

# CAS CLINIQUE / CASE REPORT

## CELIAC DISEASE AFTER INTUSSUSCEPTION RECURRENCE

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**ABSTRACT** • Intussusception (IS) is one of the most common causes of abdominal emergency in infancy.

It is rarely seen above the age of 7 years. The association between celiac disease (CD) and intussusception in the pediatric group is limited to case reports. Celiac disease should be considered in the differential diagnosis in the presence of recurrent ileal intussusception regardless of the age group.

We report the case of an 8-year-old male with recurrent IS ultimately diagnosed with celiac disease.

Keywords : intussusception; celiac disease; pediatric

### INTRODUCTION

Intussusception (IS) is one of the most common causes of abdominal emergencies in infancy. It can be idiopathic as in most of the cases in the pediatric age or it may be the result of an intra- or extraluminal intestinal lesion, malignancy, or a lead point as in older children and adults [1].

Celiac disease can present with typical or atypical gastrointestinal (GI) symptoms, non-GI manifestations, or no symptoms at all [2].

Here, we report the case of a male child with recurrent IS ultimately diagnosed with celiac disease.

### CASE HISTORY

This is the case of an eight-year-old boy who presented to our emergency room (ER) for persistent and worsening vomiting. The symptoms started two days prior to presentation when he had recurrent vomiting, eventually becoming bilious. Patient presented to the ER one day prior to presentation and was given intravenous hydration and discharged home.

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**RÉSUMÉ** • L'invagination intestinale aiguë (II) est l'une des causes les plus fréquentes d'urgence abdominale chez l'enfant.

Elle est rare chez les enfants de plus de 7 ans. L'association entre maladie cœliaque (MC) et l'invagination chez les enfants est limitée à de rares cas rapportés. La maladie cœliaque devrait être envisagée dans le diagnostic différentiel en présence d'une invagination iléale récurrente quel que soit le groupe d'âge.

Nous rapportons le cas d'un garçon de 8 ans présentant une invagination intestinale aiguë récurrente débouchant sur le diagnostic d'une maladie cœliaque.

Mots-clés : invagination intestinale aiguë; maladie cœliaque; pédiatrie

He was complaining of intermittent diffuse abdominal pain associated with obstipation and hypoactive bowel sounds on auscultation but with a soft abdomen on palpation. A KUB (kidneys, ureters, bladder Xrays) was done showing no air fluid levels. An ultrasound was done that showed an ileo-ileal intussusception (Figure 1) that required surgical reduction after failure of observation and hydrostatic enema reduction.

During surgery, a total of eight intussusceptions in the ileum were identified and reduced with no lead point recognized.



**Figure 1** Ultrasound showing a transverse section of a target lesion representing intussusception

On his second day post surgery, the patient vomited again, so an abdominal computerized tomography (CT) scan was done showing jejunal wall thickening. Hence, a gastroscopy plus biopsy were subsequently done showing partial villous atrophy with increase intraepithelial lymphocytes (50%) in favor of celiac disease. Genetic testing (HLA haplotype) was ordered and came out to be HLA-DQ8/DQ2 trans carrier phenotype. The association between pathology and genetic testing made the diagnosis of celiac disease highly probable despite a negative serology (EMA & TTG).

To note that four years ago, he had a surgery for an ileo-ileal intussusception episode. During the interim, he presented several times to the emergency room for recurrent and severe vomiting that required intravenous hydration. A small bowel series was done at that time and was found to be normal.

Although our patient was short for his age, he was not considered to have failure to thrive as he was growing appropriately on the respective growth curve.

After initiating a gluten free diet, our patient did not experience any other similar episode over the past twelve months.

#### DISCUSSION

Intussusception, which is the invagination of one part of bowel into another, is caused by the irritation of the lumen of bowel wall altering the normal peristalsis [1].

It is the second most common cause of gastrointestinal obstruction in young children, and the most common cause of small bowel obstruction in children ages 3 months to 5 years with an approximate incidence of 22 to 56 cases per 100,000 per year [3]. But it is only seen in 1 to 5 percent of cases in adults with obstruction [1].

The triad that is present in 50% of the cases characterizes it: abdominal pain, palpable mass and current jelly stools [4]. The diagnosis of obstruction is done by plain abdominal film and an ultrasound or a CT scan if needed confirms intussusception.

The treatment of ileo-ileal ones is either by observation and conservative management, reduction by enema, or by operative techniques [5].

In contrast to the adult population where a lead point such as Meckel's diverticulum or lymphoma may be present, no cause can be identified in 90% to 95% of pediatric cases [1]. Its incidence of recurrence after reduction in children varies from 2% to 20% [6] generating the need to seek for an underlying disease that is initiating the intussusception in such cases [7].

This is present in 25% of overall cases. Several un-

derlying etiologies were noted, one of which is celiac disease that would induce the invagination because of the dysmotility, excessive secretions, or bowel wall thickness [8].

Celiac disease, also known as the gluten-sensitive enteropathy, is a genetically autoimmune related chronic disorder that manifests after the ingestion of gluten proteins found in wheat, rye, barley and other grains triggering mucosal inflammation and villous atrophy [9]. It occurs in 1% of the general population. The symptoms vary according to age group [10]. It is detected by an increase level of IgA antibodies to tissue transglutaminase and endomysium (with 100% accuracy) [10] and diagnosed by a biopsy taken from the small intestine that reveals villous atrophy [11].

Celiac disease is described as being a cause of IS in the older population which is a rare condition in that age group. The association between CD and IS in the pediatric group is limited to case reports only compared to adults [3]. Altaf *et al.* reported in their article two cases of infants who were diagnosed with celiac disease after developing recurrent intussusceptions and mentioned two possible theories regarding the mechanism of intussusception in CD: inflammation causing unbalanced peristaltic waves or hypotonic intestinal loops disturbing the normal peristaltism [7].

Intussusception is rarely seen in the older age group as mentioned (only 5% of all intussusception cases are seen in adults [1]). It is mostly idiopathic between 6 months and 2 years [5]. Our patient's age made this case peculiar: he had his episodes at an age exceeding the age interval mentioned above which warranted the need to look for an underlying disease.

Despite the atypical presentation of CD in our patient, the absence of failure to thrive, diarrhea, bloating, or any specific features of the disease, a high index of suspicion led us to the diagnosis.

#### CONCLUSION

It is important to consider celiac disease in the differential diagnosis of recurrent ileal intussusception regardless of the age group.

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