



Polymorphous low grade Adenocarcinoma of Tongue Case Report

SACHIN SHARAD GANDHI, NILANJAN PRASHANT BHOWMICK, NEEHARIKA GUNTURU, SUJIT JOSHI

Department of ENT, Deenanath Mangeshkar hospital, Pune

Abstract

Polymorphous low-grade adenocarcinoma (PLGA) is a low grade malignant neoplasm that occurs almost exclusively in the minor salivary glands, primarily those in the palate. We report a case of PLGA that arose at the base of the tongue. The tumor was excised by diode laser with adjuvant radiotherapy. This case shows that PLGA, which has a variable morphologic appearance, can occur at rare sites like base of tongue other than the salivary glands of palate.

Key words: Polymorphous low-grade adenocarcinoma (PLGA), Minor salivary gland, base of tongue

Introduction

Polymorphous low-grade adenocarcinoma (PLGA) is a malignant neoplasm that occurs almost exclusively in the minor salivary glands, especially those in the palate(1). This tumor is of particular interest because of its low degree of aggressiveness, its slow growth, and its conspicuous architectural polymorphism--features that for many years have complicated its recognition as a tumor type distinct from other adenocarcinomas(1). We report a case of PLGA that arose in an unusual location--the base of the tongue without any metastasis elsewhere in the body.

Case report

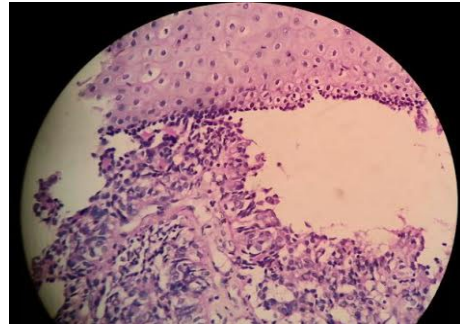
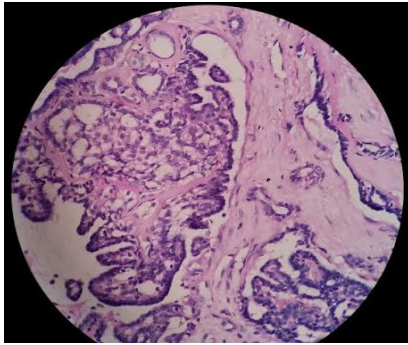
A 73-year-old woman presented to our institution with complaints of dysphagia & change in voice(hot potato voice) that had developed within one year. She underwent biopsy of the same lesion one year back with histopathological study revealing haemangioma & she had CT scan

which showed a 3.4/2.6/2.5 cm mass occupying posterior 1/3rd of tongue. A biopsy was repeated few weeks later which showed glomangioma of tongue (tumour was of pericytic origin).

Immunohistochemistry showed positivity for SMA & negativity for CD 34 with KiB-1 proliferative index 03%

She had no cervical lymphadenopathy at that time. Videolaryngostroboscopic examination showed a mass over base of tongue which bled to touch. Patient underwent microlaryngoscopic surgery with diode laser debulking & multiple chunks of tissues were taken & again sent for histopathological study.

Histopathological examination revealed hyperplastic stratified squamous epithelium arranged in trabecular pattern & small duct like structures lined by uniform round cells having bland nuclei & scanty eosinophilic cytoplasm. In few areas papillary arrangement, mitotic activity not significant.



Immuno histochemistry result showed strong positivity for CK,SMA & weak positivity for S-100 protein and negativity for CD31,CD34 & EMA.

Final diagnosis was Polymorphous low grade adenocarcinoma of minor salivary gland origin.

Patient underwent radiation therapy . Follow up endoscopy done after 6 months showed no recurrence.

