

A Case Report of Pulmonary Hemosiderosis Associated with Celiac Disease

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Abstract

Lane Hamilton Syndrome is an extremely rare combination of Idiopathic Pulmonary Hemosiderosis (IPH) and Celiac Disease (CD) [1-3]. We report a case of 4-years old male admitted for evaluation refractory iron anemia without response to iron treatment, the boy diagnosed with IPH and concomitant with CD.

Keywords: Lane-Hamilton Syndrome; Idiopathic Pulmonary Hemosiderosis; Celiac Disease

Introduction

Idiopathic pulmonary hemosiderosis is rare disease characters by the trade of iron deficiency anemia, recurrent episodes of hemoptysis and diffuse alveolar infiltration [1,2].

Celiac disease is an enteropathy, characterized by intolerance to ingested gluten [5]. The simultaneous occurrence of these condition is rare (prevalence 1%) and it's called Lane-Hamilton syndrome that described the first time in 1971. Significant improvement occur with GFD and corticosteroid, not the intestinal symptom but also the pulmonary [1,2,8,9].

Case Presentation and Discussion

A 4-years old male admitted for evaluation refractory iron anemia without response to iron treatment, he was sent to our department, mainly because for a persistent cough and refractory iron deficiency.

On physical examination, vital signs were normal, the body weight 16 kg and height 110 cm, overt pallor. In palpation, liver was palpation 3 cm below the right subcostal margin. In auscultation, bilateral fine crackles heard.

Laboratory tests were performed and revealed microcytic iron deficiency anaemia (Hct: %, Hgb: 6.3 g/dL, MCV: 57, Fe: 24 mg/dL). Ferritin, B12, reticulocyte count, immunoglobulin level and complement and folic acid levels were within normal range.

Peripheral smear showed hypochromic microcyte anemia.

Laboratory tests for collagen vascular disease and vasculitis, including antinuclear (ANA), antineutrophil cytoplasm (ANCA), and anti-glomerular basement membrane antibodies, were also negative. The tuberculin skin test was negative.

Pulmonary function tests (PFTs) were performed and showed normal FVC (%), FEV1 (%) and FEV1/FVC ratio (%).

The radiographic investigation; echocardiography was normal, chest X-ray showed bilateral alveolar interstitial infiltration (Figure 1).

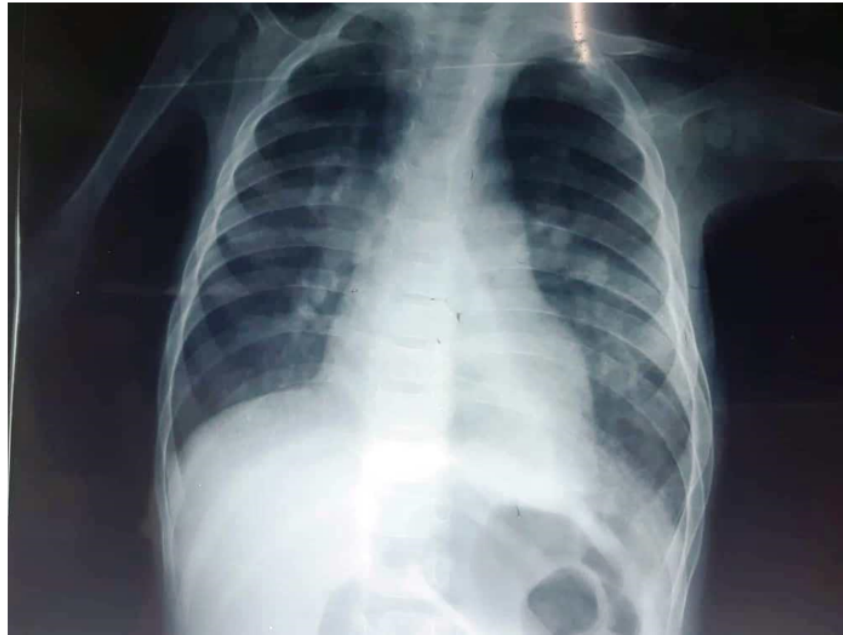


Figure 1

The high-resolution computed tomography (HRCT) showing diffuse bibasilar ground-glass opacities (Figure 2).

