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Cystic intestinal pneumatosis revealed by peritonitis in perforated peptic ulcer: A case report

Perfore peptik ülserde peritonit ile ortaya çıkan kistik intestinal pnömatozis: Olgu sunumu

Ismaël Dandakoye Soumana [1], Anas Belhaj [1], Moussa Sylla [1], Somuah Tenkorang [2], Mouaqit Ouadii [1], Ibn Majdoub Hassani [1], Imane Toughrai [1], Hassan Moulay Farih [1], Khalid Mazaz [1], Ait Taleb Khalid [1]

[1] Visceral and Endocrinological Surgery Service II, Chu Hassan II, Fes, Morocco

ORCID ID of the authors
IDS: 0000-0003-2451-4199
AB: 0000-0002-1118-1594
MS: 0000-0002-5739-7382
ST: 0000-0001-5657-7253
MO: 0000-0003-3286-7568
IMH: 0000-0002-0421-7296
IT: 0000-0003-0401-3012
HMF: 0000-0001-8997-5717
KM: 0000-0001-7779-7802
ATK: 0000-0002-0920-796X

Abstract

Cystic intestinal pneumatosis is a rare condition characterized by the presence of cyst-like formations in the walls of the digestive tract. Cystic intestinal pneumatosis can affect any part of the digestive tract. However, the small and large bowels are the most affected. This pathology is more common in men and after the age of 50 years. Cystic pneumatosis of the intestines may be idiopathic or most often secondary to various diseases. The primitive forms preferentially affect the left colon with essentially submucous gaseous cysts, whereas the secondary intestinal cystic pneumatosis tend to affect the small intestines especially in the subserosa. The origin of this pathology is multifactorial but the main cause is not definitively proven. The long list of pathological associations has led to the development of various etiopathogenic theories that are not antinomic since some mechanisms may be associated.

Cystic intestinal pneumatosis is most often asymptomatic and therefore fortuitously discovered. This pathology could have clinical signs like bloody and glairy stools, abdominal pain, and diarrhea. Endoscopic and radiological examinations are easy to diagnose and avoid unnecessary laparotomy explorations in benign pneumoperitoneum. Treatment varies according to its etiology. For primary forms, antibiotic therapy to reduce the hydrogen-producing colonic flora is indicated as first-line treatment. If unsuccessful, oxygen mask or hyperbaric, which will promote the replacement of hydrogen with oxygen, must be attempted. For secondary forms, the treatment is that of the causal affection. In most cases, cystic intestinal pneumatosis is asymptomatic and no treatment is needed. Surgery remains reserved for serious forms of this disease.

Keywords: Cystic intestinal pneumatosis, Cysts, Pneumoperitoneum, Perforated peptic ulcer

Öz

Kistik bağırsak pneumatosisis sindirim sistemi duvarlarında kist benzeri oluşumların varlığı ile karakterize nadir bir durumdur. Kistik bağırsak pnömotozu sindirim sisteminin herhangi bir bölümünü etkileyebilir. Bununla birlikte, küçük ve büyük bağırsaklar en çok etkilenenlerdir. Bu patoloji erkeklerde ve 50 yaşından sonra daha yaygındır. Bağırsakların kistik pnömatozu, idiopatik veya çoğu zaman çeşitli hastalıklara ikincil olabilir. İlkel formlar, esas olarak submukozlu gazlı kistler ile sol kolonunu tercih ederler, oysa sekonder intestinal kistik pnömatozlar, özellikle ince bağırsakların subserozasını etkileme eğilimindedir. Bu patolojinin kökeni çok faktörlüdür, ancak ana sebep kesin olarak kanıtlanmamıştır. Patolojik ilişkilerin uzun listesi, çeşitli etiyopatojenik teorilerin gelişmesine yol açmıştır.

Kistik bağırsak pnömotozu en sık asemptomatiktir ve bu nedenle tesadüfi bir şekilde saptanır. Bu patoloji kanlı dışkılama, karın ağrısı ve diyare gibi klinik belirtilere sahip olabilir. Endoskopik ve radyolojik incelemeler benign pnömoperitoneumda gereksiz laparotomi eksplorasyonlarını tespit etmek ve önlemek için faydalıdır. Tedavi etiyolojisine göre değişir. Birincil formlar için, hidrojen üreten kolonik florayı azaltmak için antibiyotik tedavisi, birinci basamak tedavi olarak belirtilmektedir. Başarısız ise, oksijen ile oksijen değiştirilmesini teşvik edecek oksijen maskesi veya hiperbarik tedavi denenmelidir. Sekonder formlar için tedavi, nedensel olarak değişkendir. Çoğu durumda, kistik bağırsak pnömatozu asemptomatiktir ve tedaviye gerek yoktur. Cerrahi, bu hastalığın ciddi formları için ayrılmıştır.