DISCUSSION:

Polymorphous low-grade adenocarcinoma (PLGA) is a malignant neoplasm that occurs almost exclusively in the minor salivary glands, especially those in the palate.(1). The most common sites of tumors of the minor salivary glands are, in order, the palate, the oral mucosa, lip, retromolar trigone & cheek(2). In very rare cases it can occur in the tongue. This distribution probably reflects the higher number of glands in this area. Approximately 80% of minor salivary gland tumors are malignant; adenocarcinomas account for 25% of these malignancies.(3)

Cancer of the tongue is the second most common type of cancer arising in the upper digestive tract, with approximately 40% arising from the tongue base (4). In addition, lesions located in the base of tongue are more likely to be malignant (85%) than those located in the oral tongue (20%)

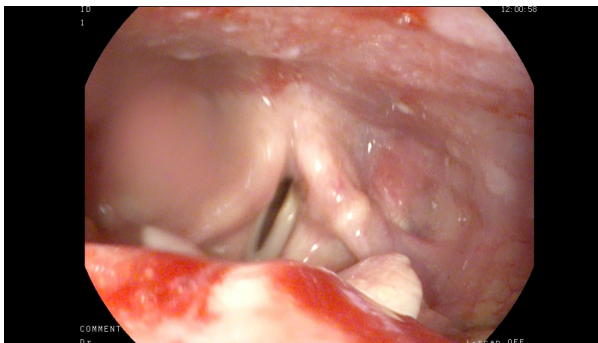
(5). Most tongue cancers (more than 95%) are histologically characterized as squamous cell carcinoma; adenocarcinoma of the tongue, often arising from minor salivary glands, occurs in fewer than 2% of all tongue malignancies(4).

Minor salivary gland malignancies are themselves rare, representing only 5%–10% of all malignancies in the head-and-neck region. Many histologic subtypes of minor salivary gland tumours in the oropharynx have been documented, mucoepidermoid and adenoid cystic carcinoma being the most common, followed by the more rare subtypes such as adenocarcinoma and acinic cell carcinoma (4,5).

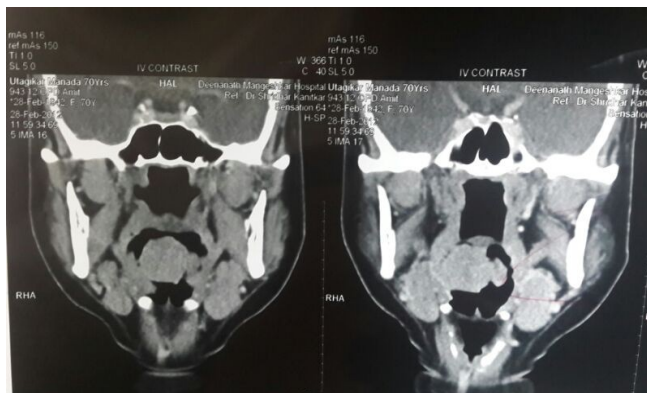
Several risk factors have been identified with respect to the development of oropharyngeal cancer, Which include smoking, alcohol consumption(6) and a diet deficient in fruits and vegetable. However, infection with the human papilloma virus has recently emerged as a major contributor to the development of squamous cell oropharyngeal cancer, including base-of-tongue cancers(7), particularly in younger populations(8). The same association has not been made with adenocarcinoma of the oropharynx.

The prognosis for adenocarcinoma is more favourable than that for squamous cell carcinoma(9,10) and reported 3-, 5-, and 10-year survival rates are approximately 50%(11).

However, survival varies greatly depending on tumour stage, grade, and treatment modalities used.



Video Stroboscopic picture showing mass at base of the tongue which bleeds on touch



CT Scan pictures showing lesion at base of tongue

PLGA has a male preponderance (2:1), commonly manifesting during the seventh decade of life. PLGA at base of tongue mostly cause symptoms of discomfort while swallowing & occasional bleeding(12).Change in voice associated with nasal obstruction & otalgia may also occur. PLGA had previously been referred to as terminal duct carcinoma in view of its probable origin in the ductal system of the salivary glands.(13) The term polymorphous low-grade adenocarcinoma was first used in 1984 by Evans and Batsakis to describe a tumor of the salivary glands that had as its primary histologic characteristic a variety of architectural patterns associated with cytologic uniformity(14).Video laryngostroboscopy will help identifying the tumour in terms of its extent,size & its gross appearance. But however CT Scan helps in better understanding of extent/local spread/deep invasion & about nodal metastasis. The recommended treatment for PLGA is Wide local excision with adjuvant radiotherapy may be used in cases of local recurrence and/or lymph node metastasis.(15,16,17)

