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Mini Review.

Association of midgut malrotation with intussusception

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Abstract

Background: The cause of intussusuception in most infants is unclear. Intestinal malrotation has been postulated as a possible cause in some infants. Waugh,s syndrome is the association of intestinal malrotation with intussusception.
Patients/Methods: Of 18 children with intussusception managed in the Paediatric Surgical Unit of our hospital over a 3-year period, eight had abnormality of intestinal rotation and fixation. Their case notes, operation notes and discharge summary sheets have been retrospectively reviewed.
Results: There were five boys and three girls. Their ages ranged from 13 days – 12 months (median 10 months). The main clinical features were bilious vomiting, blood stained diarrhoea and abdominal distension. In two infants, the intussusceptions had prolapsed through the anus at presentation. One neonate had ruptured omphalocoele minor containing a caeco-colic intussusception that had perforated at presentation. Two other infants had mid gut volvulus, one as a simultaneous finding with intussusception while the other one 72 hours after operative reduction of intussusception. All had laparotomy. In six infants, the intussusception was ileo-colic while in two it was caeco-colic. There was no lead point in any infant. Four infants had successful operative reduction while four had bowel resection with end-to-end anastomosis. All had Ladd's procedure. One child died of overwhelming sepsis following resection of gangrenous bowel.
Conclusion: Intestinal malrotation may be associated with idiopathic intussusception. It is important to look for this association when managing infants with intussusception.
Key words: Waugh's syndrome, intussusception, malrotation, midgut volvulus, Ladd's procedure.

Introduction

It is not clear whether intestinal malrotation predisposes to intussusception in infants. Some previous reports had suggested a strong association between these two conditions and referred to the association as Waugh's syndrome[1-3]. This is a report of eight infants with this association managed in our unit over a 3-year period

Patients and Methods

Eight infants with idiopathic intussusceptions in whom abnormalities of intestinal rotation and fixation were present were managed in the Paediatric Surgery Unit of the Jos University Teaching Hospital, Jos, from January 2001 to December 2003. During this period 18 children with intussusception were managed by the unit. The case records, operation notes and discharge summary sheets of these eight infants were reviewed and form the basis of this report.

Results

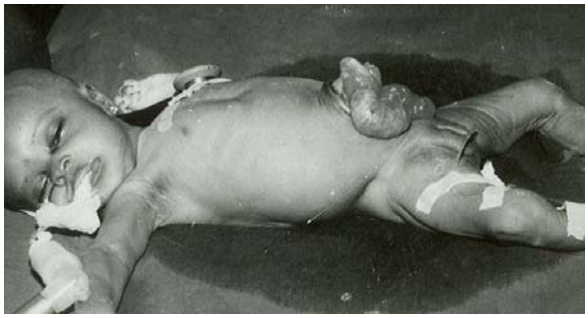
There were five boys and 3 girls. Their ages at presentation ranged from 13 days to 12 months (median 10 months). The duration of symptoms before presentation ranged from 2 – 14 days (median 7 days). The classical triad of bile stained vomiting, abdominal colic and blood stained loose stool occurred in six infants. In two infants, the intussusception had prolapsed through the anus at presentation. One neonate had ruptured omphalocoele minor at presentation with a caeco-colic intussusception that had perforated and was discharging faecal matter at the tip (fig 1). There was no vitello-intestinal tract or meckel's diverticulum seen in this neonate. One child had an operative reduction of the intussusception

without taking note of the associated malrotation. Seventy-two hours after operation, he started having bilious vomiting associated with abdominal distention. A re-laparotomy revealed a midgut volvulus with gangrenous bowel and the duodeno-jejunal junction to the right of the midline. The haemograms of the infants ranged from 6.0 – 10g/dl (median 8.0g/dl). There was neutrophilia with toxic granulation and a shift to the left in two infants. The remaining six infants had normal white cell counts and normal platelets on admission. The electrolytes values were normal in two infants but there was elevation of urea levels in the remaining six infants, probably due to dehydration.On admission to hospital all the children were resuscitated using paediatric saline to which 50% glucose was added to make 8.0% dextrose in saline. When the urine output was between 2-3 ml/kg/hr, laparotomy was done in all. None had an attempted air-enema reduction as facilities for this is not available at our hospital. Laparotomy was done through a right transverse supra-umbilical incision. The operative findings, procedures performed and outcome in the eight infants are summarized in Table I. In six infants, intussusception was ileo-colic while two had caeco-colic intussusception (fig1). An associated midgut volvulus was found at operation in one child. There was no identifiable pathology at the lead point or mesenteric lymph node enlargement in any of the infants. Apart from the child with ruptured ompahlocole, there were no other associated congenital abnormalities seen in any of the infants.

