Case Report / Olgu Sunumu

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Primary adenocarcinoma of the base of tongue: a rare case

Dil kökünün primer adenokarsinomu: Nadir bir olgu

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Abstract

Head and neck cancers are the sixth most common type of cancer in world; and of these, less than 10% are oropharyngeal cancers. The most common malignant tumor of oropharynx, even though it is accepted as a part of digestive system, is squamous cell carcinoma. On the other hand, adenocarcinoma, a common histopathological type for digestive system, is very rarely reported at base of tongue. Therefore, this article was aimed to present a 72-year-old male patient with a primary adenocarcinoma of the base of tongue and discuss the clinical presentation, radiological evaluation, histopathological examination, treatment modalities and prognosis with the literature.

Keywords: Head and neck neoplasms, oropharyngeal neoplasms, adenocarcinoma.

Özet

Baş boyun kanserleri, tüm kanserler arasında sıklığı itibariyle altıncı sıradadır ve bu kanserlerin %10'undan azını orofarenks kanserleri oluşturmaktadır. Orofarenks her ne kadar sindirim sisteminin bir parçası olarak kabul edilse de, en sık rastlanan malign tümörü yassı hücreli karsinomdur. Ancak, nadiren de olsa bu bölgede sindirim sisteminde sık rastlanan adenokarsinom da gelişebilmektedir. Bu makalede, primer dil kökü adenokarsinomu olan 72 yaşında erkek olgu sunumu ile nadir rastlanan bu kanserin klinik tablosu, radyolojik değerlendirmesi, histopatolojik incelemesi, tedavisi ve prognozu tartışılmıştır.

Anahtar sözcükler: Baş boyun neoplazmları, orofarengeal neoplazmlar, adenokarsinom.

Oropharyngeal cancers constitute less than %10 of all head and neck cancers despite of an increasing incidence worldwide.[1] A vast majority (≥70%) of patients with oropharyngeal cancers are male. The main etiological factors of oropharyngeal cancers are: smoking and alcohol consumption, and human papilloma virus infection.[1,2] Unfortunately, most of the patients with oropharyngeal cancers, especially patients with a malignant lesion at the base of tongue, are diagnosed at a locally advanced stage or with cervical/distant metastases.[3] The main reason of delay in diagnosis is generally related to the asymptomatic development of the tumoral mass; however, odynophagia, dysphagia and referred pain may be reported at early stages. The major treatment modality for locally advanced oropharyngeal cancers is concomitant radiotherapy or surgical resection with adjuvant radiochemotherapy. However, the surgical treatment of this region is challenging due to its anatomical difficulties during surgery and may cause functional problems such as chronic aspiration, dysphonia and dysphagia.

The predominant histopathological type of oropharyngeal cancers is squamous cell carcinoma; and approximately 9 times more frequent when compared with other types.[4] On the other hand, adenocarcinoma, a common histopathological type for digestive system, is very rarely reported at this region, even though oropharynx is accepted as a part of digestive system. In literature, there is lim-

