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Parotid Neoplasms in Pediatric Population: A Single Institute Experience

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ABSTRACT

Background: salivary gland neoplasms are rare in pediatric age group with pediatric parotid neoplasms account for most of them, mucoepidermoid carcinoma is the most common malignant variant while pleomorphic adenoma is considered the most common benign variant among them.
Methods: this is a retrospective cohort study that included 41 pediatric patients with parotid lesions the aim is to present our institutional experience with the evaluation of pathological features and management of pediatric parotid gland tumors.
Results: the median age was 12 years old. (26.8%) were female and (73.2%) were male, radiological investigations were CT & MRI in (17.1%), ultrasound alone in (12.2%), ultrasound & CT in (68.3%), ultrasound & MRI (2.4%); pathological investigations were FNAC alone in (80.5%), FNAC and true cut biopsy in (19.5%) cases. pathological results; adenoid cystic carcinoma (7.3%), cavernous hemangioma in (2.4%), Hodgkin lymphoma in (12.2%), mucoepidermoid carcinoma in (29.3%), myoepithelioma in (2.4%), pleomorphic adenoma in (34.1%), rhabdomyosarcoma (anaplastic variant) in (2.4%) and rhabdomyosarcoma (embryonal variant) in (7.3%) cases.
Conclusion: 58.5% of pediatric parotid neoplasms were malignant with Mucoepidermoid carcinoma is the commonest and 41.5% were benign; were the most frequent was the pleomorphic adenoma.
Keywords: mucoepidermoid – parotid – pediatric – pleomorphic

Introduction

Salivary gland neoplasms are uncommon in children representing about 3% of all head and neck tumors (1&2), and constitutes less than 10% of pediatric head and neck tumors (3-6). In the general population, about 5% of salivary gland neoplasms are encountered in pediatric population (1-4). Pediatric salivary gland neoplasms are more commonly seen in female children ranging in age from more than 10 years old and less than 18 years old (2&4). Pediatric parotid gland neoplasms compose 90% of pediatric salivary gland neoplasms with malignancy rate varying in-between 23% up to 40% (7&8). The most frequently diagnosed malignant pediatric parotid gland neoplasm is mucoepidermoid carcinoma, followed by Adenoid cystic carcinoma, Acinic cell carcinoma and rhabdomyosarcoma (1&9). Surgery is the gold standard procedure for treatment of pediatric parotid neoplasms. The facial nerve to be spared unless it is invaded (7). Superficial parotidectomy with preservation of the facial nerve is the basic surgical procedure required for diagnosis and treatment of pediatric parotid neoplasms (10). Adjuvant radiotherapy is considered beneficial in certain malignant cases such as positive surgical margins, persistent lymph node metastases, perineural invasion or high-grade tumors despite the risk of post-irradiation complications including facial deformities, secondary malignancies, trismus and hyposialia. There is no role for radiotherapy in benign parotid neoplasms (11-13). Neoadjuvant chemotherapy is indicated for malignant subtypes of parotid gland tumors which are rapidly progressing, recurrent, metastatic and non-resectable lesions (14). The majority of literature reviews about pediatric parotid neoplasms are based on retrospective studies encompassing small

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numbers of cases and case reports. The goal of this study is to report our institution's experience with evaluation and management of pediatric parotid neoplasm.

Methodology

Medical records of 41 pediatric patients with parotid lesions were collected and reviewed from the period of January 2008 to January 2018; patients with parotid lesions in pediatric age group are included in this study while patients excluded from this study are those who are more than 18 years old. The primary end point for this study is to report our experience in National cancer institute, with the aim to estimate the incidence, pathologic type, clinical presentation, imaging characteristics, pathological features and treatment outcomes of pediatric parotid tumors.

Ethical issues

Consent forms have been signed by all patients and their parents before enrolment into this study along with the approval of National cancer Institute ethical committee. Subject and consent on the medical photos was obtained from patients and their parents.

