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# Imaging of multi-etiological pneumatosis intestinalis in children with AIDS: case report and review

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## ABSTRACT

**Introduction:** Pneumatosis intestinalis is a condition where there is gas inside the intestinal wall. Pneumatosis intestinalis or intramural gas is a particular sign of radiology and is not a disease. Pneumatosis intestinalis in patients with very poor clinical conditions, accompanied by symptoms of abdominal pain or distension, is possible because of the presence of bowel ischemia. One of the causes of Pneumatosis intestinalis is immunodeficiency, such as in AIDS.

**Case Presentation:** Here in we present a 7-year-old HIV AIDS positive male presented with abdominal distension, acute abdomen, and sepsis. His first abdominal radiographs showed intramural gas in linear and circular form along the descending colon, which migrates to the ascending colon after few days. Intramural gas was also seen on his abdominal CT therefore concluded bowel ischemia diagnosis without life threatening signs such as portal vein gas, pneumoperitoneum, or portomesenteric gas. His previous chest CT revealed multiple cystic bronchiectasis in both lung bases.

**Discussion:** Underlying cause of Pneumatosis intestinalis in this case is HIV AIDS related which causing both lung and bowel disease resulting in intramural gas, hence bowel ischemia diagnosis was made.

**Conclusion:** Thorough imaging evaluation is needed in diagnosing pneumatosis intestinalis moreover in HIV AIDS patient to determine prognosis and treatment plan.

**Keywords:** Pneumatosis intestinalis, AIDS, abdominal radiology.

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## INTRODUCTION

Pneumatosis intestinalis (PI) is a condition where there is gas inside the intestinal wall. PI may occur because of a mild process without clinical manifestation until a description of ischemic condition of the intestine.

PI can be divided into two groups, i.e. primary which occurs about 15% and secondary 85%.<sup>1</sup> The gas in the intestinal wall may be a condition occurring locally in the gastrointestinal tract or correlated with the disease of the respiratory system. Primary PI have no basic conditions of either respiration or gastrointestinal, and usually occur in adults which tend to appear in colon. The secondary PI mostly occurs in the small intestine and occurs due to many underlying diseases.

Intramural gas is a collection of air found in subserosa and less frequently in submucosa and muscular layers, the gas can appear in a variety of shapes and sizes

from a few millimeters to a centimeter. PIs are often found on the side of the intestinal mesentery, but it can also be found in antimesenteric or on the mesentery itself.

Interpretation of the radiological findings of PI should be correlated with the patient's clinical condition. Previous studies reported that the image of intestinal pneumatosis can be seen from 22-71% of plain abdominal images that have been confirmed via CT scan.<sup>2</sup>

PI may occur in asymptomatic patient or present in non-harmful diseases, sometimes even sudden findings. PI in patients with very poor clinical conditions, accompanied by symptoms of abdominal pain or distension, is possible because of the presence of bowel ischemia. Meanwhile, PI alone can be caused by many conditions outside the age of the neonate, especially in immunodeficiency condition.

One of the causes of PI is immunodeficiency, both in AIDS, leukemia, post organ transplant, long-term steroid use, and other diseases. However, several literatures had recently observed the presence of PI in cases of patients with AIDS, which are usually associated with underlying opportunistic infections in the gastrointestinal tract. PI in AIDS patients had varying rates of incidence, a report by Woods accounted for 25% of all inpatient AIDS patients whereas in other institutions only 9% were documented.<sup>3</sup>

### The underlying etiology and theory

The cause of PI can be categorized into 4, first is intestinal necrosis, which describes bowel ischemia. 2. Mucosal disruption by ulcers, biopsy procedures, trauma, enteric tube, or inflammatory bowel disease. 3. Increased mucosal permeability due to immunosuppressive conditions such as AIDS, post organ transplant, or chemotherapy. 4. Lung disease resulting in

alveolar disruption and air dissection of the pulmonary interstitium to the intestinal wall, usually found in patients with chronic obstructive pulmonary disease, asthma, cystic fibrosis, barotrauma and chest trauma.<sup>4,5</sup>

