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Tumors in the parotid are not relatively more often malignant in children than in adults

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ABSTRACT

Introduction: Tumors of the parotid gland in children are rare and very little data has been published regarding the incidence of these tumors. We present a nationwide survey on this topic.
Methods: Data regarding benign and malignant tumors in the parotid gland in children from January 1st, 1990 to December 31st, 2005 in Denmark was collected retrospectively from nationwide registries. This generated 61 patients for inclusion in this study.
Results: 85% of the tumors were benign and the malignant tumors made up the last 15%. The most common of the malignant tumors was the acinic cell carcinoma ($n = 4$) followed by the mucoepidermoid carcinoma ($n = 3$) and adenoid cystic carcinoma ($n = 2$). The overall female-to-male ratio was 1.18, with a ratio of 1.08 and 2.0 in the benign and malignant groups, respectively. At the end of follow-up (August 1st, 2014) two patients had died, one with adenoid cystic carcinoma and one with mucoepidermoid carcinoma. Both patients had perineural invasion and involved resection margins at presentation. The incidence was 0.12 and 0.53 per 100,000 children of the malignant and benign tumors, respectively.
Conclusion: Pleomorphic adenomas were the predominant neoplasm in the parotid gland in children. The most frequent of the malignant tumors was the acinic cell carcinoma, which is in contrast to previous studies. The proportion of malignant-to-benign parotid gland tumors is in contrast to earlier study reports not higher in children than in adults.

1. Introduction

Diagnosis and treatment of parotid gland tumors is a challenge for clinicians and pathologists due to the rarity of these tumors [1]. Salivary gland tumors (SGT) are especially rare in children and very little data has been reported in the literature regarding population-based demographic data. Most studies are single-center experiences [1–8] and are therefore selected materials. We want to gain a better understanding of the true incidence of these tumors as well as the histopathologic distribution in a nationwide population.

The parotid gland is the most commonly involved site for the salivary gland tumors [3,6,7,9–15] and our study focuses on tumors in this particular gland in children. Earlier studies have reported a larger fraction of malignant salivary gland tumors in children than in adults calling for alertness of parotid masses in children [10,13,15,16].

Denmark has unique nationwide registries allowing complete epidemiological studies without selection or bias. These registries allow access to raw data from all hospitals in the country and all specimens for pathology examination. This enables us to find the overall relationship between benign and malignant SGT in our population without exceptions.

We have therefore conducted a nationwide study of benign and malignant childhood tumors in the parotid gland in Denmark in a 16-year-period from January 1st, 1990 to December 31st, 2005.

2. Methods

The Ethics Committee of the Capital Region of Denmark approved this protocol (H-1-2012-061 add#3) and all specimens

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were processed according to the World Medical Association Declaration of Helsinki (version 2008).

The search for peer-reviewed literature was conducted via PubMed using the MeSH term "Parotid neoplasms" and the subheadings "epidemiology", "statistical and numerical data" and "classification". We defined our population as being below 18 years of age.

Data regarding malignant parotid gland tumors in the Danish population was collected from three nationwide registries: The Danish Cancer Registry, The Danish National Pathology Registry and The Danish Patient Registry, and included children diagnosed with a salivary gland carcinoma in the period from January 1st, 1990 to December 31st, 2005. Patient records were retrieved from the Danish head and neck oncology centers and local departments of ENT Head and Neck Surgery. This yielded nine patients for inclusion in our study. A detailed description of how the data was collected can be found in "Salivary gland carcinoma in Denmark 1990–2005: a national study of incidence, site and histology. Results of the Danish Head and Neck Cancer Group (DAHANCA)" by Bjørndal et al. [17].

Data regarding all benign parotid gland tumors in the 2005 WHO classification was extracted from The Danish National Pathology Registry concerning children diagnosed from January 1st, 1990 to December 31st, 2005. 51 pleomorific adenomas and one adenolymphoma (Warthin's tumor) were identified and included in this retrospective study.

As benign tumors are not registered in The Danish Cancer Registry or Patient Registry, only The Danish National Pathology Registry was available for data extraction. Nonetheless, it is an almost complete database of all diagnoses and their topography, making us confident of the completeness of our data [18].

