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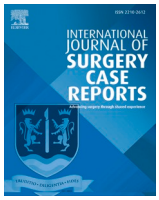

Case report

Extensive inflammatory adhesion of small bowel with massive Pneumatosis Intestinalis in a patient with gastric outlet obstruction: A rare case report

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ABSTRACT

Introduction: Pneumatosis Intestinalis (PI) is a rare disease, majority of which are self-limited processes, in which the intestinal sub mucosa and sub serosa are filled with gas-filled cysts. The exact cause and pathogenesis is not well known yet but there are different theories. The two well accepted fundamental pathogenesis is: mechanical and bacterial.

Case presentation: Here we report a case of a 25 years old patient presented with history of persistent vomiting, intermittent abdominal cramp and significant weight loss over three months. The primary diagnosis was made as gastric outlet obstruction with concomitant small bowel extensive PI.

Discussion: Primary PI has no known cause while secondary type has proposed underlying pathologies with different theorized pathogenesis. The current case report has an underlying pathology of long standing peptic ulcer disease with recent diagnosis of gastric outlet obstruction in favor of the mechanical theory.

PI has a broad spectrum of clinical symptoms; ranges from asymptomatic patients to non-specific gastrointestinal symptoms like diarrhea, abdominal distention, weight loss, bloody or mucous stool. Patients with underlying pyloric stenosis, peptic ulcer disease presents with more of upper GI symptoms.

Conservative management is usually the treatment of choice. However, surgery must be considered if peritoneal irritation or bowel obstruction appears overt.

Conclusion: Concomitant occurrence of gastric outlet obstruction with small bowel PI is not uncommon disease but severe and extensive inflammatory adhesion was rarely reported. Therefore surgical intervention is mandated for the former or both depending the severity of the PI.

1. Introduction

Pneumatosis Intestinalis (PI) is characterized by gas-filled cysts in the intestinal sub mucosa and sub serosa [1]. The exact cause is not known but there are two main hypotheses regarding the fundamental pathogenesis of PI: mechanical and bacterial [2]. Both highlighted as mechanisms for the loss of integrity of the intestinal mucosal layer, resulting in leakage and formation of gas within the intestinal wall [3,4].

PI has been descriptively divided into primary (idiopathic) and secondary pneumatosis, with approximately 85 % of cases considered being of secondary nature. Primary PI typically consists of colonic cystic collections of air and can be identified either radiologically or pathologically. Secondary PI has been attributed to a host of clinical diseases with a wide range of severity and is identified by linear or circumferential air in any part of the gastrointestinal (GI) tract [4,5]. Secondary PI has been reported in association with gastrointestinal conditions including inflammatory bowel disease, diverticular disease, ischemic enterocolitis, pseudomembranous colitis, Hirschsprung's disease, sigmoid volvulus, pyloric stenosis, surgical anastomoses, and non-gastrointestinal conditions such as collagen vascular diseases, chronic obstructive pulmonary disease, asthma, and cystic fibrosis [6].

Good results can be achieved in most cases by conservative means, but surgical treatment may be necessary in some cases. Surgery should be avoided unless there are signs of severe inflammation, metabolic

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acidosis or portal venous gas, which are indicators of more serious diseases [7]. Here we report a case of 25 year old patient presented with long standing symptoms of Gastric outlet obstruction with current exacerbation. This case report was organized in line with SCARE checklist [8].

2. Presentation of the case

A 25 years old male patient come from a rural area of Jarso district of East Hararge Zone which is located at an altitude of 2076 m above sea level with a long standing history of dyspepsia and current exacerbation with persistent vomiting of ingested matter for more than two weeks. He had history of intermittent abdominal cramp, poor appetite and significant weight loss. He had no history of known chronic illnesses. He had no history of surgery. No identified history of drug allergy or any immune suppressing drug use.

Physical examination revealed generally wasted muscles. Vital signs: BP 105/77 PR 101 RR 20. There was decreased air entry over right lower chest. Cardiovascular system examination was unremarkable. Abdomen looks Scaphoid, no mass or tenderness appreciated. Succussion splash test is positive. Laboratory and imaging works: WBC 7.37 $\times$ 10^3/µl, N 64 % Hgb 14.8 g/dl Platelet count 490 $\times$ 10^6/μl, AST 41 IU/l, ALT 35 IU/l, Total bilirubin 0.542 mg/dl, Direct Bilirubin 0.179 mg/dl, serum Creatinine 0.6 mg/dl, serum electrolytes were normal. Abdominal Ultrasonography scan revealed dilated stomach filled with thick debris. CT scan of the abdomen showed signs suggestive of Gastric outlet obstruction and air filled multiple cystic lesions located between the dome of right diaphragm and liver (Fig. 1).

