

Case Report

Rare tracheal mucoepidermoid carcinoma with invasion of thyroid gland initially misdiagnosed as invasive thyroid cancer

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Received January 8, 2016; Accepted March 23, 2016; Epub June 15, 2016; Published June 30, 2016

Abstract: Mucoepidermoid carcinoma of the trachea is a rare tumor. We report an 80-year-old male with mucoepidermoid carcinoma in the upper trachea with invasion of the isthmus of thyroid gland. The patient's major symptom was dyspnea with coughing. Computed tomography scan showed a protruding mass into the trachea suspected of thyroid cancer. Ultrasonography also revealed the suspicion of an invasive thyroid cancer. Under diagnosis of invasive thyroid cancer, total thyroidectomy, tracheal resection and reconstruction were performed. Although pre-operative imaging studies indicated that the lesion was primary thyroid cancer, later histopathologic examination revealed that the tumor was rare trachea mucoepidermoid carcinoma. We should consider the possibility that tumors regarded as thyroid cancer with tracheal invasion under imaging studies could be a primary tracheal cancer, especially in patients with obstructive airway symptoms.

Keywords: Mucoepidermoid carcinoma, thyroid cancer, tracheal neoplasms

Introduction

Mucoepidermoid carcinomas (MEC) originate from the serous and mucous glands of the upper airway. The incidence of these tumors is rare, consisting of only 0.1 to 0.2% of primary lung cancers and 1 to 5% of tracheobronchial adenoma [1]. These tumors are frequently misdiagnosed as asthma, bronchitis, pneumonia, or as aggressive thyroid cancer when at the level of the thyroid cartilage due to the rare incidence. We demonstrate successful surgical treatment in a patient with MEC of the trachea with thyroid invasion that was initially misdiagnosed as invasive thyroid cancer.

Case report

An 80-year-old male, who weighed 55 kg and was 165 cm tall, presented with dyspnea even at resting state, of 2 months prior to onset. He was a 60 pack-year ex-smoker who had stopped smoking 3 months ago. Apart from dyspnea, he complained cough and sputum but not hemop-

tysis, dysphagia or hoarseness. He had taken medications for chronic obstructive pulmonary disease for approximately 1 year 2 months prior. Pulmonary function test demonstrated that the ratio of forced expiratory volume in 1 s to forced vital capacity was 51%. A chest radiograph showed a well-defined intratracheal mass like opacity at the T2 level. Chest computed tomography (CT) showed a marked thyroid mass that was an approximately 3.4 cm heterogeneous enhancing mass in the right thyroid gland with invasion of the thyroid cartilage and narrowing upper trachea (**Figure 1A, 1B**). Additionally, thyroid ultrasonography detected a 2.6 cm suspicious mass with tracheal invasion in the right lower thyroid that was an irregular ill-defined hypoechoic nodule with microcalcifications. The patient was scheduled for a total thyroidectomy and tracheal resection and reconstruction under the diagnosis of invasive thyroid cancer without fine needle aspiration (FNA) biopsy of the thyroid and bronchoscopic biopsy.

