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Case Report

Ileo-Ileal Intussusception Presenting as Cullen's Sign: A Case Report

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Abstract

Intussusception is defined as the telescoping of one segment of intestines into another causing intestinal obstruction. This condition although common in children is considered a rare condition in adults and is usually present secondary to a pathological lead point like a neoplasm in adults. It usually presents with non-specific complaints like abdominal pain and as such peri umbilical ecchymosis (Cullen's sign) has not been seen as a presenting symptom of intussusception. Here we present a rare case of acute abdominal pain with contrast imaging of abdomen revealing ileo ileal intussusception. The patient had periumblical ecchymosis (Cullen s sign) on abdominal examination.

Keywords: reverden suture; caesarean section; endometritis

Introduction

Intussusception is the telescoping of a one segment of bowel into its adjacent segment causing intestinal obstruction which can further cause bowel ischemia, bowel necrosis, sepsis and fatal gangrene. It is a known cause of abdominal pain in pediatric population however it is rarer in adults and since abdominal pain is the only presenting symptom which makes it a challenging diagnosis in adults. Radiological imaging especially CT is usually needed to make a definitive diagnosis of intussusception. Definitive treatment involves surgical intervention although it argued idiopathic intussusception which doesn't involve neoplasms as a lead point can be managed by supportive care also [1] Cullen's sign is defined as a superficial edema with bruising in the subcutaneous fatty tissue around the peri-umbilical area. This sign is most often identified in hemorrhagic pancreatitis. The sign usually takes 2-3 days to appear and is used as a clinical sign for the diagnosis of acute pancreatitis mainly [3] We searched the literature with the keywords intussusception and Cullen's sign and could not find the literature in which Cullen's sign was associated with intussusception. Here we present a case of abdominal pain with cullens sign in a young male who turned out be a case of ileo ileal intucesseption on contrast imaging of abdomen.

Case

A 24-year-old male with no co-morbidities presented to accident & emergency department with acute abdominal pain of 2 days duration. It was associated with one episode of non-projectile vomiting containing food contents. General physical examination was unremarkable except for tachycardia. On per abdominal examination there was peri-umbilical ecchymosis consistent with Cullens sign with diffuse abdominal tenderness and guarding.

Figure 1: Peri umbilical ecchymosis (Cullen's sign).

There was no history of fever,arthralgias, bleeding per rectum or hematuria and no evidence of rash on lower limbs or buttocks. Having acute abdominal pain with periumbilical ecchymosis (Cullen sign) a possibility of acute pancreatitis was made.serum amylase and lipase were normal and ultrasonography of abdomen revealed no gross pathology in pancreas. The patient was subjected to contrast CT abdomen which revealed features of ileo-ileal intussusception with normal pancreas (figure- 2)

Figure 2: Contrast enhanced axial scans of abdomen showing ileo ileal intussuception

The patient underwent exploratory laparotomy and a 5 cm loop of ileo-ileal intussusception with gangrenous changes was found. Primary resection and end to end anastomosis was done and abdomen closed back in layers. The post-op course remained uneventful and on day 3 of exploration the peri-umbilical ecchymosis disappeared(fig 3). Orals were started on day 3 and patient was discharged from hospital in a hemodynamically stable condition after 5 days.

Figure 3: Disappearance of peri-umbilical ecchymosis (Cullens sign) on 3rd postoperative day.

