months without toxic effects, and cultures of sputum became negative after the fourth month. The patient's functional status stabilized, and she regained most of her lost weight. Unfortunately, she died from progressive pulmonary hypertension in June 1991.

**DISCUSSION**

Sporotrichosis is most often acquired as a primary infection from the inhalation of conidia. Two radiologic patterns are recognized: (1) involvement of the tracheobronchial lymph nodes; and (2) chronic cavitary disease. This patient represents an 8-year course of chronic cavitary disease with fever, cough, malaise, weight loss, and nodular apical masses associated with thin-walled cavities, fibrosis, and pleural thickening. Her smoking history is consistent with other reported cases and seems to confer susceptibility to infection. Although her history of exposure to tuberculosis was strong, she was repeatedly evaluated for tuberculosis over the 8-year period, and all cultures were negative.

Medical treatment of chronic cavitary pulmonary sporotrichosis has been unsatisfactory. Unlike the cutaneous form of the disease, SSK1 usually is ineffective. Amphotericin alone, even in doses greater than 2 g, has been associated with failure, especially if surgical resection cannot be performed. Most reviews emphasize that surgical removal of the infected tissue has proven to be the most effective therapy.

The use of azole antifungal agents for sporotrichosis has met with partial success for cutaneous disease but not for pulmonary disease. Calhoun et al10 reported that 8 of 11 patients with deep-seated sporotrichosis (no pulmonary cases) had a response to ketoconazole, 400 to 800 mg/day, when treated for more than 1 year. Dall and Salzman11 reported failure of ketoconazole in chronic pulmonary forms of the disease.

Itraconazole is an investigational triazole antifungal agent with in vitro activity against sporotrichosis at achievable serum levels. Restrepo et al treated 17 patients with lymphocutaneous disease with 100 mg/day for an average of 115 days. All had resolution of their disease. Lavalle et al12 and Borelli13 also reported success with itraconazole in cutaneous or deep subcutaneous disease. Baker et al14 reported a case of fungemia with an amphotericin B-resistant isolate of *S. scendent*. This was treated with itraconazole. We believe that this is the first case of chronic cavitary disease caused by *S. scendent* to be treated with itraconazole, with apparent microbiologic response, as gauged by the sterilization of sputum cultures, and a clinical response, as gauged by the patient’s subjective recovery and gain in weight; however, by the time she received this agent, irreversible lung damage had occurred, and end-stage pulmonary hypertension prevented subsequent attempts at cure by surgical resection. We advocate early use of itraconazole in the initial treatment of pulmonary sporotrichosis.

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**Respiratory Dyskinesia Presenting as Acute Respiratory Distress**

Matthew Ioanovich, M.D.; Russell Glantz, M.D.; Roger C. Bone, M.D., F.C.C.P.; and Peter Sizdom, M.D.

Incapacitating respiratory distress was the presenting manifestation of a choreaiform movement disorder. Because the patient also had asthma, respiratory distress was at first mistakenly attributed to this condition. Despite vigorous asthma management, there was no improvement. However, once the neurologic condition was recognized, use of specific therapy (haloperidol and reserpine) resulted in rapid and sustained remission of respiratory symptoms. *(Chest 1993; 103:314-16)*

A case of severe respiratory dyskinesia is reported, occurring in a patient who had asthma but who was unaware that he also had mild limb and trunk chorea. Involvement of respiratory muscles in chorea can lead to severe respiratory distress that can be effectively treated if drugs directed at correction of the movement disorder are employed. It is important to recognize this possibility to be able to institute specific therapy.

**CASE REPORT**

The patient is a 35-year-old black man who had a childhood history of asthma that did not require continuous medication and that disappeared during puberty. There was a history of minimal cigarette smoking and substantial alcohol and intravenous opiate use until approximately five years prior to hospital admission.

