

# Metastasizing Pleomorphic Adenoma of the Tongue: A Case Report

B. Khademi,\* B. Geramizadeh, \*\*  
B. Gandomi,\* S. Ghanbarian\*\*\*

## Abstract

A 19-year-old girl referred to our clinic complaining of a painless lump on the left side of her tongue. The patient had no history of dysphagia, lingual paresthesia or bleeding. Physical examination revealed the presence of multiple enlarged lymph nodes on the left side of her neck. Histopathology of lingual mass and lymph nodes showed pleomorphic adenoma. To our knowledge, this is the first case of pleomorphic adenoma of the tongue with metastasis to the cervical nodes.

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**Keywords** • Metastasizing mixed tumor • adenoma, pleomorphic • tongue neoplasm

## Introduction

**P**leomorphic adenoma is the most frequent tumor arising in human salivary glands, accounting for 40-70% of all salivary gland neoplasms. It is most frequently localized in the parotid and submandibular glands, although an origin in the intraoral salivary and submucosal glands may occur. Microscopy shows a characteristic combination of epithelial, myoepithelial and stromal components, hence the tumor is also known as benign mixed tumor. Three types of mixed salivary gland tumors with metastasizing potential have been described. The first group consists of carcinomas arising in a benign pleomorphic adenoma (carcinoma ex pleomorphic adenoma) occurring in 2.9% of all pleomorphic adenomas.<sup>1</sup> Metastases of these tumors are composed of carcinomatous elements. The second group comprises carcinosarcomas; these are true malignant mixed tumors containing both malignant stromal and malignant epithelial (carcinomatous) components. The third group, which is the rarest, is distant metastasis of a benign pleomorphic adenoma. The histology of the metastatic tumor is identical to that of the primary tumor.<sup>1</sup>

To date, metastasizing pleomorphic adenoma of the tongue has not been reported. We present a case of pleomorphic adenoma of the tongue with metastasis to the cervical lymph nodes.

## Case Report

The patient was a 19-year-old girl who presented with a painless lump of lateral part of the tongue on the left side. She gave no history of dysphagia and lingual paresthesia or bleeding. Also, there was no history of any manipulation of the tongue. Physical examination

\*Department of Otorhinolaryngology,  
Shiraz University of Medical Sciences,  
Shiraz, Iran.

\*\* Department of Pathology, Shiraz  
University of Medical Sciences, Shiraz,  
Iran.

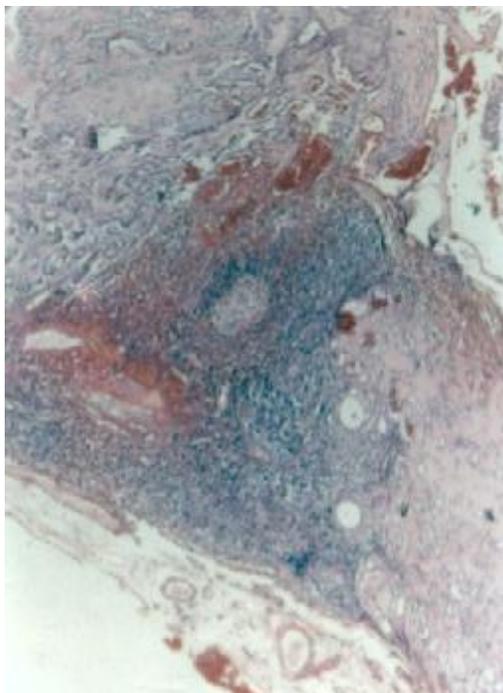
\*\*\*Department of Otorhinolaryngology,  
Fassa School of Medicine, Iran.

