Oat Cell Carcinoma with Multiple Tracheobronchial Papillomatous Tumors*

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Multiple papillomas of the tracheobronchial tree are rare in children and adults. We describe a 42-year-old woman with multiple papillomatous-like tumors involving the trachea and bronchi. The limited tissue samples obtained during fiberoptic bronchoscopy initially lead to a benign histopathologic diagnosis; concurrent cytology studies strongly suggested oat cell carcinoma. The biopsy specimen obtained by rigid bronchoscopy ultimately demonstrated unequivocal oat cell carcinoma. This case illustrates an unusual and previously unreported manifestation of oat cell carcinoma—an endoscopic presentation in a papillary form suggesting benign tracheobronchial papillomatosis. Cytology studies were of increased diagnostic significance in this clinicopathologic setting.

Papillary tumors of the respiratory tract which occur most frequently during childhood are usually multiple and benign. In children the laryngeal area is the primary site for these tumors. Only rarely (2 to 3 percent) are papillomas found involving the lower respiratory tract.¹

In adults papillomas more commonly present as solitary lesions. They may, however, involve multiple areas of the lower respiratory tract. Approximately 50 percent of adult solitary papillomas become malignant epidermoid carcinomas.² In reviewing the literature, no other report of an association between oat cell carcinomas and papillomas was found.

The present report describes a patient with multiple polypoid tumors affecting the lower respiratory tract, without laryngeal involvement. An initial diagnosis of papillomatosis was based on the benign histology of the small tissue samples obtained endoscopically with fiberoptic bronchoscopy. Subsequent biopsies obtained during rigid bronchoscopy unequivocally demonstrated oat cell carcinoma.

CASE REPORT

A 42-year-old woman was referred to the University Hospital of the New Jersey Medical School with an abnormal chest roentgenogram showing atelectasis of the left lung (Fig 1).

The patient’s symptoms had begun with wheezing on exertion approximately ten months prior to the referral. Over a period of several months, her symptoms progressed, until she noticed wheezing even at rest. She also had a cough productive of white sputum and had pleuritic chest pain and fever. Symptomatic treatment had been given and had resulted in some improvement of cough and sputum production.

Two months prior to her admission, the cough worsened. She experienced moderate dyspnea which necessitated her quitting her job as a laboratory technician in an aluminum powder plant. She also began to have episodes of hemoptysis. Symptoms persisted despite antibiotic therapy, and her chest roentgenogram, which previously had been described as normal, now showed atelectasis of the left lung (Fig 1). She was referred to the University Hospital for evaluation.

The patient had a 20 pack-year (2 packs/day × 10 years) history of cigarette smoking, but had stopped using tobacco approximately five years prior to the onset of her symptoms. She also experienced a 35-lb weight loss over a period of one year, but stated that her weight loss was deliberate and related to dieting.

Physical examination showed her to be well-developed, mildly obese, oriented, and in no acute distress. Her vital signs were within normal limits. Pertinent physical findings were confined to the chest, where decreased expansion of the left chest was noted, with a dull percussion note. Breath sounds were decreased to absent, and fremitus was diminished.

The clinical impression was atelectasis of the left lung caused by bronchial obstruction. Fiberoptic bronchoscopy was performed and revealed a normal larynx, but multiple, soft, friable, polypoid lesions were observed involving the entire length of the trachea from a point just below the vocal cords to the carina (Fig 2). The left main bronchus was occluded by confluent tumors extending from the carina and left tracheal wall. A solitary polypoid tumor was also noted.

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FIGURE 1. PA and left lateral chest roentgenograms reveal atelectasis of the left lung. Note marked elevation of the left diaphragm as outlined by the gastric air shadow and the shift of the right border of the cardiac silhouette.
in the right intermediate bronchus. Multiple forcep biopsies of these tumors were taken. Histopathology of the biopsies was reported as papillomas with diffuse submucosal inflammation and granulation tissue with no evidence of malignancy. Bronchial washings for cytologic studies obtained at the time of bronchoscopy and processed according to the modified Papanicolaou method were reported as suspicious for carcinoma.2

Several weeks after her initial fiberoptic bronchoscopy, the patient underwent therapeutic rigid bronchoscopy, with removal of multiple tumors from the mid-trachea, and particularly the lower left trachea and carina, in a successful attempt to reestablish patency of the left main bronchus. Full reexpansion of the collapsed left lung resulted. Tissue obtained during this procedure was submitted for histologic examination, and the results supported the previous findings except that there was a marked crushed artifact. Cytology studies were not done.

Following rigid bronchoscopy, the patient’s symptoms improved dramatically. However, multiple sputa collected for cytologic study as an outpatient were reported as highly suspicious for malignancy. A repeated fiberoptic bronchoscopy was performed and again revealed polypoid lesions involving the lower trachea, carina, and right bronchial tree. The left main bronchus was markedly narrowed but patent. Cytologic evaluation of the brush biopsy smears of this narrowed bronchus was reported as being highly suspicious for a malignancy (oat cell carcinoma). The patient was again admitted to the hospital, where ptosis of the left eye developed, and she complained of diplopia. Her chest roentgenogram revealed prominence of the left hilum, and a CT scan and brain scan suggested metastatic brain lesions. A bone scan revealed an area of abnormal uptake where bone marrow biopsy specimens revealed foci of small cell carcinoma. Bronchial biopsies again were obtained through a rigid bronchoscope. At this time the diagnosis of oat cell carcinoma of the lung was histologically confirmed (Fig 3).