Anahtar kelimeler: Kistik bağırsak pnömatozu, Kist, Pnömoperiton, Peptik ülser perforasyonu

Corresponding author / Sorumlu yazar:
Ismaël Dandakoye Soumana
Address / Adres: Visceral and Endocrinological Surgery Service II, Chu Hassan II, Fes, Morocco
e-Mail: sairaisma14@gmail.com

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Introduction

Cystic intestinal pneumatosis (CIP), which is characterized by the presence of pseudocysts or air bubbles in the subserosa or submucosa of the digestive wall, is a rare benign condition [1]. It can be localized in the various segments of the digestive tract, from the esophagus to the rectum [2,3]. The small intestine appears to be more affected than the colon. Both small intestine and colon can be affected in 20% of cases [4,5].

This pathology is often asymptomatic and therefore discovered fortuitously. Endoscopic and radiological examinations are easy tools to diagnose this pathology. These diagnostic tools help to avoid unnecessary laparotomy in benign pneumoperitoneum. In most cases, CIP is asymptomatic and no treatment is needed. Surgery is reserved for particularly serious forms [4-7]. We report the case of a 37-year-old chronic smoker admitted to the emergency department for management of diffuse abdominal pain with incidental discovery of CIP.

Case presentation

A 37-year-old patient, with a 20 year history of cigarette smoking, presented with epigastric pain associated with a subocclussive syndrome, fever and a few episodes of vomiting. These symptoms had begun two days prior to his admission to the emergency department. Clinical examination found a feverish, conscious patient with a body temperature of 38 °C. Abdominal examination revealed a diffuse abdominal sensitivity more accentuated at the epigastric level. Laboratory test were within normal limits except C-reactive protein at 32mg/l and renal insufficiency. Plain abdominal X-ray objectified multiple air fluid levels without pneumoperitoneum. Abdominal non contrast computed tomography (CT) scan performed objectified pneumoperitoneum with suspicion of intestinal pneumatosis (Figure 1).

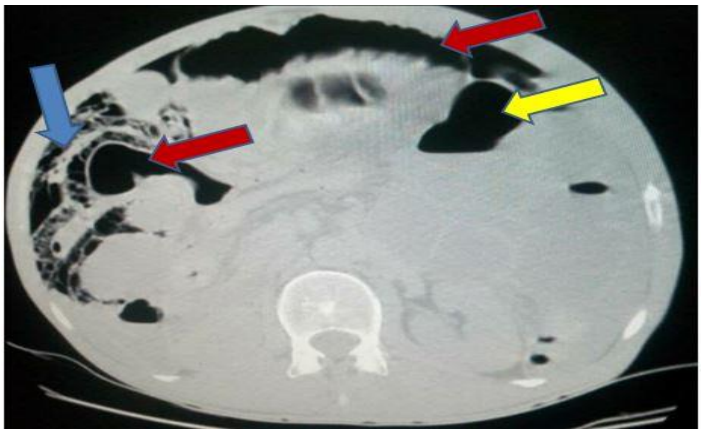

Figure 1: Abdominal computed tomography of the patient. Blue arrow: Suspicion of parietal pneumatosis, red arrow: gas in the small intestine, yellow arrow: hydro-aeric level colic type

Surgical exploration revealed a 1 cm perforation on the anterior surface of the duodenal bulb (Figure 2). We also found a cystic pneumatosis affecting 1.3m of the small bowel. The perforated zone was repaired with abundant rinsing and drainage of the peritoneal cavity. No action was taken for the CIP.

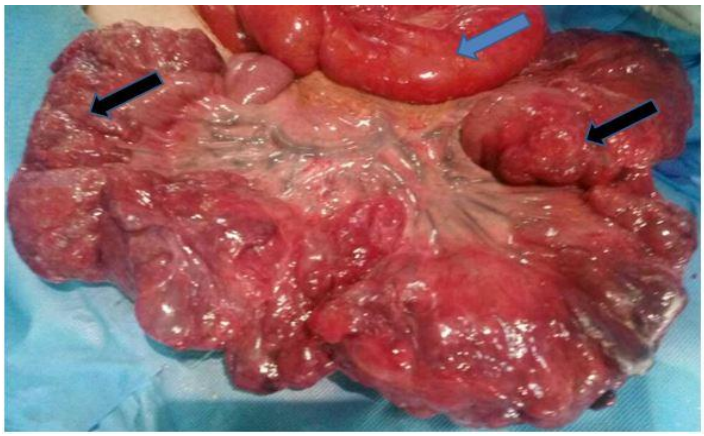

Figure 2: View in operation. Blue arrow: normal small intestine, black arrow: pathological small intestine

The postoperative course was marked with no incident. The patient was discharged on the 3rd day after surgery.

