

Case Report

Celiac Disease with Intestinal Malrotation - A Rare Phenomenon.

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Abstract

Introduction: Celiac disease has immune based autoimmune disease which is noticed in genetically predisposed persons and is due to gluten allergy. It was initially thought that it is disease of children and presents with typical or classical symptoms associated with gastro-intestinal tract like pain abdomen, diarrhea or constipation. Now, it has been proven that it is commonly seen in adult age also and in half of cases presents with atypical or unclassical symptoms which are mainly due to nutrients or minerals deficiency. Thus, patients can have neurological, psychiatric, dental or reproductive problems. Intestinal malrotation is seen in many cases but in association with celiac disease is rare and very few cases are reported in literature.

Case Report: We report a case of sixteen-year-old child who was known case of celiac disease for last three years but was intermittently taking gluten in diet. He presented with severe pain abdomen for last two months and his serum IgATTG antibody were raised to 80 I.U. He was restarted on strict gluten restriction but pain abdomen did not subside. Moreover, his pain abdomen was severe, requiring injectable analgesics intermittently. His pain abdomen was different which is usually seen in celiac disease i.e. generalized and dull aching. His ultrasonogram abdomen was normal but in view of non-resolution of pain abdomen, he was subjected to contrast enhanced computed tomography scan which showed intestinal malrotation, all his small bowel was on left side and large bowel was on right side. He was advised surgical consultation and has yet to come for review after surgical consultation.

Conclusion: Malrotation can be seen more commonly than expected. Even though it has traditionally been thought as a disease of the infants, it can present later in life and tends to have non-specific gastrointestinal symptoms. Clinicians need a high index of suspicion when assessing patients with these symptoms which poses more challenge in background of celiac disease.

Key words: Celiac disease, Gluten, Intestinal malrotation, Serum IgATTG antibody, Endoscopy

INTRODUCTION

Celiac disease has immune based autoimmune disease which is noticed in genetically predisposed persons and is due to gluten allergy [1-3]. The global prevalence of CeD has been estimated at around 1.7% based on positive serology and 0.7% based on biopsy-confirmed CeD [4]. It was initially thought that it is disease of children and presents with typical or classical symptoms associated with gastro-intestinal tract like pain abdomen, diarrhea or constipation. Now, it has been proven that it is commonly seen in adult age also and in half of cases presents with atypical or unclassical symptoms which are mainly due to nutrients or minerals deficiency. Thus, patients can have neurological, psychiatric, dental or reproductive problems. Celiac disease and intestinal

malrotation can coexist and pose diagnostic challenges, as both can cause gastrointestinal symptoms, but they are distinct conditions. Celiac disease is an autoimmune disorder triggered by gluten that damages the small intestine's villi. Intestinal malrotation is a congenital condition where the intestines don't form or position correctly in the abdomen, which can lead to twisting and blockage. Some research suggests a potential association, though not a direct causal link, making careful evaluation for both conditions important, especially with non-specific gastrointestinal symptoms.

CASE REPORT

A fourty two-year-old male presented with unexplained transaminitis for last one year and after ruling out all other

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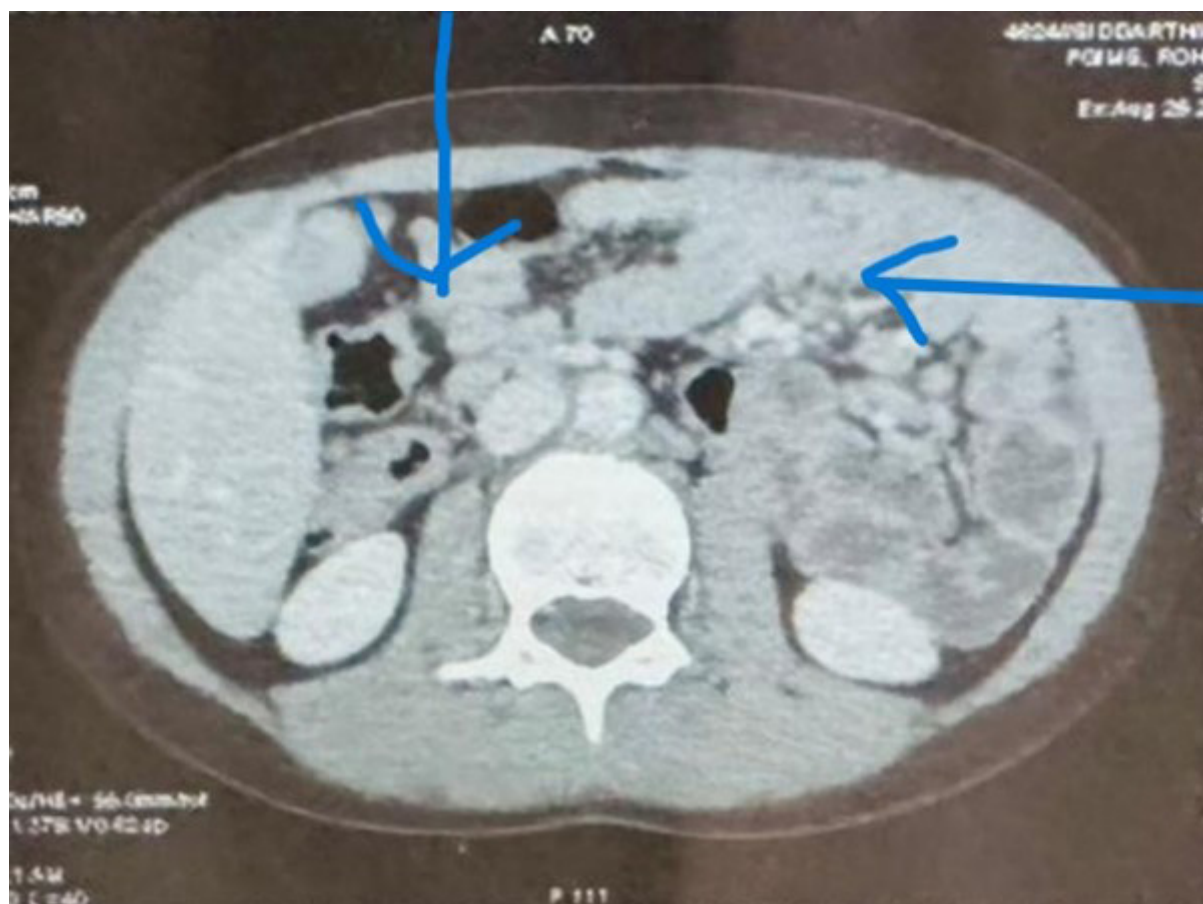
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causes, he was diagnosed to be celiac disease on basis of serological, endoscopic and histopathological examination of duodenal biopsy. He was put on strict gluten restricted diet and within three months transaminases became normal and after one year of follow up on strict gluten restricted diet, his transaminases are normal. As per protocol, his family was screened for celiac disease and his wife was normal but his seven-year-old son who was asymptomatic, showed significant rise of serum IgATTG antibody level of 110 IU/ml, whereas normal range was 0-20 IU/ml. He was subjected to upper gastro-intestinal endoscopy which showed moderate scalloping of duodenal folds and on histopathological examination proved it to be celiac disease with Marsh Grade 3 stage. On general physical examination, the teeth of this child had dental enamel defect selectively in upper incisor and canines which parents admitted that it was for last two years. His rest examination including systemic one was normal. All his biochemical tests including hemogram, blood sugar, thyroid profile was normal except for slightly low level of serum calcium and serum Vitamin D3 level. He was immediately put on strict gluten restricted diet, along with calcium and vitamin D3 supplementation. After six months of gluten restricted diet, his Serum IgATTG antibody, Calcium, Vitamin D3 levels were found to be normal but there was no improvement in teeth. He was reviewed again after a gap of two years and there was definitive change on positive side in dental enamel defect in upper incisors and canines.