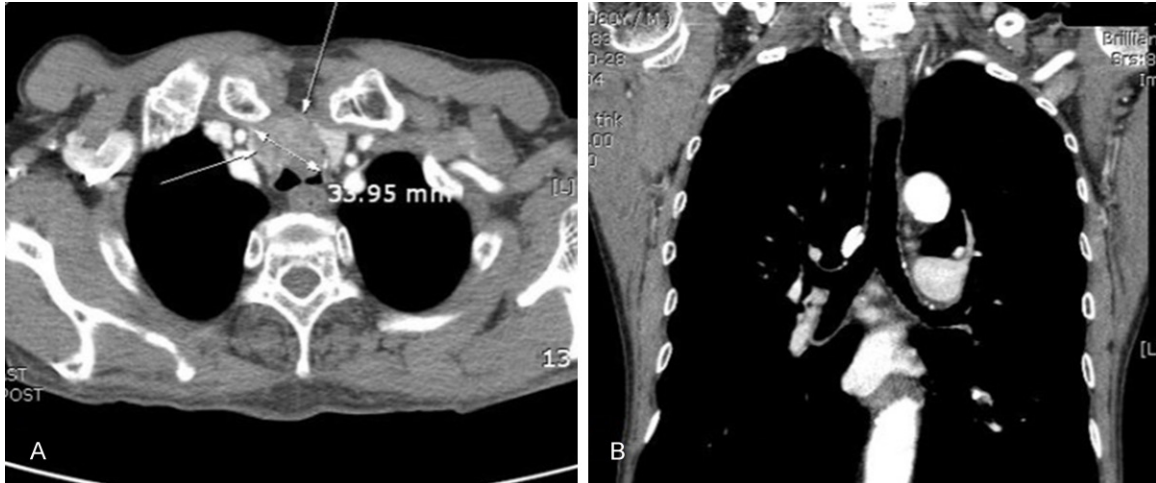


Figure 1. Computed tomographic scan. A. Axial section shows the narrowest point of the trachea. B. Coronal section shows the tumor lying 8 cm above the carina at the level of thyroid cartilage.

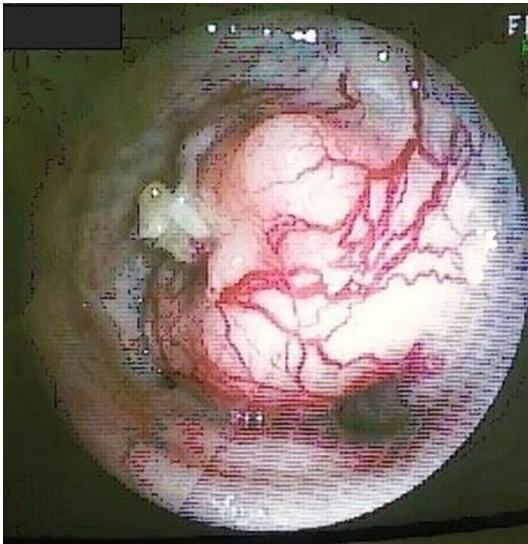


Figure 2. Bronchoscopy showing a tracheal mass. Reddish surface with mesh-like dilated vessels is shown.

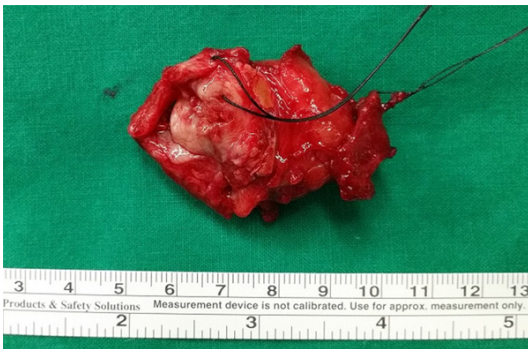


Figure 3. Intraoperative gross photo of the tracheal mass.

Because of tracheal narrowing, difficulty of intubation was expected, we decided to perform an awake fiberoptic intubation. Beyond the vocal cord, bronchoscopy identified a large polypoid, localized mucosal red mass, located at the anterolateral portion of the trachea that obstructed the tracheal lumen (**Figure 2**) and a reinforced endotracheal tube (6.0 mm internal diameter) could be passed around the mass. Under general anesthesia, the thyroid gland was partially detached and tracheal resection was proceeded on the attached isthmus portion of the thyroid gland due to the directly infiltrating lesion. The trachea was transected distally below the inferior border of the tumor. Following withdrawal of the upper orotracheal intubated tube up to the level above the lesion, a reinforced tube was inserted into the opened trachea, distal to the site of resection across the operative field. The trachea was then transected proximally (the portion of cricoid cartilage), the tumor was removed with the thyroid gland. The brownish protruding tumor mass was measured to be 3.3 × 2.8 × 2.2 cm (**Figure 3**). With removal of the tube that was distally intubated directly into the trachea, the orotracheal tube was reinserted into the trachea passed the site of anastomosis using the fiberoptic bronchoscopy, and the cricotracheal anastomosis was completed.

Histopathologic examination presented the intermediate grade MEC. For differential diagnosis, immunohistochemistry was performed using thyroid transcription factor 1 (TTF-1). Con-

