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ORIGINAL RESEARCH

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Pediatric parotidectomy outcomes: A 14-year multicenter study

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Abstract

Objectives: Parotidectomy increases childhood challenges. This study aimed to determine the clinical profiles, investigations, and outcomes of pediatric patients who had undergone parotidectomy.

Methods: A multicenter retrospective review of parotidectomy in pediatric patients between 2007 and 2020.

Results: In 108 parotidectomies, the final diagnoses were benign (47.22%), malignant (36.11%), and non-neoplastic (16.67%). The incidence of facial palsy was 37.03%, which was significantly lower in the superficial group than that in the total parotidectomy group ($p = .021$). The incidence of facial nerve palsy was significantly higher in the malignancy group than that in the benign group ($p = .035$). Magnetic resonance imaging (MRI) detected malignancy with 92.8% overall accuracy, 83.3% sensitivity, and 100% specificity. The sensitivity and specificity of fine-needle aspiration (FNA) were 54.2% and 92.7%, respectively.

Conclusions: Parotidectomy is commonly performed for benign and non-neoplastic diseases in pediatric patients. Facial nerve palsy is significantly associated with malignant tumors and total parotidectomy. MRI is the most accurate imaging modality for diagnosing malignant lesions. FNA exhibits moderate agreement with the final pathology.

Level of Evidence: Level IV.

KEYWORDS
facial palsy, parotid cytology, parotid tumor, parotidectomy, pediatric

1 | INTRODUCTION

Parotid gland surgery is a common procedure in head and neck surgeries. The indications for parotidectomy include neoplasms, chronic inflammation, sialolithiasis, infection, and abscess formation. Parotidectomy is more challenging in pediatric patients than in adult. Juvenile parotid gland disease is far less common than that in adults; therefore, reaching a high-volume pediatric parotidectomy is unachievable. The etiology of parotid gland disease in childhood is predominantly congenital and inflammatory, whereas neoplasms are more frequent in adults. Parotid gland resection varies in complexity depending on the spectrum of its associated pathologies.

Parotid gland resection with facial nerve dissection remains the standard procedure for parotidectomy. Iatrogenic facial nerve injury is a devastating complication resulting in long-lasting cosmetic and functional morbidities. According to previous studies, 14–64% and 0–9% patients report permanent facial nerve weakness following parotidectomy.[1–5] With respect to the pediatric population, it is essential to carefully attend to the facial nerve because of the minute nerve caliber and the surrounding anatomy. Additionally, the continued maturation of the mastoid process contributes to a more superficial location of the facial nerve.[6]

The published literature on parotidectomy in childhood includes a small number of case series. Data on clinical investigations and surgery-related complications are lacking. Therefore, the Thai Society for Head Neck Oncology research network conducted a multicenter study to retrieve data from five tertiary care centers to determine the clinical profiles, investigations, and surgical outcomes of pediatric patients who underwent parotidectomy.

2 | MATERIALS AND METHODS

This multicenter study was conducted in Thailand. The study centers included tertiary/university hospitals across the country: (1) Faculty of Medicine, Chulalongkorn University; (2) Faculty of Medicine, Siriraj Hospital, Mahidol University; (3) Faculty of Medicine, Chiang Mai University; (4) Faculty of Medicine, Prince of Songkla University; and (5) Faculty of Medicine, Khon Kaen University. This was a retrospective chart/electronic medical record review of patients aged ≤18 years who had undergone partial superficial, superficial, or total parotidectomy between 2007 and 2020.

Information on patient characteristics, preoperative imaging, fine-needle aspiration (FNA) results, type and extent of surgery, final pathological report, survival, and postoperative complications were collected. Owing to the retrospective nature of this study, FNA, final pathology, and imaging results were based on a previous official report and were not re-evaluated. Pathological diagnoses were categorized into three groups: (1) non-neoplasm, (2) benign, and (3) malignant for subgroup analysis. Patients who underwent a biopsy and those with incomplete medical records were also excluded.

