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Case Report

Polymorphous Low-grade Adenocarcinoma of the Larynx: A Rare Case Report

Amir Hosein Jafarian MD[1], Kamran Khazaeni MD[2], Amin Rahpeyma DDS[3], Saeedeh Khajehahmadi DDS•[4]

Abstract

Laryngeal polymorphous low-grade adenocarcinoma is a rare malignant tumor with a predilection for intraoral sites. Polymorphous low-grade adenocarcinoma mostly occurs during the sixth to eighth decades of life with a female predilection. Although histopathologic evaluations enabled the definite diagnosis of this tumor, it might be mistaken with adenoid cystic carcinoma or mixed cellular tumor especially in situations when specimens with small sizes are available. We present a rare case of laryngeal polymorphous low-grade adenocarcinoma in a 55-year-old female patient.

Keywords: Immunohistochemistry, laryngeal tumor, polymorphous low-grade adenocarcinoma

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Introduction

Polymorphous low-grade adenocarcinoma (PLGA) is a malignant epithelial neoplasm which originates from reserve cells of salivary glands.[1] PLGA was introduced by the World Health Organization (WHO) as a new entity in 1990.[2] PLGA is almost exclusively a tumor of the minor salivary glands. Palate is the most common site for minor gland tumors. The next common site is the buccal mucosa. PLGA occurs predominantly during the sixth to eighth decades of life and has a 3 : 1 female to male ratio.[3]

Tumor cells can show different growth patterns such as solid pattern or cordal, ductal, or cribriform patterns. Thus the term "polymorphous" is applied to such malignancies. Adenoid cystic carcinoma has a mean proliferation index of 12.5 % while PLGA's proliferation index is less than 6.4 %.[4]

The overall prognosis of PLGA is good and local lymph node metastasis took place in less than 10 % of cases.[5] Metastasis to the lung is uncommon. The papillary type of this tumor has more aggressive behavior. [6]

We present a very rare case of PLGA in the larynx of a female patient and discuss its clinical and histopathologic features, as well as immunohistochemical findings and differential diagnoses.

Case Report

A 55-year-old woman with a 2 year history of hoarseness was referred to our clinic. The patient complained of progressive dysphagia for solid food. Axial computed tomography (CT) scan revealed a supraglottic mass passing the midline (Figure 1). Ultrasonographic evaluations showed no abnormalities in the liver, gallbladder, kidneys, and bladder. Also, no lymphadenopathies were present in the neck region. Laboratory and radiographic (chest X-ray) findings were normal. Only a slight elevation in ALT (SGPT) was reported. A flexible fiberoptic scope examination showed normal vocal cord movements. Direct laryngoscopy and mapping was done, and a biopsy was obtained from the lesion. Histopathologic examination of the specimen proposed PLGA as the definite diagnosis. Under general anesthesia, supracricoid laryngectomy with clear margins was performed. The gross specimen was consisted of a firm gray-white mass with 30 × 25 × 15 mm diameters (Figure 2). Microscopic examination showed that the tumor cells were arranged in varied patterns including tubular, trabecular, and papillary. The tumor cells were monomorphic in shape, round to polygonal with eosinophilic cytoplasm.

The stroma between the solid areas demonstrated hyalinization and involvement of laryngeal cartilage. Tumoral cells forming tubules and trabeculae were cuboidal or columnar with eosinophilic cytoplasm and round or oval nuclei. The lesion was diagnosed as infiltrative neoplasm without involvement of the overlying nasopharyngeal epithelium with clear margins (Figure 3A). laryngeal cartilage and perineural invasion were present. Immunohistochemical staining for CK7 was also positive (Figure 3B) while immunoreactivity for glial fibrillary acidic protein (GFAP), c-kit, and CK20 were negative. However, the proliferation rate -Ki67 staining of PLGA was measured as 6.2 % and the definite diagnosis was PLGA.

Discussion

PLGA of the salivary glands was initially introduced in 1983 by two scientific groups, Freedman and Lumerman, which worked independently.[7] They described twelve salivary gland tumors, called lobular carcinoma. One month later Batsakis, et al. used terminal duct carcinoma to describe a similar lesion.[8] Six months

Authors' affiliations: [1]Department of Pathology, Ghaem Hospital , Mashhad University of Medical Sciences, Mashhad, Iran, [2]Oral and Maxillofacial Diseases Research Center, Ghaem Hospital, Mashhad University of Medical Sciences, Mashhad, Iran, [3] Dental Research Center, School of Dentistry, Mashhad University of Medical Sciences, Mashhad, Iran, [4] Oral and Maxillofacial Diseases Research Center, School of Dentistry, Mashhad University of Medical Sciences, Mashhad, Iran.

