

Pneumatosis Coli Secondary to Eosinophilic Colitis: Report of a Refractory Case

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Abstract

Introduction: Pneumatosis intestinalis (PI) is a broad term that describes the presence of gas in the bowel walls. The colon is affected in 36% of cases. Eosinophilic colitis (EC) is a rare form of colitis that is characterized mainly by eosinophilia in blood and affected tissues. Although eosinophilic colitis responds well to conservative treatment, yet the association of PI could be an indication of emergent surgery.

Case Presentation: A 48-year-old female patient with a history of recurrent attacks of abdominal pain and distention that did not respond to conservative treatment, was hospitalized and investigated thoroughly. The result of abdominal computed tomography (CT) scan indicated the presence of pneumatosis coli. Based on the findings of the CT scan, colonoscopy was done revealing hyperemic and hypertrophied mucosa at the hepatic flexure, from which multiple punch biopsies were taken for histopathological assessment. The pathological examination of these biopsies detected a diffuse active colitis of moderate severity with excess eosinophils (10 - 15/HPF), which is consistent with the diagnosis of EC. The patient improved dramatically on fluid therapy and antibiotics with complete resolution of pneumatosis coli in the follow up CT scan. However, the patient developed a severe relapse of symptoms once oral intake was resumed and this time the conservative treatment failed to improve the condition. Surgery was indicated and laparoscopic right hemicolectomy with primary anastomosis and covering ileostomy was done. The patient was free of symptoms after the operation and her postoperative course was uneventful with no complications encountered.

Conclusions: Eosinophilic colitis usually has a good prognosis, however its association with pneumatosis coli could reflect a serious damage to the colonic mucosa. Although eosinophilic colitis responds well to medical treatment, yet in certain cases where clinical signs are evident and pneumatosis coli is present, surgical intervention is the treatment of choice.

Keywords: Pneumatosis, Coli, Eosinophilic, Colitis

1. Introduction

Eosinophilic colitis (EC) is a specific type of colitis that occurs in healthy infants with acute self-limited bloody diarrhea or in young adults as chronic relapsing colitis. The pathophysiology of EC is explained by altered hypersensitivity, as food allergy in infants and T lymphocyte-mediated reaction in young adults. Treatment of EC includes a dietary protocol to avoid food hypersensitivities, corticosteroids, antihistamines, leukotrienes receptors antagonists, and recently monoclonal antibodies against IgG1K (omalizumab) (1).

Pneumatosis intestinalis (PI) is a broad term that describes the presence of gas in the bowel walls extraluminally. This condition commonly affects the small bowel in about 42% of cases, followed by the large bowel in 36% of cases, whereas both are affected in the remaining 22%. Pneumatosis coli is a term used exclusively to describe the presence of gas in the wall of the colon (2).

The gas-filled cysts of PI are usually located on the antimesenteric border within the submucosal layer or the muscularis propria of the bowel (3). Regarding the etiology of PI, it can occur as a primary disease or secondary to multiple conditions. Primary cases, which represent only 15% of all cases, are considered to have an autonomous disease (4). Secondary cases, which are more common, are caused by damage to the intestinal mucosal that is induced by several traumatic, inflammatory and infectious etiologies and collagen diseases (5).

Sources of the mural gas include the intraluminal gas of the gastrointestinal tract, gas of bacterial fermentation and pulmonary gas. The mechanism of gas entry into the bowel wall is still controversial as it is not fully clear if it is due to increased intraluminal pressure, injury to intestinal mucosa or even a combination of these factors (6).

Symptoms of PI range from mild symptoms including abdominal pain and distention, change in the bowel habits and weight loss to serious symptoms such as bleed-

ing, acute abdominal pain and ileus, which reflect life threatening complications including intestinal ischemia, perforation and secondary peritonitis. Pneumatosis intestinalis can be an asymptomatic condition in some cases (2).

Computer tomography (CT) scan is the corner stone in the diagnosis of PI with more diagnostic accuracy than plain films or ultrasound (6). Management of PI is essentially conservative including antibiotics, fluid therapy and aggressive oxygen therapy (7). However, surgery is indicated in patients with symptoms and signs of peritonitis and patients with metabolic acidosis, which is a sign of bowel ischemia (8).

2. Case Presentation

This report describes a 48-year-old female, who had a complaint of recurrent attacks of colicky pain that she felt diffusely all over her abdomen associated with abdominal distention for two months. The patient sought medical advice and she was preliminarily diagnosed with colitis and was managed conservatively by antispasmodics and bulking agents.

