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Bronchiectasis combined with chronic sinusitis following Celiac disease: a case report

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Abstract

Introduction Celiac disease is a disease triggered by a protein called gluten. Celiac disease has intestinal and extraintestinal manifestations. Bronchiectasis is a permanent dilation of the bronchi that causes symptoms, such as cough producing a large amount of sputum, recurrent respiratory infections, and breathlessness. In addition, bronchiectasis can present in 60% of cases with chronic rhinosinusitis.

Case presentation A 40-year-old Arab woman presented with a worsening old cough with an increased amount of sputum; the patient was diagnosed with Celiac disease 7 months prior. Investigations started with laboratory tests followed by a computed tomography scan for the head and chest, bronchoscopy, bronchoalveolar lavage, and spirometry; the final diagnosis was bronchiectasis with chronic rhinosinusitis. She was advised to commit to the gluten-free diet, in addition to the medications prescribed for her bronchiectasis and chronic rhinosinusitis.

Conclusion Celiac disease and bronchiectasis might share an immunologic disturbance that caused both entities, so Celiac disease should be kept in mind as an etiology for pulmonary diseases.

Keywords Bronchiectasis, Chronic sinusitis, Celiac disease

Introduction

Celiac disease (CD) is a multisystem condition that occurs owing to abnormal immune response to gluten. CD has intestinal manifestations, such as diarrhea, weight loss, loss of appetite, and abdominal pain, in addition to extraintestinal manifestations, such as bone loss, stomatitis, anemia, and others [1, 2]. Bronchiectasis is defined as a permanent dilation of the bronchi and has several reasons, such as infections, immunodeficiency, and ciliary clearance disturbance. The symptoms are cough, a large amount of sputum, breathlessness, and recurrent infections [3–5]. Bronchiectasis can accompany chronic rhinosinusitis (CRS) in more than 60% of cases [6]. According to our research in literature, we have found only few cases that describe the association between bronchiectasis and CD. We present a case of a 40-year-old woman who came to the hospital with a cough and increased sputum. She was diagnosed 7 months earlier with CD and our diagnosis for the recent complaint is bronchiectasis with CRS.

Case presentation

A 40-year-old Arab woman presented to the hospital with a worsening old cough and increased amount of white-yellowish sputum, accompanied by multiple bronchitis exacerbations during the last 6 months, with chest pain that is mostly worse with cough. The patient does not use tobacco or alcohol or take chronic medications. She is married and has six children. Her previous six pregnancies and deliveries were normal.

On physical examination, mild pallor was noticed and coarse crackles were heard on auscultation. Vital signs were measured: (blood pressure: 100/70 mmHg, heart

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rate: 111/minute, respiratory rate 36/minute, and saturation on room air 91%).

Regarding investigations, we started with laboratory tests (Table 1). Then, bronchoscopy revealed heavy thick secretions in both left and right bronchus, sputum, and bronchoalveolar lavage cultures showed growth of streptococcus. A chest computed tomography (CT) scan with contrast confirmed the diagnosis of bronchiectasis (Fig. 1). The CT scan of the sinuses showed chronic inflammation in the maxillary, right frontal, right sphenoidal, and ethmoidal sinuses (Fig. 2). Pulmonary function test showed these values: forced expiratory volume (FEV)1/forced vital capacity (FVC) was 72.7%, FEV1 was 1.44 (39%) and FVC was 1.98 (44%), which is consistent with combined restrictive and obstructive patterns.

Table 1 Laboratory tests

Laboratory test	Test value	Normal values
White blood cell count	11,350/mm^3	4500–10500/mm^3
Red blood cell count	3.5 million/mm^3	4.2–5.4 million/mm^3
Hematocrit	33.5%	36–48%
Hemoglobin	11 g/dL	12–16 g/dL
Mean corpuscular volume	96 fL	60–100 fL
Platelet count	566,000/mm^3	150,000–450,000/mm^3
Erythrocyte sedimentation rate	75 mm/hour	less than 20 mm/hour
Serum urea	20 mg/dL	6–24 mg/dL
Serum creatinine	0.49 mg/dL	0.6–1.1 mg/dL
Serum glucose	68 mg/dL	70–110 mg/dL
Serum lactate dehydrogenase	215 U/L	140–280 U/L
Serum creatine kinase	83 U/L	22–198 U/L
Serum total protein	7.4 g/dL	6–8.3 g/dL
Serum albumin	2.1 g/dL	3.4–5.4 g/dL
Serum total bilirubin	0.21 mg/dL	0.1–1.2 mg/dL
Serum direct bilirubin	0.12 mg/dL	Less than 0.3 mg/dL
Serum alanine transaminase	39 IU/L	4–36 IU/L
Serum aspartate transaminase	45 IU/L	8–33 IU/L
Serum sodium	140 mEq/L	135–145 mEq/L
Serum potassium	4.6 mEq/L	3.5–5.5 mEq/L
Serum chloride	102 mEq/L	95–105 mEq/L
Serum calcium	9 mg/dL	8.5–10.5 mg/dL
Serum phosphorus	3.4 mg/dL	3.4–4.5 mg/dL
Prothrombin time	15 seconds	11–13.5 seconds
Partial thromboplastin time	28.9 seconds	25–35 seconds
International normalized ratio	1.14	0.1–1.2
C-reactive protein	3.9 mg/dL	Up to 5 mg/dL
Serum vitamin D	10.43 ng/mL	25–70 ng/mL
Serum magnesium	2.01 mg/dL	1.5–2.5 mg/dL
Serum IgM	142 mg/dL	40–230 mg/dL
Serum IgG	2393 mg/dL	700–1600 mg/dL