Results

The studied patients median age was 12 years old ranging from 2 to17 years old with mean age (11.685) and standard deviation (3.4888), 11 cases (26.8%) were females and 30 cases (73.2%) were males. all the included cases had negative family history and past medical history for malignancy , radiological investigations and pathological evaluation were done for all patients; radiological investigations were CT & MRI in 7 cases (17.1%), ultrasound alone in 5 cases (12.2%), ultrasound & CT in 28 cases (68.3%), ultrasound & MRI in one case (2.4%); pathological diagnosis was determined by FNAC alone in 33 cases (80.5%), FNAC and true cut biopsy in 8 cases (19.5%), 22 cases (53.7%) had left side lesions and 19 studied cases (46.3%) had their lesions in the right side, pathological results: adenoid cystic carcinoma 3 cases (7.3%), cavernous hemangioma in one case (2.4%), Hodgkin lymphoma in 5 cases (12.2%), mucoepidermoid carcinoma in 12 cases (29.3%), myoepithelioma in one case (2.4%), pleomorphic adenoma in 14 cases (34.1%), rhabdomyosarcoma (anaplastic variant) in one case (2.4%) and rhabdomyosarcoma (embryonal variant) in 3 cases (7.3%) Table (1). 14 cases (34.1%) were diagnosed as pleomorphic adenoma, clinically 3 cases were T1N0M0 and 11 cases were T2N0M0, all cases underwent superficial parotidectomy only, all cases are free of tumor during follow up period ranging from 2 to 10 years with mean value 3.7 and median value 3. 12 cases; (29.3%) were diagnosed as mucoepidermoid carcinoma, 10 cases were grade 1 and two cases were grade 2, clinically 9 cases were T2N0M0, one case was T1N0M0 and two cases were T2N1M0. 11 cases had total parotidectomy with facial nerve preservation and ipsilateral supra-omohyoid neck dissection Figure (1). Only one case had total parotidectomy with facial nerve preservation without neck dissection, among the 11 cases that underwent neck dissection all of them had negative lymph nodes (pathologically proven). Regarding adjuvant treatment 4 cases received adjuvant RTH due to positive margins of resection. In-between mucoepidermoid carcinoma cases during the follow up period ranging from 2 to 10 years with (mean value 3.7) and (median value 3), 3 cases had recurrence; 2 of them had nodal recurrence and they received RTH after the initial surgery and one had local recurrence, all of them underwent surgical treatment and the one who had the local recurrence received adjuvant RTH with good response

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and they are all tumor free till now Figure (2). 4 cases (9.7%) were diagnosed as rhabdomyosarcoma, one case was anaplastic variant and 3 cases were embryonal variant. Anaplastic variant which is a high grade variant , clinically was staged as T2N0M0, received neoadjuvant CTH followed by total parotidectomy with facial nerve preservation and ipsilateral supra-omohyoid neck dissection, some of the lymph nodes were positive (pathologically proven) then received adjuvant RTH, during follow up the patient developed metastatic recurrence to the lung within 2 years ; then received CTH with poor response and mortality occurred within one year of metastatic recurrence. 3 cases (7.3%) were diagnosed as embryonal variant rhabdomyosarcoma, one case was high grade and two cases were low grade, clinically 2 cases were T1N0M0 and one case was T3N0M0, all cases received neoadjuvant CTH followed by total parotidectomy with facial nerve preservation and ipsilateral supra-omohyoid neck dissection followed by adjuvant RTH, all cases had negative lymph nodes, all cases are free during the follow up period ranging from 2 to 10 years. 3 cases (7.3%) were diagnosed as adenoid cystic carcinoma, 2 cases were high grade and one case was low grade, clinically 2 cases were T2N0M0 and one case was T1N0M0, 2 cases underwent total parotidectomy with facial nerve preservation and ipsilateral supra-omohyoid neck dissection and one case underwent superficial parotidectomy without neck dissection, among the 2 cases who underwent neck dissection both of them had positive lymph nodes (pathologically proven) and received adjuvant RTH after the initial surgery. Among adenoid cystic carcinoma cases during follow up 2 cases had metastatic recurrence in the lung within 2 years of the initial surgery and received CTH with poor response and mortality within 2 years of the recurrence, one case had local recurrence within 3 years of the initial surgery and underwent completion parotidectomy with ipsilateral supra-omohyoid neck dissection and the LNs were negative and didn't receive adjuvant RTH with good response and is free of tumor till now. 5 cases (12.2%) were diagnosed as lymphoma; all cases received CTH as initial treatment after surgical diagnostic Procedure in the form of superficial parotidectomy as there was no peripheral lymphadenopathy.