After three months of conservative treatment, the patient did not notice any improvement in her condition, thus she was asked for some investigations on a follow up visit. These investigations included series of laboratory tests such as blood picture, liver functions tests, serum creatinine level and Widal and Brucella tests, yet no abnormalities were detected in all of these tests.

Radiological investigations such as gastrografen enema and computed tomography (CT) scan of the abdomen and pelvis were done for the patient. While the gastrografen enema was perfectly normal, the CT scan revealed a dilated ascending colon and hepatic flexure with mural gas bubbles (Figure 1) that was suggestive of pneumatosis coli.

Based on the findings of the CT scan, the patient was prepared for colonoscopy, which revealed hyperemic and hypertrophied mucosa at the hepatic flexure, from which multiple punch biopsies were taken for histopathological assessment. The pathological examination of these biopsies detected a diffuse active colitis of moderate severity with excess eosinophils (10 - 15/HPF), which agree with the diagnosis of EC.

The patient was then hospitalized and managed conservatively with intravenous (IV) fluid therapy, antispasmodics, and antibiotics in the form of IV levofloxacin 200 mg every 12 hours and IV metronidazole 500 mg every eight hours. After 72 hours, the patient showed a marked clinical improvement with complete relief of the abdominal pain. The improvement was not only clinical, but

also radiological improvement was also noted in the follow up CT scan done on the 5th day of medical treatment, which showed marvelous improvement with complete disappearance of the previously described pneumatosis coli (Figure 2).

As the patient improved, oral fluids intake was resumed seven days after starting the medical treatment. Two days later, the patient developed recurrent bouts of severe abdominal pain with diffuse tenderness all over the abdomen. The oral intake was ceased and conservative treatment was resumed. As the patient failed to respond to the second course of conservative treatment and the abdominal signs became prominent, the decision of surgical intervention was made. Laparoscopic assessment of the entire abdomen followed by right hemicolectomy with ileo-tranverse anastomosis was done. A covering ileostomy was also performed, as the colon was not well prepared.

The postoperative course was uneventful, the patient started taking oral fluids once the ileostomy was functioning (the second postoperative day) and the abdominal drain was removed on the third postoperative day. No morbidities were encountered during the patient's convalescence period.

The postoperative histopathological examination of the resected specimen reported thinning of the mucosal lining of the colon, submucosal air-field space and an inflammatory reaction with excess eosinophils, giving a clear diagnosis of pneumatosis coli secondary to EC.

The patient was invited to visit the outpatient clinic every week for one month, then every month for the next three months. During each visit the patient reported marked improvement of her condition without recurrence of the previous attacks of pain.

An informed consent was obtained from the patient regarding the publication of her condition.

3. Discussion

The EC is characterized by dense infiltration of colonic wall with eosinophils, which can be a primary disease or secondary to parasitic infestations, vasculitis, and drugs such as carbamazepine, naproxen and tacrolimus (1). The clinical picture depends on the layer that is most affected by the eosinophilic infiltration; if it is the mucosa then patients present malabsorption, diarrhea, and protein-losing enteropathy, in serosal involvement eosinophilic predominant ascites is the key clinical feature, (9) and in transmural affection, colonic wall thickening and intestinal obstruction occurs (10). Diagnosis of EC is done by measuring serum IgE levels, skin prick tests to identify specific allergens, endoscopy and biopsy, which typically shows sheets

