WebLog Journal of Pediatrics

Case Report
Published: 21 Nov, 2025

Hemolytic Uremic Syndrome Complicated with Colonic and Jejunal Strictures in a Toddler Girl with Hypoganglionosis

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WebLog Open Access Publications
Article ID : wjp.2025.k2102
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Abstract

An unusual rare case of hemolytic uremic syndrome in a toddler girl who was a known case of hypoganglionosis initially mimicked appendicitis and intussusception with pulmonary infection but finally leading to very rare complications of sigmoid colonic and jejunal strictures which required operative intervention by laparotomy with resections of the strictures and end to end anastomosis has been reported with complete recovery.

Keywords: Appendicitis; Colon; Complications; *Escherichia Coli* Intussusception; Jejunum; Hemolytic Uremic Syndrome; Hypoganglionosis; Laparotomy; Resection; Sigmoid; Stricture

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Received Date: *13 Nov 2025*
Accepted Date: *20 Nov 2025*
Published Date: *21 Nov 2025*

Citation:
Patel R, Govani DR, Swamy KB, Midha PK, Govani ND, Panchasara NG, et al. Hemolytic Uremic Syndrome Complicated with Colonic and Jejunal Strictures in a Toddler Girl with Hypoganglionosis. WebLog J Pediatr. wjp.2025.k2102. https://doi.org/10.5281/zenodo.17687294

Introduction

Right sided pulmonary infection may mimic appendicitis [1]. Appendicitis is very common in a child presenting with triad of symptoms of abdominal pain, vomiting, fever and diarrhea associated with peritonism signs and can mimic intussusception when there is blood in the stool and a palpable abdominal mass [2-3]. We wish to report a toddler girl who was a known case of conservatively treated hypoganglionosis and initially mimic appendicitis, intussusception and pulmonary infection but finally diagnosed with hemolytic uremic syndrome (HUS) which got complicated by sigmoid colonic and jejunal strictures.

Case Report

A 3-year-old girl, a known case of hypoganglionosis treated conservatively, who had lower abdominal pain, vomiting, fever, rigors and diarrhea with signs of peritonism presented to a general practitioner. Acute appendicitis was suspected and referred to us.

On examination, patient looked ill and by this time patient had blood in stools and a palpable tender mass in left lower abdomen and fine crepitations and reduced air entry to right chest. An intussusception with chest infection was suspected.

Abdominal ultrasound showed normal appendix and right iliac fossa and a thickened sigmoid colon with 5 mm thickness of wall suggestive of acute sigmoid colitis but could not see any target or kidney sign in anteroposterior or transverse views to suggest intussusception (Figure 1 A & B). Color doppler studies showed acutely inflamed sigmoid colon with thick wall and increased vascularity (Figure 1C). Chest radiograph showed right middle lobe collapse consolidation with moderate right sided pleural infusion (Figure 1D).

Urine output was reduced and it was dark yellow or tea-colored. Urinalysis was normal and culture negative. Complete blood count showed severe anemia with hemoglobin of 71 g/L; platelet count of 35000/µL; creatinine of 154 mmol/L; and urea of 16.6 mmol/L with 20% hematocrit and leukocyte count 18760/µL. Peripheral blood smear study showed severe hemolytic anemia associated with fragmented red blood cells and thrombocytopenia. Biochemical tests showed acute renal injury. Stool culture grew Escherichia coli O157:H7 *E. Coli* and Shigella dysenteriae type 1 with

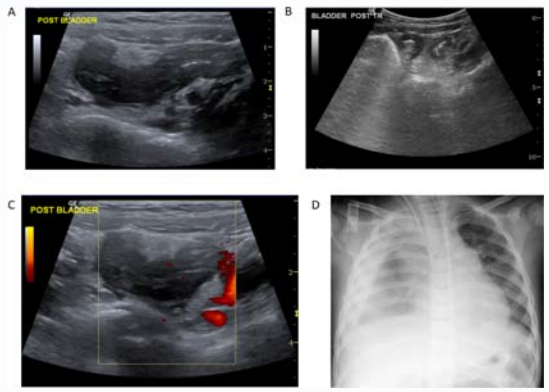

Figure 1: Ultrasound and chest radiograph A. posterior bladder view showing severe sigmoid colitis and enteritis bowel loops B. transverse view with no target sign C. color doppler showing severe enterocolitis D. Chest radiograph demonstrating right mid zone collapse consolidation and moderate pleural effusion.

