Pleomorphic Adenoma in Cheek: An Uncommon Finding

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Pleomorphic adenoma (PA), also called benign mixed tumor, is the most common benign salivary gland neoplasm. About 90% of these tumors occur in the parotid gland and 10% in the minor salivary glands. Review of literature, clinical features, pathology, radiological findings, and treatment of these tumors are discussed. We report a case of PA of minor salivary gland of cheek in 16-year-old female patient.

Keywords: Adenoma, Salivary glands, Tumor

INTRODUCTION

Pleomorphic adenomas (PAs) are benign salivary gland tumors that represent about 3-10% of the neoplasm of the head and neck region.1,2 PA is the most common benign neoplasm of the salivary glands. The palate is considered as the most common intraoral site (42.8-68.8%), followed by the upper lip (10.1%) and cheek (5.5%).3 Other rare sites include the throat (2.5%), retromolar region (0.7%), floor of the mouth and the alveolar mucosa.4 PA usually presents as a mobile slowly growing, painless firm swelling that does not cause ulceration of the overlying mucosa.5 PA consists of cells with epithelial and mesenchymal differentiation (mixed tumor). The highly variable morphology of this neoplasm is the result of interplay between these elements. Now it is widely accepted that both epithelial and mesenchymal (myxoid, hyaline, chondroid, osseous) elements often arise from same cell clone, which may be a myoepithelial or ductal reserve cell. There is no difference in behavior of this neoplasm based on proportion of various elements.6 Variants of PA include PA with lipomatous change,7 myxoliopmatous PA, PA with squamous differentiation and benign metastasizing mixed tumor.8

The mucosa of the cheek is an uncommon site of occurrence for intraoral PA9 and most of these cases have been reported in adults.10,11 In children, very few cases have been reported to date. Here, we report a case of PA in a 16-year-old girl. The relevant studies have been discussed.

CASE REPORT

A 16-year-old girl presented with a slowly growing painless swelling in the left cheek of 4 years duration. Clinical examination (Figure 1) revealed a 3 × 2.5 cm, firm, and mobile mass in the left cheek. There was no history of trauma, fevers, disturbance of salivation, or oral surgeries. The laboratory tests were unremarkable. Conventional panoramic view did not reveal any calcific or other abnormality within the swelling.

Further diagnostic aspiration was done to estimate the vascular presence within the mobile mass, which was negative. The differential diagnosis was buccal space abscess, dermoid cyst, foreign body reaction, fibroma, lipoma, neurofibroma, rhabdomyosarcoma, mucoepidermoid carcinoma, and adenoid cystic carcinoma. The possibility of buccal space abscess was ruled out due to absence of any sign of inflammation. The lack of ulceration of the buccal mucosa, pain, paresthesia or invasion of the surrounding tissue rules out the possibility of any malignant transformation.

Figure 1: Clinical intra oral view with visible swelling over left cheek mucosa, overlying mucosa with no abnormality in color or texture noted

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In account of the small size and mobile nature of the lesion, the mass was excised with safety margins under local anesthesia, by placing incision over the overlying cheek mucosa. Grossly, the lesion was in the form of an ovoid well-demarcated, encapsulated, gray-white partly myxoid, partly rubbery mass, and with solid cut surface (Figure 2). On histopathological examination, a well-circumscribed growth was seen. The neoplastic proliferation had biphasic populations of epithelial and mesenchymal cells. The former was composed of glandular structures lined by round, oval cells having large hyperchromatic nuclei, pink cytoplasm, and myoepithelial basal cell layer. The stroma was myxoid, hyaline and chondroid. No mitotic figures or necrosis were seen (Figure 3). Thus, diagnosis of PA of minor salivary gland was made.

Postoperative period was uneventful. The patient was followed-up over a period of 1 year and no recurrence was observed.

DISCUSSION

PA occurs more frequently in women than in men and is most common from the fourth to sixth decades with a mean age of 43-46 years. Salivary gland tumors are rare in children and when they do arise, they preferentially affect major salivary glands, but minor salivary gland tumors have also been reported.

