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Pan-Uveitis in Celiac Disease: A Case Report

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Abstract **Case Report**

Cealiac disease (CD) is an autoimmune disease, where autoantibodies (anti-transglutaminase (TG)2/anti- endomysial antibodies) attack mainly the intestinal tract. This disease is associated with several other autoimmune diseases. Ocular involvement in CD is often secondary to the intestinal malabsorption syndrome following the vitamin deficiency, so uveitis is rarely reported in the literature. We report the case of a 14-year-old girl, followed for coealiatic disease and type 1 diabetes, admitted for panuveitis of the left eye resistant to corticosteroid therapy, with a well response to a strict gluten-free diet. Uveitis secondary to coeliac disease is probably under-diagnosed, and some studies suggest the necessity to try a strict gluten-free diet in all idiopathic cortico-resistant uveitis.

Keywords: Cealiac disease (CD), anti-transglutaminase, autoimmune diseases, Ocular involvement.

INTRODUCTION

Celiac disease (CD) is a chronic autoimmune enteropathy, occurs in about 1% of the general population with a genetic predisposition [1], characterized by food intolerance to gluten, the treatment is mainly based on a strict gluten- free diet [2]. It is a polymorphic clinical entity involving several organs, mainly the intestinal tract (small intestine) [3]. CD has been associated with a wide spectrum of extraintestinal manifestations, such as dermatological, neurological and hepatic, which may occur in up to 50% of patients [4]. Ocular involvement is rare, and may be secondary to intestinal malabsorption syndrome by dry eye, nyctalopia, cataract, or to other autoimmune diseases associated with celiac disease, notably autoimmune thyroiditis by an orbitopathy, type 1 diabetes by diabetic retionpathy, neuromyelitis optica, orbital myositis or central retinal vein occlusion [5].

Then, uveitis during celiac disease is rarely described and is characterized by inflammation of the uveal tract [6]. We report a case of panuveitis in a young girl, followed for celiac disease since 4 years well balanced on a strict gluten- free diet.

CASE REPORT

A fourteen-year-old girl was admitted to the ophthalmology department because of sudden vision loss in her left eye. The medical history revealed a control of a CD since 1 year by a strict gluten-free diet without any episode of diarrhea or abdominal pain, and a type 1 diabetes mellitus for one month with good glycemic control. Ocular examination of the right eye was unremarkable with a best corrected visual acuity at 10/10. The best corrected visual acuity of the left eye was 1/20, with a macular syndrom with metamorphopsia and alteration of contrast. Slit lamp examination revealed some small and fine keratic precipitates, with tyndall in the anterior chamber at two cross, without any irian nodules or posterior synechiae, foundus examination found a hyalitis at two cross, a papillitis and an important serous retinal detachment, that was confirmed by angiography and macular optical coherence tomography (Figure 1- 2).

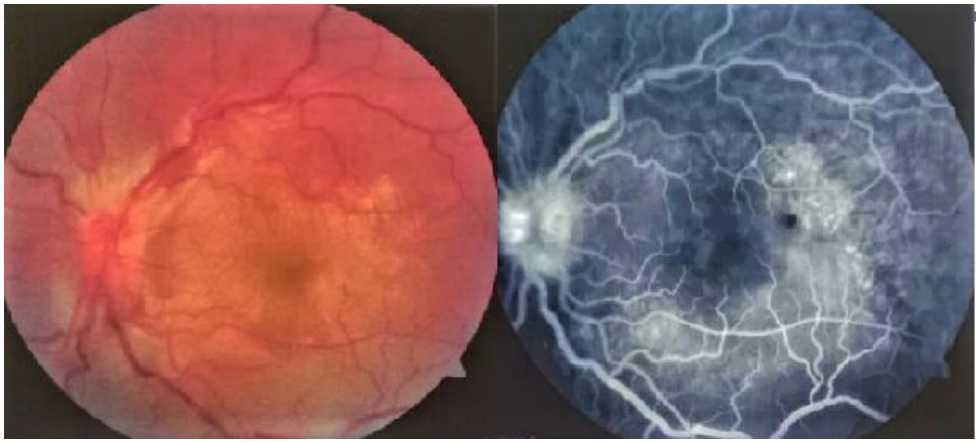


Figure 1: Fluorescein angiography: Papillitis, large serous retinal detachment, with the typical smokestack

