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Pediatric Pleomorphic Adenoma of Parotid Gland: A Case Report

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Abstract: Pleomorphic adenoma is a type of tumor that affects the parotid glands. It is also called a benign mixed tumor (BMT) because it has both epithelial and myoepithelial cells. This type of tumor is the most frequent parotid gland tumor, making up about 66% of all such tumors in parotid glands. We reported a case of 12-year-old male patient with a lump on his parotid gland, measured 7x3x3cm at Prof. Dr. R. D. Kandou Hospital Manado. CT Scan results of neck region of the patient revealed that there was a mass in the right side *colli* area and right frontal sinusitis A histopathological examination of the tumor tissue showed that it was encapsulated by connective tissue and composed of proliferating epithelial and myoepithelial tumor cells, some with clear nuclei, arranged in solid and microcystic patterns, and interspersed with fibromyxoid stroma. The patient was diagnosed as parotid gland adenoma, therefore, a parotidectomy was performed on him. He was discharged home and advised to return for a check-up one week later to remove the stitches. At the two-month follow-up visit, he was stable and did not require any additional therapy. In conclusion, a parotidectomy shows good result in the patient with parotid gland adenoma. However, due to the recurrence rate of parotid gland malignancies, several oncologists prefer to observe the patient periodically trough follow up at policlinic. Early detection and right management are required to achieve the best prognosis.
Keywords: pleomorphic adenoma; pediatric patient; parotid gland

INTRODUCTION

A pleomorphic adenoma is a type of tumor that affects the parotid glands. It is also called benign mixed tumor (BMT) because it has both epithelial and myoepithelial cells, and is the most common salivary gland neoplasm, affecting approximately 2-3.5 people per 100,000 per year.[1,2]

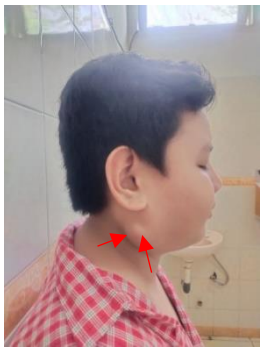
Parotid tumors are extremely rare in children, with fewer than 5% occurring in children compared to adults. Tumors in the minor parotid glands are uncommon, and pleomorphic adenomas are the most common benign tumors of the parotid gland. Pleomorphic adenoma has a remarkable degree of morphological diversity, with the essential components being the capsule, epithelial and myoepithelial cells, and the mesenchymal or stromal elements. There was a higher prevalence of females (with a female-to-male ratio of 1.43:1) in children, similar to the ratio seen in adult cases. In this study, the parotid glands were the most frequently affected salivary glands (56.7%), though this is a lower rate compared to findings from other reports that focused on cases involving children, adolescents, and adults. Benign salivary gland tumors are uncommon in children compared to adults. The most frequent type is pleomorphic adenoma, which accounts for 66.6% to 90% of these tumors in pediatric patients. The majority of benign salivary gland tumors are found in the major salivary glands, with approximately 70% to 85% of pleomorphic adenomas occurring in the parotid gland.[3-5]

However, the majority of patients seeking medical attention for this condition present with a painless mass on the side of the face.[3] As such, it is incumbent upon primary care providers and nurse practitioners to be knowledgeable of the differential diagnosis of such lesions and make appropriate referrals. The United Kingdom National Multidisciplinary Guidelines recommend complete excision for all first-presentation pleomorphic adenomas, with no need for routine adjuvant. Options include enucleation, a procedure which has fallen out of favour due to high recurrence rates, superficial parotidectomy, or total parotidectomy. Recently, extracapsular dissection has become more common, involving excision of the encapsulated tumor with a rim of healthy tissue, without dissection of the nerve.[1,6]

Pleomorphic adenomas have a propensity for local recurrence, with rates ranging from 20 to 45% after enucleation to between 2 and 5% following superficial parotidectomy treatment. The elimination of the superficial lobe ensures a very low incidence of tumor recurrence, but it can cause a number of major and minor complications due to the dissection of the facial nerve and the loss of at least three quarters of the parotid gland volume. The knowledge of the potential risks and complications associated with superficial parotidectomy may be relevant to offer better preoperative counseling to patients, improve preoperative planning, and achieve better long-term results.[1,6] This study discussed a case of pleomorphic adenoma in a child at Prof. Dr. R. D. Kandou Hospital, Manado.

