Recurrent Respiratory Infections in a Patient With Chronic Diarrhea

Infecciones respiratorias de repetición en paciente con diarrea crónica

To the Editor:

Good’s syndrome (GS) is a primary immunodeficiency characterized by thymoma and humoral immunodeficiency. It is the most unusual form of the parathymic syndrome, after far behind myasthenia gravis or pure red cell aplasia.¹ The most common forms of clinical presentation are recurrent infections, hematological changes, and chronic diarrhea.¹,²

We report the case of a 76-year-old man with a history of arterial hypertension, a former smoker of 20 pack-years, with chronic diarrhea which yielded Campylobacter coli on culture. He was referred to the respiratory medicine department for repeated respiratory infections. Forced spirometry showed mild obstruction: FEV₁/FVC 0.67, FEV₁ 2.1 L (87%), FVC 3.17 L (98%). Skin prick tests for airborne allergens were negative. Clinical laboratory tests showed hemoglobin 11.7 g/dl with normal corpuscular volume, with no impact on platelet levels, and markedly reduced CD19 lymphocytes, with a CD4/CD8 ratio of 1.04. Immunoglobulin levels were low: IgA < 5 mg/dl, IgG < 74 mg/dl, IgM < 5.3 mg/dl. Methicillin-resistant Staphylococcus aureus was isolated from repeated sputum cultures. Computed tomography (CT) of the paranasal sinuses showed occupation of the maxillary sinuses, while the chest CT revealed mild bronchiectasis in the middle lobe, lingula and both lower lobes, and a solid multilobulated mass in the anterior mediastinum suggestive of thymoma. VATS was performed, confirming the histological diagnosis of polygonal cell cortical thymoma. With these findings, the patient was determined to have GS, and immunoglobulin replacement therapy was started. He showed good progress and the number of infections fell.

GS mostly appears in patients in their 30s or 40s (unlike our patient), and affects men and women equally.¹ This immunodeficiency is caused by an antibody deficit, and is currently classified as a different entity to common variable immunodeficiency (CVID).³ It accounts for 2% of cases of primary antibody deficiency treated with immunoglobulin replacement therapy.¹

The most common clinical manifestation is recurrent respiratory infection, and the major pathogens are Haemophilus influenzae and Pseudomonas spp. The chronic diarrhea presented by 50% of patients appears to have an autoimmune basis, and the isolation of pathogenic agents is anecdotal.³

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Table 1
Clinical Laboratory Tests on Admission and Arterial Blood Gases.

<table>
<thead>
<tr>
<th></th>
<th>Hb (g/dl)</th>
<th>Hct (%)</th>
<th>Urea (mg/dl)</th>
<th>Cr (mg/dl)</th>
<th>Na (mEq/dl)</th>
<th>K (mEq/dl)</th>
<th>Urea (mg/dl)</th>
<th>Cr (mg/dl)</th>
<th>Na (mEq/dl)</th>
<th>K (mEq/dl)</th>
<th>pH</th>
<th>pCO₂ (mmHg)</th>
<th>Bicarbonate (mMol/l)</th>
<th>Base Excess (mMol/l)</th>
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<tbody>
<tr>
<td>Case 1</td>
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<td>88</td>
<td>0.91</td>
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<td>2.81</td>
<td>28</td>
<td>1.65</td>
<td>140</td>
<td>3.1</td>
<td>7.61</td>
<td>46</td>
<td>46.2</td>
<td>24.8</td>
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<tr>
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<td>49</td>
<td>127</td>
<td>2.86</td>
<td>128</td>
<td>2</td>
<td>39</td>
<td>1.43</td>
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<td>7.47</td>
<td>45.90</td>
<td>32.7</td>
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<tr>
<td>Case 3</td>
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<td>53.8</td>
<td>94</td>
<td>1.43</td>
<td>131</td>
<td>4.36</td>
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<td>1.05</td>
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<td>3.91</td>
<td>7.48</td>
<td>33</td>
<td>24.3</td>
<td></td>
</tr>
</tbody>
</table>

Cr, creatinine; ER, emergency room; HB, hemoglobin; Hct, hematocrit; K, potassium; Na, sodium.