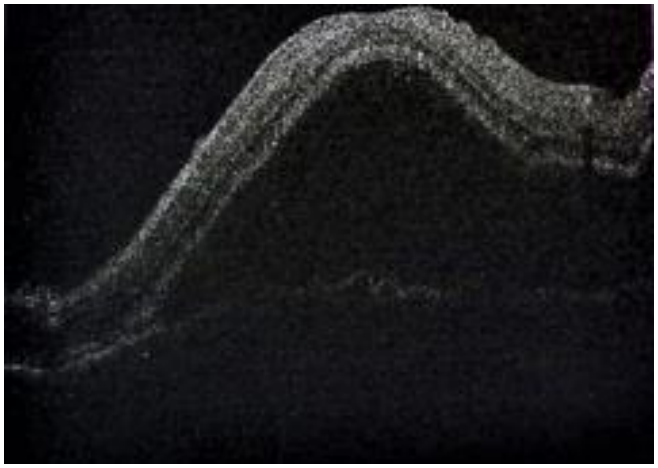


Figure 2: Optical coherence tomography: Important serous retinal detachment

The patient was hospitalized, where biological tests were performed to exclude an infectious (tuberculosis, syphilis) or inflammatory (sarcoidosis, juvenile idiopathic arthritis) causes of the panuveitis. No etiology was found, and the patient did not respond to treatment with topical and systemic corticosteroids. While hospitalized, our patient presented a severe abdominal pain with diarrhea. A biopsy of the intestinal tract was indicated to clarify the digestive involvement, in particular by eliminating chronic inflammatory bowel disease (Crohn's disease and ulcerative colitis), the biopsy objectified a subtotal villous atrophy with an increase in intraepithelial lymphocytes, which confirms the diagnosis of CD, we completed by anti-transglutaminase antibodies which were positive. The rest of the biological test showed a hypochromic microcytic anemia due to iron deficiency at 8g/dl, a vitamin deficiency (Vitamin A - Vitamin D - Vitamin B12), and recurrent episodes of hypoglycemia were found during her hospitalization. This significant biological metabolic malabsorption syndrome testified of a poor control of her celiac disease, despite the gluten-free diet followed by the patient.

On the therapeutic plan, we proceeded to a progressive degression of the topical and systemic corticotherapy, then the patient benefited from a nutritional consultation, her body mass index was (BMI) 17.5 kg/m2, the nutritional investigation found the consumption of some foods containing gluten unknown by the patient, such as brewer's yeast that the patient took to gain weight, so a normocaloric, hyperproteinic and strict gluten-free diet was instituted. After 4 weeks, we have a regression of abdominal pain and episodes of diarrhea from 4 episodes per day to 3 per week, with a regression of the retinal serous detachment and persistence of a small retro-foveal fibrosis, a vitreous tyndall at 0.5 cross and an anterior segment without anomalies. At the end of the 6th month, under diet, the patient was doing well on a general level with a BMI of 22kg/m2, and a total acalmia of her uveitis without any relapse, with a visual acuity of 8/10.

