

CASE REPORT

Case report of polymorphous low-grade adenocarcinoma in the trachea with metastasis to the right middle lobe bronchus

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Keywords

Adenoid cystic carcinoma; bronchus; misdiagnose; polymorphous low-grade adenocarcinoma; trachea.

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Received: 26 June 2014;

Accepted: 25 July 2014.

doi: 10.1111/1759-7714.12157

Thoracic Cancer 6 (2015) 220–223

Abstract

Polymorphous low-grade adenocarcinoma (PLGA) is a low-grade malignant infiltrative tumor of the minor salivary glands. According to data from PubMed (National Center for Biotechnology), one case of PLGA involving the left main bronchus and one case involving the right main and upper lobar bronchi have previously been reported. Here, we describe the first case of PLGA originating in the trachea with metastasis to the right middle lobe bronchus, all initially misdiagnosed as adenoid cystic carcinoma (ACC). It is particularly important to distinguish this tumor from other types of salivary gland tumors, especially ACC. Complete surgical excision is the curative treatment of choice for PLGA.

Introduction

Polymorphous low-grade adenocarcinoma (PLGA) is a low-grade malignant infiltrative tumor of the minor salivary glands.¹ It was initially described in 1983 as lobular carcinoma¹ and terminal duct carcinoma² of the minor salivary gland. The term *polymorphous low-grade adenocarcinoma of minor salivary glands* was coined in 1984 to reflect its morphological appearance and clinical behavior.³

According to data from PubMed (National Center for Biotechnology Information), one case of PLGA involving the left main bronchus,⁴ one case involving the right main and upper lobar bronchi,⁵ one case involving the lungs,⁶ and three cases of metastases to the lung^{7–9} have previously been reported.

Here, we report the first case of PLGA originating in the trachea with metastasis to the right middle lobe bronchus, all initially misdiagnosed as adenoid cystic carcinoma (ACC).

Case report

A 53-year-old woman presented to our hospital with a four-month history of intermittent hemoptysis and shortness of breath. Bronchoscopic examination revealed a 1.5-cm endobronchial mass that partly occluded the distal part of the right middle lobe bronchus and an operative scar below the glottis (Fig. 1a, b). A computed tomography (CT) scan and three-dimensional CT reconstruction of the chest confirmed a mass (Fig. 1c, d). Endobronchial biopsy revealed adenoid cystic carcinoma (ACC) (Fig. 2a). A right middle lobectomy and mediastinal lymph node dissection were performed. The patient recovered well after surgery. The mass to the cut margin of the bronchus measured 0.8 cm. Histopathologically, the bronchial cut margin was free from tumor infiltration, and there was no evidence of tumor invasion to the visceral pleura, the lymphovascular structures or the perineural structures. There were a total of six dissected lymph,