The median follow-up period was 174 (range: 5–273) and 213.5 (range: 106–294) months for the malignant and benign tumors, respectively. Follow-up ended on the 1st of August 2014 or at the time of death.

Fishers exact test was used to test for differences in frequencies with a significance level of $p < 0.05$.

Incidences were calculated according to data from Statistics Denmark regarding the population divided into age groups; 9–17 years of age in the benign group, and 11–17 years in the malignant group. The mean population of the 9–17 year-olds between 1990 and 2005 was calculated, and the same for the 11–17 year olds. The incidence was then calculated as number of tumors (52 benign and 9 malignant) multiplied by 100,000 divided by the mean population in the respective age groups multiplied by 16 years.

The risk of recurrence in relation to pleomorphic adenomas was calculated using the summarized time of follow-up: (4 patients/10,784 months) × 12 months × 100%.

3. Results

Of 61 children, 52 (85%) were diagnosed with benign tumors and nine (15%) with malignant tumors (a ratio of 5.67). The mean age was 15.2 (range: 9–17) years. The overall female-to-male ratio was 1.18 (33 female vs. 28 male).

In the benign group the mean age was 15.3 (range: 9–17) years; of these patients 27 were female and 25 male (a ratio of 1.08). The incidence of benign tumors was calculated to 0.53 per 100,000 children from the ages 9–17 years.

51 of the patients were diagnosed with pleomorphic adenomas and one with Warthin's tumors. Of the 51 patients with pleomorphic adenomas four (7.8%) experienced recurrences and the risk of recurrence is 0.45% per year of follow-up after surgery for these tumors.

Exact age, sex and histological types of the malignant tumors are shown in Table 1. In the malignant group the mean age was 14.3 (range: 11–17) years; of these patients six were female and three male (a ratio of 2.0). The following tumors were found: acinic cell carcinoma ($n = 4$; 44%), mucoepidermoid carcinoma ($n = 3$; 33%) and adenoid cystic carcinoma ($n = 2$; 22%). The incidence of malignant parotid tumors was calculated to 0.12 per 100,000 children from the ages 11–17 years.

Of the nine patients with malignant tumors, one (11%) was lost to follow-up and three (33%) had recurrences. All three recurrences were M-site (patient nr. 1, 2 and 8, Table 1), two of these were also N-site (patient nr. 2 and 8) and only one was a T-site recurrence (patient nr. 2). The two patients experiencing both M-site and N-site recurrences later died of the disease. The two tumors causing death were adenoid cystic carcinoma (diagnosed in a year 16 old male; patient nr. 2) and mucoepidermoid carcinoma (diagnosed in a 14 year old male; patient nr. 8). In both cases perineural invasion was present and the resection margins were involved upon surgical removal of the tumors. In addition, the mucoepidermoid carcinoma (patient nr. 8) presented with invasion of the mandibula (T4a), lymph node metastases (N2b) and metastases to the lungs and bones (M1).

Table 1
Malignant tumors.

Patient nr.	Gender	Age (years)	Histotype	Tumor type/grade	TNM classification	Resection margins	Perineural invasion	Vascular invasion	Recurrence	Death by tumor
1	Female	11.7	Adenoid cystic carcinoma	Tubular–cribriform	T1N0M0	Close	Yes	No	Yes	No
2	Male	16.2	Adenoid cystic carcinoma	Tubular–cribriform	TxN0M0	Involved	Yes	No	Yes	Yes
3	Female	17.7	Acinic cell carcinoma	–	T1N0M0	Free	No	No	No	No
4	Female	15.2	Acinic cell carcinoma	–	T1N0M0	Free	Not described	Not described	No	No
5	Female	15.2	Acinic cell carcinoma	–	Not described	Not described	Not described	Not described	No	No
6	Female	12.1	Acinic cell carcinoma	–	Not described	Involved	No	Not described	No	No
7	Female	14.3	Mucoepidermoid carcinoma	High	T3N0M0	Involved	Yes	Yes	No	No
8	Male	14.4	Mucoepidermoid carcinoma	Low	T4aN2bM1	Involved	Yes	Yes	Yes	Yes
9	Male	11.7	Mucoepidermoid carcinoma	Specimens from this patient could not be recovered for revision histology						