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RESEARCH ARTICLE

CASE REPORT OF A CHALLENGING CYSTIC INTESTINAL PNEUMATOSIS OF THE SMALL GUT ASSOCIATED WITH ULCER PERFORATION

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Abstract

Introduction:-Cystic Intestinal Pneumatosis represents a challenging clinical condition with various clinical onsets and complications. Altogether, CIP is prone to misdiagnosis and or mistreatment due to delayed management. The aim of our study is to report the challenging diagnosis of a pneumatosis intestinalis clinical case, the therapeutic strategy we chose to adopt and the outcome.

Material and Methods:-Forty-five-year-old manwith no past medical and surgical history who presented aggravated acute abdominal pain and absence of bowel movement associated with vomitus in the last 5 days earlier. Physical examination unveiled unstable patient with cardiovascular choc syndrome and generalized abdominal contraction. Prompt medical care was started with abundant IV fluids repletion and antibiotics. Abdominal X-ray without barium showed the presence of air in the abdominal cavity. Diagnosis of pneumoperitoneum was confirmed, and due to the unstable condition, we decided to undergo surgical assessment first without abdominal CT scan. Therefore, laparotomy was carried on. We found perforated anterior duodenal ulcer associated with cystic bubbles filled with air along the gut wall. We decided to preserve the affected segment with cystic pneumatosis as there was no significant signs of inflammation, perforation nor ischemia. Post-operative course was uneventful, and the patient was discharged on day 5. Long-term follow-up at 4 months was uneventful.

Discussion:-The abstract underlined the misleading radiology imaging of CIP, highly like the one of pneumoperitoneum. The etiology behind Pneumatosis intestinalis is yet to be understood. Multiple theories have been described, including mechanical disruption of mucosa, the spread of intraparietal gas to operate via lymphatic drainage, pulmonary pathogenesis and bowel necrosis, or finally idiopathic. Although it is a benign condition there arepotentially associated complications requiring both adequate diagnosis and management.

Conclusion:-To the best of our knowledge, we report the third case in literature of associated cystic intestinal pneumatosis with perforated duodenal ulcer.Intestinal preservation in cystic intestinal pneumatosis seems to be the best suited approach, both in an elective setting and

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emergency setting. Though more clinical data and extend follow-ups on this matter should be held in order to affirm the safety of this approach.

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Introduction:-

Cystic Intestinal Pneumatosis or Pneumatosis cystoids intestinalisis defined by the presence of gas bubbles within the intestinal wall (1). Though it has been abundantly described in literature, it is still a challenging clinical condition. Indeed, Cystic Intestinal Pneumatosis can manifest under various clinical onsets, such as superior mesenteric ischemia, intestinal perforation due to wall necrosis, intestinal obstruction or necrotizing enterocolitis (1). Hence it is associated with multiple complications. Pneumatosis cystoids intestinalis is also characterized by an uncertain etiology that is yet to be understood. All these factors taken together create a challenging entity prone to misdiagnosis and or mistreatment due to delayed management. Radiologic exploration techniques are the gold standard in diagnosis, though endoscopy is gaining interest in adequate pre-operative evaluation (2). Radiological techniques include abdominal X-ray andabdominal CT scan. Radiologic assessment findings are described as typical pattern of gas bubbles in the wall of the intestine (1). The aim of our study is to report the challenging diagnosis of a pneumatosis intestinalis clinical case, the therapeutic strategy we chose to adopt and the outcome.

Material And Methods:-

We admitted a 45-year-old man who presented to the emergency room with aggravated acute abdominal pain in the last 5 days earlier. The patient described clinical evolution to be marked with absence of bowel movement associated with vomitus. The patient reported no past surgeries, and medical history was uneventful. Physical examination revealed an unstable patient with cardiovascular choc syndrome made of tachycardia, low blood pressure and diaphoresis.Abdominal assessment found generalized abdominal contraction. Prompt medical care was started with abundant IV fluids repletion and antibiotics. Abdominal X-ray without barium showed the presence of air in the abdominal cavity (figure 1). Diagnosis of pneumoperitoneum was confirmed, and due to the unstable condition, we decided to undergo surgical assessment first without abdominal CT scan. Therefore, laparotomy was carried for suspected perforation of the gut. Indeed, upon initial abdominal exploration we found perforated anterior duodenal ulcer (figure 2) associatedwith cystic bubbles filled with air along the gut wall (figure 3). We carried on with sutures of the ulcer and abundant peritoneal lavage. We decided to preserve the affected segment with cystic pneumatosis as there was no significant signs of inflammation, perforation nor ischemia. Histology analysis confirmed the presence of cystic pneumatosis of the small gut without any sign of intestinal wall perforation. Post-operative course was uneventful, and the patient was discharged on day 5. Long-term follow-up at 4 months was uneventful.

