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Recurrent Pneumonia in Relapsing Polychondritis

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RELAPSING POLYCHONDRITIS is a systemic disease characterized by recurrent inflammatory lesions of cartilaginous structures throughout the body. Inflammation typically involves the external ear, nose, eyes, larynx, trachea, ribs, and joints.[1-3] Respiratory tract involvement is common. Secondary infections of both upper and lower parts of the respiratory system can occur. It is unusual for pneumonia to be the presenting symptom, but in the case we describe, recurrent unexplained pneumonia was the key to the diagnosis.

Report of a Case

The patient, a 38-year-old man, was admitted to R. E. Thomason General Hospital, El Paso, Texas, because for three days he had had cough, pleuritic chest pain, dyspnea on exertion, subjective fever, chills, and a discharge from the left ear. He admitted to hoarseness of his voice for several years and mild knee pain without swelling, especially after walking long distances. There was no history of redness, pain, or swelling of his ears or nasal septum nor any dryness of his eyes or mouth. His medical history was notable for insulin-dependent diabetes mellitus and five previous admissions in the past two years for community-acquired pneumonia (Table 1).

On admission the patient's blood pressure was 100/67 mm of mercury, pulse rate was 112 beats per minute, temperature was 36.8°C (98.2°F), and the respiratory rate was 20 per minute. His left ear showed a purulent discharge. There was no laryngotracheal swelling or tenderness present. Bronchial breath sounds were audible in the right mid-lung field. A 2/6 decrescendo diastolic murmur was heard in the aortic area. There was no evidence of joint tenderness or swelling.

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Laboratory tests showed a leukocyte count of 11.2 × 10^9 cells per liter (11,200 per mm^3) with 0.75 (75%) segmented forms and 0.19 (19%) bands, a hemoglobin level of 149 grams per liter (14.9 grams per dl), and a hematocrit of 0.442 (44.2%). The serum glucose level was 30.6 mmol per liter (552 mg per dl), with serum ketones present in 1:8 dilutions. A urinalysis was normal. A chest roentgenogram showed an infiltrate in the left upper and lower lobes and right middle lobe. Gram's stain of the sputum showed mixed bacteria, and cultures of specimens from tracheal aspirate and broncheoalveolar lavage did not grow any bacteria. Gram's stain of a specimen of the ear discharge showed gram-positive cocci in pairs; culture grew *Streptococcus pneumoniae.* He was treated with a regimen of intravenous fluids, insulin, and cefuroxime.

The finding that the patient had several episodes of pneumonia prompted a further workup. Bronchoscopy showed laryngeal and perilaryngeal edema, inflamed vocal cords, thin translucent tracheobronchial mucosa, and pronounced tracheobronchial collapse on coughing. A bronchial biopsy specimen showed chronic inflammation. On computed tomographic scan of the thorax, he had a minimal infiltrate in the right middle lobe, without any evidence of tracheal stenosis or collapse. Echocardiogram showed a large aortic root with aortic root sclerosis and mild aortic and mitral regurgitation. On ophthalmologic evaluation there was an old uveitis. An erythrocyte sedimentation rate was 100 mm per hour. A test for anti-SS-A (Ro) antibody was positive. Tests for antinuclear antibody, anti-SS-B (La) antibody, rheumatoid factor, the human immunodeficiency virus, HLA-B27, and a VDRL test were negative. The results of complement studies—CH50, C3, and C4—quantitative immunoglobulins, and serum protein electrophoresis were normal. Sinus series and a roentgenogram of sacroiliac joints and the lumbosacral area were normal. Pulmonary function tests showed a mild restrictive pattern.

A diagnosis of relapsing polychondritis was made on the basis of the tracheobronchial thinning and collapse during bronchoscopy and by the presence of otitis media, uveitis, and aortic root dilatation. Steroid therapy was offered and discussed with the patient, but he declined.

Discussion

Relapsing polychondritis is a rare disease of unknown cause. An autoimmune pathogenesis has been supported

TABLE 1.—*Repeated Hospital Admissions for Pneumonia*

Date	*Location of Infiltrate on Roentgenogram*	*Organism Identified in Sputum Specimen*
4/8/91	Right peribronchus	Specimen not collected
10/15/91 ..	Both lungs, patchy	*Staphylococcus aureus*
12/13/91 ..	Left lower lobe	*Haemophilus influenzae,* biotype 2
2/10/92 ...	Left lower lobe	*H influenzae,* biotype 3
12/4/92 ...	Left basilar and right cardiophrenic angle	*S aureus*
2/21/93 ...	Left upper and lower lobes and right middle lobe	None in sputum, but *Streptococcus pneumoniae* in middle ear

TABLE 2.—*Presenting Symptoms or Signs in 159 Patients With Relapsing Polychondritis**

Signs or Symptoms	*Patients, No.*	*Percentage*
Auricular chondritis	35	26
Arthritis	32	23
Ocular inflammation	19	14
Respiratory tract involvement	19	14
Nasal chondritis	18	13
Audiovestibular damage	8	6
Miscellaneous	6	4

*Adapted from McAdam et al.[6]

by the presence of antibodies to type II collagen in two thirds of patients.[4] The syndrome was first described in 1923 by Jaksch-Wartenhorst.[5] The diagnosis of relapsing polychondritis can be made on clinical grounds when at least three of the following six features are present[6]: bilateral auricular chondritis; nonerosive, seronegative, inflammatory polyarthritis; nasal chondritis; ocular inflammation; respiratory tract chondritis; and audiovestibular damage. Common clinical features of relapsing polychondritis are shown in Table 2.

Respiratory tract involvement occurs in as much as 50% of patients as a result of compromise of the cartilage of the laryngotracheobronchial tree and is the leading cause of death.[6] Lower airway disease may be asymptomatic and may be found only on tomography and pulmonary function testing.[7] All reported cases of subglottic strictures[7] and tracheal stenosis[2] were symptomatic. The most common symptoms in relapsing polychondritis indicating airway involvement are breathlessness, hoarseness, cough, stridor, wheezes, and tenderness over the laryngotracheal tree; these tend to occur later in the illness and are harbingers of increasing morbidity.[8]

Many associated diseases have been described among patients with relapsing polychondritis.[1,3,6] Our patient had insulin-dependent diabetes mellitus and a positive SS-A test without xerostomia or xerophthalmia. There are no specific laboratory abnormalities diagnostic of relapsing polychondritis. An increased sedimentation rate, anemia, and leukocytosis have been consistently reported during active disease. The evaluation of respiratory tract symptoms should include conventional radiography, computed tomography, pulmonary function tests including flow-volume curves, and bronchoscopy.[8] The mainstays of treatment include the administration of parenteral steroids, nonsteroidal anti-inflammatory agents, and cytotoxic drugs and the surgical treatment of complications.

Airway involvement in relapsing polychondritis can occur with or without minimal ear and nasal involvement.[9] Chest physicians should be aware of the possibility of relapsing polychondritis in patients presenting with recurrent pneumonia; this syndrome has not been described previously. We considered the diagnosis of middle lobe syndrome, immunoglobulin deficiency, and recurrent aspiration until the diagnosis of relapsing polychondritis was made.

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