Case Report

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# A rare complication of small bowel intussusception: Report of a case and review of literature

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## ABSTRACT

Volvulus and intussusception are rare conditions in children. We describe an unusual case of intussusception due to a solitary Peutz-Jeghers type hamartomatous polyp complicated by volvulus, which occurred in an 11-year-old girl. A laparotomy allows to successfully treat the pathology. The postoperative course was favourable. We discuss the clinical findings and the values of the preoperative instrumental diagnosis. The literature is reviewed. Identifying a midgut volvulus, as complication of a small bowel intussusception, during the diagnostic phase could help in choosing the most appropriate surgical approach.

**Key words:** Children, hamartomatous polyp, intussusception, volvulus

## INTRODUCTION

Intussusception is one of the most common causes of bowel obstruction in children, which needs a prompt diagnosis and an adequate management. If not recognized it may cause bowel obstruction and mesenteric compromise with intestinal necrosis. The majority of cases of intussusception during the childhood are ileocolic type, while small bowel intussusception (SBI) is uncommon and more frequent in children older than 2 years.[1-2] Usually, an SBI is associated with a lesion of the bowel acting as an anatomical lead point.

Midgut volvulus is characterised by an intestinal segment twisted on its mesentery.

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The association of SBI and midgut volvulus is a rare condition in children and has been reported few cases at the time. This condition can represent the complication of SBI and mean a diagnostic dilemma.

Our paper describes a case report of SBI due to solitary hamartomatous polyp (SHP) and midgut volvulus occurring in an 11-year-old girl. We discuss the unusual condition, clinical and radiological findings, and the value of a pre-operative detailed diagnosis. The literature is reviewed.

## CASE REPORT

An 11-year-old girl was admitted to our hospital. The clinical history revealed severe abdominal pain starting 2 days before. The pain was located to the mesogastric region and to the left side. The intestinal transit was blocked since 24 hours. No fever. Gastric vomiting was reported, no bilious vomiting was present. Physical examination revealed a deep tenderness at the palpation of the mesogastrium and the left side, without resistance. Laboratory findings disclosed a white blood cell count of 19,600 mmc, with 85% neutrophils, C-reactive protein (CRP) of 2.35 mg/dL and erythrocyte sedimentation rate at 1 hour of 44.

An ultrasound (US) examination was performed demonstrating dilated fluid-filled small bowel loops, moderate amount of free fluid and a mass in the mesogastrium. No classic findings of intussusception as the "target" or the "doughnut" sign were found. Thus, the radiologist suggested to integrate the US study with a Computed Tomography scan (CT scan).

The CT scan showed a bowel obstruction and a U-shaped loop of the small bowel within the centre of abdomen, consistent with volvulus [Figure 1]. The lumen of this loop contained fluid, soft tissue and fat density suggestive of an ileo-ileal intussusception [Figure 2]. The patient underwent surgical treatment with a diagnosis of ileo-ileal intussusception and

volvulus. A laparotomy was performed through a median incision and the peritoneal fluid was aspirated. The ileal loops were dilated and a volvulus of the ileum was found in a clockwise direction and was reduced without difficulties. The bowel immediately resumed its normal colour. The ileo-ileal intussusception was located approximately 30 cm from the ileo-caecal valve and it was manually reduced. The invaginated intestine was initially intensely purple, but it recovered its viability. The revision of the intestinal coil showed a polypoid mass within ileal lumen approximately 50 cm proximal to the ileo-caecal valve. The mass was resected together with 7 cm of small bowel and the continuity was restored by end-to end anastomosis. Histology assessment of the mass showed findings suggestive of solitary hamartomatous polyp (SHP). The patient's postoperative course was uneventful. Three months after surgery, the patient underwent a contrast study of the bowel, an upper gastrointestinal endoscopy and colonoscopy that excluded the presence of intraluminal lesions. At follow-up of 48 months, the patient was well without recurrence neither other lesions at instrumental follow-up.

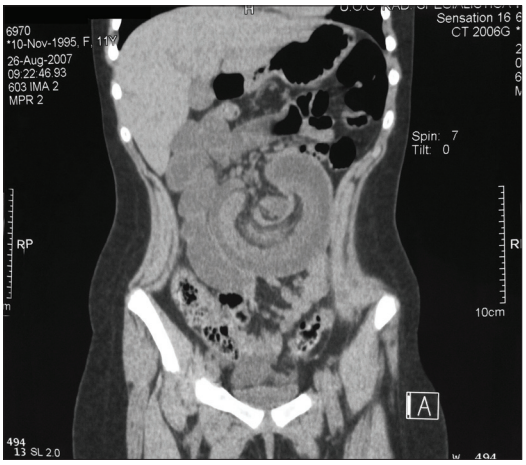



**Figure 1: Non-contrast computed tomography coronal view showing a bowel obstruction and a U-shaped loop of the small bowel within the centre of abdomen consistent with midgut volvulus**