CASE REPORT

We reported a case of a 12-year-old boy who presented with a lump on the right side of his face, under his ear for four years (since 2019) before being admitted to the hospital. The lump first appeared after the patient had a fever, cough, and runny nose, and it was initially small like a marble but gradually grew larger. The patient did not experience any pain, fever, feeling of tightness, weight loss, or appetite loss due to the lump, which was movable and firm to touch. The color of the lump matched the surrounding skin (Figure 1). A physical examination revealed a lump measuring 7x3x3 cm. A CT-scan of the neck in November 29, 2023 suggested a parotid abscess, a mass in the neck area, and sinusitis in the frontal area (Figure 2). The patient's laboratory tests were normal. The patient was diagnosed with a suspected malignant parotid tumor on the right side and underwent surgery to remove the mass (Figure 3). A histopathological examination of the tumor tissue showed that it was encapsulated by connective tissue and composed of proliferating epithelial and myoepithelial tumor cells, some with clear nuclei, arranged in solid and microcystic patterns, interspersed with fibromyxoid stroma (Figure 4). The diagnosis was parotid gland adenoma. The patient was followed up for two months after surgery and showed no complications or complaints.



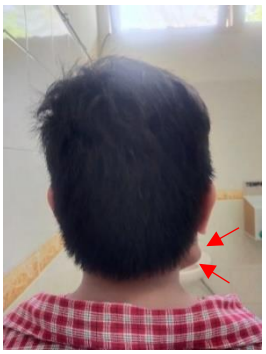


Figure 1. Clinical photo of the patient before parotidectomy, bumps and lumps under the right ear; (left) lateral view, (right) back view

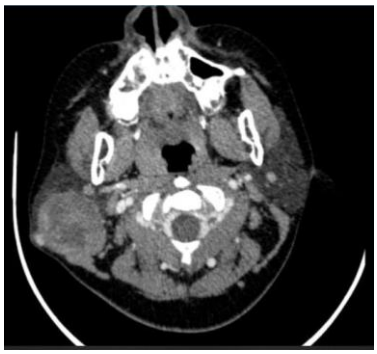


Figure 2. CT Scan results of colli on patient, there is a mass in the right side colli area, and right frontal sinusitis

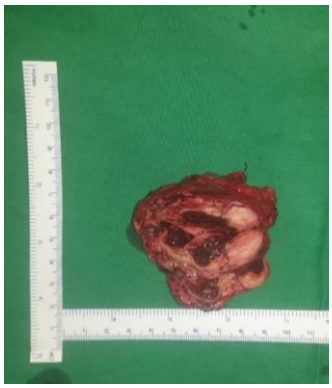
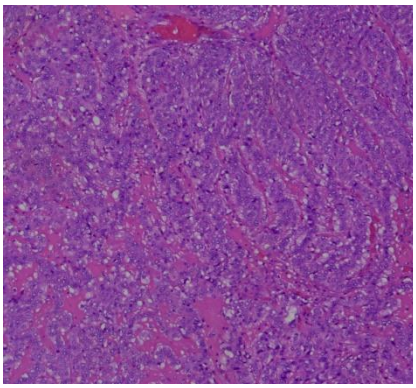


