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Case Report

Juvenile buccal pleomorphic adenoma: a rare case report

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ABSTRACT

Salivary glands tumors are relatively uncommon in pediatric age group. Less incidence is expected in minor salivary glands of pediatric patients. In the buccal minor salivary glands only five cases reported as pleomorphic adenoma in english literature. Herein we present an 11 years old boy presented with right side buccal mass which is mobile on physical examination with normal overlying buccal mucosa. Histopathology revealed a pleomorphic adenoma. Such case is rare in this part of oral cavity especially in young age group. our case is the third youngest patients with buccal pleomorphic adenoma. Reporting it will add a material to the literature for further studies on such disease presentation.

Keywords: Pleomorphic adenoma, Buccal mass, Juvenile, Salivary gland tumor

INTRODUCTION

Minor salivary gland are distributed in the oral cavity and referred as palatal, buccal, labial, lingual, glossopalatine and retromolar minor salivary glands. Pleomorphic adenoma is a benign tumor. It is considered the most common salivary gland tumor representing 45-75% of all salivary gland neoplasms.[1,2] It is most common in 3rd to 6th decades.[3] The palate is considered as the most common intraoral site, followed by the upper lip and cheek. It is presentation is usually with mobile, firm, painless mass of slow progression.[8] In children, only five cases were reported as buccal salivary glands pleomorphic adenoma.[4-8] Most of the reported cases are elder than the case we are presenting here at the age of presentation. The relevant studies were discussed.

CASE REPORT

An 11 years old boy with no clinically significant past history presented with 4 months history of right cheek swelling incidentally discovered by his mother. It was increasing gradually in size with no associated symptoms apart from mass effect disturbing the chewing process. On presentation the mass was in right buccal area, submucosally seated, firm, mobile, rounded, 2×2 cm, non-tender, with normal non-adherent covering skin and mucosa, and no sinus or fistula. No palpable neck masses or lymph nodes.

Workup yielded a vaguely delineated non-enhancing mass in the right buccal region about 2×2 cm without adjacent structure involvement with no cervical lymphadenopathy on the Computed Tomography of neck as shown in Figure 1. Fine needle aspirate cytology (FNAC) was suggestive of pleomorphic adenoma with no evidence of malignancy.

Patient then taken to operation theatre, mass excised transorally with 1-2 millimeter safe-margin under general anesthesia. Histopathologically there was a salivary gland parenchyma which consist of epithelial and stroma mesesnchyma as seen in Figure 2, confirming the FNAC findings, and the diagnosis of pediatric buccal pleomorphic adenoma was established. Thereafter, three years follow up showed no recurrence.

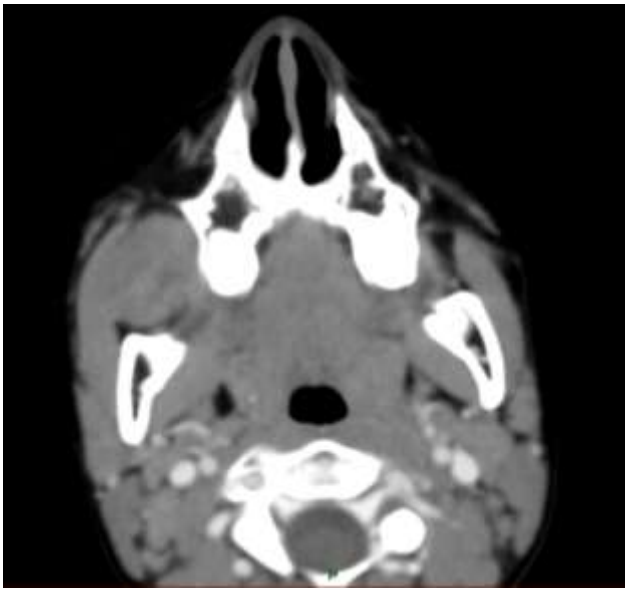

Figure 1: CT- scan of neck shows a 2×2 cm rounded, non-enhancing mass at right buccal part of oral cavity (arrow) with no cervical lymphadenopathy and intact adjacent structure.