Figure 1. CECT Scan abdomen coronal section showing small bowel on left side(horizontal blue arrow) and large bowel on right side (vertical blue arrow) on right side



Figure 2. CECT Scan abdomen transverse section showing small bowel on left side (horizontal blue arrow) and large bowel on right side (vertical blue arrow) on right side



DISCUSSION

Celiac Disease is an autoimmune disorder where consuming gluten (found in wheat, barley, and rye) triggers an immune response that damages the small intestine's villi. The symptoms include chronic abdominal pain, diarrhea, fatigue, weight loss, growth failure in children, and skin rashes. The diagnosis involves blood tests for antibodies (serum IgATTG antibody test) and an endoscopic biopsy to confirm damage to the small intestine. Intestinal Malrotation is a birth defect where the intestines don't rotate and fix into their proper anatomical positions during fetal development which can lead to volvulus, causing a life-threatening obstruction and requiring urgent surgical correction. The reported prevalence is 1 in 500 live births; however, some studies report a prevalence as high as 1% of the population [5,6]. Traditionally it is thought as a disease of the pediatric population presenting as bilious emesis during the first year of life but can present with different symptoms even in adulthood. Presentation in older populations tends to be atypical, chronic abdominal pain, nausea and diarrhea being the most commonly reported symptoms [7]. The diagnosis includes imaging tests, such as computed tomography (CT) scans, are used to identify abnormalities in intestinal position and fixation. The relationship between celiac Disease and

intestinal malrotation can be challenging as both conditions can present with vague gastrointestinal symptoms. There are case reports of celiac disease with underlying intestinal malrotation [8]. If a patient with suspected celiac disease develops symptoms that don't improve with a gluten-free diet, or if there are signs of intestinal obstruction, imaging should be done to rule out malrotation. The treatment for malrotation was described by William Ladd in 1938. The Ladd's procedure includes detorsion of the volvulus in case is present, lysis of the Ladd's bands from the cecum to the right abdominal wall, widening of the small bowel mesentery to prevent volvulus, appendectomy and placing the small bowel to the right and colon to the left leaving the bowel on non-rotation [9]. Multiple retrospective studies have showed the safety of laparoscopic Ladd's procedure. This technique has improved pain, earlier full enteral feeds, shorter hospital stay and decreased number of postoperative small bowel obstructions compared to the traditional open approach [10]. The main pitfall of the laparoscopic approach is the increased number of volvulus post procedure which has been explained by the lack of adhesions formed during a minimal invasive approach. One should also have in mind that the reported conversion rate is 25%. In our case, clinching point was severe pain abdomen even after strict gluten restriction and CT scan abdomen revealed all small intestine on left side and large

intestine on right side which is reverse of what is normally seen in intestinal malrotation.

CONCLUSION

Malrotation can be seen more commonly than expected. Even though it has traditionally been thought as a disease of the infants, it can present later in life and tends to have non-specific gastrointestinal symptoms. Clinicians need a high index of suspicion when assessing patients with these symptoms which poses more challenge in background of celiac disease.

Conflict Of Interest

No conflict of interest and prior permission from patient and relatives was taken before publishing the case report.

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