Figure 3. Mass photo after tumor removal



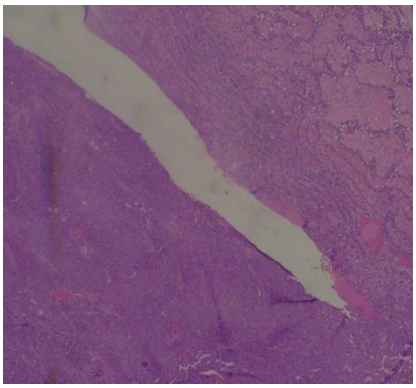
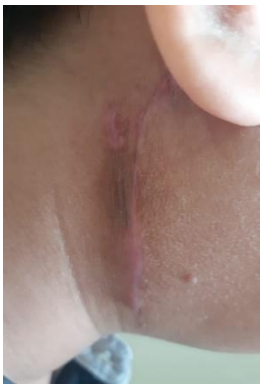


Figure 4. Histological photo biopsy showed proliferation of epithelial and myoepithelial tumor cells arranged solidly, trabecular microcytic between the stroma of fibromyxoid connective tissue with pleomorphic conclusions of parotid gland adenoma. (left) 100x magnification, (right) 250x magnification

The patient was discharged home and advised to return for a check-up one week later to remove the stitches. At the two-month follow-up visit, he was stable and did not require any additional therapy (Figure 5). He was scheduled to have regular follow-ups every six months.



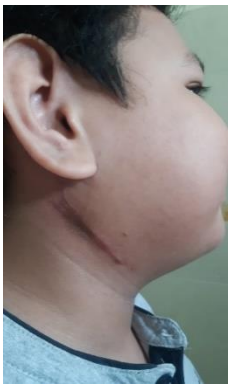


Figure 5. Clinical photo of the patient after parotidectomy

DISCUSSION

Parotid gland tumors are rare, despite in some cases they could develop into malignancies, but it is reported for only 0.3% of all malignancies. They occur in about 5% of head and neck cancers, with less than 5% of cases occurring in children. It is important to note that the majority of parotid tumors (about 80%) are benign, while submandibular gland tumors are more likely to be malignant.[7] Early detection and treatment are important for managing parotid gland tumors. In case of any unusual lumps or swelling in the head or neck area, it is important to consult a

healthcare provider for further evaluation.

Benign tumors that commonly occur in the parotid gland are pleomorphic adenoma and pilomatrixoma. On the other hand, mucoepidermoid carcinoma is the most frequent malignant tumor that affects the parotid gland.[8] Many patients with parotid tumors usually report a gradually enlarging mass, similar to what our patient experienced, while others are diagnosed incidentally during routine medical examinations conducted by their doctors. According to some reports, ultrasound imaging can be used as a first step to distinguish cystic or vascular lesions from solid masses. However, MRI is a more comprehensive diagnostic tool that helps in preoperative assessment of the extent of the mass, detection of facial nerve and regional lymph node involvement, and differentiation from other malignancies or vascular tumors.[7] Following surgical intervention, the recurrence rates for these tumors are remarkably low, ranging from 0–2%. In children, the facial nerve is situated more superficially and is more susceptible to dissection and stretching compared to adults. Therefore, it is recommended to utilize facial nerve monitoring during parotidectomy to mitigate the risk of facial nerve palsy or paresis.[8] In patients with salivary gland tumors, systemic therapy is usually reserved for those with rapid disease progression or advanced or incurable disease. It can also be used in patients with recurrent disease that is not amenable to surgery and/or radiation therapy.[9]

Ultrasound-guided fine needle aspiration cytology (FNAC) helps in determining the subtype of the parotid tumor on the basis of histopathology. It can be conducted only after vascular tumors are ruled out. Parotid gland tumors (both benign and malignant) have been classified into 24 distinct histological subtypes by the World Health Organization.[9,10] In our patient, FNAC findings showed the presence of epithelial cells, myoepithelial cells, and mucoid material. Hence, the tumor was classified as pleomorphic adenomas of the parotid gland. Superficial parotidectomy with en-bloc excision of the tumor mass, with preservation of the facial nerve, is the surgery of choice for parotid pleomorphic adenoma involving the superficial lobe.[7]

