Juvenile Pleomorphic Adenoma: A Case Report

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Abstract Pleomorphic adenoma is a benign salivary gland tumor that represents less than 5% of benign tumors in childhood. World literature suggests salivary gland tumors account for less than 3% of the head and neck tumors and benign Pleomorphic adenoma of minor salivary glands arising de novo is very rare. Here we report a case of Pleomorphic adenoma of minor salivary glands occurring on the palate of 12 year old female. Salivary glands may present with a diverse range of lesions presenting a challenge to even the most experienced clinician and pathologist. Resection with surrounding dispensable normal tissues is the key to successful treatment of such tumors.

Keywords: children, Pleomorphic adenoma, salivary gland tumor


1. Introduction

Neoplasm’s originating from salivary glands are relatively uncommon in childhood, constituting less than 5% of the total number of salivary gland neoplasms [3,7]. Pleomorphic adenomas occur at any age but more common in patients between 30-60 years old and slightly more frequent in women (male-female ratio 1:1.39). Pleomorphic adenoma in children is rare [6]. Pleomorphic adenoma is of glandular origin in the head and neck, usually presenting as a mobile slowly growing painless firm swelling that does not cause ulceration of the overlying mucosa [5]. The most common sites of Pleomorphic adenoma of minor salivary glands are the palates followed by lips and cheeks [4]. Microscopically, benign mixed tumors are characterized by variable, diverse structural histological patterns [2,8].

The purpose of this case is to report a case of Pleomorphic adenoma of minor salivary gland with the characteristic features.

2. Case Report

A 13 year old female referred to the department of oral medicine and radiology with an asymptomatic swelling on the upper part of the jaw with duration of 7-8months. Her past medical/dental history was not significant. There was no history of pain or sensory changes or trauma.

Physical examination revealed a solitary swelling on the right side of the palate, oval in shape and had a smooth surface. The mucosa over the swelling appeared normal. There was no signs of inflammation or ulceration (Figure 1). On palpation, the swelling was non-tender, firm in consistency, non-fluctuant, non-reducible, compressible and non-pulsatile. There was no history of bleeding. Panoramic radiography revealed no abnormality.

Fine needle aspiration cytology revealed mixture of epithelial and myxoid stroma. Epithelial cells are scattered freely and are also in small cohesive clusters. Cells are uniform in size, round to oval with eccentrically placed nucleus, inconspicuous nucleoli and moderate to densely stained cytoplasm (plasmacytoid). Mild to moderate nuclear atypia observed. Background shows fibrillary fibromyxoid stroma. Features were suggestive of Pleomorphic adenoma.

Based on clinical and cytological findings, a provisional diagnosis of benign Pleomorphic adenoma of minor salivary gland was made. Surgical excision for tumor was performed under general anesthesia. Microscopically, the tumor showed cells forming ducts, small cellular nests, and solid sheets of cells and anastomosing cords. Large portion of the tissue showed sheets of loosely cohesive cells with formation of occasional tubular structures. Few areas showed ductal structures resembling normal salivary gland intercalated ducts. Myxoid component was noted in few areas. Islands of mucous cells were also seen if few
are uncommon neoplasms of upper aero digestive tract sites. Tumors originating in the minor salivary glands, palate and anterior soft palate being the most common for approximately 10% of PA’s with the posterior hard adenomas arising from oral minor salivary glands account gland, which accounts for 60-80% of PA’s. Pleomorphic adenomas appear to be completely separated from the main tumor common feature and sometimes lobules of tumor may encapsulated but extension of tumor into the capsule is a Pleomorphic adenomas are usually well demarcated or attain a size greater than 1-2cm in diameter. Because this combination of epithelial and stromal components. Revealed a well defined capsulated tumor mass with the myoepithelial cells are often polygonal with a pale eosinophilic cytoplasm giving an epithelioid or plasmacytoid phenotype.

Our diagnosis was confirmed by tubular and ductal patterns lined by single to double layered cuboidal cells with large darkly stained nucleus in the center. Connective tissue capsule is absent. Although uncommon, Pleomorphic adenoma should be considered in the differential diagnosis of young patients with swellings in the oral cavity, particularly in the palate, lips, tongue and buccal mucosa. The biological behavior in the young patients seems to be similar to that in adults, with a very low recurrence rate after complete surgical resection.

3. Discussion

Pleomorphic adenoma is more prevalent in the 4-6th decades of life; however cases in the first two decades have been reported and are more common in females than males. Salivary gland Pleomorphic adenomas are extremely rare in children and adolescents. Pleomorphic adenomas are the most common types of salivary tumors (40-60%) and mostly occur in the parotid gland, which accounts for 60-80% of PA’s. Pleomorphic adenomas arising from oral minor salivary glands account for approximately 10% of PA’s with the posterior hard palate and anterior soft palate being the most common sites. Tumors originating in the minor salivary glands are uncommon neoplasms of upper aerodigestive tract.

Children account for less than 5% of all patients with salivary gland tumors, and within the first decade of life, less than 0.25% were found. The most common symptom of PA reported was a sub mucosal lump, although few cases showed ulceration, pain and bleeding. Pleomorphic adenoma is a benign tumor with slow growth; it seems not to change for many years and express itself simply as a non painful mass. Pleomorphic adenomas are usually well demarcated or encapsulated but extension of tumor into the capsule is a common feature and sometimes lobules of tumor may appear to be completely separated from the main tumor mass.

PA of intraoral accessory glands seldom is allowed to attain a size greater than 1-2cm in diameter. Because this tumor causes the patient difficulties in mastication, talking and breathing, it is detected and treated earlier than tumors of major glands. Histopathological examination revealed a well defined capsulated tumor mass with combination of epithelial and stromal components. Foote and frazell 1954 categorized the tumor into the following types.

Principally Myxoid
Myxoid and cellular components present in equal proportion. Predominantly cellular and Extremely cellular. The epithelial component forms ducts and small cysts that may contain an eosinophilic coagulum, the epithelium may also occur as small cellular nests, sheets of cells, anastomosing cords and foci of keratinizing squamous or spindle cells. Myoepithelial cells are a major component and are responsible for characteristic mesenchyme like changes. When the pleomorphic pattern of stroma is absent, and the tumor is highly cellular, it is often referred to as cellular adenoma. In the minor glands, lesions are often more solid or cellular than those seen in the major glands, and the myoepithelial cells are often polygonal with a pale eosinophilic cytoplasm giving an epithelioid or plasmacytoid phenotype.

4. Conclusion

The salivary glands may show a diverse range of lesions presenting a challenge to even the most experienced clinician and pathologist. Pleomorphic adenoma of minor salivary gland is a tumor of rare occurrence and a diagnosis should be made carefully lest a major salivary gland be resected. High index of suspicion and an adequate clearance of the tumor with a cuff of surrounding dispensable normal tissues is the key to successful treatment of such tumors.

References