



## Case Series

# Pneumatosis cystoides intestinalis with pneumoperitoneum secondary to stenosing pyloro-duodenal peptic ulcer: Case series of three patients and literature review



Khalid El Hattabi <sup>a,b</sup>, Oussama Lafkih <sup>a,b,\*</sup>, Fatimazahra Bensardi <sup>a,b</sup>, Abdelilah Elbakouri <sup>a,b</sup>, Mounir Bouali <sup>a,b</sup>, Abdelaziz Fadil <sup>a,b</sup>

<sup>a</sup> Department of General Surgery, University Hospital Centre Ibn Rochd, Casablanca, Morocco

<sup>b</sup> Faculty of Medicine and Pharmacy, Hassan II University, Casablanca, Morocco

## ARTICLE INFO

## Article history:

Received 16 February 2021

Received in revised form 11 March 2021

Accepted 11 March 2021

Available online 13 March 2021

## Keywords:

Pneumatosis cystoides intestinalis

Pneumoperitoneum

Diagnosis

Peptic ulcer

Case series

## ABSTRACT

**INTRODUCTION:** Pneumatosis cystoides intestinalis (PCI) is a rare condition, which can affect the entire gastro-intestinal tract. It can be idiopathic or most often secondary to various diseases. The causes remain multiple and the stenosing peptic ulcer is one of them. We report three case reports of pneumatosis cystoides with pneumoperitoneum intestinalis secondary to stenosing pyloro-duodenal peptic ulcer.

**CASE PRESENTATION:** We report a case series of three patients with PCI that presented to the emergency department with a reassuring clinical picture with the discovery in imaging of a pneumoperitoneum. All our patients presented with a pyloro-duodenal stenosis secondary to a complicated peptic ulcer, one of which was treated surgically and the other two endoscopically.

**DISCUSSION:** The objective of this report is to provide an update on pneumatosis cystoides intestinalis secondary to stenosing pyloro-duodenal peptic ulcer, by specifying its etiopathogenic, diagnostic and therapeutic characteristics.

**CONCLUSION:** Knowledge of this pathology is necessary in order to avoid unnecessary abusive surgery.

© 2021 The Author(s). Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

## 1. Introduction

Pneumatosis cystoides intestinalis (PCI) is defined by gas-filled cysts in the wall of the gastro-intestinal tract [1]. First described in 1730 by the French DUVERNOY during an autopsy report [2]. PCI is classified as primary (15%) or secondary (85%) depending on its etiology [3]. The pathogenesis of PCI has long been discussed. However, the number of reported observations has led to the development of different theories that provide a pathophysiological approach and a better understanding of the etiopathogenic mechanisms underlying the formation of intestinal cysts. Among these, we believe that mechanical theory best explains the situation of our patients. This theory suggests that an increase in intraluminal pressure and a rupture of mucosal integrity (peptic ulcers with pyloric stenosis in our patients) allow intraluminal gas to penetrate the intestinal wall [4]. It is a rare condition, but probably underdiagnosed due to its low symptoms. The treatment has not

yet been codified but remains mainly medical. Surgical indications are reserved for complicated forms [3]. Knowledge of this pathology is necessary in order to avoid unnecessary abusive surgery [1]. The objective of this report is to provide an update on pneumatosis cystoides intestinalis secondary to stenosing pyloro-duodenal peptic ulcer, through a retrospective case series of three patients collected in the department of surgical emergency of Ibn Rochd University Hospital Center Casablanca between 2016 and 2020, with a literature review.

This case series has been reported in line with the PROCESS Criteria [5].