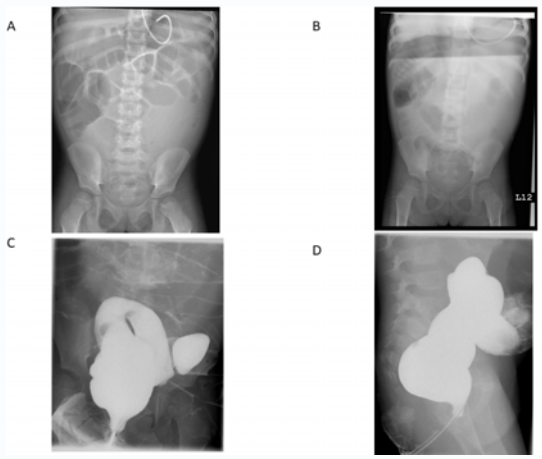

Figure 2: Plain and contrast radiographs A Supine AXR. Note dilated featureless proximal jejunum in upper central abdomen with dilated colon with haustra up to left colon. B Antero-posterior and lateral views of contrast enema showing complete stricture of proximal sigmoid colon with beaking effect.

Verotoxin/ Shiga toxin (StX) isolated associated with severe HUS enterocolitis, confirming the diagnosis of Shiga-like toxin producing E coli hemolytic-uremic syndrome (STEC-HUS or typical HUS).

Patient was resuscitated and supportive treatment for severe enterocolitis, pulmonary infection and HUS that required ventilation, intravenous fluids, broad spectrum antibiotics, parenteral nutrition, transfusion support and renal replacement therapy. Patient was discharged home after 35 days.

At 3 months after discharge, patient developed subacute intestinal obstruction and presented again and plain abdominal supine and erect films clearly showed dilated featureless proximal jejunal loops in upper central abdomen with gasless lower central abdomen and dilated large bowel loops up to left colon and gasless left paracolic area and pelvis (Figure 2 A and B). Water soluble contrast enema showed very tight colonic stricture at proximal sigmoid colon with beaking sign and no contrast could be passed beyond the stricture (Figure 2 A and B). Small intestinal stricture was suspected based on the plain radiographs.

Patient underwent exploratory laparotomy which showed proximal sigmoid and jejunal strictures which were resected and primary anastomosis carried out uneventfully. Histopathological examination confirmed the presence of sigmoid colonic and jejunal strictures with severe inflammation, chronic ischemia and necrosis. Long term follow-up at 10 years showed patient to be asymptomatic, thriving well, normal hematological and renal functions with normal abdominal and renal ultrasound scans.

Discussion

Most children with HUS and its complications recover without long term effects. HUS is most common in young children between 6 months and 4 years and a triad of microangiopathic hemolytic anemia, thrombocytopenia and an acute kidney injury [4]. Most often, infection with certain strains of *E. coli* bacteria is the cause. However, other infections, certain medications or comorbidities such as pregnancy, cancer or autoimmune diseases may have HUS.

During acute phase interpretation of laboratory results needs special skills to address the challenges and intensive care unit treatment is ideally suited [5]. Triad of macroangiopathic hemolytic anemia, thrombocytopenia and acute kidney injury together with isolation of bacteria and toxin confirms the diagnosis [6]. Abdominal symptoms are predominant in the prodromic phase, simulating appendicitis, intussusception, or acute inflammatory bowel disease [7].

Treatment of acute phase of HUS is mainly conservative and supportive as in our case. Although anti-inflammatory therapy with intravenous immunoglobulin and methylprednisolone pulses has been reported with good results [8].

Most survivors of HUS have no immediate catastrophic complications, about 3-5% are left with long-term sequelae to organs other than the kidneys, especially to the gut, pancreas or brain as extrarenal organs. Similar percentage may have severe kidney damage and, require dialysis and renal transplant later on.

With aggressive treatment, more than 90% of patients survive the acute phase of HUS, and only about 9% may develop ESRD. Roughly one-third of persons with HUS have abnormal kidney function many years later, and a few require long-term dialysis.

Conclusion

Combined colonic and jejunal strictures following severe enterocolitis is a very rare complication of HUS. The risk factors include female sex, younger age, high leukocyte count/ hematocrit and severe enterocolitis. The parents, carers and patients should be alerted about this complication and high index of suspicion will allow early detection. Cardinal symptoms of bowel obstruction, plain radiographs and contrast enema will allow accurate diagnosis. Surgical treatment includes laparoscopic or open resection of stricture/s and primary anastomosis with excellent results.

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