

Case Report

A Polymorphous Low-Grade Adenocarcinoma of the Tongue

Aykut BOZAN^{1,a}, Ayşe POLAT², Denizhan DİZDAR³, Hayrettin Cengiz ALPAY³

¹Özel Medical Park Tarsus Hastanesi, Kulak Burun Boğaz Kliniği, Mersin, Türkiye

²Mersin Patoloji Laboratuvarı, Patoloji, Mersin, Türkiye

³İstanbul Kemerburgaz Üniversitesi Tıp Fakültesi, Kulak Burun Boğaz Kliniği, İstanbul, Türkiye

ABSTRACT

A polymorphous low-grade adenocarcinoma arising from a minor salivary gland is a rare malignancy of the aerodigestive system. Most such adenocarcinomas develop in the hard palate. Surgical excision constitutes adequate treatment. A 41-year-old male presented with a mass 1 × 0.7 cm in size at the left side of the tongue near the papilla circumvallata and underwent total resection and primary closure. The pathological diagnosis was a low-grade polymorphous adenocarcinoma and the surgical margins were negative. We present the case and review the management of low-grade polymorphous adenocarcinomas of (usually) the hard palate as described in the literature; such tumours are rarely encountered.

Keywords: Minor Salivary Gland, Polymorphous Low-Grade Adenocarcinoma, Tongue.

ÖZET

Dilin Polimorfoz Düşük Dereceli Adenokarsinom

Minör tükürük bezinden kaynaklanan Polimorfoz düşük dereceli adenokarsinom üst aerodijestif yolun nadir görülen malign bir tümördür. Bu tür Adenokarsinomların çoğu sert damakta gelişirler. Tedavide cerrahi eksizyon yeterlidir. Yazımızda total rezeke edilen ve primer olarak kapatılan dil sol tarafta papilla sirkumvallata komşuluğunda 1x0.7 cm çaplı kitlesi olan 41 yaşında erkek hasta sunulmuştur. Patolojik tanı düşük dereceli polimorfoz adenokarsinom ve cerrahi sınırlar negatiftir. Bu makalede literatürde genellikle sert damakta tanımlanan ve seyrek olarak karşılaşılan düşük dereceli polimorfoz adenokarsinom olgusu ve tedavi yönetimi tartışıldı.

Anahtar Sözcükler: Minor Tükürük Bezleri, Polimorfoz Düşük Dereceli Adenokarsinom, Dil.

A polymorphous low-grade adenocarcinoma (PLGA) is the second most common malignancy of the minor salivary glands (after a mucoepidermoid carcinoma); the hard palate is the most frequently involved head-and-neck site (1). PLGAs affect patients of all ages, from 16 to 94 years reported, with a mean age of 59 years, and exhibit a female predilection. The typical presentation is an indolent submucosal mass, which may occasionally be painful or even ulcerated (2). The most common site of a PLGA is the palate, followed by the buccal mucosa, the upper lip, the retromolar triangle, and the tongue (3). We present a (rare) case of tongue PLGA in which total surgical excision was performed.

CASE REPORT

A 41-year-old male presented to our otolaryngological department with a mass at the left side of the tongue. He complained of no symptom other than the mass. On rigid laryngoscopy, a tumour 1 cm in diameter with an intact overlying mucosa was evident at the left side of the tongue near the papilla circumvallata (Figure 1). Neither neck palpation nor ultrasonic neck evaluation revealed any cervical lymphadenopathy. Computed tomography of the head-and-neck was performed after

administration of intravenous contrast, and revealed that the tongue mass was both superficial and small. The patient underwent surgical excision under general anaesthesia both as treatment and to allow pathological

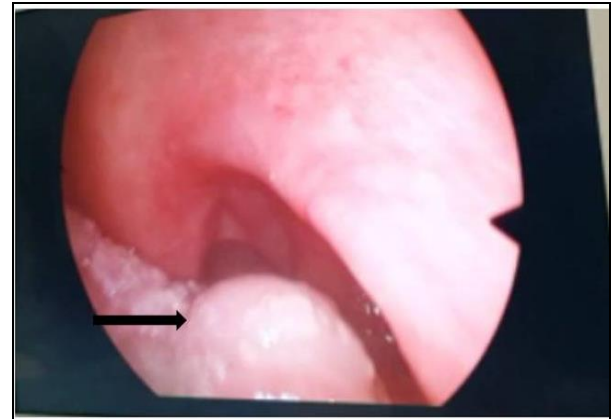


Figure 1. A tumor 1 cm in size with intact overlying mucosa, was found at the left side of the tongue near the papilla circumvallata (as shown by black arrow).

diagnosis. The mass was hard and the overlying mucosa was clearly distinguishable from the surrounding normal mucosa. After resection, we performed primary wound closure. The pathological diagnosis was a PLGA and the resection margins were negative.

^aYazışma Adresi: Aykut BOZAN, Özel Medical Park Tarsus Hastanesi, Kulak Burun Boğaz Kliniği, Mersin, Türkiye

Tel: 0324 241 4141

Geliş Tarihi/Received: 29.06.2016

e-mail: aykbzn@gmail.com

Kabul Tarihi/Accepted: 04.08.2017

The lesion was encapsulated; the cells formed tubular, cribriform, and trabecular patterns (Figure 2).

