

# Primary epithelial salivary gland tumours in children and adolescents

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**Abstract.** Primary epithelial tumours of the salivary glands are very rare in paediatric patients. The aim of this study was to evaluate the clinical course, treatment, and outcomes of these uncommon neoplasms based on the authors' experience and the recent literature. The medical charts of 12 female patients and seven male patients with primary epithelial salivary gland tumours were reviewed. All were under 19 years of age and underwent surgical treatment between 1994 and 2016. The results of this group of paediatric patients were compared with those of 621 adult patients. The two most common tumours in the paediatric patients were pleomorphic adenoma and mucoepidermoid carcinoma (89.4%;  $P = 0.004$ ). The incidence of facial nerve palsy following surgery of the parotid tumours was similar in the two groups ( $P = 1.000$ ). The most common primary cancer in the paediatric group was mucoepidermoid carcinoma (77.8%), while in the adult group, adenoid cystic carcinoma was most common ( $P < 0.001$ ). The paediatric group had only low-grade cancers in early stages ( $P < 0.001$ ), with an overall 5-year survival rate of 100%. These results show that the incidence of malignant salivary gland tumours is higher in paediatric patients than in adult patients. This should be taken into account during diagnosis and therapy.

**Key words:** salivary gland tumours; pleomorphic adenoma; mucoepidermoid carcinoma; children; adolescents.

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Salivary gland tumours are a heterogeneous group of neoplasms that can present diagnostic and therapeutic challenges for both the pathologist and surgeon. The incidence of all salivary gland tumours varies from 0.4 to 13.5 cases per 100,000 population, and close to 80% of these cases are benign<sup>1</sup>. Thus salivary malignancies are quite rare; they comprise only 0.3% of all malignancies and 6% of

head and neck cancers in the USA<sup>1</sup>. Even rarer are malignant epithelial tumours of the salivary glands in paediatric patients, with an annual incidence of 0.8 cases per 1 million children and adolescents<sup>2</sup>. In fact, most salivary gland neoplasms occur in adults, with less than 5% developing in children and adolescents<sup>3</sup>.

The distribution of benign and malignant tumours differs in adults and

children. In paediatric patients, malignant neoplasms can reach 73% of epithelial salivary gland tumours<sup>4</sup>. For this reason, knowledge regarding the pathological aspects and treatment options in this group of patients is very important. A few large series of salivary gland tumours in paediatric patients have been published in the English language literature. However, only Sultan et al. compared the results from a

group of children and adolescents to a group of adult patients suffering from malignant salivary gland tumours<sup>2</sup>.

This study presents 23 years of experience with primary benign and malignant salivary gland tumours in patients under 19 years of age. Additionally, a review of the recent literature was conducted in order to evaluate the clinical course, treatment, and outcomes of these rare neoplasms.

## Materials and methods

Between January 1994 and July 2016, 776 patients were treated for salivary gland tumours in the Department of Cranio-Maxillofacial Surgery of the Jagiellonian University in Cracow. The tumours were primary epithelial salivary gland neoplasms in 640 cases, 19 (2.9%) of which were diagnosed in children and adolescents. One 15-year-old girl was excluded from the study due to a secondary tumour in the parotid gland (metastasis of the mucoepidermoid carcinoma of the upper eyelid).

The medical charts of the patients were evaluated according to demographic characteristics, clinical presentation, histopathological aspects, methods of treatment, recurrences, follow-up, and outcomes. Results from the group of paediatric patients were compared with those of 621 adults older than 19 years of age. This study was approved by the institutional review board. Because only medical files were obtained, the review board approved the study without the need for patient consent as long as all personal information was kept confidential and any facial features or other identifying marks were removed and/or covered. In addition to the review of medical charts, the English language literature published from January 2000 to July 2016 was reviewed to identify epidemiological data for these uncommon tumours.