•Corresponding author and reprints: Saeedeh Khajehahmadi DDS, Oral and Maxillofacial Diseases Research Center, Mashhad University of Medical Sciences, Vakilabad Blvd, P. O. Box: 91735-984, Mashhad, Iran. Tel: +98-511-8829501, Fax: +98-511-8829500,
E-mail: khajehahmadis@mums.ac.ir, saeedeh.ahmady@yahoo.com
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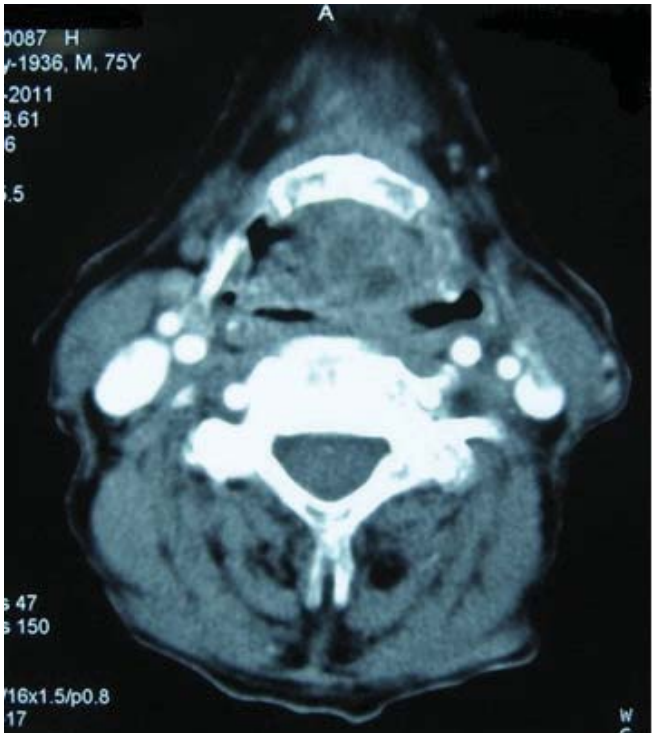


Figure 1. External view of neck, without any abnormal findings in clinical examination; Axial CT scan shows a supraglottic mass in the anterior wall of the left hypopharynx without any invasions to adjacent fat tissue that passed the midline.

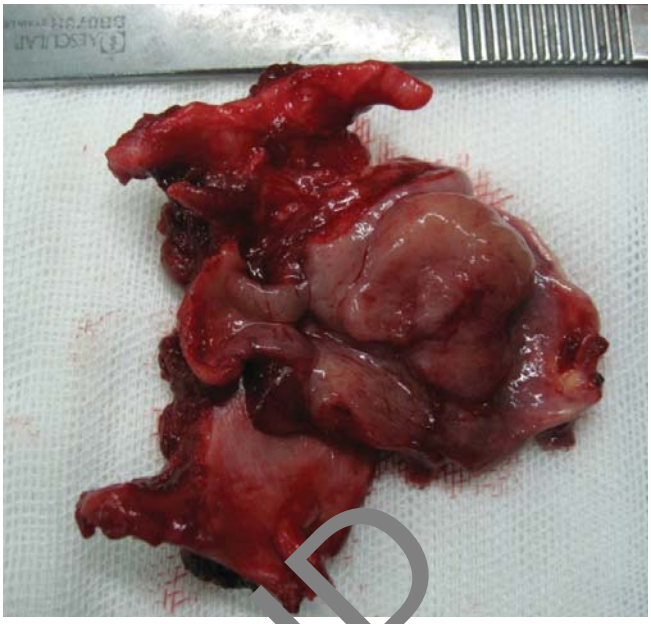
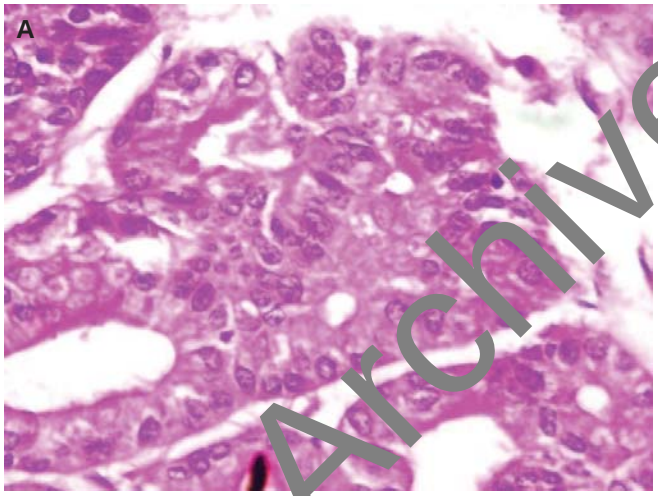


