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# Pica Syndrome Revealing Celiac Disease: A Case Report

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**Abstract** | **Case Report**

Celiac disease is an autoimmune mediated chronic inflammatory enteropathy primarily affecting the small intestine, occurring in genetically predisposed individuals and caused by a permanent sensitivity to ingest gluten cereals. Pica is the compulsive eating of nonnutritive substances, and may be either a cause or a result of iron deficiency, with the latter being a possible presenting sign of Celiac Disease. We report the case of an 18-year-old female patient with history of depression and pica disorder. Laboratory investigations, serology testing and duodenal biopsy showed arguments of celiac disease with spectacular improvement noted after initiating the gluten free diet.

**Keywords:** Celiac disease, pica syndrome, Behavioral disorder, Malabsorption syndrome.



## INTRODUCTION

Celiac disease (CD) is an autoimmune chronic disorder developed as result of interplay between genetic and environmental factors [1]. It's an inappropriate immune response to gluten. Patients may present typical symptoms like diarrhea, steatorrhea, abdominal distension, edema and weight loss. In some cases, it may be oligosymptomatic, or may present atypical manifestation such as emotional disorders and psychiatric disturbances, including eating disorders (EDs) like Pica syndrome [2]. The term "pica" comes from the medieval Latin word for magpie, a bird known for its random and large appetite [3]. Pica is an eating disorder, commonly seen in children and pregnant women, and defined as the excessive and repeated ingestion of non-nutritive or atypical food combinations over the course of 1 month such as chalk, soil, wool, gypsum, pebbles or rocks [4, 5]. A specific name is given to describe each different eating preferences, trichophagia describes preference to ingest hair or wool such as the case of our patient who used to eat her duvet. The exact cause of pica remains unclear, but it is significantly associated with nutritional deficiencies especially iron deficiency anemia which may be either the result or the etiology of pica [6].

## CASE REPORT

We report a case of 18-year-old female patient with a history of depression and pica disorder who used to eat her duvet when she was alone. The patient was referred for evaluation of persistent abdominal pain with an anemic syndrome, weight loss and asthenia, associated with mood swings. Clinical examination revealed a cachectic patient with a BMI at 23kg/m2 and a pallor. Vital parameters were within the normal range; without any abdominal sensitivity or defense.

At admission, laboratory tests showed a microcytic hypochromic anemia (hemoglobin = 9g /dl, MCV = 75 u3, CCMH = 28%), a normal white cell and platelet count with signs of malabsorption: Iron at 13 mg/dL, ferritin at 9 ng/mL (NR 12-150 ng/mL) hypo albuminemia at 29 mg/l. Other blood tests, notably, *liver* and *kidney function tests* were normal.

Serology testing showed an elevated level of anti-endomysial antibodies (IgA EMA) (and IgA anti-tissue transglutaminase *antibodies* (*tTg*-IgA). Celiac disease was suspected. An esophagogastroduodenoscopy (EGD) was realized with the obtention of multiple biopsies. Histopathological analysis revealed normal esophagus and stomach. On duodenal level, there were intraepithelial lymphocytosis (IEL>30/100 epithelial cells), and subtotal villous atrophy.

The histological findings, positive serology testing and clinical features all supported the presumptive diagnosis of CD.

A gluten-free diet was initiated with the resolution of all symptoms including the pica disorder. The patient's follow up was very promising as she gained 6kg. Hb was 11g/l and antibodies levels normalized. A duodenal biopsy was performed 1 year later, revealing a normal intestinal mucosa.

## DISCUSSION

Celiac disease (CD) is an inappropriate immune response to gluten, which is an autoimmune chronic disorder developed as a result of interplay between genetic and environmental factors [1]. There is a concept known as the "celiac iceberg" in which the whole prevalence of CD equates to the total size of the iceberg with the submerged portion equating patients with undiagnosed CD and the visible tip of the iceberg representing the diagnosed cases [7]. This fact proves that celiac disease, also known as gluten-sensitive enteropathy, often goes unnoticed, as was the case with our patient where the diagnosis of CD was confirmed after an initial discovery of atypical eating disturbance, PICA Syndrome.

Cause-and-effect relationship between PICA syndrome and CD remains unclear, and is often a result of metabolic disturbances due to malabsorption syndromes resulting in deficiencies of iron, thiamine (Vitamin B1), tryptophan, folic acid (Vitamin B9), pyridoxine (Vitamin B6), and cyanocobalamin (Vitamin B12), and not primary psychological disorders [8].

Diarrhea and lethargy are the most common symptoms of CD but other gastrointestinal signs like abdominal discomfort or pain, constipation or vomiting may also exist. Unexplained weight loss in adulthood is the main aspect of the history, while in childhood the corresponding sign would be failure to thrive. Extraintestinal signs are also possible and may include an anemia due to defective absorption of iron, vitamin B12 or folate, a coagulopathy due to impaired absorption of vitamin K, osteoporosis, or neurological symptoms like paresthesia and seizures [9, 10]. In fact, in many cases, CD can be revealed by atypical, paucisymptomatic or extradigestive manifestations. It has long been associated with emotional, cognitive, and neurodegenerative disorders and studies have outlined a psychiatric history in a considerable proportion of newly CD diagnosed adults [11, 12].

CD had been reported to occur in association with eating disorders (ED) in some case reports [8, 13]. Some studies have examined the link between ED and CD, and found an increase in altered or disordered eating behavior in CD patients compared with healthy controls and a higher-than-expected rate of ED in adolescents with CD [14-16].

Iron deficiency can be a presenting sign of CD as a result of malabsorption. In the same time, it has been proposed as one cause of Pica, and the complete resolution of pica disorder has been reported in 3 children with comorbid CD and pica following the initiation of a gluten free diet [17].

Basically, even though the connection between eating disorders and CD are controversial, it is possible that an incorrect diagnosis of Pica syndrom can obscure the diagnosis of CD, while on the other hand CD can produce eating disordered behaviors [18,19].

A positive association between CD and eating disorders such as Pica, is not supported by many studies. A retrospective study reviewing 494 individuals who had been evaluated for an eating disorder and screened for CD, found 2% (equals 10 patients) who tested positive for anti-tTG antibodies. 4 of these patients had biopsy confirmed CD via endoscopy thus the overall prevalence rate being at 0.8%. This prevalence almost equals the rate found in the general population. Therefore, there has been no suggestion of routine screening of individuals with EDs for CD [20].

## CONCLUSION

Our case highlights that CD may manifest quite abruptly with a severe malabsorption syndrome, that is, electrolyte abnormalities and hypoproteinaemia. That's why clinicians should remain vigilant to evoking CD when a patient presents with symptoms of an eating disorder such as Pica, while also exploring the possibility of disordered eating behavior in patients with CD.

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