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Case Report Open Access

Capsule Endoscopy Detection of Ileal Mucosa Alterations due to Cow Milk Malabsorption in a Celiac Patient

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Abstract

We report a case of a 77-year-old man suffering from celiac disease admitted as an outpatient to our clinic for the persistence of bloating and diarrhea without macroscopic steatorrhea, weight loss or an increase in circulating carbohydrate antigen 19-9. An esophagogastroduodenoscopy showed *H. pylori* negative antral gastritis and atrophy of the duodenal mucosa and the results of a colonoscopy were normal. Abdominal contrast-enhanced computed tomography did not show any parenchymal alteration of the liver and pancreas. The patient underwent a capsule endoscopy and the examination showed the presence of villous atrophy with microerosions at the level of the medial jejunum, in the ileum, and some areas of lymphangiectasia. A repeated capsule endoscopy after starting a lactose-free diet showed the normalization of the jejunum-ileum villous atrophy. At present, the patient is in good general health and continues a gluten- and cow milk-free diet. We should be aware that celiac patients may have lactose intolerance that cause malabsorption; the transient increase of CA 19-9 in celiac patients may be due to causes other than the presence of gastrointestinal cancer; the capsule endoscopy is useful in detecting ileal mucosa abnormalities which disappear after going on gluten- and cow milk-free diet.

Keywords: Celiac disease; Cow milk intolerance; Wireless capsule endoscopy; Outcome

Introduction

Lactose intolerance is a frequent cause of gastrointestinal symptoms worldwide, including bloating, flatulence and diarrhea, appearing after the ingestion of food products containing lactose [1]. The degree of discomfort depends primarily on the amount of lactose ingested as well as the sensitivity to lactose [2]. The association of cow milk intolerance in celiac disease patients has been reported [3] by means of histology and disaccharidase activities of duodenal mucosal biopsies. To this end, we report the case of lactose intolerance in a patient with celiac disease in whom the diagnosis of lactose intolerance was assessed by means of a lactose breath test, and ileal alterations were detected by using capsule endoscopy.

Case Report

We describe, after had he given informed consent, the case of 77-year-old man suffering from celiac disease which had been histologically diagnosed at the age of 68 years and who was on gluten-free diet. He was admitted as an outpatient to our clinic in September 2010 for the persistence of bloating and diarrhea without macroscopic steatorrhea, weight loss (about 4 kilos) and an increase in circulating carbohydrate antigen 19-9 (CA 19-9) levels (72 U/mL, upper reference limit <37) that were apparently not due to other medical problems.

The patient was compliant with the gluten-free diet as was also confirmed by his spouse. Blood examinations were unremarkable as was the total IgE (53 U/L, upper reference value 100), the serum IgA were 241 mg/dL (reference range 70-400) and the serum determination of both immunoglobulin A antibodies to deamidated gliadin peptides (0.8 U/mL, upper reference value: 25 AU/mL) and to tissue transglutaminase (0.5 U/mL, upper reference value: 7 U/mL). Fecal occult blood examination was also negative and fecal elastase 1 was within the normal limit. An esophagogastroduodenoscopy showed *H. pylori* negative antral gastritis and atrophy of the duodenal mucosa and the results of a colonoscopy were normal (biopsies showed a normal colonic mucosa). Abdominal contrast-enhanced computed tomography did not show any parenchymal alteration of the liver and pancreas. To better investigate the etiology of the diarrhea, the patient underwent an H_2 breath test with lactose. After a baseline breath sample was measured, 25 g of oral lactose was administered and breath samples were taken every 30 minutes; the presence of hydrogen in parts per million (ppm) was determined by using a QuinTron MicroLyzer (Milwaukee, WI); this examination revealed milk-induced malabsorption because there was a rise in hydrogen excretion of more than 20 ppm above the baseline (Figure 1).

In order to evaluate the extension of small bowel lesions, the patient underwent a capsule endoscopy (Given Imaging, Yoqneam, Israel). This examination showed the presence of villous atrophy with microerosions at the level of the medial jejunum and in the ileum, and some areas of lymphangiectasia (Figure 2).

After starting a lactose-free diet, the gastrointestinal symptoms rapidly disappeared; CA 19-9 returned to normal values (27 U/mL) and the patient regained his normal body weight. A capsule endoscopy was carried out again after six months; this examination showed the normalization of the previously described jejunum-ileum villous atrophy (Figure 3). The six months post milk free diet lactose breath test was also normal (Figure 4). At present, the patient is in good general health and continues a gluten- and cow milk-free diet.

Discussion

This case was of particular interest for the following reasons:

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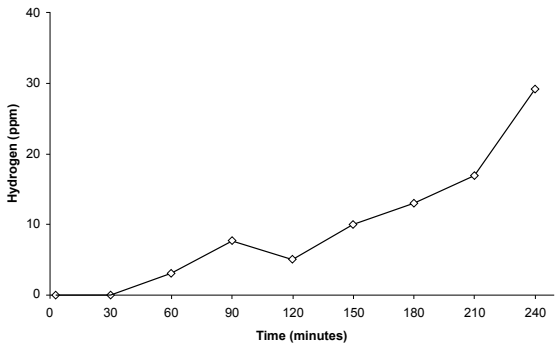
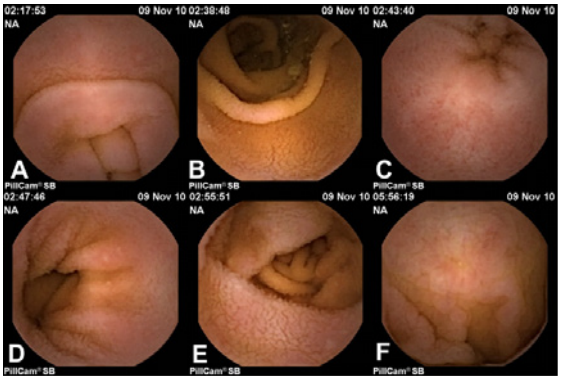
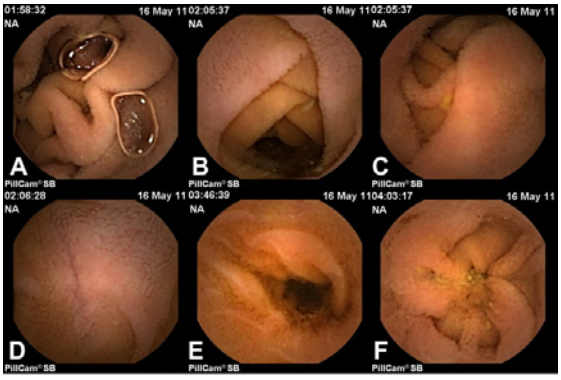