Discussion

CIP is characterized by the presence of cysts-like formations localized in the submucosa or subserosa of the digestive tract. These formations measure from a few millimeters to several centimeters. The cysts may concern a segment of the intestine or be diffused in the intestine extending to several meters, sometimes unrelated to the site of the causal lesion. The small bowel are the most frequent affected (42%) followed by the colon (32%); It is relatively less frequent to have both small and large bowels affected. When the large bowel is involved, sigmoid colon is the most frequent affected (70% of cases), followed by the descending colon in 40% of cases, and other segments in 15 to 25% of cases, including 10% for the caecum [6].

CIP are generally submucous in the colon, appearing as sessile polypoid nodules. They are more often subserosal in the small intestine, taking the form of air bubbles in grape-like clusters and are mostly localized on the mesenteric edge [6]. The wall of cysts is sometimes very thin and can be broken, either spontaneously or after an endoscopic biopsy causing pneumoperitoneum [8-10].

In 85% of cases, CIP secondary or associated with other gastrointestinal pathologies (inflammatory bowel disease, peptic ulcer, pyloric stenosis, abdominal trauma) or extra gastrointestinal (chronic obstructive pulmonary disease, heart disease, cystic fibrosis, lupus, periarteritis nodosa); primitive forms account for only 15% of cases reported [8-9]. In our patient, we found cystic pneumatosis linked to a peptic ulcer.

Many theories have been proposed to explain how CIP occur. Currently the most likely pathogenesis combines mechanical and bacterial theories: association of mucosal lesions, an increase in digestive intraluminal pressure allowing anaerobic bacteria (producing hydrogen) to enter the intestinal wall. The principal mechanism of this pathology is the mucous breach which seems indispensable [11-13]. For others, pneumatosis is explained by a deficit in hydrogen reducing bacteria: methanogenic bacteria [13].

CIP is usually pauci-symptomatic. Most authors report nonspecific signs in 30% of cases: diarrhea, bloody or glairy stools, meteorism, vomiting, constipation, tenesmus [6]. Abdominal meteorism is found in 38% of cases in the Jamart

series [6]; luminal occlusion associated with cysts may be responsible for transit disorders. Some rare complications related to cystic volume have been described (3%): volvulus, intussusception, perforation, hemorrhage. These complications may require segmental intestinal resections [14].

Plain X-rays of the abdomen often show pneumoperitoneum due to rupture of subserosal cysts in the peritoneal cavity. Cystic pneumatosis is the first cause of pneumoperitoneum without digestive perforation. It is present in 15% of the cystic pneumatosis in the small bowel and in 2% of the affected colon [11,12]. Cysts are best seen on the colon wall. They are effected by the presence of air formations contiguous to each other having the appearance of 'grape-like clusters' in the wall of the colon [11,15].

Computed tomography with intestinal opacification has a good diagnostic accuracy [16]. It reveals gaseous density images in the digestive wall, well detectable in a cross-sectional and lung window view [17,18]. The presence of these images associated with an asymptomatic pneumoperitoneum is almost pathognomonic of CIP [6]. An ultrasound appearance has been described associating a thinning of the intestinal wall and echogenic gas bubbles often seen as a circle [19]. There is an important diagnostic criterion, which is the gas in the portal vein (unlike intestinal gangrene) on the CT scan or ultrasound [20]. In endoscopy, cysts correspond to large hemispherical sessile polyps, covered with pale and transparent mucosa, which is sometimes ulcerated. Typically, the cyst is collapsed by puncture or biopsy with a burst sound effect [21].

The treatment of CIP is most often medical. Its purpose is to reduce or eliminate cysts by reducing the anaerobic bacteria that cause them. Anti-anaerobic antibiotherapy with metronidazole is often effective [12,15], but other antibiotics such as ampicillin or fluoroquinolones have been successful [11]. Hyperbaric oxygen therapy is used for its anti-anaerobic power and for its ability to collapse cysts by promoting exchanges with blood [12,22]. Other therapeutic options such as octreotide or endoscopic fenestrations have been used with varying results [12]. Surgical treatment is indicated in case of complications and in case of symptoms that are resistant to medical treatment [12,22]. It involves resecting the intestinal segment through laparotomy or via laparoscopy. The latter is preferred because of the benignity of the pathology and the favorable local conditions (absence of inflammation, absence of adhesion attributable to this pathology) [22,23].

In conclusion, cystic pneumatosis of the small intestine is a rare benign affection that is diagnosed with the means of radiological techniques. It should not be confused with a simple pneumoperitoneum or pneumatosis by intestinal gangrene. For this, the multidetector CT scan is extremely efficient to detect air of air in the digestive serosa, the different peritoneal and extra spaces. The CT scan also guides the surgeon to treat a possible complication such as peritonitis by perforation of the digestive tract.

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