnostic accuracy, and there are controversies on the clinical significance of FNA biopsy in the diagnosis of salivary gland masses (4, 5).

Chronic sialadenitis and sialolithiasis are mostly seen in the submandibular glands, and surgical excision is considered a standard and safe method for patients not responsive to conservative treatments and minimally invasive techniques (6, 7). However, submandibular gland surgery carries a risk of operative complications like damage to the facial nerve's mandibular branch, hypoglossal nerve, and lingual nerve. Smaller incisions and meticulous dissection are usually preferred, but there is a considerable risk of postoperative complications like salivary fistula, wound infection, scar formation, and disease recurrence (8).

Surgical management is the most valid method in the treatment of submandibular gland diseases. However, there are still operative and diagnostic pitfalls that might lead to significant complications. Our study aimed to review our 14 years of experience in submandibular gland surgery and investigate the clinical, surgical and pathological outcomes of our cases.

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## MATERIAL AND METHODS

This study was designed as a retrospective clinical study. A total of 158 patients who underwent submandibular gland excision from January 2007 to April 2020 at a tertiary academic center were included. Demographic data, preoperative FNA cytology results, clinical findings, surgical procedure, postoperative histopathological diagnosis, and postoperative morbidity were obtained from the digital database.

### Ethical considerations

All procedures performed in studies involving human participants were carried out according to the institutional and/or national research committee's ethical standards and the 1964 Helsinki declaration and its more recent amendments or comparable ethical standards. Informed consent for the surgery was obtained from all subjects.

### Procedure and instrumentation

Conventional FNA biopsy was performed, and aspiration was accomplished with 4 to 8 passes using 10 ml disposable plastic syringes and 22 to 23-gauge needles. Then air-dried and alcohol-fixed smears were sent for histopathological examination. The preoperative FNA biopsy results were categorized as benign, suspicious, malignant, and non-diagnostic.

All surgeries were performed under general anesthesia. After a skin incision of 3 to 4 cm length parallel to the skin crease and 2-3 cm below the mandible, the platysma muscle was divided, and the upper part was carefully pulled upwards. The marginal branch of the facial nerve was identified but not dissected extensively. The facial vein was tied over the submandibular gland, and the retained upper part was gently retracted. If the facial artery was not associated with the mass, it was preserved. The submandibular gland was carefully dissected from the surrounding tissue and excised with its deep part and canal extending under the mylohyoid muscle. The hypoglossal and lingual nerves were routinely identified and preserved. Negative pressure or a penrose was placed in the surgical field and removed on the second postoperative day if no hemorrhage or seroma was observed. Oral amoxicillin/clavulanic acid 2 g/day were administered to all patients, for five to seven days after the operation.

Routine follow-up examinations for malignant cases were performed every 2-3 months in the first and second year, every six months until the end of the fifth years and annually thereafter. A complete head and neck examination was performed with ultrasound at every visit, and chest X-ray or computerized tomography was obtained annually for metastasis screening. For benign cases, a postoperative and a six-month follow-up examination was performed.

### Statistical analysis

Statistical analysis was carried out using computer software (SPSS version 22.0, SPSS Inc. Chicago, IL, USA). The Shapiro-Wilk test was used for determining the distribution pattern of the data. The distribution of the groups was parametric. Descriptive statistics were used to describe the data expressed as "mean±standard deviation." The FNA biopsy's primary outcomes

were the sensitivity, specificity, positive and negative predictive value. The suspicious cytology was regarded as malignant in calculating the sensitivity, specificity, and predictive values.

## RESULTS

One hundred and fifty-eight patients were included in our study. The mean age of the patients was 54.4±14.7 years (range 19-86 years). Eighty-nine of the patients were male (56.3%), and 69 were female (43.7%). The average follow-up time was 88.9±44.0 months (range 8-167).

Regarding FNA cytology results, 26 of the aspirates were benign cytology, seven were malignant cytology, seven were suspicious cytology, and four were non-diagnostic cytology. In three of seven patients with suspicious cytology on FNA results, the final histopathology resulted in malignancy. In one patient with preoperative benign FNA cytology, the final histopathology was low-grade mucoepidermoid carcinoma. The sensitivity of FNA biopsy in our case series was 83.3% (10/12), specificity was 78.1% (25/32), positive predictive value was 71.4% (10/14), and the negative predictive value was 96.1% (25/26). The results of FNA cytology and its correlation with final histopathologic diagnoses are shown in Table 1.