Discussion

Intussusception is a condition in which a proximal part of the intestine (commonly small intestine) gets folded into the adjacent distal part which can cause bowel obstruction as well as intestinal ischemia as the blood supply to the affected bowel is compressed and subsequently compromised which progressively leads to necrosis followed by gangrene, perforation and sepsis. Symptoms of intussusception include crampy abdominal pain like in our case which might be intermittent or constant, vomiting (can be bilious), bloating and even bloody stools[7,8,9] Fever is usually not a symptom of intussusception, it occurs only after necrosis and intestinal perforation[10] Signs and symptoms which suggest decompensation can also occur such as hypothermia, hyperthermia, hypotension and tachycardia secondary to complications like bowel necrosis or sepsis[2] As the adult intussusception presents with variable presentation , it makes the preoperative diagnosis a challenging task. Plain abdominal films are the first investigation since in most cases the obstructive symptoms are the chief complaints. Such films usually show signs of intestinal obstruction and provide information regarding site of obstruction also[4,12] Upper Gastrointestinal contrast series shows a “stacked coin” or “coil spring” appearance. Barium enema is suitable for patients with colo-colic or ileo-colic intussusception which demonstrates a “cup-shaped” filling defect or “coil spring” or “spring” appearance[4,13,14] USG is also a sensitive investigation for intussusception in both children and adults. It shows the classical imaging features of intussusception like the “dough-nut” sign or the “target” sign on transverse view and “kidney” sign or “hayfork” sign in longitudinal view[15,16] USG has certain drawbacks like it is heavily observer or operator dependent investigation which requires an experienced handler and interpreter like in our reference case ultrasound did not pick any abnormality[9] Abdominal CT is currently considered the most sensitive investigation to confirm intussusception. The characteristic features of CT scan include an unhomogeneous “target” or “sausage”- shaped soft- tissue mass with a layering effect. A CT scan may define the location, the nature of the mass, its relationship to surrounding tissues and additionally it may help staging the patient with suspected malignancy causing the intussusceptions [9,4] Treatment in adults usually consists of surgical intervention as the preoperative diagnosis is usually missed or delayed owing to its non specific presentation also adult intussusception occurs mainly secondary to a pathological lead point like a malignancy or a structural abnormality and hence surgery remains the definitive choice of treatment. In contrast intussusception in pediatric patients usually presents as a primary and benign condition and preoperative reduction with barium or air can be done. In adults reduction and manipulation is not done due to risks like intraluminal seeding and venous dissemination of tumor, perforation and seeding of micro-organisms and tumor into the peritoneal cavity and increased risk of anastomotic complications of the manipulated friable and edematous bowel tissue [4,17,18,19,20] Moreover reduction should not be attempted if there are signs of inflammation or ischemia of bowel wall [21]

Therefore, in patients with ileo-colic, ileo-cecal and colo-colic intussusceptions especially those more than 60 years of age and due to the high incidence of bowel malignancy formal resection using appropriate oncologic techniques are recommended with the construction of a primary anastomosis between healthy and viable tissue [4,19,20,22,23,24]

Cullen’s sign is defined as the hemorrhagic discoloration of the peri-umbilical area due to intra peritoneal hemorrhage from any cause [11] Cullen sign is usually seen in acute hemorrhagic pancreatitis, ruptured ectopic pregnancy, perforated bowel viscus and abdominal trauma. Here we present a rare case of acute abdomen with Cullen sign which turned out be ileo ileal intussusception on contrast imaging of abdomen. The possible cause behind the Cullen’s sign in intussusception could be that when the bowel loops compress on each other the blood supply gets compressed and compromised subsequently which leads to ischemia of the bowel segment and if the intussusception continues it leads to necrosis. As this process of vascular insult occurs it leads to the release of the inflammatory mediators from the local damaged tissue which further leads to vasodilation and subsequently increases the vascular permeability and hence leading to accumulation of blood around the peri-umbilical region. Also, as the structural integrity of the bowel tissue is compromised secondary to ischemia, it leads to extravasation of pancreatic enzymes which may also cause local fat necrosis and present as tender erythematous lesions of subcutaneous tissue.

Conclusion

Cullen sign is a peri umbilical ecchymosis seen in acute abdominal conditions like pancreatitis, abdominal trauma and perforated bowel viscus. we present a rare a case of acute abdomen turned out to be ileo ileal intussusception with Cullen sign on abdominal examination. More cases need to be seen before cullens sign can be certainly attributed to intussusception.

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Conflict of interests:

None declared

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