

Lithophagia as a clue for celiac disease: a case report and literature review

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Abstract

Lithophagia is a type of pica that might be resulted from Iron Deficiency Anemia (IDA) which is the frequent presenting signs of Celiac Disease (CD). A 5-year-old child with a two year history of the lithophagia with a, refractory IDA, abdominal distention and constipation. The child did not grow well and had failure to thrive. With suspicion to CD, TTg IgA level was measured and due to an increase of TTg IgA level the patients were undergone esophagogastroduodenoscopy and jejunal biopsy. The biopsy showed severe villous atrophy and an increase in lymphoplasmic cells. Biopsy confirmed diagnosis of CD and gluten free diet was initiated finally. Six months after diagnosis and commencing the gluten free diet, the lithophagia and constipation in patient eradicated completely. IDA and failure to thrive were improved and the level of TTg IgA was reached to the normal. The case demonstrated the relationship between lithophagia and CD in anemia. Therefore, in the same cases such as our case should be considered CD as the most important causes of lithophagia.

Keywords: Lithophagia, Pica, Celiac disease, Iron deficiency anemia

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Introduction

Celiac disease (CD) is an autoimmune mediated chronic inflammatory gluten sensitive bowel disorder with strong evidence of T- cell mediation, occurring in genetically predisposed individuals. (1 ,2). Its prevalence varies in different subgroups from 0.5 to 12 percent (3, 4 ,5).

CD in children could present with extra intestinal signs, such as short stature, delayed puberty, Iron Deficiency Anemia (IDA), Eating Disorders (ED) and Osteopenia (6,7,8,9,10,11). From the extra gastro-intestinal point of view, IDA is the most involvement reported; also this IDA is not responsible for Iron supplementation (8). As mentioned, IDA and eating disorders are signs of malabsorption, even in patients without gastrointestinal symptoms (8,10).

Pica is an irresistible desire for consumption of non-nutritive and unusual substances, such as soil, chalk, gypsum, ice and pebbles or rocks which could be documented in IDA, ED, chronic renal failure, and pregnancy (10,12). Lithophagia, a type of Pica, is an extremely rare reported desire of eating pebbles or stone fragments (12). Some associated

complications have been reported with lithophagia, including intestinal obstruction (13) and colon perforation (14).

This report describes a child with long term history of lithophagia with the ultimate diagnosis of CD.

Case Report

A 5-year-old boy with a two year history of the lithophagia, was treated for IDA several times. He had been diagnosed with hypochromic microcytic anemia, but he showed no response to repeated therapeutic iron supplementation. His history revealed several episodes of abdominal distention and long standing constipation that aggravated with iron supplements. Also, he had temptation to lithophagia. Recently, he developed abdominal pain and distention and no defecation for at least 15 days. On arrival, he seemed chronically ill, but his weight and height were acceptable for his age (height=110Cm, weight=20 Kg); moreover, he belonged to a high socio-economic family.

Physical examination documented abdominal distention with decreased bowel sounds but no evidence of organomegaly. Due to his avoidance of rectal examination, plain abdominal X-ray was taken for evidence of fecal impaction, revealing multiple, well-defined radio-opaque foreign bodies in all parts of his colon. (Figures 1, 2)

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Table 1. Patient's laboratory data

Indices	Early measure	After 6-months
Heamoglobin (g/dL)	80	120
mean circular volume (fl)	55	78
RBC (/μL)	3300000	4100000
WBC (/μL)	10000	7000
RDW (fL)	18	14
PLT (/μL)	600000	300000
Albumin (mg/ml)	4	4.5
Total protein (mg/ml)	6.5	7
TTG IgA (ng/ml)	400	8
Total IgA (ng/ml)	130	125
Weight (kg)	20	26
Length (cm)	110	112

According to his laboratory assessments, he had heamoglobine 80g/dl, mean circular volume 55fl, RBC 3300000, WBC 10000, RDW 18, total protein 65g/l, and albumin 40g/l. With this history of IDA unresponsive to iron supplementations, we were suspicious to CD and total IgA and Anti Tissue Trans-Glutaminase Antibody (TTG-IgA) was measured; TTG-IgA was 400 (normal=20) and total IgA was in a normal range. After that, Esophagogastrointestinal endoscopy was done and multiple biopsy was obtained. Histological examination revealed normal esophagus and stomach but duodenum biopsies documented Marsh-III villus atrophy with an extensive epithelial lymphocyte infiltration in favor of CD. Table(1)

The subject commenced lactolose with a dose of 3 ml /Kg/ day divided into two equal doses for disimpaction and Gluten free diet. The patient passed rocks without any complication 3 weeks after initiation of lactolose (Figure 3) and his desire to lithophagia was diminished with Gluten free diet.

Discussion

Celiac disease is a frequently observed genetic disorder with a prevalence of 2.67% in the general population and the characteristic immunologic response to gluten that might be silent. (5,15) There is a documented association between CD and iron deficiency anemia and ED. (9, 10) Pica is an eating disorder presented with consumption of strange non-nutritive substances like soil, chalk, gypsum, ice and pebbles or rocks. (7,9,10)

This case was a 5-year-old patient with acceptable growth and development according to his age that presented with lithophagia, IDA, constipation, and abdominal distention.

Eyad Altamini reported a 4.5 year old girl with lithophagia and IDA that was ultimately diagnosed as CD although that girl had poor weight gain, edema in lower extremities, hypoalbuminemia, chemical and radiological rickets, and growth parameter lower than the 3rd percentile (12). But nowadays

there are studies documented CD in patients with normal growth, even obese subjects (7,9).