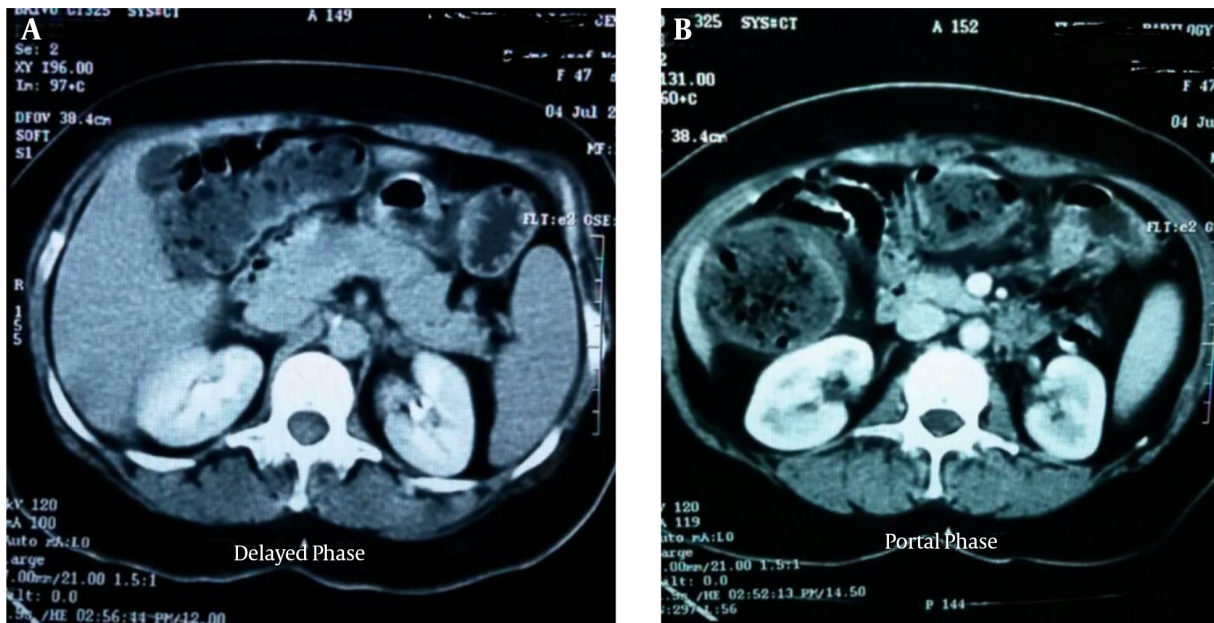


Figure 1. Computerized Tomography Scan of the Abdomen Showing the Dilated Right Side of the Colon With Mural Gas Bubbles

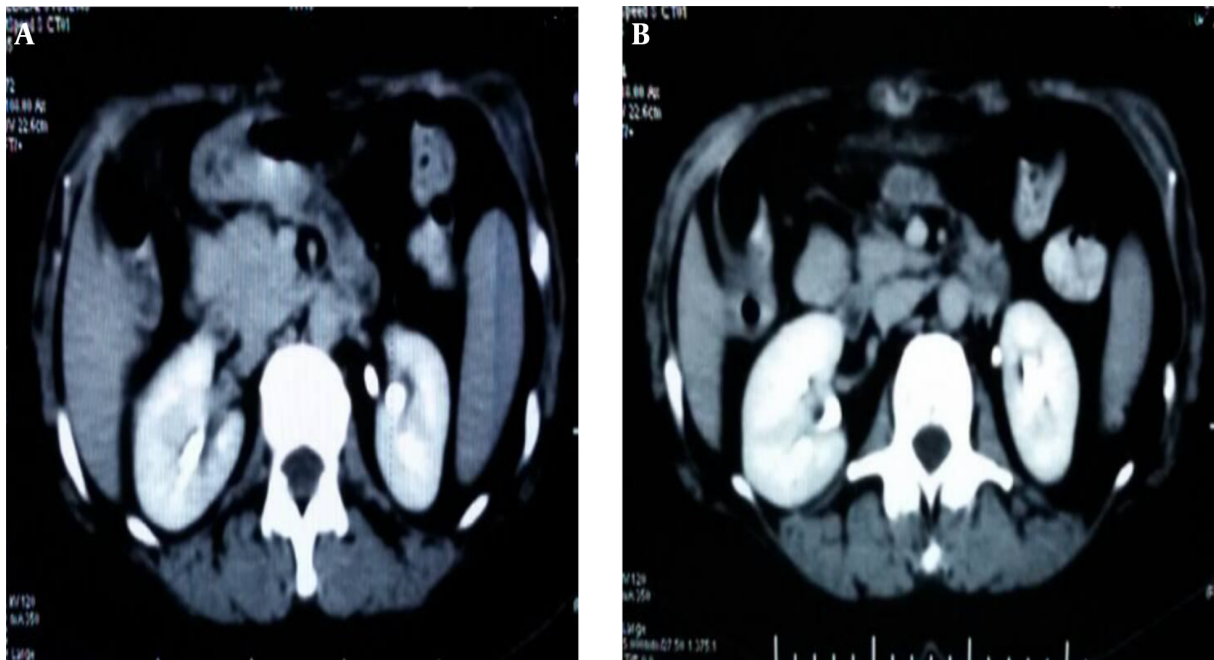


Figure 2. Follow up Computerized Tomography Scan After Starting Medical Treatment Showing Disappearance of the Mural Gas Bubbles

of eosinophils infiltrating the lamina propria extending into the sub-mucosa (1).

Pneumatosis intestinalis is a clinical condition defined as the extra-luminal presence of gas in the bowel wall,

whether the small or large bowel. It is considered an ominous radiological finding rather than a disease per say (11).