**Table 1: Results of preoperative fine-needle aspiration (FNA) cytology and their correlation with final histopathological diagnoses. Diagnosis of FNA Cytology**

	Number (n)	Final Histopathological Diagnosis	
		Benign (n)	Malign (n)
Benign	26	25	1*
Intermediate/Suspicious	7	4	3
Malign	7	0	7
Non-diagnostic	4	3	1#
Total	44	32	12

\*: Low-grade mucoepidermoid carcinoma

#: Adenoid cystic carcinoma

Regarding final histopathologic diagnoses, 66 patients (41.7%) had sialolithiasis, 47 patients (29.7%) had chronic sialadenitis, 44 patients (27.8%) had submandibular mass, and one patient (0.6%) had IgG4 related inflammatory disease (Kuttner's tumor). Pleomorphic adenoma was found in 30 patients (18.9%) that had been operated on for submandibular mass. Lymphoma was the postoperative histopathological diagnosis in three patients (1.8%), adenoid cystic carcinoma in three patients (1.8%), mucoepidermoid carcinoma in three patients (1.8%), lymphoepithelial cyst in two patients (1.2%), pleomorphic adenoma ex carcinoma in one patient (0.6%), adenocarcinoma in one patient (0.6%) and myoepithelial carcinoma in one patient (0.6%). The final histopathologic diagnoses of the cases are shown in Table 2.

Perineural invasion was observed in postoperative pathology in one patient diagnosed with adenoid cystic carcinoma. This patient received postoperative radiochemotherapy after gland excision. Low-grade mucoepidermoid carcinoma was detected

**Table 2: Final Histopathologic Diagnoses of the Cases**

Histopathological Diagnosis	Number (158)	%
Inflammatory	<b>114</b>	<b>72.1</b>
Sialolithiasis	66	41.7
Chronic Sialadenitis	47	27.8
IgG4 related disease	1	0.6
Benign	<b>32</b>	<b>20.2</b>
Pleomorphic adenoma	30	18.9
Lymphoepithelial cyst	2	1.2
Malignant	<b>12</b>	<b>7.5</b>
Lymphoma	3	1.8
Adenoid cystic Carcinoma	3	1.8
Mucoepidermoid carcinoma	3	1.8
Pleomorphic adenoma ex carcinoma	1	0.6
Adenocarcinoma	1	0.6
Myoepithelial carcinoma	1	0.6

in one, and gland excision was performed. For two patients diagnosed with high-grade mucoepidermoid carcinoma, we performed supraomohyoid neck dissection on both of them in the third postoperative week in addition to the gland excision. In these two patients, lymph node metastasis was not detected in the initial histopathology.

None of the nine patients diagnosed with submandibular gland-derived carcinoma had clinical or pathological lymph node metastasis, and no local recurrence or disease-related mortality was observed during follow-up. In the seventh-year follow-up of the patient diagnosed with adenoid cystic carcinoma with perineural invasion, no recurrence was observed, and the follow-up continued.

Lingual or hypoglossal nerve damage was not observed in any patient. Transient marginal mandibular nerve palsy was observed in 14 patients (8.8%). Only two patients diagnosed with malignancy (1.2%) developed permanent facial nerve marginal mandibular branch palsy. Major arterial hemorrhage and mortality due to operation in the early postoperative period were not detected in any patients. In seven of the 113 patients diagnosed with submandibular sialadenitis and sialolithiasis, purulent discharge from the incision was observed in the early postoperative time. Also, in four patients diagnosed with submandibular sialadenitis and sialolithiasis, seroma formation was observed. All of these 11 patients responded well to the intravenous antibiotic treatment and daily sterile dressings. No additional revision surgery was performed in this patient group. Also, none of the patients developed a saliva fistula at the operation site. None of any patients developed hypertrophic or severely undesired scar formation and underwent scar revision surgery in the postoperative follow-up period.

## DISCUSSION

Salivary gland disorders and their treatment is one of the most frequently encountered diseases in an otolaryngologist's

practice. Since surgery is indicated in most submandibular gland pathologies, excision of the gland is performed in many clinics by both senior and junior surgeons. Our current study represented our clinic's experience of 158 patients who had undergone submandibular gland excision due to different indications. The conventional lateral surgical approach to the submandibular gland has remained the most preferred and standard procedure for over nearly three decades (9), and we were able to review the long-term outcomes of a large group of homogenous patients that we had operated on for 14 years in our clinic.