Figure 1: Lactose breath test. After a baseline breath sample was measured, 25 g of oral lactose was administered, and breath samples were taken every 30 minutes and the presence of hydrogen in parts per million (ppm) was determined by gas chromatography. Milk-induced malabsorption was diagnosed as demonstrated by the rise in hydrogen excretion of more than 20 ppm above baseline.



Panels A-D: jejunum with villous atrophy and microerosions (D)
Panels E,F: ileum with villous atrophy

Figure 2: Capsule endoscopy at diagnosis.



Panels A-D: normal jejunum
Panels E,F: normal ileum

Figure 3: Capsule endoscopy after 6 months.

first, the appearance of gastrointestinal symptoms, such as bloating, diarrhea and weight loss, due to the appearance of lactose intolerance [4]; second, the transient increase of CA 19-9 in celiac patients which was due to causes other than the presence of gastrointestinal cancer, which usually indicates in the general population [5], and third, capsule endoscopy was useful in detecting ileal mucosa abnormalities which disappear after going on a gluten- and cow milk-free diet.

Nonresponsive celiac disease is a common problem affecting 7-30% of celiac patients. Gluten exposure was the most common cause in one-third of nonresponsive celiac disease, but other causes can be recognized, such as irritable bowel syndrome, refractory celiac disease, microscopic colitis and lactose intolerance in about 8% of the cases [6]. It has also been reported that disaccharidase activity analysis offers additional information for evaluating the severity of mucosal villous atrophy especially during follow-up in patients with celiac disease; in fact, decreased activity of maltase or sucrase in these patients may be responsible for mucosal villous atrophy [3]. In addition, no patient with normal disaccharidase activity had severe villous atrophy [3]. Why cow milk intolerance can appear during the course of celiac disease is still under debate. Some authors have claimed that there are gluten-free peptides present in cow milk [7]; others have found that no significant amount of high molecular weight glutenin proteins can be detected in bovine milk, and the intolerance of celiac disease patients to bovine milk is not due to the presence of T-cell stimulatory epitopes of gluten [8]. The fact that the immunoglobulin A antibodies to deamidated gliadin peptides and the tissue transglutaminase antibodies were normal in our patient could support this latter hypothesis. Finally, because the H_2 breath test with lactose after eliminating the cow-milk from the diet was normal, the gluten proteins contamination in the milk lead to the patient's symptoms and findings.

Another interesting aspect of this case is the transient increase of serum CA 19-9; the levels of this marker returned to normal after he went on a cow milk-free diet. CA 19-9 is synthesized by normal human pancreatic and biliary ductular cells as well as by gastric, colonic, endometrial, and salivary epithelia [9]. Persistent high serum levels of this cancer marker have also been reported in serum CA 19-9 with no evidence of malignant disease [5] as well as in patients with non-malignant diffuse lung diseases, such as idiopathic pulmonary fibrosis, bronchiectasis, cystic fibrosis [10] and diabetes mellitus [11]. It has been shown that increased CA 19-9 serum levels are present in patients with hypothyroidism, and the gradual improvement of the thyroid function resulted in the resolution of the elevated CA 19-9 levels [12]. Thus, it is possible that, as in our patient, the serum levels of CA19-9 normalized after the disappearance of the ileal lesions.

Finally, current guidelines recommend that patients with refractory celiac disease undergo capsule endoscopy [13], even if the role of this technique in celiac patients is complementary. When careful history and serological markers rule out noncompliance with a gluten-free diet, capsule endoscopy is indicated for detecting complications, such as cancer, ulcerative jejunitis or ileitis and other associated conditions such as collagenous sprue [13]. Our report suggests the use of capsule

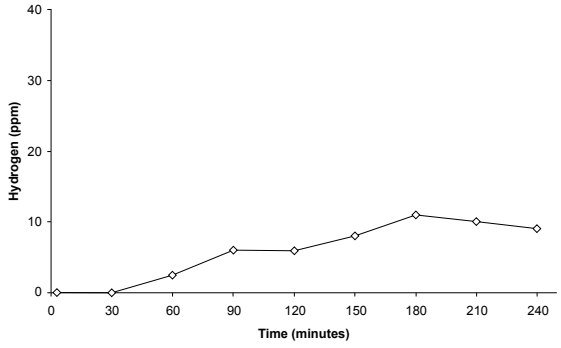


Figure 4: Lactose breath test after six months from the elimination of the cow-milk from the diet. The test demonstrated a normal hydrogen excretion.

endoscopy in this diagnostic approach can be utilized in celiac patients with symptomatic lactose intolerance for the detection of ileal mucosa abnormalities and their subsequent disappearance after starting cow milk-free diet.

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