PI has been reported to have a special correlation with certain infections in people with AIDS, such as cytomegalovirus (CMV), mycobacterium avium complex (MAC), and cryptosporidium. In addition, PI was also found in patients with immunodeficiency other than AIDS, namely leukemia, lymphoma, long-term steroid use, and organ transplant.<sup>6</sup>

### Pathophysiology

#### Bacterial Theory

A clinical and experimental study has revealed that the gas in pneumatosis comes from bacteria. The analysis of the gas in the cyst in the intestinal wall showed that approximately 50% is composed of hydrogen compared to the normal hydrogen percentage in intestine which is only 14%, thus confirming that the production is produced by bacteria. This pneumatosis is also accompanied by condition of an inflammatory reaction around the cyst. Hyperbaric oxygen has been used for the treatment of pneumatosis and clostridium infection. In some cases, pneumatosis is also a response to antibiotic therapy.<sup>5,7</sup>

#### Mechanical Theory

The gas entering the intestinal wall is caused by direct trauma or increased pressure. Mechanical disruption of the intestinal wall is a major cause of pneumatosis in cases of trauma, surgery, and endoscopy, and is a condition of PI predisposition that correlates with obstruction.<sup>5,7</sup>

#### Mucosal Disruption Theory

Mucosal disruption is the primary cause of almost all PI cases, in which bacteria or gases can forcefully enter the intestinal wall. Mucosal disruption is also the main contributing factor of PI due to inflammation and ischemia. In cases such as Chron's disease and steroid use, depletion of Peyer's plaques may result in PI.<sup>5,7</sup>

#### Pulmonary Disease Theory

Pulmonary diseases that can cause PI include chronic obstructive pulmonary disease, asthma, and cystic fibrosis. In addition, PI may occur due to barotrauma and after thoracic tube insertion.<sup>5,7</sup>

#### PI in AIDS Patient

In children with AIDS, gastrointestinal disorder results mainly from the presence of acute and chronic enteric infections, and less frequently because of gastrointestinal neoplasms. CMV infection can also affect the stomach and colon. CMV colitis is also common and usually leads to toxic clinically and leads to PI, stricture, toxic megacolon, and perforation. Chronic diarrhea is one of the most visible manifestations in AIDS patients and its etiology can be caused by Cryptosporidium, *Isospora belli*, Salmonella, Shigella, Campylobacter, and Giardia. PI in children with AIDS is usually affecting cecum and ascending colon more and patients tend to recover with conservative therapy. Immunodeficiency plays an important role in lymphoid depletion of the intestine and leads to reduced intestinal wall integrity.<sup>8</sup>

The PI gas pattern in patients with AIDS is usually linear and high risk with perforation due to the presence of epithelial necrosis. In Wood's study, 4 of 6 patients had PI linear and 2 of 6 patients had intra and retroperitoneal gas appearances. Additional observations were also made on PI patients in AIDS, that PIs in AIDS were more commonly found in the right colon (80%).<sup>3,9</sup>

The mechanical theory with the presence of mucosal integrity damage coupled with an increase in intraluminal pressure will facilitate dissection gas in the intestinal wall, this theory is very important and explains the association of PI coli and infectious diarrhea, because of damage from the mucosal layer as seen in AIDS patients with opportunistic infections, i.e. CMV infection, where there is an increase in secondary intraluminal pressure from diarrhea with increased peristaltic. In addition to MAC bacterial infection, there is lymphatic obstruction, which causes an increase in intraluminal pressure causing PI. In cryptosporidium and pneumatosis coli infection, it is more

difficult to analyze the underlying cause of PI theory, although cryptosporidium can cause large volume diarrhea and increase intraluminal pressure.<sup>8</sup>

#### Imaging

In both plain radiograph and CT, PI appears radiolucent or hypodense with cystic/bubbly or linear features in the intestinal wall. The PI pattern can be a combination of both linear and bubbly. The circular collection of gases on the intestinal wall may also be a PI pattern. The presence of a mixture of intestinal contents with air or air trapped between the folds of the mucosa may resemble a PI, thus misinterpreting it as PI. During CT scan, image analysis should be performed on the lung window to confirm PI detection, especially in the colon of Figure 4. Because CT is more sensitive than plain radiograph, it can be used to clarify PI detection in ambiguous plain radiograph and can detect potential causes.<sup>7</sup>

