

## CASE REPORT

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# Pleomorphic Adenoma of Minor Salivary Glands in Child

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## ABSTRACT

**Introduction:** Tumors of salivary glands are very rare in children overall, and rarely in minor salivary glands. Among them pleomorphic adenoma (PA) is the most common benign tumor of salivary glands in children and it accounts 60% of all salivary neoplasms, frequently found in major salivary glands (85 %) and rarely in minor salivary glands (10-15%). PA appears as painless oval mass, elastic consistency and smooth surface. It is characterized by slow evolution of growing. The diagnosis can be confirmed only by histopathological features, after total excision of the lesion. **Case report:** This case report presents the uncommon case of the formation well circumscribed in upper lip region, nearby the frenulum labii oris superior in 10 years old school girl. After clinical intraoral examination of the formation approximately 2cm in size the surgical treatment has been done and histopathologic analysis resulted as PA. **Conclusion:** Long term follow-up examination is necessary due to possible recurrence or malignancy alteration.

**Keywords:** tumor, salivary glands, pleomorphic adenoma.

## 1. INTRODUCTION

Tumours of salivary glands are very rare in children overall, and rarely in minor salivary glands (1, 2). Among them pleomorphic adenoma (PA) is the most common benign tumor of salivary glands in children and (3, 4). It accounts 60% of all salivary neoplasms (5), frequently found in major salivary glands (85 %) and rarely in minor salivary glands (10-15%) (3). Hard and soft palate are the commonest places of its appearance while in the lips PA is presented in 20- 40% of all intraoral PA, with higher frequency in upper than lower lip (6, 7). Etiology of PA remains unclear although exposure to radiation and virus infection (SV40) has been reported as a causative role in development of PA (8). The histology shows two cell types:

The ductal epithelial cell and myoepithelial cell, which may vary in variety of cell lines, considering that pleomorphic means many forms. The morphological variety, exists among the tumor between individuals and glands, and even within the same tumor, which actually explains the terminology (9,10). It has connective tissue capsule which may be incomplete which explains frequent relapse of this pathology. Recurrence and sometimes malignancy transformation to adenoid cystic carcinoma or

ex mucoepidermoid carcinoma is predisposed when leaving residual tumor while surgical procedure (11). Modality of treatment is complete surgical excision with margin of normal uninvolved tissue.

## 2. CASE REPORT

Osteomas A 10- years old school-girl has been referred to the Department of Oral Surgery in University Dental Clinical Centre of Kosovo complaining for appearance of the formation in the upper lip which has been progressively grown in past three years (Figure 1). The anamnesis reported an asymptomatic increase of the mass in upper lip in previous 3 years. Clinical examination shows the 2 x1,5 cm firm, nodular well circumscribed and non-tender lesion present in the region corresponding frenulum of the upper lip which was clinically suspected to be as lipoma. The overlying mucosa was without physiological changes. No regional lymphadenopathy was not observed. No anamnestic features related to systemic disease were registered.

The Surgery was performed in the operating theater regarding to the standard infection control guidelines for surgery using Chlorhexidine solution 1% for intra- and extraoral surgical field disinfection and covered with sterile surgical drapes. Ap-

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Figure 1. Preoperatively situation



Figure 2. Resected specimen of the PA



Figure 3. Excision during the surgery



Figure 4. Suturing with 3/0 silk suturing material

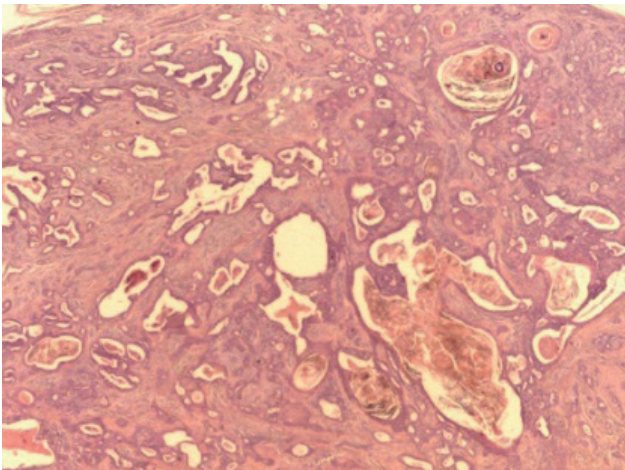


Figure 5. Hematoxylin & Eosin, 2.5x

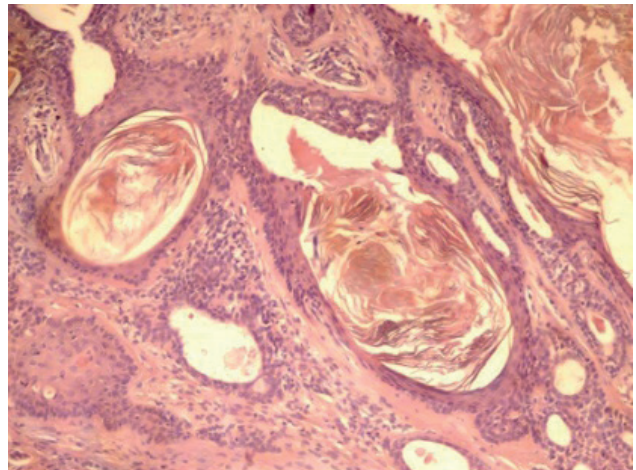


Figure 6. Hematoxylin & Eosin, 5x

appropriate local analgesia was secured using 4 ml of 2o/o Lidocaine with 1:800,000 Adrenaline (Alkaloid, Skopje) for appropriate anesthesia of ramii alveolaris superior anterior and nervus nasopalatinus scarpae.