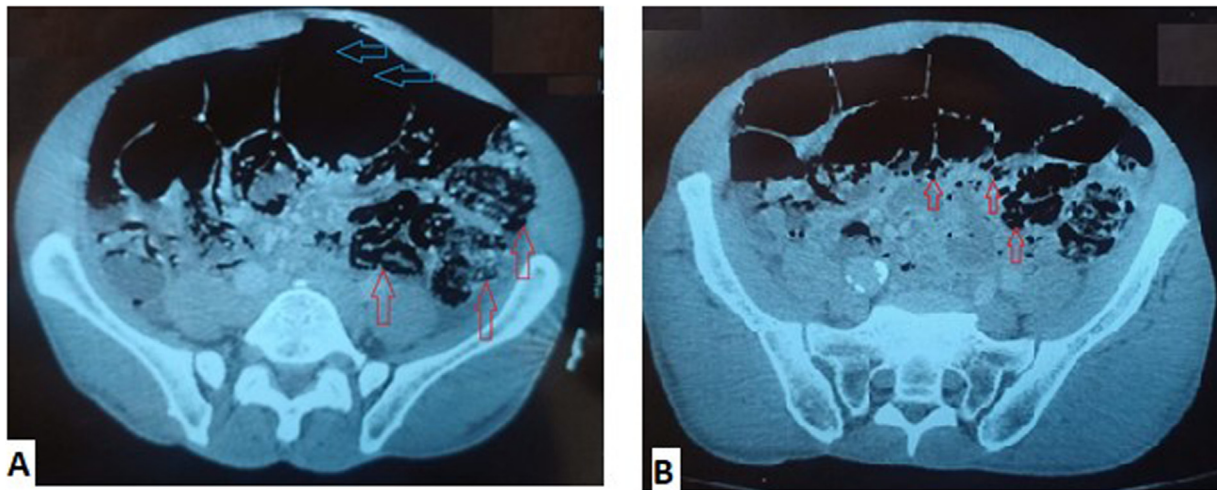
## 2. Case presentation

### 2.1. Case 1

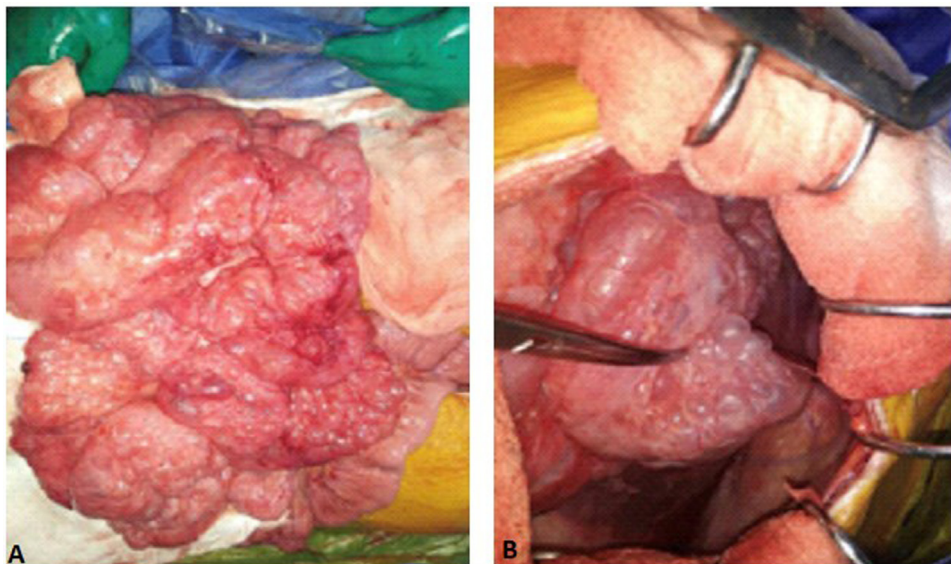
A 44-year-old male patient, chronic tobacco user, Admitted to the emergency for mild abdominal pain, and post-prandial vomiting since a month. Operated 10 years ago for peritonitis secondary to perforation of a gastric peptic ulcer, by midline laparotomy. Physical examination revealed slight abdominal distension without tenderness. Body temperature was 37.2 °C. Abdominal CT (Fig. 1)

\* Corresponding author at: les collines Sidi Maarouf residency, appt 2 Imm F, Casablanca, Morocco.

E-mail address: [lafkih27@gmail.com](mailto:lafkih27@gmail.com) (O. Lafkih).



**Fig. 1.** Abdominal CT scan showing abundant intra-abdominal free-air (blue arrows) and cyst like structures located trough the small bowel and mesentery (red arrows).