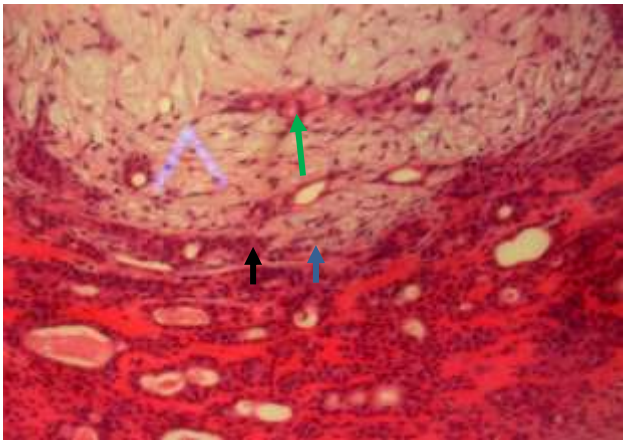

Figure 2: Histopathologic section showing the diagnostic findings of pleomorphic adenoma consisting of epithelial/myoepithelial areas (black arrow), myxochondroid areas of fibrous stroma (green arrow) and gland like tubular structure (blue arrow).

DISCUSSION

Minor salivary glands are distributed all through oral cavity, oropharynx, nasaopharynx, hypopharynx and nasal cavity. Pleomorphic adenoma is the most common tumours (50%) of the major and minor salivary gland, representing 3-10% of the neoplasm of the head and neck.[9,10] It is a benign tumour. In adults, oral cavity minor salivary gland pleomorphic adenomas are commonly seen in the palate in up to 69%, followed by the upper lip (10.1%) and buccal area (5.5%).[11,12] Other rare sites include the throat (2.5%), retromolar region (0.7%), floor of the mouth and the alveolar mucosa.[13] It is very rare in minor salivary glands of pediatric patients in general, especially in the buccal minor salivary glands as there are only five cases are reported up to date, 3 of them are older than our patient.[4-8] It usually present as a mobile slowly growing, painless, firm swelling that does not cause ulceration of the overlying mucosa.[8] The differential diagnosis of juvenile Pleomorphic adenoma includes abscess, foreign body reaction to substances like fish bone, fibroma, dermoid cyst, lipoma, rhabdo-mayosarcoma, mucoepidermoid carcinoma and adenoid cystic carcinoma. Although pleomorphic adenoma grossly appears encapsulated, it has a highly lobular architecture with microscopic pseudopods. Therefore simple enucleation is not recommended, as the recurrence rates approach 40%.[14,19] Kusum et al stated that the FNA-C is an ideal, fairly accurate preoperative procedure for the diagnosis of pleomorphic adenomas.[15] Certain diagnostic problems occur in differentiating pleomorphic adenomas from adenoid cystic carcinoma, monomorphic adenoma and mucoepidermoid carcinoma.[15] Malignant transformation of pleomorphic adenoma is rare and occurs most frequently in patients with long-standing tumors. The risk of malignant transformation in pleomorphic adenoma is 1.5% within the first 5 years of diagnosis but increases to 10% if observed for more than 15 years.[16] The most common symptom of salivary gland tumors in children is submucosal lump, with few cases showing ulceration or bleeding.[8] Although some Pleomorphic adenoma in pediatric tend to grow rapidly within short period but most of it enlarge in slowly as in adult age group.[8] In our case, the age of presentation is relatively very young among the reported cases and the mass was discovered incidentally by the patient's mother, relatively rapidly growing. FNAC was reliable in diagnosing buccal pleomorphic adenoma. A safe margin of 1-2 millimeter was sufficient as a safe margin as a preventative measure for recurrence.

CONCLUSION

Pleopmorphic adenoma should be considered one of the differential diagnosis of oral cavity in early age group although it is very rare in the buccal oral cavity subunit as in our case. Follow up with clinical assessment and radiological study is highly recommended especially in pediatric age group because of highly chance of recurrence. Reporting such case is very important in aiding studies' literature review on pediatric salivary gland tumors.

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