PAROTIDECTOMY AND SUBMANDIBULECTOMY IN THE MATERIAL OF THE OTOLARYNGOLOGY CLINIC OF THE MILITARY INSTITUTE OF AVIATION MEDICINE IN THE YEARS 2014-2022. RESULTS OF RETROSPECTIVE ANALYSIS

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 - Introduction: Tumors of the salivary glands constitute 3-6% of head and neck tumors. The majority of these are benign lesions, although recently there has been an observed increase in the incidence of tumors in this region of the body. The aim of the study is to analyze patients operated on for submandibular and parotid gland tumors and to compare the concordance between preoperative fine-needle aspiration biopsy and postoperative histopathological examination.
 - **Methods:** The medical records of all patients who underwent parotidectomy or submandibulectomy were retrospectively analyzed. The data obtained included age, sex, date of surgery, results of preoperative fine-needle aspiration biopsy, and postoperative histopathological examination.
 - **Results:** The medical histories of 146 patients, 77 women and 69 men, with an average age of 53.7 years were analyzed. Histopathological examination revealed that 137 tumours were benign, while 9 cases were malignant. The most frequently diagnosed benign tumors of the parotid gland were pleomorphic adenoma and Warthin's tumor. The most prevalent malignant tumors were mucoepidermoid carcinoma, acinic cell carcinoma, and squamous cell carcinoma. Tumor recurrences were observed in three patients. The concordance between the results of preoperative fine-needle aspiration biopsy and histopathological examination was 83.5%. The sensitivity and specificity in diagnosing tumors were 66.7% and 93.4%, respectively.

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- **Conclusions:** Performing cytological and imaging studies is essential for properly planning the scope of the surgery. The results obtained in this study mostly reflect data from other centers.
 - Keywords: salivary gland, parotid gland, submandibular gland, salivary gland tumors, parotidectomy, submandibulectomy, mucoepidermoid carcinoma, salivary gland tumors, retrospective analysis

INTRODUCTION

Salivary gland tumors constitute 3-6% of head and neck tumors, and up to 8.5% in Europe; occurring in 70% of major and 30% of minor salivary glands [16,24]. They originate from various types of glandular cells, hence their morphological diversity. Over the past forty years, an increase in the incidence of head and neck cancers has been observed, which is associated with alcohol consumption, tobacco use, and the spread of the HPV virus [10]. Smoking increases the risk of developing a benign salivary gland tumor eightfold [3]. Additional etiological factors include specific gene mutations [27]. Treatment options include surgical resection, radiotherapy, chemotherapy, and multimodal therapy. Although tumors are less common in minor salivary glands, their presence in these glands is associated with a 50% risk of malignancy compared to 20% in major glands. Tumors of the sublingual gland are almost always malignant [27].

According to the 5th edition of the WHO classification of salivary gland tumors, there are currently 15 benign and 21 malignant epithelial tumors of the salivary glands, one benign mesenchymal tumor, and two non-neoplastic epithelial lesions [25].

The aim of this study was to analyze patients who underwent surgical removal of the salivary gland at the Otolaryngology Clinic between 2014 and 2022, with a focus on the correlation between the results of fine-needle aspiration biopsy (FNAB) and postoperative histopathological examination (HP).

METHODOLOGY

The study was based on a retrospective singlecenter analysis of 146 patients who underwent removal of the parotid or submandibular gland at the Otolaryngology Clinic of the Military Institute of Aviation Medicine between 2014 and 2022.

Each operation was planned based on FNAB, as well as imaging studies such as ultrasound (USG), computed tomography (CT), and/or magnetic resonance imaging (MRI). Submandibulectomy was performed in a typical manner, without the use of a neurostimulator.

