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Case Report (Pages: 6041-6046)

A 13.5- Year Old Boy with Abdominal Pain and Weight Loss and Chronic Intussusception

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Abstract

We present a 13.5 years old boy with chronic intussusception that was misdiagnosed as Crohn disease and received treatment for the wrong diagnosis. Because his condition didn't improve, more work-up were performed and the radiologic work-ups revealed the diagnosis of a missed intussusception with fistula.

We suggest that prolonged intussusception should be considered in the list of differential diagnosis for the refractory abdominal pain and distention, bloody diarrhea, and radiologist consultancy should be requested, as Crohn disease, infectious gastroenteritis, abdominal malignancies and prolonged intussusception should be carefully investigated and considered in such situations.

***Key words*:** Crohn disease, Intussusception, Burkitt lymphoma.

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1- INTRODUCTION

Intussusception is an important disease with considerable mortality during childhood and it may become chronic and misdiagnosed. The common clinical presentation with some prevalent diseases such as gastroenteritis is a challenging issue for the pediatricians. Crohn disease with the clinical presentations such as abdominal pain, bloody diarrhea could also be misdiagnosed with gastroenteritis and intussusception. The detailed medical history accompanied by clinical and para-clinical evidences should be used to make the correct diagnosis. One of the important tools in the correct diagnosis is the precise interpretation of radiologic imaging.

2- CASE REPORT

A 13.5- year- old boy was referred to the emergency clinic of our hospital (Children's Medical Center, Tehran-Iran) with, abdominal pain and weight loss (15 kg in the last 10 months). He didn't have diarrhea, but had occasional episodes of vomiting after eating food. The pain was located at the periumbilical region, radiated to the flanks, and aggravated by eating. On physical exam, he was pale, cachectic, weak and ill appeared. The exam of heart and respiratory system was normal, no lymphadenopathy was detected, and abdomen was soft, without distension, organomegaly or tenderness. His weight was 20 kg (below the 3rd percentile). The vital signs were stable and axillary temperature was 36.6 centigrade degree.

The illness had been started from 11 months ago with abdominal pain followed by bloody diarrhea a few minutes after the belly pain. The blood appeared as spots mixed with the bulk of stool. He was treated for infectious diarrhea without any progress. Because of refractory diarrhea, abdominal pain, weight loss and abdominal mass (abscess and granuloma) in the right upper quadrant on the imaging studies (**Figure.1**), later was diagnosed with Crohn disease and treatment started with Mesalamine, Azathioprine and Prednisolone. The mass was considered to be the complication Crohn disease and it was considered that removing the mass may cause more complications such as fistula formation. His condition got partially better for about 2 months, but gradually he started having abdominal pain, bloating and distension that started about a few minutes after eating food. Infliximab was added to the list of the medications, with the hope of controlling the disease but no improvement was seen. The tests are as follows:

White blood cells=19,550/microliter, red blood cells=3,540,000/microliter, hemoglobin=7 gram/deciliter, MCV=68.2 femtoliters, MCH=19.8 picograms, MCHC=29gram/deciliter, Platelet=727,000/microliter, neutrophil=84.5%, lymphocyte=9.1%, Monocyte=6.3%, Basophil=0.1%.

Aspartate aminotransferase=9 unit/liter, Alanine aminotransferase=7 unit/liter, Alkaline phosphatase=336 unit/liter, lipase=11 unite/liter (13-60), Amylase=40 unit/liter (up to 60), gamma-glutamyl transferase (GGT) =34 IU/liter (10-71). C-reactive protein=120 milligram/liter (up to 6). P-ANCA, C-ANCA and ASCA were negative. Stool examination reported as: >+3 for occult blood, many white blood cells with pus cells and many red blood cells.

PT, PTT and INR, Blood urea nitrogen, creatinine, sodium, potassium, albumin, uric acid, and lactate dehydrogenase were also normal. An abdominal ultrasonography was performed and revealed; a hypoechoic mass with lobulated border, measured86*48*47 millimeter, located in the right side of the midline, extended to right upper quadrant and below the liver, and color Doppler ultrasonography showed an hypoechoic, highly vascularized mass, suggesting

inflammatory mesenteric mass, without any collection inside. It was considered to be related to the complications of Crohn's disease which previously diagnosed. A computerized tomography had been performed in the another medical center (**Figure.1**) which showed a lobulated soft tissue mass in transverse and descending colon with central lumen in favor of intussusception, however it was not diagnosed previously. The contrast media at the distal part of the obstruction due to the old fistula is seen. The image had been interpreted mistakenly as granuloma caused by Crohn disease.