After proper preoperative preparations done, the patient was taken to operation room and laparotomy was done under general anesthesia. Intraoperative finding showed hugely dilated stomach with thickening and narrowing of gastro-duodenal junction. There were extensive, 0.5 cm–3 cm sized air filled cysts along the serosa and mesentery of distal small bowel sandwiched between the right dome of diaphragm and liver. There was dense inflammatory adhesion causing severe narrowing of the involved bowel lumen (Fig. 2). There was no other gross pathology seen in abdominal cavity. The operation was performed by consultant General surgeon assisted by Surgery residents at a tertiary teaching hospital.

Gastro-jejunal bypass along with truncal vagotomy and Braun's anastomosis was performed for the gastric outlet obstruction and resection of the significantly stenosed and distorted 100 cm of distal ileum 6 cm from ileocaecal junction was done with hand sewn ileo-ileal anastomosis. The excised tissue was sent for histopathological examination which has confirmed the diagnosis of Pneumatosis Intestinalis (Fig. 3). The patient had spent smooth postoperative course and discharged on 5th postoperative day. He had follow-up at two weeks, two months and six months after surgery with no symptom of recurrence and significant weight gain.

3. Discussion

Pneumatosis Intestinalis is an uncommon disease without clear pathogenesis, characterized by air filled cysts within the submucosa or subserosa of intestine [9,10]. It is categorized as primary or secondary with 15 % and 85 % rate of occurrence respectively. Primary PI, also known as Pneumatosis Cystoides Intestinalis has no known underlying causes while the secondary type has proposed underlying pathologies with different theorized pathogenesis [11]. The current case report has an underlying pathology of long standing peptic ulcer disease with recent diagnosis of gastric out let obstruction which makes it a secondary type of PI. It is favored by *the mucosal disruption theory*, that attempts to explain the presence of gas by a mechanical breakdown, which includes PI secondary to obstruction, peptic ulcer disease, endoscopy,

