

CASE REPORT

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Intussusception Caused by *Yersinia enterocolitica* Enterocolitis in a Patient with Sickle Cell Anemia

Geetanjali Gupta¹, Shailesh Kumar², Reecha Singh¹ and Kathirvelu Shanmugasamy¹

¹Department of Pathology, Mahatma Gandhi Medical College and Research Institute, Puducherry, India 607402.

²Department of Microbiology, Mahatma Gandhi Medical College and Research Institute, Puducherry, India 607402.

Corresponding author email: gng_82@yahoo.co.in

Abstract: *Yersinia enterocolitica* intussusception is rarely encountered in patients without an underlying susceptibility and is most frequently reported in iron-overloaded patients. This is thought to be related to the unusual use of iron by this microorganism. We present a case of a 5-year old child with intussusception of the terminal ileum caused by *Y. enterocolitica* whose past medical history was significant for sickle cell disease. This type of presentation is extremely rare. His monthly blood transfusions may have put him at risk for *Y. enterocolitica* enterocolitis. The pathogenesis of this disease relates to the role of iron as an essential growth factor for *Y. enterocolitica*, and this patient's transfusions left him in an iron overloaded state despite treatment with Deferoxamine. Our patient's unusual presentation of intussusception was secondary to the mass effect caused by lymphoid hyperplasia, specifically hypertrophied Peyer's patches in the ileum caused by the *Y. enterocolitica* infection. We believe that our case demonstrates that *Y. enterocolitica* should be considered a possible pathogen in patients with sickle cell disease, especially if symptoms occur shortly after blood transfusion.

Keywords: *Yersinia enterocolitica*, intussusception, sickle cell anemia

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Introduction

Patients who are homozygous for sickle cell anemia are at risk for *Yersinia enterocolitica*. We present a case of a 5-year-old with intussusception of the terminal ileum whose past medical history was significant for sickle cell anemia. His monthly blood transfusions may have put him at risk for *Y. enterocolitica* enterocolitis. The pathogenesis of this disease relates to the role of iron as an essential growth factor for *Y. enterocolitica*, and this patient's transfusions left him in an iron overloaded state, despite treatment with Deferoxamine. Our patient's unusual presentation of intussusception was secondary to the mass effect caused by lymphoid hyperplasia, specifically hypertrophied Peyer's patches in the ileum caused by *Y. enterocolitica* infection. To our knowledge, this is the first case of intussusception as a complication of *Y. enterocolitica* enterocolitis in a patient with sickle cell anemia.

Case Presentation

A 5-year-old boy was hospitalized with a 3-day history of abdominal cramps, diarrhoea, bloody stools with mucus, intermittent emesis and fevers to 40 °C. His past medical history was significant for sickle cell disease, and he had received blood transfusions once every 3–4 weeks for the 3 years previous. His last transfusion (225 ml of packed cells) had occurred 5 days prior to admission. He also received weekly Deferoxamine treatment over the previous 1.5 years.

On admission, the child appeared ill and was dehydrated and tachypneic. His abdomen was distended, with diffuse tenderness, guarding and peritoneal signs in the right lower quadrant but with positive bowel sounds. A computed tomography scan of the abdomen revealed an inflammatory process in the right lower quadrant, with a small amount of free air and fluid collection that was consistent with an abscess. The patient was taken for exploratory laparotomy, ileocecectomy and ileostomy, during which time a 3 cm ileocolic intussusception was manually reduced. Mesenteric adenitis was noted. The appendix was erythematous and bulging. A preoperative stool culture in Cefsoludin-Irgasan-Novobiocin (CIN) agar grew *Y. enterocolitica* (serotype 0:3).

The patient was treated with antimicrobials and had an uneventful postoperative course.

Materials and Methods

The surgical specimens were fixed in formalin and embedded in paraffin. The sections were stained with routine Hematoxylin and Eosin stains. Special stains were performed including Brown and Hopps stain for *Yersinia*. A culture was grown from samples taken from the mesenteric lymph node.

Results

Surgical specimens included a section of terminal ileum measuring 18 × 4 × 4 cm that telescoped into the ileocecal region (Fig. 1). Other specimens included a section of the cecum and ascending colon measuring 9 × 5 × 4 cm, and another section of the cecum attached to the vermiform appendix that measured 5 × 1 × 1 cm. All serosal surfaces were erythematous. Multiple discrete lymph nodes were seen in the mesentery. Microscopically extensive, severe, transmural, acute and chronic inflammation was present in the terminal ileum, cecum, right colon and appendix, as were ulceration, abscess formation, lymphoid hypertrophy, focal necrosis and focal peritonitis (Figs. 2, 3, 4 and 5). Lymph nodes showed necrotizing lymphadenitis and extensive infarction. The Brown and Hopps stain revealed gram negative bacilli. The culture of the mesenteric



Figure 1. Photograph of the terminal ileum telescoped into the ileocecal region.