Stanley Korman presented a case series of three patients with long standing IDA and lithophagia as Pica that had characteristics of CD in serology and jejunal biopsy; he concluded that CD must be noticed in any child manifesting with Pica and IDA, especially in growth retarded patients (6).

Daniel, et al. demonstrated the relationship between CD and EDs in a series of 10 patients, stating the need for multi-disciplinary evaluation and care for these patients (9).

The clinical picture of IDA varies from case to case and symptoms depend on the severity, underlying disease and patient's characteristics. A proved symptom of IDA is Pica that is due to reduction of iron – containing enzymes and this pica responds quickly to treatment (8,9). IDA patients without bleeding causes, such as menstruation or occult blood loss, were referred to a gastroenterology consultation; more than half of them had pica that can be an explanation for lithophagia in CD (9). IDA is abundant in digestive pathology with two reasons of a reduction in quality of life and a consequence of gastrointestinal diseases such as CD. IDA is the most frequent extra-intestinal manifestation of CD, so it is crucial (8).

Many studies have been reported the relationship between EDs and CD and their association with personal behaviours, other personality disorders and mal-absorption due to CD, as causative factors (9). Consumption of strange substances such as rocks, ice and soil in the domain of EDs has been reported. A scarce report has evaluated the relationship of gastrointestinal pathology in the presentation of EDs and Functional Gastrointestinal disorders. It is greatly documented that Pica could be due to metabolic disturbances but not primary behavioral or psychological disorders (9,10).

Evaluation of this case suggests the necessity of serological and histological evaluation for CD in children with Pica, especially lithophagia along with IDA even if their growth parameters are in a normal range.



Figure 1. Erect anteroposterior radiograph that shows rocks in all parts of colon



Figure2. Lay down anteroposterior radiograph that shows rocks in all parts of colon



Figure3. Anteroposterior radiograph after treatment that shows no rocks in colon

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References

1. Shahramian I, Dehghani SM, Haghighat M, Noori NM, Teimouri A, Sharafi E, et al. Serological evaluation of Celiac disease in children with congenital heart defect; a case control study. *Middle East J Dig Dis* 2015; 7: 98-103.
2. Noori NM, Teimouri A, Nakhaey Moghaddam M, Shahraki T. The Prevalence of Celiac Disease in Down syndrome Children with and without Congenital Heart Defects. *Int J Pediatr* 2016; 4: 2143-52.
3. Shahramian I, Dehghani SM, Haghighat M, Noori NM, Teimouri AR, Sharafi E, et al. Serologic evaluation of Celiac disease in patients with beta thalassemia major and control. *Gastroenterol Hepatol Bed Bench* 2015; 8: 153-9.
4. Shabbazkhani B, Forootan M, Merat S, Akbari MR, Nasserimoghaddam S, Vahedi H, et al. Celiac disease presenting with symptoms of irritable bowel syndrome. *Aliment Pharmacol Ther.* 2003; 18: 231-5.
5. Husby S, Koletzko S, Korponay-Szabó IR, Mearin ML, Phillips A, Shamir R, et al. European Society for Pediatric Gastroenterology, Hepatology, and Nutrition guidelines for diagnosis of Celiac disease. *J Pediatr Gastroenterol Nutr* 2012; 54: 136-60.
6. Korman SH. Pica as a presenting symptom in childhood celiac disease. *Am J Clin Nutr* 1990; 51: 139-41.
7. Fotoulaki M, Panagopoulou P, Efstratiou I, Nousia-Arvanitakis S. Pitfalls in the approach to pica. *Eur J Pediatr* 2007; 166: 623-4.
8. Bermejo F, García-López S. A guide to diagnosis of iron deficiency and iron deficiency anemia in digestive diseases. *World J Gastroenterol* 2009; 15: 4638-43.
9. Leffler DA, Dennis M, Edwards George JB, Kelly CP. The interaction between eating disorders and Celiac disease: An exploration of 10 cases. *Eur J Gastroenterol Hepatol* 2007; 19: 251-55.
10. Louw VJ, du Preez P, Malan A, van Deventer L, van Wyk D, Joubert G. Pica and food craving in adult patients with iron deficiency in Bloemfontein, South Africa. *S Afr Med J* 2007; 97: 1069-71.
11. Njiru H, Elchalal U, Paltiel O. Geophagy during pregnancy in Africa: a literature review. *Obstet Gynecol Surv* 2011; 66: 452-9.
12. Altamimi E. Lithophagia in iron-deficient patient with Celiac disease. *J Pediatr Gastroenterol Nutr* 2014; 59: e49.
13. Chand M, Livesey E, McCarthy L, Davenport M. A sticky case of intestinal obstruction. *Pediatr Surg Int* 2007; 23: 707-9.
14. Ilhan Y, Cifter C, Doğru O, Akkuş MA. Sigmoid colon perforation due to geophagia. *Acta Chir Belg* 1999; 99: 130-1.
15. Rostami Nejad M, Aldulaimi D, Ishaq S, Ehsani-Ardakani MJ, Zali MR, Malekzadeh R, et al. Geographic trends and risk of gastrointestinal cancer among patients with celiac disease in Europe and Asian-Pacific region. *Gastroenterol Hepatol Bed Bench.* 2013; 6: 170-7.