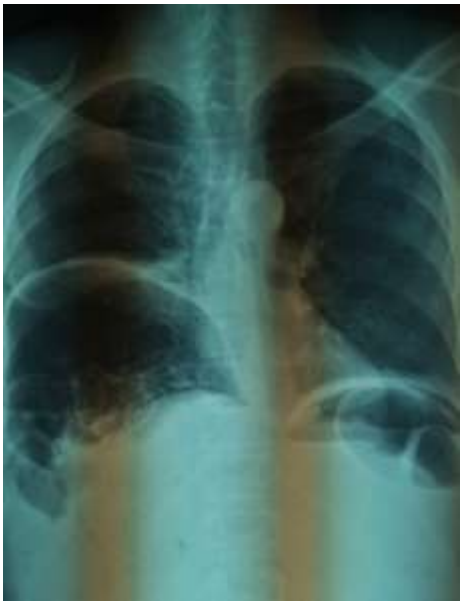

Figure1:-Abdominal X-ray without barium showed the presence of air in the abdominal cavity

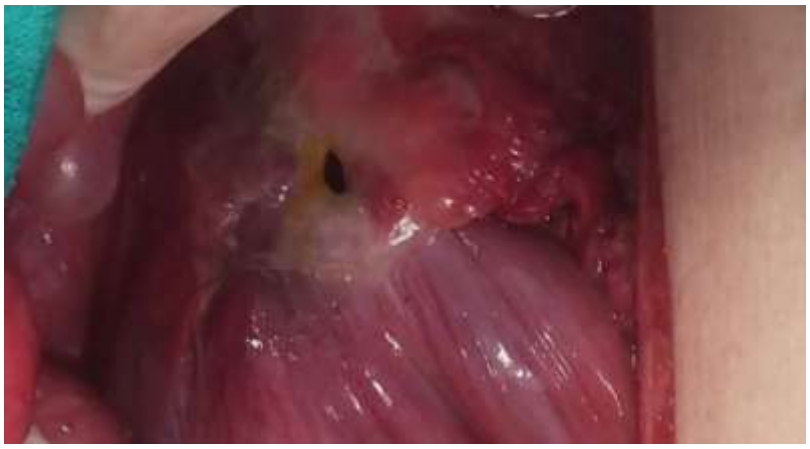

Figure 2:-Anterior duodenal ulcer perforation

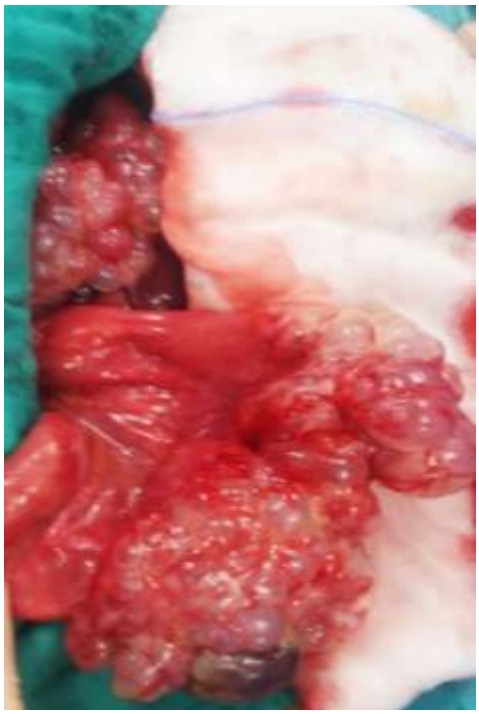

Figure 3:-cystic bubbles filled with air along the gut wall

Discussion:-

Pneumatosis Intestinalis, or Cystic Intestinal Pneumatosis is defined as a collection of gas in bullous cysts within the wall of the gastrointestinal tract. And although this entity has been only recently heavily described, it was first described since 1730 under multiple appellationsuch as “gas cysts of the intestine” and “pneumatosis cystoids intestinalis” (3). 1926 marked the first manuscript in literature that manifested the challenge of this pathologic entity, in a French publication that was published in both Radiology and the American Journal of Roentgenology and Radium Therapy (3). The abstract underlined in fact that the appearance of the abdomen is most often of timesidentical to the one of pneumoperitoneum (3, 4).

The etiology behind Pneumatosis intestinalis is yet to be understood. Multiple theories have been described, including mechanical disruption of mucosa. According to the later theory, a break in the integrity of the gastric mucosa allowed incarceration due to pressure of luminal gas and spread with the peristalsis from the stomach to other portions of the gastrointestinal tract (3). A second theory implies the spread of intraparietal gas to operate via lymphatic drainage, but this concept was abandoned in favor of spread along the mesentery (3,5). Other theories

have been described throughout history, including pulmonary pathogenesis and bowel necrosis (3). Finally, idiopathic causes have been retained and justified with the lack of evidence otherwise. It is a benign condition of parietal bowel cysts filled with gas, but due the potential associated complications as well as the multiple etiologies it can become an emergency requiring both adequate diagnosis and management.

To the best of our knowledge, we report the third case in literature of associated cystic intestinal pneumatosis with perforated duodenal ulcer. The first two were published in 1998 and 2012 (6, 7). Interestingly, Ohashi H et al reported a young 31-years old male diagnosed with both entities during a routine health check-up. As opposed to the acute onset of symptoms experienced by our reported case, Ohashi H et al had the opportunity to assess the patient using a CT scan exploration and upper gastrointestinal endoscopy, which confirmed the diagnosis of PCI associated with non-perforated gastric ulcer. This allowed them to manage in a more non-invasive strategy and reported a positive follow-up. Due to the emergent nature of our case and the prompt surgical management, we decided to carry surgical treatment of the imminent issue of perforated ulcer and to preserve the concerned segment of the small intestine. In fact, intestinal preservation in cystic intestinal pneumatosis seems to be the best suited approach, both in an elective setting and emergency setting. Though more clinical data and extend follow-ups on this matter should be held in order to affirm the safety of this approach.

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