DISCUSSION

This case illustrates that an inflammatory laryngotracheostenosis may develop insidiously during a phase of apparent remission of Wegener's granulomatosis. The vocal chords and distal trachea were relatively spared, while the subglottic larynx and uppermost trachea were constricted by fibrosis, edema, and inflammatory mononuclear cell infiltrate. Owing to the slowly progressive nature of the constricting lesion, the patient was able to adjust his breathing pattern gradually without complaint, until a critical point of stenosis was reached.

The subglottic site was noted to be a target for Wegener's granulomatosis in 1954 by Godman and Churg, who showed that in two of their seven cases, "striking lesions of the larynx and trachea" were present consisting of "edema, congestion, and extensive ulceration of the mucosa, particularly in the subglottic area." In one of their cases, stridor occurred paroxysmally. The histologic picture typically lacks the necrotizing granulomas or vasculitis seen in other organs. The ulcerative lesions of the trachea and larynx seen in up to 30 percent of untreated cases seem to be infrequent in the era of immunosuppressant treatment. Rarely, in a case of active disease, the larynx, trachea, and bronchi may be diffusely affected with ulcers, infiltration, and cicatricial stenotic lesions. Hypothetically, the granulomatous stenosis seen in our patient may represent smoldering disease activity, which in the untreated state would have resulted in a florid ulcerative lesion.

It is unlikely that the subglottic lesion results from any cause other than Wegener's granulomatosis itself. Histologic and bacteriologic studies have failed to identify likely infectious agents. Relapsing polyarthritis, amyloidosis or sarcoid, conditions linked rarely to laryngostenosis, were not in the clinical picture. Posttraumatic stenosis caused by cicatrized granulomas induced by endotracheal intubation is unlikely, since this typically occurs within three weeks of extubation, and in most cases the intubation has lasted longer than 24 hours. However, in a few cases in which steroids or immunosuppressants were administered (not for Wegener's granulomatosis), there developed a delayed fibrous stenosis of the larynx as late as five months after extubation. Several cases of subglottic stenosis have occurred in Wegener's granulomatosis in the absence of a history of intubation, occasionally as a presenting finding.

This type of subglottic lesion may be seen more frequently in the future as patients with Wegener's granulomatosis live longer in immunosuppressant-induced remissions. The case report illustrates the need for vigilance in discovering and monitoring target sites during and after treatment of this disease.

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Myocarditis in Legionnaires' Disease*

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A case of Legionnaires' disease is described in which the characteristic features of multilobar pneumonia, rhabdomyolysis, renal failure, hepatic and CNS involvement are accompanied by the previously undescribed complication of myocarditis. Clinical and laboratory findings of myocardial involvement included overt heart failure, a new gallop, an abnormal ECG, elevated myocardial specific enzymes and an abnormal thallium scan. All of these abnormalities resolved completely after recovery.

Legionnaires' disease is usually associated with multiorgan involvement, including pneumonia and renal and hepatic abnormalities. We present a case that included myocarditis, a complication not previously described to our knowledge.

CASE REPORT

A 51-year-old woman factory worker was hospitalized in May 1979 with complaints of fever, multiple rashes, myalgias, weakness, nausea and nonproductive cough. She had been in

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Figure 1. Serial ECGs. Second week, normal QRS complexes with prominent U waves and QT prolongation. Fifth week, marked ST-T abnormalities, with T wave inversions in anterior-lateral leads. By eighth week, ECG had returned to normal.

good health until three days earlier. There was no history of recent travel or exposure to air conditioning or to construction sites. She smoked one pack of cigarettes per day for 30 years and drank moderately on weekends.

On admission, the blood pressure was 98/70 mm Hg; pulse, 120 beats per minute and regular; respiratory rate, 28 breaths per minute; and temperature, 40.7 °C. The sclera were mildly icteric. Signs of consolidation were present at the left lung base. The result of cardiac examination was normal, without gurps or murmurs. The total liver span measured 14 cm at the midclavicular line. Severe proximal muscle weakness was noted. Initial laboratory studies included a hemoglobin of 13.2 g/dl; hematocrit, 40 percent; WBC count, 9,700/dl, with 50 percent polymorphonuclear leukocytes, 41 percent bands, and 9 percent lymphocytes. Serum sodium level was 128 mEq/L; potassium, 3.3 mEq/L; BUN, 17 mg/dl; creatinine, 2.0 mg/dl; SCOT, 570 μ/ml; lactic acid dehydrogenase (LDH), 1503 μ/ml; alkaline phosphatase, 59 μ/ml; bilirubin, 3.8 mg/dl; and phosphorus, 1.9 mg/dl. The blood pH was 7.52; Pco2, 31.7 mm Hg; and Po2, 42 mm Hg. The ECG showed sinus tachycardia, but was otherwise within normal limits. A dense alveolar infiltrate in the left lower lobe was present on chest roentgenogram. Sputum and blood cultures showed no growth.

