Case Report

Cytomegalovirus as a Cause of Colonic Stricture-Simulating Hirschsprung’s Disease

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INTRODUCTION

Colon is the least common site of congenital bowel stenosis/atroies and accounts for only 5%–15%.[1,2] The reported incidence is approximately 1 of 40,000 of live birth.[3] Colonic stenosis is rarer than atresia and mostly occurs in ascending and transverse colon.[4] Sigmoid colonic stenosis is extremely rare. The length of the stenotic segment may range from 3 to 15 cm.[5] Gastrointestinal sequelae of cytomegalovirus (CMV) are rare, usually associated with significant immunocompromise and frequently require surgical intervention for diagnosis and management.[6] We encountered an interesting case of colonic stricture due to localized CMV infection mimicking Hirschsprung’s disease. In babies <6 months of age, only one similar case has been reported in literature by Ekema et al.[7]

CASE REPORT

A 5-month-old female child, weighing 4.8 kg, presented with complaints of recurrent abdominal distension, vomiting, and constipation for 2 months. Her antenatal period was uneventful. TORCH test was not done in the antenatal period. She was full-term normal vaginal delivery and perinatal period was uneventful. Birth weight was 3 kg. The baby passed meconium within 24 h of birth. On examination, the child was anemic and had soft-distended abdomen with visible bowel peristalsis. Plain X-ray abdomen showed dilated bowel loops. Ultrasound abdomen was suggestive of grossly dilated gas-filled large bowel up to descending colon and collapsed sigmoid colon and rectum with thickened cecum and descending colon. Anorectal manometry showed the presence of rectoanal inhibitory reflex. A barium enema showed smooth passage of contrast in the rectum and sigmoid colon. There was a short segment of narrowing at the colosigmoid junction with proximal dilatation of the bowel [Figure 1]. In view of anemia (Hb – 8.4 g%) and hypoalbuminemia (albumin – 2.5), blood and albumin were transfused. Thyroid function tests were normal. The baby also had raised total leucocyte counts for which parenteral antibiotics were started. Blood culture showed no growth. In view of persistent abdominal distension and intermittent vomiting with visible peristalsis, baby was taken up for surgery. At surgery, frozen sections of the rectal biopsy showed absent ganglion cells. A diagnostic laparoscopy showed a dilated proximal colon up to the descending and collapsed distal colon [Figure 2]. There were some adhesions in this area. Multiple colonic and rectal biopsies were done. On frozen section, there were few ganglion cells in the colon at the level of peritoneal reflection and abundant ganglion cells in the sigmoid colon. On exploration, there was a stricture in the sigmoid colon. The biopsy showed localized CMV infection.

KEYWORDS: Colonic, cytomegalovirus, stricture

Colonic stenosis/atroies account for only 5%–15% of all atresias. Colonic stenosis is rarer than atresia and mostly occurs in ascending and transverse colon. Gastrointestinal sequelae of cytomegalovirus (CMV) are rare, frequently requires surgical intervention for diagnosis and management. We describe a 5-month-old female child with complaints of recurrent abdominal distension, vomiting, and constipation for 2 months simulating Hirschsprung’s disease. After barium enema, the baby was taken up for surgery. Intraoperatively, we found a colonic stricture in the descending colon. The biopsy showed localized CMV infection.

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descending colon which was resected and a colostomy was created [Figure 3]. From postoperative day 2, colostomy started functioning, and the baby was allowed oral feeds. At paraffin section, ganglion cells were present throughout the colon and rectum. Histopathological examination suggested acute colitis with marked fibrosis compatible with a stricture measuring 28 mm, which was CMV positive on staining [Figure 4a-c]. Since CMV infection has been reported with colonic strictures,[7] CMV DNA polymerase chain reaction was performed in the baby’s oral secretions and mother’s expressed breast milk, which were positive. However, the blood samples of both baby and mother were negative for CMV. Subsequently, the baby has had a colostomy closure and has recovered uneventfully and is now 7 months old and 7 kg.