Two days after the admission in our hospital, an ultrasonography guided abscess drainage in the operating theatre planned and the radiologist consultant reported a missed invagination, surrounded by granulation tissue and the possibility of fistula formation and recommended the urgent need for rolling out the possibility of a malignant leading point.

Because of the mass, the surgeon consultants had planned to perform a laparotomy for removing the mass, but with the suggestion of a prolonged intussusception in ultrasonography, they first decided to post pone the surgery until completing a 14 days course of antibiotics but later a biopsy taken from the mass. After 14 days of treatment with antibiotics, a laparotomy operation was performed and ileocolic intussusception detected and a reduction operation tried and cecum and ascending colon was resected.

Hematology consultant suggested a bone marrow aspiration which reported normal. The pathology report of the mass was "highly suspicious to leukemia or lymphoma" which later reported to be Burkitt lymphoma with the following flow cytometry: CD3=74.49% (52-78), CD-16-56 10.50% (normal range: 3-17%), CD-19=7.59% (normal range: 8-24%), CD4=32.19% (normal range: 25-48%), CD8=40.88% (normal range: 9-35%).

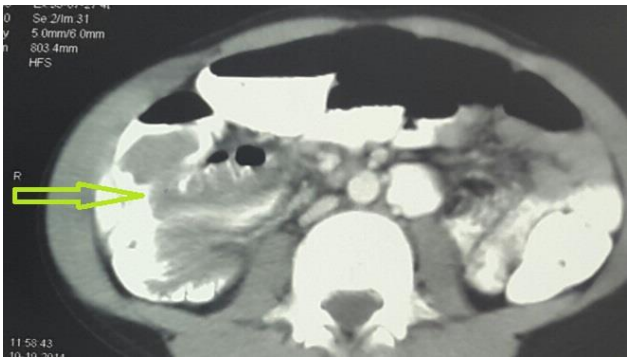

Fig.1: The CT scan shows; lobulated soft tissue mass (the arrow) in the transverse and descending colon (intussusceptum) in favor of intussusception.

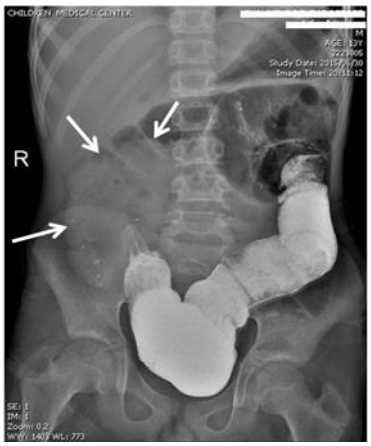

Fig.2: Gastrointestinal follow through shows a soft tissue mass (the three white arrows) in the right side of the abdomen in favor of intussusception.

3- DISCUSSION

Intussusception was described in 1674 for the first time and in 1876, Hirschprung introduced the method of "hydrostatic reduction" for its treatment (1). Intussusception is one of the common causes of bowel obstruction in children with noticeable mortality if not promptly treated. It is defined as the invagination of a proximal segment into a distal segment of the bowel (2). This disease is the most prevalent cause of bowel obstruction between 5 month and 3 year of age; 60% of these children are below 1 year old. Males are afflicted 3 times more than females. Without treatment, Ileocolic intussusception may cause intestinal infarction, perforation, peritonitis and death. The etiology of about 90% of the intussusception cases is idiopathic.