**Correspondence:** B. Khademi, M.D,  
Department of Otolaryngology, Khalili  
Hospital, Shiraz, Iran.

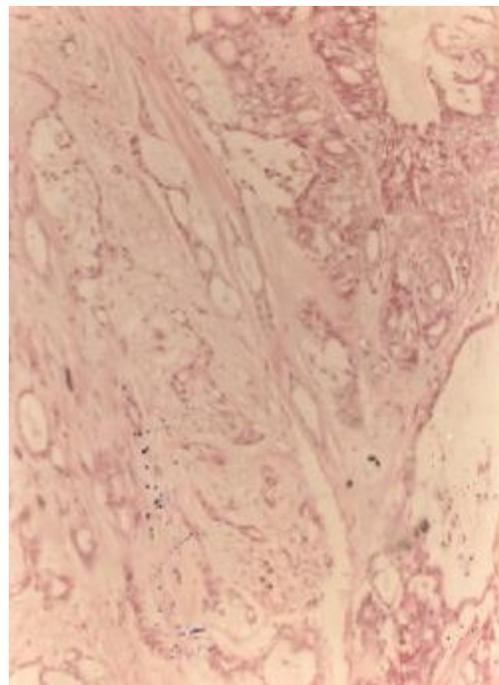
**Tel:** +98-711- 6279372

**Fax:** +98-711 6279372

**E-mail:** [Khademib@yahoo.com](mailto:Khademib@yahoo.com)



**Fig 1:** Section from the primary lesion in the tongue, showing infiltration of muscle fibers by tumor cells (H&E  $\times$  200).



**Fig 2:** Histopathologic section from the neck node showing metastatic pleomorphic adenoma (H&E  $\times$  200).

revealed a sessile round mass on the middle third of the left lateral side of the tongue that was covered with normal lingual mucosa. Physical examination revealed the presence of multiple asymptomatic lymph nodes on the left side of the neck (submandibular as well as upper and midjugular chains). Parotid and submandibular glands were normal. Under local anesthesia, biopsies of the lingual lump and midjugular lymph nodes were taken and studied histopathologically. Microscopic study of the biopsied specimens from the lingual mass showed typical pleomorphic adenoma (Fig 1). Sections from lymph nodes showed a metastasizing benign pleomorphic adenoma with no evidence of anaplasia (Fig 2). Under general anesthesia, the lingual mass was excised with a cuff of normal tissue. A left sided neck dissection was also performed and histopathologic findings were similar to previous biopsy. Although many sections of the whole specimen were obtained, no sign of malignancy was detected. The patient was followed for 2 years, during which no recurrence of the primary site was detected. There was development of cervical lymph node enlargement (in the left suprahyoid region and right midjugular chain).

Therefore, left supraomohyoid neck dissection and right sided functional neck dissection were performed which revealed the same histopathologic picture as the previous one, i.e. pleomorphic adenoma.

## Discussion

Pleomorphic adenoma usually presents in the tail of the parotid gland as a painless, slowly growing mass. There is a predilection for women in the fifth decade of life although the tumor can occur at any age. Approximately, 6.5% of pleomorphic adenomas occur in the minor salivary glands and are usually located in the palate.<sup>2</sup> Pleomorphic adenoma is generally regarded as a benign tumor however occasionally some types of this tumor have malignant microscopic features or behave in a malignant fashion. The most common malignant form is carcinoma ex-pleomorphic adenoma. Only the epithelial component transforms into a malignancy and is capable of metastasis. Another histologically malignant type is malignant mixed tumor where both the epithelial and mesenchymal-like components develop a malignant microscopic appearance and metastasize together. There has

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also been a small series of cases of metastasizing pleomorphic adenoma in which both the primary and the metastasis maintained their benign microscopic appearance.<sup>3</sup> Patients invariably have had a mixed tumor removed from the parotid or some other salivary gland and metastatic spread is usually preceded by multiple episodes of local tumor recurrence.<sup>4</sup> Pleomorphic adenoma is also the most common lesion in the minor salivary glands, however, approximately 50% of all minor salivary gland tumors are malignant.<sup>5</sup> Common sites of minor salivary gland tumors are the palate, followed by the maxillary sinus, lip, cheek, tongue and others.<sup>5</sup> In the tongue, malignant neoplasms of salivary gland origin such as adenoid cystic carcinoma, adenocarcinoma, and mucoepidermoid carcinoma are predominant over benign tumors.<sup>5</sup> Our case is insofar unique since the primary site of the tumor is minor salivary glands of the tongue as opposed to previous reports of parotid origin.<sup>2</sup> In all cases of metastasizing pleomorphic adenoma reported previously, patients were treated for local pleomorphic adenoma by surgical excision or enucleation prior to the first presentation of distant metastasis. Many patients had also undergone additional treatments for one or more local recurrences before presentation of distant metastasis. It has been suggested that metastasis of a pleomorphic adenoma can only occur if

previous surgical manipulation has provided the opportunity for the pleomorphic adenoma cells to enter lymphatic or blood vessels, thus giving rise to systemic metastasis.<sup>1</sup> In our case, both the primary lesion and the cervical nodal metastasis presented nearly simultaneously and there was no history of any previous manipulation.

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