as hemorrhagin is the possible mechanism of hemothorax in this patient. DIC causes fibrin deposits in the microcirculation, platelets and coagulation factors consumption with secondary fibrinolysis leading to bleeding.

Systemic envenomation by snakes can affect various organs of the body due to disturbances in the coagulation pathways. Hemothorax developing as a complication of snake bite has not been reported in the past and should be considered as a possible complication following snake bite.

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Lane-Hamilton Syndrome: Association or Coincidence?

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The combination of idiopathic pulmonary hemosiderosis (IPH) and celiac disease (CD) is rare. The clinical importance of this association is that a significant improvement can be obtained with gluten free diet not only in intestinal but also in pulmonary symptoms. A four and half-years old girl was admitted with complaints of cough, difficulty in breathing and paleness. She had intermittent episodes of abdominal pain and diarrhea. She had dyspnea and tachycardia, and oxygen saturation 88%. The patient was diagnosed with CD and concomitant IPH. With gluten-free diet and corticosteroid treatment, both intestinal and pulmonary symptoms were controlled.

Key words: *Celiac disease, Gluten-free diet, Pulmonary hemosiderosis.*

Idiopathic pulmonary hemosiderosis is a rare disease characterized by recurrent episodes of hemoptysis, pulmonary infiltration and iron deficiency anemia [1-4]. Celiac disease is an enteropathy characterized by life-long intolerance to ingested gluten in genetically susceptible people [5]. The simultaneous occurrence of these conditions is rare and is called Lane-Hamilton syndrome [6]. Significant improvement can be obtained with gluten-free diet, not only in intestinal but also in pulmonary symptoms [1,4,7].

CASE REPORT

This four and half years old girl was admitted with complaints of cough, difficulty in breathing and pallor. She had intermittent episodes of abdominal pain and diarrhea. The body weight and height were 16 kg and 105 cm (both at 3rd percentile). The heart rate, respiratory rate, blood pressure and oxygen saturation were 168 per min, 54 per min, 80/40 mm Hg and 88% respectively. The patient showed severe pallor. Fine crepitations were heard and liver was palpable 2 cm below the right subcostal margin. Investigations showed hemoglobin level of 2.5 g/dL, leukocytes 8300/cu mm and platelets 296000/cu mm. The mean corpuscular volume was 59 fl, reticulocyte count 10%, plasma iron 13 µg/dL, iron biding capacity 296 µg/dL and ferritine 74 µg/L. The peripheral smear examination showed hypochromic microcytic anemia. The chest radiograph showed bilateral infiltrates; echocardiography was normal. The findings improved after blood transfusion and the

pathologic radiological findings disappeared within a few days.

On follow-up, the patient showed recurrence of anemia, steatorrhea, abdominal pain and diarrhea. The levels of serological markers of celiac disease (anti-gliadin antibody IgA, IgG and antiendomysial antibody IgA) were >12 U/mL (normal range 0-12 U/mL). She was diagnosed as celiac disease based on the presence of crypt hyperplasia and partial villous atrophy and marked lymphocytic infiltration of the villous epithelium. The patient received gluten-free diet with improvement in intestinal symptoms. The patient was admitted again with anemia and respiratory distress. The hemoglobin level was 3 g/dL; chest radiograph showed bilateral patchy infiltrates. Hemosiderin laden macrophages were seen on gastric aspirate. Histologic examination of lung biopsy specimen revealed hemosiderin-laden intraalveolar macrophages, but there was no evidence of pulmonary vasculitis or granulomatous inflammation. Coagulation studies, levels of vitamin B12, folic acid, α-1 antitrypsin, immunoglobulin and complement were normal; serology for systemic lupus and other vasculitis were negative. RAST test against cow milk was negative. The patient was treated with oral methylprednisolone, at a dose of 2 mg/kg/day and gluten-free diet, with recovery of gastrointestinal and pulmonary symptoms. At follow up, the disease was controlled with low dose steroids and a gluten-free diet. The height was within 10 to 25 percentile.

DISCUSSION

Idiopathic pulmonary hemosiderosis with concomitant celiac disease is a rare condition [3,4,7,8]. Both conditions are considered to be immune-mediated, although the causal relationship is not clear [2-4,6,7]. Accumulation of immunocomplexes (including food allergens) in the alveolar capillary basement membrane, and cross reaction between antireticulin antibodies and alveolar basal membrane antigens are presumed to be pathogenic [3].

Use of gluten-free diet results in cessation of lung hemorrhage, reduced need for blood transfusions [2-4,7] and improved respiratory functions in Lane Hamilton syndrome. Gluten-free diet alone may not be sufficient, and patients require immunosuppressive therapy [1].

In two studies that screened patients with idiopathic pulmonary hemosiderosis for celiac disease [1,9] an accompaniment rate of 3/7 and 3/10, respectively was found . Two other studies showed that the two conditions may be associated in 6.6-8.7% cases [8,10]. Some researchers suggest that all patient with IPH should be screened routinely for CD, even in the absence of gastrointestinal symptoms [1-4,7,10]. Our patient had growth retardation, chronic anemia, recurrent abdominal pain and diarrhea. The diagnosis of celiac disease was based on positive serologic test, intestinal biopsy, and clinical and serological remission with gluten-free diet.

However, patients who have the Lane Hamilton syndrome show satisfactory improvement of pulmonary and gastrointestinal symptoms on a gluten-free diet. We propose that patient with idiopathic prolonged hemosiderosis should be screened for celiac disease, even in absence of gastrointestinal symptoms.

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