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CASE REPORT

PNEUMATOSIS CYSTOIDES INTESTINALIS RELATED TO GASTRIC OUTLET OBSTRUCTION: A CASE REPORT

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A b s t r a c t: Pneumatosis remains a rare condition presenting with multiple gas filled cysts at various parts of the GIT. It islmost always a secondary finding in a already active disease. It is very usual for it to be found occasionally at a routine examination.

In the following study we present a case of intestinal pneumatosis in a 49-year-old female patient who underwent routine surgery for gastric dilatation as a complication of a chronic peptic ulcer. After exploration of the abdominal cavity, a polycystic tumor formation was found at the terminal ileum. It was further resected and sent for pathohystology analysis according to which it was stated that it was a cystoid intestinal pneumatosis on a terminal ileum.

The presented case went in favour of the mechanical theory which states that pyloric gastric outlet obstruction is the most common cause of intestinal pneumatosis.

Key words: Pneumatosis cystoides, pyloric stenosis, terminal ileum, mechanical theory.

Introduction

This was identified as entity for the first time in 1730 by Du Veroni, who defined pneumatosis as the presence of gas in the GIT. [1] Later it was determined that it represents a rare condition presenting with multiple gas filled cysts in separate segments of the GIT[2] It is estimated that the incidence of pneumatosis in the overall population is 0.03%.

[3] According to one source in both males and females it is equally most common from the fourth to the seventh decade, [3] according to others it is more common in males than females with peak incidence from the third to fifth decade of life. [4] The reasons for pneumatosis are divided into benign and life-threatening diseases. The cysts in cystic pneumatosis are located mainly in the subserosal or submucosal wall and rarely in the muscle tissue of the bowel. They vary in shape and dimension from microscopic to several centimetres. They can appear in any segment of the GIT, but the most common location is the jejunum, the ileocoecal region and the colon. [5, 6] Extraintestinal structures like the mesentery, peritoneum or falciform ligament can also be involved in the process. The pathogenesis has been debated for decades and several explanations have been offered. It is very important to understand that pneumatosis is a condition, not a disease, and should always be connected with the overall clinical condition of the patient. [3, 7, 8]

Traumatic and Mechanic	***Inflamatory***	***Infective***	***Pulmonal***
GOO	IBD	Clostridium difficile	Asthma
Gastroduodenal ulcer	–	Klebsiella	COPD
Blunt abdominal trauma	Diverticulitis	Mycobacterium tuberculosis	Cystic fibrosis
Surgical anastomosis	Appendicitis	Cytomegalovirus	–

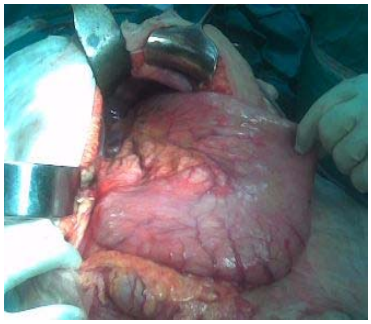
Three possibilities are suggested for for a cyst-forming gas source: intraluminal, pulmonal and microbe produced gas. First, the mechanic theory says that GOO is the most common reason for pneumatosis. This theory was proved by Hughes and associates and Gillon and associates [9, 10] separately noticing that cystic pneumatosis occurs when the partial pressure of hydrogen and methane in the bowel is similar to that in the cysts. In this case the origin of the intramural gas is well known and the mechanisms of its entrance are probably elevated intraluminal pressure or mucosal injury as well as a combination [11, 12]. On the other hand, research by Florin and associates [13] shows that an increased concentration of hydrogen in the lumen of the colon can provoke counterperfusion supersaturation that induces gas-filed cysts.

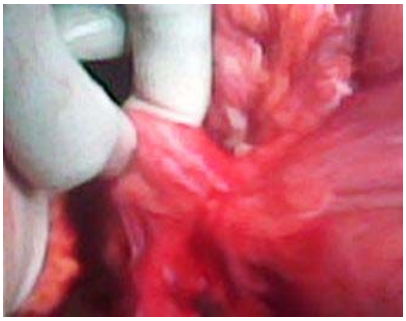
The theory of the development a PCI with gas of pilmonal origin is based on the assumption that the gas is leaking from ruptured alveoli [14] dissecting through vascular channels in the mediastinum and caudaly into the retroperitoneum and bowel mesentery. [1]

