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Case Report

Multiple Intussusceptions with multiple polyps as identifiable lead point: A curious case report ☆☆☆

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ABSTRACT

Intussusception is a condition in which the proximal segment of the bowel telescopes into the distal segment. 90% of the cases, especially in infants and toddlers, do not have any lead point and are due to lymphoid hypertrophy, following a viral infection. The presence of polyps in the form of lead point in children is rare. In fact, multiple polyps presenting as multiple sites intussusception are much rarer. We report a case of multiple intussusceptions in a 11-year-old female who presented with complaints of pain abdomen and blood-mixed stool. On imaging examination, she was found to having multiple polyps serving as a possible lead point, and was managed with hydroreduction. Imaging in case of multiple intussusception typically reveals concentric rings of bowel within the bowel giving a “target sign” on axial scans and a “pseudo-kidney sign” on coronal/sagittal CT scans. Our case report shows that, multiple intussusception due to multiple polyps can be a possibility for acute pain abdomen in children.

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Introduction

Intussusception is a condition in which the proximal segment of the bowel telescopes into the distal segment [1]. The donor loop is termed “intussusceptum” and the receiving loop is termed “intussusciptens” [2]. According to the site, it is classi-

fied into 3 types: small bowel, large bowel, or both [1]. It is the most common cause of intestinal obstruction among infants and toddlers between 6 and 18 months of age [3]. About 90% of the cases do not have any lead point and are due to lymphoid hypertrophy, following a viral infection [4]. Adult Intussusceptions account for only 5% of the cases [5]. Intussusception in adults is rare and is due to well-defined structural abnormal-

Abbreviations: US, Ultrasonography; CT, Computed Tomography; CECT, Contrast Enhanced Computed Tomography.

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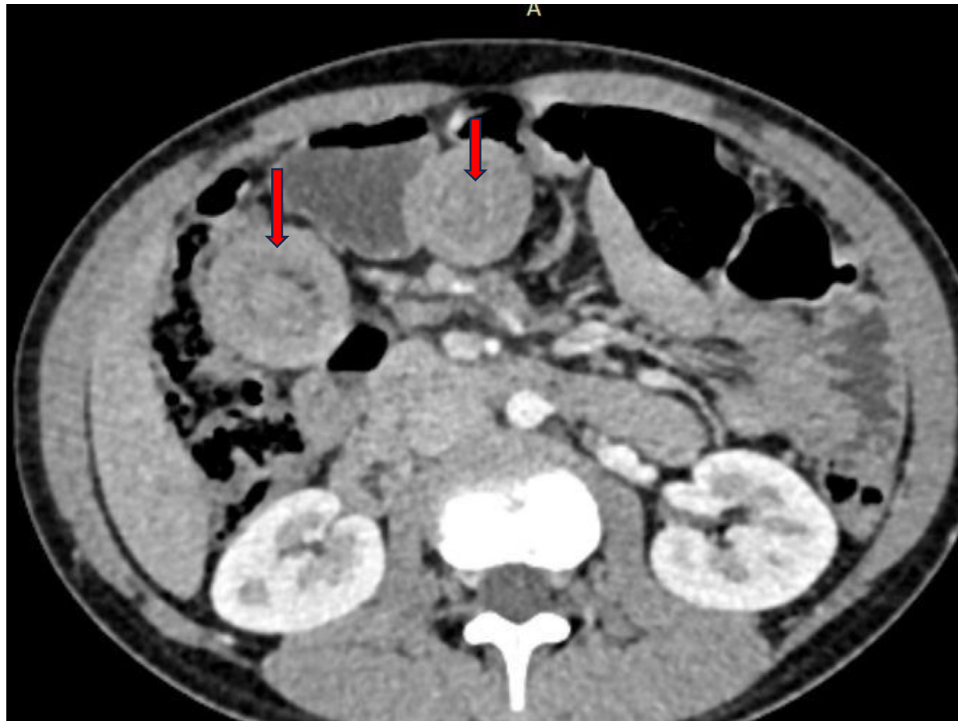


Fig. 1 – CECT abdomen showing concentric layer of bowel within bowel in subhepatic and epigastric region (marked by red arrow).

ity serving as lead point [6]. Recurrent intussusception occurs in 8%-15% of cases and generally in patients older than 1 year of age [7]. Imaging typically reveals concentric rings of bowel within the bowel giving a “target sign” on axial scans and a “pseudo-kidney sign” on coronal/sagittal scans [8]. The presence of free fluid between bowel loops in intussusception is a sign that indicates the irreducibility of hydro-reduction [9]. The identification of lead points by imaging modalities like US and CT play a pivotal role in diagnosing Intussusception with high sensitivity and specificity [10].

Intussusceptions are usually single. We report a case of multiple intussusceptions in a 11-year-old female due to multiple polyps serving as a possible lead point.