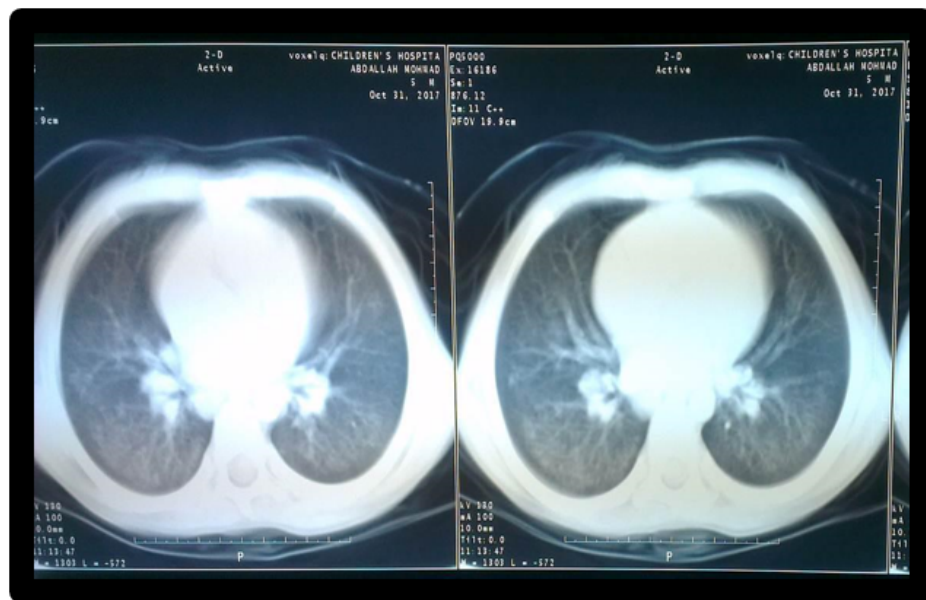


Figure 2

Bronchoalveolar lavage showed numerous hemosiderin-laden macrophage was confirming (IPH) (Figure 3).

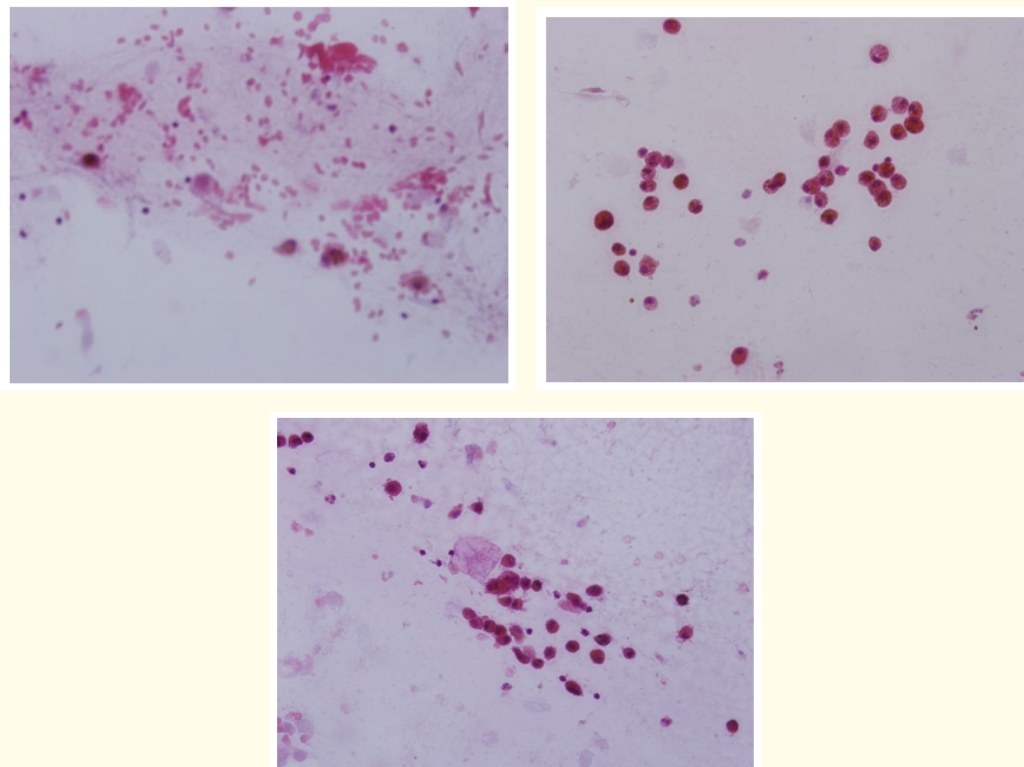


Figure 3

In search of an etiology of IPH an extensive workup performed, and the investigation of celiac disease was positive (serum IgA anti-endomysial and tissue transglutaminase antibodies at a titer of 300 IU/ml normal 0 - 10).

Duodenal biopsy showed villous atrophy, that confirmed CD, and the diagnosis of Lane Hamilton Syndrome was established (Figure 4).