The appearance of PI on necrotic intestine may appear as a fixated part and distension locally, with other parts being decompressed. The fixated part of the intestine may be a symptom of impending intestinal perforation, which should be evaluated with a lateral left decubitus radiograph or cross-table.<sup>10</sup>

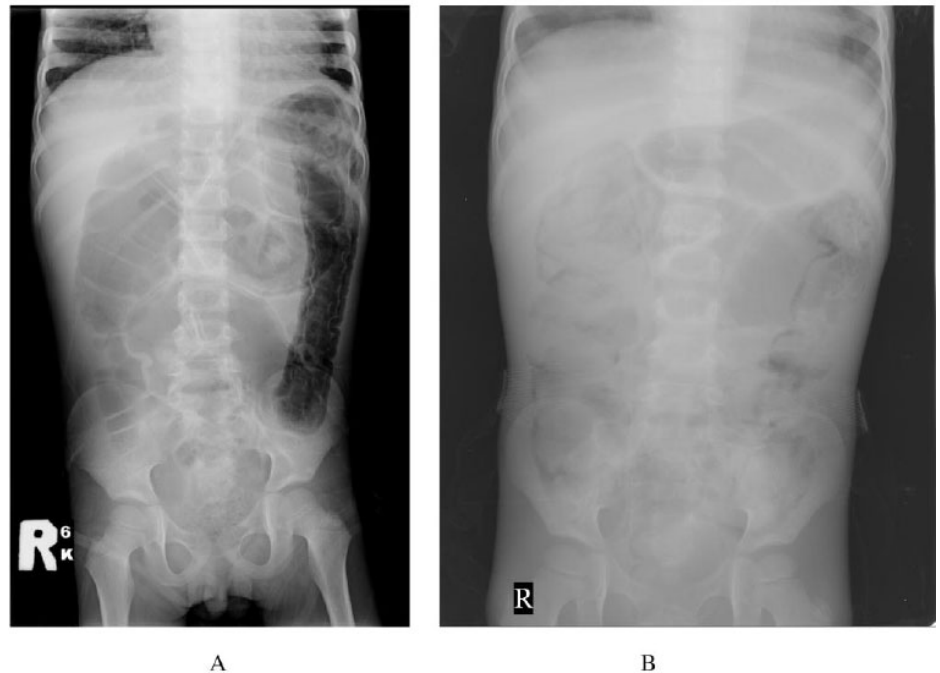
Circular form of PI is usually caused by something simple and non-harmful, often accompanied by pneumatosis cystoides intestinalis (PCI). Both minor and life-threatening diseases can cause linear and bubbly type appearance of PI. Intestinal appearance is usually normal in PI caused by mild disease, but other discovery such as intestinal wall thickening, loss or more well-defined mucosa, intestinal dilatation, occlusion of artery or vein, ascites and portal hepatic gas or mesenteric portal may increase the likelihood of PI caused by life-threatening disease.<sup>11</sup> PIs that are only localized to a specific intestinal region in a specific vascular distribution may be caused by ischemia. Intra or extraperitoneal free air appearance can be seen in PI conditions caused by both simple and life-threatening disease.<sup>12</sup> The correlation of spontaneous pneumoperitoneum and PI is usually due to the presence of serosal and subserosal cysts rupture in the intestinal wall.<sup>7</sup>

Several studies expressed the capability of CT in distinguishing images of early nontransmural mesenteric ischemia from irreversible transmural infarct with full thickness. Linear PI is more common in transmural infarct patients. Other studies have also revealed that the presence of PI with postmesenteric gas is strongly correlated with intestinal transmural infarction, where PI without portomesenteric gases is more frequent under nontransmural ischemic condition.<sup>7,13</sup>

