

A Case Report of Juvenile Intraoral Pleomorphic Adenoma

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Abstract

Pleomorphic adenoma (PA) is the most common tumour of the salivary glands. About 90% of these tumours occur in the parotid gland and ten % in the minor salivary glands. The most common sites of Pleomorphic adenoma of the minor salivary glands are the palate, followed by lips and cheeks. Throat, floor of the mouth, tongue, tonsil, pharynx, retro molar area and nasal cavity are rarely involved. Pleomorphic adenoma typically present as a slow growing, unilateral and painless mass of salivary glands, with a predilection for recurrence and risk of malignant transformation (about 1.5% up to five years and increases to 9.5% after more than 15 years). This case report describes a case of Pleomorphic adenoma of buccal mucosa in a 13-year-old male patient.

Key-words: Buccal mucosa, Pleomorphic adenoma, Salivary gland tumor

Introduction

Pleomorphic adenoma (PA) has been defined by World Health Organization in 1972 as a circumscribed tumour characterized by its pleomorphic or mixed appearance, clearly recognizable epithelial tissue being intermingled with tissue of mucoid, myxoid and chondroid appearance¹. Pleomorphic adenoma is one of the most common benign tumour affecting salivary glands. It accounts for 53–77% of parotid tumours, 44–68% of submandibular tumours, 6.4% occur in the minor salivary glands. Pleomorphic adenoma arising from minor salivary glands in the nasopharynx and parapharyngeal spaces has also been reported although in rare instances². It appears as a unilateral painless, slowly growing, firm mass typically in adults from the third to fifth decades and 60% of the cases are reported in females³. The other intraoral sites are lips, buccal mucosa, tongue, floor of the mouth, and retromolar trigone (0.7%)⁴. Histologically, PA consists of cells

with epithelial and mesenchymal differentiation. The treatment of choice for PA is surgical removal with safety margins, to prevent the recurrence. Recurrence rate of 5 -30% has been found for PA, so a periodic follow-up is must, due to the important relapse potential and aggressiveness of these lesions⁵. As the mucosa of the cheek is a relatively rare site of occurrence for intraoral pleomorphic adenoma, the authors present a case in a 13 year old male patient.

Case History: A 13 year-old male patient with a painless swelling on the left buccal mucosa was presented to the Department of Periodontology. History of presenting illness revealed that the swelling was gradual in onset and had grown slowly to attain the present size. There was no history of fever, bleeding, pain, sensory changes and disturbance of salivation or trauma. The past dental history and medical history was unremarkable and no other abnormalities were found. General physical examination showed that patient was of moderate built, height and well-oriented to time and place and all the vitals were in normal limits. On intraoral examination, there was a two by three cm dome shaped mass on the left buccal mucosa in the region of 23,24,25. The surface was smooth and on palpation, the lesion was firm, non-tender, and mobile over the underlying tissues. No discharge was present. The colour of the overlying

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mucosa was normal. Based on the history and clinical findings, differential diagnosis consisting of fibroma, and mucocele were considered. Patient was advised to get blood investigations done (Complete Blood Count, Bleeding Time/Clotting Time, Haemoglobin, Prothrombin Time/International Normalised Ratio). A verbal and written consent form was obtained from patient's guardian. Wide excision of the lesion with primary closure was then carried out. The excised specimen was sent to the Department of Oral Pathology and Microbiology, for histopathological examination. Microscopic findings showed well encapsulated mass of epithelium circumscribing connective tissue. Epithelium appeared to be glandular in origin forming duct like structures with eosinophilic coagulum within. Tumour cells were present in the form of sheets, cords and nests of cells with atypical features. Connective tissue showed myxoid areas and endothelial lined blood vessels. An overall diagnosis of Pleomorphic adenoma was suggested. The patient was followed up for a period of three months during which satisfactory healing of the surgical site was observed and no evidence of recurrence was noticed.

Discussion

The minor salivary glands are small, independent, predominantly mucous glands that are found in every part of the upper respiratory tract. The majority of the minor salivary glands are located in the palate, while the others are found in the submucosa of the uvula, inner surface of the lips, around the opening of the parotid duct, in the mucous membrane of the cheek, floor of the mouth, palatoglossal folds, superior pole of the tonsils (Weber's glands), on the inferior surface of the tongue, near the frenulum of tongue, and within the palatine tonsil⁶. Pleomorphic adenoma is the most common salivary gland tumor. The main site of occurrence is the parotid gland, affecting patients of any age, more frequently between the third to fifth decades of life⁷. In 2002, Jansisyanont et al⁸ in a study concluded that a total of 80 minor salivary gland tumors were identified in 49 female patients and 31 male patients and the ratio range from 1.2:1 to 1.9:1, suggesting a more female predilection for this clinical entity. PA is a benign tumor consisting of cells capable of differentiating to epithelial (ductal and nonductal) cells and mesenchymal (chondroid, myxoid and osseous) cells⁹. A study

conducted on a Mexican sample showed that in minor salivary gland tumors, 64.2% are benign neoplasms and only 35.8% were malignant¹⁰. Variants of pleomorphic adenoma include Pleomorphic adenoma with lipomatous change, myxolipomatous Pleomorphic adenoma, Pleomorphic adenoma with squamous differentiation, and benign metastasizing mixed tumour¹¹. There are three histologic subtypes, myxoid (80% stroma), cellular (myoepithelial cells predominant), and mixed (classic)³. The majority of intraoral mixed tumours are less than three cm in diameter. They are usually solitary and well-circumscribed. The findings of the case presented here is in agreement with those of other investigators^{4,12,13}. The differential diagnosis of PA buccal mucosa includes buccal abscess, dermoid cyst, sebaceous cyst, mucocele, neurofibroma, lipoma, mucoepidermoid carcinoma and polymorphous low grade adenocarcinoma⁸. The possibility of buccal space abscess was ruled out due to absence of signs of inflammation. The solid nature of lesion coupled with lack of tissue representing the three germ layers rule out the possibility of mature dermoid cyst. Absence of punctum and freely movable nature of the mass differentiate PA from sebaceous cyst. As on histological picture both epithelial and myoepithelial cells were seen, which rules out mucoepidermoid carcinoma. The negative slip test clinically and absence of lipomatous component histologically rules out lipoma. The absence of perineural invasion and mitotic figures obscure the chances of polymorphic low grade adenocarcinoma¹⁴. Pleomorphic adenoma is known to produce recurrence either due to spillage, inadequate removal or enucleation at the time of operation, but is not known to produce distant metastasis¹⁵. A recurrence rate of two–44% has been reported in the literature. The ideal treatment of choice for PA is wide local excision with good safety margins and follows-up for at least three-four years¹⁶. In the present case report, wide local excision and primary closure was done and case was followed up till three months from the date of surgical intervention, and presently the case is still under follow-up procedure.

Conclusion:

Pleomorphic adenoma, though a benign tumor of salivary gland, should be diagnosed at an early stage and complete local surgical excision with negative microscopic margins is recommended. Care must be

taken to remove the lesion entirely to avoid recurrence and malignant transformation.

Declaration of Patient consent:

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that his name and initials will not be published and due efforts will be made to conceal his identity, but anonymity cannot be guaranteed.

Conflicts of Interest: Nil

Source of Funding: Self

Acknowledgement: Department of Pediatric and Preventive Dentistry, School of Dental Sciences, Sharda University, Greater Noida.

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