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An Unusual Presentation of Ulcerative Colitis in a Young Girl: A Case Report

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ABSTRACT

Pulmonary manifestations of ulcerative colitis have been well recognized, but incidence of bronchiectasis is rare, particularly in children.

In this study, a 16-year old girl is presented with rectorrhagy and abdominal cramp, who developed pulmonary symptoms as productive cough and dyspnea after a short period of time. Arterial blood gas (ABG) analysis showed hypoxia and hypercapnea. Chest x- ray and high resolution computerized tomography (HRCT) demonstrated honey comb pattern and changes indicating bronchiectasis in lower segments of both lungs.

Ulcerative colitis pattern was present in rectosigmoid biopsy. Open lung biopsy was performed to achieve definite diagnosis. Pathologic findings along with clinical and radiological presentations revealed a bronchiectatic pattern.

The patient underwent treatment with bronchodilators, sulfasalazine and prednisolone ; consequently, the symptoms were relieved. ***(Tanaffos 2005; 4(14): 71-74)***

Key words: *Ulcerative colitis, Bronchiectasis, Inflammatory bowel disease*

INTRODUCTION

Patients with ulcerative colitis (UC) may have a presentation dominated by extraintestinal manifestations (1). The association of respiratory disorders with enteric disease is uncommon but well recognized (2, 3). Up to 25% of patients with inflammatory bowel disease have respiratory involvement but these manifestations particularly bronchiectasis are rarely seen in pediatric patients (4).In this report, a 16-year-old girl with ulcerative colitis and coexistent bronchiectasis is presented. She developed unexplained productive cough and other respiratory symptoms after the appearance of colonic disease. This condition was repeated four times during 2 years but every time only respiratory symptoms were appreciated and rectorrhagy was missed; until the patient was admitted in our center, when we found every exacerbation of respiratory symptoms had been preceded by rectorrhagy and enteric manifestations which had been begun before the first presentation.

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CASE REPORT

A 16-year-old nonsmoker girl was well until

December 2002 when referred to our hospital with a history of chronic productive cough, recurrent respiratory infections, and no history of pulmonary disease in her family. There was also a history of recurrent rectal bloody diarrhea and crampy abdominal pain since 3 months before the first respiratory presentation. In physical examination, she had tachypnea, mild anemia and mild clubbing in fingers. There was no evidence of organomegaly but mild tenderness in lower parts of abdomen. Crackles and rhonchi were heard during lung auscultation. Arterial blood gas (ABG) analysis showed hypoxia and hypercapnea. Chest x-ray and HRCT showed evidences of hyperinflation and bronchiectatic lesions in both lower lungs. Bronchoscopy was performed and the specimen was not satisfactory for interpretation. Considering history of rectal bleeding, rectosigmoidoscopy was performed and severe continuous rectosigmoid ulcerations mostly consistent with ulcerative colitis were observed. Eight endoscopic biopsies were obtained from the involved area. On microscopic examination, the biopsies showed an increase in basal lymphoplasmocytes together with architectural distortion of crypts, cryptitis and focal crypt abscess, which were consistent with the diagnosis of ulcerative colitis (Fig 1,2).

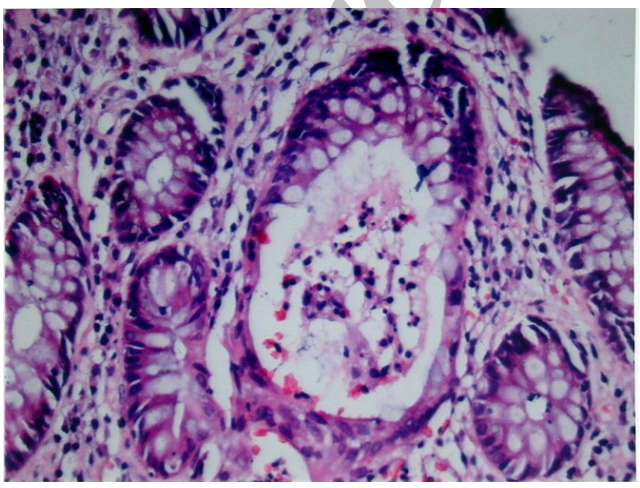

Figure 1. Crypt abscess formation in active phase of UC (H&E×400)

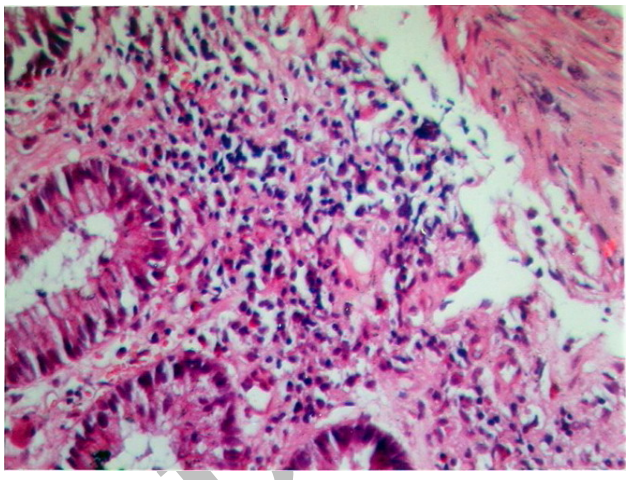

Figure 2. Basal lymphoplasmocytosis in chronic UC (H&E×400)