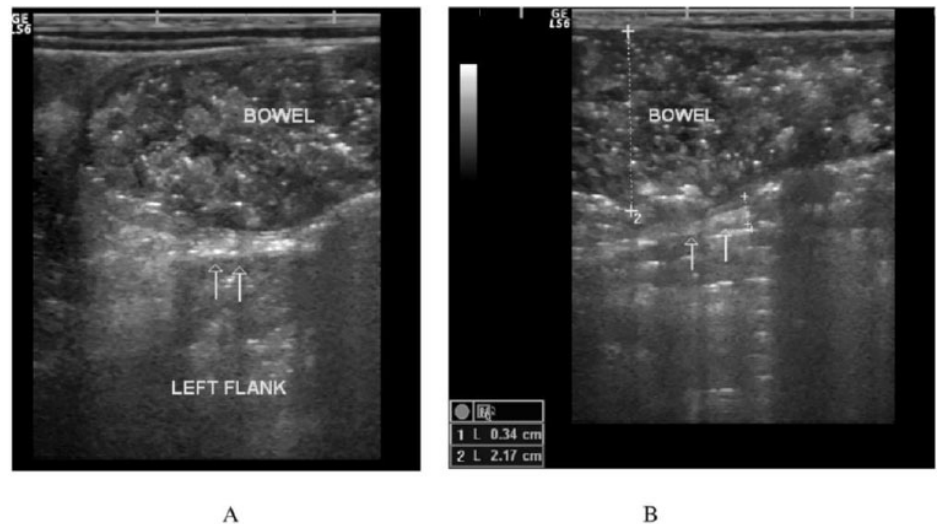
Ultrasound can also be used in detecting PI.<sup>7</sup> The use of ultrasound is prioritized in pediatric patients where radiation can be reduced. PI appearance in ultrasound is usually described as linear and focal echogenic areas within the intestinal wall. It may also appear as a continuous echogenic ring on the intestinal wall.<sup>7,14</sup>

MRI was less chosen as a modality in detecting PI, but there have been several cases reported in the literature that PI appearance in MRI appears as a collection of air close to each other in echo gradient sequence (GRE) due to the presence of blooming artifacts associated with inhomogeneous magnetic space at the air and tissue interface.

CT findings that correlate closely with life-threatening PIs are mild (> 3 cm-6 cm) and large (> 6 cm) intestinal dilatation, as well as intestinal wall thickening (> 0.5 cm).<sup>15</sup> MDCT with intravenous contrast is an alternative diagnostic modality. CT findings on acute intestinal ischemia are 1) diffused thickening of the intestinal wall, usually 8-9 mm, and may occur up to 15 mm, 2) thinning of the intestinal wall may occur in acute arterial occlusion due to loss of intestinal muscular tone and loss of tissue volume by constriction of blood vessels, 3) low attenuation of the intestinal wall due to edema, 4) high attenuation of the intestinal wall due to intramural bleeding, 5) lack of stinging of the intestinal wall is a very specific finding in acute ischemia, 6) PI with intestinal thickening indicates a transmural infarction, 7) dilatation of the intestinal wall with adynamic ileus, 8) mesenteric vessels with emboli or thrombus that fail to sting post-contrast and 9) mesenteric fat strands and ascites are also usually seen.<sup>10</sup>



**Figure 1.** (A) Supine Abdominal X-Ray. Showed Linear and circular PI along the descending colon without any sign of pneumoperitoneum or portal vein gas. (B) AXR 5 days later revealed the migration of PI up to ascending colon.

