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Adult intussusception caused by myoepithelial hamartoma in the small bowel: A case report

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ABSTRACT

INTRODUCTION: Intussusception is rare in adults and can have acute, intermittent or chronic presentations. We present an unusual case of intussusception in an adult.
PRESENTATION OF CASE: A 25 year old male presented with sudden severe abdominal pain and vomiting. He had no relevant medical history. Mechanical small bowel obstruction was confirmed on imaging and laparotomy revealed a nodular lead point in the submucosa of the ileum. He had resection of a segment of small bowel with a primary anastamosis. Histopathology of the lesion demonstrated myoepithelial hamartoma (MEH), a rare cause of intussusception.
DISCUSSION: Although intussusception is not uncommon in children, it is rare in adults. Management delays are a major cause of morbidity. This report details our management of a case of intussusception caused by MEH in an otherwise healthy adult.
CONCLUSION: Intussusception caused by MEH is a rare but serious cause of mechanical bowel obstruction. We propose that surgeons should consider this diagnosis in atypical cases of bowel obstruction and expedite laparotomy when it is suspected.

1. Introduction

Intussusception is a relatively common cause of acute abdomen in the paediatric age group, but it is rare in adults [1,2]. It is a disorder in which part of the bowel slides into adjacent bowel. This then leads to bowel obstruction and its sequelae. In children, it causes sudden severe progressive abdominal pain with nausea and vomiting. In later stages, red currant jelly stools are sometimes seen. In adults, pain is often intermittent, and it can be associated with a variety of other symptoms [3]. In stable children, non-operative reduction with hydrostatic or pneumatic enemas under ultrasound or fluoroscopic guidance is an appropriate initial approach. However, in adults, surgical treatment is the norm, and it is common to find a well defined lesion serving as the lead point [4,5].

Ultrasound is the method of choice for diagnosis [6]. Although it is operator dependent, it can reliably confirm or exclude the diagnosis without the use of radiation. Plain abdominal radiography is less sensitive and less specific [7], and therefore, should be avoided if intussusception is suspected. Computed tomography (CT) scanning can also reliably achieve the diagnosis [3], although its use is limited in children due to concerns regarding radiation and the need for sedatives.

2. Presentation of case

A 24 year old man presented with sudden onset of severe colicky central abdominal pain. There was associated anorexia and nausea and the patient vomited about ten times. At presentation, his symptoms were of 12 h duration. His complaints were progressively worsening. He was an otherwise healthy adult and had no previous surgery. On examination, he had a distended abdomen with mild generalised tenderness. There were no signs of peritonitis, and there were no detectable masses or hernias.

White cell count was elevated while other blood investigations were within normal limits. Plain abdominal X-ray and CT scan (Fig. 1) showed mechanical small bowel obstruction.

He was managed initially with intravenous fluids and opiate analgesia. He had placement of a nasogastric tube and a urinary catheter. Over the following hours, his pain worsened although his vital signs remained stable, and he did not develop peritonitis. He underwent an emergency laparotomy at 12 h following hospital admission. We did not persist with conservative management as

Abbreviations: MEH, myoepithelial hamartoma.

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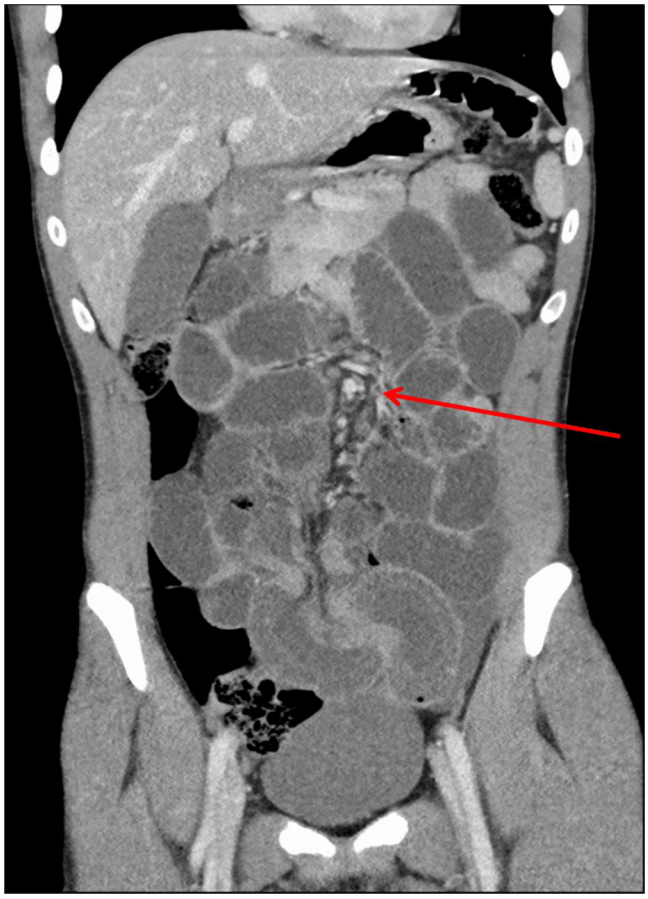

Fig. 1. Contrast enhanced computed tomography scanning image demonstrating mechanical small bowel obstruction with a transition zone (arrow).

there was obvious mechanical obstruction with worsening symptoms and with no obvious cause.