Due to reticular pattern in CT scan and suspicion of interstitial lung disease (ILD), open lung biopsy was performed. Histology revealed dilated bronchioles sometimes up to 4 times greater than normal size , intense acute and chronic inflammatory exudation in the walls of the bronchioles associated with desquamation of lining epithelium , areas of necrotizing ulceration with wall destruction and fibrosis (Fig3,4) which were in accord with bronchiectasis. Other lab exams were as follows:

Hct	31.1 %	ANA	-
Hgb	8.7 g / dl	Anti DNA	-
WBC	16000	ESR	30

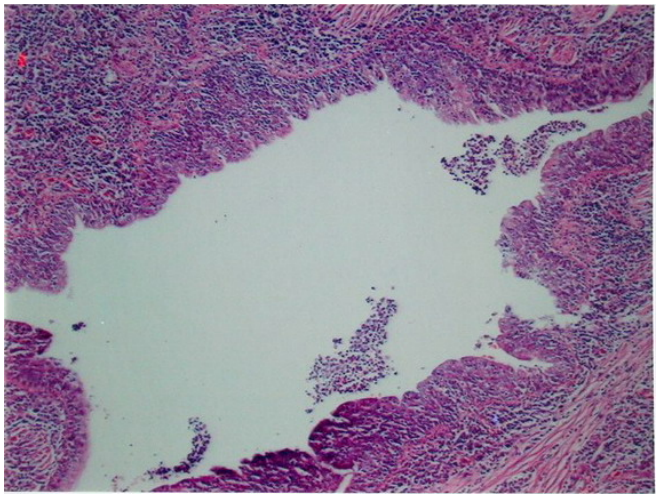

Figure 3. Dilated and destructed bronchiole with severe inflammation (H&E×100).

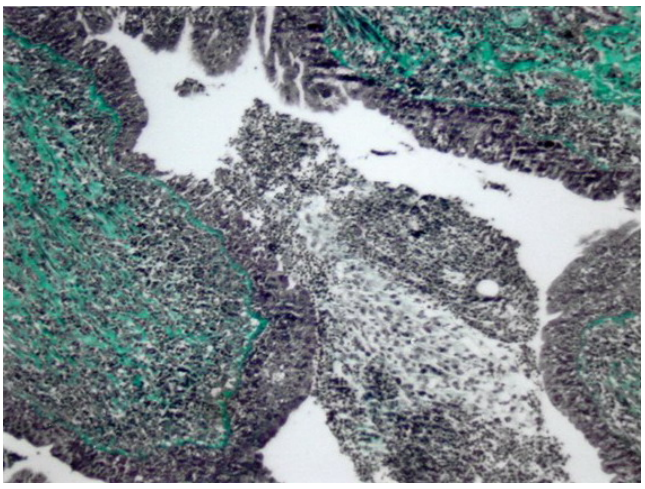

Figure 4. Trichrome staining of another bronchiole (Trichrome ×100)

Renal profile and liver function tests were normal.

She was discharged on the following drug regimen: prednisolone, theophylline, salbutamol and sulfasalazine.

DISCUSSION

Various respiratory complications have been reported in association with ulcerative colitis, albeit in a sporadic pattern. These include bronchiolitis obliterans, bronchitis, pleuritis, lung vascular involvement, bronchiectasis, inflammatory tracheal stenosis, chronic pneumonia and interstitial fibrosis (2, 3, 5, 6).

To the best of our knowledge, there is a very rare report of bronchiectasis complicating ulcerative colitis in adolescents. This link may be coincidental, related to the basic disease, its complications or its treatment but in our case, she was neither diagnosed nor treated for UC; therefore, relation to treatment can be ruled out. Complications and extraintestinal manifestations may precede or follow the diagnosis of IBD and may occur either with exacerbations of bowel symptoms or independently (7). The pathogenesis of respiratory complications in inflammatory bowel disease is largely unknown though postulated links including infiltration of the air way by immune effector cells such as lymphocytes enhanced immune activity as a part of the underlying disease (8).

The prevalence of autoimmune disorders is three times greater than expected in patients with ulcerative colitis but in this patient the absence of autoantibody partially argues against an autoimmune hypothesis(9).

Abnormalities of cell–mediated and humoral immunities, regulatory pathways, along with inflammatory mechanisms have been demonstrated in IBD and are related to various extraintestinal manifestations.

Excess formation of free radicals resulting in tissue damage has been documented in both bronchiectasis and ulcerative colitis.

Close temporal relationship between the two disorders and the relative infrequency of both conditions in adolescent population, along with previously published respiratory complications of inflammatory bowel disease, suggest a common denominator.

Considering this observation, we recommend that in approach to a bronchiectatic patient, every clinical history of abdominal pain or rectorrhagy (even transiently) must necessitate ruling out the inflammatory bowel disease. Early detection is important as both the pulmonary and gastrointestinal manifestations often respond well to steroids (particularly prednisolone).

Documentation of coexistence of above-mentioned disorders in a single patient is warranted in similar case reports in future.

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