Idiopathic Ileo-ileal Intussusception in Early Infancy

The classic 'idiopathic ileocolic intussusception' is a common entity. On the other hand, small bowel intussusceptions (SBI) are rare and constitute only 1-8.3% all intussusceptions(1,2). Amongst the SBI, ileo-ileal group constitute a bulk. The available literature categorizes SBI into three situations: SBI in children aged over 2 years, SBI due to pathological lead point, and SBI occurring in postoperative period(3). We believe that apart from the above three categories, there exists another definite entity 'idiopathic ileo-ileal intussusception', a term that has been coined the term by Sheung-Fat, *et al.*(4) recently. They described 8 cases of idiopathic ileo-ileal intussusception in their series of 19 cases of surgically proven small bowel intussusception(4). We present here our experience with four patients with 'idiopathic ileo-ileal intussusception' presenting in early infancy. This rarely described entity bears epidemiological semblance to 'idiopathic ileocolic intussusception', but has clinical presentation described previously for SBI.

During a four-month period (April to August 2004), four infants of 'idiopathic ileo-ileal intussusception' were managed. We may state here that only the symptomatic patients of ileo-ileal intussusception that were confirmed at surgery have been mentioned. The so-called short, transient, inconsequential small bowel invaginations that are picked up on high-resolution abdominal ultrasound and CECT are not included here. Three of them were males. All of them presented between 4 to 6 months. Three of them had presented in early summer. All four had been completely weaned off breast feeds by the age of 3-4 months. Three of these patients had history of diarrhea presenting just prior to the intussusception. The average duration of symptoms before presentation was 4½ days (range 1-7 days). All four presented with bilious vomiting and abdominal distention; two had colicky abdominal pain also. Only one of them had bleeding per rectum. Abdominal mass could not be palpated in any of them. Abdominal roentgenograms revealed multiple air-fluid levels suggestive of intestinal obstruction in all the cases. Abdominal ultrasound was done in three and was diagnostic of intussusception in only one patient. Two required intestinal resection and end-to-end anastomosis for ischemic bowel, while in two patients, reduction at surgery was possible. None of these patients had demonstrable lead point. Though all the patients survived, one of them developed wound dehiscence. This patient had iatrogenic bowel injury at the time of secondary abdominal closure that led to a third surgery.

Idiopathic ileo-ileal intussusception appears to have many epidemiological similarities with classical idiopathic ileocolic intussusception. The male preponderance described in patients with ileocolic variety was also noted in our series. Similarly, the increased incidence in early summer months is common to both 'idiopathic ileocolic intussusception' and 'idiopathic ileo-ileal intussusception'. Relationship to change in diet, loss of passively acquired maternal immunity (early weaning away from breast milk), acute enteritis with hyper peristalsis and mesenteric adenitis have been known to be present in patients with 'idiopathic ileocolic intussusception'(3); similar has been our experience with our four patients of 'idiopathic ileo-ileal intussusception'.

We suspect that these two entities may be representing the same pathology. Ileo-ileal invagination due to hyper-peristalsis could be the initial step in either state. In some cases, the intussuscipiens manages to advance into

a mobile cecum leading to ileocolic intussusception. On the other hand, in case of ileo-ileal intussusception, the neck of the intussusceptum is very tight and therefore obstruction to blood supply and gangrene occur early. This tightness prevents advance of intussuscipiens, so ileo-cecal and ileo-colic intussusception cannot occur. The presentation of ileo-ileal intussusception in early infancy (4-6 months) could be merely reflective of early weaning.

Y.K. Sarin,
N.D. Nagdeve,
Department of Pediatric Surgery,
M.A.M.College and Associated Hospitals,
New Delhi 110 002, India.
E-mail: yksaring@hotmail.com

REFERENCES

1. Kim YS, Rhu JH. Intussusception in infancy and childhood: analysis of 385 cases. Int Surg 1989: 74: 114-118.
2. AI-Salem AH, Habash 8M. Ileoileal intussusception: A report of four cases. Ann Saudi Med 2000; 20: 310-312.
2. Stringer MD, Pablot SM, Brereton RJ. Pediatric intussusception. Br J Surg 1992; 79: 867-876..
4. Sheung-Fat Ko, Tze-Yu Lee, Shu-Hangn Ng, Yung-Liang Wan, Min-Chi Chen, Mao-Meng Tiao, *et al*. Small bowel intussusception in symptomatic pediatric patients: Experience with 19 surgically proven cases. World J Surg 2002, 26: 438-444.

Hereditary Methemoglobinemia in an Infant

The rare causes of cyanosis in the absence of pulmonary or cardiac disease are methemoglobinemia, sulphemoglobinemia and carboxyhemoglobinemia(l).

Hereditary or congenital forms of methemoglobinemia are extremely rare(l) and may occur due to NADH-cytochrome b5 reductase deficiency or Hb M disorders.

In view of rare reporting of this condition(2-4), we narrate our experience with an infant with congenital methemoglobinemia.

A 10-month-old baby girl presented with history of fever, cough and respiratory distress for last five days. In the past five months she had similar attacks of respiratory distress twice, that was treated with oral medication. There was no history of any significant drug intake or exposure to well water. Birth, developmental and dietary history was non-contributory. There was history of consanguinity. Mother was married to her own maternal uncle. The baby had a bluish hue since birth, which used to increase during attack of respiratory distress. On examination she had raised temperature, mild respiratory distress and central cyanosis. She had rhonchi and crepts in both the lung fields but other systems were within normal limits. Her pulse-oximetry reading showed oxygen saturation was 85%, Arterial blood gas analysis showed PH 7.38, PaO_2 82, PCO_2 34, HCO_3-24, SaO_2 88%. Colour of blood was dark, which was noted during sampling.

She had hemoglobin of l3 gmidL, TLC l6000/cmm, DLC-NeutrophiI 30, Lymphocyte 70, ESR-8 mm, PCV 44, and total erythrocyte count $4.6 \times l0^{12}$cells/L. Chest *X*-ray and electrographic tracing and echocardiographic finding was normal. Hemoglobin