**Discussion**

Colonic atresias/stenosis forms 5% to 15% of all gut atresias. Isolated colonic stenosis is even rarer than colonic atresias. The most common presentation of these infants is complete intestinal obstruction in cases of colonic atresia and partial intestinal obstruction in cases of colonic stenosis.[9] In case of colonic atresia, the mean age of presentation was 2.4 days (standard deviation ± 1.2 days), and in case of colonic stenosis, the age of presentation could vary from 3rd day of life to 8 months of age.[9]

Left-sided colonic stenosis is less common than right-sided colonic stenosis. It may be due to vascular insult in utero.[10] Depending on the severity of the vascular insult, stenosis or atresia may occur. The patient with colonic stenosis may present early or late in life depending on the severity of stenosis and other concomitant factors. In the literature, the usual lengths of reported stenosis in colon may vary from 3 to 15 cm.[10] The treatment option used in most of the cases of colonic atresia and stenosis is end-to-end colonic anastomosis with or without enterostomy.

CMV is usually transmitted by direct human-to-human contact through vertical or horizontal routes. An infected person can excrete CMV in urine, saliva, semen, cervical secretions, or breast milk. The virus establishes latent infection through blood products. Solid organs can also transmit CMV. In the surgical literature, intestinal CMV-infected cells in infants are prevalent in neonatal necrotizing enterocolitis. Very few cases of primary CMV infection of the gastrointestinal tract have been reported.
tract of surgical interest in immunocompetent neonates have been reported. Ekema et al.[7] described a neonate with congenital or perinatal CMV infection with gastrointestinal involvement who developed a colonic stricture and manifested a clinical picture similar to Hirschsprung’s disease. The intestinal lesion was a localized segmental CMV infection of the colon in which inflammation dominated the histopathologic finding.[7]

In our case, there was an unusual clinical presentation. Intraoperatively, we found a colonic stricture in the descending colon which on histopathology was classical of CMV. We are recommending that any baby with a colonic stricture presenting after the neonatal period must be tested for CMV. Since there was no CMV detected in the blood of either baby or mother, no antiviral therapy was given.

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Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Sax EJ. Pediatric case of the day. Congenital colonic stenosis. AJR Am J Roentgenol 1991;156:1315-7.
2. Powell RW, Raffensperger JG. Congenital colonic atresia. J Pediatr Surg 1982;17:166-70.
3. Franken EA. Gastrointestinal Imaging in Pediatrics. 2nd ed. New York, USA: Harper & Row; 1982. p. 286-7.
4. Ruggeri G, Libri M, Gargano T, Pavia S, Pasini L, Tani G, et al. Congenital colonic stenosis: A case of late-onset. Pediatr Med Chir 2009;31:130-3.
5. Galván-Montaño A, Suárez-Roa Mde L, Carmona-Moreno E. Congenital stenosis of the colon with foreign bodies. Case report. Cir Cir 2010;78:259-61.
6. Arnold M, Itzikowitz R, Young B, Machoki SM, Hsiao NY, Pillay K, et al. Surgical manifestations of gastrointestinal cytomegalovirus infection in children: Clinical audit and literature review. J Pediatr Surg 2015;50:1874-9.
7. Ekema G, Pedersini P, Milianti S, Ubertazzi M, Minoli D, Manciana A, et al. Colonic stricture mimicking Hirschsprung’s disease: A localized cytomegalovirus infection. J Pediatr Surg 2006;41:850-2.
8. Louw JH. Congenital intestinal atresia and stenosis in the newborn: Observation on its pathogenesis and treatment. Ann R Coll Surg Engl 1959;25:209-34.
9. Mirza B, Iqbal S, Ijaz L. Colonic atresia and stenosis: Our experience. J Neonatal Surg 2012;1:4.
10. Hall TR, Zaninovic A, Lewin D, Barrett C, Boechat MI. Neonatal intestinal ischemia with bowel perforation: An in utero complication of maternal cocaine abuse. AJR Am J Roentgenol 1992;158:1303-4.