For the last three years, there had been multiple attacks of severe respiratory distress that were diagnosed as asthma and treated in community hospital emergency departments. The patient had been hospitalized and intubated endotracheally four times. He was known to have a hiatus hernia and took metoclopramide for two years prior to our examination. Following an episode of upper gastrointestinal tract bleeding and endoscopic demonstration of peptic esophagitis, he underwent plication of the fundus. Control of gastroesophageal reflux and its presumed role in exacerbation of asthma had been part of the rationale for the surgical intervention. During this hospital admission, progress notes indicated, at times, the presence of expiratory wheezing but, by large, chart entries emphasized...
rapid respiratory rates (30 to 50/min) and diminished breath sounds. Pulmonary function tests obtained before hospital discharge are shown in Table 1 (first evaluation) and demonstrate mild airways obstruction. Three weeks after discharge, while taking albuterol, ipratropium, beclomethasone as aerosols, and theophylline and prednisone (100 mg daily) orally, the patient experienced acute severe respiratory distress and was admitted to an intensive care unit at another institution. There was little or no change in respiratory distress after two days of conventional acute asthma management that included intravenous corticosteroids. Clinical signs of airways obstruction were not described. Roentgenographic evidence of lung hyperinflation was not present. The Pco₂ was 21 mm Hg and the Po₂ was 131 mm Hg while breathing O₂ at 2 L/min by nasal cannula. The patient was transferred to our institution for further care and to undergo phrenic nerve conduction studies, since the possibility of diaphragmatic weakness had been raised clinically.

At the time of hospital admission, the patient had a croupoid appearance, tachypnea, diminished breath sounds, no wheezing, and was described to have an otherwise normal physical examination. Respiratory recordings using an impedance plethysmograph (Resigraphe, NIMS, Miami, Fl) demonstrated bursts of fast, irregular breathing rates (Fig 1), but no evidence of abnormal thoracoabdominal motion. The neurologist who had been consulted to perform phrenic nerve conduction studies recognized choreiform movements of the head and trunk and the possibility that respiratory distress was due to respiratory dyskinesia. Diffuse mild chorea involving the limbs, head, and the oropharyngeal musculature was evident. Videofluoroscopy of swallowing documented choreiform movements of the tongue and oral pharynx that were typical of tardive dyskinesia. As a therapeutic/diagnostic trial, haloperidol (1 mg three times daily) and lorazepam (0.5 mg twice daily) therapy was started. Within one week, there was considerable attenuation of the choreiform movements. Respiratory rate diminished, and dyspnea was abolished. Pulmonary function tests again showed mild airways obstruction (Table 1) (second evaluation). Reserpine was substituted for haloperidol and lorazepam. At the time of hospital discharge, chorea was no longer discernible and treatment with all asthma medications had been tapered off. One month after hospital discharge, the patient continued to be free of dyspnea and used an albuterol inhaler occasionally for mild wheezing.

**DISCUSSION**

Respiratory dyskinesia is a term used to describe involvement of respiratory muscles in choreiform disorders. Most patients are not greatly impaired: they have spells of irregular breathing associated with involuntary grunting, gasping noises, or speech dysfunction. Less frequently, respiratory dyskinesia can cause disabling attacks of hyperventilation and severe, chronic respiratory alkalosis. The mechanism of hyperventilation seems to consist of involvement of the diaphragm in repetitive bursts of rapid, effective breaths. Respiratory dyskinesia, like other choreiform movements, disappears during sleep and tends to be exacerbated by subjective distress, emotions, or exercise.

Respiratory dyskinesia was a common sequela of epidemic viral encephalitis that occurred at the beginning of this century. In contemporary clinical settings, it is seen in patients with neuroleptic-induced tardive dyskinesia. The prevalence of respiratory muscle involvement in tardive dyskinesia is fairly large, approximately 5 percent in long-term neuroleptic users. The specific cause of the chorea in our patient is difficult to determine. Metoclopramide, which the patient took for two years, is a known cause of tardive dyskinesia but it is difficult to determine, in retrospect, the onset of the abnormality. Earlier episodes of respiratory distress may have been due to asthma.

Tardive dyskinesia is thought to result from postsynaptic dopaminergic receptor supersensitivity at the striatum due to long-term neuroleptic blockade of receptor sites. In established tardive dyskinesia, haloperidol reduces chorea because it causes dopaminergic receptor blockade. However, eventually this drug induces postsynaptic dopaminergic receptor supersensitivity and ultimately worsens the syndrome. A drug such as reserpine, which depletes the central dopaminergic concentration without causing dopaminergic receptor hypersensitivity, is preferred for long-term treatment.

