

CASE REPORT



Polymorphous low-grade adenocarcinoma of parotid gland – A case report and review

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Abstract

Polymorphous low-grade adenocarcinoma (PLGA) is a low aggressive tumor commonly occurring in minor salivary glands. Its occurrence is rarely documented in major salivary glands. Diagnosis of PLGA is challenging both clinically and histologically due to its slow-growing behavior. Here, we report a case of PLGA arising from the right parotid gland in a 48-year-old female patient.

Introduction

Polymorphous low-grade adenocarcinoma (PLGA) is a slow-growing malignant tumor of low aggressiveness.^[1] It was first described in 1983 by Batsakis *et al.* as “terminal duct carcinoma” and by Freedman as “lobular carcinoma.”^[1-8] It is the second most commonly diagnosed salivary gland neoplasm, with 60% of cases located in palate. Mean age of occurrence is 59 years with a female predilection of 2:1. Even though it is predominantly seen in minor salivary glands, 32 cases of PLGA were reported in literature arising either as *de novo* or as preexisting neoplasm developing as pleomorphic adenoma (carcinoma ex pleomorphic adenoma).^[6] We report a case of PLGA developing in the right parotid gland, surgically treated by superficial parotidectomy.

Case Report

A 48-year-old female patient was referred to the Oral Medicine and Radiology outpatient clinic of the College of Dental Sciences, Davangere, Karnataka, by her physician for an asymptomatic mass on the right preauricular region which was noticed 3 years back. On eliciting the history, swelling was gradual in onset for 3 years with no history of pain, oral dryness, and difficulty

in mastication. The patient is a known case of hypertension (15 years) and diabetes (2 years).

On extraoral examination, mild asymmetry was noted on the right side of lower face [Figure 1]. A solitary diffuse mass of 2 cm × 3.5 cm seen in the right preauricular region superiorly in line with tragus and inferiorly 1 cm below the ramus of mandible. The postero extension was approximately 3.5 cm from tragus of the right ear. No evidence of secondary changes, bleeding, pus discharge, and sinus opening were noted. Mass was not prominent on clenching the teeth. Furthermore, there was no associated lymphadenopathy or facial nerve deficiency. On palpation, no rise in temperature and the swelling was non-tender, firm in consistency, and non-mobile with ill-defined borders. Swelling was non-compressible, non-reducible, and non-pulsatile. The right submandibular lymph node of size 1 cm was palpable, mobile, and non-tender.

Intraoral hard tissue examination revealed that Grade I mobility w.r.t 11, 12, 13, and 38 was migrated mesially as 37 was clinically missing and grossly decayed 18 and 44. On examination, no noticeable soft tissue changes were visible. The salivary flow and viscosity were normal. Correlating the history and clinical findings, we arrived at a provisional diagnosis of benign adenoma of parotid gland. Pleomorphic adenoma, masseteric hypertrophy, and sialosis were the differential set of diagnosis.

Ultrasonography (USG) report stated the evidence of well-defined isoechoic lesion measuring 2 cm × 1.9 cm × 1.4 cm in size in the right parotid gland with regular margins and no vascularity on color Doppler. No evidence of calcification/necrosis was found within the lesion. Region of parotid gland and submandibular gland appears normal. Other vascular structures are normal. Contralateral parotid appears normal. USG impression was suggestive of pleomorphic adenoma in the right parotid gland.

Hematological and biochemical reports were normal. Surgical removal of the right superficial gland was performed under general anesthesia. A soft tissue specimen was sent for histopathological analysis. Microscopic features of hematoxylin and eosin section of tumor proper revealed a well-circumscribed cellular lesion tissue. The cells showed indistinct cell outline with hyperchromatic nuclei. Nuclei showed polygonal with irregular chromatin clumping. In between these islands, eosinophilic aggregates of amorphous material were found which were irregular in few areas and globular in focal areas. This made it appear as swiss cheese pattern, but these were not lined by duct-like cells as in adenoid cystic carcinoma (ACC) [Figure 2]. There was sheet-like arrangement in few sites and also evident mitotic figures were seen. A lymph node attached to the lesion showed tumor cell infiltration. Histopathological features were suggestive of PLGA.

The patient was recalled after 6 months for follow-up. Operated site was healed with no secondary changes and recurrence.

