

Indistinctly Synchronous Lung and Base of the Tongue Adenocarcinoma: Case Report and Discussion of Possible Patterns of Disease Development

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ABSTRACT:

Lung adenocarcinoma accounts for about 40% of all lung cancers and is characterized by distinct cellular and molecular features. Distant metastases from primary lung cancer are frequent, however metastasis of the tongue is quite rare and unexpected. If there is existing evidence of metastasis in tongue, primary tumor is most probably generally disseminated and prognosis is poor. Such patients require a complete staging workup to evaluate the extent of disease, because tumor stage plays a major role in determining the choice of treatment. In this case report we want to present an unusual case of simultaneous lung and base of the tongue adenocarcinoma and discuss the possible patterns of disease development.

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KEYWORDS: lung adenocarcinoma, tongue, metastasis

SAŽETAK:

NEJASNO SINKRONI ADENOKARCINOM PLUĆA I BAZE JEZIKA: PRIKAZ SLUČAJA I RASPRAVA O MOGUĆIM OBRASCIMA RAZVOJA BOLESTI

Adenokarcinom pluća čini oko 40% svih karcinoma pluća i karakteriziraju ga različite stanične i molekularne značajke. Česte su udaljene metastaze iz primarnog karcinoma pluća, međutim metastaze u jeziku prilično su rijetke i neočekivane. Ako postoje dokazi o metastazama u jeziku, primarni tumor je najvjerojatnije širen, a prognoza loša. Takvi pacijenti zahtijevaju cjelovitu obradu kako bi se procijenio opseg bolesti, jer stadij tumora igra glavnu ulogu u određivanju izbora liječenja. U ovom slučaju želimo predstaviti neobičan slučaj istodobnog adenokarcinoma pluća i baze jezika i razgovarati o mogućim obrascima razvoja bolesti.

KLJUČNE RIJEČI: adenokarcinom pluća, jezik, metastaze

CASE REPORT

The 66-year-old Caucasian male patient was referred to our Department with histologically proven adenocarcinoma of the base of the tongue. His primary symptom was difficulty in swallowing and breathing although patient was not dysphonic and was on regular diet. Patient also reported occasional haemoptysis in last two months. The patient was a smoker with a long history of significant consumption of alcohol and sawdust exposure starting 45 years ago, and no history of previous tumor disease. Clinical examination revealed a left-sided tumor located at the base of the tongue, partially covering the epiglottis. MSCT scans revealed an exophytic, mushroom-like tumor of the left side of the base of the tongue connected to the epithelium by a thin pedicle with practically no infiltration of the surrounding tissue. No suspected positive lymph nodes were found in the neck.

Furthermore, routine chest MSCT scan additionally revealed a tumor measuring 30 mm in diameter located in the lower left lung. Subsequently CT-controlled transthoracic fine needle biopsy was performed and histopathological analysis revealed a tiny focus consistent with adenocarcinoma. Routine PCR analysis for EGF receptor was performed, revealing negative results, but the specimen was inadequate for further immunohistochemical analysis to confirm the primary lung carcinoma. PET-CT showed no other tumor masses, except for the pulmonary and base of tongue tumor. At that stage the patient refused any kind of surgery and he was presented and discussed at the oncology group meeting, where the diagnosis of pulmonary adenocarcinoma cT1 N0 M1b, stage IVa, was made. Patient was treated with pemetrexed in combination with cisplatin chemotherapy protocol. After receiving two cycles of chemo, patient refused further therapy and medical assistance due to personal reasons. Two months later the patient presented to outpatient clinic complaining of having difficulty swallowing. MSCT scan showed a reduced size of pulmonary tumor with slightly increased tumor size at the base of tongue. Considering localization and need for subsequent reconstruction, comprehensive surgical procedure followed, including partial glossectomy, selective neck dissection (I-IV) and reconstruction with microvascular flap (radial forearm). Histopathological examination of the surgical specimen revealed an exophytic tumor on the base of the tongue, measuring 36 mm, composed of atypical epithelial cells forming alveolar, tubular and cribriform pattern with multiple necrotic foci. Neck dissection revealed a conglomerate composed of several positive lymph nodes in regions II and III. Immunohistochemical analysis revealed negative reactivity with Napsin A, CK5/6, TTF-1 and CK20, and partially positive reactivity with CDX-2 and CK7. The diagnosis was adenocarcinoma of the tongue with lymph node metastasis. PCR analysis for EGF receptor was negative. Oncology group decided to proceed with the treatment with pembrolizumab immunotherapy according to the protocol. After receiving 5 cycles of immunotherapy, MSCT scan showed clear status intraorally and in the neck, without evidence of any suspicious pulmonary mass. At present, 12 months after the treatment there are no signs of recurrent disease neither in the lung nor in the neck.

The accurate diagnosis of histologically identical simultaneous tumors is diagnostically challenging. Primary adenocarcinoma of the tongue is exceptional, therefore presented case has been considered as primary lung adenocarcinoma. The presence of lung cancer metastasis in the base of the tongue is a rare clinical entity and according to available literature several cases have been reported¹.

Due to lack of material obtained from the lung tumor with fine needle biopsy we could not perform immunohistochemical markers to confirm lung origin of the tumor (TTF-1, Napsin A) and to establish relationship between lung and base of the tongue tumor. Although, presented case has been considered as primary lung adenocarcinoma, we established three possible scenarios for such a disease development: 1. primary pulmonary carcinoma metastasizing into the tongue, 2. primary base of tongue carcinoma metastasizing into the lung, 3. or synchronous, independent pulmonary and base of tongue carcinoma. For the third hypothesis, of two synchronous, independent tumors we do not have enough histological evidence to prove whether these tumors are of the same or different origin. The second hypothesis, where base of the tongue tumor is primary and lung is metastasis, is possible but in our opinion is not likely. Main reason for this is exophytic grow, practically without infiltration of tongue tissue. Secondly, MSCT and PET-CT negative neck at first visit. Positive neck was developed several months after initial chemotherapy.

The first hypothesis, of the tongue tumor being metastasis of the lung adenocarcinoma, in our opinion, is the most likely scenario. Possible patterns of metastasizing into tongue could be haematological spread or inoculation of cancer cells into mucosal epithelium. In our case spread through the blood stream is unlikely for two reasons. Firstly, the base of the tongue tumor would be the only metastasis and in all other reported cases tongue metastasis was one of many other metastases. Secondly, the blood stream metastasis would most likely present in the base of the tongue, submucosally, and not only intraepithelially. For these reasons we believe that the present case is not induced by hematological spread and the theory of epithelial inoculation with cancer cells originating from lung tumor is the most likely. Models investigated in animals confirm the possibility of such a pattern. We believe that the tongue metastasis was caused by inoculation of malignant cells by coughing and seeding into the lingual epithelium. Hayasaka et al.² reported a case of multiple endotracheal metastases of lung cancer after bronchoscopic intervention. Later regional neck disease was caused by lymphatic spread of tongue tumors and we base that opinion on the negative MSCT and PET-CT scans during initial and control check-ups.

To our knowledge, this is the only recorded case of simultaneous adenocarcinoma of the lung and the base of the tongue (as a single metastasis). Although, due to the lack of histological evidence, we could not establish clear pattern of disease development, we believe that this case report could contribute to a better understanding of the possible disease spread and subsequently its treatment.

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