The recognition of respiratory dyskinesia is not difficult when it occurs in settings in which it can be anticipated (eg, in institutionalized psychiatric patients taking neuroleptics). Our case is unusual because the degree of truncal and

**Table 1—Pulmonary Function Tests**

<table>
<thead>
<tr>
<th>Evaluation No.</th>
<th>FVC, L (%)</th>
<th>FEV/FVC, %</th>
<th>TLC, L (%)</th>
<th>FRC, L (%)</th>
<th>Dco, ml/min/mm Hg (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>4.2 (102)</td>
<td>71</td>
<td>5.9 (106)</td>
<td>2.12 (70)</td>
<td>26 (96)</td>
</tr>
<tr>
<td>2</td>
<td>3.6 (80)</td>
<td>72</td>
<td>5.7 (96)</td>
<td>2.36 (83)</td>
<td>27 (87)</td>
</tr>
</tbody>
</table>
cephalic chorea was mild in comparison to the intensity of respiratory muscle involvement, the presenting complaint, and because the patient also had asthma.

Increased awareness of the potential for respiratory dyskinesia to be confused with other causes of respiratory distress is necessary not only to avoid potentially harmful treatments, as exemplified in the present report, but to allow effective management to be instituted. One must keep in mind the spectrum of causes, in particular the use of neuroleptics, and the possibility that choreiform movements of limb or trunk may be mild and may not necessarily precede the onset of respiratory symptoms.

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Wegener's Granulomatosis Presenting as a Primary Seizure Disorder with Brain Lesions Demonstrated by Magnetic Resonance Imaging

K. Scott Miller, M.D., F.C.C.P.; and Joseph M. Miller, M.D.

Since the earliest attempt by Klinger in 1931 to describe the systemic vasculitis subsequently characterized in 1936 by Wegener as Wegener's granulomatosis, this disorder has been reported to present in a number of differing fashions. No previous description relates Wegener's presenting as a seizure disorder, and no reports of MRI of the brain in such patients exists. We relate such a case, along with MRI findings, prior to and after treatment, with a review of neurologic manifestations of the disorder.

(CHEST 1993; 103:316-18)

WG = Wegener's granulomatosis

Wegener's granulomatosis is characterized by widespread necrotizing granulomatous lesions and an associated systemic vasculitis. The disease usually involves the upper and lower respiratory tract and kidneys. Since its original description, multiple organ system involvement and alternative presenting manifestations have been described. Although seizures during treatment of WG have been recognized, no case of a patient initially presenting as a seizure disorder has been reported. This case report describes a patient seen primarily for a convulsion. The diagnosis was uncovered by CT and MRI, which revealed intracranial lesions thought to be the etiology of the seizures, and by open lung biopsy, which demonstrated the classic pathologic changes of WG.

CASE REPORT

A 34-year-old white man presented to the Trident Regional Medical Center after working outside all day in the summer heat. His original complaint to the triage nurse was fatigue and heat exhaustion. After a 30-min wait, a tonic-clonic seizure witnessed by the medical staff occurred and the patient was transferred to an examination room. A second seizure minutes later prompted administration of intravenous diazepam and phenytoin. He was then maintained on phenytoin without further seizures.

He had been in good health until eight weeks prior to admission, when gradual weight loss and a cough productive of foul-smelling yellow sputum, mixed occasionally with blood, was noted. Drenching sweats had occurred nightly for several weeks, and one hard shaking chill was noted four weeks prior to admission. A fall from a ladder had led to a fracture of the left hand three weeks before presentation. This was treated surgically with an uneventful recovery.

A history of chronic recurring headaches was obtained but they had not changed in character or frequency. Symptoms of sinusitis, arthritis, or prior neurologic problems were denied by the patient. A remote exposure to tuberculosis without prophylaxis was noted and he was an active smoker with a 40-pack-year history. He denied specific risk factors for HIV infection. Physical findings, including funduscopic and neurologic examinations, were within normal limits. The sinuses and lungs were without abnormality. The fingers were not clubbed. The white blood cell count was 14,400 without a left shift, and the hemoglobin value was 11 g/dl. The creatinine level was 0.7 mg/dl, and urinalysis disclosed 3 to 5 WBC and 10 to 15 RBC without casts per low-power field. Drug and alcohol screenings were negative. A left upper lobe thick-walled cavity without an air fluid level and a right middle lobe infiltrate and small cavity were

**Figure 1.** Admission posterior-anterior chest radiograph revealing left upper lobe and right middle lobe cavity lesions.