Pulmonary Phaeohyphomycosis in a Patient with Hemoptysis*

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A 79-year-old retired schoolteacher had a history of bronchiectasis. She developed recurrent hemoptysis requiring multiple blood transfusions. *Exophiala dermatitidis* was cultured repeatedly from bronchial lavages. To our knowledge, this is the first documented case of isolated pulmonary phaeohyphomycosis due to *E. dermatitidis*, and it was successfully treated with amphotericin B and 5-flucytosine. (Ches 1989; 95:1158-60)

Phaeohyphomycosis is usually a chronic, slowly progressive infection caused by a variety of closely related "black" or dematiaceous fungi which are found in plants and soil. The infection is usually initiated by traumatic implantation into the skin. Generally, the organisms remain localized to the site of inoculation, but rarely hematogenous spread to distant organs (eg, CNS) occurs. Alternatively, the infection may be acquired by inhalation of the conidia with hematogenous spread to distant organs.

Phaeohyphomycosis of the lung presented in a patient with severe hemoptysis and no identifiable source of cutaneous infection. To our knowledge, this is the first published report of an unequivocally documented case of isolated pulmonary infection causing hemoptysis due to *Exophiala dermatitidis*, and it is the first infection due to this fungus which was successfully treated.

**Case Report**

A 79-year-old white woman (an unmarried retired schoolteacher) had a history of recurrent hemoptysis. A bronchoscopy in 1979 failed to demonstrate any cause of bleeding. Cultures of the secretions taken at bronchoscopy were negative for pathogenic microorganisms. Based upon the history, findings on bronchoscopy, and x-ray films, she was thought to have and was treated for bronchiectasis involving the lingula of the lung. She continued to have occasional episodes of hemoptysis. Past medical history was otherwise not significant. Prior to admission, she had had progressive weakness and weight loss for six months and had a cough with hemoptysis for two weeks.

On examination, at the time of admission on Nov 7, 1985, temperature was 37.0°C and respiratory rate, 28 per minute. She was intermittently expectorating bloody sputum. The remainder of the physical examination was unremarkable. The chest was clear to auscultation, and there was no significant lymphadenopathy or hepatosplenomegaly.

The chest x-ray film had an infiltrate in the left lower lobe and lingula and apiapleural thickening (Fig 1) and showed no major change from previous roentgenograms. On admission, hemoglobin value was 11 g/dl with normal indices; white blood cell count was 11,600/cu mm with a normal differential; glucose value was 110 mg/dl; and albumin level was 3.7 g/dl. Immunoglobulin levels were normal. The initial Mantoux test was 14 mm, but two subsequent tests had no induration.

On the night of admission, she had a syncopal episode with documented hypotension and questionable seizure activity. At bronchoscopy, there was profuse bleeding from the left lung, probably originating in the lingula. She was thought to have tuberculosis and was treated with isoniazid, ethambutol, rifampin, and multiple blood transfusions. However, two weeks later, a dematiaceous fungus grew from the specimen obtained at bronchoscopy. This was identified as *Exophiala dermatitidis* on the basis of morphology (Fig 2) and physiologic characteristics by both the Clinical Mycology Section at the National Institute of Allergy and Infectious Diseases and the Illinois Department of Public Health Laboratory. The antituberculosis medications were stopped. A repeat bronchoscopy was done on Nov 26, 1986, and subsequent cultures from the lavage of the lingula again grew *E. dermatitidis*. Direct microscopic examination of the lavage showed a moderate

**FIGURE 1. Infiltrate in the lingula and lower left lobe with pleural thickening over both apices.**

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The number of septate hyphae (Fig 3). She had a CT scan of the head and a lumbar puncture but both were unremarkable. She was treated with amphotericin B (40 mg IV three times a week for three months with a total dose of 1.536 g) and 5-fluocytosine (1.5 g po, qid for three months). The patient improved and had resolution of her cough and hemoptysis. She gained weight and strength and resumed normal activities. Sputum specimens from September of 1986 and 1987 were negative for fungi. As of June 1988, she has had no recurrence of hemoptysis or of symptoms suggestive of pulmonary infection.

**DISCUSSION**

Phaeohyphomycoses are cutaneous and systemic infections characterized by darkly pigmented, septate hyphae present in the tissue. They are caused by a heterogeneous group of imperfect black (dematiaceous) fungi which contain a cytoplasmic melanin-like pigment that appears to have etiologic significance by decreasing susceptibility of these organisms to antifungal agents.** Taxonomic characterization of these fungi is controversial, so the names given to these fungi are extremely variable and still changing as taxonomic criteria improve. Some commonly recognized species included in this group are *Exophiala dermatitidis* (synonymous with *Wangiella dermatitidis*), Cladosporium, Curvularia, and Drechslera species.