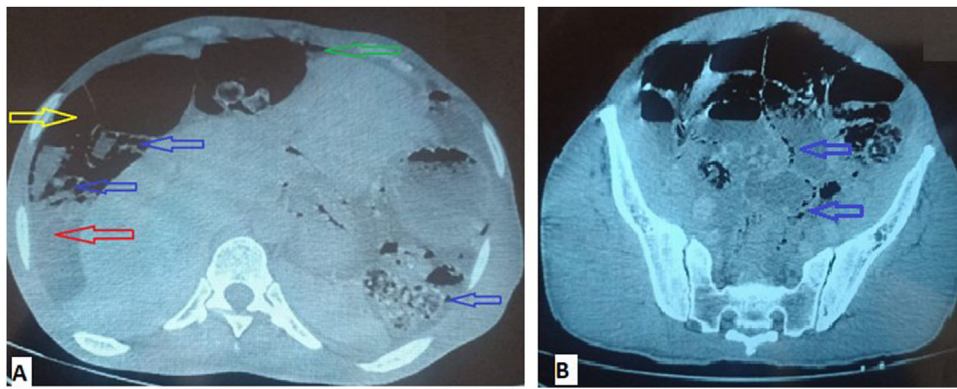
**Fig. 2.** Intra-operative findings: a gas-filled, thin-walled, cyst like structures located trough the small bowel and mesentery.

showed abundant pneumoperitoneum with moderate peritoneal free fluid and Intramural bowel gas. The laboratory analysis found normal white blood cell (WBC = 6200/ $\mu$ L), and C-reactive protein was 10.2 mg/l. In front of these findings, the diagnosis of PCI was the top of differential. Gastrointestinal perforation was rejected because of the absence of clinical and biological signs of peritonitis. Esophago-gastro-duodenoscopy showed gastric stasis with complete pyloric obstruction, due to peptic ulcer. Several biopsies were performed. Pathological examination revealed non-specific inflammation without signs of malignancy. Ascites puncture was realized, which was exudative and predominantly lymphocytes with normal adenosine deaminase activity. A delayed explorative laparotomy was performed, under general anesthesia with endotracheal intubation, by an assistant professor, a 5th year and a 4th year surgical resident. Preoperative prophylactic antibiotics were administered (Cefoxitin 2 g + Metronidazole 1g: a single dose injection). Intra-operative findings (Fig. 2) were: abundant ascites, a gas-filled, thin-walled, cyst like structures located trough the small bowel and mesentery. Gastrojejunostomy was carried out using an omega loop with a biopsy of cystic mesentery lesions. The pathological study showed chronic fibro-inflammatory changes without signs of specificity or malignancy. Postoperative course was simple.