Parotidectomy included superficial lobe (including lateral and partial), deep lobe, and extra-

Analysis of salivary gland removal surgeries	Parotid	Submandibular	Total	
2014	1	4	5	
2015	10	5	15	
2016	22	8	30	
2017	16	5	21	
2018	8	9	17	
2019	15	б	21	
2020	10	2	12	
2021	8	2	10	
2022	13	2	15	
Total number	103	43	146	
Men	41	28	69	
Women	62	15	77	
Age (range; average)	16-83; 54.6	18-77; 51.6	16-83; 53.7	
Left side	46	24	70	
Right side	57	19	76	

Tab. 1. Demographic structure of operated patients.

Comparison of FNAB and HP before and after surgery	Parotid FNAB	Submandibular FNAB	Total	Parotid HP	Submandibular HP	Total
PA	40	5	45	43	6	49
Warthin	30	3	31	38	3	41
Chronic calculi, nonspecific or hypertrophic inflammation	-	-	-	2	27	29
Lipoma	3	-	3	4	-	4
Basal cell adenoma	1	-	1	4	-	4
MEC	-	-	-	1	2	3
Inflammatory cyst	5	-	5	2	1	3
Squamous cell carcinoma	2	-	2	2	-	2
Acinic cell carcinoma	-	-	-	2	-	2
Küttner tumor	-	-	-	-	2	2
Papillary cystadenoma	-	-	-	1	1	2
PA and Warthin	-	-	-	1	-	1
Fibrotic branchial cleft cyst	-	-	-	1	-	1
DLBCL lymphoma	-	-	-	-	1	1
Myoepithelioma	-	-	-	1	-	1
Carcinoma in tumore mixto	-	-	-	1	-	1
No FNAB performed	17	33	49	-	-	-
Adenoid Cystic Carcinoma (AdCC)	1	-	1	-	-	-
Atypia/cancer cells/indeter- minate	5	2	7	-	-	-
Liquid	1	-	1	-	-	-

Tab. 2. Comparison of FNAB and histopathological results.

capsular tumor removal. The European Salivary Gland Society recommended the classification of parotidectomy based on the five-level anatomy of the parotid gland [26], but this analysis did not include it as it predates this classification. The extent of the procedure depended on the initial cytological and imaging diagnosis. Available FNAB and HP results were compared.

RESULTS

The study included 146 salivary gland surgeries, with 77 performed on women and 69 on men. There were 43 submandibular gland surgeries and 103 parotid gland surgeries. The average age of the patients was 53.7 years (\pm 15.7 years; range from 16 to 83 years). The structure of both operated groups is presented in Table 1.

The most common benign tumor in both groups was pleomorphic adenoma (PA) (33.6%), followed by Warthin's tumor (28.1%). Among the histopathological diagnoses of removed parotid glands, mixed tumor (PA) and Warthin's tumor predominated (78.6%), while chronic inflammation prevailed in submandibular glands (62.8%). Among malignant tumors, mucoepidermoid carcinoma (MEC) slightly predominated (2.1%), along with squamous cell carcinoma (1.4%) and acinic cell carcinoma (AciCC) (1.4%).

The concordance between FNAB and HP results reached 83.5%. The sensitivity and specificity in diagnosing tumors were 66.7% and 93.4%, respectively. The decrease in the number of surgeries performed in 2020 and 2021 was due to the SARS-CoV-2 pandemic. The list of preoperative FNAB and postoperative HP results is presented in Table 2.

DISCUSSION

Among the 80% of primary benign tumors, pleomorphic adenoma (PA) is noted in 65%, both in major (55%) and minor salivary glands (50%), with a female predominance of 1:1.4 [21]. In our analysis, among individuals operated on for parotid gland tumors, PA accounted for 41.7%, with a male-to-female ratio of 1:1.87. Although it is a benign tumor, it has a tendency for recurrence and malignant transformation, especially when incompletely excised [14].

