OPEN ACCESS

# Greenfort International Journal of Applied Medical Science

Abbreviated Key Title: Grn Int J Apl Med Sci | ISSN 3048-6904(P) | ISSN 2584-2420(O) |
Greenfort International Publisher, Assam, India
Journal homepage: https://gipublisher.com/journals/gijams

**Research Article** DOI: 10.62046/gijams.2024.v02i05.006

## Salivary Gland Tumours in Children and Adolescents-A Study in Shaheed Monsur Ali Medical College & Hospital, Uttara, Dhaka, Bangladesh

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**Abstract:** **Introduction:** Salivary gland carcinomas (SGCs) are rare neoplasms in adults, but are exceedingly infrequent in children, with a reported annual incidence of 0.8–1.4 per million populations under 20 years old. Like other pediatric very rare tumors, SGCs in children and adolescents often present diagnostic and therapeutic challenges for pathologists, surgeons, and pediatric oncologists. **Objective:** To assess the salivary gland tumours in children and adolescents. **Methods:** A cross-sectional observational study was carried out at Dept. of ENT, Shaheed Monsur Ali Medical College & Hospital, Uttara, Dhaka, Bangladesh from January to June 2023. Total 60 patients included in our study. Our pre-operative clinical diagnoses were based on size, shape, mobility, fixity of the tumour to the underlying structures, ulceration of overlying skin, involvement of facial nerve by its malignant transformation and fine needle aspiration cytology. All patients, or their guardians, gave their informed consent for data collection within the national rare tumour group and analysis. **Results:** Total sixty (60) cases of salivary gland tumours were available for our study. Out of 55% were male and 45% were female patients. Incidence of tumours is highest in the age group of 41-50 with 13 cases (21.6% of the total). The least affected age group was the 0-10 group with only one patient (3.3%). Women accounted for 16 cases of the total 33 benign neoplasias (55% of the total). Of the 33 benign tumour, 18 were in males (54.5% of all benign tumours) and 15 were in females (45.5%). Total 60 operated cases). Of these, 32 were pleomorphic adenomas (53.3%) and 1 was an adenolymphoma (1.6%). the total number of malignant tumours confirmed by HPE were 27 (45%). Of these, most numerous was adenoid cystic carcinoma in 8 cases (13.3%). squamous cell carcinoma and muco epidermoid carcinoma were found in 6 cases each (10%). There were 4 and 3 cases of adenocarcinoma (6.6%) and acinic cell carcinoma (5%) respectively. The parotid glands were also the sole location for the 3 acinic cell carcinomas. No tumours were found in the sublingual gland. The majority of the tumours found in the parotid gland were benign tumours, mainly pleomorphic adenomas with 24 cases (75% of the total parotid tumours). **Conclusions**: The tumours were equally distributed between males and females. The tumours were most commonly found in the third to fifth decades of life. The most common benign tumour and the most common tumour overall was pleomorphic adenoma making up 54.5% of all tumours. Surgical excision was the most commonly done treatment. The most commonly done surgical procedure was superficial parotidectomy.

**Keywords:** lower third molar, mandibular canal, impacted, orthopantomogram, retrospective.

**Citation:** Atiur Rahman. Salivary Gland Tumours in Children and Adolescents-A Study in Shaheed Monsur Ali Medical College & Hospital, Uttara, Dhaka, Bangladesh. Grn Int J Apl Med Sci, 2024 Sep-Oct 2(5): 197-204.

## INTRODUCTION

Salivary gland carcinomas (SGCs) are rare neoplasms in adults but very rare in children. The annual incidence is 0.8–1.4 per 1 million inhabitants under the age of 20 [1]. Their incidence in the pediatric population does not exceed 0.8 per 1,000,000 [2], accounting for less than 10% of all head and neck cancers in children [3]. As with other very rare tumors in children, SGCs in children and adolescents often pose diagnostic and therapeutic challenges for pathologists, surgeons, and pediatric oncologists. In the absence of specific standardized pediatric guidelines, treatment is often determined on a case-by-case basis based on guidelines validated for adult patients. However, some differences have been reported between children and adults regarding the histological spectrum and clinical behavior of SGCs [4]. When treating cancers in children and adolescents, special consideration should also be given to possible long-term effects [5]. What SGC has in common with other very rare pediatric tumors (VRTs) is not only its low incidence but also that it has always been an “orphan” disease, i.e. few clinical and biological details are available and specific protocols to help pediatric oncologists and surgeons define the best treatment have not been developed for a long time. Pediatric SGC is often diagnosed in the second decade of life (median age 15 years) [6, 7]. It is very rare under 10 years of age, but it is more likely to