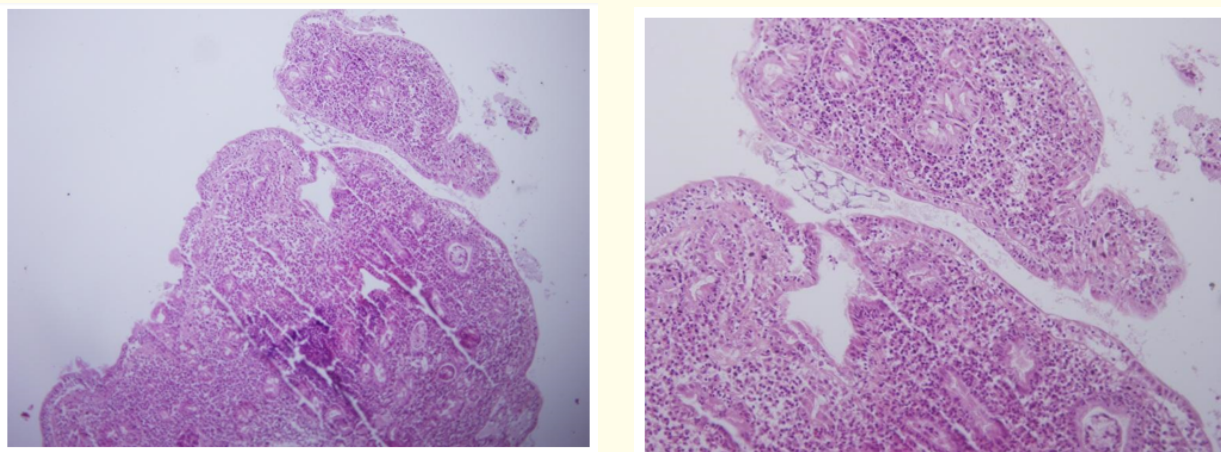


Figure 4

The child treated with prednisone at a dose of 1 mg/kg/day, gluten-free diet and iron supplement, with recovery growth, anemia and chest X-ray finding.

At follow up, within 4 months, the disease has controlled with low dose of steroids and gluten-free diet. On his last follow up after 1 year of treatment, the child gained weight and height, haemoglobin was 11 g/dl and improvement the radiography finding CXR and CT.

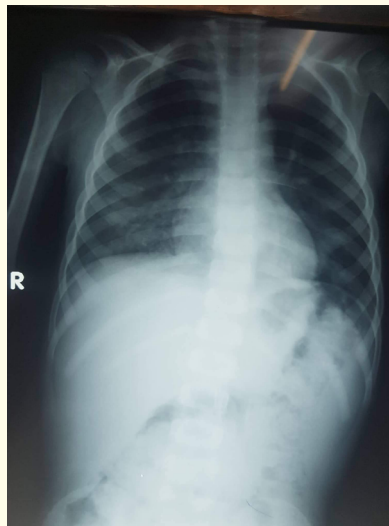


Figure 5

IPH is a rare disorder (the incidence of IPH between 0.24 and 1.26 patient per million children) [2,3].

The triad of hemoptysis, iron deficiency anemia and pulmonary infiltrates on chest imaging suspects diagnosis and the finding of haemosiderin laden macrophages in bronchoalveolar lavage fluid confirm the disease [4].

CD is an immunologically mediated enteropathy of small intestine, characterized by life long intolerance to the gliadin [5]. The combination of IPH and CD has been rarely reported in the literature [6,9]. The pathogenetic link between them is not clear [8].

Lane-Hamilton syndrome (association of IPH and CD) is reported in adults, so an evaluation for celiac disease is appropriate, if not already performed.

In our case the child had no signs of classic or non-classic CD, he had only refractory iron anaemia. When IPH has diagnosed and because of CD has been previously reported in association with IPH, we looked for CD and confirmed diagnosis.

Conclusion

Although the association is extremely rare, celiac disease should be consider in a patient with idiopathic pulmonary hemosiderosis, despite the absence of digestive symptom. Due to the rarity of this disease, diagnosis is usually delayed, but early diagnosis and treatment with corticosteroids and gluten free diet prevent further episode of recurrent alveolar hemorrhage and improve the outcome (both pulmonary and intestinal symptom).

Bibliography

1. Milman N and Pederson FM. "Idiopathic pulmonary haemosiderosis. Epidemiology, Pathogenic aspects and diagnosis". *Respiratory Medicine* 92.7 (1998): 902-907.
2. Kjellman B, *et al.* "Idiopathic pulmonary haemosiderosis in swedish children". *Acta Paediatrica Scandinavica* 73.5 (1984): 584-588.
3. Ogha S, *et al.* "Idiopathic pulmonary haemosiderosis in japan: 39 possible cases from survey questionnaire". *European Journal of Pediatrics* 154.12 (1995): 994-998.
4. Collard HR and Schawarz MI. "Diffuse alveolar hemorrhage". *Clinics in Chest Medicine* 25.3 (2004): 583-592.
5. Khemiri M, *et al.* "Screening for celiac disease in idiopathic pulmonary hemosidrosis". *Gastroentérologie Clinique et Biologique* 32.8-9 (2008): 745-748.
6. Bailey P and Gorden BM. "Idiopathic pulmonary heamosiderosis: report of two cases and review of the literature". *Postgraduate Medical Journal* 55.642 (1979): 266-272.
7. Bouros D, *et al.* "Bronchoalveolar lavage finding in a young adult with idiopathic pulmonary haemosiderosis and celiac disease". *European Respiratory Journal* 7.5 (1994): 109-107.
8. Ammar J, *et al.* "Idiopathic pulmonary haemosiderosis associated with celiac disease supre in child". *Revue Française d'Allergologie et d'Immunologie Clinique* 47 (2007): 511-514.
9. Reading R, *et al.* "Pulmonary haemosiderosis and gluten". *Archives of Disease in Childhood* 62.5 (1987): 513-515.

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