Case Report

A Case of Sarcoidosis Associated with Bronchial Asthma

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A 54-year-old woman was treated for bronchial asthma for 14 yr. In March of 1989, chest roentgenography and computed tomography (CT) revealed development of bilateral pulmonary hilar lymph node enlargement. Positive 67Ga uptake was observed in bilateral pulmonary hilus. Although levels of serum angiotensin converting enzyme (ACE) and lysozyme were within normal range, biopsy specimen of scalene lymph nodes showed noncaseating epitheloid-cell granuloma, leading to the diagnosis of sarcoidosis. Steroid therapy ameliorated both sarcoidosis and bronchial asthma. Although the association of sarcoidosis and bronchial asthma is uncommon, there may be an etiological relationship between them.

Key words: Bilateral hilar lymphadenopathy, Bronchial hyperreactivity, Scalene lymph node

Although the etiology of sarcoidosis is still unknown, an allergic mechanism may play some role in its development (1, 2). It is known that patients with sarcoidosis show immunological abnormalities such as negative tuberculin reaction and abnormal lymphocyte subsets. In addition, there are some reports that patients with sarcoidosis show bronchial hyperreactivity and tend to develop obstructive pulmonary change (3-5), while others have reported that this is not the case (6). Thus, the association of sarcoidosis with bronchial hyperreactivity still seems controversial. However, in fact, the association of sarcoidosis with bronchial asthma is unusual (7). Here we report a rare association of sarcoidosis during the course of bronchial asthma.

CASE REPORT

A 54-year-old woman (a housekeeper) was admitted to our hospital because of bilateral hilar lymph node enlargement. She had been healthy until the age of 39 (1975), when she started to experience frequent attacks of bronchial asthma some of which were provoked within 30 min by aspirin intake, but not by ibuprofen or diclofenac sodium. In 1986, when she was 50 yr old, she had a severe attack of bronchial asthma and was brought to our hospital by an ambulance. She was severely cyanotic and lost consciousness. Oxygen inhalation and administration of adrenalin, theophyllin, and hydrocortisone restored her. From that time she was treated in our outpatient clinic and had several attacks that were not related to any drug intake. The attack was typical of bronchial asthma in that it was characterized by periodic paroxysms of dyspnea with intervals of nearly complete remission and that marked elongation of expiratory time and expiratory wheezing were recognized and were dramatically improved by the administration of beta adrenergic stimulant and/or theophyllin. In November, 1988, chest roentgenogram revealed bilateral hilar lymph node enlargement which had not been present before. However there was no other remarkable change at that time. In March 1989, she was admitted to our hospital to determine the cause of pro-
gressive hilar lymph node enlargement. There was no history of atopy or bronchial asthma in her family. She was 152 cm tall and weighed 55.5 kg. Blood pressure was 130/75 mmHg. Pulmonary auscultation revealed slight expiratory sibilant rhonchi. Her abdomen, skin, and eyes including optic fundi showed no abnormality. No otolaryngeal abnormality such as nasal polyp or sinusitis was found. Analysis of urine and stool specimen, serum hepatic enzymes, renal function, various electrolytes in the blood including calcium and phosphate levels, and electrocardiograms were all normal. Blood cell counts were normal except that an increased number of eosinophilic leukocytes (480/μl) was obtained. Pulmonary function test showed a decrease in forced expiratory volume in one second. Radioallergosorbent test disclosed no positive allergen. Incorporation of thymidine into patient's lymphocytes was not stimulated by aspirin, although the aspirin was capable of provoking the attacks in her history. In this case, the negative family history of atopy, adult onset of asthma, no known external allergen, negative radioallergosorbent test for common allergens such as dust, pollen and ticks, association of no other allergies, and serum IgE level of 83 IU/ml (normal value is less than 250) are all suggestive of intrinsic-type bronchial asthma (8). Bilateral hilar lymph node enlargement was observed on plain roentgenogram (Fig. 1), chest tomogram, and chest CT scan. Whole body 67Ga citrate scintigraphy revealed abnormal accumulation in bilateral pulmonary hili (Fig. 2). Tuberculin reaction was 11 x 9.5 mm. Serum angiotensin converting enzyme and lysozyme levels were 18.5 IU/l/37°C (normal range 8.3–21.4) and 9.2 μg/ml (normal range 5.0–10.2), respectively. The values of tumor markers such as CEA, CA19-9, AFP, TPA, NSE and SCC were normal. Biopsy specimen of scalene lymph nodes showed multiple noncaseating epitheloid-cell granulomas, leading to the diagnosis of sarcoidosis (Fig. 3). She was discharged from the hospital without any medication for sarcoidosis in April 1989. In June 1989, bronchial asthma worsened and she experienced daily attacks despite...
continued administration of beta adrenergic stimulant, theophyllin, and ketotifen. The effect of inhalation of beta adrenergic stimulant became blunt and asthma became nearly continuous rather than intermittent as before. Prednisolone treatment of bronchial asthma was started in July 1989 and was successful. In March 1990, roentgenological improvement of bilateral hilar lymph node enlargement was observed.

DISCUSSION

An uncommon case of sarcoidosis during the course of bronchial asthma is reported. In this case, sarcoidosis was diagnosed by bilateral hilar lymphadenopathy and positive scalene biopsy. Although the sarcoidosis seemed to be active as evidenced by positive $^{67}$Ga uptake, the other features of sarcoidosis, such as skin and eye lesions, negative tuberculin reaction, and elevated serum angiotensin converting enzyme and lysozyme level were not observed. Since serum angiotensin converting enzyme level is decreased in bronchial asthma, apparently the normal angiotensin converting enzyme level in active sarcoidosis may be due to the association of the bronchial asthma.

Although the etiology of sarcoidosis is unknown, accelerated humoral immunity and decreased cellular immunity are often present and it is possible that allergic responses to exogenous stimuli play an important role in the development of sarcoidosis (1, 2). Recent reports suggest the presence of bronchial hyperreactivity in sarcoidosis and some etiological relationship between sarcoidosis and bronchial asthma (3–5). Abnormal immunologic reactions present in sarcoidosis cause activation of B lymphocytes to produce immunoglobulin (1, 8) and thus may accelerate IgE-mediated allergic reaction. In the present case, however, bronchial asthma preceded the development of sarcoidosis. Therefore it is unlikely that the abnormal immunologic reaction of sarcoidosis caused bronchial asthma in this patient, although it is possible that sarcoidosis is responsible for the exacerbation of pre-existing bronchial asthma. It is also possible that the granuloma of sarcoidosis caused airway obstruction which provoked the rather continuous bronchial asthma in this case. It would be unlikely that an underlying immunological abnormality in bronchial asthma caused sarcoidosis, because the association of sarcoidosis with bronchial asthma is uncommon (7). However, it is still possible that some antigenic stimulus, which we could not detect in the typical clinical tests, caused both the bronchial asthma and sarcoidosis.

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REFERENCES