The disease prevalence has its peaks in fall and winter. Previous or concurrent respiratory infection with adenovirus has correlation with the incidence of intussusception. Swollen Peyer patches in the terminal ileum caused by gastrointestinal infection or receiving new food proteins, and lymphoid nodular hyperplasia are among of the risk factors for the intussusception. Recognizable lead points for the intussusception, like a Meckel diverticulum, neurofibromas, intestinal polyp, leiomyomas, anastomotic suture line, hemangioma, duplication cysts, enterostomy tube, lymphoma, are found in 2-8 % of the patients. Lead points are more common in older children. The most prevalent type of intussusception is ileocolic (3). Intestinal tumors account for less than 5% of the cases (4). Lymphoid hyperplasia caused by Yersiniaenterocolitica has been reported as a cause of intussusception in a patient with sickle cell anemia who was in an iron overload state because of several transfusions. Yersinia infection is more prevalent in iron overload condition, as it uses iron as a growth factor (5).

Juvenile polyps that happen usually in the 1st decade of life, can also act a recognizable lead point for colocolic intussusception (6). The clinical presentation of the intussusception can be similar to gastroenteritis which may occur previous to the disease (7), and include; sudden onset of severe paroxysmal colicky pain episodes with asymptomatic intervals between paroxysms of pain (8). The duration of pain is about 4-5 minutes and intervals of about 20 minutes (8). With the progression of the disease the child become weaker and lethargic, and shock-like state, fever and peritonitis may appear.

Vomiting, bloody stool with mucus (in 60% of the infants) and a palpable abdominal mass are the clinical manifestations. Less than 30% of the patients are presented with the classic triad of bloody or currant jelly stool, pain and a palpable sausage-shaped abdominal mass (3). Chronic intussusception manifests with vomiting, abdominal pain, and diarrhea, blood in rectum and weight loss (1). Ultrasonography with a sensitivity of 98-100% and a specificity of 98% can diagnose the intussusception. In plain abdominal radiography a dense region may appear in the area of intussusception (3).

Treatment of the intussusception is an emergency. In case of prolonged intussusception, shock, and peritoneal manifestation, hydrostatic reduction should not be tried and the patient should be prepared for surgery. If there are multiple recurrences and a suspected lead point is considered, surgical treatment should be performed (3). The success rate of reduction of intussusception with air or barium saline enema and hydrostatic saline reduction under ultrasound, are equal (9). When reduction with a contrast enema is successful or when there is concern about damage to the intestines, surgery should be performed (10).

Our patient was firstly diagnosed with infectious diarrheal disease and as the signs and symptoms were refractory to treatments, Crohn disease was considered later. Children with Crohn disease may have cramping, abdominal pain, and intermittent abdominal distension caused by partial small bowel obstruction. Intraabdominal abscess with pain and fever may also occur in Crohn disease. Small bowel lymphoma is less common in children than Crohn disease. In Crohn disease, the medical history may reveal abdominal pain, especially in the right lower quadrant, diarrhea, and vomiting, anorexia, and weight loss (11). In our hospital, radiologist consultant was requested to perform and abscess drainage under ultrasonography guide, while a missed intussusception was detected and later work-up revealed Burkitt lymphoma as the main cause of all problems.

4- CONCLUSION

Performing abdominal ultrasonography for detecting intussusception has been previously suggested in children with bloody diarrhea, colicky abdominal pain and distension who do not respond to antibiotics (12), and we also suggest that prolonged intussusception should be considered in the list of differential diagnosis for the refractory abdominal pain and distention, bloody diarrhea, and radiologist consultancy should be requested as Crohn disease, infectious gastroenteritis, abdominal malignancies and prolonged intussusception should be carefully investigated and considered in such situations.

5- ABBREVIATIONS

MCV: Mean Corpuscular Volume,

MCH: Mean Cell Hemoglobin,

MCHC: Mean Cell Hemoglobin Concentration,

P-ANCA: Perinuclear Anti-Neutrophil Cytoplasmic Antibodies,

C-ANCA: Cytoplasmic antineutrophil cytoplasmic antibodies,

ASCA: Anti-Saccharomyces cerevisiae antibodies,

PT: Prothrombin time,

PTT: Partial thromboplastin time,

INR: International normalized ratio,

CD3: cluster of differentiation 3,

CD4: cluster of differentiation 4,

CD8: cluster of differentiation 8,

CD-19: cluster of differentiation 19,

CD-16: cluster of differentiation 16.

6- CONFLICT OF INTEREST: None.

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