Letters to the Editor

Fibromyalgia and Chronic Fatigue Syndrome Caused by Non-Celiac Gluten Sensitivity

Dear Editor:

Sensitivity to gluten with negative celiac disease testing or non-celiac sensitivity to gluten is a recently recognized problem with clinical manifestations that are superimposed with those of fibromyalgia, chronic fatigue and irritable bowel syndrome.

We present the case of a 40-year-old woman who came to the clinic with a 7-year history of generalized pain and chronic fatigue. She had been diagnosed with fibromyalgia by several rheumatologists and complied with the 1990 American College or Rheumatology criteria. She also presented chronic fatigue syndrome criteria. She had concentration and memory problems, «foggy mind», and intermittent diarrhea. The severity of the affection led to limitation in her daily activities which limited her to bed rest in spite of several visits to specialists in rheumatology, gastroenterology and alternative medicine/homeopathy. In addition to the typical symptoms of fibromyalgia, chronic fatigue and intermittent diarrhea, she had oral ulcers, autoimmune hypothyroidism and a history of iron deficiency. She had undergone multiple studies with normal findings, including anti-transglutaminase IgA antibodies to rule out celiac disease.

We suspected sensitivity to gluten and more studies were performed. Laboratory studies showed iron deficiency and low vitamin D levels. On a screening test for anti-transglutaminase and anti-deamidated gliadin peptide antibodies, both IgG and IgA were negative. HLA typing showed the presence of DQ2 (DQA1*05 DQB1*02). Gastroscopy showed small atrophic lesions on the duodenal bulb. Duodenal biopsies showed normal villi structure and lymphocytic duodenitis with apical redistribution, 28 CD3 lymphocytes for every 100 enterocytes [stage I Marsh lesions]. Urease testing for Helicobacter pylori was positive. Celiac disease was ruled out due to the absence of specific antibodies or intestinal villi atrophy, though we still suspected sensitivity to gluten. A gluten-free diet was recommended without treating the infection by Helicobacter pylori.

Six months after starting the diet there was a marked improvement in all of the symptoms, with remission of the oral ulcers; the patient had gone back to work after a long disability period. When she consumed small amounts of gluten she experimented a relapse of all of the symptoms. Iron and mineral supplementation was carried out. 2 years after she started the diet there was remission of fibromyalgia, she continued working and practiced sports. During that time her daughter had been diagnosed with celiac disease, presented positive anti-transglutaminase antibodies and villi atrophy on the duodenal biopsy.

The existence of non-celiac gluten sensitivity is recognized based on the observation of patients whose symptoms respond to a gluten-free diet but have negative celiac disease tests. Lymphocytic duodenitis is a characteristic of celiac disease that may present without villi atrophy in non-celiac gluten sensitivity, although it is not a specific finding as it may have other causes, such as Helicobacter pylori. When, as in this case, a patient with gluten sensitivity has an HLA susceptibility and Marsh 1 enteropathy, there is such a diagnostic proximity to celiac disease that the term of Marsh 1 celiac disease has been proposed. A gluten-free diet was attempted in this patient without treating Helicobacter because it was unlikely that treatment would improve the fibromyalgia.

Non-celiac gluten sensitivity has recently been described as a cause of fibromyalgia. This case reinforces this hypothesis due to the characteristic clinical response, a relapse after gluten consumption, the HLA typing, the presence of lymphocytic duodenitis in the duodenal biopsy and the latter diagnosis of celiac disease in her daughter.

References


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What Is the Outcome of Undifferentiated Arthritis?

¿Cuál es la evolución de las artritis indiferenciadas?

To the Editor:

The use of a coding system in rheumatology improves the performance and quality of patient care. In the rheumatology department of our hospital, which has a reference area of 850,000 inhabitants, coding is carried out since 1984. Several rheumatologists prospectively collect, at 6 months of the first visit, filiation and diagnosis of patients seen in the clinic, patients admitted to hospital beds and admitted to other services that request an interconsultation. The nomenclature of the American College of Rheumatology (ACR) of 1983 was used and suitably modified to include a section of undiagnosed arthritis where mono, oligo and polyarthritis without an identified cause are included. Between 2006 and 2011, 13,767 first consultations were carried out and 154 patients with undiagnosed arthritis (1.12%) were classified. They retrospectively reviewed the medical records of these patients and the location of arthritis, follow up and final diagnosis reached up until December 2013 were collected, with an elapsed minimum 2½ years since the first visit (range 2.5–5 years).