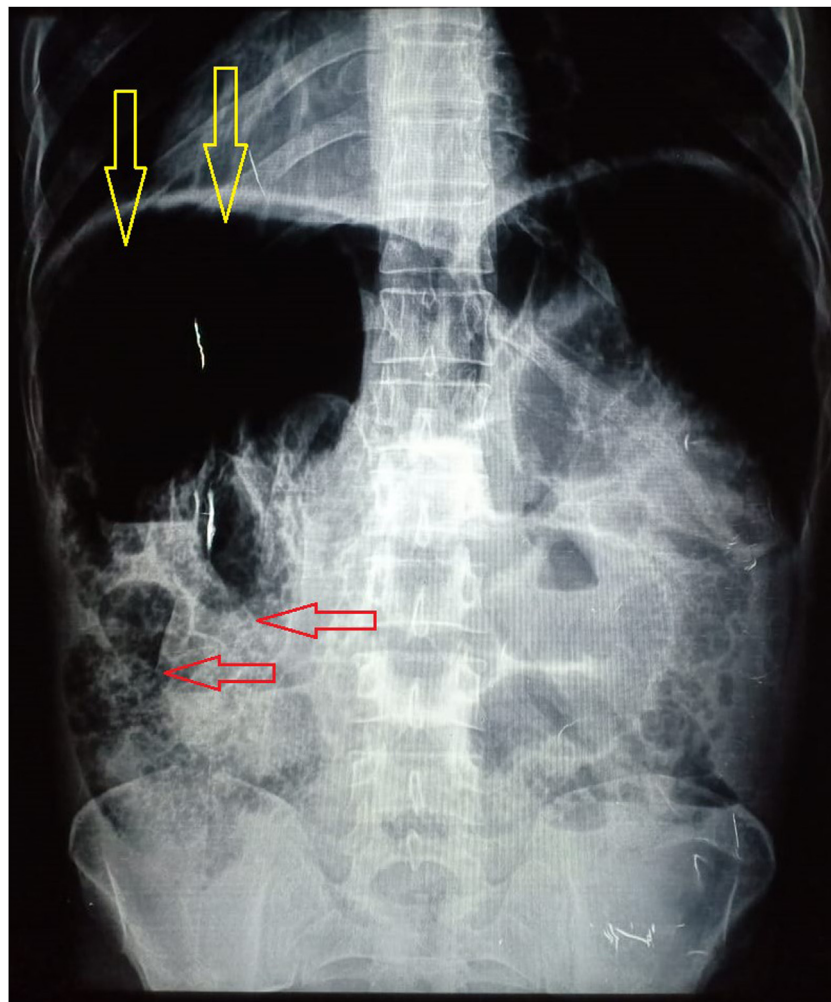
The patient was discharged on the seventh postoperative day, then he was lost to follow-up. He did not show up for his post-operative appointments.

## 2.2. Case 2

57-year-old male patient, with a 20-pack-year smoking history, admitted to the emergency for diffuse abdominal pain, starting epigastric, and vomiting with mild abdominal distension associated with chronic constipation, for two months. Operated in 2009 for perforated pyloric peptic ulcer, by midline laparotomy. On examination, his vital signs were: temperature 37.3 °C, pulse 90/minute, respiratory rate 18/minute, and blood pressure 110/60 mmHg. An abdominal examination revealed a mild distension of his abdomen without signs of peritonitis. His abdomen was tympanic to percussion. Abdominal CT (Fig. 3) showed a low abundance pneumoperitoneum with moderate abundance intraperitoneal effusion. With small bubbly cysts beneath the intestinal serosa in pelvic location. The colon was interposed between the liver and diaphragm (Chilaiditi Syndrome) and a good opacification of mesenteric vessels. Blood biochemistry revealed normal level of white blood cells (8940/ $\mu$ l), with normal C-Reactive Protein (0.5 mg/l). In



**Fig. 3.** A- Abdominal CT showing: low abundance pneumoperitoneum (green arrow) with intraperitoneal effusion (red arrow). With the demonstration of small bubbly cysts beneath the intestinal serosa (blue arrow) and Chilaiditi Syndrome (red arrow).