Case presentation

An 11-year female presented to the emergency department with a history of pain abdomen and blood-mixed stool sometimes for 3 days. The pain was of increasing intensity. There was no history of vomiting, fever, or diarrhea excluding acute gastroenteritis. There was no history of chronic cough or any mass protruding from the anus excluding rectal prolapse. According to the mother, she has a recurrent history of pain abdomen, most of the time relieved by pain medications. This time, the pain was not relieved so she was brought to emergency. On examination, mild tenderness was noted in the abdomen in the right upper quadrant and pelvic region. Vitals were within normal limits. The rest of the systemic examinations were insignificant. Lab parameters were within normal

limits. US of the abdomen revealed concentric rings of bowel within the bowel appearance giving a “Target sign” in the subhepatic region and pelvic region. Based on this, a diagnosis of Intussusception was made and the patient was planned for hydro-reduction. Based on the fact of the presence of multiple intussusceptions in our patient and taking into consideration the unusual age of the presentation of Intussusception, a CT of the abdomen was done to identify any lead point. CT showed multiple intussusception and multiple polyps within the small bowel in subhepatic, epigastric and pelvic regions (Figs. 1-4). Hydro-reduction was done which turned out to be successful in relieving the symptoms and the patient was referred to pediatric surgery department for further management.

Discussion

Small bowel intussusception is rare in adults, comprising about 5% of all the cases [6]. Multiple simultaneous small bowel intussusceptions are rare. Small bowel intussusception in adults is mostly attributed to the presence of pathological lead point in single region of the bowel. However, the presence of multiple site lead point is a rare occurrence. The mechanism behind the development of intussusception from the lead point is still unknown. But it has been postulated that, the presence of lead point or any pathological lesion alters the normal peristaltic pattern of the bowel causing the bowel to be invaginated into the following bowel segment, thus leading to the formation of intussusception. In cases where no lead point



Fig. 2 – CECT abdomen showing another similar lesion in the right pelvic region (marked by yellow arrow).

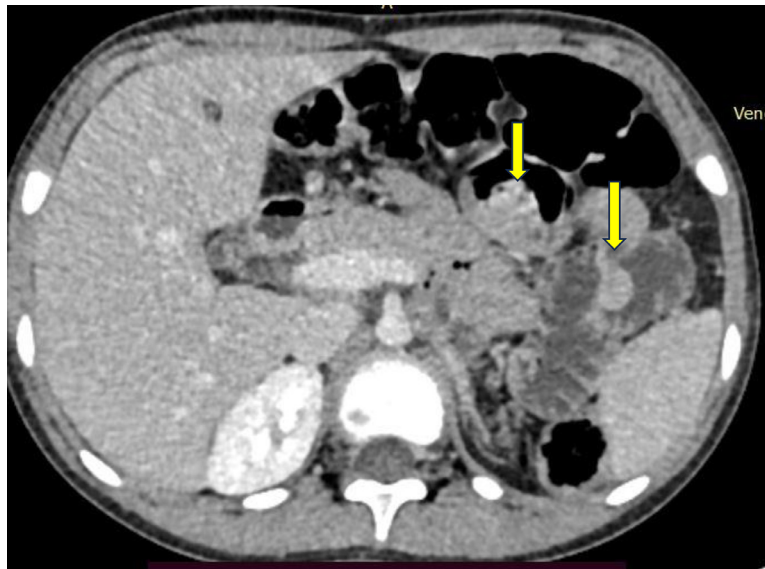


Fig. 3 – CECT abdomen showing multiple enhancing polyps in small bowels (yellow arrow).

can be identified, intussusception can be due to submucosal bowel edema, fibrous adhesions or dysrhythmic contractions [11]. The lesions that serve as the lead point include coeliac disease, Crohn's disease, Meckel's diverticulum, adhesions, endometriosis, enteric polyps, tuberculosis, adenocarcinoma, carcinoid tumors, gastrointestinal stromal tumors, metastatic carcinoma and neuroendocrine tumors [12]. Among all the lesions serving as the lead point, benign lesions are most common (60%), followed by small bowel malignancy (30%), with 10% of the cases being idiopathic [13]. The malignancies may include either the primary lesions or the metastatic lesions from the lung, breast, and malignant melanoma [14]. Apart from these causes, there is term named “functional intussusception” which occurs in the setting of metabolic derangement such as severe hyperglycemia, metabolic acidosis, or hyperkalemia, is rare [15]. Lee et al. [16] reported a case of mul-

tiple intussusception (jejunojejunal and ileoileal) in a 9-year-old male following blunt abdominal trauma. Our case was unique in the fact that our patient, although a child had intussusception in multiple areas of small intestine due to the presence of multiple polyps, the occurrence of which is rare in children. Most of the cases of intussusception occurs in the ileo-cecal region. However, the incidence of it in children is usually associated with history of upper respiratory tract infections and viral gastroenteritis [17]. Following are the factors that are responsible for the pathophysiology of intussusception: (a) mature lymphoid tissue may develop into lead points of intussusception in the setting of viral/bacterial infection causing lymphoid hyperplasia, (b) the thin wall of the small intestine makes it easy to invaginate, (c) narrow ileal lumen which is obstructed and develop into the lead point of intussusception, (d) underdeveloped fixation of the ileocecal