Phaeohyphomycosis involving infections of the paranasal sinuses, mucous and mucocutaneous membranes, subcutaneous tissues, and the central nervous system including brain abscesses, meningitis, and mycotic aneurysm have been reported. Reports of dematiaceous fungi of the respiratory system include the incidental finding of *Cladosporium cladosporoides* cultured at autopsy from a previously existing lung cavity and the isolation of various species from nasal septa and/or colonization of maxillary sinuses. These were all isolated infections, and hence, were probably acquired by inhalation of the fungi.

In a review of *E dermatitidis*, Hohl et al documented the first subcutaneous knee infection in the United States with *E dermatitidis*. Subsequently, Vartian et al reported endocarditis caused by this fungus in an intravenous drug abuser.

In our case of phaeohyphomycosis, *E dermatitidis* was repeatedly cultured from bronchial lavages of the patient who had presumed bronchiectasis but no other recognized predisposing illness. Usually patients with phaeohyphomycosis in sites other than the subcutaneous tissues are immunocompromised. However, in our patient, there was no evidence for diabetes or other condition associated with immunodeficiency. Focal lung disease from bronchiectasis probably predisposed the patient to this unusual infection.

To the best of our knowledge, this is the first reported case of isolated pulmonary phaeohyphomycosis caused by *E dermatitidis*. It was the most likely agent inciting severe hemoptysis. Hohl et al reported that medical treatment of infections due to *E dermatitidis* has failed in all patients, but our patient improved when treated with amphotericin B and 5-fluocytosine. She has now been followed for 2½ years with no recurrence of hemoptysis and no recurrence of symptoms of suggestive of pulmonary infection.
Corticosteroid-induced Myopathy and the Respiratory Muscles

Report of Two Cases

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Two women with connective tissue disease developed a characteristic steroid-induced myopathy. Reduced maximal transrespiratory pressures indicated reduced respiratory muscle strength. Gradual steroid dosage tapering resulted in prompt clinical improvement and marked increases in respiratory muscle strength, maximal inspiratory pressure increasing by 33 percent in one patient and by 70 percent in the other. This reversible steroid-induced respiratory muscle weakness may be of great significance in reconsidering long-term steroid therapy in patients with underlying lung disease.

Systemic corticosteroid therapy is known to cause many unwanted side effects, among which muscular weakness and wasting have been observed since its earliest days.1,2 Experimental steroid myopathy3-5 was shown to share many of the features of its clinical counterpart, which was extensively described by Afifi et al.4 A more precise definition of the biochemical abnormalities of this myopathy, a better correlation between clinical and laboratory findings, and a useful approach for early detection, differential diagnosis, and management were provided by Askari et al.7

Steroid-induced myopathy in the respiratory muscles, to our knowledge, has never been reported. We were fortunate to make observations on respiratory muscle strength in two patients with steroid myopathy and complaints of dyspnea and on the changes caused by progressive tapering of the steroid therapy.

CASE REPORT

Case 1

In January 1978 this 58-year-old woman developed typical skin lesions of dermatomyositis and was followed up as an outpatient in the dermatology department. Clinical and biochemical investigation was negative, and spirometric test results were within normal limits.

Corticosteroid treatment (50 mg of prednisone daily) was started in March 1978 and over the following years continuously adjusted and tapered.

In March 1986, however, steadily worsening disease activity (reappearance of pathognomonic dermatologic features) necessitated an increase in prednisone dose, up to 60 mg daily. For the first time the patient developed rapidly progressing muscle weakness and was referred to us because of prominent dyspnea. Her nutritional status was normal. Muscle enzyme values remained normal, but 24-h creatine excretions were clearly elevated (Fig 1). Pulmonary function test results revealed both notably decreased maximal inspiratory pressure, Pimax, being 52 percent predicted and maximal expiratory pressure, Pmax, being 40 percent predicted. After gradual reduction of the prednisone dose to 7.5 mg daily, the creatinuria substantially decreased, and both Pmax and Pmax increased by 33 percent.

Case 2

In May 1986 the diagnosis of "unidentified connective tissue disease" was established in this 50-year-old woman with chronic pylonephritis and secondary hypertension. Spirometric test results were normal. Prednisone treatment (50 mg daily for four weeks, gradually tapered to 30 mg) resulted in rapid regression of skin lesions and she was followed up as an outpatient in the division of nephrology.

After ten weeks of treatment, she complained of general fatigue, dyspnea on exertion, and muscle weakness. Serum enzyme values remained normal, but spirometric study results now showed clearly...