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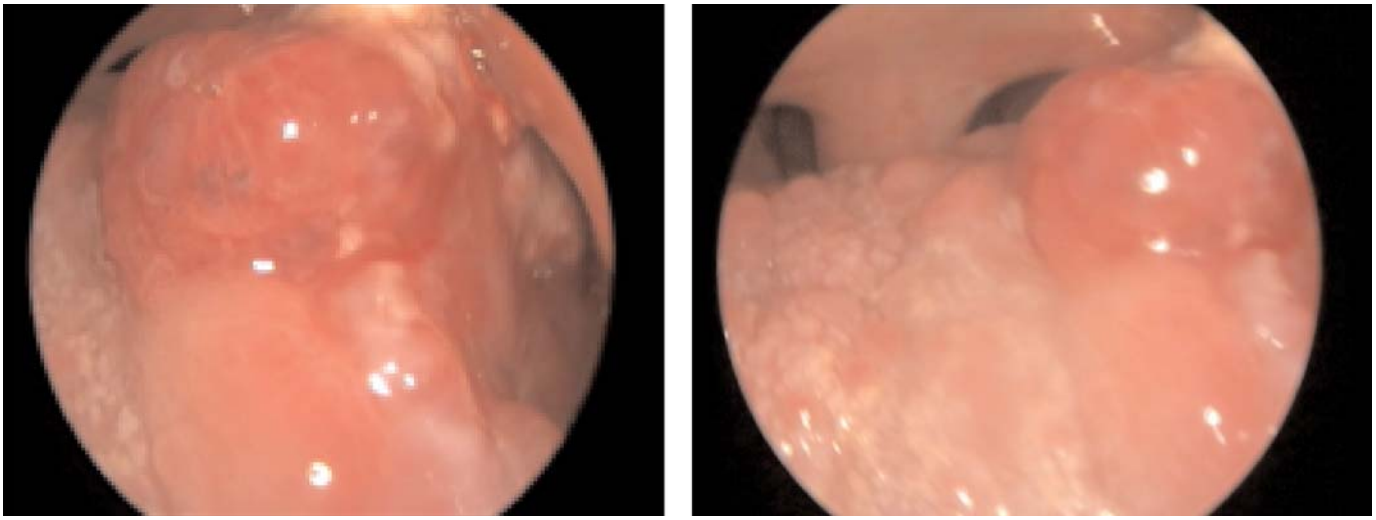

Fig. 1. The view of tumoral mass located at the left side of the base of tongue.

ited data about the clinical presentation, histopathology and treatment modalities for the primary adenocarcinoma of the base of tongue. Therefore, this article was aimed to present a case with a primary adenocarcinoma of the base of tongue and discuss the clinical presentation, histopathological examination, treatment and prognosis with the literature.

Case Report

A 72-year-old male patient with a history of choking and pain on left side of his throat while swallowing administered to an ENT specialist in elsewhere. An incisional biopsy from the lesion, which was located at the base of tongue, was performed and histopathological examination was reported as highly suspicious for a malignancy. The patient, whose symptoms relieved for a duration, administered to our center 18 months after the initial biopsy because of an increase in his complaints of odynophagia and otalgia. The physical examination revealed a mass of 3 cm in size located at the left side of the base of tongue (Fig. 1). No lymphadenopathy was detected in neck examination, and the patient was normal otherwise. He was a smoker (1 pack of cigarette/day for fifty years) and regular drinker (4 glasses of alcohol/day for fifty years). His history revealed no surgical procedure or any additional disease.

The magnetic resonance imaging demonstrated a potentially malignant mass with a diameter of 22×15×22 mm at the base of tongue, and the lesion was located very close to the lateral pterygoid muscle (Fig. 2). An incisional biopsy, which was reported as malign epithelial tumor, was performed. The PET/CT was suggested in order to determine the presence of local or distant metastases. A hypermetabolic mass at the base of tongue with a standardized uptake value (SUV) of 14 and a millimetric lymphadenopathy with a slight increase in SUV (2.8) on left side of the neck was detected and no sign of distant metastases was determined (Fig. 3a). Thereby, the patient was clinically staged as cT2N1M0 (Stage III) oropharyngeal carcinoma (at the base of tongue), and surgery with adjuvant radiochemotherapy was recommended. In surgery, laser-assisted tumor excision with primary reconstruction, and left modified radical neck dissection (sternocleidomastoid muscle, internal jugular vein and accessory nerves were protected), and right elective (supraomohyoid) neck dissection were performed. The postoperative follow-up of the patient was uneventful, and no problem in swallowing and phonation was observed. The histopathological examination of the patient was reported as "*adenocarcinoma*". In addition, the immunohistochemical evaluations were as follow: mono-CEA (+), AE1/AE3 (positive in each cell), synaptophysin focal weak positive, chromogranin (-), NSE (-), HMWCK (-), chromogranin ((-) 2 pieces), (Figs. 4a and b). Both histopathological and immunohistochemical evaluations demonstrated that the tumor was not originated from a minor salivary gland; therefore it was considered as "*primary adenocarcinoma of the base of tongue*". In addition, cervical metastasis was detected on one of the lymph nodes of the left neck. Therefore, the pathological stage of the patient was also evaluated as