be highly malignant and lead to a poor prognosis [8]. Although cases of familial aggregation of SGC have been reported, no genetic predisposition syndromes have been reported, and the etiology of SGC may be related to multiple susceptibility genes and/or environmental factors [9-11]. A personal history of cancer and its treatment are considered to be potential risk factors for the development of SGC [12]. However, a clear association with increased risk could only be demonstrated for previous radiation exposure [13-15]. Atypical distribution of tumor sites has been noted in some of the above studies. The high proportion of palatal tumors in B. minor glands suggests selection bias [13], while other cohorts included young adults and patients with benign tumors such as pleomorphic adenoma [16]. In addition, concerns regarding treatment-related late effects (especially those related to radiation therapy in survivors) may affect the treatment approach in different age groups. The differential diagnosis of parotid tumors includes benign adenomas (e.g., pleomorphic adenoma) and metastases of other cancers such as lymphoma, sarcoma, and neuroblastoma. In contrast to adults, salivary gland tumors in children are more often malignant (10–25% vs. 50%), and therefore a possible or probably malignant diagnosis should be considered for noninflammatory salivary gland masses to avoid diagnostic and therapeutic abuse [17,18].

## MATERIALS & METHODS

**Methods:** A cross-sectional observational study was carried out at Dept. of ENT, Shaheed Monsur Ali Medical College & Hospital, Uttara, Dhaka, Bangladesh from January to June 2023. Total 60 patients included in our study. Our pre-operative clinical diagnoses were based on size, shape, mobility, fixity of the tumour to the underlying structures, ulceration of overlying skin, involvement of facial nerve by its malignant transformation and fine needle aspiration cytology. All patients, or their guardians, gave their informed consent for data collection within the national rare tumour group and analysis. Histological diagnoses were made by local pathologists and centrally reviewed by national reference committees. Histological diagnoses (histological type and grade) were classified according to the 2005 World Health Organization (WHO) Classification of Tumors of the Head and Neck and the 2008 Armed Forces Institute of Pathology (AFIP) Atlas of Tumor Pathology - Tumors of the Salivary Glands. Disease stage was determined according to the Union for International Cancer Control (UICC) Tumor-Node-Metastasis (TNM) staging system (8th edition).

In the absence of specific pediatric guidelines, treatment was based on the recommendations of national VRT groups, which were not yet uniform during the period described here. In addition, some patients were treated based on the decisions of their local tumor boards, and contact with national bodies was only made after surgery. After preoperative evaluation, extensive tumor removal with sialodenotomy was recommended. In cases of primary parotid gland involvement, partial or total parotidectomy (depending on tumor size and anatomical extent) with the aim of achieving free margins was the mainstay of treatment. Referral to a surgical center with an experienced pediatric surgeon or ENT surgeon specializing in the treatment of SGC was strongly recommended. Tumor resection with simultaneous facial nerve monitoring was recommended.

## RESULTS

Total sixty (60) cases of salivary gland tumours were available for our study. Incidence of tumours is highest in the age group of 41-50 with 13 cases (21.6% of the total). Out of 55% were male and 45% were female patients. The 21-30 and 31-40 age groups are the second most afflicted group with 14 cases (23.3%) each. The least affected age group was the 0-10 group with only one patient (3.3%). The youngest patient was a male child of age 5 years and the oldest was a female patient of age 75 years. The breakup between age groups among males also follows that of the total distribution with majority of cases numbering in the age group 21-30, 31-40 (table-1).