In four infants, operative reduction was effected and a Ladd's procedure was performed. The child with volvulus after initial operative reduction of his intussusception had resection of the gangrenous bowel with end-to-end bowel anastomosis. A Ladd's procedure was also performed. The child with a simultaneous volvulus had resection of the gangrenous bowel, end-to-end bowel anastomosis and Ladd's procedure. The child with associated ruptured omphalocoele and bowel perforation had bowel resection and end-to-end bowel anastomosis. A Ladd's procedure and fascial closure of the omphalocoele were also done.

Postoperatively one child had persistent vomiting for three days and one other child had that responded to treatment with broad spectrum antibiotics. The child that had bowel resection for gangrenous bowel died 48 hours after operation due to overwhelming sepsis. Five infants were lost to follow-up after discharge and are presumed well. The other two infants were followed up for 6 months and had remained well

fig 1: A newborn with ruptured omphalocoele containing a caecolic intussusception that had perforated and malrotation



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Discussion.

Intussusception is a common cause of intestinal obstruction in infants[4-6]. Most of these intussusceptions are considered to be idiopathic in origin as there is usually no identifiable pathology at the lead point. The association between intussusception and malrotation in infants has not been widely reported. Brereton et al[1] in a report of 18 infants with this association referred to it as Waugh's syndrome. Recent reports from South Africa[2] and Turkey[3] also used the same terminology for the association. It has been suggested that malrotation is associated with a mobile right colon which may be a pre-requisite for intussusception[2]. Eight (44%) of the 18 children treated for intussusception in this report have this association. This compares well with Brereton et al's incidence of 40% in a study of 49 children with intussusception. In a review of 64 children with intussusception in our hospital, 41% had no identifiable pathology at the lead point[5]. The authors however did not look for malrotation in their series. This association may be more common than reported in the literature.

Seven of the children in this report had unfixed caecum and mobile right colon with the duodenojejunal junction located to the right of the midline. There was no identifiable pathology at the lead point in any of them. This would strongly support the opinion of Breckon and Hadley that malrotation by its nature is associated with a mobile right colon which may predispose to intussusception. In two of the infants in this report, intussusception had prolapsed through the anus. In our environment, delayed presentation and an unfixed caecum with a mobile right colon may be responsible. Intussusception is rare during the neonatal period[7]. One of our patients was a neonate.In one of the patients, volvulus of the midgut occurred simultaneously with intussusception. This rare occurrence had been reported previous by others[2,8-10]. Air-enema reduction of intussuception is widely practiced in other centres. Air-enema reduction of intussusception may be complicated by midgut volvulus in children with Waugh's syndrome and should be investigated to rule this out after the procedure. In one of the patients in this report, volvulus of the midgut occurred after operative

reduction. Malrotation should be looked for and treated if present when reducing idiopathic intussusception to prevent midgut volvulus after operation

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Table I **Operative findings, procedures and outcome in 8 children with intussusception associated with malrotation**

Age	Sex	Operative findings	Procedure performed	outcome
13 days	f	Caeco-colic intussusception Perforated intestine Ruptured omphalocoele Malroation	Bowel resection + anastomosis, Ladd's procedure + fascial closure	Survived
6 months	m	Ileo-colic intussusception DJ on right of midline Caecum in RUQ	Operative reduction Ladd's procedure	Survived
8 months	m	Ileo-colic intussusception DJ on the right of midline Caecum in RUQ	Operative reduction Ladd's procedure	Survived
10 months	f	Ileo-colic intussusception DJ on the right of midline Caecum in mid-abdomen	Operative reduction add's procedure	Survived
10 months	m	Ileo-colic intussusception Volvulus, gangrenous bowel *DJ to right of midline Caecum *RUQ	Bowel resection + anastomosis Ladd's procedure	Died
11 months	f	Ileo-colic intussusception DJ to right of midline Caecum inRUQ	Bowel resection + anastomosis Ladd's procedure	Survived
12 months	m	Ileo-colic intussusception DJ in midline	Operative reduction Ladd's procedure	Survived
12 months	m	Caeco-colic intussusception Re-laporotomy showed Volvulus + malrotation DJ to right of midline Caecum mid-abdomen	Operative reduction bowel resection + anastomosis + Ladd's procedure	Survived

* DJ Duodenojejunal junction., *Right upper quadrant

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