Pathologic anatomic examination revealed incomplete resection of the tumor, so we decided to add postoperative radiotherapy. After 2 years, in March 1993, the patient was still alive and doing well without evidence of recurrence.

**DISCUSSION**

Regressive tetraplegia after tracheal resection is rare, and as far as we know, it has been mentioned only once in the literature. In neurosurgical and cardiosurgical procedures, it is a well-known complication. Especially in aortic reconstruction with prolonged cross clamping of the aorta or division of one or more intercostal arteries at the level of the lower thoracic vertebra, the blood supply of the medulla is jeopardized.

Our patient showed a complete loss of motor activity below level C7, except for the activity of the pelvic muscles. A possible explanation for this phenomenon may be the development of a relative stenosis of the cervical medullary channel as a result of the maximal anterior flexion of the neck. This could have caused a partial compression of the anterior spinal artery, which became symptomatic during the period of hypotension. On an MRI scan, a relative stenosis of the vertebral channel by spondylisis was seen on level C7. There was no compression of the medulla.

Obstruction of the anterior spinal artery occurs in several diseases, ie, embolization during or following aortic surgery, arteritides, aortic dissections, cervical spondylisis, cervical intervertebral disk hernia, syphilis, and compression by tumor.

In general, occlusion of the anterior artery leads to a loss of pain and temperature sensation below the level of the lesion in combination with a paralysis of motor function and a relative or absolute sparing of the proprioceptive sensation. The prognosis depends on the duration of occlusion and the existence of anastomoses.

In our patient, there was a decreased blood flow in the anterior spinal artery, probably caused by flexion of the neck in combination with hypotension, resulting in an isolated paralysis of the muscles below the involved segment. Theoretically this isolated symptom can be explained by the localization of the pyramidal tract, which is vascularized by the end branches of the anterior spinal artery, and in this way the most vulnerable structure in cases of decreased blood flow in the anterior spinal artery. Recovery is variable, but as shown in our patient, it can be complete.

**REFERENCES**


**Acute Airway Obstruction and Necrotizing Tracheobronchitis From Invasive Mycosis**

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Necrotizing tracheobronchitis and acute airway obstruction from invasive mycosis developed in a patient who had undergone bone marrow transplantation. The infectious process obstructed the airway and disintegrated the walls of the right mainstem bronchus and pulmonary artery. The airway was cleared using rigid bronchoscopy to extract the detritus. The patient died of hemorrhage after rupture of the pulmonary artery through the right mainstem bronchus.

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Immunocompromised patients may develop invasive aspergillosis, frequently with fatal results. The lung parenchyma usually is involved, but only rarely do patients develop ulcerative tracheobronchitis as a local manifestation of the infection. The patient described here rapidly developed distal tracheal obstruction and necrotizing tracheobronchitis from an Aspergillus infection after bone marrow transplantation.

CASE REPORT

A 20-year-old white man presented with weakness, fatigue, and shortness of breath. His white blood cell (WBC) count was 2,000/mm³. His hemoglobin level was 5 g/dl. His platelet count was 20,000/mm³. A bone marrow biopsy revealed hypocellular marrow with megaloblastic maturation of the red cell line and scattered normal-appearing megakaryocytes. The remainder of the evaluation, including chest x-ray film, blood chemistry values, and human immunodeficiency virus test, was normal. The patient's physical examination disclosed no further abnormalities, and the diagnosis was aplastic anemia of idiopathic origin. Bone marrow cytogenetics did not reveal any evidence of preleukemia.

Therapy with cyclophosphamide, 60 mg/kg, was begun 4 days prior to bone marrow transplantation. On the 4th day of this treatment, the patient received total lymphoid irradiation (7.5 Gy as a separate mantle abdominal field, inverted Y). Bone marrow from a human leukocyte antigen-identical relative was transplanted into the patient the next day. The patient was discharged 2 weeks later with a white blood cell (WBC) count of 5,800/mm³, a hemoglobin level of 9.1 g/dl, and a platelet count of 31,000/mm³; there were no signs of graft versus host disease. Immunosuppression was achieved with cyclosporine and steroids.

The patient was evaluated 3 weeks later for night sweats, shortness of breath, and throat tightness. The patient was afebrile; with clear lung fields bilaterally, no lymphadenopathy, and a soft abdomen without guarding. The chest x-ray film (Fig 1) and ECG were reportedly normal. The WBC count was 5,400/mm³ (absolute neutrophil count, 5,000/mm³—91 percent neutrophils; hemoglobin, 8.4 g/dl; and lactate dehydrogenase, 4,058 IU/L [normal range: 313 to 618 IU/L]). A ventilation-perfusion scan showed irregular perfusion defects from prior chemotherapy and radiation therapy. Oxygenation was not impaired. The patient was not treated for pulmonary emboli. Pyogenic bronchitis with fever developed with significant bronchial hyperreactivity. Treatment with bronchodilators and broad-spectrum antibiotics (vancomycin, 2 g every 12 h, and cefazidime, 2 g every 8 h) was begun for Gram-positive cocci and Gram-negative rods. A right lower lobe infiltrate developed 1 week later. Fiberoptic bronchoscopy revealed markedly inflamed distal tracheal mucosa and a mass nearly occluding the entire right mainstem bronchus. A biopsy specimen and brushings revealed the septated branching fungal hyphae of Aspergillus. Therapy with intravenous administered amphotericin B was begun (1 mg/kg/d).