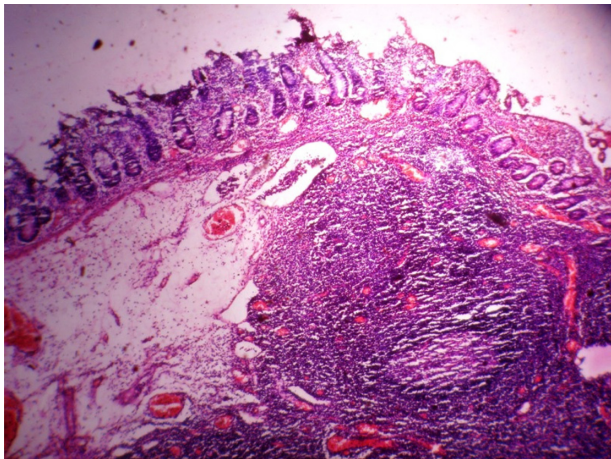


Figure 2. Microscopic view (Hematoxylin and Eosin stain; 200X magnification) showing severe transmural acute and chronic inflammation, ulceration, abscess formation and lymphoid hypertrophy.

lymph node (Fig. 6) yielded *Y. enterocolitica* of the same serotype, biotype and isotype as the organism isolated from the stool sample.

Discussion

Y. enterocolitica is a gram-negative coccobacillus with worldwide distribution that commonly causes gastroenteritis and mesenteric adenitis in children and adolescents. Reported complications have included severe enterocolitis involving the ileocecal region, appendicitis, small bowel gangrene, intestinal perforation, peritonitis, hepatic and splenic abscesses, chronic abscess

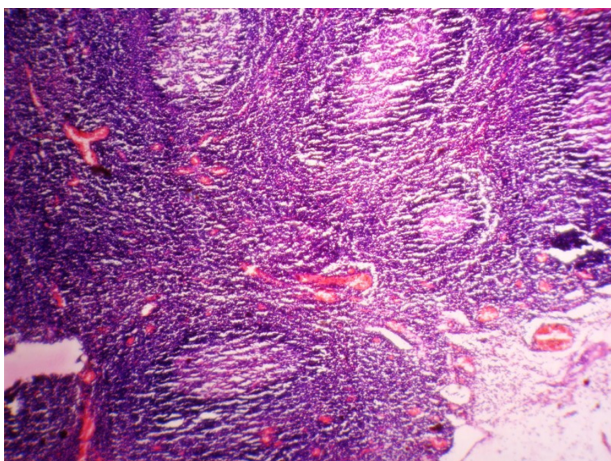


Figure 3. Microscopic view (Hematoxylin and Eosin stain; 200X magnification) showing lymphoid hypertrophy, and lymphoid hyperplasia with prominent germinal centers in the lamina propria.

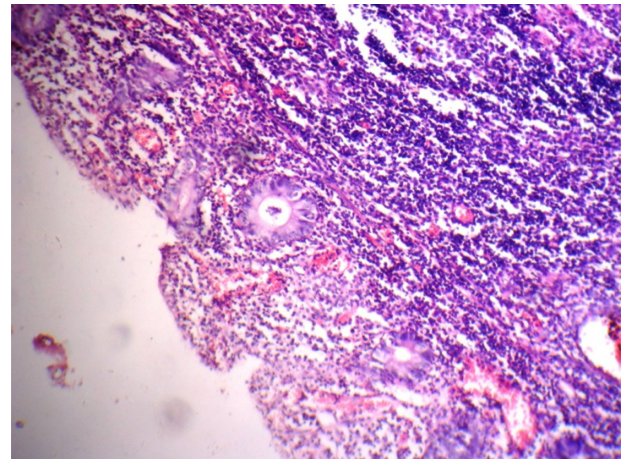


Figure 4. Microscopic view (Hematoxylin and Eosin stain; 200X magnification) showing mucosal ulceration, cryptitis and occasional cryptic abscess.

formation in the inguinal region and fatal septicaemia.¹ Bacterial infection in an intussusception has been described only sporadically. *Y. enterocolitica* is the only bacterial agent responsible for infected intussusceptions in children. *Y. enterocolitica* has been reported in three previous cases in North American literature and seven previous cases worldwide.²⁻⁶

Y. enterocolitica is spread primarily via the fecal—oral route, and infection most often results from human consumption of contaminated meat products (especially pork), milk products, water and vegetables. Less commonly, exposure to human and zoonotic fecal carriers such as pigs, dogs, cats