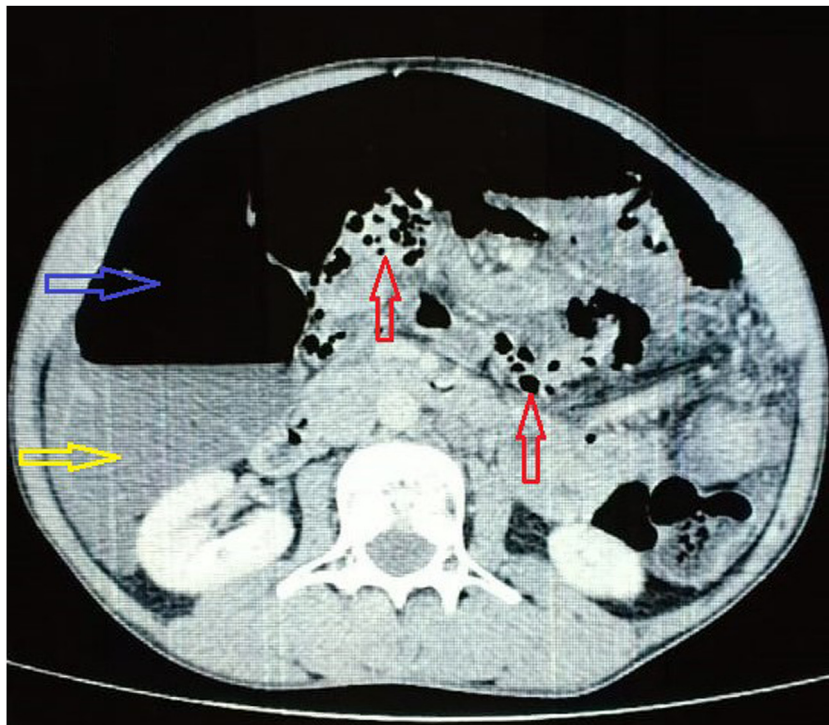


**Fig. 4.** Abdominal X-ray showing a very abundant pneumoperitoneum under diaphragm (yellow arrows) multiple small gas cysts lining the intestinal (red arrow).

front of these findings, the diagnosis of PCI was the top of differential. Gastrointestinal perforation was rejected because of the absence of clinical and biological signs of peritonitis. A nasogastric tube was placed with antibiotic therapy (Metronidazol 500 mg: 3 times/day during 10 days) was administered. Eso-gastro-duodenoscopy was realized, which showed gastric stasis with antral gastritis, and large bulbar duodenal stenotic ulcer which remains passable. Several biopsies were performed. The pathological examination showed non-specific inflammation, with the

presence of *Helicobacter Pylori*. The patient received medical treatment for his *Helicobacter Pylori* (HP) gastritis: Proton pump inhibitor: Omeprazol (20 mg), Clarithromycin (500 mg), Amoxicillin (1 g) twice daily during 14 days, and was then referred to the gastro-enterology department for endoscopic dilation sessions. He was seen in two follow-up visits at 2 weeks and 4 weeks, he finished his treatment of HP gastritis and benefited of two endoscopic dilatation sessions. The course was marked by good clinical outcomes.





**Fig. 5.** Abdominal CT scan showing the presence of abundant pneumoperitoneum (blue arrow) multiple air-filled cysts in the intestine wall (red arrows), and moderate ascites (yellow arrow).

### 2.3. Case 3

44-year-old male, who was admitted to our surgical department with diffuse abdominal pain associated with postprandial vomiting and few episodes of liquid diarrhea. He was followed in gastro-enterology department for a gastric ulcer on proton pump inhibitors for 3 years. Without any previous surgical history. On examination his blood pressure was 120/60 mm Hg, heart rate was 75 beats/min, body temperature was 37 °C, and respiratory rate was 18 breaths/min. Abdominal examination found a distended abdomen, without tenderness and tympanic on percussion. The abdominal x-ray (Fig. 4) revealed abundant free-air under the diaphragm. Abdominal CT scan (Fig. 5) showed abundant pneumoperitoneum identified in the perihepatic and inter-loop location, multiple air-filled cysts in the intestine wall, and moderate ascites with good opacification of the mesenteric vessels. The laboratory test had noted a normal white blood cell level at 9180/ $\mu$ l, and normal CRP at 2.7 mg/l. In front of these findings, the diagnosis of PCI was the top of differential. Gastrointestinal perforation was rejected because of the absence of clinical and biological signs of peritonitis. A nasogastric tube was placed and antibiotic therapy (Metronidazole 500 mg: 3 time/day during 10 days) was administered. Eso-gastroduodenal fibroscopy was performed, and showed the presence of bulbar duodenal narrowing which remains passable, with erythematous fundic and antral gastritis. Several biopsies were performed, the anatomopathological study showed non-specific inflammatory change, with moderate chronic gastritis, testing for *Helicobacter pylori* was negative. The patient was then referred to the gastroenterology department for endoscopic dilation sessions, and he was seen in two follow-up visits at 4 weeks and 6 weeks with good clinical evolution.

