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CASE REPORT

Celiac Disease. Does the Honeymoon Exist?

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Abstract

We present the case of a 7-year-old patient diagnosed with coeliac disease at the age of 6 months with compatible clinical, laboratory and histological findings. Improvement after gluten withdrawal. Because of the early diagnosis, gluten was reintroduced at 4 years of age and she remained asymptomatic for 3 years, at which time she began again with symptoms of abdominal pain, diarrhoea and stature stagnation. Laboratory tests showe delevated anti-celiac antibodies. This leads us to believe that there is a honey moon phase of the disease that has not been described in theliterature.

Keywords: Celiac disease, Child, debut, honey moon period.

1. Back Ground

Celiac disease (CD) is a systemic immunological process triggered by gluten consumptionin genetically pre disposed individuals. The average ageat diagnosis is 3.7 years in our country,which is almost exceptional below one year of age. The pathogenesis of this disease is not well understood. Patients may present with varying degrees of intestinal inflammationas well as variable extra intestinal symptoms.

Small bowel biopsy has beenthe gold standard for the diagnosis of CD since ancient times. Additionally, highly sensitive and specific serological tests are available for diagnosis and follow-up. The treatment of CD involves following a gluten-free diet, achieving clinical improvement, negativization of antibodies, and normalization of the histology. We present the case of a 7-year-old patient diagnosed with 6 months of CD who reintroduced gluten inherdiet at the age of 4years, with successive controls over 3 years in the normal range; therefore, for which reason we wonder if there is, as in other auto immune diseases such as diabetes, a period of partial regression of the disease or "honey moon period" orifit was a change of disease phenotype from an active to an asymptomatic, sub clinical or silent form.

2. Clinical Case

A 7-year-old girl with no personal or obstetric history, consulted Primary Care at 6 months of age for vomiting and refusal feedings two weeks after starting complementary feeding with gluten-containing cereals. After the withdrawal of gluten from the diet, she had clinical improvement. It was decided toper form a provocation test with gluten-containing cereals, and the previous symptoms re appeared together with ponderal stagnation.

From Primary Care, analytical study was requested with antigliadin antibodies of 54 IU/ml (normal value< 7 IU/ml) and IgA anti-transglutaminase antibodies 0.3 IU/ml (normal value< 7 IU/ml). Serum IgA, IgG and IgM were with int he normal values. It was requested a his to compatibility study (HLA) that showe positive riskalleles for CD DQ2 (HLA-DQA1 * O1:O1P HLA-DQA1 * O5:O1/ HLA-DQB1 * O2:O1 * HLA-DQB1 O5: O1). She was referred othepediatric gastroenterology service. At 8 months

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of life, a digestive endoscopy showed severe villous a trophy, cry pthyperplasia and intra epitheliallym phocytes in the pathological range, corresponding to a March IIIc grade (figure 1 and 2). Gluten was removed from the diet with clinical improvement at 12 months of life and normalization of analyticaltests at 14 months of life, remaining symptom-free in successive annual controls until 7 yearsof age. At 4years of age, gluten was reintroduced cover tly into the patient's diet, remaining asymptomatic and with serology in normal range for 3 years.

At 7.5 years of age, she presented clinical symptoms of abdominal pain and occasional diarrhea after meals, as well as growth retardation, with a drop from the 14th percentile (-1.06 SD) tothe 3rd percentile (-1.93 SD) (Figure 3). In the analytical control, an increase in IgA anti transglutaminase antibodies up to 101 IU/ml and positive anti endo mysial antibodies (++) was found.