**Table-1: Distribution of the patient's age**

| Age Group in Years | N | % |
|---|---|---|
| 0-10 | 2 | 3.33 |
| 11-20 | 5 | 8.3 |
| 21-30 | 14 | 23.3 |
| 31-40 | 14 | 23.3 |
| 41-50 | 13 | 21.6 |
| 51-60 | 7 | 11.6 |
| 61-70 | 2 | 3.3 |
| 71 And above | 3 | 5.0 |

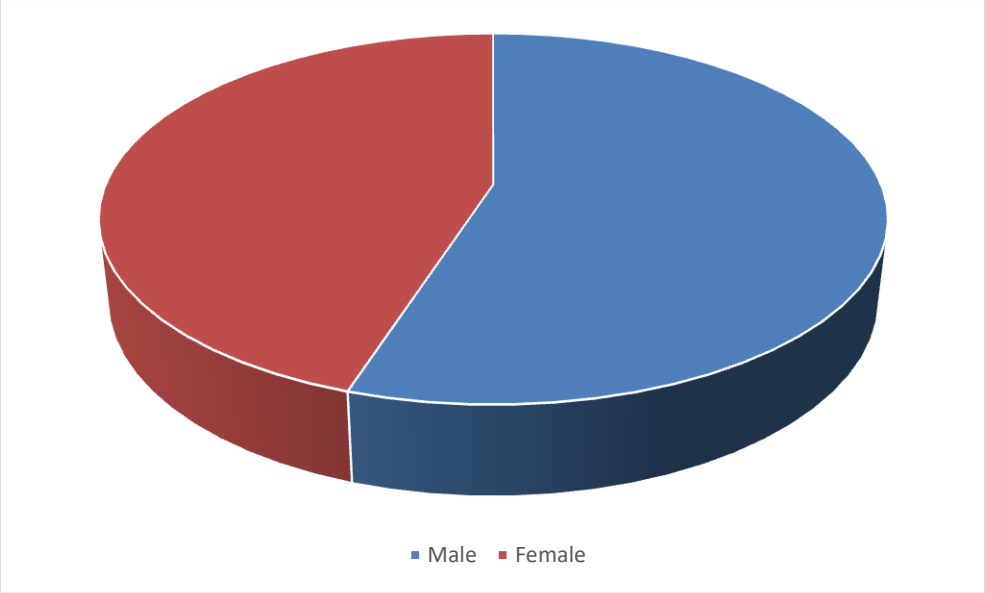



**Fig-1: Sex distribution of the study patients.**

**Table-2: Showing the Age Distribution of HPE Proven Benign and Malignant Tumours**

| Age Group | Benign | | Malignant | |
|---|---|---|---|---|
| | Male | Female | Male | Female |
| 0-10 | 1 | - | 1 | - |
| 11-20 | 2 | 1 | 1 | 1 |
| 21-30 | 6 | 4 | 2 | 2 |
| 31-40 | 3 | 3 | 5 | 3 |
| 41-50 | 2 | 5 | 4 | 2 |
| 51-60 | 2 | 2 | 2 | 1 |
| 61-70 | 1 | - | - | 1 |
| >70 | 1 | - | - | 2 |

Women accounted for 16 cases of the total 33 benign neoplasias (55% of the total). Men had the remaining18 cases (54.4%). So, the female: male ratio is 1.2:1 in case of benign tumours. Men accounted for 15 out of the total 27 cases of malignant tumours (55.5% of the total). Women made up the rest 12 cases (44.4%) the men to women ratio are 1.25:1. Overall, most of the benign cases are found in the 21-30 age group. The maximum number of malignant cases was found in the 31-50 age group with 13 cases (48.1% of the total 27 cases) (table-2).

**Table-3: Showing the distribution of HPE confirmed benign and malignant tumours among the sexes**

| Sex | Male | Female | Total Tumour Types |
|---|---|---|---|
| Benign | 18 | 15 | 33 |
| Malignant | 15 | 12 | 27 |
| Total tumours among sexes | 33 | 27 | 60 |

Table-3 shows that, of the 33 benign tumour, 18 were in males (54.5% of all benign tumours).15 were in females (45.5%). 15 of the 27 cases of malignant tumours were in males (55.5% of all malignancies). 12 cases were found in females (44.4%). Of the 27 cases in females, there were 15 benign (55.5%) and 12 malignant tumours (44.4%). Of the 33 operated cases in males, there were 18 (54.5%) and 15 malignant tumours (45.4%).

