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Pulmonary Metastases of Basal Cell Adenocarcinoma Presenting as Hemoptysis

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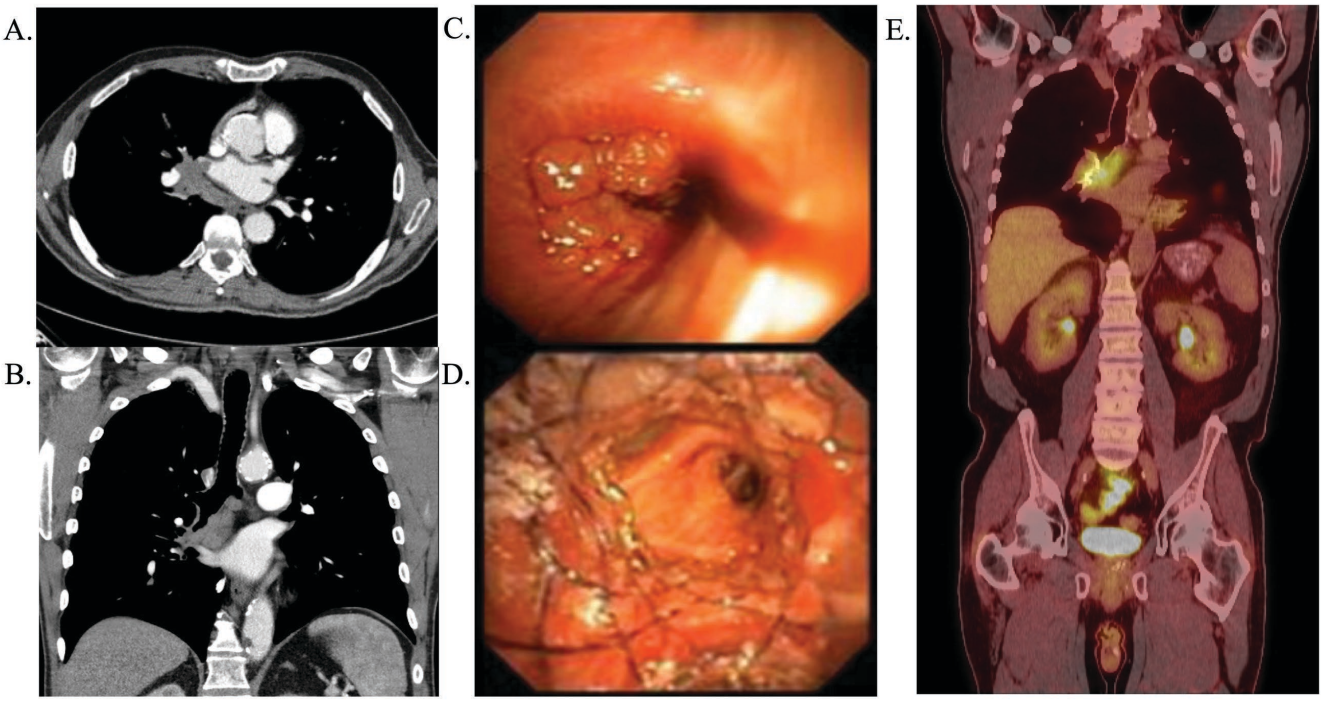

Figure 1: Computed tomography (CT) chest revealing tumor invasion with intraluminal thrombus of the right superior pulmonic vein (**A** and **B**). Bronchoscopy revealing luminal compromise (**C**) prompting tracheal stent placement (**D**). Positron emission tomography (PET) imaging ultimately showed increased tracer uptake concerning for malignancy (**E**).

INTRODUCTION

Basal Cell Adenocarcinoma (BCAC) is a rare malignancy, only accounting for approximately 2% of all salivary neoplasms[1]. Considered the malignant counterpart of basal cell adenoma, it most commonly presents at 60 years of age without gender predilection. Sites of involvement frequently includes the parotid gland, but sites in the minor salivary glands, nasopharynx, buccal mucosa, and tongue have also been reported[2,3]. Often regarded as an indolent malignancy, BCAC can occasionally cause invasive disease and infrequently, metastatic disease[2]. Among all solid tumors, endobronchial metastases is quite a rare occurrence, contributing to approximately 4% of endobronchial biopsies[4]. Most common sites of metastases in BCAC include cervical lymph nodes with sparse reports of pulmonary, hepatic and cutaneous involvement[5]. We present a case of endobronchial metastases from BCAC of the base of the tongue.

CASE PRESENTATION & OUTCOME

A 76-year-old male with a past medical history of BCAC who underwent right tongue base excision with bilateral neck dissection followed by adjuvant radiotherapy in 2018 and prostate cancer treated with radiotherapy who presented with a one-day history of hemoptysis. Two weeks prior to initial presentation, he underwent a CT-guided lung biopsy for a right upper lobe nodule that was nondiagnostic. There was concern that his hemoptysis was related to his recent procedure, so he was transferred to the academic medical center for further evaluation.

On presentation, the patient felt well overall and had not experienced any additional occurrences of hemoptysis. Vital signs were stable, and physical examination was significant for decreased breath sounds in the right lower lung field. CT chest with contrast was performed and revealed an enlarging mediastinal and right infra-hilar lymph node causing severe stenosis of the bronchus intermedius, right middle, and right lower lobe bronchus. Findings also included direct invasion with an intraluminal thrombus of the right superior pulmonic vein (**Figures 1A, 1B**).

On hospital day 2, patient underwent bronchoscopy that revealed extensive endobronchial tumor involvement of the bronchus intermedius resulting in approximately 80% luminal compromise (**Figure 1C**). Tumor debulking was performed as well as tracheal stent placement (**Figure 1D**). Patient was discharged following the procedure, and an outpatient PET scan confirmed malignant involvement (**Figure 1E**). Pathology of the endobronchial mass revealed a poorly differentiated basaloid epithelial neoplasm that was morphologically similar the prior tongue biopsy. Of note, histology revealed an exceptionally high proliferation index (Ki67 > 90%). Comprehensive genomic profiling did not reveal any potential targeted therapies. After discussion with the multidisciplinary tumor board, the decision was made to pursue palliative radiotherapy followed by pembrolizumab monotherapy. The patient expired at home approximately two months after initial diagnosis.

DISCUSSION & KEY POINTS

The management of BCAC, as with all salivary gland neoplasms, is particularly challenging due to the phenotypic heterogeneity and low clinical incidence. The most common strategy includes surgical resection with adjuvant radiotherapy. While distant metastases is an uncommon recurrence, local recurrence is quite common despite tumor-free surgical margins with rates of upward to 50%[2]. Chemotherapy has been found to be of limited use, predominately in the setting of recurrent or metastatic disease. Currently, there are no National Comprehensive Care Network (NCCN) recommendations for specific regimens. The most common agents include cisplatin, doxorubicin with either 5-fluorouracil or cyclophosphamide. However, no regimen has produced significant and reproducible improvements in overall survival or disease-free survival[6].

This case demonstrates several unique features. This patient did not experience local recurrence, but rather distant metastasis to a novel location. Also, this patient featured an exceptionally high proliferation index, indicating that BCAC has the potential for clinical heterogeneity. Finally, this case highlights the need for further research into the role of imaging surveillance during remission.

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