most recovered completely. Previous reports have not emphasized respiratory symptoms as a manifestation of babesia infection. However, it has been described with *P. falciparum* malaria and may range from minimal upper airway complaints to fatal ARDS. Two forms of ARDS in malaria have been described. One is secondary to volume overload following aggressive fluid replacement, while the other appears to be related to increased capillary permeability. The patient described developed acute pulmonary edema in the presence of normal pulmonary capillary wedge pressure and cardiac index, suggesting that the cause of the pulmonary edema was secondary to an increase in capillary membrane permeability similar to patients with malaria and ARDS.

The possible mechanisms for ARDS in both malaria and babesiosis remain speculative. It has been suggested that the increased vascular permeability may be secondary to microemboli, disseminated intravascular coagulation and immune complex disease. Experimentally, the effects of babesiosis have been likened to those of endotoxin shock, and injury to vascular endothelium has been demonstrated. Except for its presence in red blood cells within the pulmonary vasculature, direct infestation of the parasite in lung tissue or pleural fluid does not seem to play a role.

In summary, this case presents a previously undescribed complication of infection with *Babesia microti*, acute respiratory failure due to noncardiogenic pulmonary edema. For most patients, babesiosis is a self-limiting febrile illness of varying severity. In splenectomized patients, however, the illness can be acute and more fulminant. Although our patient had an intact spleen, she appears to have fallen into this latter group.

As awareness of this disease increases, the number of patients diagnosed with babesiosis will probably increase. This case highlights the need for careful monitoring of chest roentgenograms, arterial blood gases and, if indicated, pulmonary artery wedge pressure measurement in patients with symptomatic infection due to *B microti*.

REFERENCES


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Lymphomatoid Granulomatosis Presenting as Central Neurogenic Hyperventilation*

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A patient with lymphomatoid granulomatosis and focal transformation to lymphoma limited to the central nervous system presented with severe central neurogenic hyperventilation. The hyperventilation resolved as the underlying pathologic condition was treated with prednisone and cyclophosphamide.

Central neurogenic hyperventilation (CNH) has been defined by Plum and Swanson as a syndrome comprising normal arterial oxygen tension (PaO₂), decreased arterial carbon dioxide tension (PaCO₂), and respiratory alkalosis in the absence of cardiac or pulmonary abnormalities that would stimulate a compensatory hyperpnea. We report here an awake patient with CNH associated with lymphomatoid granulomatosis (LC) confined to the central nervous system.

**Case Report**

A 41-year-old white male electrician presented to his local hospital with a three-day history of uncontrollable rapid breathing. His past medical history revealed a grand mal seizure one year prior to this admission which was not evaluated. At the outside hospital, the patient was awake and alert. Arterial blood gas levels breathing room air revealed pH, 7.62; PaO₂, 120 mm Hg; PaCO₂, 7 mm Hg with a bicarbonate of 12 mEq/L. His white blood cell (WBC) count was 26,600/cu mm with a hemoglobin of 17.1 g/dl. The patient underwent a lumbar puncture which revealed glucose 52 mg/dl, protein, 90 mg/dl, and 15 cells (87 percent lymphocytes). Computerized tomography (CT) of the brain revealed multiple small contrast enhancing lesions. He was treated with chloramphenicol and tobramycin. Three days later, the patient was found to be confused and his mentation was slow. He was transferred to the University Medical Center for further evaluation.

Physical examination revealed a temperature of 36.7°C with a pulse rate of 85 beats per minute and a blood pressure of 110/70 mm Hg. The patient was found to be breathing rapidly with a respiratory rate of 26 per minute. He was awake and alert but had a flat affect. He was oriented to person, but he had a decrease in immediate memory. His coordination was poor. The reflexes were all intact and four plus bilaterally. His plantar reflexes were flexor bilaterally. His admission chest roentgenogram was normal. Arterial blood gas levels breathing room air revealed pH, 7.55; PaO₂, 113 mm Hg, and PaCO₂, 16 mm Hg. A repeat spinal fluid examination revealed 10 WBCs (100 percent lymphocytes), protein, 109 mg/dl; glucose, 44 mg/dl; and a negative VDRL. Sedation with morphine sulfate failed to control the hyperventilation. Repeat blood gas values obtained 24 hours later revealed pH, 7.67; PaO₂, 124 mm Hg; PaCO₂, 6 mm Hg; bicarbonate, 7 mEq/L; and the anion gap was 11 mEq/l. Repeat chest roentgenogram was normal. Because of the severe alkalosis, the patient was intubated, mechanically ventilated, and paralyzed with pancuronium bromide. A CT scan of the brain revealed decreased left frontal density with diffuse irregular contrast enhancement of both hemispheres that was greatest in the frontal areas (Fig 1). No mass