**Table-4: Showing the incidence of individual lesions as confirmed by HPE in the operated cases**

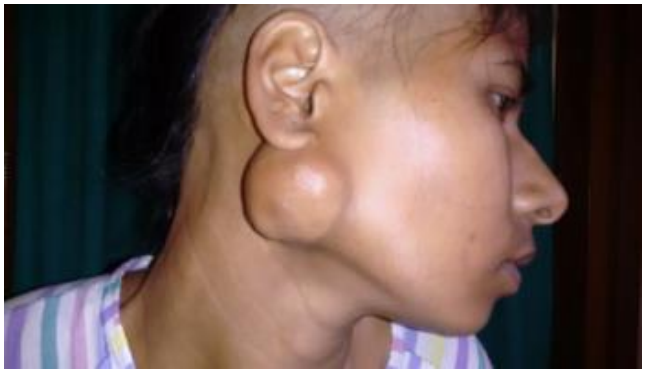
| Type | Total No. Of Cases | No. Of Individual Cases - % Of Total 60 Operated Cases | Percentage (%) Of The 60 Operated Cases |
|---|---|---|---|
| Benign | 33 | Pleomorphic Adenoma –32-53.3 | 55.0 |
| | | Adenolymphoma –1-1.7 | |
| Malignant | 27 | Adenoid Cystic Carcinoma –8-13.3 | 45.0 |
| | | Squamous Cell Carcinoma –6-10.0 | |
| | | Muco-Epidermoid Carcinoma–6-10.0 | |
| | | Adenocarcinoma –4-6.6 | |
| | | Acinic Cell Carcinoma –3-5.0 | |

Of the 60 operated cases, HPE confirmed 33 cases as benign (55% of the total 60 operated cases). Of these, 32 were pleomorphic adenomas (53.3%) and 1 was an adenolymphoma (1.6%). The total number of malignant tumours confirmed by HPE were 27 (45%). of these, most numerous was adenoid cystic carcinoma in 8 cases (13.3%). Squamous cell carcinoma and muco epidermoid carcinoma were found in 6 cases each (10%). There were 4 and 3 cases of adenocarcinoma (6.6%) and acinic cell carcinoma (5%) respectively.

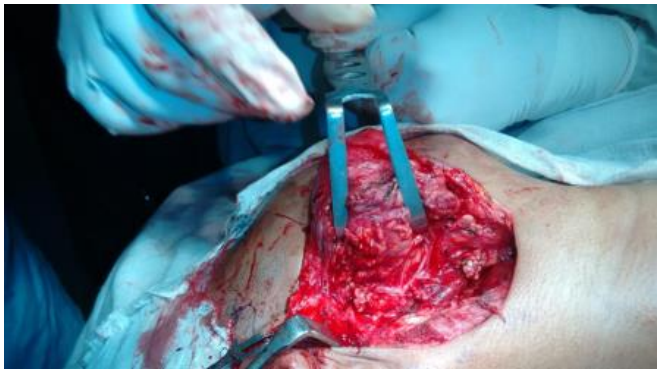
**Table-5: Showing the distribution of the HPE confirmed tumours depending on their site**

| Histopathology | Parotid | Submandibular Gland | Sublingual Gland | Minor Salivary Gland | Total |
|---|---|---|---|---|---|
| Pleomorphic Adenoma | 24 | 7 | - | 1 | 32 |
| Adenolymphoma | 1 | - | - | | 1 |
| Adenocystic Carcinoma | 3 | - | - | 5 | 8 |
| Squamous Cell Carcinoma | 5 | 1 | - | - | 6 |
| Muco-Epiderrmoid Carcinoma | 6 | - | - | - | 6 |
| Adenocarcinoma | 1 | 3 | - | - | 4 |
| Acinic Cell Carcinoma | 3 | - | - | - | 3 |
| Total | 34 | 9 | - | 5 | 60 |

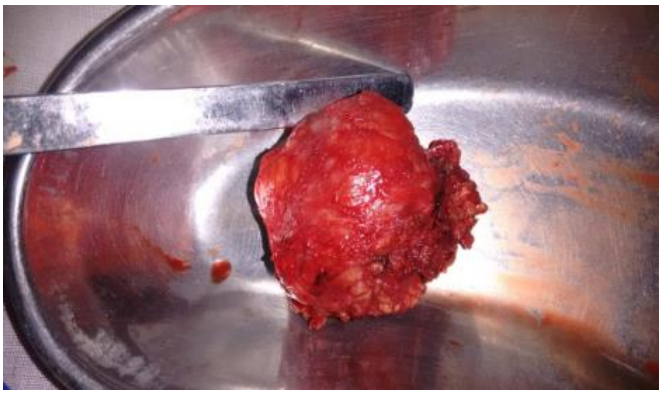
Of the 32 cases of benign pleomorphic adenoma, 24 were in the parotid gland, 7 in the submandibular gland and 1 in the minor salivary gland. The only adenolymphoma was in the parotid gland. The minor salivary glands were the site for 5 cases of adenoid cystic carcinoma while the parotid gland made up the other 3 cases. Of the 4 cases of squamous cell carcinoma, 5 were in the parotid gland and 1 was in the submandibular salivary gland. The 6 cases of muco-epidermoid carcinoma were found exclusively in the parotid gland. The parotid glands were also the sole location for the 3 acinic cell carcinomas. No tumours were found in the sublingual gland. The majority of the tumours found in the parotid gland were benign tumours, mainly pleomorphic adenomas with 24 cases (75% of the total parotid tumours).