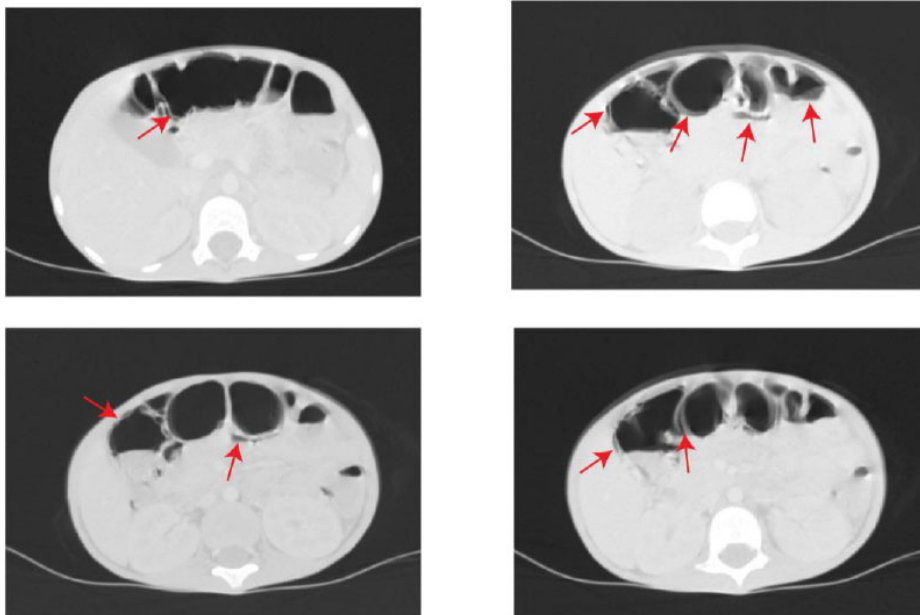


**Figure 2.** Abdominal Ultrasound showed multiple linear foci hyperechoic lesion within the intestinal wall both left (A) and right (B) bowel, with the presence of intraluminal feces in mild dilated colon.

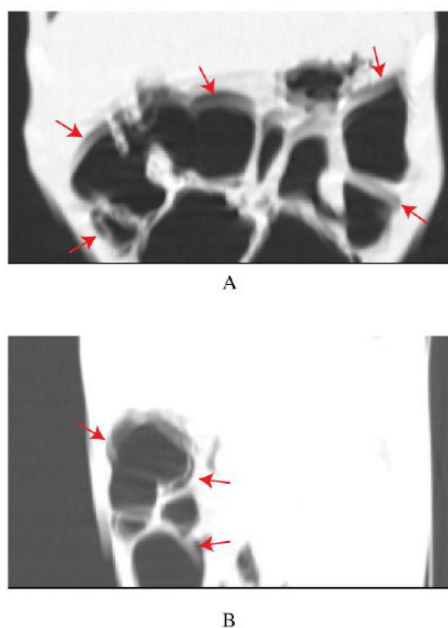
## CASE REPORT

A 7-years-old boy with HIV/AIDS and sepsis presented with abdominal distension, acute abdomen, and malnutrition. His last CD4 cell count was 26 cells/ul (Normal: 380-1704) and CD4% was 3.09% (Normal: 30.13-60.23). A plain abdominal

radiograph was taken and showed linear and circular PI along the descending colon without any signs of portal vein gas or pneumoperitoneum. After about 5 days another plain abdominal X-Ray was taken and demonstrated the PI migration to the ascending colon with appearance of feces in the mild dilated colon. An abdominal



**Figure 3.** Abdominal CT Axial reformat, showed linear and circular hypodense lesion within ascending-transverse-descending colon with air density, which resembles intramural gas (arrow).



**Figure 4.** Abdominal CT(A) Coronal, B) Sagittal. Circular linear intramural gas (arrow).

ultrasound was done which further confirm the presence of intramural gas in the colonic wall through the appearance of multiple linear foci within the intestinal wall both right and left bowel. Abdominal CT was also confirmed the presence of

hypodense lesion with air density within the intestinal wall of both sides colon in linear and circular pattern. No red flag signs, such as pneumoperitoneum, portal vein gas and porto-mesenteric gas, were observed from all of his imaging.

Multiple cystic lesions were demonstrated both on his chest X-ray and Chest CT-scan hence cystic bronchiectasis conclusion was made. Unfortunately the exact underlying disease of his cystic bronchiectasis was not revealed due to his unfavorable condition to perform any other study.

## DISCUSSION

PI in AIDS patient usually has linear and cystic form which appear on this case, although often the distribution of PI in AIDS patients often in the right colon but it can be present first in the left side of colon. PI in the right side of colon correlates with extraluminal gas, which indicates the life threatening conditions. The first appearance of PI in this case was on the left side but after 5 days it migrates to the right side, which explains the patient worsening condition even though none of the red flag imaging signs were detected,