The sample size calculation used the incidence of pediatric parotid malignancy and postoperative facial nerve palsy of 16% and 21%, respectively, from studies by Orvidas et al.[7] and Owusu et al.,[8] respectively. The confidence level was set at 95%, $z = 1.96$, and the precision (e) was set at 10%. The calculations revealed a minimum sample size of 64 patients. FNA results were categorized and converted into Milan system reporting.[9] Milan II and IVa were considered negative tests, and Milan III, V, and VI were considered positive tests to calculate sensitivity, specificity, and accuracy, as the final histopathology is the gold standard. Pearson's chi-squared test was used to assess the incidence of postoperative facial palsy between the diagnosis and surgical extent. Statistical significance was set at $p < .05$. Statistical analyses were performed using IBM SPSS Statistics version 22. software (SPSS Inc., IBM).

This study was approved by the Institutional Review Board of (1) Faculty of Medicine, Chulalongkorn University (COA No. 1535/2021); (2) Faculty of Medicine Siriraj Hospital, Mahidol University (COA Si948/2021); (3) Faculty of Medicine, Chiang Mai University (Research Ethics Committee No.467/2021); (4) Faculty of Medicine, Prince of Songkla University (REC.64–420–13-3); and (5) Faculty of Medicine, Khon Kaen University (Ethics Committee for Human Research IRB00001189). The need for informed consent was waived by the Institutional Review Board.

3 | RESULTS

A hundred and eight parotidectomies were performed in 61 male (56.48%) and 47 female (43.51%). The median age of the patients was 16 years in male and 15 years in female, ranging from 1 to 18 years. The Pearson correlation of the non-neoplasm to the malignant group and the benign to the malignant group were 2.76 ($p = .096$) and 4.45 ($p = .035$), respectively. Pearson correlation values of the partial superficial and superficial parotidectomy to total parotidectomy group were 1.16 ($p = .281$) and 5.31 ($p = .021$). The final diagnoses were mainly benign in 51 (47.22%), malignant in 39 (36.11%), and non-neoplastic in 18 (16.67%) cases. The third most common diagnosis in the non-neoplasm group was Kimura disease, followed by branchial cyst anomalies and chronic sialadenitis. Most benign neoplasms were pleomorphic adenomas, accounting for up to 80% of all benign cases, and schwannomas and hemangiomas ranked second by far, at 5.8% each. Nearly half of the cases in the malignant group were of mucoepidermoid carcinomas (51.28%) followed by acinic cell carcinomas (23.07%). The most common procedure was superficial parotidectomy (59.26%), followed by total parotidectomy (33.33%), and partial superficial parotidectomy (7.40%). Neck dissection was performed in five cases. The overall incidence rate of facial palsy was 37.03%. The incidence was significantly different between superficial and total parotidectomies ($p = .021$). According to histopathological diagnosis, parotidectomy in the malignancy group had a 51.28% chance of palsy. Benign and non-neoplastic tumors showed a comparable incidence of postoperative palsy of 29.41% and 27.78%, respectively. The incidence of benign and malignant palsies was significantly different ($p = .035$). The results are presented in Table 1.

Malignant tumors were found in half (50%) of all postoperative facial palsy cases. No facial nerves were deliberately sacrificed for the clearance of malignancy. Malignant patients with palsy also had the highest probability of becoming permanent (15.15%). Palsy was frequently found in the lower facial nerve division (72.5% of the marginal mandibular branch and 35% of the buccal branches) compared to the upper nerve division (15% of the zygomatic branch and 22.5% of the temporal/frontal branch). One case of low-grade mucoepidermoid tumor presented with facial palsy before surgery. Three patients underwent revision parotidectomy (two with acinic cell carcinoma and one with Kimura disease) without any postoperative facial palsy. The results are presented in Table 2.