### 3. Discussion

Pneumatosis cystoides intestinalis is a rare condition that is always underestimated because of its frequently asymptomatic

nature [6]. In the literature, studies are dominated by case reports and small series with few retrospective studies [7]. PCI lesions can sit from the esophagus to the rectum over the entire gastrointestinal tract [8]. The most common symptoms of PCI are diarrhea or constipation, bloody stools, abdominal pain, asthenia or weight loss. It has been suggested that these symptoms are caused by mechanical effects of the cysts, but in many cases even large cysts remain asymptomatic [9]. PCI is diagnosed using imaging tests (x-rays or CT scan). An x-ray of the abdomen shows multiple small gas cysts lining the intestinal walls with pneumoperitoneum. A CT scan shows multiple gas cysts in the intestinal walls. PKI is sometimes accompanied, as an indirect sign, by pneumoperitoneum, gas in the portal vein or Chilaiditi syndrome (found in one of our patients). In the cases we presented, massive pneumoperitoneum was found in all 3 cases with multiple small gas cysts in the wall of the small bowel and colon on the CT scan. Notably in all 3 cases, ascites was observed on the CT scan. The appearance of gas cysts on the CT scan can be described as linear, circular and bubbly; however, it is not useful for determining the severity of PCI [3]. The more widespread use of CT scans in recent years has led to increased recognition of this pathology. Careful correlation of the clinical presentation with corroborating biological tests should determine whether a conservative medical approach or emergency surgical exploration is appropriate [10].

PCI is often benign and only conservative treatment, and follow up is warranted. Treatment of PCI is generally reserved for symptomatic patients [11,12]. A serious cause of intestinal pneumatosis that requires immediate medical and/or surgical management must first be eliminated. The CT scan findings (associated signs of severity) must be compared with the clinical and biological data. Then, benign causes can be suggested and the treatment can be that of the cause (stenosing duodenal peptic ulcer in our cases) [13,14]. Medical treatment can sometimes be proposed: Antibiotics (metronidazole, tetracycline, ampicillin and vancomycin) reduce the production of hydrogen, but have an inconstant effect. Oxygen therapy hyperbaric has a bactericidal effect [15]. In our first

case, surgery was only indicated to manage his complete stenosing duodenal peptic ulcer.

#### 4. Conclusion

PCI is a benign pathology that can be secondary to several diseases including peptic ulcer, its treatment is essentially based on the treatment of the underlying cause (ulcer stenosis), it can be medical or surgical. Knowledge of this rare pathology makes it important to avoid unnecessary exploratory laparotomy.

#### Declaration of Competing Interest

The authors report no declarations of interest.

#### Sources of funding

No sources of funding.

#### Ethical approval

I declare on my honour that the ethical approval has been exempted by my establishment.

#### Consent

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

#### Author contribution

Oussama Lafkih: Corresponding author writing the paper, Khalid El Hattabi: writing and correction of the paper, Fatimazahra Bensardi: correction of the paper, Abdelilah Elbakouri: correction of the paper, Mounir Bouali: correction of the paper, Abdelaziz Fadil: correction of the paper.

#### Registration of research studies

Not applicable.

#### Guarantor

Dr. Oussama Lafkih.

#### Provenance and peer review

Not commissioned, externally peer-reviewed.

#### CRediT authorship contribution statement

**Khalid El Hattabi:** Conceptualization, Methodology, Validation, Formal analysis, Investigation, Resources, Data curation, Writing - original draft, Writing - review & editing, Visualization, Project administration. **Oussama Lafkih:** Conceptualization, Methodology, Software, Validation, Formal analysis, Investigation, Resources, Data curation, Writing - original draft, Writing - review & editing, Visualization, Funding acquisition. **Fatimazahra Bensardi:** Validation, Resources, Writing - review & editing, Visualization, Project administration. **Abdelilah Elbakouri:** Visualization. **Mounir Bouali:** . **Abdelaziz Fadil:** Supervision.