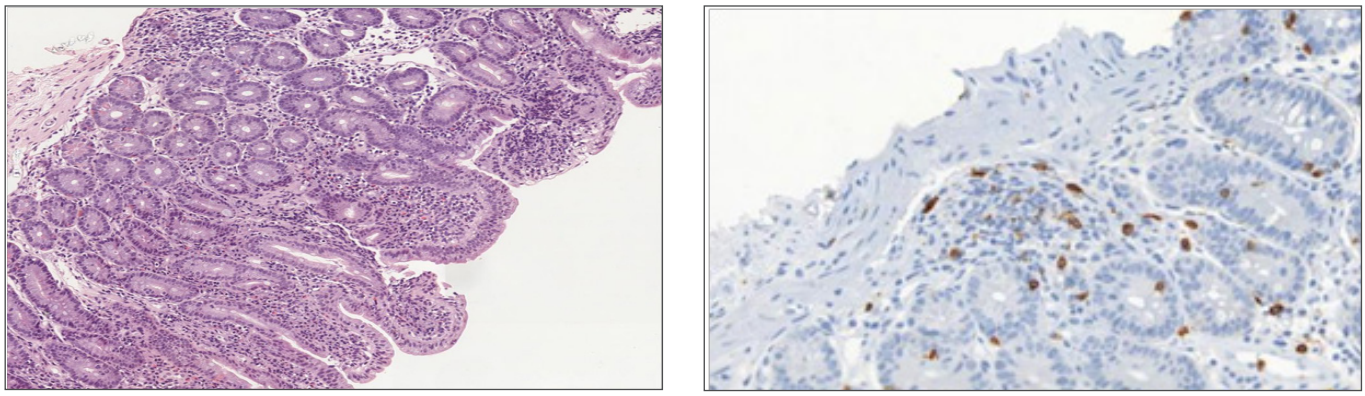

Figure 1, 2. *Severe villousatrophy, with crypthyperplasia and presence of intraepitheliallymphocytes/intraepitheliallymphocytosis in pathological range*

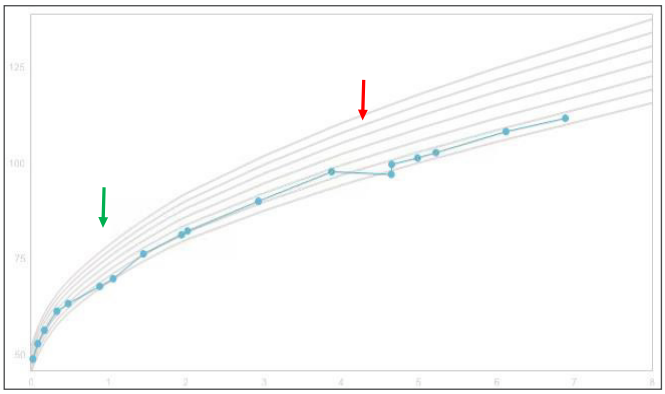

Figure 3. *Evolution of the size of the patient. Percentile drop at 6 months of age and recovery with the elimination of gluten from the diet (green arrow). Percentile drop with there introduction of gluten into the diet (red arrow)*

3. Discussion

Celiac disease is a systemic disease that causes intestinal malabsorption and can lead to a deficient state and/or severe malnutrition in infants. This is responsible for numerous clinical manifestations, some of which are paucis into matic,such as a failure to the rive.

According to the REPAC study[3], the a geat diagnosis is approximately 3.7 years, with diagnosis below 12 months of age being exceptional. The most common form in our country at this age is the classic form (71%, 91% in children under 2 years of age). The patient presented clinical signs of malabsorption, positive antibodies and villousatrophy. In our case, we high light a form of classic celiac disease presentationin a 6-month-old infant, the youngest in our cohort, which is rarely described in the literature.

CD is related too the autoimmune diseases such as auto immunethyroid disease, systemic lupus erythematosus, selective IgA deficiency, and type 1 diabetes mellitus. In the latter, there is a phase called the partialregression period of the disease or "honey moon phase", which has not been described in other autoimmune diseases such as celiac disease.

For diagnosis, we performed several tests: serological markers, histology and genetic study. Serological markers include IgA anti transglutaminase antibodies (together with the determination of total IgA that excludes IgA deficiency) and in children under 2 years of age, deaminated antiglidin antibodies.

According to the ESPGHAN[4] (European Society for Paediatric Gastroenterology, Hepatology and Nutrition) criteria updated in 2020, it is not necessary to performa biopsy with positive IgA anti transglutaminase antibodies with values10 times above the normal value and with positive anti-endo mysial antibodies in a second blood test.

HLA-DQ2 and/or DQ8 negative individuals have a very low risk of CD; a positive result does not confirm the diagnosis, but has a high negative predictive value Currently, the only the rapeutic solution to this pathologyis a gluten-free diet for life once the diagnosis is confirmed, which is why it is important toen sure it at such an early age in life, when most patients, despite the criteria described above, will under go an endoscopic study with biopsy.

4. Conclusion

The fact of having such an early diagnosis of celiac disease in our patient made us consider there introduction of gluten in thediet at 4 years of age. The patient remained clinically asymptomatic from the age of 12 months, although weight-stagnation was noted 3 years after starting to consume gluten again, which is why we think that during this time, the patient presented a latentor subclinical formor a type of honey moon phase of celiac disease not described in the literature.

Therefore, if a patient under one year of ageis diagnosed with CD and it is decided to reintroduce diet as a provocation test, the consequences of malabsorptive syndrome should be assumed and an even closer follow-up of the disease should be carried out. It would also be interesting to consider a special study and follow-up protocol for patients diagnosed with CD under one year of age.

5. Acknowledgement

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