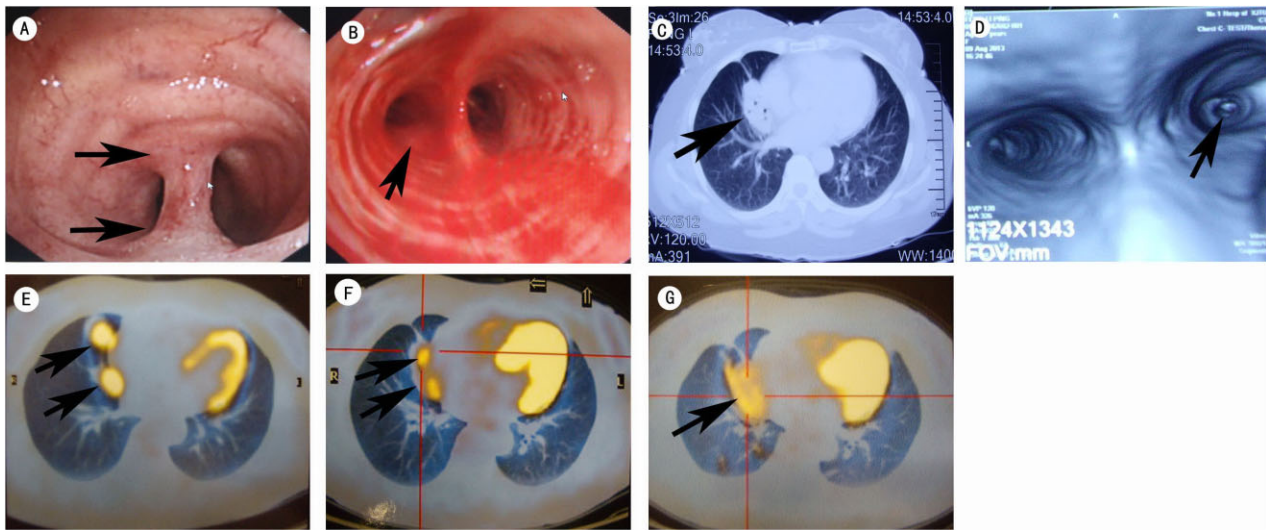


Figure 1 (a) Bronchoscopic examination revealed an operative scar below the glottis (abnormal healing after the tracheal tumorectomy). (b/d) Bronchoscopic examination and three-dimensional computed tomography reconstruction demonstrated a mass in the distal part of the right middle lobe bronchus. (c) A computed tomography CT scan showed atelectasis of the right middle lobe. (e-g) Positron emission tomography (PET)-CT scans (2010, 2012, 2013) demonstrated two masses in the medial segment of the right middle lobe.

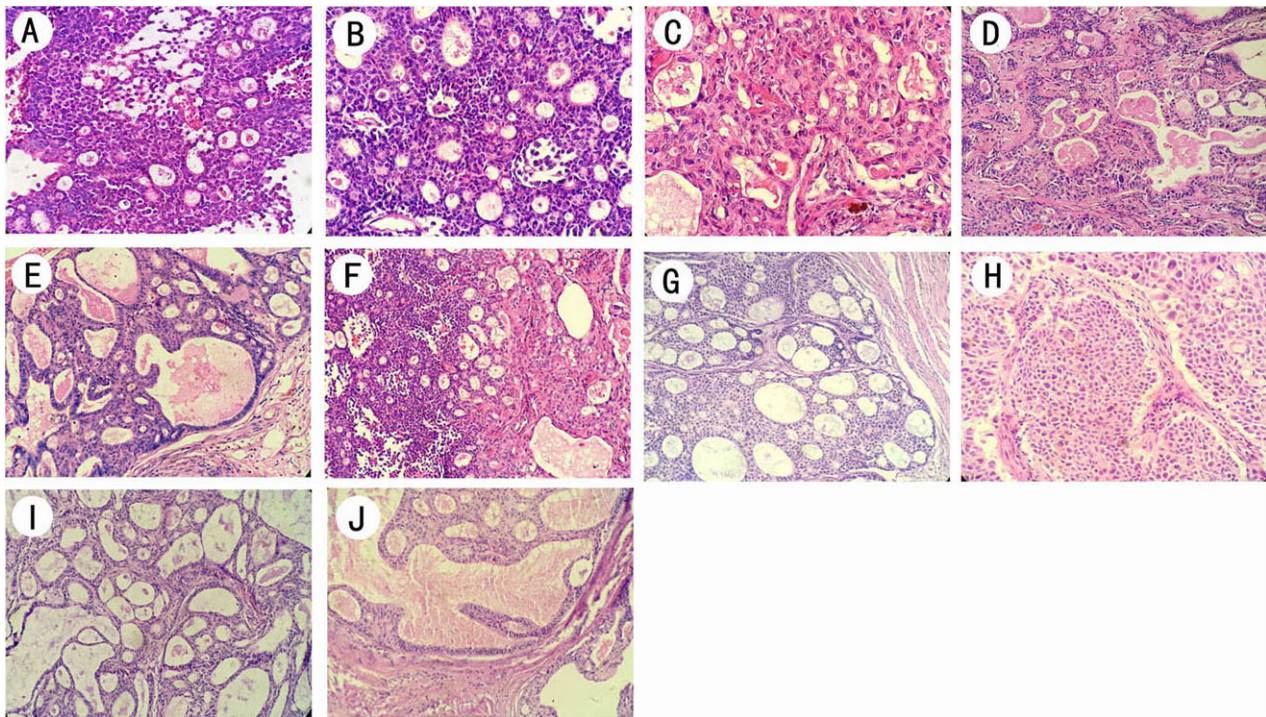


Figure 2 (a) Endobronchial biopsy of the right middle lobe bronchus. (b-f) Histopathological examination after right middle lobectomy. (b-e) The organizational structure of the tumor cells is diversiform, including solid, complex adenoid, papillary, and sieve-like structure. (f) The transitional region, from the cell enrichment region to the structurally diverse region. (g) Endotracheal biopsy. (h-j) Histopathological examination after tracheal tumorectomy.

tracheobronchial, and subcarinal nodes, and all were negative for tumor invasion. Postoperative pathology also revealed ACC (Fig. 2b). Positron emission tomography (PET)-CT scans, which were regularly performed and reviewed (2010, 2012, 2013), demonstrated two masses in the medial segment of the right middle lobe (Fig. 1e–g) with no abnormality at other sites.

