Pulmonary artery stent for bronchial adenoid cystic carcinoma causing pulmonary artery stenosis

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Keywords
Bronchial adenoid cystic carcinoma, bronchoscopy and interventional techniques, lung cancer, pulmonary circulation and pulmonary hypertension, rare lung diseases.

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Received 02 December 2013; Accepted 10 December 2013

Respirology Case Reports 2014; 2(1): 39–41

doi: 10.1002/rcr2.42

Abstract
A 46-year-old woman presented with a 6-month history of dyspnea and weight loss on a background of previous pneumonectomy for bronchial adenoid cystic carcinoma 14 years beforehand. Several years prior to this presentation, she had developed left vocal cord palsy and a metastatic lesion to the right buttock confirmed at resection. Investigations included CT pulmonary angiography and right heart catheterisation and demonstrated pulmonary artery stenosis suggestive of vascular encasement, severe pulmonary arterial hypertension and pulmonary nodules. Subsequent pulmonary artery stenting markedly improved both pulmonary artery pressures and the patient’s symptoms. The diagnosis of pulmonary artery stenosis due to mediastinal infiltration by metastatic bronchial adenoid cystic carcinoma was based on these findings as well as the presence of the pulmonary nodules and the previous mediastinal (recurrent laryngeal nerve) and metastatic complications. This case is the first report of successful pulmonary artery stenting for this rare complication.

Introduction
Bronchogenic malignancies have the potential to invade and compress the mediastinal vessels, and acquired pulmonary artery stenosis can occur as a recognized complication. Most of these cases are due to non-small cell lung cancer. This complication of adenoid cystic carcinoma is far more rare, and to our knowledge, only one case has been reported previously [1].

We present the case of a patient with exertional dyspnea due to pulmonary artery stenosis with pulmonary hypertension, which presented 14 years after left pneumonectomy for a bronchial adenoid cystic carcinoma. Compression by recurrence had caused a severe focal stenosis of the right pulmonary artery, and stenting of this lesion resulted in significant clinical and hemodynamic improvement.

Case Report
A 46-year-old non-smoking woman presented with 6 months of progressive dyspnea on exertion associated with 3 kg of weight loss. She had undergone left pneumonectomy 14 years prior for bronchial adenoid cystic carcinoma; the bronchial resection margin was positive. She remained well until 10 years after surgery, when she developed left vocal cord palsy. A year later, she developed a painful lump in her right buttock, subsequently confirmed as metastatic bronchial adenoid cystic carcinoma after resection.

Examination revealed a loud pansystolic murmur, enhanced on inspiration, and signs consistent with previous thoracic surgery and left pneumonectomy. Computed tomography (CT) of the chest demonstrated a 2-cm pulmonary nodule anterior to the heart. Transthoracic echocardiography demonstrated severe pulmonary hypertension with the pulmonary artery systolic pressure (PASP) estimated at 110 mmHg. Right heart catheterization confirmed pulmonary hypertension with a significant gradient between the proximal and distal pulmonary artery suggestive of pulmonary artery stenosis. CT pulmonary angiography showed no evidence of pulmonary emboli but demonstrated marked enlargement of the right atrium, right ventricle, and proximal right (single remaining) pulmonary artery as well as a 2-cm...
stenosis extending 5 mm into the pulmonary artery with an appearance suggestive of tumor encasement (Fig. 1A).

Formal pulmonary angiography confirmed a focal stenosis just distal to the origin of the right pulmonary artery (Fig. 1B), with a peak gradient of 80 mmHg. This lesion was treated by balloon dilatation and placement of a 39-mm stent (Palmaz Genesis Stent; Cordis Corporation, Bridgewater, New Jersey) initially deployed on a 10-mm balloon. This stent was subsequently post-dilated with 14-mm and 18-mm balloons with a good final angiographic result and a mild degree of residual stenosis (Fig. 1C). Hemodynamics improved following stent insertion with the peak gradient reduced to 55 mmHg and PASP to 74 mmHg. Serum pro-brain natriuretic peptide fell from 3107 to 375 ng/L post stenting. Dyspnea and exercise tolerance improved, and the patient was able to resume work as a farmer.

The patient remained stable for six months until anorexia and weight loss developed. CT chest showed multiple new pulmonary nodules and new osteoblastic lesions in the thoracic vertebrae (Fig. 1D). Transthoracic echocardiography at that time estimated the PASP to be stable at 80 mmHg. Chemotherapy did not induce a response and the patient succumbed to metastatic disease several months later.

Discussion

Bronchial adenoid cystic carcinoma is a rare and slowly progressive disease. Recurrence is usually local and sometimes hematogenous [2]. Pulmonary artery compression due to mediastinal bronchogenic lung cancer [3] and other malignant processes [4] has been reported. Pulmonary artery stenosis caused by locally recurrent mediastinal bronchial adenoid cystic carcinoma has been reported only once previously [1], occurring five years after pneumonectomy. Successful percutaneous pulmonary artery stenting has been reported in the management of pulmonary artery compression due to other mediastinal malignant processes [3, 5], but not bronchial adenoid cystic carcinoma.

Figure 1. (A) Computed tomography (CT) pulmonary angiogram showing posterior displacement of the mediastinum and stenosis of right pulmonary artery. (B) Pulmonary angiography demonstrating right pulmonary artery stenosis (arrow). (C) Pulmonary angiography post stent insertion. (D) CT chest 12 months after pulmonary artery stenting showing evidence of widespread pulmonary nodules and the right pulmonary artery stent in situ.
This case presented with pulmonary hypertension due to pulmonary artery stenosis secondary to locally recurrent bronchial adenoid cystic carcinoma, in the context of proven hematogenous metastasis, positive bronchial resection margin, and two clinical phenomena consistent with mediastinal invasion: left vocal cord palsy and pulmonary artery stenosis. Pulmonary hypertension responded well to the insertion of a percutaneous pulmonary artery stent. To our knowledge, this is the second report of pulmonary artery stenosis due to recurrent mediastinal bronchial adenoid cystic carcinoma and the first report of successful pulmonary artery stenting for this complication.

Disclosure Statements

No conflict of interest declared.

Appropriate written informed consent was obtained for publication of this case report and accompanying images.

Acknowledgment

The authors acknowledge the support of the patient’s family and the consent given to submit this case for publication.

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