Case report

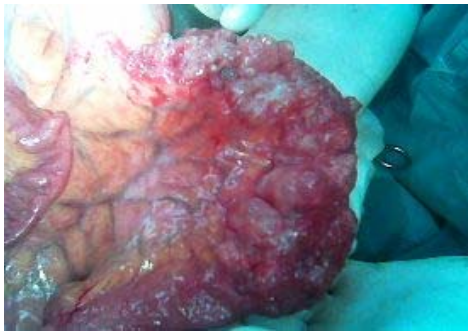
A female patient aged 49 was hospitalized for elective treatment of previously diagnosed GOO and gastrectasia due to a chronic peptic ulcer. At the

time of arrival the patient reported supraumbilical pain and vomiting in the past few weeks. The problems of abdominal discomfort and pain connected with taking food had started 13 years earlier, with intermittent remissions. A few months before, the intensity of the symptoms had increased and continued with nausea, vomiting and weight loss. The patient declared that she had undergone a gastroscopy a few years earlier when a diagnosis of duodenal ulcer was determined. After that the patient underwent conservative treatment. On physical examination at first admission the abdomen was soft and relaxed, palpable and painless at superficial palpation, at the level of thorax with pain in the epigastrium on deep palpation. Laboratory analysis showed a slight decrease of Haemoglobin (97 g/l) and packed cell (0.33). The patient was directed to further investigations where gastoscopy and gastorduodenography were performed and a diagnosis of pyloric stenosis and gastrectasia was confirmed. The patient was then admitted and prepared for the operating theatre where the operation was started. A supraumbilical median laparatomy was performed. After opening the abdominal cavity and exploration, the following finding was present: stenotic pylorus with an abnormally increased stomach (27 cm l; 10 cm d);

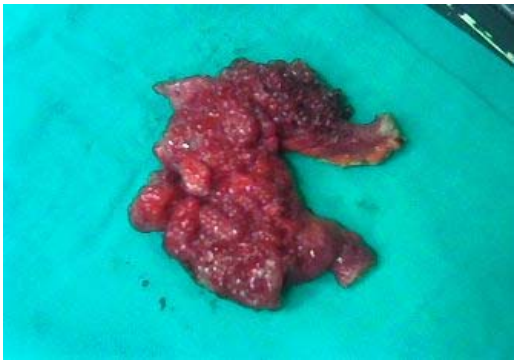




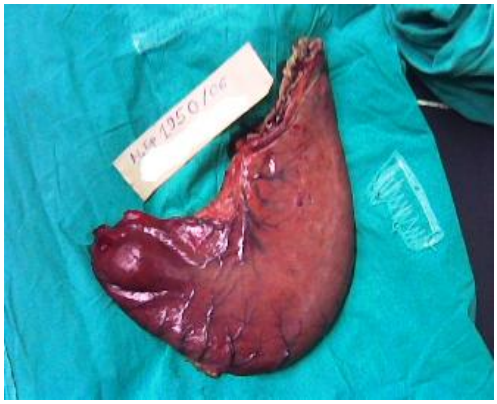
Tumefaction at the terminal ileum distant 20 cm from the valvula Bauchini to a length of 15 cm with numbered cystic macroscopic formations of different sizes.



After that we decided to make the first resection of that part of the ileum then T-T anastomosis. Then followed the gastrectomy Bilroth II. Subhepatic drainage was set.



Definitive exploration was then done and the operative wound was then closed with haemostatic control. The operative material was sent to the Medical Faculty Institute for Pathologic Anatomy in Skopje, where following finding was concluded: At first, operative material of stomach with increased dimensions, tall and prominent mucosal folds without macroscopic erosions and ulcerations, and astenotic place on the pylorus with the lumen passable for 0.5 cm.



Microscopic, there was seen chronic inflammatory infiltration with lymphocytes and intestinal metaplasia into the antrum with bleeding erosions focus and an oedematic edge of the pylorus. The second operative material resected segment from the ileum: macroscopic increase of size was notified with the presence of serous cystic formations like clusters and macroscopic intact mucosa. Under the microscope, a numbered airway cyst was noted, subserous and submucose of a different size covered with endotel. Also, it was confirmed

that the cysts did not communicate with the lumen of the bowel or with each other, which means that the finding at the terminal ileum was pneumatosis cystoides intestinalis. The postoperative follow-up was orderly and the patient left the clinic on the eighth post-operative day. She came twice to follow-up examinations at which orderly findings were affirmed.