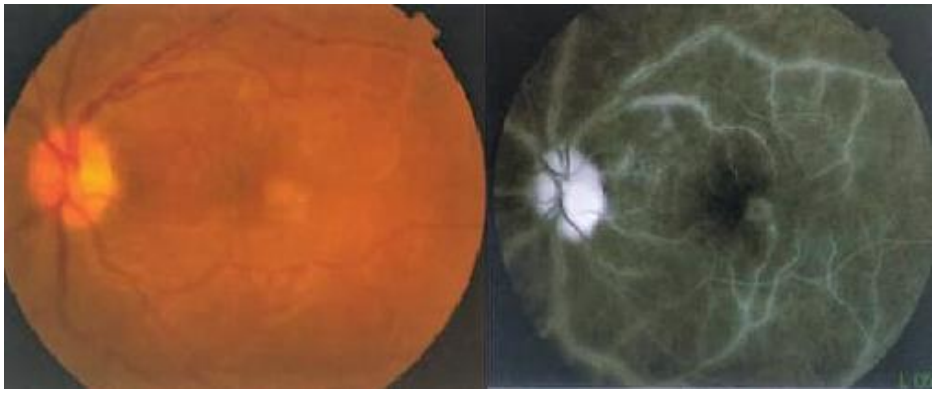
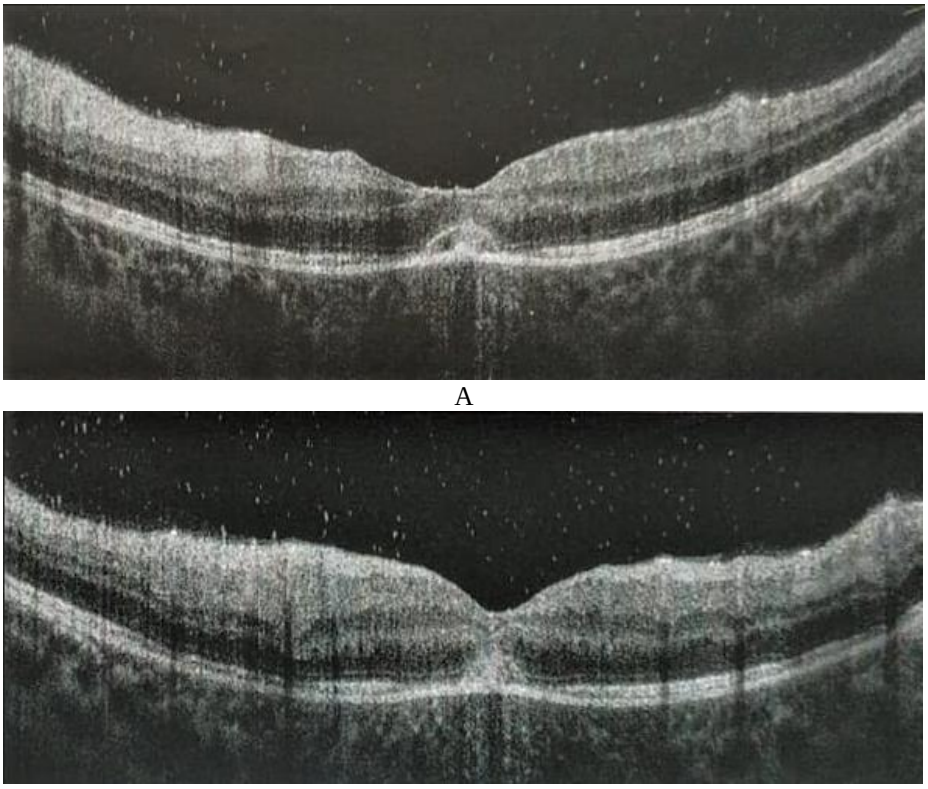


Figure 3: Fluorescein angiography: Total regression of the papillitis, and partial regression of the serous retinal detachment



A

B

Figure 4: Optical coherence tomography: A =1 month: Regression of the serous retinal detachment / B = 6 month: Persistance of a retro-foveolar fibrosis

DISCUSSION

Celiac disease (CD) was initially considered as a gluten sensitivity responsible for digestive symptomatology, nowadays it falls within the spectrum of autoimmune diseases, where gluten and prolamins are responsible for a systemic immune-mediated cascade in genetically sensitive individuals, this immune response produces specific autoantibodies [anti-transglutaminase (TG)2/anti-endomysial antibodies (EMA)], of HLA-DQ2 and/or DQ8 haplotypes responsible for different degrees of enteropathy, ranging from lymphocytic infiltration of the epithelium to complete villous atrophy [1, 7].

The autoimmune character of CD is further confirmed by its close association with certain autoimmune disorders, such as insulin-dependent diabetes mellitus -as in the case of our patient-, thyroiditis (Hashimoto's thyroiditis), dermatitis herpetiformis, Addison's disease, autoimmune myocarditis, autoimmune hepatitis [8].