Therapy was initiated with oral erythromycin and cephalothin. On the third hospital day, the patient became delirious and oliguric. Lumbar puncture results were normal. Urinalysis revealed 2+ protein, 4+ occult blood, many WBCs, pigmented casts, and three to four RBCs per high-power field. Urine sodium level was 84 mEq/L. The serum was of normal color. Serum aldolase was 200 μ/ml, and creatine phosphokinase (CPK) was 13,650 μ/ml, with an MB fraction of 4,200 μ/ml. Cardiac LDH isoenzymes were notably increased. After one week, the patient’s ECG showed new QT interval prolongation and prominent U waves (Fig 1). The hematocrit decreased to 25 percent by the seventh day, and then stabilized at 17 percent. There was no evidence of blood loss or hemolysis. The patient also experienced diarrhea for several weeks.

The patient remained oliguric and underwent peritoneal dialysis twice. Gram-negative peritonitis developed, necessitating treatment with amikacin and chloramphenicol. Congestive heart failure and bilateral transudative effusions developed despite adequate fluid removal during dialysis. On the 18th day, erythromycin was increased to 4 g intravenously per day. On the 34th day, a prominent S4 gallop was noted and the ECG disclosed deep T wave inversions in the anterolateral leads (Fig 1). The patient complained of precordial pressure, but the cardiac enzymes continued to decrease. Over the next three weeks, the urine output increased, and azotemia resolved. A resting thallium perfusion scintigram showed patchy defects in the inferolateral areas of the heart (Fig 2). By the eighth week, her ECG normalized. After five months, the patient is asymptomatic except for continuing

Figure 2. Resting thallium scintigram in anterior (ANT) and left anterior oblique (LAO) views. Myocardial perfusion defects (arrows) in inferior and posterolateral walls.
myalgias. Initially, Legionnaires' titers in the patient's blood were 1:64, but three weeks later they were 1:2048.

**DISCUSSION**

This patient had most of the characteristic clinical and laboratory findings of Legionnaires' disease, including multilobar pneumonia, rhabdomyolysis, renal failure, hepatic and CNS involvement, hypophosphatemia, and hyponatremia. Clinical myocarditis has not previously been described, although there is an autopsy report of inflammatory infiltrates of the heart.3

Our patient had an early increase of the muscle enzymes, aldolase and CKP, with great increases of the cardiac specific enzymes CPK-MB and LDH 1 and 2. In addition, there were ECG ST-T repolarization changes and QT prolongation. She had chest pain and heart failure, a gallop rhythm, and had abnormalities on thallium scan. These findings suggested the clinical diagnosis of myocarditis.

ECG changes have been described in two patients with Legionnaires' disease.7 Myocarditis may be associated with many viral, bacterial, fungal, and parasitic infections.4 Often, the only clues are the presence of nonspecific ECG changes, inappropriate tachycardia, atrial or ventricular irritability, and gallops. Heart failure may be overt or covert.4,6 It is important not to erroneously diagnose ischemic heart disease. Our patient had no arrhythmia or conduction blocks. We do not believe that she had a myocardial infarction, since her ECG normalized after eight weeks without persistent ST-T abnormalities or Q waves. In addition, the temporal dissociation between the enzymatic and ECG changes is not suggestive of myocardial infarction. Furthermore, the defects on the thallium scan were more patchy than that normally seen in ischemic heart disease and resolved partially after convalescence.

We conclude that the spectrum of Legionnaires' disease may include myocarditis.

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**Heparin-Induced Thrombotic Thrombocytopenia**

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Following subcutaneous therapy with heparin, the patient developed signs and symptoms of vascular occlusion in both legs. This was accompanied by thrombocytopenia and platelet aggregation when the patient's platelets were incubated with heparin. The clinical features and implications of this syndrome are discussed.

**H**eparin-induced thrombocytopenia is a well-recognized phenomenon.3 Marked thrombocytopenia induced by heparin, with life-threatening thrombotic complications attributable to platelet aggregation, is a rare occurrence and not widely appreciated.5,2

**CASE REPORT**

The patient was a 66-year-old man admitted to Maimonides Medical Center, Brooklyn, NY, because of severe substernal pressure in the chest and pain in the left arm of several hours' duration. The patient's history was not contributory, except for 40 pack-years of smoking. Physical examination on admission revealed blood pressure of 90/60 mm Hg and a heart rate of 80 beats per minute. There was no distention of the veins in the neck at 45°. The lungs were clear. A fourth heart sound was heard. The peripheral pulses were intact except for absent dorsalis pedis pulses bilaterally. The electrocardiogram revealed acute anterior wall myocardial infarction.

The patient was admitted to the coronary intensive care unit and was treated with prophylactic administration of lidocaine and subcutaneous injection of heparin (5,000 units every 12 hours). He was treated for resistant ventricular arrhythmia with procainamide hydrochloride (Pronestyl; 500 mg orally every four hours). On the 11th day after the myocardial infarction, therapy with heparin was discontinued. On the 13th day the patient complained of soreness in the toes of his left foot. No objective findings were present. On the 15th day the pain in his left foot intensified. There was transient loss of the left posterior tibial pulse with persistent cyanosis and coolness of the lateral aspect of the left foot and the small toe of his right foot (Fig 1). At this time, full therapy with heparin was instituted, with 5,000 units given intravenously every four hours. Drug-induced digital vasculitis was considered, and therapy with procainamide hydrochloride was discontinued. The cyanosis and severe pain did not abate. The platelet count on the fifth day was 242,000/cu mm and on the 16th day was 62,000/cu mm.

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