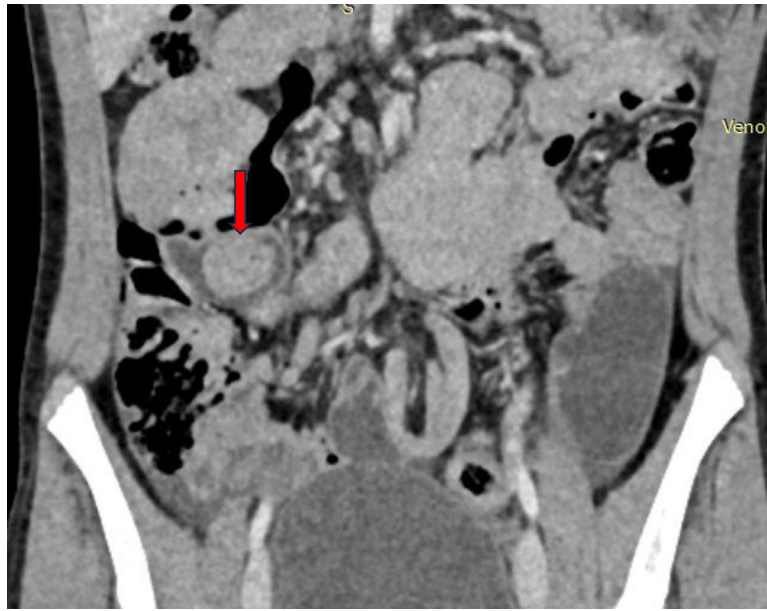


Fig. 4 – CECT abdomen (coronal image) showing another polyp in small bowel loop (red arrow).

region [18]. The presentation of intussusception in children is often acute, with a triad of abdominal pain, palpable abdominal mass and bloody mucus like bowel movement often termed as “currant jelly” [19]. Adults with intussusception may present with abdominal pain, diarrhea, constipation, gastrointestinal hemorrhage, and sometimes abdominal mass [19]. Our patient had pain abdomen and blood-mixed stool as the major complaints. On physical examination, patient may show localized or diffuse abdominal tenderness and pressure pain [20]. The diagnosis of intussusception can be made clinically in only 50% of the cases. The diagnostic evaluation thus depends on the imaging tools for making the correct diagnosis [3]. The features of Intussusception in US include the “target sign” and “pseudo-kidney sign” with a hypoechoic outer ring and hyperechoic center [2]. It has proved to be beneficial in adult intussusception by allowing better visualization of all planes of the abdominal wall and allows for a timely diagnosis when modalities like CT are not available [21]. CT abdomen is currently considered as the most superior modality for diagnosing adult intussusception owing to its higher sensitivity [21]. It shows a characteristic “target mass”, describing a concentric, alternating layers of hyperechoic and hypoechoic shadowing within the intestine [21]. The ‘target’ shaped mass may contain mesenteric fat and vessels within it [1]. Additionally, a CT scan can define the site and the nature of the mass and its relationship with the surrounding tissues. It may also help in staging of the patients with suspected malignancy serving as a lead point for intussusception [20]. US and CT scan of abdomen was used as the diagnostic modalities in our case which showed the presence of multiple intussusceptions. Contrast enema can be useful in diagnosing colonic intussusceptions in adults. However, it does not exclude the presence of small bowel intussusceptions. Also, it does not have the same therapeutic importance as that in adults [22]. Due to frequent association with lead points and the possibility of malignant tumor, surgery is considered to be

the mainstay of treatment in case of adult intussusception. However, hydrostatic reduction is the mainstay of treatment in case of children [7]. Also, multiple intussusceptions often do not reduce with hydrostatic enema and thus require surgery [8]. Surgery involves the resection of the affected portion of the bowel with or without anastomosis [9]. The symptoms in our patient was relieved after hydro-reduction but she was referred to pediatric surgery department in view of the presence of multiple polyps in the small intestine.

Conclusion

Lead point in the form of multiple polyps within the small intestine causing multiple intussusceptions could also be a possibility of acute pain abdomen in children although the occurrence of polyps in the form of lead point is rare in children. This thing must be taken into consideration by the treating physicians while approaching the case of pain abdomen in case of children.

Ethical approval

The study is exempt from ethical approval in our institution.

Patient consent

Written informed consent was obtained from the patient's parents for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Provenance and peer review

Not commissioned, externally peer-reviewed.

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