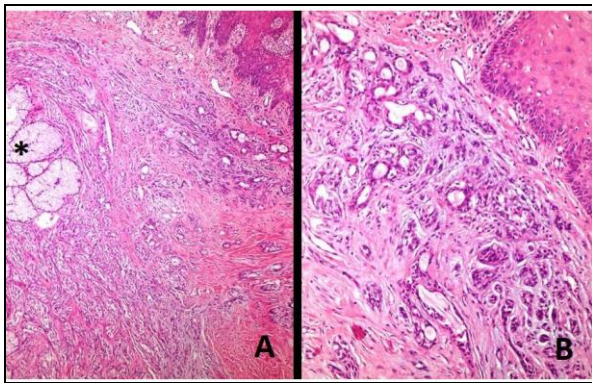


Figure 2. A and B. Polymorphous low grade adenocarcinoma: the invasive tumor was near the mucous salivary glands (*) and Tumour cells arranged in tubular, cribriform, solid, trabecular pattern.

The patient did not complain of postoperative dyspnea or pulmonary aspiration. He has received close follow-up, and no recurrence has been observed during the first postoperative year.

DISCUSSION

A PLGA is a low-grade malignancy, first described by Evans and Batsakis (4) as a malignant tumour arising in the minor salivary glands; the condition was previously considered to be a lobular carcinoma or a terminal duct carcinoma. PLGA constitutes around 19–26% of the malignant tumours of the minor salivary glands

(5), developing most commonly in the hard palate, principally at the base of the tongue (6). We here describe a PLGA in a rare location.

Histopathologically, a PLGA is characterised by cytologically uniform, anonymous round cells arranged in several architectural patterns (hence the polymorphous descriptor). The cell growth patterns include solid, trabecular, tubular, cribriform, microcystic, and papillary presentations. In the present case, tubular, trabecular, and cribriform growth patterns were evident among cells of the periphery.

Regional and distant metastases develop in 5–15% (7) and 0.6–7.5% (8) of patients, respectively. Therefore, elective neck dissection is not recommended when treating early T- stage tumours (9). The prognosis is relatively good and wide surgical resection is the recommended primary treatment. Any role for radiotherapy remains controversial. However, postoperative radiotherapy may be considered if the surgical margins are positive (8). We found no perioperative cervical lymphadenopathy; we thus considered that the condition was benign and did not perform neck dissection. We did not schedule radiotherapy because the surgical margins were negative upon postoperative histopathological examination.

Long-term follow-up is essential to prevent transformation of the condition into a high-grade malignancy, and to detect recurrence (10). We have followed-up the patient closely; there has been no recurrence to date, 1 year postoperatively.

REFERENCES

1. Olusanya AA, Kadiri OA, Akinmoladun VI, Adeyemi BF. polymorphous low grade adenocarcinoma: Literature review and report of lower lip lesion with suspected lung metastasis. *J Maxillofac Oral Surg* 2011; 10: 60-3.
2. Paleri V, Robinson M, Bradley P. Polymorphous low grade adenocarcinoma of the head and neck. *Curr Opin Otolaryngol Head Neck Surg* 2008;16: 163-9.
3. Barasoain AM, Martin VFJ, De La Fuente GE, Santamaria SJ, Pampin-Franco A, Lopez-Esteban JL, et al. Polymorphous low-grade adenocarcinoma in the upper lip: a well-described but infrequently recognized tumour. *Dermat Online J* 2013; 19: 192-5.
4. HL Evans, JG Batsakis. Polymorphous low-grade adenocarcinoma of minor salivary glands. A study of 14 cases of a distinctive neoplasm. *Cancer* 1984; 53: 935-42.

5. Takubo K, Doi R, Kidani K, Nakabayashi M, Ohtake F, et al. Polymorphous low grade adenocarcinoma arising at the retromolar region: A rare case of high grade malignancy. *Yonago Acta Medica* 2007; 50: 17-22.
6. De Diego JI, Bernaldez R, Prim MP, Hardisson D. Polymorphous low grade adenocarcinoma of the tongue, *J Laryngol Otolol* 1996; 110: 700-3.
7. Pogodzinski MS, Sabri AN, LewisJE, Olsen KD. Retrospective study and review of polymorphous low-grade adenocarcinoma. *Laryngoscope* 2006; 119: 2145-9.
8. Castle JT, Thompso LD, Frommelt RA, Wenig BM, Kessler HP. Polymorphous low grade adenocarcinoma: a clinicopathologic study of 164 cases. *Cancer* 1999; 86: 207-19.
9. Paleri V, Robinson M, Bradley P. Polymorphous low grade adenocarcinoma of the head and neck, *Curr Opin Otolaryngol Head Neck Surg* 2008; 16: 163-9.
10. Fife TA, Smith B, Sullivan CA, Browne JD, Waltonen JD. Polymorphous low-grade adenocarcinoma: a 17 patient case series. *Am J Otolaryngol* 2013; 34: 445-8.