The ophthalmic manifestations of CD are mainly secondary to the intestinal malabsorption syndrome and are responsible for ocular dryness, cataract, xerophthalmic fundus by hypovitaminosis A [9]. Uveitis during CD is poorly reported in the literature, so we have no idea about the type of uveitis that CD can cause but all the uveal tract can be affected,

so these uveitis can be anterior, intermediate, posterior or mixed [6, 10]. Several studies consider that uveitis associated with CD is under-diagnosed, and that all idiopathic uveitis not responding to corticosteroid therapy should be evaluated in the sense of looking for asymptomatic or minimally symptomatic CD underneath, and the response of the uveitis to the gluten-free diet is a test treatment that allows us to orient ourselves towards this origin [11]. Kaziwe *et al.*, performed a cohort study to investigate the risk of developing uveitis in a patient with CD compared to an age- and sex- matched population. The hazard ratio HR is 1.32, with a risk that remains significant even 5 years after the initial diagnosis of CD, The absolute risk of uveitis was 50/100 000 person-years in CD, corresponding to a moderat risk of developping an uveitis in CD [3].

CONCLUSION

Coealiatic disease should be one of the diagnoses to be evoked in front of uveitis, mainly when the uveitis is considered idiopathic, in spite of the absence of digestive signs. The response of the uveitis to a trial treatment with a strict gluten-free diet will allow to orientate the diagnosis, and to complete it by the dosage of antibodies anti- transglutaminase (TG) 2/anti-endomysial antibodies and a biopsy of the intestinal tract.

REFERENCES

1. Rubio-Tapia, A., Ludvigsson, J. F., Brantner, T. L., Murray, J. A., & Everhart, J. E. (2012). The prevalence of celiac disease in the United States. *Am J Gastroenterol*, 107(10), 1538–1544.
2. Rubio-Tapia, A., Hill, I. D., Kelly, C. P., Calderwood, A. H., & Murray, J. A. (2013). ACG clinical guidelines: diagnosis and management of celiac disease. *Am J Gastroenterol*, 108(5), 656–676.
3. Mollazadegan, K., Kugelberg, M., Tallstedt, L., & Ludvigsson, J. F. (2012). Increased risk of uveitis in coeliac disease: a nationwide cohort study. *British journal of ophthalmology*, *96*(6), 857-861.
4. Laurikka, P., Nurminen, S., Kivelä, L., & Kurppa, K. (2018). Extraintestinal manifestations of celiac disease: early detection for better long-term outcomes. *Nutrients*, *10*(8), 1015.
5. Fousekis, F. S., Katsanos, A., Katsanos, K. H., & Christodoulou, D. K. (2020). Ocular manifestations in celiac disease: an overview. *International Ophthalmology*, *40*(4), 1049-1054.
6. Lachar, R., Ouazzani, BT, Laghmari, M., Ibrahimy, W., Cherkaoui, O., Skiker, H., ... & Daoudi, R. (2008). 635 Ocular damage in celiac disease. *French Journal of Ophthalmology*, *31*, 193. DOI : 10.1016/S0181-5512(08)71234-8
7. Troncone, R., & Jabri, B. (2011). Coeliac disease and gluten sensitivity. *Journal of internal medicine*, *269*(6), 582-590. doi.org/10.1111/j.1365-2796.2011.02385.x
8. Bizzaro, N., Villalta, D., Tonutti, E., Tampoia, M., Bassetti, D., & Tozzoli, R. (2003). Association of celiac disease with connective tissue diseases and autoimmune diseases of the digestive tract. *Autoimmunity reviews*, *2*(6), 358-363. doi.org/10.1016/S1568-9972(03)00055-7
9. dos Santos Martins, T. G., de Azevedo Costa, A. L. F., Oyamada, M. K., Schor, P., & Sipahi, A. M. (2016). Ophthalmologic manifestations of celiac disease. *International Journal of Ophthalmology*, *9*(1), 159-162. doi: 10.18240/ijo.2016.01.26
10. Krifa, F., Knani, L., Sakly, W., Ghedira, I., Essoussi, A. S., Boukadida, J., & Hamida, F. B. H. (2010). Uveitis responding on gluten free diet in a girl with celiac disease and diabetes mellitus type 1. *Gastroentérologie clinique et biologique*, *34*(4-5), 319-320.
11. Klack, K., Pereira, R. M. R., & de Carvalho, J. F. (2011). Uveitis in celiac disease with an excellent response to gluten-free diet: third case described. *Rheumatology international*, *31*(3), 399-402. DOI: 10.1007/s00296-009-1177-z