One individual underwent three surgeries due to recurrent PA (in 2015, 2020, and 2021), first at the age of 24. After the third surgery, she developed grade III facial nerve palsy according to the House-Brackmann scale. Another patient underwent three surgeries for recurrent PA (in 1994, 2000, and 2010), and the fourth surgery in 2017 at our institution revealed histopathological findings of carcinoma ex pleomorphic adenoma. Warthin tumor accounts for approximately 20% of benign salivary gland tumors, and up to 50% considering only the parotid glands. Although the diagnosis can be made based on FNAB and ultrasound, a high percentage of nondiagnostic results are observed in the literature [9]. Among patients with both FNAB and HP results, the concordance rate was 80.6%, consistent with the sensitivity reported in the literature [17,22]. The development of Warthin tumor is mainly observed in the fifth and sixth decades of life, with a male predominance (2.6:1-10.1:1) [12]. In our analysis, we did not observe such a result; the male-to-female ratio was 1:1.16.

Recently, there has been an increasing trend in the percentage contribution of Warthin tumor (papillary cystadenoma lymphomatosum) in the group of benign salivary gland tumors [8]. Due to the very low risk of malignant transformation, delaying surgery and monitoring the tumor using imaging studies is now acceptable [5,18].

The risk of recurrence of Warthin tumor is low, mainly associated with incomplete tumor resection [20]. Among men, one reoperation for recurrent Warthin tumor was performed 12 years after the initial removal.

The occurrence of multiple synchronous tumors in one parotid gland is observed in up to 5-10% of benign tumors and most commonly involves Warthin tumor [11]. Unilateral synchronous tumors with different histopathological differentiation are less common, and they most often occur in combination with Warthin tumor and pleomorphic adenoma [19]. In our analysis, one such case was noted, where the preoperative FNAB did not clearly define the nature of the lesion.

In four patients, FNAB described pleomorphic adenoma, while histopathological examination confirmed Warthin tumor.

Three individuals were operated on for mucoepidermoid carcinoma (MEC), two of whom had the tumor located in the submandibular gland. MEC is the most widespread malignant salivary gland tumor [13], that is usually located in the parotid gland, accounting for up to 80%, followed by 8-13% in the submandibular gland and 2-4% in the sublingual gland [4].

AciCC accounts for 6-7% of all salivary gland tumors and mainly involves the parotid gland (>90%), making it the second most common malignant tumor in children [23]. Usually occurring in women around the age of 50, AciCC was documented in our material in two women aged 41 and 63 with tumors located in the right parotid gland. One of them was qualified for reoperation, which involved total parotidectomy with facial nerve resection and selective lymphadenectomy II-IV (parotidectomy I-IV (CNVII) with SND II-IV).

Primary squamous cell carcinomas of the salivary glands most commonly occur in the seventh and eighth decades of life and are associated with poor prognosis. Typically, their detection results from metastasis from the throat or skin [1]. Among the patients studied, one case of primary squamous cell carcinoma was noted in a woman, while in another patient, it was a distant metastasis after the removal of squamous cell carcinoma of the prostate. Both tumors were located in the left lobe of the parotid gland at the ages of 75 and 70, respectively.

The most common reason for submandibular gland removal is stone formation causing chronic inflammation [12]. This accounted for 64.3% of the reasons for surgery in the analysis.

Lymphomas account for 2% of salivary gland tumors while being the second most common malignant lesion in the head and neck region [6]. They occur in the parotid gland in 83% of cases, in the submandibular gland in 9%, and in the sublingual gland in 0.3%. The predominant subtypes of benign lymphomas are marginal zone lymphomas and follicular lymphomas, while the more aggressive subtypes include mainly diffuse large Bcell lymphomas (DLBCL), mantle cell lymphomas, and T-cell lymphomas [7]. In the Clinic's material, one case of DLBCL involving the left submandibular gland in a 66-year-old man was detected, and the FNAB was consistent with the histopathological findings.

Küttner tumor, also known as chronic sclerosing sialadenitis of the submandibular gland, classified as an IgG4-related disease, is a benign tumor that was recognized as a distinct entity by the WHO in 1992. Küttner tumor typically affects the submandibular glands and presents as a hard, painless swelling, challenging to distinguish from malignant salivary gland lesions [15]. In the described material, two such diagnoses were obtained in men.