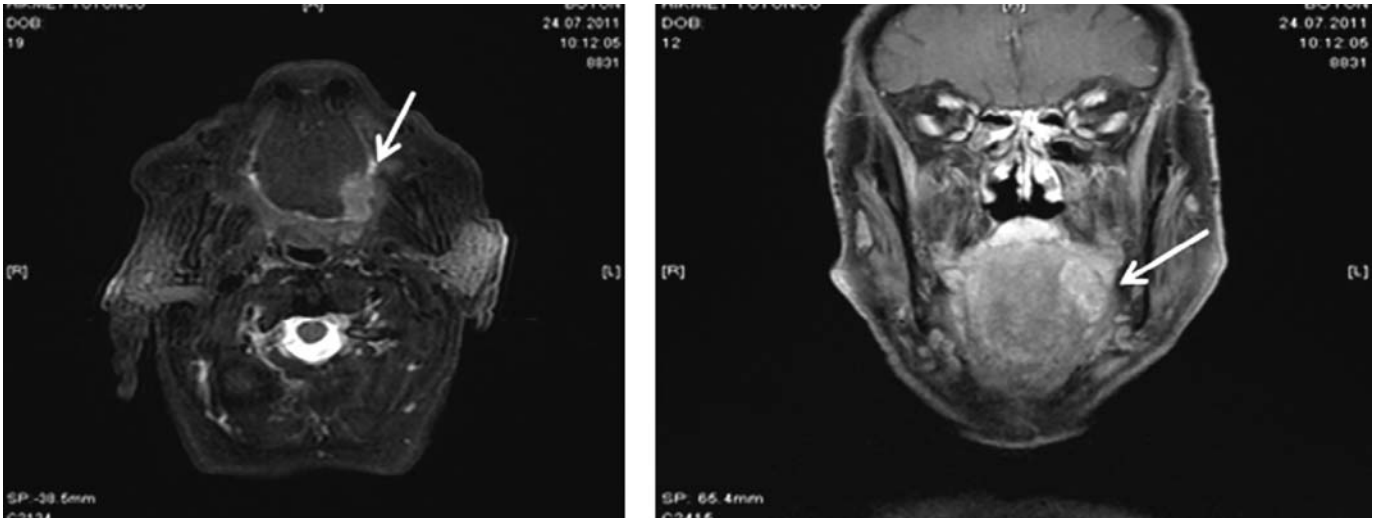

Fig. 2. The magnetic resonance imaging of the lesion that was marked by a white arrow.

pT2N1M0 (Stage III). An adjuvant treatment with intensity- modulated radiation therapy (IMRT) and chemotherapy (cisplatin) was recommended; however, only IMRT was applied and chemotherapy could not be performed due to patient's performance status. Patient was regularly follow-up after the treatment; however, lung and brain metastasis were detected at the fifth month of surgery. (Fig. 3b). Therefore, a palliative chemotherapy was considered; unfortunately, patient died due to cardiac arrest before the application of chemotherapy.