Bronchoscopic examination revealed an operative scar below the glottis as a result of treatment performed 10 years earlier. The patient had been hospitalized with a one-year history of shortness of breath and a two-month history of blood in the sputum. A CT scan demonstrated a 1.7 cm cauliflower-like soft-tissue mass in the trachea at the level of the thyroid gland. An endotracheal biopsy revealed ACC (Fig. 2g). A tracheal tumorectomy with end-to-end anastomosis was performed. The patient recovered uneventfully after surgery. In terms of gross appearance, the mass was found to be 3 cm in diameter with a pedicle of approximately 2 cm × 1 cm. The microscopic finding revealed that the mass had invaded through the full thickness of the trachea wall. Histopathological examination after the operation also revealed ACC (Fig. 2h–j).

To determine whether the tumor of the right middle lobe bronchus was homologous with that of the trachea, hematoxylin and eosin staining of the bronchoscopic and postoperative specimens of the trachea and the right middle lobe bronchus were retrospectively analyzed, and immunohistochemical examination of all specimens were performed. The results revealed that the organizational structure of the tumor cells was diversiform, including solid, complex adenoid, papillary, and sieve-like structures. The nuclei of the tumor cells were round and uniform (Fig. 2c). Immunohistochemically, the tumor cells were all positive for cytokeratin (CK), CK8, periodic acid schiff, and alcian blue, and all negative for carcinoembryonic antigen, GCDF15, S-100, actin, smooth muscle actin, P63, NapsinA, and thyroid transcription factor-1. PET-CT was performed as blood in the sputum was again found four years ago. The blood in the sputum disappeared after another period of radiotherapy (60 CG). The final diagnosis was tracheal PLGA, therefore, the patient underwent periodic review.

Discussion

Primary tracheal tumors have various histological features. They can arise from the respiratory epithelia, salivary glands, and mesenchymal structures of the trachea.¹⁰ Tracheal tumors arising from salivary glands include pleomorphic adenoma, mucoepidermoid carcinoma, ACC and PLGA. Pleomorphic adenoma is benign, whereas mucoepidermoid carcinoma and ACC are malignant tumors and demonstrate more aggressive behavior compared to PLGA.

The tumor is locally invasive, with a low predilection for recurrence or lymph node metastases. The organizational structure of PLGA cells is diversiform, as demonstrated in this case. The nuclei of PLGA cells are slightly larger, rounder, and more uniform compared with those of ACC. The cytoplasm of PLGA cells is more eosinophilic than that of ACC.

Complete surgical excision is the curative treatment of choice for PLGA.^{10,11} A large study of 164 cases¹² showed that 97.6% of all patients were alive or had died of unrelated disease after a mean follow-up of 115.4 months following treatment with surgical excision only.

The characteristics of this case are as follows:

- 1 Regions of sieve-like structured cell enrichment (Fig. 2a, b, g–j), such as those found in ACC, mislead pathologists. The transitional region, from the cell enrichment region to the structurally diverse region (Fig. 2f), and the uniform nuclei confirm a diagnosis of PLGA (Fig. 2b–f). Immunohistochemical staining indicates that the tumors are homologous.
- 2 The final pathological diagnosis was tracheal PLGA and metastasis to the right middle lobe bronchus.
- 3 No PLGA in the trachea has been reported previously; this is the first reported case of PLGA originating in the trachea.
- 4 Castle *et al.*¹² argued that tumors localized to the hard palate were statistically significantly more likely to be associated with tumor recurrence/persistence or patient death, but not those in other anatomic locations. It is not well known whether PLGA in the tracheobronchus is recurrent or metastasizes.

Conclusion

This is the first reported case of PLGA originating in the trachea. Complete surgical resection is the treatment of choice. Whether adjuvant radiotherapy could be beneficial for treatment requires further study.

Disclosure

No authors report any conflict of interest.

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