Among the 43 individuals who underwent submandibular gland removal, 33 did not have FNAB performed. In five individuals, a diagnosis of pleomorphic adenoma (PA) was obtained, while in three, Warthin tumor was diagnosed, and in two, atypical neoplastic cells were described. The lack of FNAB was due to the detection of stones in the duct or within the submandibular gland, either palpably or based on ultrasound findings, directly qualifying the patients for surgery.

CONCLUSIONS

Our results regarding the concordance between FNAB and HP were consistent with the available literature data. Proper qualification for surgery with the performance of FNAB and imaging studies is essential to differentiate between benign and malignant tumors and to plan the scope of the operation.

AUTHORS' DECLARATION:

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REFERENCES

- 1. Al-Khafaf AE, Al-Shahrestani F, Baysal Y, Rahbek Gjerdrum LM, Heegaard S, Pedersen LM, Homøe P. Lymphomas of the salivary glands: a systematic review. Acta Oto-Laryngologica. 2023; 21:1-7.
- Baj A, Beltramini GA, Demarchi M, Combi VA, Giannì AB. Bilateral SMAS rhytidectomy in parotid recurrent pleomorphic adenoma. Acta Otorhinolaryngologica Italica. 2011; 31(4): 256.
- 3. Baloch Z, Lubin D, Katabi N, Wenig BM, Wojcik EM. The Milan system for reporting salivary gland cytopathology. InThe Milan system for reporting salivary gland cytopathology. Cham: Springer International Publishing. 2023; 1-13.
- 4. Barsouk A, Aluru JS, Rawla P, Saginala K, Barsouk A. Epidemiology, Risk Factors, and Prevention of Head and Neck Squamous Cell Carcinoma. Medical Sciences. 2023; 11(2): 42.
- 5. Blanco M, Mesko T, Cura M, Cabello-Inchausti B. Chronic sclerosing sialadenitis (Kuttner's tumor): unusual presentation with bilateral involvement of major and minor salivary glands. Annals of diagnostic pathology. 2003; 7(1): 25-30.
- Cabeçadas J, Martinez D, Andreasen S, Mikkelsen LH, Molina-Urra R, Hall D, Strojan P, Hellquist H, Bandello F, Rinaldo A, Cardesa A, Ferlito A. Lymphomas of the head and neck region: an update. Virchows Arch. 2019 Jun;474(6):649-665. doi: 10.1007/s00428-019-02543-7. Epub 2019.
- Cavaliere M., De Luca P., Scarpa A., Savignano L., Cassandro C., Iemma M. Acinic cell carcinoma of the parotid gland: From pathogenesis to management: A literature review. Eur. Arch. Oto-Rhino-Laryngol. 2020; 277: 2673-2679. doi: 10.1007/ s00405-020-05998-6.
- FHKC AB, van Hasselt MMed CA. If cytology of Warthin tumor is accurate, can management be conservative?. Ear, Nose & Throat Journal. 2016; 95(4/5): 185.
- Fois P, Mureddu L, Manca A, Varrucciu S, Crescio C, Gallus R, Rizzo D, Cossu A, Bussu F. Preoperative Diagnosis of Warthin Tumors Combining Cytological, Clinical and Ultrasonographic Information within a Multidisciplinary Approach in a Lump Clinic. J Pers Med. 2023; 13(7): 1075, doi: 10.3390/jpm13071075.
- 10. Franzen A, Koegel K. Synchronous double tumors of the parotid gland. Laryngo-rhino-otologie. 1996; 75(7): 437-40.
- 11. Franzen A, Lieder A, Guenzel T, Buchali A. The heterogenicity of parotid gland squamous cell carcinoma: a study of 49 patients. in vivo. 2019; 33(6): 2001-6.
- Gatta G, Guzzo M, Locati LD, McGurk M, Prott FJ. Major and minor salivary gland tumours. Critical reviews in oncology/ hematology. 2020; 152: 102959.
- 13. Kalwaniya DS, Meena R, Kumar D, Tolat A, Arya SV. A Review of the Current Literature on Pleomorphic Adenoma. Cureus. 2023; 15(7).
- 14. Lin HH, Limesand KH, Ann DK. Current state of knowledge on salivary gland cancers. Critical Reviews™ in Oncogenesis. 2018; 23(3-4).
- 15. Ochal-Choińska A, Bruzgielewicz A, Osuch-Wójcikiewicz E. Synchronous multiple unilateral parotid gland tumors of benign and malignant histological types: Case report and literature review. Braz J Otorhinolaryngol 2016. doi: 10.1016/j.bjorl.2016.03.002.
- 16. Neville WB, Damm DD, Allen CM, et al. Salivary gland pathology. 2nd edn. Oral And Maxillofacial Pathology. 2002: 420-3.
- 17. Nishimura H, Kawata R, Kinoshita I, Higashino M, Terada T, Haginomori SI, Tochizawa T. Management for Warthin Tumor of the Parotid Gland: Surgery or Observation. A 21-Year Retrospective Study of 387 Cases. Ear, Nose & Throat Journal. 2022: 01455613221080927.
- Pires FR, Chen SY, Da Cruz Perez DE, et al. Cytokeratin expression in central mucoepidermoid carcinoma and glandular odontogenic cyst. Oral Oncology 2004; 40: 545-51.