The surgery was realized with intraoral approach. Total exision of the well encapsulated mass approximately 2 cm in size was done (Figure 2) and macroscopy with no invasion on the adjacent structures (Figure 3), followed by suturing as shown in Figure 4. Histopathology of the tumoral tissue shows Tumor tissue displaying cystic and ductal structures (Figure 5), Hematoxylin & Eosin, 2.5x) lined by keratinizing squamous (Figure 6), Hematoxylin & Eosin, 5x) and cuboidal (Figures 7 and 8) Hematoxylin

& Eosin, 5x) epithelium. The patient was examined one week later and suture has been removed. The follow-up examination one year later shows no signs of its recurrence (Figure 9). Considering the possibility of tumor recidivation, patient will be in routine control every 12 months in the next two years.

### 3. DISCUSSION

PA is the most common salivary gland, mainly in the major salivary glands. They are comm, only occur in women 4th-6th decade of life (1, 2). Laccourreye et al reported ratio 1:1,4 male to female (12). The incidence of salivary gland tumors in children is low and it differs



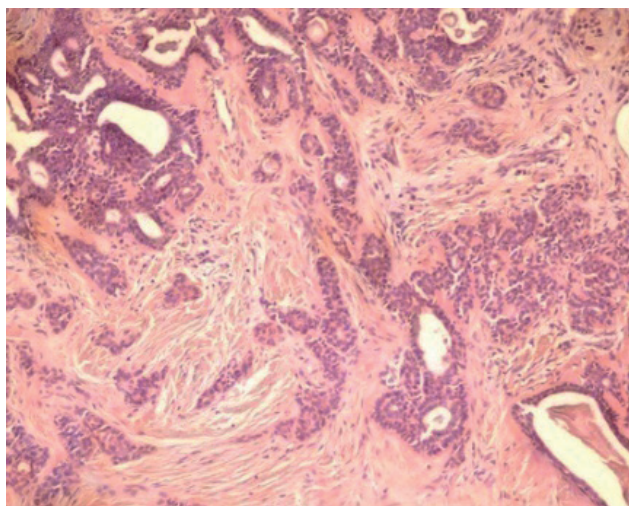


Figure 7. Hematoxylin & Eosin, 5x

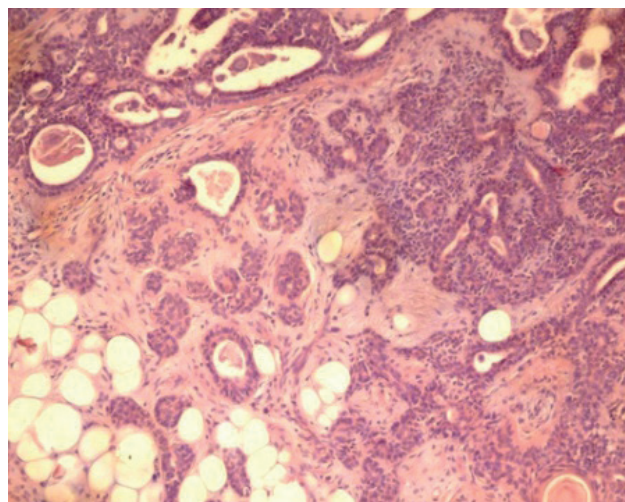


Figure 8. Hematoxylin & Eosin, 5x



Figure 9. The follow-up examination one year later

from the adults less than 5%. First decade of life was found to be 0,25 % represented for PA appearance (13, 14, 15). Krolls et al. in 430 diagnosed cases of salivary gland tumors in children, found 11 PA of the minor salivary gland (16). In literature most of the PA originated from the minor salivary glands in children was recorded between 10 and 18 years of ages (2, 14, 17). Palate is the most common site of MSGTs, and is followed by the lips and cheek (1, 2, 18). The upper lip has higher prone for developing benign neoplasms including PA compare to lower lip 6:1, whereas malignant tumors are predominate. This can be explained due to embryonic development differences (19, 20).

PA are benign salivary gland tumor, slow-growing, painless and in most of the cases they are well encapsulated. Its growth within many years and in the most common cases is presented as a submucosal lump, rarely with pain, ulceration and bleeding of overlying mucosa (21). The PA is usually encapsulated and the treatment of choice is surgical excision with adequate surrounding tissue margins. tumors of the minor salivary glands has lower risk for recurrence. The main cause of its recurrence is reported inappropriate surgery procedure of total tumor excision. main cause is reported inadequate (21). Recurrence of PA rare in minor salivary gland tu-

mors is of lower risk as also malignant transformation to malignant adenoid cystic or mucoepidermoid carcinoma. The recurrence rate of PAs with two or more years follow-up is 13.0%.

Clinical examination and histopathologic study is required for definitive diagnosis of PA because of its rare appearance in this location.

Due to functional difficulties as eating and talking, PA of minor salivary glands in upper lip usually can be diagnosed earlier than PA in major salivary glands, also treated before arising the high dimensions (19).

PA in upper lip in young patients should be consider in differential diagnoses with, neurofibroma, lipoma, dermoid cyst, mucocele, fibroma, adenoid cystic carcinoma and mucoepidermoid carcinoma and swelling regarding to other etiologies (17, 22, 23).

The treatment of choice of PA is complete excision of the tumor and its capsule. Recurrence and malignant transformation is a concern of this pathology therefore long- term follow-up is recomanded (23, 24).

#### 4. CONCLUSION

Frontal In most of the cases PA does not recur after adequate surgical tretament. Complete surgical excision will provide the definitive diagnosis with histopatologic analysis and treatment of PA.

- Conflict of interest: The authors declare that they have no conflict of interest.
- Authors contributions: All authors participated in each step of research process.

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