Tuberculosis tests including Mantoux test and Gene Xpert on the bronchoalveolar lavage were negative. The echocardiogram showed an ejection fraction of 35% with moderate diastolic dysfunction. The bone density was measured and the patient was found to have osteoporosis. Treatment with a nasal spray of budesonide 50 µg for CRS, inhalers containing formoterol and ipratropium in addition to levofloxacin for bronchiectasis, spironolactone, sacubitril, valsartan, and bisoprolol were prescribed for the heart condition, and vitamin D for osteoporosis.

The patient also mentioned being hospitalized 7 months ago owing to a history of unintentional weight loss of 15 kg in 6 weeks, accompanied by fatigue, nausea, vomiting, and diarrhea. CD was suspected back then. Anti-TTG immunoglobulin A (IgA) was 340 IU/L. An upper endoscopy was done, and biopsies confirmed CD. The patient was asked to commit to a gluten-free diet. After that, she gained weight, became less fatigued and her stool returned to normal.

Regarding the follow-up, the patient came back to the hospital twice thereafter complaining about diarrhea, but she admitted not committing to the gluten-free diet. However, respiratory symptoms were better.

Discussion

CD is a multisystem condition that is caused by abnormal T cell-mediated immunological response to gluten [1]. Gluten is a water-insoluble protein found in wheat, rye, barley, spelt, and kannt [2, 7]. Gluten is one of the digestive-resistant proteins, which is chronically consumed. The final products of the partial digestion of gluten is a combination of peptides that trigger the host response and lead to changes in the epithelium surface and lamina propria through immune-mediated mechanisms [2, 7]. CD is one of the most common autoimmune diseases with a prevalence of 0.5–1% of the population [2].

The clinical manifestations of CD include intestinal and extraintestinal manifestations. Intestinal symptoms among children include diarrhea, loss of appetite, abdominal distension, and failure to thrive, while adults complain about diarrhea, bloating, constipation, abdominal pain, and weight loss. Extraintestinal signs are anemia, bone loss as osteopenia or osteoporosis, aphthous stomatitis, neurological symptoms, such as headache, paresthesia, and depression, and CD might affect menstrual cycle and fertility [1, 2]. CD might accompany many autoimmune diseases, such as dermatitis herpetiformis, Hashimoto's thyroiditis, selective IgA deficiency, connective tissue diseases, and others [2].

The criteria to diagnose CD is to achieve at least four out of five of the following: typical symptoms and signs, antibody positivity, existence of HLA-DQ2 or/and HLA-DQ8, intestinal damage, such as villous atrophy, and