Discussion

Intestinal pneumatosis can occur in irregularly medicated cases of peptic ulcer [15, 16, 17, 18], it being assumes that perforation multiple times, where perforation stubs into the "tecta" perforation that conditioned the symptoms remission. Some older studies have given data that showed 60% of cases reported by the time of appearance of pneumatosis were connected with pyloric stenosis. [18] Others have suggested that reason for maintained pneumatosis in the distal parts of the bowel is mucosal distension which allows disruption and conditions the spreading of gas through mesenterial lymphatics. [7, 16, 18, 19] In 1952 the pathologist Koss made the most complete study until then of intestinal pneumatosis, reporting a case of a patient with pyloric stenosis as a complication of a chronic peptic ulcer. [7] Later, with the implementation of modern diagnostic and intervention procedures, the spectrum of pneumatosis has expanded, [20] new causes for the occurrence of the condition have been found and therapeutic indications have been expanded. However, the lack of symptoms and a real clinical picture very often do not indicate the existence of this condition with an already complicated peptic ulcer.

Clinical manifestations of a patient with pneumatosis are almost always the consequence of a previously existent pathologic condition that induces pneumatosis, not the condition itself, which is why it frequently happens to be found occasionally intraoperative [11]. Although the condition is most benign, 27% of patients with benign forms of pneumatosis are presented with a wrong diagnosis of an acute abdomen and undergo unnecessary surgical treatment. [3, 21]

The cystoid pneumatosisas in our caseis only a subgroup of intestinal pneumatosis whose reason for occurrence is mostly a benign disorder of the GIT. Determination whether the condition is benign and should be treated only with conservative treatment or not, depends on laboratory findings, physical findings and radiology investigation. [5, 21, 22, 23] Our case was admitted to hospital for elective treatment of pyloric stenosis and gastrectasia as a complication of peptic ulcer disease, of which the symptoms had existed for more than 13 years. Apart from the symptoms and discomfort that clearly indicated GOO and gastrectasia, other symptoms and problems were not found. That is why diagnostic investigations and interpretations where targeted at the primary

disease. At the time of surgery, on exploration of the abdominal cavity, the tumefaction described in the text above was found. Because of the inability to assess morphologically whether it was a benign or malignant condition and because of the size of the tumefaction we approached radical resection of the mentioned part of the ileum so that optimal radicalism was assured even if it was an eventual malignity. In the cases of occasionally found intraoperative pneumatosis and the character of the change cannot be 100% sure, operative removal is recommended. The focus of the treatment should completely be directed to the primary disease. Some authors takes a position that occasionally found intestinal intraoperative pneumatosis should always be surgically treated. [24]

Conclusion

Intestinal Pneumatosis is a condition almost always caused by an already existing disease. It is rare and mostly found occasionally either intraoperative leading to surgical treatment of primary disease or radiological treatment.

The etiopathogenesis of the presented case goes in the direction of the mechanical theory which says that pyloric stenosis is the most common reason for intestinal pneumatosis.

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Резиме

PNEUMATOSIS CYSTOIDES INTESTINALIS АСОЦИРАНА СО ПИЛОРНА СТЕНОЗА: ПРИКАЗ НА СЛУЧАЈ

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Вовед: Пнеуматозата претставува ретка состојба презентирана со мултипни цисти исполнети со гас во одделни сегменти на гастроинтестиналниот тракт. Скоро секогаш е секундарен наод на веќе постоечко заболување. Ретка е и најчесто се открива случајно.

Приказ на случај: Презентираме случај на интестинална пнеуматоза кај 49-годишна пациентка, која што се јавила за елективен оперативен третман на претходно дијагностицирана пилорна стеноза и гастректазија, како компликација на хроничен пептичен улкус. На експлорација на абдоминалната празнина, на терминален илеум е најдена полицистична туморозна формација, при што истата е ресецирана и испратена на патохистолошка анализа според која е утврдено дека се работи за цистоидна пнеуматоза на терминалниот илеум.

Заклучок: Презентираниот случај, етиопатогенетски оди во прилог на механичката теорија која вели дека пилорната стеноза е најчеста причина за интестинална пнеуматоза.

Клучни зборови: Цистоидна пнеуматоза, пилорна стеноза, терминален илеум, механичка теорија.

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