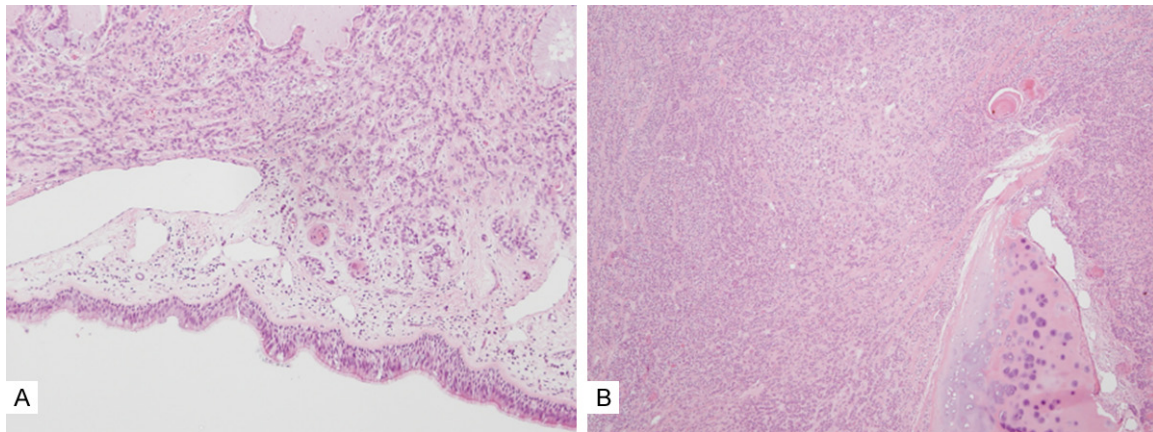


Figure 4. Tracheal mucoepidermoid carcinoma identified by immunohistochemistry for TTF-1. (A) The MEC seemed to arise at subepithelial lesion at trachea. (B) The MEC invaded the hyaline cartilage of trachea and penetrated to thyroid gland. [Original magnifications (A), (B) 40 ×].

trary to the expectation of primary thyroid cancer, the tumor cells did not show immunoreactivity to TTF-1, suggesting the idea that they originated from not thyroid but trachea (**Figure 4A, 4B**). Postoperative bronchoscopic examination revealed no complication of the surgical tracheal reconstruction site. He was discharged without chemotherapy or radiotherapy.

Discussion

MEC in the lung was first reported in 1952 by Smetana [2]. It is uncommon in the tracheal bronchial tree and accounts for 0.2% of all lung tumors, and 1 to 5% of all tracheobronchial adenomas [1]. The 3 main histologic cell types consist of squamous, mucus-secreting, and intermediate cell. The squamoid cell proportion on tumor histology may be an indicator of the level of tumor malignancy [3]. Prognosis depends on histological grading of the tumor and the possibility of complete surgical resection. Complete resection of low and intermediate MEC can effectively minimize the need for further therapy. On the other hand, because high grade tumor is frequently not indicated for complete resection, adjuvant chemotherapy and radiotherapy may be beneficial in high grade tumor.

Although thyroid carcinoma with tracheal invasion is uncommon, it is considerably more prevalent than primary tracheal tumors. The incidence of primary thyroid carcinoma is 13.5:100,000 people per year [4], compared to 0.1:100,000 people per year in the primary tra-

cheal malignancy [5]. Additionally, primary tracheal cancers are observed less frequently than laryngeal or bronchial cancers. These differences in incidence can result in initial misdiagnosis in patients with thyroid mass as our case.

Imaging studies such as CT scan or ultrasonography are useful for providing information on lesion size and location and evaluating the extensive tumor involvement. Both flexible and rigid bronchoscopy provide a definitive diagnosis of tracheal obstruction and treatment plan to secure the airway and approach the surgical site. All these are important considerations for anesthetic and surgical treatment strategies, as well as for preoperative diagnostic evaluation. However, as our case, only imaging studies cannot offer accurate information about primary origin of tumor in case of involvement of adjacent organs, so alternative diagnoses should be considered.

Scherl et al. reported that patients were misdiagnosed preoperatively with differentiated thyroid cancer with aggressive invasion of the trachea, by FNA biopsy and imaging study, while the postoperative final diagnoses were primary tracheal tumor [6]. FNA biopsy, used in the workup for thyroid cancer patients with abnormal findings on physical or radiologic examinations, has a high specificity of 90 to 99%, and results in a small, but relatively high false-positive proportion of 9.9% [7]. In our case, the patient was also misdiagnosed preoperatively as thyroid cancer based on imaging studies,

although FNA biopsy was not performed. We did not make an accurate diagnosis until evaluating the postoperative histopathologic results that identified specimens as primary tracheal tumor *i.e.* mucoepidermoid carcinoma.

In conclusion, although the tumor is regarded as aggressive thyroid cancer with tracheal invasion under imaging studies, we should consider the possibility of a primary tracheal lesion, especially in patients showing obstructive airway symptoms. That may be more helpful for surgeons and anesthesiologists to perform surgical planning and more targeted resection of the tumor for desirable prognosis.

Disclosure of conflict of interest

None.

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