The distribution of arthritis was: 36 (23.3%) monoarticular, 71 (46.1%) oligoarticular and 47 (30.5%) polyarticular. Monoarticular forms affected the following joints: in 21 cases (58.3%) knee; 4 (11.1%) metatarsophalangeal; 3 (8.3%) proximal interphalangeal; 2 (5.5%) tibioperoneoastragalus; 2 (5.5%) carpus; 2 (5.5%) metacarpophalangeal; 1 (2.7%) tarsus, and 1 (2.7%) elbow. The evolution of arthritis is summarized in Table 1.

It is noteworthy that of the 36 patients with monoarthritis, 4 evolved to seronegative oligo or polyarthritis and 3 oligoarthritis patients progressed to undifferentiated polyarthritis, also undifferentiated.

We conclude that undifferentiated arthritis may progress to a classifiable inflammatory disease, self-limit or persist undifferentiated, representing a challenge for the rheumatologist.

*The percentage of spontaneous resolution in our series of cases is 44.4%, 9.8% and 14.9% for mono, oligo and polyarthritis, respectively. The former are more inclined to resolve spontaneously, whereas oligoarticular and polyarticular onset forms are more likely to become chronic. In the present series an established diagnosis was reached in 57 patients (65%) after an average of 8 months; mostly had an oligoarticular origin (65%). 11 patients (19%) were diagnosed with spondyloarthritis, whose onset was characterized as mono or oligoarticular; conversely, the 9 patients (16%) with a subsequent diagnosis of rheumatoid arthritis (RA) had an oligo or polyarticular onset. In addition, 10 patients (17.5%) were diagnosed with microcrystalline arthritis with a mono, oligo or polyarticular onset.

Most undiagnosed polyarthritis in the first 6 months, continued without cause after follow up. Interestingly, the percentage of

Table 1

<table>
<thead>
<tr>
<th>Evolution</th>
<th>Monoarticular 36 (23.3%)</th>
<th>Oligoarticular 71 (46.1%)</th>
<th>Polyarticular 47 (30.5%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Undifferentiated 67 (43.5%)</td>
<td>9</td>
<td>27</td>
<td>31</td>
</tr>
<tr>
<td>5 follow controls</td>
<td></td>
<td>16</td>
<td>11 follow controls</td>
</tr>
<tr>
<td>4 do not follow controls</td>
<td></td>
<td>11 do not follow controls</td>
<td>20 do not follow controls</td>
</tr>
<tr>
<td>self-limited 30 (19.5%)</td>
<td>16</td>
<td>7</td>
<td>7</td>
</tr>
<tr>
<td>Diagnosis established 57 (65%)</td>
<td>11</td>
<td>37</td>
<td>9</td>
</tr>
<tr>
<td>3 spondyloarthritis</td>
<td></td>
<td>8 spondyloarthritis</td>
<td>2 rheumatoid arthritis</td>
</tr>
<tr>
<td>2 microcrystalline</td>
<td></td>
<td>7 rheumatoid arthritis</td>
<td>2 microcrystalline</td>
</tr>
<tr>
<td>2 juvenile idiopathic arthritis</td>
<td></td>
<td>6 microcrystalline</td>
<td>Lupus</td>
</tr>
<tr>
<td>4 others *</td>
<td></td>
<td>3 palindromic rheumatism</td>
<td>1 palindromic rheumatism</td>
</tr>
<tr>
<td></td>
<td></td>
<td>2 sarcoidosis</td>
<td>Psoriatic arthritis</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Still’s disease</td>
<td>Two others *</td>
</tr>
</tbody>
</table>

* Arborescent lipoma, undifferentiated granulomatous synovitis and mechanical causes.

† Paraneoplastic, TBC, reflex sympathetic dystrophy, SAPHO syndrome, vasculitis and osteoarthritis.

‡ Polygala gia rheumatic and osteoarthritis.

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