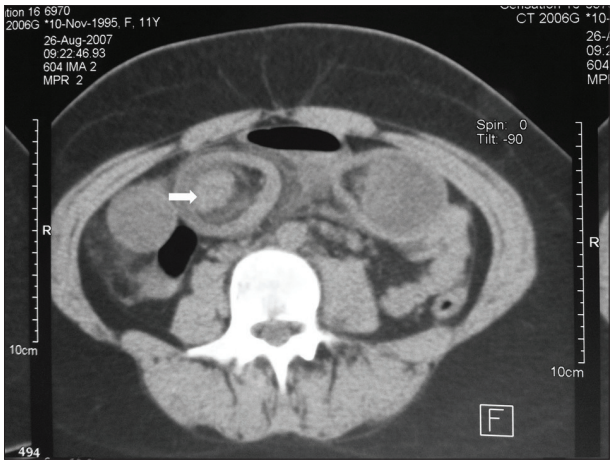



**Figure 2: Non-contrast computed tomography transverse view showing the presence within the bowel lumen of fluid, soft tissue and fat density as small bowel intussusceptions. The arrow shows intussusception**

## DISCUSSION

SBI is uncommon. Kornecki observed an SBI in 14% of 310 patients with intussusceptions (2). Moreover, SBI is easily clinically overlooked with the risk of a delayed diagnosis and consequent bowel ischemia and necrosis. The midgut volvulus is a rare complication of SBI, the first case was described by Young in a 15-year-old girl.[3]

The association of ileocolic intussusception and intestinal malrotation has been described as Waugh's syndrome by Dr. George E. Waugh who first described three cases in 1911. The Waugh's syndrome may be complicated by a midgut volvulus.

In our case, SBI was complicated by a midgut volvulus; this condition was not due to intestinal malrotation. Young, describing a patient with SBI complicated by midgut volvulus due to a polyp, concluded that "total mass was very bulky and solid and the weight of this in an active gut combined to provoke the volvulus".[3]

The literature review showed only five cases of SBI complicated by midgut volvulus in children;[4-7] our patient is the sixth. The pathological lesion of the bowel, acting as an anatomical lead point, was reported in three patients; the present case is the fourth [Table 1].

The small bowel is also the most difficult intestinal segment to evaluate because of its central position, length and complex looped morphology.

**Table 1: Review of literature**

| Author | Age | Operative findings | Led point |
|---|---|---|---|
| Young HB. BMJ 1951;3:226-7.[3] | 15 y | SBI and volvulus | Polyp |
| Tagart REB. Br Med J 1953; 29:475.[4] | 9 y | SBI and volvulus | Meckel's diverticulum |
| Viaggio J. Prensa Med Argent 1957; 44(19):1434 38.[5] | 7 y | SBI and volvulus | Idiopathic |
| Lo Bello G. Radiol Med 2003; 105(3):246-69.[6] | 13 y | SBI and volvulus | Myoepithelial hamartoma |
| Dawrant MJ. Pediatr Surg Int 2005; 21: 730-32.[7] | 8 y | SBI and volvulus | Idiopathic |
| Present case, 2007 | 11 y | SBI and volvulus | Solitary hamartomatous polyp |

The diagnosis of SBI is not easy. Usually it is based on the clinical findings and imaging. Abdominal US has become the first-line diagnostic tool for SBI with a rate of sensitivity of 84%.[8] The diagnosis of volvulus can be reliable with US of colour Doppler image.[9] It is also reported that US is very useful in the evaluation of gastrointestinal obstruction and can suggest the diagnosis in atypical clinical presentation of midgut volvulus.[10-12]

The CT scan does not usually represent the initial diagnostic tool in children with small-bowel obstruction, although it can be used in patients with a long-standing obstruction and marked bowel distension and may be helpful to confirm an SBI after ultrasonographic evidence.[1,12]

In our case, US was unable to identify the characteristic signs of intussusception, showing only marked bowel distension and a mass in the mesogastrium. The CT scan, performed as second step after US examination, was enlightening showing the presence within the lumen of fluid and soft tissue, and the fat density suggested an SBI and a U-shaped loop of the small bowel consistent with volvulus.

Abdominal CT scan was very important in preoperative study in determining the cause of small bowel obstruction. It is particularly helpful when US is ineffective, avoiding reduction enema and reducing the waiting time for surgical treatment.

A lead point of SBI was found at surgery. The histological assessment of the mass showed findings suggestive of a solitary Peutz-Jeghers type hamartomatous polyp. It is still debated if SHP represents an incomplete form of Peutz-Jeghers syndrome or a different entity. The microscopic appearance and the clinical diagnosis enable to differentiate it from a Peutz-Jeghers syndrome. In our case, no clinical sign as peri-orificial mucocutaneous pigmentation was observed. Family history was negative and no recurrence was demonstrated at follow-up after 48 months.

In conclusion, despite the midgut volvulus being a rare complication of the SBI, its early identification allows the surgeon to choose the most appropriate management.

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