Table (1); variants of the Pathological parotid lesions

Pathological types.	**N = 41 (%)**
Adenoid cystic carcinoma	3 (7.3%)
Cavernous hemangioma	1 (2.4%)
Epidermoid cyst	1 (2.4%)
Hodgkin lymphoma	5 (12.2%)
Mucoepidermoid carcinoma	12 (29.3%)
Myoepithelioma	1 (2.4%)
Pleomorphic adenoma	14 (34.1%)
Rhabdomyosarcoma (anaplastic variant)	1 (2.4%)
Rhabdomyosarcoma (embryonal variant)	3 (7.3%)

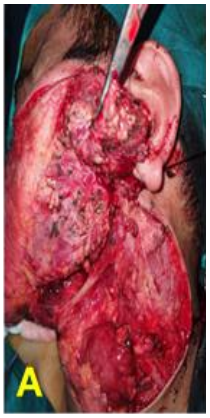

Figure (1): male child with left parotid mucoepidermoid carcinoma underwent left total parotidectomy and left supraomohyoid neck dissection (A): dissection of the lesion from left facial nerve, (B): left supraomohyoid neck dissection (C): postoperative image post left total

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parotidectomy with facial nerve preservation and left supraomohyoid neck dissection (D): specimens of the left total parotidectomy and left supraomohyoid neck dissection