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glioncentric, angiodestructive infiltrate of atypical lymphoreticular cells predominantly affecting the lungs, skin, central nervous system, and kidneys that was first described in 1972. Disease presenting in or confined to the central nervous system is rare, although central nervous system involvement occurs in 23 to 30 percent of patients with LG. The disease process can evolve into a malignant lymphoma if not vigorously treated with a combination of cyclophosphamide and prednisone. Combination chemotherapy of the resultant malignant lymphoma with several drug regimens has been unsuccessful in achieving remission. A recent reevaluation of the syndrome has suggested that a diagnosis of malignant lymphoma or malignant lymphoproliferative syndrome can be made instead of lymphomatoid granulomatosis if multiple samples of tissue are examined.

Central neurogenic hyperventilation has been reported in association with primary brainstem astrocytoma, and malignant cerebral reticulosis. Our review of the literature failed to reveal a similar case of central hyperventilation due to lymphomatoid granulomatosis of the central nervous system with or without transformation to lymphoma.

Acute respiratory alkalosis decreases cerebral blood flow, shifts the oxygen hemoglobin dissociation curve to the left, and increases anaerobic metabolism of glucose in the brain. Our patient remained awake despite a PaCO₂ of 6 mm Hg and pH of 7.67. Our patient continued to hyperventilate in spite of sedation and required total paralysis to correct his severe respiratory alkalosis. The hyperventilation resolved, and the patient was successfully weaned off the panceurion and the respirator after eight days of cyclophosphamide and prednisone therapy for lymphomatoid granulomatosis.

The histologic findings in our patient revealed lymphomatoid granulomatosis. Bone marrow examination and CT scans of the chest and abdomen showed no evidence of lymphoma. The initial CT scan of the brain (Fig 1) revealed contrast enhancement similar to that of a case reported previously by Sackett et al. One patient with systemic LG who developed a central nervous system immunoblastic sarcoma five months after initial diagnosis has been reported.

Plum and Swanson, in their analysis of nine patients with...
Conduction Disturbance In Behcet's Disease* 

Association with Ruptured Aneurysm of the Sinus of Valsalva into the Left Ventricular Cavity

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Cardio-Behcet's disease is not often reported; only one case to our knowledge has described complete atrioventricular (AV) block, without pathologic details.1 We document, from direct observation during surgery and pathologic examination of associated lesions, a case whose conduction disturbance was possibly caused by direct extension of Behcet's disease itself into the conduction system.

Behcet's disease, which was originally described by Halusi Behcet in 1937,2 is a generalized chronic inflammatory disease characterized by recurrent oral and genital ulcerations and dermal and ocular manifestations. Involvement of the heart is called cardio-Behcet's disease; its prognosis has been extremely poor.3

We treated a patient with aortic valve regurgitation and conduction disturbance successfully with a prosthetic valve replacement and a permanent pacemaker implantation. Upon surgery it became clear that there were pathologic changes in the aortic cusps, aortic ring, and sinus of Valsalva; and an aneurysm of the sinus of Valsalva had ruptured into the upper portion of the muscular ventricular septum and perforated into the left ventricular (LV) cavity. Conduction disturbance in this case was presumably caused by an extension of the inflammatory extension of Behcet's disease, the same as in other generalized disease such as systemic lupus erythematosus4 and mixed connective tissue disease.5

CASE REPORT

A 47-year-old man had complaints of shortness of breath and palpitations upon admission. Since 32 years of age, recurrent arthralgia of the ankle and knee joints, and since 37 years of age, recurrent oral aphtha, genital ulcers, skin eruptions, and scleritis had been noted. Since 43 years of age, shortness of breath and palpitations had been noted and progressed, requiring repeated hospitalization at the medical center. In January 1981 massive melena developed, and he was admitted to a medical service. A barium enema revealed multiple ulcerations of the ileum and cecum. On this hospitalization Behcet's disease was diagnosed at first on

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