Laparotomy confirmed ileo–ileal intussusception about 2–3 feet from the ileocaecal valve (Fig. 2). Once it was reduced, a segment of gangrenous bowel was found. The lead point of the intussusception was a submucosal nodule measuring approximately 1.2 cm (Fig. 3).

The patient underwent a small bowel resection and primary anastomosis and made a full recovery.

The histology of the submucosal nodule showed ductular structures some of which were cystically dilated and lined by pancreatic duct type epithelium (Fig. 4) and surrounded by whorling smooth muscle fibres (Fig. 5) consistent with the diagnosis of myoepithelial hamartoma causing the intussusception.

3. Discussion

Intussusception is not uncommon in children, but it is rare in adult [4]. Intussusception occurs when a proximal segment of bowel becomes telescoped in to the bowel distal to it [4]. There is a lead point, termed the intussusceptum, which gives rise to the intussusception, and it is at that point that the pathology initiating the process is usually found [5].

Diagnosis of adult intussusceptions is less frequently made preoperatively and mechanical intestinal obstruction will usually be the reason for surgical intervention. Most cases of intussusception in adult have a demonstrable cause unlike what occurs in the paediatric population [4,5]. Lymphoid hyperplasia, Meckel's diverticulum, polyps, lymphoma, lipoma, Peutz–Jegher polyps, primary adenocarcinoma and myoepithelial hamartoma are listed as some of the causes of intussusception [2,4]. Myoepithelial

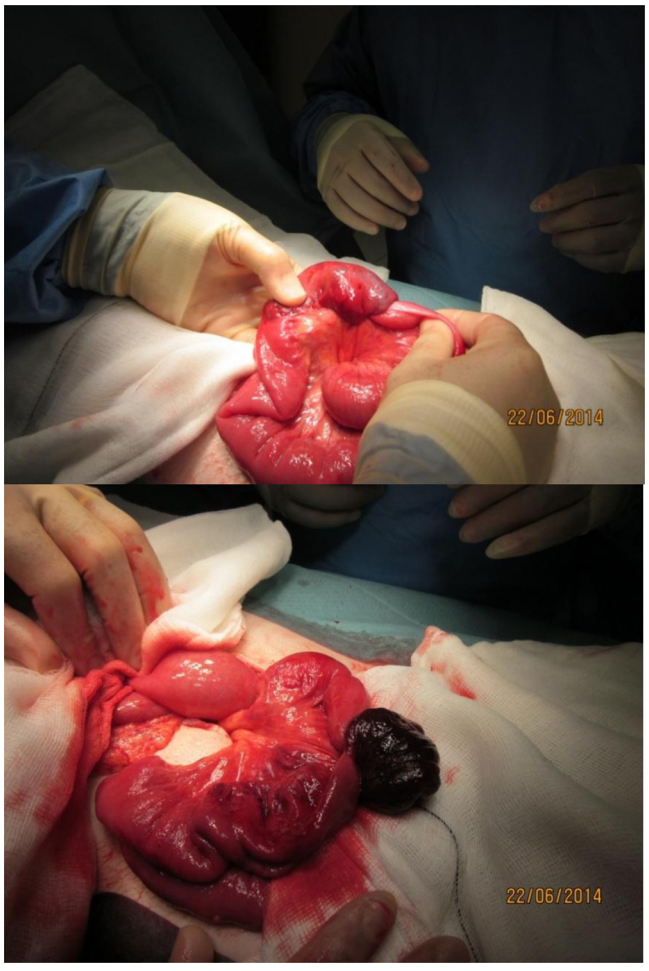

Fig. 2. Intussusception as initially found at laparotomy and after reduction showing gangrenous bowel.

hamartoma(MEH) as found in this patient is a rare cause [2] of intussusception.

Myoepithelial hamartoma has been called by various names in the literature. In addition to myoepitialial hamartoma, names such as adenomyoma, ectopic pancreas, and foregut choristoma (coined from the fact that all tissues are from the embryologic for gut) have been used [2,4]. The following are the pathologic features that

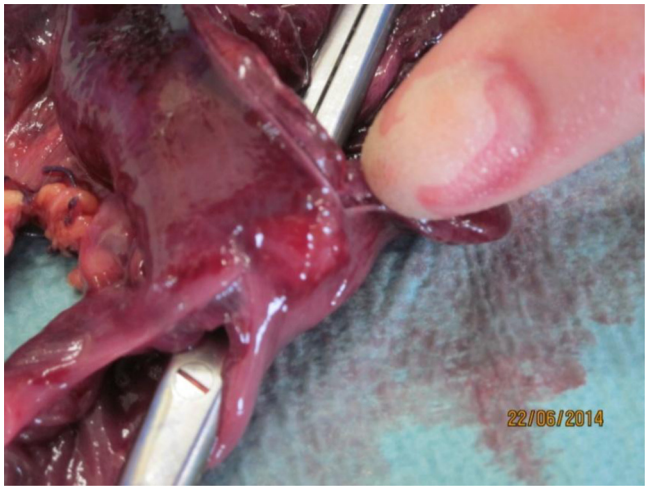

Fig. 3. Submucosal nodule.