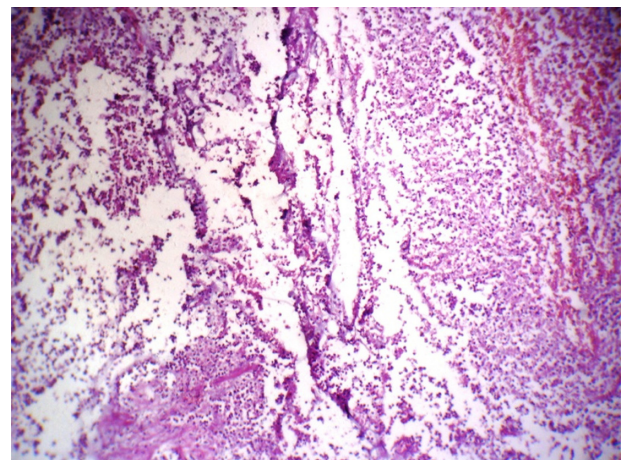


Figure 5. Microscopic view (Hematoxylin and Eosin stain; 200X magnification) showing areas of necrosis and congestion.

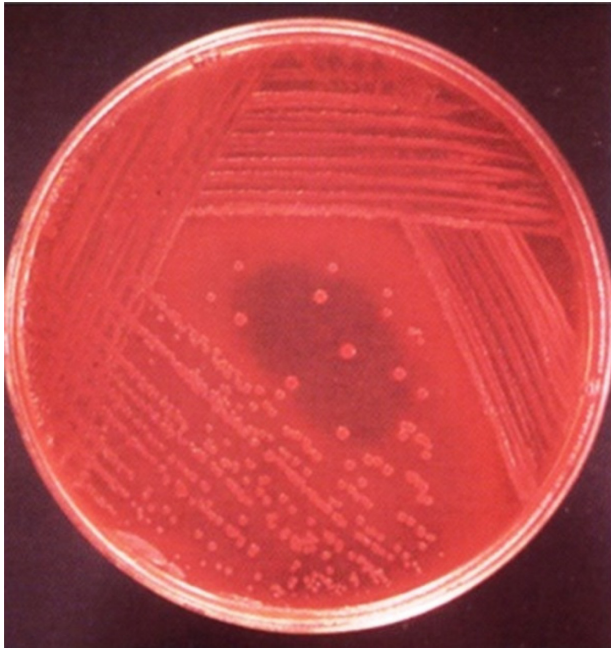


Figure 6. “Bull’s-eye view” of a colony of *Yersinia enterocolitica* on Cefsulodin-Irgasan-Novobiocin (CIN) agar.

and rats results in this disease.⁷ Our patient acquired the infection from contaminated water.

Intussusception is more common in infants aged 4 to 10 months, with a peak at 7 months. It is proposed that hypertrophied Peyer’s patches, possibly resulting from viral infection, act as a mechanical lead point for intussusceptions. The intussusception in our case was in the terminal ileum, secondary to mass effect caused by lymphoid hyperplasia. In one prospective study of 261 patients (228 children and 33 adults) with intussusceptions, 88% of the cases were idiopathic, without any definitive lead point. In these cases, the ileocecal area was the site most commonly involved (82%), hypertrophic Peyer’s patches of the terminal ileum being responsible for 39% of the idiopathic intussusceptions of the ileocecal area. Other studies had similar findings.^{8–11}

Patients who are homozygous for sickle cell anemia are at risk for serious *Y. enterocolitica* infections.¹² The cause of this increased susceptibility is related to systemic iron overload and the use of iron chelators. Pathogenesis of *Y. enterocolitica* infection relates to the role of iron as an essential growth factor. Bacteria produce iron by producing and releasing high-affinity chelators called siderophores. These bind and

solubilize ferric iron by competing with transferrin. The iron-siderophore complexes enter the bacterial cells following binding to specific receptors. Deferoxamine is a bacterial siderophore obtained from *Streptomyces pilose* that is used as a chelating agent in the treatment of acute iron toxicity and in chronic iron overload. *Y. enterocolitica* is unusual among bacteria in that it requires iron for growth and has receptors for siderophores but cannot produce siderophores endogenously.¹

Conclusions

This is the first case, to our knowledge, of intussusception as a complication of *Y. enterocolitica* enterocolitis in a patient with sickle cell disease. Recommendations from this and previous articles indicate that in a patient with iron overload, the clinician should be alerted to the possibility of *Y. enterocolitica* infection when abdominal symptoms and fever occur. The index of suspicion should be raised for patients with sickle cell disease especially if the symptoms occur shortly after a blood transfusion.

Disclosures

This manuscript has been read and approved by all authors. This paper is unique and is not under consideration by any other publication and has not been published elsewhere. The authors and peer reviewers of this paper report no conflicts of interest. The authors confirm that they have permission to reproduce any copyrighted material.

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