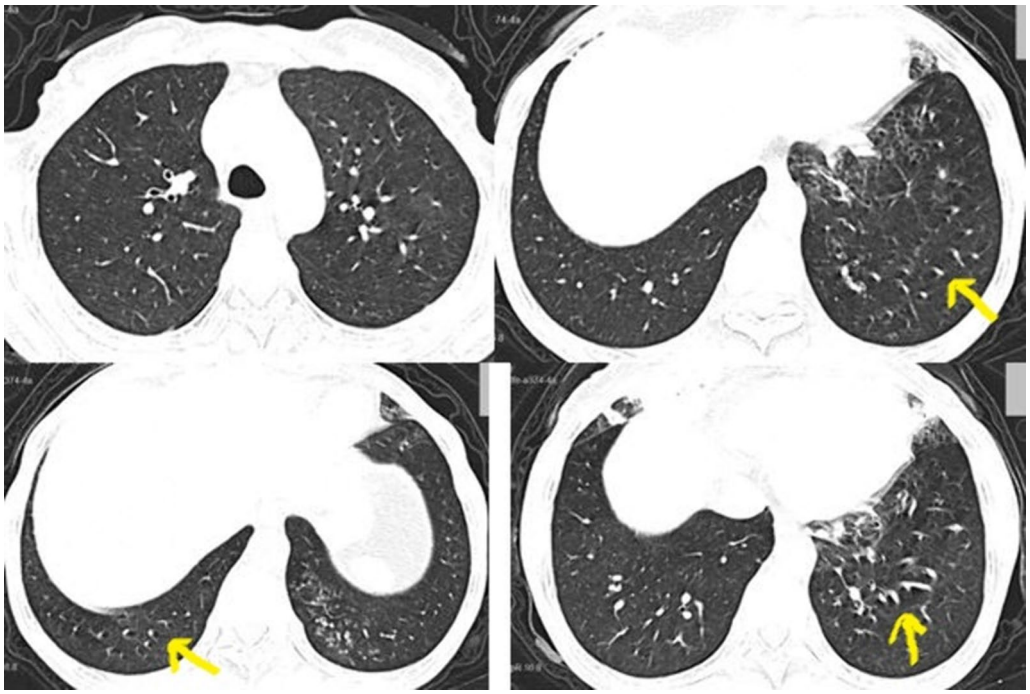

Fig. 1 High-resolution Computed tomography with contrast of the chest, showing the dilated bronchi (the arrows are defining the dilated bronchi)

clinical response to gluten-free diet [2]. The decision to diagnose CD depends on serology testing; including tissue transglutaminase IgA more than three times the normal limit, endomysial antibodies, antigliadin and antideaminated gliadin peptides. In addition, histological changes of the biopsy taken from the duodenum through endoscopy should reveal intraepithelial lymphocytes with more than 25 lymphocytes for every 100 enterocytes, partial or total villous atrophy, crypt hyperplasia, and chronic inflammation in the lamina propria [2, 7].

CD complications contain hyposplenism, which can cause infections by encapsulated bacteria, refractory CD, which is defined as persistent symptoms and atrophic villi despite the commitment to the gluten-free diet, intestinal lymphoma, small bowel adenocarcinoma, and ulcerative jejunoileitis [2, 7].

Bronchiectasis is a permanent dilation of the bronchi and bronchioles as a result of destruction of the elastic and smooth muscle tissue, and that occurs owing to infectious or noninfectious reasons [3]. The etiology is kind of a vicious circle, which begins in the accumulation of mucus followed by chronic inflammation and infection on the mucus and then airway destruction owing to the infection, then mucus accumulation again and so on [4]. The causes are 40% idiopathic, 30% after an infection, such as pneumonia or tuberculosis, 5% immunodeficiency [either primary or secondary to human immunodeficiency virus (HIV), chemotherapy, or immunosuppressant], other causes contain ciliary clearance disturbance, such as primary ciliary dyskinesia or cystic fibrosis, or airway structure damage, such as foreign body, chronic obstructive pulmonary disease COPD, tumor, recurrent aspiration and alpha-1-antitrypsin deficiency [4, 5].

Bronchiectasis patients can present with chronic cough producing massive amounts of sputum, recurrent respiratory infections, hemoptysis, breathlessness, and chest pleuritic pain [4]. The gold standard imaging for bronchiectasis is high-resolution computed tomography (HRCT), which should reveal airway dilation besides a bronchial-arterial diameter ratio more than 1 [4]. Investigations to discover the cause of bronchiectasis include complete blood count (CBC), serum immunoglobulins, serum protein electrophoresis, HIV, sputum culture for tuberculosis (TB), sodium chloride sweat test, cystic fibrosis transmembrane conductance regulator (CFTR) mutation analysis, alpha 1 antitrypsin, bronchoscopy, and test assessing ciliary structure and function [4].

The treatment purpose is to relieve symptoms, have a better quality of life, less exacerbations, and make disease progression slower. Mucus clearance has to be done daily through aerobic exercise, active cycle of breathing, positive expiratory pressure device, and postural drainage. Smoke cessation and vaccinations are important. Antibiotics are used to treat exacerbations, less frequent exacerbations, and to eradicate pseudomonas. Other than