The pathophysiology of PI is still controversial. Sources of extra luminal gas and mechanism of its entry are debat-

able, yet the main event is the breakdown of the mucosal barrier of the intestine accompanied by increased intraluminal pressure (12). The insult of the intestinal mucosal barrier is usually related to variable causes, however in almost 15% of cases it can be an idiopathic phenomenon (4).

Pneumatosis Intestinalis can be secondary to mechanical factors such as endoscopy and carcinoma; inflammatory conditions such as necrotizing enterocolitis, Cohn's disease and ulcerative colitis; autoimmune conditions such as scleroderma; infections such as *Clostridium difficile*, HIV and cytomegalovirus; pulmonary causes such as chronic obstructive pulmonary disease and asthma; cytotoxic drugs and immunosuppression (2).

The diagnosis of PI depends mainly on abdominal CT scan, which is the most frequently used modality for diagnosis. Computerized-tomography scan is superior to abdominal radiography as it is more sensitive at detecting not only PI but also portal and porto-mesenteric venous gas, which may indicate the presence of serious causes of PI (13). In CT imaging, PI usually has a linear pattern or a bubbly pattern of gas of low-density inside the bowel wall; intramural circular collections of gas can also exist (14). Sonography is also used to detect PI mainly in pediatric patients to avoid the risk of ionizing radiation that appears on sonography as linear or focal echogenic areas within the bowel wall (15). Magnetic Resonance Imaging can be sometimes used in the diagnosis of PI revealing a circumferential collection of air inside the bowel more clearly on gradient echo images (16).

Not every patient with PI requires an emergent surgery. Asymptomatic patients or those with mild symptoms are usually managed conservatively with no need of specific therapy, whereas patients with serious clinical presentation such as acute abdominal pain or bleeding are candidates for surgical intervention as their symptoms suggest intestinal mucosal injury.

While some authors (2, 17) tend to consider PI a medical condition that should be treated conservatively, the patient in our report showed initial response to the medical treatment then the symptoms relapsed after resumption of oral intake with no response to medical therapy necessitating surgical intervention.

The patient initially presented abdominal colic that recurred regularly, especially after meals, associated with abdominal distention; this presentation was suggestive of colitis, yet the failure to respond to ordinary medications used in treatment of colitis and Irritable Bowel Syndrome (IBS) drew our attention to the possibility of an underlying specific pathology. The aforementioned investigations concluded the presence of EC.

In the patient of our report the right side of the colon was only affected, which agrees with the fact that

eosinophilic colitis usually involves the proximal colon (18). The infiltration of mucosa by eosinophils and the toxicity of eosinophil granule proteins to the tissues are responsible for the mucosal damage inflicted by eosinophilic colitis. This mucosal insult associated with increased intra-luminal pressure during colonic contractions initiated the sequence of PI in the patient of this report (19).

In a review of 25 cases of pneumatosis coli over 30 years, colitis was responsible for only 12% of cases and colectomy was only done in two patients (20). This review reported a high recurrence rate that ranged between 50% - 78% similar to the patient in our report, who relapsed within a week, however the review stated that in 96% of cases the left side of the colon was affected, whereas the right side was affected in our patient.

The initial clinical and radiological improvement of the patient's condition with conservative management can explain that the extent of mucosal damage induced by colitis was not remarkable; however, starting oral feeding at an early point caused exacerbation of the inflammatory process and recurrence of PI in a more aggressive pattern. As the medical treatment failed to achieve any improvement and as the abdominal signs became more prominent, surgical intervention was deemed the decision of choice. The reason we decided surgical intervention was that we decided prolonged medical treatment will not improve the condition as the abdominal signs became more prominent making us suspect an impending colonic perforation, that is why corticosteroids were not a part of the treatment plan, fearing it might induce colonic perforation as the presence of PI indicated a transmural affection of the colon rather than a simple mucosal injury.

We preferred to start with laparoscopic exploration of the abdomen to assess the extent of the problem and to proceed afterwards. The right side of the colon appeared dilated and pathologic, thus right hemicolectomy was performed with a covering ileostomy. The patient recovered from the surgery quickly and smoothly with no postoperative morbidity.

Eosinophilic colitis is a rare entity that has a good prognosis, however its association with pneumatosis coli could reflect a serious damage to the colonic mucosa, which can be correlated with the clinical findings. Although EC responds well to medical treatment, yet in certain cases where clinical signs are evident and pneumatosis coli is present, surgical intervention is the treatment of choice.

Footnote

Authors' Contribution: Hossam Elfeki performed the data acquisition; Sameh Emile conducted the analysis and

interpretation of the data and drafting of the manuscript; Hosam Elbanna conducted the critical revision of the manuscript for important intellectual content.

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