



## Intussusception Secondary to Pneumatosis Intestinalis: A Rare Case Report

Authors

Dr Arvind Kumar Prabhat<sup>1</sup>, Dr Niranjan Dash<sup>2</sup>, Dr J.M. Gadekar<sup>3</sup>,  
Dr Mrs. V.J.Thipse<sup>4</sup>

<sup>1</sup>Postgraduate Student, <sup>2</sup>Professor, <sup>3</sup>Professor and Head of Department, <sup>4</sup>Professor  
Department of General Surgery, DVVPF'S Medical College and Hospital, Ahmednagar, Maharashtra, India

### Abstract

*Pneumatosis intestinalis (PI) is a very rare intestinal disease, characterized by presence of multilocular cysts in the intestinal wall. Complications occur due to PI in about 3% of cases<sup>(1)</sup> and include intussusception, intestinal obstruction, volvulus and intestinal perforation. We report a 39 years old female patient who presented in our hospital with acute abdominal pain in the epigastric region with vomiting and loose stools mixed with blood since 2 days. USG & CECT abdomen revealed colo-colic intussusception of transverse colon. Right hemicolectomy with ileo-transverse anastomosis was performed. The immediate post-operative was uneventful and patient discharged on 12<sup>th</sup> post-operative day. Histopathology report of resected segment of colon was PI.*

**Keywords:** Intussusception; Ileo-transverse anastomosis; Pneumatosis intestinalis (PI)

### Introduction

PI presents as idiopathic and secondary forms of the disease. The primary idiopathic form has multiple thin-walled cysts present in the subserosa or submucosa of the gut wall. This form is usually asymptomatic and diagnosed incidentally through upper GI scopy, colonoscopy or radiological imaging.<sup>(2)</sup> Secondary form is associated with necrotic and obstructive gastrointestinal disease.<sup>(3)</sup> In our case, intussusception was caused by small and large cystic lesions of secondary forms, which were present in the submucosa of transverse colon.

PI was first described by D U Vernoy in 1730 from cadaveric dissection.<sup>(4)</sup>

Clinical presentation of PI is variable and remains a difficult condition to diagnose clinically. It

presents a challenge to the surgeons for management in both an acute and outpatient setting.

Pneumatosis intestinalis has numerous etiologies resulting in a spectrum of clinical presentation ranging from no overt symptoms to bowel obstruction. In our case, the patient presents as acute abdominal pain, vomiting along with loose stools mixed with blood because of colo-colic intussusception.

### Case Report

A 39-years-old female patient, presented to the casualty in DVVPF'S Medical College and Hospital with acute abdominal pain in the epigastric region, five episodes of vomiting and loose stools mixed with blood since two days. On

clinical examination, pain and tenderness was present in epigastric region. Patient was admitted in surgical ICU. X- ray erect abdomen (Fig 1) showed multiple air-fluid level. USG of abdomen and pelvis revealed bowel within bowel appearance in the periumbilical region along with few dilated bowel loops with maximum diameter 4.1 cm and inter bowel free fluid suggestive of intussusception. CECT abdomen and pelvis (Fig 2,3) showed long segment bowel within bowel appearance in the region of transverse colon with involved segment measuring approximately 10-12 cm. There is proximal dilation of caecum and ascending colon seen with maximum caecal diameter measuring 5.8 cm. All routine investigations were within the normal limits except haemoglobin (HB : 8.6 gm/dl) and TLC count(TLC :18400/cmm). Two units of whole blood were transfused. Pre-anaesthetic check-up done and patient was taken up for surgery. On exploratory laparotomy (Fig 4), colo-colic intussusception of transverse colon with dilation of caecum and ascending colon was found. Right hemicolectomy and ileo-transverse anastomosis was performed. Resected segment of bowel was sent for histopathology. The immediate post operative period was uneventful. Oral feeds were started on 5<sup>th</sup> postoperative day. The patient recovered satisfactorily and discharged after removal of skin sutures on 12<sup>th</sup> postoperative day. During the follow-up visit after one month, the operative scar was well healed and patient was absolutely asymptomatic.

#### **Histopathology Report**

Gross: Resected segment of intestine measured 30 cm in length (Fig 5) along with caecum and appendix. Mucosal surface showed polypoidal and cystic lesion measuring 3 cm in diameter, adjoining mucosal folds were raised and dilated.

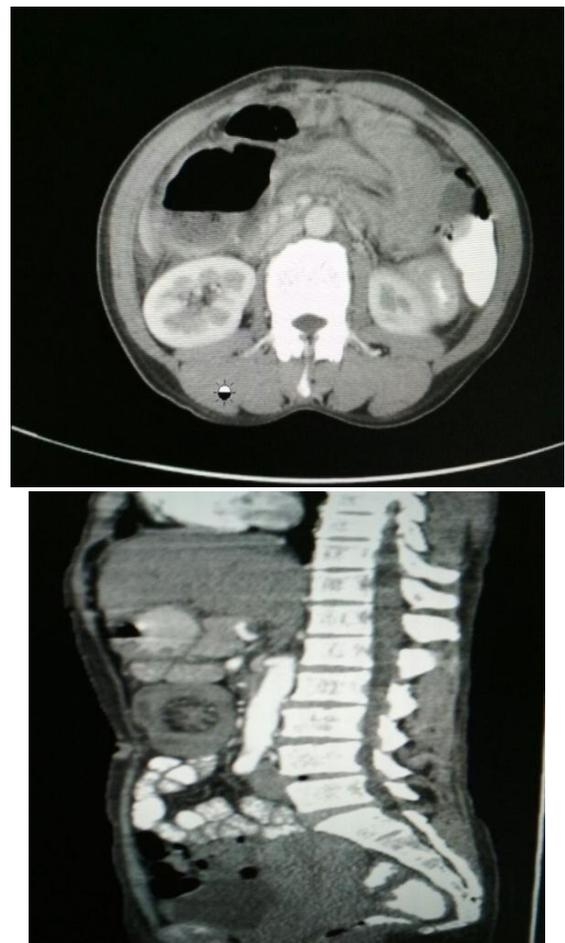
Cut-Section: Revealed multiple cysts overlined by mucosa (submucosal location).