**Pic-1: Parotid Pleomorphic Adenoma**



**Pic-2: Intra-Operative Picture of Parotid Pleomorphic Adenoma**



**Pic-3: Specimen Picture of Parotid Pleomorphic Adenoma**

## DISCUSSION

Salivary gland cancer (SGC) is rare in children and adolescents. Therefore, there are no standardized recommendations for the diagnosis and treatment of pediatric SGC, and pediatric oncologists and surgeons usually follow adult guidelines. Inflammation of these glands is called sialadenitis. Salivary gland disease is not common in children. If there is a problem, it is usually due to a bacterial or viral infection. Both types cause painful swelling in the affected gland, which can make eating difficult. Salivary gland carcinoma (SGC) is a rare neoplasm in adults but is very rare in children. The annual incidence is 0.8-1.4 cases per million inhabitants under the age of 20 years [11]. Sialolithiasis or lithiasis is the most common benign disease affecting the salivary glands. Its prevalence is estimated at 1.2%. The overall prognosis of primary SGC in children and adolescents is good. The 10-year overall survival (OS) is more than 90% [19]. However, locoregional recurrence and distant metastases are not uncommon, and survival rates for recurrent patients vary greatly depending on the histological type and grade of the tumor. Poor outcomes after salvage therapy strongly support the need for appropriate initial treatment with an aggressive approach, especially for high-risk tumors. However, overtreatment of low-risk tumors must be avoided because of the potential risk of late side effects. Nevertheless, prognostic stratification of pediatric patients with SGC remains a major challenge due to the rarity of these tumors, the difficulty of accurate histological grading, and the overlap between histological and intrahistologic subtypes. Sixty cases of salivary gland tumors were available for our study. Incidence of tumours is highest in the age group of 41-50 with 13 cases (21.6% of the total). The 21-30 and 31-40 age groups are the second most afflicted group with 14 cases (23.3%) each. The least affected age group was the 0-10 group with only one patient (3.3%). The youngest patient was a male child of age 5 years and the oldest was a female patient of age 75 years. The breakup between age groups among males also follows that of the total distribution with majority of cases numbering in the age group 21-30, 31-40. A study in Brazil by Solanger (2005) on 493 cases of salivary gland tumours resulted in a female: male ratio of 1.25:1[20]. Another study carried out in Brazil by Felipe et al (2012) on 245 cases obtained a female: male ratio of 1.5:1[21]. Another study done in India by maya et al (2014) on 30 cases of intraoral salivary gland tumours had only ten cases as males making the female: male ratio of 2:1[22]. Women accounted for 16 cases of the total 33 benign neoplasias (55% of the total). Men had the remaining18 cases (54.4%). So, the female: male ratio is 1.2:1 in case of benign tumours. Men accounted for 15 out of the total 27 cases of malignant tumours (55.5% of the total). Women made up the rest 12 cases (44.4%) the men to women ratio are 1.25:1. Overall, most of the benign cases are found in the 21-30 age group. The maximum number of malignant cases was found in the 31-50 age group with 13 cases (48.1% of the total 27 cases). In the other Brazilian Study by Felipe et al, they found a female preponderance amongst both benign and malignant cases. The female to male ratio in benign neoplasms was 1.25:1 while in