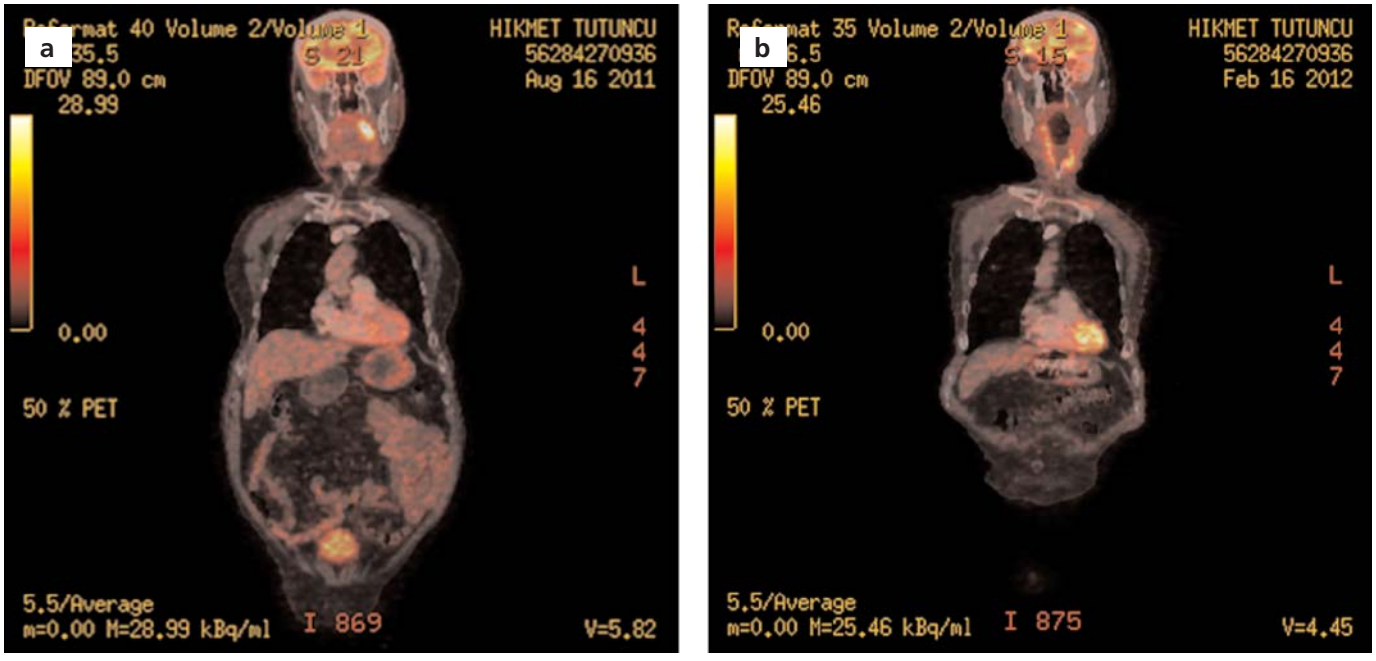

Fig. 3. Preoperative PET/CT demonstrated the mass at the base of the tongue with a hypermetabolic tumoral bulky lesion (SUV:14) and a lymphadenopathy (7 mm in size) at the left neck with a slight increase in SUV (a). Postoperative 5th month PET/CT: hyper metabolic nodules, that were considered in the favor of lung metastasis, were observed in both lungs when compared with the previous PET/CT, although no sign of disease was seen at the base of tongue and neck.