References

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Diagnosis is based on the clinical picture and immune response studies. The latter are characterized by reduced B cells and hypogammaglobulinemia with reduced CD4 T cells and an inverted CD4/CD8 ratio.¹

Treatment of choice is regular intravenous administration of gammaglobulins to treat the humoral immunity, providing clinical benefit in most cases.² Resection of the thymus is also indicated, to prevent the potential risk of locally invasive growth and metastatic dissemination, although the procedure does not seem to improve immunodeficiency.²

References


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Chronic Lung Infection Caused by Trichosporon mycotoxinivorans and Trichosporon mucoides in an Immunocompetent Cystic Fibrosis Patient

*Infección pulmonar crónica causada por Trichosporon mycotoxinivorans y mucoides en un paciente inmunocompetente con fibrosis quística*

To the Editor:

Very few studies have been published in the medical literature on systemic infections caused by Trichosporon spp in immunocompetent patients, and even fewer in cystic fibrosis (CF) patients. This species is often associated with acute processes with poor prognosis.¹–⁴

We report a review of the literature and a case study of a CF patient with chronic bronchial infection (CBI) caused by Trichosporon mycotoxinivorans (T. mycotoxinivorans) and Trichosporon mucoides, who progressed well during follow-up.

This is a 37-year-old man, diagnosed at the age of 4.5 months with CF (F508del/G542X), with mild pulmonary and gastrointestinal involvement, and a history of CBI due to methicillin-sensitive Staphylococcus aureus and intermittent bronchial infection with Pseudomonas aeruginosa resolved in 2005. He continued to receive rapid-action bronchodilators, physiotherapy, pancreatic enzymes, and liposomal vitamins.

Five years ago, during a routine visit, T. mucoides was isolated from a microbiological culture of the sputum. In the complementary examinations, spirometry was normal (FEV1: 2.71 l/65%) with basal oxygen saturation (SO₂) 96%. In view of this finding, itraconazole 200 mg/24 h was started. In successive sputum cultures, T. mycotoxinivorans was isolated and persisted until the patient’s last visit. No radiological (Bhalla: 16) or functional (FEV1: 3.25 l/95% and basal SO₂: 97%) worsening was observed. Mean exacerbations/year in the 5-year follow-up was 1.2, all of which were mild, treated with oral antibiotic therapy according to the sensitivity profile, similarly to previous years.

The first human infection in CF with T. mycotoxinivorans was a case of pneumonia with fatal outcome, published in 2009. Cases published subsequently also had very poor prognosis.¹²⁴ Shah et al.² reported a series followed up for a maximum of 6 years, of which 4 patients had CBI due to T. mycotoxinivorans, and in another, it was isolated once. No correlation was found between this infection and the very high number of subsequent exacerbations, but it can be supposed that T. mycotoxinivorans played a part,² both in the clinical symptoms and the prognosis.

Although it remains to be clarified, workplace exposure, transplantation and treatment, diabetes, inhaled and systemic corticosteroid, malnutrition, severely compromised lung function, intrinsic drug resistance to mycotic infections, or the chronic use of inhaled or systemic antibiotic treatment, may be risk factors for developing T. mycotoxinivorans infection in CF.¹—⁴ Our patient did not present any risk factors or clinical, radiological or functional worsening in the 5 years before the appearance of Trichosporon spp.

We believe that the change of species in our case was related with 2 events. The first was that the Trichosporon genus was recently reorganized.¹ The second was the method of identification used, phenotyping techniques (API®, VITEK®) and mass spectrometry (MALDI-TOF).³

After close examination of our case, a CBI with no clinical repercussions, and after reviewing the literature, we conclude that Trichosporon spp, and in particular T. mycotoxinivorans, are associated with widely varying clinical manifestations in CF, although to date most cases have been severe and fast progressing with a fatal outcome. Under the right circumstances,¹,³ patients may present chronic infection caused by this fungus.

References


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