malignancies it was 1.11:1 paralleling our study [23]. In our study benign tumours were not discovered in the first decade of life. This is in keeping with the findings of Solanger et al [24]. Similarly, we did find two cases of malignant neoplasms in the first two decades of life, one of them was however lost for follow-up so it was not histopathologically proven and the other was a case of adenocarcinoma in the parotid gland in a female. This was in contrast to their study. Of the 33 benign tumour, 18 were in males (54.5% of all benign tumours). 15 were in females (45.5%). 15 of the 27 cases of malignant tumours were in males (55.5% of all malignancies). 12 cases were found in females (44.4%). Of the 27 cases in females, there were 15 benign (55.5%) and 12 malignant tumours (44.4%). Of the 33 operated cases in males, there were 18 (54.5%) and 15 malignant tumours (45.4%). This is in keeping with the results reported by most studies as mentioned by Barnes et al [25]. A high incidence of pleomorphic adenoma was found in patients of the fourth and sixth decade was reported by Eneroth (1971) [26]. Studies by Potdar et al [27] and Verma et al., report a male preponderance in their studies. On the contrary, Castro and his associates (1972) reported a high incidence in children and young adults [28]. Of the 60 operated cases, HPE confirmed 33 cases as benign (55% of the total 60 operated cases). of these, 32 were pleomorphic adenomas (53.3%) and 1 was an adenolymphoma (1.6%). the total number of malignant tumours confirmed by HPE were 27 (45%). of these, most numerous was adenoid cystic carcinoma in 8 cases (13.3%). squamous cell carcinoma and muco epidermoid carcinoma were found in 6 cases each (10%). There were 4 and 3 cases of adenocarcinoma (6.6%) and acinic cell carcinoma (5%) respectively. This is in keeping with most studies by workers such as khan et al, Bradley et al, Terhaard et al and Watkinson et al., which mentions that about 30 percent of salivary malignancies is made up of adenoid cystic carcinomas. Most of these arise in the minor salivary gland, about 60 percent. Between 25 to 33 percent arise in the parotid gland making a very small proportion of the parotid gland neoplasms [29-32]. According to literature, this tumour is the most common malignant tumour of the submandibular salivary gland, however we found no cases of submandibular salivary gland adenoid cystic carcinomas in our study. Furthermore, it should account for approximately 10—12% of all malignant salivary gland tumours, which is much less than our 43.58%. Of the 32 cases of benign pleomorphic adenoma, 24 were in the parotid gland, 7 in the submandibular gland and 1 in the minor salivary gland. The only adenolymphoma was in the parotid gland. The minor salivary glands were the site for 5 cases of adenoid cystic carcinoma while the parotid gland made up the other 3 cases. Of the 4 cases of squamous cell carcinoma, 5 were in the parotid gland and 1 was in the submandibular salivary gland. A single case of the tumour was detected in the submandibular salivary gland making up 11.11% of all cases of submandibular salivary gland tumours. As quoted in scott-brown, squamous cell carcinoma is very unusual representing less than 1.1% of all salivary gland tumours [33]. The findings of our study in contrast to most studies as adenocarcinoma is the second, as quoted by Barnes et al [25] or the third, as earlier quoted by Irving et al [33] most common salivary gland malignancy. Most studies have also found the same incidence of over half of the submandibular salivary gland tumours being pleomorphic adenomas. The most common site for the minor salivary gland tumours in our study was the palate, which is in keeping with most of the results in literature such as those by Waldron et al, Toida et al and Yih et al, that mentions more than 33 percent of intraoral malignant tumours of salivary gland to be located in the palate [34-36]. Most work shows that minor salivary gland tumours are majorly found in females, except in case of adenoid cystic carcinoma which has no sex predilection. In our study too, the single minor salivary gland pleomorphic adenoma was found in a female and the four adenocystic carcinoma was equally divided between the two sexes. Tumours of sublingual glands are extremely rare as reported by Eveson et al and Perez et al [37,38]. We too did not find any sublingual salivary gland tumours in our study.

## CONCLUSIONS

We came to the following conclusions: A total of 50 salivary gland tumors were examined in this study. The tumors were evenly distributed between men and women. The tumors most often appeared between the third and fifth decades of life. Benign tumors were more common than malignant tumors, accounting for 55% of all tumors. Benign tumors most often appeared in the third decade of life, while malignant tumors most often appeared in the fourth and fifth decades of life. The most common benign tumor and the most common tumor overall was pleomorphic adenoma, accounting for 54.5% of all tumors. The most commonly performed treatment was surgical excision. The most commonly performed surgical procedure was superficial parotidectomy.

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