References

- 1) Vincent SD, Hammond HL, Finkelstein MW. Clinical and therapeutic features of polymorphous low-grade adenocarcinoma. *Oral Surg Oral Med Oral Pathol* 1994;77:41-7.
- 2)Waldron CA, el-Mofty SK, Gnepp DR. Tumors of the intraoral minor salivary glands: A demographic and histologic study of 426 cases. *Oral Surg Oral Med Oral Pathol* 1988;66:323-33.
- 3) Spiro RH, Koss LG, Hadju SI, Strong EW. Tumors of minor salivary origin. A clinicopathologic study of 492 cases. *Cancer* 1973;31:117-29.
- 4) Attner P, Du J, Näsman A, et al. Human papillomavirus and survival in patients with base of tongue cancer. *Int J Cancer*. 2011;128:2892–7. doi: 10.1002/ijc.25625
- 5) Warnakulasuriya S. Causes of oral cancer—an appraisal of controversies. *Br Dent J*. 2009;207:471–5. doi: 10.1038/sj.bdj.2009.1009.
- 6) Biller HF, Lawson W, Baek SM. Total glossectomy. A technique of reconstruction eliminating laryngectomy. *Arch Otolaryngol*. 1983;109:69–73. doi: 10.1001/archotol.1983.00800160003001.
- 7) Dupont JB, Guillaumondegui OM, Jesse RH. Surgical treatment of advanced carcinomas of the base of the tongue. *Am J Surg*. 1978;136:501–3. doi: 10.1016/0002-9610(78)90269-6
- 8) Frazell EL. Observations on the management of salivary gland tumors. *CA Cancer J Clin*. 1968;18:235–40. doi: 10.3322/canjclin.18.4.235.
- 9) Evans M, Powell NG. The changing aetiology of head and neck cancer: the role of human papillomavirus. *Clin Oncol (R Coll Radiol)* 2010;22:538–46. doi: 10.1016/j.clon.2010.05.024

- 10) Evans HL, Batsakis JG. Polymorphous low-grade adenocarcinoma of minor salivary glands. A study of 14 cases of a distinctive neoplasm. *Cancer* 1984;53:935-42.
- 11) Batsakis JG, Pinkston GR, Luna MA, et al. Adenocarcinomas of the oral cavity: A clinicopathological study of terminal duct carcinomas. *J Laryngol Otol* 1983;97:825-35.
- 12) Regezi JA, Zarbo RJ, Stewart JC, Courtney RM. Polymorphous low-grade adenocarcinoma of minor salivary gland. A comparative histologic and immunohistochemical study. *Oral Surg Oral Med Oral Pathol* 1991;71:469-75.
- 13) Kennedy KS, Healy KM, Taylor RE, Strom CG. Polymorphous low-grade adenocarcinoma of the tongue. *Laryngoscope* 1987;97:533-6
- 14) Colmenero CM, Patron M, Burgueno M, Sierra I. Polymorphous low-grade adenocarcinoma of the oral cavity: A report of 14 cases. *J Oral Maxillofac Surg* 1992;50:595-600.
- 15) Wenig BM. *Atlas of Head and Neck Pathology*. Philadelphia: W.B. Saunders, 1993
- 16) Muir C, Weiland L. Upper aerodigestive tract cancers. *Cancer*. 1995;75(suppl 1):147-53. doi: 10.1002/1097-0142(19950101)75:1+<147::AID-CNCR2820751304>3.0.CO;2-U
- 17) Goldblatt LI, Ellis GL. Salivary gland tumors of the tongue. Analysis of 55 new cases and review of the literature. *Cancer*. 1987;60:74-81. doi: 10.1002/1097-0142(19870701)60:1<74::AID-CNCR2820600113>3.0.CO;2-