Parotidectomy, can be done partial or complete and was performed for a variety of reasons, such as inflammatory conditions, infections, congenital anomalies and tumor, wether benign or malignant. The complexity of the surgery necessitates highly skilled surgeons, especially because of the proximity of the facial nerve to the gland. The most critical aspects of the procedure are the identification and preservation of the facial nerve, which is paramount to oncological safety when malignancy is involved. This focus on identification is essential as it is often challenging to accurately predict the tumor's position relative to the facial nerve preoperatively unless there is pre-existing facial paralysis caused by the tumor.[7] The most common reason for a parotidectomy is to remove the neoplasm. If the deeper lobe is involved, then total or radical parotidectomy is the procedure of choice, depending on the extent of the tumor mass and involvement of the facial nerve. Following surgery, recurrence rates are as low as 0–2%. The facial nerve in children is located more superficially and is more sensitive to dissection/stretching than in adults. Hence, facial nerve monitoring during parotidectomy is recommended to reduce the risk of facial nerve palsy/paresis.[7] The most frequently observed complication after performing either superficial or total parotidectomy was temporary facial muscle weakness. All affected patients regained complete facial nerve function by their initial postoperative appointment, which occurred one week after surgery. One case required the removal of the superior branches of the facial nerve, followed by nerve graft repair after the tumor was excised. This patient regained partial facial function, with synkinesis being observed.[11]

As most patients can tolerate pleomorphic adenoma of the parotid for a long duration without any symptoms of discomfort, they often delay medical advice. However, such delays increase the risk of malignant transformation.[12] The most frequent malignant tumor of the salivary glands in children is mucoepidermoid carcinoma, followed by adenocarcinoma and acinic cell carcinoma. In cases where children have mucoepidermoid carcinoma of the parotid gland, the histological assessment is typically of low grade, and the prognosis is generally positive with suitable treatment. Studies that look at all pediatric salivary gland masses, rather than just those where a

parotidectomy sample was taken, indicate that benign lesions are overwhelmingly more common. For instance, in a case series from the Massachusetts Eye and Ear Infirmary involving 22 children who had persistent parotid mass over an 8-year period, only one child was found to have a malignant tumor (mucoepidermoid carcinoma).[13] As in the case of benign parotid tumors, surgery is also the mainstay in the management of malignant parotid tumors.[14] Simultaneous neck dissection is indicated in high-grade malignancies, wherein lymph node involvement is confirmed both clinically and radiologically.[15] The decision to administer postoperative radiation therapy is generally made by the treating radiation oncologist in collaboration with the head-and-neck surgeon. Radiation therapy is applied to the parotid bed, with the technique varying based on the timing of treatment, extent of the disease, and the judgment of the radiation oncologist. The treatment area is designed to encompass the entire parotid bed with margins of 2–3 cm. Treatment is administered using a continuous-course approach with daily sessions. The median radiation dose is 5,000 cGy, ranging from 4,500 to 6,000 cGy, and is determined at the discretion of the radiation oncologist.[16] Adjuvant radiotherapy is considered in cases of incomplete surgical resection, persistent lymph node involvement, perineural invasion, or an aggressive histological grade tumor.[17] However, the risk of post-irradiation complications such as facial/dental deformities, secondary malignancies, trismus, and hyposialia should be carefully evaluated. Perioperative systemic chemotherapy is reserved for malignant forms of parotid gland tumors which are rapidly progressing, recurrent, metastatic, incurable, or unresectable.[7]

CONCLUSION

We reported a case of 12-year-old male patient with a lump on his parotid gland. CT Scan results of neck region of the patient revealed a mass in the right side colli area and right frontal sinusitis A histopathological examination of the tumor tissue showed a diagnosis as parotid gland adenoma, therefore, a parotidectomy was performed on him. The patient was followed up for two months after surgery and showed no complications or complaints. Due to the recurrence rate of parotid gland malignancies, several oncologists prefer to observe patient periodically trough follow up at policlinic. Early detection, and right management are required due to patient best prognosis.

Conflict of Interest

The authors affirm no conflict of interest in this study.

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