Microscopy: Lesional tissue showed cystic lesion large as well as small in the submucosa. These were overlined by normal appearing mucosa and muscularis mucosae. At the base, muscularis

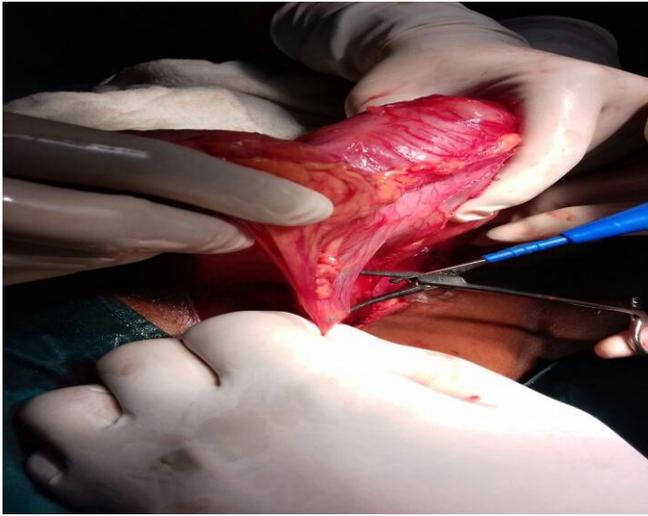
propria and serosa were noted. Adjoining tissue showed dilated lymphatics and blood vessels. All above findings were suggestive of Pneumatosis intestinalis.



**Fig 1:** X-ray erect abdomen: shows multiple air-fluid level



**Fig 2 & 3:** CECT abdomen and Pelvis: Shows colo – colic intussusception



**Fig 4** Intraoperative : Segment of intussusception



**Fig 5** Resected segment of colon of length approximately 30 cm containing cecum, appendix and ascending colon.



**Fig 6:** Segment of colon showing multiple cysts

### Discussion

PI is a very rare disease characterized by the presence of gaseous cysts filled with hydrogen,

nitrogen and carbon dioxide in the gut wall.<sup>(5)</sup> Gaseous cysts are located beneath the serosa and mucosa of the gut wall. Colonic localization of gaseous cysts is due to an increase in number of the therapeutic and diagnostic procedure with upper GI scopy, barium and colonoscopies.<sup>(6)</sup> Colonoscopy increases the intraluminal pressure. In our case, gaseous cysts were present in the submucosa of transverse colon.

The exact etiology of the PI is still unknown. PI may be due to colonoscopies,<sup>(6)</sup> ileal surgery,<sup>(7)</sup> chronic pulmonary disease,<sup>(8)</sup> connective tissue disorder,<sup>(9)</sup> and ingestion of lactulose/sorbitol.<sup>(10,11)</sup> In our case, the etiology was also unknown.

To explain the etiology of PI, various theories have been proposed: Pulmonary, Mechanical and Bacterial theory.

The Pulmonary theory explains, in patients of asthma and chronic bronchitis, the gas released by the rupture of the alveoli, travels through the mediastinum into the retroperitoneal space and then comes through the perivascular space in the intestinal wall.<sup>(12)</sup> No pulmonary pathologies were present in our case.

The Mechanical theory explains, bowel gas is pushed through a mucosal defect into lymphatic channels and then distributed distally by peristalsis. This may occur secondarily to intussusceptions, bowel obstruction and perforation.<sup>(13)</sup> In our case, the main bowel pathology was intussusception of transverse colon.

The Bacterial theory explains that, submucosal localization of fermenting bacteria like E.coli and clostridia leads to production of gas which is retained by the submucosa and lymphatic channels.<sup>(8)</sup> In our case, PI may be due to colonic bacterial overgrowth.

There is no characteristic clinical presentation of PI. Patients may be asymptomatic or complain of acute abdominal pain, abdominal distension, diarrhea and rectal blood loss. In our case, all these clinical features were present.

X-ray erect abdomen may show a change in the characteristics of the intestinal wall in 2/3<sup>rd</sup> of patients of PI. However 1/3<sup>rd</sup> of the patients do not

have suggestive x-ray and require CECT/MRI, which show a thickened bowel wall containing gas.<sup>(14)</sup> In our case x-ray erect abdomen showed multiple air-fluid levels. CECT abdomen revealed bowel within bowel appearance in region of transverse colon.

Knechtle et al<sup>(15)</sup> establishes a correlation between the clinical presentation, the need for surgery and the final outcome. It is essential to evaluate these parameters, abdominal pain, diarrhea, fever, tenderness, rectal blood loss and hypotension and their severity coupled to laboratory tests. In our case, patient presented with acute abdominal pain, tenderness, vomiting and rectal blood loss along with low hemoglobin and raised TLC count. Patient was evaluated properly and managed successfully.

### Conclusion

The PI is a very rare disease and diagnosis should be based on radiological imaging and histopathological report. The management can be conservative or surgical, depends upon the severity of disease. We have managed our case surgically and have got good result.

### Ethical approval

Written informed consent was obtained from the patient for publication of this case report.

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