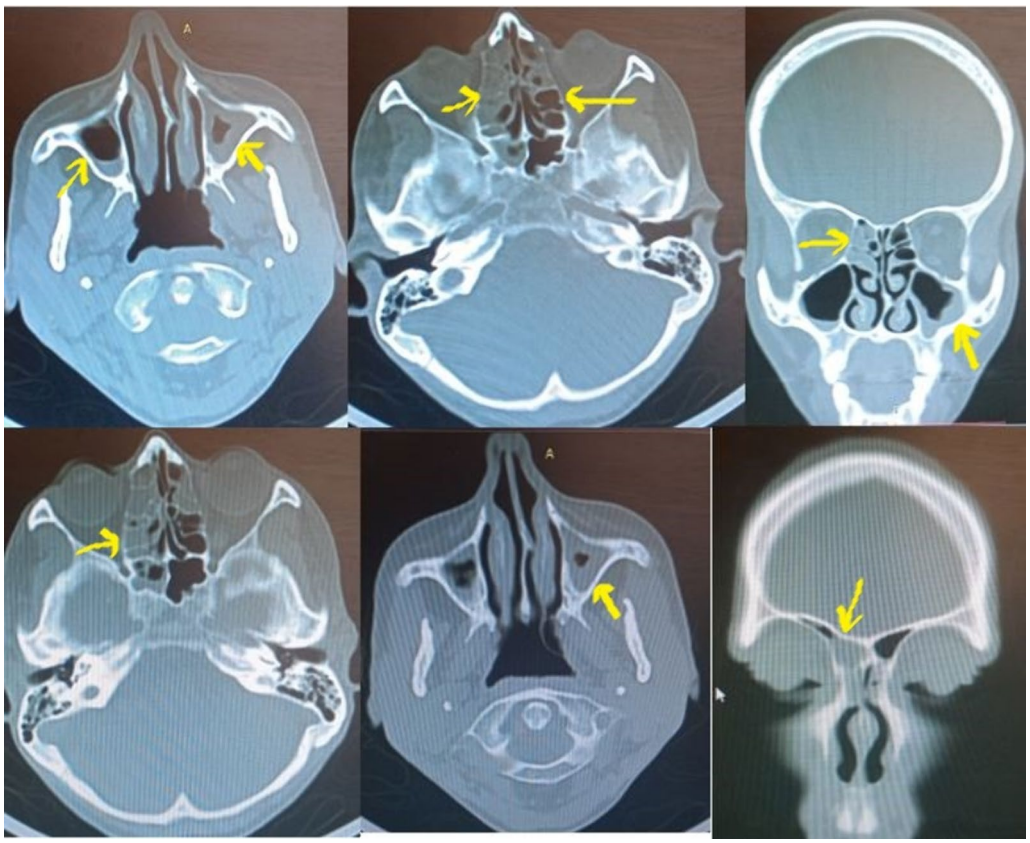

Fig. 2 Sinonasal computed tomography scan, showing the chronic rhinosinusitis manifestations, including thickened lining of the maxillary, right frontal, right sphenoidal, and ethmoidal sinuses, which is consistent with chronic inflammation (the arrows are defining the radiological signs of chronic rhinosinusitis)

exacerbations or the existence of pseudomonas, antibiotics are not indicated [5].

We might give saline or mannitol as mucoactive agents to increase the ciliary clearance of the mucus. Short-acting bronchodilators are used before mucolytics and inhaled antibiotics. There is no proof to support using inhaled corticosteroids and long-acting bronchodilators in bronchiectasis patients [5].

CRS is defined as having two or more of the following upper respiratory symptoms: nasal congestion and blockage, facial pain, and reduced smell sensation; these symptoms last more than 12 weeks. CRS accompanies bronchiectasis in 62% of cases and patients who have bronchiectasis with CRS combined have a more severe condition than patients without CRS, in addition, CRS without polyps is the type of CRS that usually accompanies bronchiectasis [6].

To our knowledge and after searching literature, bronchiectasis accompanying CD has been mentioned multiple times, but CRS has never been part of this combination. The first case is in 1998, a 48-year-old woman presented with bronchitis for 6 years and she was also diagnosed with CD [8]. The second case is of a 10-year-old boy who presented with recurrent diarrhea, wheezing, and respiratory infections. He was diagnosed with bronchiectasis and cystic fibrosis, but after treatment with enzyme supplements, no benefit was noticed. A genetic analysis showed no cystic fibrosis, and a diagnosis of CD was made [9]. The third case is of a 6-year-old girl who was diagnosed with bronchiectasis with CD [10]. Another case was published in 2012 of a 40-year-old man, who was also diagnosed with bronchiectasis and CD [11]. A study conducted in 2022 revealed that bronchiectasis accompanied CD in nine cases [12].

CD can accompany pulmonary diseases, such as asthma, chronic cough, diffuse pulmonary nodules, interstitial fibrosis and alveolitis, and pulmonary hemosiderosis [10, 13]. The reason for the association between CD and lung diseases is not clearly defined yet. However, a theory said that the absorption of an extrinsic allergen or immune complex through the abnormal gastrointestinal mucosa might cause lung disease and the association between CD with HLA and autoimmune diseases suggests a common disturbance in the immunity and this

can be the underlying cause for both CD and bronchiectasis [10].

The limitation of this case is that we could not do some diagnostic tests, including genetic testing, because of the costs.

Conclusion

CD and bronchiectasis might share an immunologic disturbance that caused both entities, so CD should be kept in mind as an etiology for pulmonary diseases.

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Author contributions
Ali Ghassa gathered the data, researched the literature, and wrote the manuscript.

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Availability of data and materials
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Declarations

Ethics approval and consent to participate
Exempted.

Consent for publication
Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Competing interests
The author declares that there is no conflict of interest to be reported.

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