In the last two decades, minimally invasive surgical procedures have gained importance with better cosmetic outcomes and reduced morbidity in the otolaryngology practice. The smaller incisions parallel to the skin crease are preferred by many (10). On the other hand, intraoral or submental approaches (11, 12) or retroauricular hairline incisions (13) are described to reduce the conventional procedure's cosmetic morbidity. Also, an endoscope assisted submandibular gland excision procedure was reported to be feasible (14). We represent the results of submandibular gland excision via the transcervical approach with the minimal skin incision. Submandibular gland excision is associated with the risk of permanent nerve palsy, reported in around 7% of cases (8). Paralysis of the hypoglossal and lingual nerve is rare (15, 16). In our study, only two patients (1.2%) developed permanent paralysis of the mandibular nerve. There was no patient with permanent lingual or hypoglossal nerve paralysis. Our case series' low nerve paralysis rate was due to the careful identification and meticulous dissection of the facial vein and mandibular branch. We think that optimum surgical view can be achieved with minimal cervical skin incision without harming the marginal mandibular nerve. The submandibular gland can also be removed safely from lingual and hypoglossal nerves with meticulous dissection and gentle retraction of the gland.

In our study, symptomatic sialolithiasis and sialadenitis was the most common indication for submandibular gland resection, similar to previous studies (1, 6, 7, 16). Diagnostic or interventional sialendoscopy can be an option for some cases, but it carries some limitations (17). After the excision of an inflamed submandibular gland, patients can develop a seroma, salivary fistula, or surgical site infection. Also, severe adhesions due to chronic inflammation can be challenging during surgery. We observed surgical site infection and purulent discharge in seven patients and seroma in four patients. None of the patients developed salivary fistula, or undesired scar formation in long term follow-up. We observed that a transcervical approach with minimal skin incision offered safe removal of the inflamed submandibular gland and resulted in less morbidity and favorable cosmetic outcome.

Twelve (27.2%) of the 44 patients who underwent excision due to a mass in the submandibular gland were diagnosed with malignancy. Nine (20.4%) of these patients had a diagnosis of primary submandibular gland-derived carcinoma. None of these nine patients had lymph node metastasis. Perineural invasion was detected in a patient diagnosed with adenoid cystic carcinoma. Adenoid cystic carcinoma is reported as the most common pathological diagnosis, followed by mucoepidermoid

carcinoma (18, 19). The prognosis of adenoid cystic carcinoma varies, and its long-term management can be challenging due to the perineural invasion or distant metastasis. In our patient group, all three patients diagnosed with adenoid cystic carcinoma had early stage diseases without distant or occult metastases, and complete removal of the tumor with the submandibular gland was successful in disease control. Early-stage low-grade mucoepidermoid carcinoma and the other submandibular gland derived carcinomas were treated successfully with submandibular gland excision. We performed selective neck dissection in two of our patients diagnosed with high-grade mucoepidermoid carcinoma. No lymph node metastasis was present at the postoperative histopathology, and disease recurrence was not observed in their third and fifth-year controls. No mortality due to the primary disease was found.

FNA biopsy was performed on patients diagnosed with submandibular mass. The FNA cytology resulted in malign or suspicious in ten of twelve patients, and FNA's sensitivity in submandibular gland malignancy was 83.3% with 71.4% (10/14) positive predictive value. Our results were similar to our studies (4, 20). In one patient, who was later diagnosed with low-grade mucoepidermoid carcinoma, the preoperative FNA cytology was benign. Also, non-diagnostic cytology was reported in an adenoid cystic carcinoma case. The most common benign pathology was pleomorphic adenoma in our case series, similar to the literature (1, 6, 7, 16). FNA cytology specificity was 78.1%, with a high negative predictive value (96.1%). FNA cytology had favorable accuracy in detecting or excluding malignancy in diagnosing a submandibular gland neoplasm, but low patient number meant making a certain conclusion was difficult on its clinic role. Also, since the final histopathological examination always plays a crucial role in selecting the treatment modality, excision of the submandibular originated masses has to be considered as the gold standard procedure. Overall, as a diagnostic tool during the preoperative planning, FNA cytology is a reliable method and mandatory in the evaluation of a submandibular mass.