One case of postoperative sialocele was reported, which was managed conservatively. No surgery-related infections or hematomas were observed. There was only one case of mortality due to adenoid cystic carcinoma. The patient presented with a parotid mass that persisted for 7 months. FNA suggested pleomorphic adenoma; however, total parotidectomy revealed adenoid cystic carcinoma. Radiotherapy was administered at a dose of 50 Gy. Two years later, chest radiography revealed cannonball metastases in both lungs without local recurrence in the surgical bed. The disease did not respond to chemotherapy, and the patient died within 1 year.

TABLE 1 Demographic data and factors affecting post-operative facial nerve palsy

	N (%)
Gender[a]	
Male	61 (56.5)
Female	47 (43.5)
Factors affecting post-operative facial nerve palsy	
Pathological diagnosis	
Non-neoplasm	18 (16.7)
Kimura	6
Other (Branchial anomaly, etc.)	12
Benign	51 (47.2)
Pleomorphic adenoma	41
Other (Schwannoma, etc.)	10
Malignant	39 (36.1)
Mucoepidermoid CA	20
Acinic cell CA	9
Other (Adenoid cystic CA, etc.)	10
Extent of surgery	
Partial superficial parotidectomy (PSP)	8 (7.4)
Superficial parotidectomy (SP)	64 (59.3)
Total parotidectomy (TP)	36 (33.3)

[a]The value of the K–S test statistic (*D*) is 0.17874. The *p*-value is .00142. This provides good evidence that your data is not normally distributed.

Computed tomography (CT) was performed in 32 (28%) children, ultrasonography in 17 (15%), and magnetic resonance imaging (MRI) in 14 (12.3%). The criteria for diagnosis of malignancy by CT according to the Parotid Imaging and Data System (PI-RADS)[10] were PI-RAD 4 (probable malignancy) and PI-RAD 5 (highly suggestive of malignancy). The sensitivity of CT for detecting malignant lesions using the PI-RADS criteria was 87.5%, with 81.2% specificity, 82.3% PPV, 86.6% NPV, and 84.3% overall accuracy. The sensitivity of MRI for detecting malignant lesions was 83.3%, with 100% specificity, 100% positive predictive value, 88.8% negative predictive value, and 92.8% overall accuracy. Despite the lack of specificity of ultrasound owing to the subjectivity of the examiner, there was a sensitivity of up to 100% for the confirmation of intraparotid lesions. Nevertheless, further CT and/or MRI imaging was required in 12 patients.

Of the 113 patients in our study, 73 underwent FNA of the parotid gland. The FNA results were classified using the Milan system (Table 3). The correlation between the FNA results and definite pathology is shown in Table 4. Eleven patients had false-negative malignancies on FNA, including five with low-grade mucoepidermoid carcinomas, two with secretory carcinomas, and one each with adenoid cystic carcinoma, lymphoma, squamous cell carcinoma, and leiomyosarcoma. The sensitivity, specificity, positive predictive value, and negative predictive value were 54.2% (95% CI: 32.8%–74.4%), 92.7%

TABLE 2 Detail of facial nerve palsy

	Palsy (%)	Permanent (%)[a]	M (%)	B (%)	Z (%)	T/F (%)
Non neoplasm	5 (12.50%)	3 (9.09%)	3 (7.5%)	3 (7.5%)	2 (5%)	3 (7.5%)
Benign	15 (37.50%)	2 (6.06%)	14 (35%)	2 (5%)	2 (5%)	2 (5%)
Malignant	20 (50%)	5 (15.15%)	12 (30%)	9 (22.5%)	2 (5%)	4 (10%)
Total			29 (72.5%)	14 (35%)	6 (15%)	9 (22.5%)

Abbreviations: B, buccal branch; M, marginal branch; T/F, temporal/frontal branch; z, zygomatic branch.
[a]Available data $n = 33$.