#### References

- [1] C. Laclotte-Duhoux, M.-A. Bigard, Pneumatose kystique intestinale, *EMC - Gastro-Entérologie* 5 (3) (2010) 1–8, janv.
- [2] G. Du Vernoi, Aer intestinorum tam subextima quam intima tunica inclusus, *Obsergatinonae Anatomicae Acad. Sci. Imp. Petropol.* 5 (1730) 213–215.
- [3] K. Wada, N. Takeuchi, M. Emori, M. Takada, Y. Nomura, A. Otsuka, Two cases of pneumatosis cystoides intestinalis with intraperitoneal free air, *Gastroenterol. Res.* 10 (3) (2017) 208–211.
- [4] C.W.Y. Wong, P.H.Y. Chung, K.K.Y. Wong, P.K. Tam, Pneumatosis intestinalis presenting as pneumoperitoneum in a teenage girl with pyloric stenosis, *BMJ Case Rep.* (2015) [Internet]. 26 févr [cité 11 juill 2020]; 2015. Disponible sur: <https://www.ncbi.nlm.nih.gov/pmc/articles/PMC4342669/>.
- [5] R.A. Agha, C. Sohrabi, G. Mathew, T. Franchi, A. Kerwan, N. O'Neill, et al., The PROCESS 2020 guideline: updating consensus Preferred Reporting Of Case Series in Surgery (PROCESS) guidelines, *Int. J. Surg.* 84 (2020) 231–235, 1 déc.
- [6] B. Hicham El, Abdelmalek, B. El Bachir, M. Karim Ibn, M. Khalid, T. Khalid Ait, Pneumatose kystique iléale révélée par un volvulus du grêle, *Pan Afr. Med. J.* 6 (2010) [Internet]. 12 août [cité 12 juill 2020]. Disponible sur: <https://www.ncbi.nlm.nih.gov/pmc/articles/PMC3063497/>.
- [7] I. Serraj, L. El Kihal, R. Mohcine, A. Essaid, Pneumatose kystique intestinale avec ascite: association exceptionnelle, *Acta Endosc.* 36 (3) (2006) 357–362, juin.
- [8] K.I.M. Hassani, I. Yazough, S.A. Laalim, I. Toughrai, K. Mazaz, La pneumatose kystique intestinale secondaire à un ulcère peptique: à propos de trois cas, *Pan Afr. Med. J.* (2015) [Internet]. 17 févr [cité 12 juill 2020]; 20. Disponible sur: <https://www.ncbi.nlm.nih.gov/pmc/articles/PMC4919673/>.
- [9] E. Schröpfer, T. Meyer, Surgical aspects of pneumatosis cystoides intestinalis: two case reports, *Cases J.* 2 (1) (2009) 6452.
- [10] D. Kancherla, S. Vattikuti, K. Vippera, Pneumatosis cystoides intestinalis: is surgery always indicated? *Cleve Clin. J. Med.* 82 (3) (2015) 151–152, mars.
- [11] H. Pülat, G. Çelik, M.Z. Sabuncuoğlu, M.F. Benzin, O. Karaköse, R. Çetin, Pneumatosis cystoides intestinalis: a rare cause of intraabdominal free air, *Turk. J. Surg.* 33 (4) (2015) 315–317, 14 juill.
- [12] P. Devgun, H. Hassan, Pneumatosis cystoides intestinalis: a rare benign cause of pneumoperitoneum, *Case Rep. Radiol.* 2013 (2013) 1–3.
- [13] L. Foessel, V. Faucher, M. Lujic, J. Charton, M.-Y. Jeung, C. Roy, Tour d'horizon de la pneumatose intestinale, *Feuill Radiol.* 55 (2) (2015) 67–81, avr.
- [14] L.M. Ho, E.K. Paulson, W.M. Thompson, Pneumatosis intestinalis in the adult: benign to life-threatening causes, *AJR Am. J. Roentgenol.* 188 (6) (2007) 1604–1613, juin.
- [15] P. Guillem, Radiologic pneumoperitoneum without perforation of a hollow viscus, *J. Chir. (Paris)* 139 (1) (2002) 5–15, févr.

#### Open Access

This article is published Open Access at [sciencedirect.com](https://www.sciencedirect.com). It is distributed under the [IJSCR Supplemental terms and conditions](#), which permits unrestricted non commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.