The clinicopathologic features of the case reported here concurs with previous studies. Yamamoto et al. reported a 9-year-old Japanese girl with PA of the cheek mucosa. Cohen and Kronenberg reported two more cases of juvenile PAs of the cheek (girls, age <18 years). Dhanuthai et al. reported the first case of palatal PA in a 13-year-old child. In children, Jorge et al. reported five cases of intraoral PA in patients under 18 years of age in two Brazilian institutions. Four patients were females and one was male; two cases affected the palate, two the upper lip and one the tongue. The cases were treated by local excision and long follow-up showed no recurrences were observed. They concluded that intraoral PA seems to have similar biological characteristics as in adults, with low recurrence rates after surgical resection. The surgical treatment for the PA in both juvenile and adult patients is principally the same and includes complete wide surgical excision with good safety margins. Inadequate resection or rupture of the capsule or tumors spillage during excision can lead to local recurrence as these tumors often have microscopic interruptions in the capsule.

In adults, van Heerden and Raubenheimer examined the clinicopathologic features of the oral salivary gland neoplasms. Seventy cases were diagnosed during 8 years period. PA was the most common entity that accounted for 48% of all tumors, but none of these lesions affected the cheek mucosa. Polymorphous low-grade adenocarcinoma comprised 15.7% of the tumors.

In 2002, Jansisyanont et al. reported a total of 80 minor salivary gland tumors in 49 female patients and 31 male patients and the ratio ranged from 1.2:1 to 1.9:1. In 2009, Oliveira et al. reported in their review of 599 salivary gland tumors, 78.3% cases were benign. According to Vicente et al. in 2008, most studies of salivary neoplasms include both the major and the minor salivary glands, and few articles focus only on minor salivary gland tumors.

The differential diagnosis of the juvenile PA of the cheek includes buccal space abscess, dermoid cyst, foreign body reaction, fibroma, lipoma, neurofibroma, rhabdomyosarcoma, mucoepidermoid carcinoma, adenoid cystic carcinoma, polymorphous low-grade adenocarcinoma and carcinoma ex-PA. The possibility of buccal space abscess was ruled out due to absence of signs of inflammation. The solid nature of the lesion coupled with
the lack of tissue representing the three germ layers rule out the possibility of mature cystic teratoma (dermoid cyst). The lack of ulceration of the buccal mucosa, pain, paresthesia or invasion of the surrounding tissue rules out the possibility of malignant transformation. Carcinoma ex PA is characterized by the presence of malignant epithelium (salivary duct carcinoma, undifferentiated carcinoma, adenocarcinoma not otherwise specified, terminal duct carcinoma or myoepithelial carcinoma) with benign stroma. Carcinoma ex PA is extensively infiltrative tumor with perineurial invasion, frequent mitotic figures, marked nuclear atypia. Adenoid cystic carcinoma usually shows cribriform, solid or tubular pattern similar to cylindromas of skin. It is composed of small bland myoepithelial cells with scant cytoplasm and dark compact angular nuclei that surround pseudoglandular spaces with PAS+ excess basement membrane material and mucin. Peripheral perineural invasion and small true glandular lumina are sometimes seen, but no squamous differentiation; or extensive necrosis is usually absent. Adenoid cystic carcinoma has high proliferative index, high p53 immuno-reactivity, intense staining for BCL-2 but negative reactivity for glial fibrillary acidic protein. In contrast, PA is not invasive, show no perineural invasion; has squamous metaplasia and mesenchyme-like areas. Characteristically, PA has strong glial fibrillary acidic protein in the myxochondromatous areas. The polymorphous low-grade adenocarcinoma is usually nonencapsulated tumorstroma by basement membrane. The polymorphous low-grade adenocarcinoma is usually nonencapsulated tumorstroma by basement membrane. The polymorphous low-grade adenocarcinoma is usually nonencapsulated tumorstroma by basement membrane. The polymorphous low-grade adenocarcinoma is usually nonencapsulated tumorstroma by basement membrane.

Conclusively, juvenile PA is a rare neoplasm and therefore its diagnosis requires a high index of suspicion. Complete wide surgical excision is the treatment of choice. Recurrence after many years of surgical excision as well as malignant transformation should be a concern and therefore long-term follow-up is necessary.

**TAKE HOME MESSAGES**

1. Juvenile PA of the cheek is a rare neoplasm and therefore its diagnosis requires a high index of suspicion.
2. Majority of tumors occurring in minor salivary gland are malignant. The salivary glands tumours may present with a diverse range of lesions presenting a challenge to even the most experienced surgeon and pathologist.

3. PA should be considered in the differential diagnosis of cheek masses in youngsters. Wide local excision is to be recommended as the treatment of choice. A close follow-up is necessary postoperatively.

**REFERENCES**


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