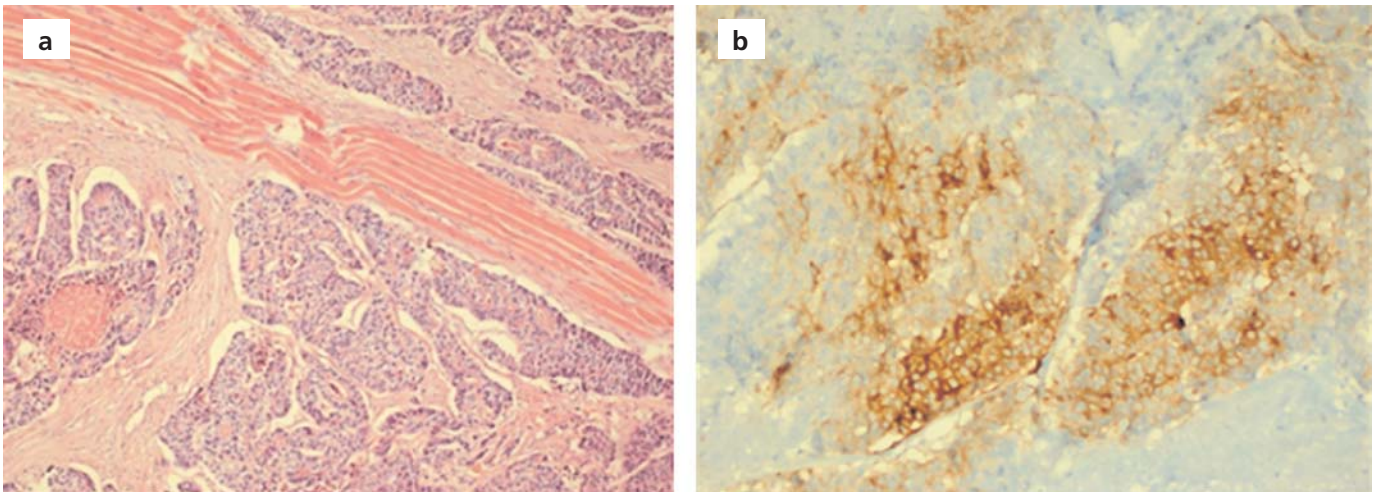

Fig. 4. The tumor cells with hyperchromatic nucleus and eosinophilic cytoplasm, which forms gland structures, and thereby creating lumen fiber formations and showing invasions between muscle (H&E, x100) (a). Intracytoplasmic and membranous monoclonal CEA in tumor cells (mCEA x200) (b).

Discussion

Adenocarcinoma is a relatively rare histopathological type of head and neck cancers. deVries et al. determined only 6 cases with adenocarcinoma of the base of tongue in their retrospective review of medical records betwen 1955 and 1985;[5] and, all of the lesions in this study were originated from minor salivary glands. On the other hand, primary adenocarcinoma of the base of tongue is very unusual. In the current article, a case with a locally advanced primary adenocarcinoma of the base of tongue with a regional cervical lymph node metastasis, which is a characteristic feature of malignancies of base of tongue, has been presented. Adenocarcinomas generally contain glycogen before and after digestion with diastase. On the other hand, most of other tumors have very little or no glycogen content. In addition, intracytoplasmic mucins are not evident with mucicarmine or alcian blue stain.[6] In this case, the diagnosis of adenocarcinoma was determined depending on the morphologic and immunohistochemical evaluations (mono-CEA and AE1/AE3 positivity). In addition, no evidence of minor salivary gland was detected; therefore it was considered as "*primary adenocarcinoma of the base of tongue*".

According to NCCN 2012 guidelines, the recommended treatments for locally advanced oropharyngeal cancer are definitive radiotherapy, surgical resection with neck dissection and adjuvant radiotherapy when at least one positive lymph node is detected, and definitive radiotherapy with systemic therapy.[7] However, these treatment modalities are especially suggested for squamous cell carcinoma; and, unfortunately, optimal treatment of adenocarcinoma of base of tongue has not been established due to its rarity. Kessler et al. offered that combined approach with surgery, radiotherapy and chemotherapy if required, can be applied for the treatment of malignant salivary gland tumors of the base of tongue.[8] Therefore, we also preferred to perform wide tumor resection with neck dissection and adjuvant radiochemotherapy, although chemotherapy could not be performed due to performance status of the patient. The follow-up of our patient demonstrated no local or regional recurrence; however, distant metastasis of lung and brain was determined soon after the treatment. Unfortunately, the distant metastasis is high and survival rate is relatively low in malignant tumors of the base of tongue regardless of the histopathological type and treatment. In a retrospective study, that included 262 cases of squamous cell carcinoma of the base of tongue, the 5 year-survival rate for all stages was %49.6.[9] In another study that examined the efficacy of treatment modalities on prognosis in patients with squamous cell carcinoma of the base of tongue, 5-year survival rate for patients treated by irradiation was 41%, for patients treated by surgery was 58%, and for untreated patients was 9%.[3] They emphasized that a remarkable difference was observed in survival rate between treated and untreated patients, although no obvious difference in the efficacy of treatment modalities was determined. Similarly, Kessler et al. reported the high rate of mortality (77% died in 10 years) and distant metastasis (57%) in patients with minor salivary cancers of the base of

tongue.[8] However, our literature survey demonstrated lack of evidence about the short and long term outcomes of treatment modalities and prognosis for adenocarcinoma of the base of the tongue. Therefore, multicenter and multidisciplinary studies are required other than reported case reports.

Conflict of Interest: No conflicts declared.

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