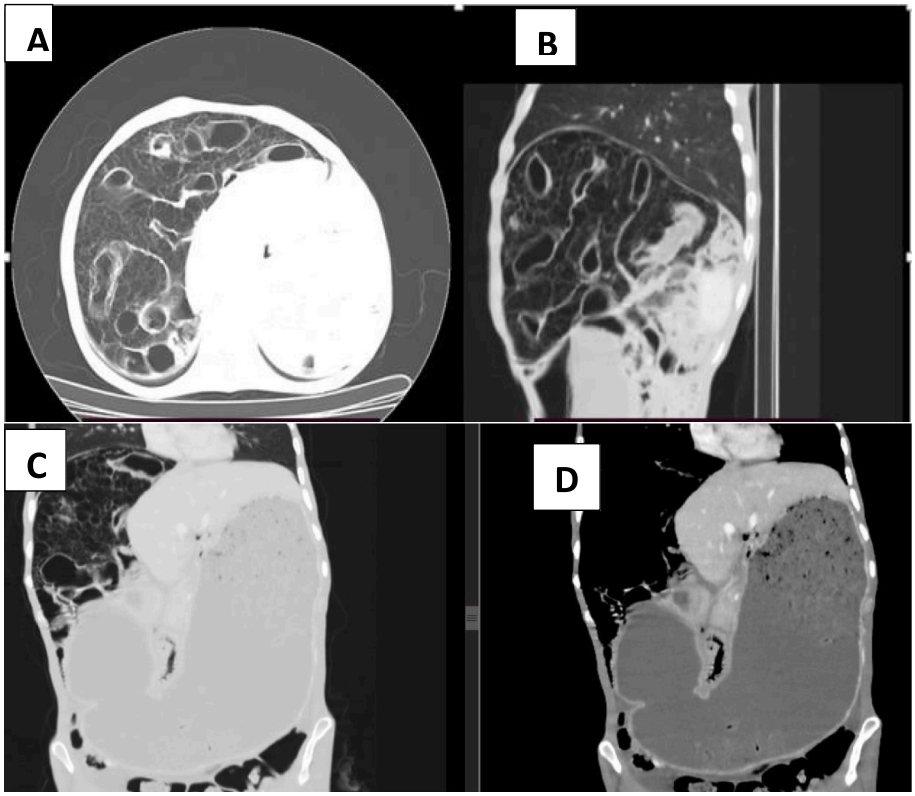

Fig. 1. CT scan of a patient presented with gastric outlet obstruction with concomitant extensive PI of small bowel. A &B axial and sagittal view showing extensive cystic area on the serosa of small bowel and its mesentery; C&D Coronal view of abdomen showing enormously distended stomach with massive cystic air filled cavities sandwiched between liver and diaphragm.

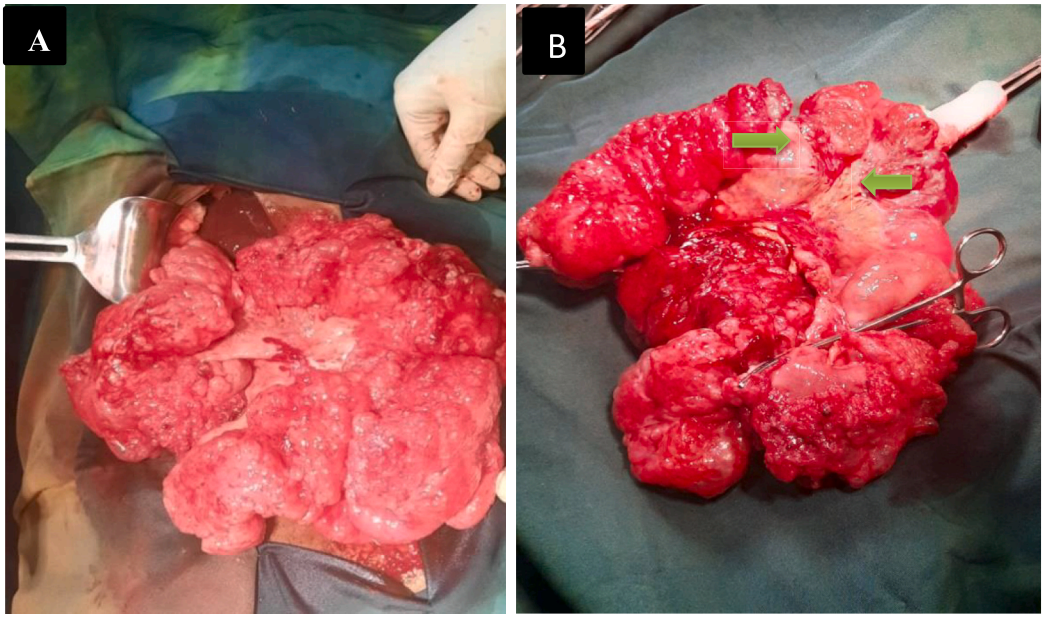

Fig. 2. A. Intra operative view of extensive cystic and bubbles of air over the serosa of distal 100 cm of ileum after delivered from right sub diaphragmatic space. B. A resected out sample with arrow showing areas of significant inflammatory adhesion and distortion.

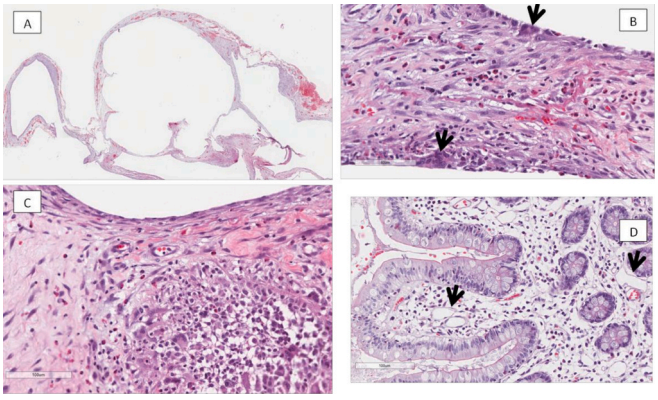

Fig. 3. A. Empty cystic spaces without epithelial lining B. Higher magnification of air filled cystic spaces with rare giant cells (arrows) and mixed cells stromal inflammation C. Foci of granulomatous reaction D. Dilated empty spaces in the lamina properia.

inflammatory bowel disease, Hirschsprung's disease, blunt trauma, surgery, and amyloidosis [11]. The bacterial theory, explains that air is produced in the bowel wall after air-producing bacteria enter into the bowel wall [12]. Chemical and nutritional deficiency theory, states that fermentation increases bowel gas. The other theory which states increased bowel mucosal permeability as a result of immunosuppressive drugs, hormones, collagen vascular disease may lead to PI were also stated [13].