There was no evidence of graft versus host disease; treatment with cyclosporine and steroids was tapered. Pulmonary function tests revealed forced vital capacity of 2.4 (94 percent), FEV₁ of 1.91 L (45 percent), and diffusing capacity for carbon monoxide of 11.9 ml/kg/min (33 percent of predicted values). No cytomegalovirus was present. Cultures of blood, bone marrow, and urine were negative for fungus and acid-fast bacilli. Legionella cultures were negative. Serologic tests to diagnose invasive Aspergillus were not done. Computed tomography of the chest (Fig 2) demonstrated necrosis of the bronchial wall and possible invasion into the right pulmonary artery. Because the mass invaded the distal trachea, right mainstem bronchus, and pulmonary artery, surgical resection (right sleeve pneumonectomy with carinal resection) was not recommended.

Maximum antifungal therapy was continued. The absolute neutrophil count was 8,700/mm³ (normal range, 1,700 to 7,500).

Ten days later, the patient experienced sentinel hemoptysis and stridor from acute airway obstruction. At bronchoscopy, an aspergilloma was noted in the left naris. Rigid bronchoscopy showed an aspergilloma that obstructed the distal trachea. Mucocoele or laser fulguration was not possible because of hypoxia. The mass (7.5×2×3.2 cm) was immediately removed. Fatal hemorrhage ensued from rupture of the right pulmonary artery wall into the proximal right mainstem bronchus.

COMMENT

The outcome of treatment of ulcerative tracheobronchitis or central airway aspergillosis is poor. Immunocompromised patients may develop disseminated pulmonary aspergillosis and, rarely, necrotizing tracheobronchitis. Antifungal agents may be helpful in treating patients with normal immune systems, but their value in treating immunocompromised patients is unknown.

Airway obstruction and necrotizing tracheobronchitis represent different stages of aspergillosis infection. Clarke et al reported nine patients with disseminated fungal infection and tracheobronchitis and proposed two morphologic patterns: (1) occlusion of the airway by pseudomembrane of necrotic tissue and hyphae, and (2) invasion of the adjacent lung parenchyma or pulmonary artery. Although the immunocompromised patient is at highest risk, other individuals with relatively less impairment also may
Figure 2. Computed tomography scan of the chest revealed destruction of the right mainstem bronchial wall and invasion of the pulmonary artery. The bronchial lumen was partially obstructed by the aspergilloma.

develop this fungal tracheobronchitis. Berlinger and Freeman described two immunocompromised patients who developed acute airway obstruction from an aspergilloma in the trachea. Transmural necrosis through the bronchus intermedius in one patient led to fatal hemorrhage from the pulmonary artery. The second patient had the aspergilloma removed but died 3 weeks later.

Baud et al noted one immunocompromised patient with central airway obstruction from Aspergillus plugs, who developed disseminated aspergillosis and died despite treatment with amphotericin B. Another patient with a normal immune system responded well to amphotericin B.

Factors that predispose to localized infection of the distal trachea and mainstem bronchus instead of the pulmonary parenchyma are unknown. Mucociliary clearance may be impaired in the patients who develop the former. Patients with a partially intact immune system may be able to control local spread of the infection and prevent airway occlusion by detritus and fungal components. With gradual enlargement of the aspergilloma, pressure necrosis occurs, causing local tissue destruction. When the bronchus or trachea overlying the pulmonary artery is infected, fatal hemorrhage may occur if the obstructing mass is manipulated.

Treatment with antifungal agents has been uniformly unsuccessful in immunocompromised patients, yet these agents represent the patient’s best treatment option for this systemic illness. Orally administered itraconazole has been used in the treatment of some forms of ulcerative tracheobronchitis following lung or heart-lung transplantation. Newer agents or aerosolized delivery of antifungal agents may be more effective. Surgery is recommended for the treatment of mechanical complications of this infection, such as persistent cavities, hemoptysis, or airway obstruction.

Immunosuppressed patients who develop central airway obstruction alone or with necrotizing tracheobronchitis from invasive fungi may have 100 percent mortality. Rapid diagnosis and initiation of antifungal therapy is needed to maintain airway control and optimize survival. Bronchoscopy should be performed at the first sign of shortness of breath. Biopsy, brushings, and cultures must be obtained for accurate diagnosis.

Surgical resection may be considered if the disease can be completely encompassed with the planned resection and the patient can tolerate such a procedure safely. Young patients without an underlying lethal disease (eg, malignancy, AIDS) may benefit from more aggressive treatment. Surgeons must consider the potential for massive hemoptysis when planning treatment for airway obstruction from Aspergillus.

REFERENCES


Pelvic Lymphangioleiomyomatosis*

Atypical Precursor to Pulmonary Disease

Jeffrey C. Ernst, B.S.; Roya Sohaey, M.D.;\(^{1}\) and Jeffrey M. Cary, M.D.

A 22-year-old woman presented with left pelvic pain and mass. Ultrasonography confirmed a multicentric left adnexal mass containing cysts of varying sizes. The patient had no pulmonary symptoms at the time of presentation. The mass was surgically excised and

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CHEST / 106 / 4 / OCTOBER, 1994 / 1267