An analysis of the relationship between clinicopathological parameters in both groups of patients was performed for categorical variables by means of the  $\chi^2$  test and Fisher's exact test for small groups. All analyses were performed using R software version 3.3.1 (R Foundation for Statistical Computing) and a *P*-value of <0.05 was considered statistically significant.

## Results

The paediatric group comprised 12 female patients and seven male patients (female to male ratio 1.7:1). The age of the patients

ranged from 8 to 18 years, with an average of 15.9 years. The mean age of patients with benign tumours was 15.6 years and of patients with malignant tumours was 16.3 years. All of the patients were of white ethnicity. Tumour size varied from 7 mm to 36 mm in diameter, with an average of 22.6 mm. The mean duration of signs and symptoms of the tumour before surgery was 14 months, ranging from 3 months to 48 months. Patient characteristics and the comparison with those of adults are presented in Table 1.

Major and minor salivary gland tumours were observed almost equally in children and adolescents. When compared with the adult population, a tendency for higher involvement of the minor salivary glands was observed in the paediatric group (*P* = 0.078). Malignancies also occurred more frequently in the paediatric group (*P* = 0.165). Still, these differences were not statistically significant. The parotid gland was the most common site of origin in both groups. The incidence of facial nerve palsy following surgery of the parotid tumours was similar in the two groups (*P* = 1.000).

When comparing the paediatric group with the adult group, a different distribution of histological types was observed. In children and adolescents, the majority of tumours were pleomorphic adenoma and mucoepidermoid carcinoma (89.4%, *P* = 0.004).

The most common primary epithelial tumour was pleomorphic adenoma, which comprised 52.6% (10/19) of all tumours and 100% (10/10) of benign tumours in young patients. Radical excision was performed in 70% (7/10) of patients, and three (30%) patients underwent a second operation due to inadequate surgical margins. None of the patients had local recurrence during a mean follow-up time of 43 months (Table 2). The most common site of pleomorphic adenoma was the parotid gland.

A different distribution of malignant tumours was also found when comparing the two age groups of patients. The most common primary cancer in children and adolescents was mucoepidermoid carcinoma (7/9; 77.8%), whereas in adults, the most common cancer was adenoid cystic carcinoma (35.1%), followed by

Table 1. Characteristics of the paediatric patients and comparison to adults; results are presented as the number of patients (%).

Characteristic	Children/adolescents	Adults	<i>P</i> -value
Sex			
Female	12 (63.2)	353 (56.8)	0.755
Male	7 (36.8)	268 (43.2)	
Site			
Major salivary gland	10 (52.6)	457 (73.6)	0.078
Minor salivary gland	9 (47.4)	164 (26.4)	
Tumour type			
Benign	10 (52.6)	436 (70.2)	0.165
Malignant	9 (47.4)	185 (29.8)	
Histological type			
Pleomorphic adenoma	10 (52.6)	289 (46.5)	0.004
Warthin's tumour	0 (0)	95 (15.3)	
Mucoepidermoid carcinoma	7 (36.8)	39 (6.3)	
Adenoid cystic carcinoma	0 (0)	65 (10.5)	
Others	2 (10.5)	133 (21.4)	
Benign tumours	<i>n</i> = 10	<i>n</i> = 436	
Pleomorphic adenoma	10 (100)	289 (66.3)	0.057
Others	0 (0)	147 (33.7)	
Malignant tumour	<i>n</i> = 9	<i>n</i> = 185	
Mucoepidermoid carcinoma	7 (77.8)	39 (21.1)	<0.001
Others	2 (22.2)	146 (78.9)	
Stage	<i>n</i> = 9	<i>n</i> = 152 <sup>a</sup>	
I	6 (66.7)	20 (13.2)	<0.001
II	3 (33.3)	29 (19.1)	
III	0 (0)	26 (17.1)	
IV	0 (0)	77 (50.6)	
Facial nerve palsy	<i>n</i> = 8	<i>n</i> = 412	
Yes	2 (25.0)	91 (22.1)	1.000
No	6 (75.0)	321 (77.9)	

<sup>a</sup> 33 patients without stage assessment.

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