In 1979 Jamart had reviewed 919 cases of the world literature on pneumotosis cystoides intestinalis and revealed the subserous cysts were more frequently found in the small bowel while the submucous localizations were predominant in the colonic wall [10]. The cysts can appear anywhere within the gastrointestinal tract, (large intestine 46 %, small intestine 27 %, stomach 5 %, mixed 7 %), nonetheless, Small bowel PI are usually located in the terminal ileum and rarely found in places like the mesentery [9,14]. The above two thoughts are supportive of our findings in which subserosal location in small bowel PI and terminal ileum were commonly affected. The cysts also had extension to the mesentery of the involved small bowel.

PI has a broad spectrum of clinical symptoms, from asymptomatic patients to non-specific gastrointestinal symptoms like diarrhea, abdominal distention, weight loss, bloody or mucous stool [9,14]. Although these symptoms are nonspecific, they parallel with the site of involvement and associated underlying diseases. Patients with underlying pyloric stenosis, peptic ulcer disease present with more of upper GI symptoms while PI involving different parts of colon manifests with lower GI symptoms like diarrhea, bloody stool and pronounced abdominal distension. Our patient had presented with these upper GI symptoms predominately sign of chronic dyspepsia and later with gastric outlet obstruction accompanied with significant weight loss. Similar case was reported by Tilahun et al. from Jimma University [15].

The laboratory examination and pathological biopsy of PI are nonspecific; the diagnosis mainly depends on colonoscopy, CT, radiography, and ultrasound findings. For the upper gastrointestinal tract, CT scan has paramount importance as endoscopy might not help unlike the colonic type of PI [5].

About 3 % of the patients with PI can present with pneumoperitoneum, volvulus, intestinal obstruction or ischemia and perforation, which may alter the usual therapy course [16,17]. In our case, the definitive diagnosis and decision was made intraoperatively after visual inspection of the extensive cysts of varies size sandwiched between the liver and diaphragm with inflammatory adhesion significantly compromising the small bowel lumen.

Conservative management is usually the treatment of choice. Oxygen and antibiotics are often sufficient with 70 % of remission rate; nonetheless, it is difficult to diagnose with certainty those who should be conservatively managed from those who need surgery. This might compel the managing team to perform unnecessary surgical procedures due to the fear of complications. However, surgery must be considered without hesitation if signs of peritoneal irritation or bowel obstruction become overt [14]. In our patient, as the main complaint was gastric outlet obstruction, surgery was decided without confusion or delay. But the decision for resection and end to end anastomosis of the segment of small bowel was made due to the extensive inflammatory adhesion and distortion of the bowel.

4. Conclusion

Concomitant occurrence of gastric outlet obstruction with small

bowel PI is not uncommon disease but severe and extensive inflammatory adhesion was rarely reported. The correct diagnosis should be made preoperatively to minimize unnecessary operations especially if there are no associated complications like obstruction, peritonitis or bowel ischemia. However, surgical intervention is mandated for the former or both depending on the severity of the PI associated pathology.

Patient consent

Written, signed and informed consent was obtained from the patient for publication of this case report and accompanying images anonymously. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

Case reports are exempted by Institutional Health Research Ethical Review Committee of Haramaya University.

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Author contribution

BB Conceptualization, acquired the data, and written the original draft of the case report **BF** Data curation, writing up of the manuscript **DWT** Data curation, editing the manuscript **BA** Data curation, edited the final manuscript **AA** Data curation, Methodology**, AT** Data curation and edited the final manuscript **AG** Methodology, writing up of the draft, Methodology. All the authors read and approved the final manuscript for publication.

Guarantor

BB.

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Not Applicable.

Conflict of interest statement

There is no conflict of interest to declare.