TABLE 3 Distribution of FNA result according to the Milan system and the risk of malignancy correlation with final histopathological results

Diagnostic category	Non-malignancy	Malignancy	Total *N* (%)	ROM (%)
I. Non-diagnosis	3	5	8	62.5
II. Non-neoplastic	8	5	13	38.5
III. AUS	0	3	3	100
IVa. Benign	30	6	36	16.7
IVb. SUMP	0	0	0	–
V. Susp. for malignancy	3	5	8	62.5
VI. Malignant	0	5	5	100
Total	44	29	73	

Abbreviations: AUS, atypia of undetermined significance; ROM, risk of malignancy; SUMP, salivary gland neoplasm of uncertain malignancy.

TABLE 4 Correlation between the FNA result and final pathology

Diagnostic FNA	Patho: non-malignancy; *N* (%)	Patho: malignancy *N* (%)	Total
Non-malignancy	38	11	49
Malignancy	3	13	16
Total	41	21	65

Note: FNA Milan: II, IVa: non-neoplastic+ benign: non-malignancy; FNA Milan III, V, VI: Malignancy; Exclude Milan I.

(95% CI: 80.1%–98.5%), 81.3% (95% CI: 54.4%–96.0%), and 77.6% (95% CI: 63.4%–88.2%), respectively. The ROC was 0.73, and the measurement agreement by kappa statistic was 0.503 (moderate agreement).

4 | DISCUSSION

This cohort had one of the largest sample sizes ($n = 108$) and the longest follow-up period (14 years) ever reported for pediatric parotidectomy. Similar to the study by Carter et al., we observed a greater prevalence of parotid masses in boys than in girls (M:F = 1.3:1).[11] The median age at diagnosis was 15 years for women and 16 years for men, which was higher than that in the previous study.[7,8,11,12]

This study found a high rate of postoperative facial nerve palsy, potentially because all conducting sites in tertiary referral centers had a higher malignancy rate than those reported in previous reports.[7,8,11,12] The marginal mandibular nerve is the most commonly affected branch because its anatomical course can vary from running directly along the angle of the mandible to a few centimeters below. This difference increases the likelihood of nerve damage. Furthermore, unlike the midfacial nerve branches, the marginal mandibular nerve lacks numerous anastomoses, causing obvious injury to this branch.[8] The growth of the mastoid and parotid area should reach adult size by the age of 12 years. However, we could not find a statistical difference in facial nerve palsy rate among the subgroups of patients aged 12 years and below versus those aged more than 12 years; T value = 1.02 and p-value = .15. We demonstrated that the extension of surgery had a greater effect on facial nerve outcomes than the effects of pediatric anatomical factors.

Apart from the possibility of facial palsy, other complications are uncommon. Only one sialocele was identified. With only one death, the survival rate could not be calculated. This finding confirmed the finding of Morse et al.[13] indicating a favorable prognosis in children with parotid carcinoma, with an overall 5-year survival rate of 93%. Li et al.[14] demonstrated that 33 pediatric patients with mucoepidermoid carcinoma had excellent prognoses with no evidence of recurrence or death. Janz et al.[15] studied 169 cases of pediatric mucoepidermoid carcinoma and discovered that the 5-year disease-specific survival rate was greater than 90%. All available data, including those from this study, indicate a favorable prognosis for pediatric parotid malignancies when treated appropriately. Any treatment plan with the potential for long-term side effects should be carefully selected.