Figure 2. The lesion was not encapsulated and had 30 x 25 x 15 mm diameters localized in the left supraglottis and passed the midline



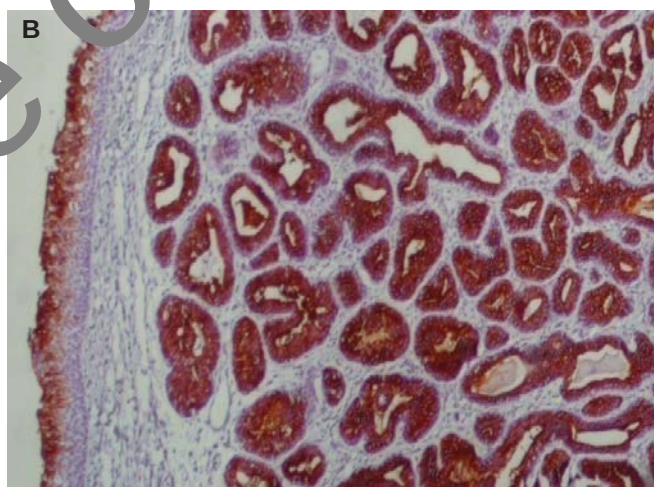


Figure 3. A) Tumor cells are large with hyperchromatic nuclei and abundant eosinophilic cytoplasm (original magnification × 400, Hematoxylin and Eosin stain); **B)** Immunohistochemical staining for CK7 was positive (original magnification × 40).

later Evans and Batsakis published a second series including 14 cases and used PLGA as the diagnosis.[9] PLGA has been reported in lacrimal glands, oropharynx (tonsil and base of the tongue), nasopharynx, uvula, vagina, and breast. PLGA has also been presented as the malignant component of carcinoma ex mixed tumor.[10] This lesion has been reported in an extended age range (16 to 94 years). However, only two cases have been reported in children.[11] Most reported cases were in the fifth to sixth decades of life and majority of them tended to be females which was in consistency with our patient who was a 55-year-old female.[2] PLGA has an infiltrative growth pattern, and commonly invades adjacent bone and cartilage.

Tumor cells are large and uniform; they have vesicular nuclei with pale cytoplasm and eosinophilic color which seems to be washed out in appearance. These cells can be arranged in clusters, sheets, trabecula, small tubules, solid islands, ducts, cystic areas, and foci with single file appearance. These microscopic features are called "polymorphous phrase".[6] In the present case, tumor cells with eosinophilic cytoplasm and round or oval nuclei were arranged in cuboids or columns.

Oncocytic changes with clear cells, squamous or mucinous metaplasia, and focal papillary changes have been reported long be-

fore. Perineural invasion is frequently seen in PLGA, as in adenoid cystic carcinoma, our patient also had perineural invasion. However, necrosis, nuclear atypia, and mitotic figures are rare. Characteristically PLGA has low stroma and CK7 positive, CK20 negative.[12] The proliferation rate of PLGA according to Ki67 staining is less than 6.4 %.[4] In our patient, immunohistochemical staining for CK7 was positive (Figure 7) while immunoreactivity to glial fibrillary acidic protein (GFAP), c-kit, and CK20 were negative. Besides, proliferation rate according to Ki-67 staining of PLGA was 6.2 %.

Histopathologic evaluations allows definite diagnosis of PLGA. However, it might be mistaken with adenoid cystic carcinoma or mixed cellular tumor when the specimen size is small.[4,13]

Multiple growth patterns, cells with eosinophilic cytoplasm, absence of angulated tumor cells, and negative immunostaining reactions for c-kit (CD117) are factors that guides us towards the diagnosis of PLGA. Obviously, although mixed tumors of minor salivary glands are not encapsulated, their periphery is distinct. Perineural invasion is absent in mixed tumors. Myxochondroid area seen in all types of mixed tumors and benign myoepithelial plasmacytoid cells are not present in PLGA. GFAP is not expressed in PLGA while nearly all mixed tumors expressed GFAP in the epithelial, myoepithelial, and stroma. In our patient, GFAP staining was negative.[13]

Extensive surgical resection accompanied with concurrent radiotherapy is recommended for positive margins or recurrent PLGA. In majority of recurrent PLGA cases, surgical resection of the lesion for the second time is also recommended. Therapeutic neck dissection is also recommended when reactive lymph nodes are present.[14] Overall survival rate for PLGA is excellent without considering the location of tumor.[5]

When PLGA is found as the malignant part of a mixed tumor, prognosis is better than other carcinomas presenting in benign mixed tumors.

PLGA is a very rare tumor in the larynx. It is important to recognize and distinguish it from other tumors occurring at this anatomic region.

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