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References

[1] Y.J. Wang, Y.M. Wang, Y.M. Zheng, H.Q. Jiang, J. Zhang, Pneumatosis cystoides intestinalis: six case reports and a review of the literature, BMC Gastroenterol. 18 (1) (2018) 100 (2018/06/28), https://doi.org/10.1186/s12876-018-0794-y.
[2] Z. Arikanoglu, et al., "Pneumatosis cystoides intestinalis: a single center experience," (in eng), World J. Gastroenterol. 18 (5) (Feb 7 2012) 453–457, https://doi.org/10.3748/wjg.v18.i5.453.
[3] S.D.S. Peter, M.A. Abbas, K.A. Kelly, The spectrum of pneumatosis intestinalis, Arch. Surg. 138 (1) (2003) 68–75.
[4] A.J. Greenstein, et al., Pneumatosis intestinalis in adults: management, surgical indications, and risk factors for mortality, J. Gastrointest. Surg. 11 (10) (2007) 1268–1274 (2007/10/01/), https://doi.org/10.1007/s11605-007-0241-9.
[5] F. Ling, D. Guo, L. Zhu, "Pneumatosis cystoides intestinalis: a case report and literature review," (in eng), BMC Gastroenterol. 19 (1) (Nov 6 2019) 176, https://doi.org/10.1186/s12876-019-1087-9.
[6] O. Shaheen, W. Ahmad, N.A. Mhammad, Gastric outlet obstruction and sigmoid volvulus in a patient with Pneumatosis intestinalis: an etiology or a complication, Case Rep. Surg. 2019 (2019) 4065749, 2019/07/10, https://doi.org/10.1155/2019/4065749.
[7] A. Saber, Pneumatosis intestinalis with complete remission: a case report, Cases J. 2 (2009) 1–4.
[8] C. Sohrabi, G. Mathew, N. Maria, A. Kerwan, T. Franchi, R.A. Agha, "The SCARE 2023 guideline: updating consensus surgical CAse REport (SCARE) guidelines," (in eng), Int. J. Surg. 109 (5) (May 1 2023) 1136–1140, https://doi.org/10.1097/js9.0000000000000373.
[9] L.L. Wu, Y.S. Yang, Y. Dou, Q.S. Liu, "A systematic analysis of pneumatosis cystoids intestinalis," (in eng), World J. Gastroenterol. 19 (30) (Aug 14 2013) 4973–4978, https://doi.org/10.3748/wjg.v19.i30.4973.
[10] J. Jamart, Pneumatosis cystoides intestinalis. A statistical study of 919 cases (in eng), Acta Hepatogastroenterol. (Stuttg.) 26 (5) (Oct 1979) 419–422.
[11] S. Bower, B.B. Simmons, Abdominal pain from pneumatosis intestinalis, Clin. Geriatr. 19 (5) (2011) 61–64.
[12] J. Gillon, K. Tadesse, R.F. Logan, S. Holt, W. Sircus, Breath hydrogen in pneumatosis cystoides intestinalis, Gut 20 (11) (1979) 1008, https://doi.org/10.1136/gut.20.11.1008.
[13] B.L. Pear, "Pneumatosis intestinalis: a review," (in eng), Radiology 207 (1) (Apr 1998) 13–19, https://doi.org/10.1148/radiology.207.1.9530294.
[14] F.X. Moyon, et al., Pneumoperitoneum and Pneumatosis cystoides intestinalis, a dangerous mixture. A case report (in eng), Int. J. Surg. Case Rep. 74 (2020) 222–225, https://doi.org/10.1016/j.ijscr.2020.07.086.
[15] T.H. Nureta, T.G. Moges, D.M. Abebe, Pneumatosis cystoides intestinalis associated with gastric outlet obstruction; a case report, Int. J. Surg. Case Rep. 111 (2023) 108828 (2023/10/01/), https://doi.org/10.1016/j.ijscr.2023.108828.
[16] S. Ko, S.S. Hong, J. Hwang, H.-J. Kim, Y.-W. Chang, E. Lee, Benign versus life-threatening causes of pneumatosis intestinalis: differentiating CT features, Rev. Assoc. Med. Bras. 64 (2018) 543–548.
[17] A. Beyene, B. Bayissa, M. Atalele, H. Umer, A. Alemu, A. Semahegn, Pneumatosis cystoids intestinalis in a patient with small bowel obstruction: a case report, J. Surg. Case Rep. 2023 (11) (2023) rjad612. England: © Crown copyright 2023.