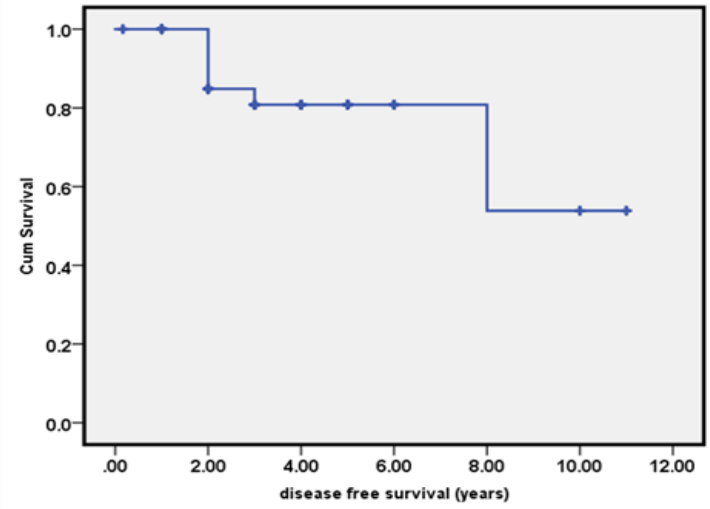

Figure (2) graph of cumulative survival and disease-free survival over years

Discussions

Pediatric parotid neoplasms are rare constituting less than 4% of all salivary gland tumors (15). Till now, very small numbers of studies have been conducted on pediatric parotid neoplasms and were less than helpful in evaluation of behaviour and prognosis of these tumors. In most of the literature reviews near about 80% of pediatric parotid neoplasms are benign and the most common type was the pleomorphic adenoma accounting for 60% of all parotid neoplasms (8). Liu et al, reported that benign pediatric parotid neoplasms represent about 65% of all parotid tumors with hemangioma being the most common followed by pleomorphic adenoma and about 35% were malignant tumors with mucoepidermoid carcinoma as commonest malignant variant (16). Byers et al, reported that mucoepidermoid type lesions are most common among malignant lesions, followed by acinar cell carcinomas (17). Xu et al reported that the malignancy rate of pediatric parotid neoplasms was 67% and they occurred mainly in second decade of life (18). In our study about 58.5% of pediatric parotid neoplasms are malignant, the most common malignant tumor was the mucoepidermoid carcinoma accounting for 29.3% of all parotid neoplasms while the benign pediatric parotid neoplasms are about 41.5% and the most frequent benign tumor was the pleomorphic adenoma representing about 34.1% of all parotid neoplasms. We reported that most of these tumors occurred within the 2nd decade of life. Valstar et al showed that recurrence rate for pediatric parotid pleomorphic adenoma was 6.7% with reported first recurrence at a median of 7 years; the study reported risk factors for the recurrence which include unclear resection margins (19). All cases diagnosed with parotid pleomorphic adenoma in our study were free of tumor during period of follow up ranging from 2 to 10 years with mean value 3.7 and median value 3 and also all the cases that underwent superficial parotidectomy with clear resection margins. Xu et al, reported that majority (about 84%) of mucoepidermoid carcinoma were intermediate grade, while that of low grade and high grade were of low frequency (18). Among our series we reported that majority (about 83.3%) of mucoepidermoid carcinoma were

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low grade and about 16.7% were intermediate grade and we didn't report high grade cases. Many studies reported that 5 years overall survival rate for pediatric parotid mucoepidermoid carcinoma was 95%: 100% (20-22). Among our series 5 years overall survival rate for pediatric parotid mucoepidermoid carcinoma was 100%- and 5-years disease free survival rate was 75%. Ryan et al, reported that mucoepidermoid carcinoma tend to have lymph node metastases and presenting with advanced stage (20). In our present study, all cases with mucoepidermoid carcinoma had undergone neck dissection with negative lymph node metastases and were admitted in early stages. Surgical intervention is the standard primary line of treatment for pediatric parotid neoplasms. Among cases of mucoepidermoid carcinoma in our series surgery was the primary line of treatment, all cases had total parotidectomy with spared facial nerve and 91.5% of them underwent additional neck dissection. Adjuvant radiotherapy still remain debatable question regarding patients with mucoepidermoid carcinoma with no consensus about certain indications of it and with reported side effects from it including radiation-induced malignancies and effect on bone growth (23). In our series 4 cases among mucoepidermoid carcinoma cases received adjuvant RTH due to positive margins of resection. Although adjuvant radiotherapy still debatable, analysis of some literatures showed that acinic cell carcinoma and adenoid cystic carcinoma are treated usually with adjuvant radiotherapy after surgery as primary line of treatment (17& 20). In our study; 2 cases of adenoid cystic carcinoma cases received adjuvant radiotherapy after surgery as they were both high grade and the patients had positive lymph node deposits.

Conclusion

In this study, we reported the clinic-pathological and prognostic characteristics of pediatric parotid neoplasms among 41 patients, 58.5% of pediatric parotid neoplasms are malignant, the most common malignant tumor was the mucoepidermoid carcinoma representing about 29.3% of all parotid neoplasms while the benign pediatric parotid neoplasms represent about 41.5% and the most common benign tumor was the pleomorphic adenoma representing about 34.1% of all parotid neoplasms. Pediatric parotid mucoepidermoid carcinoma appear to have better clinical outcome with 5 years overall survival rate was 100%- and 5-years disease free survival rate was 75%.

Conflict of interest statement

Authors declare there is no conflict of interest in this article.

Funding

This study has not received any external funding.

Informed consent

Written informed consent was obtained from all individual participants included in the study, Additional informed consent was obtained from all individual participants for whom identifying information is included in this manuscript.

Ethical approval for study protocol /study design /Methodology

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The study was approved by the institutional review board of Medical Ethics Committee of Cairo University (ethical approval code: 201819027.4).

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