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Pleomorphic adenoma of the palate in a child: A case report

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Abstract

Salivary gland tumors are rare in children and the incidence differs from the adult counterpart. When salivary gland tumors do arise in children, they preferentially affect major salivary glands, but minor salivary gland tumors have also been reported. We reported the first case of palatal pleomorphic adenoma in a 13 year-old child from Thailand. She came to Sawanpracharak hospital with the chief complaint of a swelling at the left side of the palate. The oral mucosa covering the lesion was intact. Occlusal radiograph revealed no bony destruction. Incisional biopsy was performed on this patient. The biopsy showed several ducts which were lined by cuboidal cells. These ducts were surrounded by myoepithelial cells, some of which had the plasmacytoid appearance The patient was treated by wide local excision and no recurrence was observed 8 years after the surgery. Differential diagnoses of a palatal swelling in children and treatment of pleomorphic adenoma at the palate were also discussed.

Key words: *Pleomorphic adenoma, palate, child.*

Introduction

Salivary gland tumors account for less than 3% of the head and neck tumors (1) . They are more common in adults than in children (2,3). Only 0.32-5% of all salivary gland tumors occur in children aged 16 years or younger (1,4). Among all salivary gland tumors, pleomorphic adenoma is the most frequently encountered lesion, accounting for approximately 60% of all salivary gland neoplasms (5-10). It also ranks as the most common salivary gland neoplasm in children, representing 66-90% of all salivary gland tumors (5,11). Most salivary gland tumors occur in major salivary glands, especially the parotid gland. Pleomorphic adenoma which is the most common salivary gland tumor predominantly occurs in the parotid gland. As far as the intraoral salivary gland tumors are concerned, pleomorphic adenoma also ranks as the most frequently encountered lesion (10,12,13). Palate is the most common affected site. The second most common site is the upper lip followed by buccal mucosa (10,14-16). Pleomorphic adenomas in minor salivary glands are rare in children and predominantly occur in the palatal glands. Other intraoral sites include upper lips, buccal mucosa, tongue and gingiva (17,18).

Case Report

A 13-year-old girl came to Sawanpracharak hospital with the chief complaint of a swelling at the left side of the palate (Fig.1). Her medical history was non-contributory

and she denied drug allergy. She gave the history that the swelling had been there for 3 years, but grew quite rapidly in the past 3 months. Intraoral examination revealed a soft tissue mass 1.5 cm. in greatest diameter at the left side of the hard palate. The oral mucosa covering the lesion was intact. The lesion was rubbery in consistency and no tenderness on palpation was observed. Occlusal radiograph revealed no bony destruction. Incisional biopsy was performed on this patient. Microscopically, the lesion revealed stratified squamous epithelium covering the connective tissue. The underlying connective tissue showed several ducts which were lined by cuboidal cells.

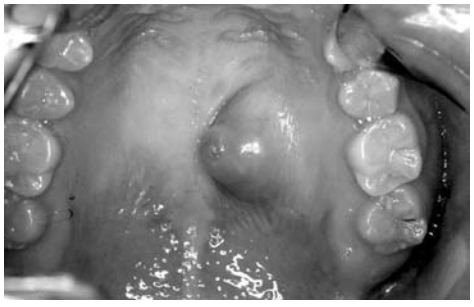

Fig. 1. Photograph showing a swelling at the left side of the palate.

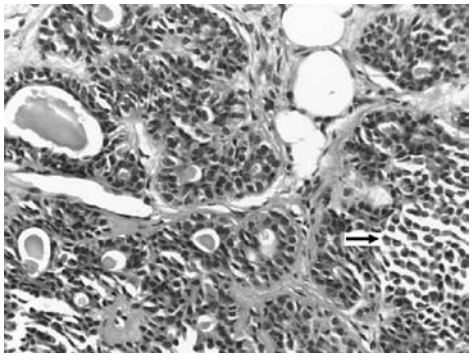

Fig. 2. Photomicrograph showing several ducts surrounded by myoepithelial cells, some of which had the plasmacytoid appearance (arrow). (Haematoxylin & eosin stain, original magnification 20X).

These ducts were surrounded by myoepithelial cells, some of which had the plasmacytoid appearance (Fig.2). Some of the ducts contained amorphous eosinophilic materials. The diagnosis was pleomorphic adenoma. The patient was treated by wide local excision with the placement of upper stent. The patient's post-operative course was uneventful. No recurrence was observed after a follow-up period of 8 years.

Discussion

For salivary gland tumors in children, it has been suggested that they have the proclivity to be malignant than to be benign regardless of the site (19). Mucoepidermoid carcinoma is the most common malignant salivary gland tumor, while pleomorphic adenoma is the most common benign counterpart (4,5,19). Pleomorphic adenomas at the palate in children and adolescents are rare. Byars et al.(20) reviewed 470 cases of salivary gland tumors in patients 18 years of age and younger and first reported 2 cases of palatal pleomorphic adenoma in children. Since then, there have been 13 additional cases in the English language literature (3,17,18,20-28). Recently, Daniels et al.(29) reported 2 more such cases, bringing the total number of cases to 17. The age of the 16 patients ranged from 5 to 17 years with the mean age of 10.75 years. The age of 10 of the 16 cases (62.5%) was in first decade of life. The age of the patient in our case is slightly older than most previous reports. The gender of the patient in our case is in accordance with previous reports since females slightly outnumber males by a ratio of 1.29:1.

The differential diagnoses for this case include palatal abscess, odontogenic and non-odontogenic cysts, soft tissue tumors and salivary gland tumors. Palatal abscess can be ruled out by clinical examination since the source of palatal abscess which is typically a nonvital tooth in vicinity or a localized periodontal defect was not found. In addition, this patient showed no sign of inflammation. Both odontogenic and non-odontogenic cysts can be ruled out at the time of exploration into the mass since it did not demonstrate cystic nature. Palatal tissues contain components of soft tissue and harbour minor salivary gland tissues. As a result, soft tissue tumors such as fibroma, lipoma, neurofibroma, neurilemmoma as well as salivary gland tumors should also be considered in the differential diagnoses for this case. Lymphoma can also present with palatal swelling in children. The fact that this patient did not exhibit sign and symptom associated with malignant tumor such as ulcer, pain or paresthesia coupled with a rather slow development of the lesion over 3 years makes the differential diagnosis of lymphoma unlikely.

The treatment of palatal pleomorphic adenoma in children is the same as in adults (29). The treatment of choice for pleomorphic adenoma in minor salivary gland is wide local excision with the removal of periosteum or bone if they are involved. Simple enucleation of this tumor is believed to lead to high local recurrence rate and should be avoided (30). Rupture of the capsule or tumor spillage is also believed to increase the risk of recurrence, so meticulous dissection is paramount. Pleomorphic adenoma generally does not recur after adequate surgical excision. Recurrence of palatal pleomorphic adenoma in children following surgical treatment has been reported in 2 cases out of 16 cases from the English language literature (3,20).

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