The patient underlying cause of PI

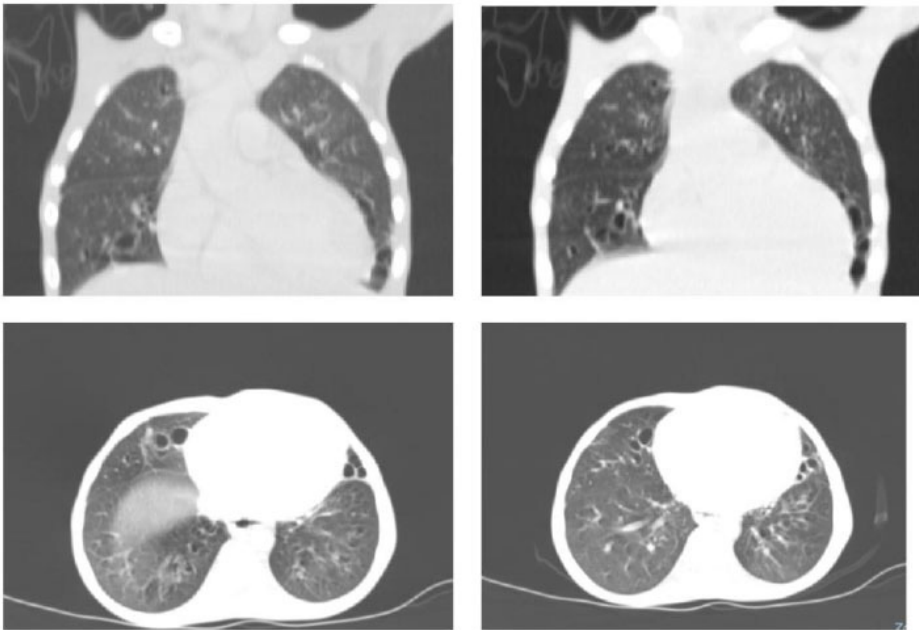
was likely to be AIDS related and there are two out of four underlying etiologies of PI on this case, which were due to the increase mucosal permeability and cystic lung conditions. Mucosal disruption is the primary cause of almost all PI cases due to inflammation and ischemia moreover in AIDS patient, in which bacteria or gases can forcefully enter the intestinal wall.

There is increased of frequency pyogenic and opportunistic pulmonary infection in AIDS patient could result in bronchiectasis complication. Bronchiectasis may cause alveolar disruption and air dissection of pulmonary interstitium to the intestinal wall, therefore explains the presence of intramural gas.<sup>5,6</sup>

The importance of knowing the presence of intramural gas is the evaluation of life threatening signs on imaging results, portal vein gas, pneumoperitoneum, and porto-mesenteric gas, to determine the need of surgery. Exploratory surgery needs to be done in the existence of these factors, while in the absence of such findings, the patient can be given antibiotic therapy, diet, oxygen therapy to relieve symptoms. Antibiotics are a fixed regimen of PI, metronidazole is a successful regimen of PI with metronidazole 500 mg 3 times daily for up to 3 months. Theoretically, antibiotics can reduce the amount of gas produced by bacteria and relieve symptoms. Oxygen inhalation is also believed to relieve symptoms by acting as a toxin against anaerobic bacteria in the intestines.<sup>15</sup>

## CONCLUSION

There are many underlying causes of both mild and life-threatening PIs. Clinical correlation, physical examination, and other supportive examinations are also indicators of the underlying cause of PI. PIs can be caused by various sources of both gastrointestinal and pulmonary. PIs resulting from immunodeficiency, especially AIDS, are often found with linear and dominant patterns on right-side colon, whereas cystic or PCI is usually based on benign causes. PIs in AIDS patients are mostly caused by opportunistic infection, and surgical therapy is not an option and should be avoided as much as possible. The most sensitive and specific modality for diagnosing PI is MDCT with



**Figure 5.** Chest CT, Axial and Coronal reformat. Cystic bronchiectasis in both lung bases.

contrast, whereas 22-71% PIs may appear in plain radiographs. Ultrasound is a non-radiation modality that can be used for pediatric patients, and MRI with a GRE sequence in certain cases can show PI. The presence of other features besides PI such as portal venous gas, ascites, and portomesenteric gas leads to intestinal ischemia.

### CONFLICT OF INTEREST

All authors declared that there is no conflict of interest regarding this publication.

### ETHICS APPROVAL

The patient had given inform consent and agreed in opening the necessary data to the authors and this case-report also had been approved by Radiology Department, Faculty of Medicine Universitas Udayana - Sanglah General Hospital Bali, Indonesia

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### AUTHORS CONTRIBUTION

All authors contributed equally in the writing process of this article.

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