The patient was given a treatment protocol requiring radiotherapy and chemotherapy consisting of cytoxan, adriamycin, vincristine, and prednisone. This resulted in complete resolution of her symptoms, disappearance of the bronchial papillomatous-like lesions, and complete reexpansion of the left lung (Fig 4).

**DISCUSSION**

This case illustrates a very unusual presentation for oat cell carcinoma and demonstrates the value of cytologic evaluation of specimens in the diagnosis of lung carcinoma. No previous reports of an association between papillomatosis and small cell carcinoma of the lung could be found in the literature. The association of papillomatosis and lung cancer has been reported on many occasions, the carcinomas being the squamous cell type.4 Though the etiology of these papillomas is not completely understood, cigarette smoking and viral infections have been suggested as possible causes.

In contrast to the equivocal results that were obtained from histologic examination of tissue, the cytology studies consistently suggested a malignancy. It is clear that crush artifact of the small pieces of biopsied tissue in some instances prevent adequate histologic evaluation. It is also possible that the tissue samples from the polypoid lesions may not have contained tumor. The papillomas which were initially reported as being composed of edematous inflammatory tissue may have developed as a result of bronchial lymphatic obstruction. It seems reasonable to speculate that the initial biopsies sampled a secondary phenomenon, whereas the cytology specimens were more helpful because they sampled the lung more generally.

The diagnostic yield when bronchial carcinomas are endoscopically visible is greater from histologic examination of biopsy tissue than from cytologic specimens.5 Improved results, however, are obtained by combining both techniques. In a report by Martini et al,4 when bronchial carcinomas were endoscopically visible, a cancer diagnosis could be confirmed by tissue biopsy in 93 percent of patients, whereas adding results of cytologic studies increased this yield to 98 percent.

Small cell carcinomas are very delicate and prone to crush artifact, especially during fiberoptic bronchoscopy.7 It is evident that recommendations suggesting greater reliance on histopathologic studies rather than cytologic studies on lung cancer diagnoses do not apply in this case.
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REFERENCES


Systolic Anterior Motion of the Mitral Valve and Outflow Obstruction After Mitral Valve Reconstruction*

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A 62-year-old woman with acute mitral regurgitation due to ruptured chordae tendineae underwent mitral valve reconstruction. Postoperatively, subaortic outflow obstruction developed. Echocardiography revealed marked systolic anterior motion of the mitral valve. We did not find a similar case in the literature.

The most common etiology of subvalvular aortic stenosis is hypertrophic obstructive cardiomyopathy (HOCM). One of the associated echocardiographic findings in this disorder is systolic anterior motion (SAM) of the mitral valve, first described by Shah et al in 1969. Since then, SAM has been described in other disorders, including mitral valve prolapse (MVP)* and atrial septal defect* as well as hypercontractile states.* We report a case of SAM with a subaortic outflow gradient following a mitral valvuloplasty and annuloplasty.

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CASE REPORT

A 62-year-old woman was referred because of the recent onset of severe dyspnea. Other than being told of a "functional murmur" as a child, she had a history of excellent health and denied any history of cardiac disease. Four days prior to hospital admission, she noted the abrupt onset of severe shortness of breath. At no time did she experience any chest pain, and there was no history of recent dental work, fever, or infection. Physical examination revealed a regular pulse rate of 108/min, a blood pressure of 110/72 mm Hg supine, a respiratory rate of 20/min, and a temperature of 37.1°C. Chest examination was remarkable for dullness to percussion and decreased breath sounds at both bases and rales to the level of the scapula bilaterally. The cardiac apex was displaced laterally. The S1 was normal, S2 was physiologically split, and an S3 was present. A grade 3/6 blowing holosystolic murmur was best heard at the apex with radiation over the entire precordium; no diastolic murmur was present. An ECG revealed sinus tachycardia, left atrial enlargement, and nonspecific ST-T wave changes. Chest x-ray film showed pulmonary vascular congestion and bilateral pleural effusions.

M-mode echocardiography revealed dilation of both the left atrium (54 mm) and left ventricle (end-diastolic dimension of 65 mm); interventricular septal thickness was normal (8 mm). The left ventricular outflow tract width, measured from the point of mitral valve closure to the left side of the septum, was also normal (32 mm). There was marked systolic prolapse of the posterior leaflet (Fig I) suggestive of mitral valve prolapse and/or a flail leaflet. Two-dimensional echocardiography was consistent with a flail leaflet. Cardiac catheterization showed 4+ mitral regurgitation and no gradient between the left ventricle and aorta.

The patient was referred for mitral valve surgery two weeks after initial hospitalization. At the time of surgery, examination of the mitral valve revealed three ruptured chordae in the midportion of the posterior leaflet, and this area was excised. The medial portion of the anterior leaflet showed elongated chordae, one of which was shortened by approximately 6 mm, and an annuloplasty was then done using a No. 32 Carpentier-Edwards ring. After removal of the patient from cardiopulmonary bypass, there was no evidence of mitral insufficiency or of a gradient across the mitral valve. However,