## CONCLUSION

The transcervical approach to the submandibular gland with minimal skin incision offers safe and reliable removal of the submandibular gland in inflammatory diseases and neoplastic tumors. It also has good postoperative outcomes with a minimal risk of permanent nerve injury. FNA cytology was found to be useful in the preoperative evaluation of the submandibular masses.

**Ethics Committee Approval:** Ethical Statement: Date: 21.01.2021 Decision No: 21-1.1T6 Institution: Ege University Faculty of Medicine Research Ethics Committees.

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## REFERENCES

1. Spiro RH. Salivary neoplasms: overview of a 35-year experience with 2,807 patients. *Head Neck Surg* 1986;8(3):177-84.
2. Pinkston JA, Cole P. Incidence rates of salivary gland tumors: results from a population-based study. *Otolaryngol Head Neck Surg* 1999;120(6):834-40.
3. Frable MA, Frable WJ. Fine-needle aspiration biopsy of salivary glands. *Laryngoscope* 1991;101(3):245-9.
4. Feinstein AJ, Alonso J, Yang SE, St. John M. Diagnostic accuracy of fine-needle aspiration for parotid and submandibular gland lesions. *Otolaryngol Head Neck Surg* 2016;155(3):431-6.
5. Schmidt RL, Hall BJ, Wilson AR, Layfield LJ. A systematic review and meta-analysis of the diagnostic accuracy of fine-needle aspiration cytology for parotid gland lesions. *Am J Clin Path* 2011;136:45-59.
6. Lustmann J, Regev E, Melamed Y. Sialolithiasis. A survey on 245 patients and a review of the literature. *Int J Oral Maxillofac Surg* 1990;19(3):135-8.
7. Marchal F, Dulguerov P. Sialolithiasis management: the state of the art. *Arch Otolaryngol Head Neck Surg* 2003;129(9):951-6.
8. Hernando M, Echarri RM, Taha M, Martin-Fragueiro L, Hernando A, Mayor GP. Surgical complications of submandibular gland excision. *Acta Otorrinolaringol Esp* 2012;63(1):42-6.
9. Beahm DD, Peleaz L, Nuss DW, Schaitkin B, Sedlmayr JC, Rivera-Serrano CM, Zanation AM, Walvekar RR. Surgical approaches to the submandibular gland: a review of literature. *Int J Surg* 2009;7(6):503-9.
10. Hussain A, Murray DP. Modified submandibular sialoadenectomy. *Ear Nose Throat J*. 2004;83(11):768, 770-1.
11. Hong KH, Kim YK. Intraoral removal of the submandibular gland: a new surgical approach. *Otolaryngol Head Neck Surg* 2000;122(6):798-802.
12. Roh JL, Yoon YH, Park C. Removal of nonthyroidal neck masses with improved cosmetic outcomes in skin-lifting surgery. *Otolaryngol Head Neck Surg* 2006;135(3):463-8.
13. Roh JL. Retroauricular hairline incision for removal of upper neck masses. *Laryngoscope* 2005;115(12):2161-6.
14. Baek CH, Jeong HS. Endoscope-assisted submandibular sialadenectomy: a new minimally invasive approach to the submandibular gland. *Am J Otolaryngol* 2006;27(5):306-9.
15. Ichimura K, Nibu KI, Tanaka T. Nerve paralysis after surgery in the submandibular triangle: review of University of Tokyo Hospital experience. *Head Neck* 1997;19(1):48-53.
16. Preuss SF, Klusmann JP, Wittekindt C, Drebber U, Beutner D, Guntinas-Lichius O. Submandibular gland excision: 15 years of experience. *J Oral Maxillofac Surg* 2007;65(5):953-7.
17. Marchal F, Barki G, Dulguerov P, Disant F, Becker M, Lehmann W. Submandibular diagnostic and interventional sialendoscopy: new procedure for ductal disorders. *Ann Otol Rhinol Laryngol* 2002;111(1):27-35.
18. Bhattacharyya N. Survival and prognosis for cancer of the submandibular gland. *J Oral Maxillofac Surg* 2004;62(4):427-30.
19. Aro K, Tarkkanen J, Saat R, Saarihahti K, Mäkitie A, Atula T. Submandibular gland cancer: Specific features and treatment considerations. *Head Neck* 2018;40(1):154-62.
20. Singh Nanda KD, Mehta A, Nanda J. Fine-needle aspiration cytology: a reliable tool in the diagnosis of salivary gland lesions. *J Oral Pathol Med* 2012;41(1):106-12.