Imaging modalities are generally required to differentiate between benign and malignant parotid gland tumors and their preoperative anatomical extents. Nevertheless, <50% of the patients received preoperative imaging regarding of availability of imaging and sedation required in pediatric patients. In our study, MRI provided insight into the benign and malignant characteristics of the parotid gland with high specificity and accuracy of 100% and 92.8%, respectively. MRI has an advantage with respect to its non-invasiveness, being free of ionizing radiation, and excellent soft-tissue resolution. CT of parotid gland neoplasms may have similar imaging features and currently lacks definition features. Applying PI-RAD (overall Cohen's Kappa test showed medium consistency; $K = 0.614$, $p < .001$, 95% CI: 0.569–0.695)[10] to evaluate the risk of malignancy may be valuable even though this has not really been validated in children yet, with sensitivity, specificity, PPV, and NPV of 87.5%, 81.2%, 82.3%, and 86.6%, respectively. However, both CT and MRI not only establish diagnoses but also help in cases with clinically questionable parotid gland lesions. Ultrasound remains the most practical imaging modality for parotid lesions in children owing to the lack of ionizing radiation and nephrotoxic contrast agents. It is commonly used in outpatient clinics and for bedside evaluations without sedation. Generally, the specificity of ultrasound is good for the effective calculation of the parotid gland and diagnostic performance improvement.[16,17] However, few patients in our study had well-documented preoperative ultrasound findings. Thus, standardized nomenclature and definition of the features of parotid lesions are recommended.

FNA is a modality used for the diagnostic evaluation of parotid tumors and is widely used because of its minimal complications. In our study, preoperative FNA was performed in 73 (64%) of 113 patients. The sensitivity, specificity, PPV, and NPV for differentiating benign and malignant lesions were 54.2, 92.7, 81.3, and 77.6, respectively. Rochi et al.[18] reported the accuracy of FNA in 34 cases of pediatric salivary gland lesions, with a sensitivity and specificity of 92% and 86%, respectively. Another larger study by Wang et al.[19] with 104 pediatric salivary glands FNA and 54 histopathologic follow-ups also reported a sensitivity and specificity of 80% and 97%, respectively, which are comparable to those of adult patients. The much lower sensitivity in this study was owing to the high false-negative results of FNA in pathological diagnostic malignant lesions, mainly low-grade mucoepidermoid carcinoma, because of the heterogeneity of tumor types with overlapping cell types in salivary gland neoplasms. The diagnosis of low-grade mucoepidermoid carcinoma is challenging because it is composed of epithelial and cystic components that make it difficult to obtain cells for cytologic smears,[20,21] leading to underdiagnosis. Garrett et al.[22] and AlGhamdi et al.[23] also reported low overall sensitivities of 39% and 46.67%, respectively, in FNA for low-grade mucoepidermoid carcinoma. The other false-negative histopathologic subtype may be due to the rarity of the disease; for example, primary squamous cell carcinoma arising in the parotid gland is quite rare in children and is a diagnosis of exclusion. Mammary analog secretory carcinomas are also rare in our region and are more common in adults.

To our knowledge, our study is one of the largest series of pediatric parotid FNA with histopathologically confirmed diagnoses. Despite the high specificity, with a moderate agreement between FNA and histopathological results, the sensitivity in our study was lower than that reported in previous studies. We suggest that FNA should be applied in pediatric parotid lesions suspected of salivary gland tumors; however, clinicians should always correlate the clinical presentation and imaging findings, especially MRI, with the FNA results, to avoid missing malignant neoplasms.

4.1 | Limitations

This study had a retrospective design and was multicenter. It had some limitations.

1. The surgery involved multiple surgeons, possibly with different skills, which might have affected the rate of facial nerve palsy.
2. Many FNA results were reported before the Milan system was implemented; therefore, some important information for the Milan classification may not have been described in the FNA reports. There was also some variation in patient demographics, referral patterns, and cytology interpretation at each referral center, which might have affected the risk of malignancy in each diagnostic category.

5 | CONCLUSION

In pediatric patients, parotidectomy is mostly performed for benign and non-neoplastic diseases. Facial nerve palsy is significantly associated with malignant tumors and total parotidectomy. MRI is the most accurate imaging modality for diagnosing malignant lesions. FNA results exhibit moderate agreement with the final pathology.

CONFLICT OF INTEREST

All authors declare that there is no conflict of interest

DATA AVAILABILITY STATEMENT

Data available on request from the corresponding authors

PATIENT CONSENT STATEMENT

The need for informed consent was waived by the institutional review board of all sites.

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