- 19. Psychogios G, Vlastos I, Thölken R, Zenk J. Warthin's tumour seems to be the most common benign neoplasm of the parotid gland in Germany. European Archives of Oto-Rhino-Laryngology. 2020; 277: 2081-4.
- Quer M, Hernandez-Prera JC, Silver CE, Casasayas M, Simo R, Vander Poorten V, Guntinas-Lichius O, Bradley PJ, Tong-Ng W, Rodrigo JP, Mäkitie AA. Current trends and controversies in the management of Warthin tumor of the parotid gland. Diagnostics. 2021; 11(8): 1467.
- 21. Rai M, Burman R. Giant submandibular sialolith of remarkable size in the comma area of Wharton's duct: a case report. Journal of Oral and Maxillofacial Surgery. 2009; 67(6): 1329-32.
- 22. Reinheimer A, Vieira DSC, Cordeiro MMR, Rivero ERC. Retrospective study of 124 cases of salivary gland tumors and literature review. J Clin Exp Dent. 2019; 11(11): e1025–32.
- 23. Teymoortash A., Krasnewicz Y., Werner J.A. Clinical Features of Cystadenolymphoma (Warthin's Tumor) of the Parotid Gland: A Retrospective Comparative Study of 96 Cases. Oral Oncol. 2006; 42: 569-573. doi: 10.1016/j.oraloncology.2005.10.017.
- 24. Skalova A, Hyrcza MD, Mehrotra R, eds. WHO Classification of Tumours Editorial Board. Head and neck tumours [Internet; beta version ahead of print]. Lyon, France, 2022, International Agency for Research on Cancer, (WHO classification of tumours series, 5th ed.; vol. 9). Available from: https://tumourclassification.iarc.who.int/chapters/52.
- 25. Valstar MH, De Ridder M, van den Broek EC, Stuiver MM, Van Dijk BA, Van Velthuysen ML, Balm AJ, Smeele LE. Salivary gland pleomorphic adenoma in the Netherlands: A nationwide observational study of primary tumor incidence, malignant transformation, recurrence, and risk factors for recurrence. Oral oncology. 2017; 66: 93-9.
- 26. Wierzbicka M, Piwowarczyk K, Nogala H et al. Czy potrzebna jest nowa klasyfikacja zabiegów chirurgicznych na gruczołach ślinowych?. Otolaryngol Pol 2016; 70(3): 8-13. doi: 10.5604/00306657.1202390.
- 27. Yoo GH, Eisele DW, Driben JS, Johns ME, Askin FB. Warthin's tumor: A 40-year experience at the johns hopkins hospital. The Laryngoscope. 1994; 104(7): 799-803.

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