Discussion

PLGA is a definite malignancy of salivary gland with low aggressive behavior.^[2,6] This tumor is commonly seen with minor salivary glands having documented few cases in major salivary glands, either arising *de novo* or as malignant component of carcinoma ex pleomorphic adenoma.^[6]

Clinically, tumor appears as a slow-growing painless swelling with female predilection.^[9] In a case series of PLGA on 64 cases by Castle *et al.*, the definitive presentation was that of an asymptomatic mass. Of 64 cases, 13 presented with mass associated with pain, bleeding, or ulceration. Nagao *et al.* stated that PLGAs of minor and major salivary glands have similar clinicopathological characteristics. Mark *et al.* stated that few differences exist with PLGA present in minor salivary glands compared to parotid gland. Even though these differences in chromosomes do not demand any contrasts in biological behavior due to its anatomical location, they support the possible origin of PLGA of parotid gland in pleomorphic adenoma. It is interesting to note that differences with biological behavior among the cases arising *de novo* and those that develop from pleomorphic adenoma have not been mentioned in literature.^[6,4] Kemp *et al.* described 22 cases which were diagnosed as terminal duct adenocarcinoma originating in the parotid gland.^[3] In the present case, an asymptomatic mass was seen on the right parotid gland of 3 years duration in a middle-aged female.

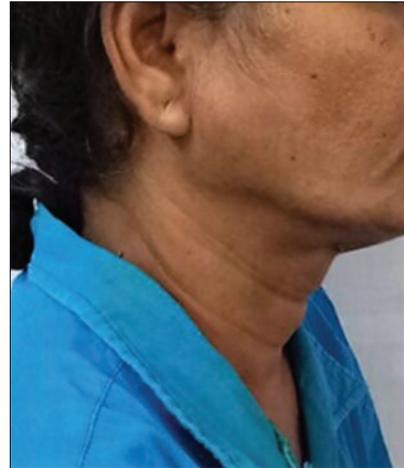


Figure 1: Mass on the right preauricular region

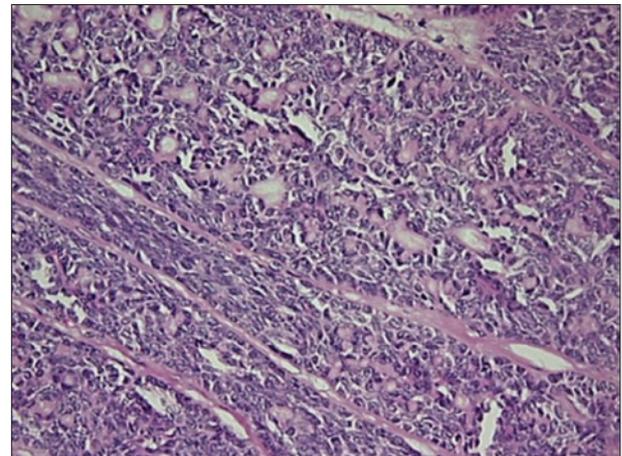


Figure 2: Tumor proper revealing Swiss cheese pattern at high resolution (×20)

PLGA is usually considered harmless in spite of its infiltrative growth pattern locally along with perineural and perivascular characteristics. It has less nodal metastasis and rare occurrence of distant spread.^[6] In our case, tumor did not show any regional and distant metastasis.

Histopathologically, architectural pattern of ACC and PLGA resembles to each other. Few differences like cells in ACC tend to be smaller with hyperchromatic nuclei and coarser chromatin, numerous mitoses. Differentiating these two entities are crucial since their prognosis and treatment are significantly different. Various architectural patterns could be seen in different areas microscopically (glandular, trabecular, tubular, cribriform, “Indian file,” and solid).^[2,10]

The mainstay of treatment remains surgical excision, irrespective of its location. In case of cervical lymphadenopathy, neck dissection should be considered. Based on the presence of perineural invasion and vascular permeation of tumor cells, post-operative radio or chemotherapy can be considered.^[6] In the present case, surgical removal of superficial parotid gland

was performed. Post-surgical follow-up was done after 6 months with no recurrence.

Conclusion

It is essential to consider the occurrence of PLGA in the major salivary glands *de novo* or as a part of a pleomorphic adenoma that does not show clinical indication of malignancy. Examining the surgical specimen is crucial as the clinical course of the PLGA is usually harmless mimicking a benign neoplasm. Once PLGA is recognized, it is important to inform the predominant histologic pattern, the occurrence of perineural invasion and vascular permeation, to determine if these criteria may be of assistance to identify its biological behavior.^[6]

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