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An Organ-Preserving Serosectomy Approach in the Management of Extensive Pneumatosis Cystoides Intestinalis: Kesmer Method

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Case Report

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Abstract

Pneumatosis cystoides intestinalis (PCI) is a rare condition characterised by the formation of gas-filled cysts in the intestinal wall. This article presents a novel organ-preserving surgical technique, previously unreported in the literature, in a patient diagnosed with PCI. A 27-year-old man presented with abdominal pain and distension caused by PCI, which had developed as a result of methamphetamine abuse. Radiological images revealed air-fluid levels, free air in the abdomen and septation-like findings. During emergency surgery, widespread cysts were observed in the small intestine. All cysts were removed via serosectomy (Kesmer method) without any bowel resection. This method, prevented the potential development of short bowel syndrome; furthermore, it eliminated the risk associated with multiple small bowel anastomoses. The postoperative period was uneventful, and the patient experienced only two episodes of ileus during the 18-month follow-up period. The episodes resolved with conservative treatment. We believe that the described serosectomy method may yield successful results in selected PCI cases to avoid bowel resection.

This technique, which preserves organ integrity, raises the question: ‘Could it serve as a model for the removal of tumour implants on the small intestine?’

Introduction

PCI is a rare condition characterized by the presence of multiple gas-filled cysts in the intestinal submucosa and/or subserosa.[1], [2] It can occur anywhere along the gastrointestinal tract from the esophagus to the rectum, including the mesentery. It is most commonly seen in the intestines. It is one of the non-surgical causes of pneumoperitoneum in neonates.[3] It is sometimes difficult to distinguish between PCI and pneumatosis intestinalis. Pneumatosis intestinalis is a life-threatening condition compared to PCI. The incidence of PCI has been reported to be 0.03%. It can be seen in all age groups. [4]It is divided into primary (idiopathic) type and secondary type. Primary type is seen in 15% and secondary type in 85% of cases. Primary PCI was first described on cadaver by Duo Vernoi in 1730. Then Koss described secondary PCI in 1952.[5] The primary type characterizes cystic-appearing air sacs and is chronic, benign and idiopathic, while the secondary type, caused by various predisposing factors, refers to radiologic findings of intramural gas with a linear, microvesicular or more circumferential appearance.[4]

Clinical presentations are very heterogeneous. PCI is usually asymptomatic, but some cases may present with abdominal pain, diarrhea, constipation, rectal bleeding, mucorrhea, malabsorption, weight loss and distension. The range of symptoms may vary depending on the location of the gas-filled cysts. [6] Some of the cases may present with spontaneous pneumoperitoneum due to rupture of the cysts, while others may present with an acute abdomen-like presentation due to air retention.

The etiology of the disease is not clear. Bacterial, chronic inflammation, mechanical, previous surgery, nutritional disorders, intestinal motility disorders, ischemic events, trauma, immune and pulmonary hypotheses have been proposed.[2], [4]

Current literature on the management of PCI is generally shaped by the location of the cysts and the clinical presentation.Computed abdominal tomography (CT) is considered the gold standard for diagnosis. It was first described by Baumann-Schender in 1939 according to radiologic findings.[7] Although it is said that the diagnosis of the disease is made by radiologic imaging or incidental, studies and case reports show that patients are mostly diagnosed during emergency operation.

Surgical intervention may be required in complex cases or in the presence of acute abdominal symptoms. The surgical approach generally requires resection of the affected bowel segment followed by anastomosis. However, intestinal resections performed in cases of PCI involving extensive segments carry the risk of ‘short bowel syndrome’, complications related to malabsorption, and increased surgical morbidity due to multiple anastomoses. Modern gastrointestinal surgical practices are increasingly moving towards ‘organ- and tissue-preserving’ models due to the long-term metabolic and functional complications associated with extensive bowel resection. In patients where the pathology is widespread but confined to the serosa, segmental resections place an unnecessary burden on the patient; they can frequently lead to problems with high morbidity and mortality, such as short bowel syndrome, malabsorption and anastomotic leaks. The current literature still contains gaps regarding bowel-preserving surgical procedures. A standard and reproducible surgical technique that can eliminate widespread serosal pathology whilst definitively preserving the serosa, mucosa, muscularis mucosae and muscularis propria—without the need for resection—has not yet been defined. Consequently, even in emergency surgical interventions, aggressive bowel resections may be performed for conditions that are merely superficial, such as in PCI patients; this clearly highlights a critical shortcoming in current organ-preserving techniques."

Particularly in cases of PCI with extensive involvement, the removal of cysts without disrupting intestinal continuity is one of the major challenges. The need for an ‘organ-preserving’ surgical approach that eliminates the risks of morbidity and mortality associated with bowel resection remains a gap in the literature.

This article presents a new serosectomy technique, in the literature, applied for the first time in a case diagnosed with widespread PCI. This method, which allows for the total excision of cysts without the need for bowel resection, represents an important alternative in terms of eliminating the risk of short bowel syndrome and preventing anastomotic complications. This technique may have the potential to serve as a new model not only in the management of PCI but also in the resection of some tumour implants on the bowel surface.

Case

A 27-year-old male patient, who was being treated for methamphetamine addiction, was referred to the emergency department of our hospital from a psychiatric hospital because of abdominal pain that had been persistent for 5 days. Physical examination revealed abdominal distension and tenderness. Abdominal PA erect view shows air fluid levels (Fig. 1). CT scan is performed to determine the etiology.

When air fluid levels, free air and grape-cluster appearance were detected in the abdomen on CT scan, the patient was considered to have delayed perforation and emergency surgery was decided (Fig. 2). In addition, the patient's acute phase reactants and renal functions were measured to be elevated. (Laboratory values; Hgb: 16.2 g/dL, Wbc: 13/µL, Plt: 848/µL, Urea;65 mg/dL, Krt: 1.68 mg/dL)

On laparotomy, there were air-filled bubbles on the small intestine, almost along the entire ileum, with intact areas of 30–40 cm between them and covering the intestine with an average length of 25 cm, reaching a size of 1x1 cm (Fig. 3). No intestinal content or dirty fluid was seen in the abdomen. Given the spread of the disease, segmental resection in four different regions of the small intestine was avoided in order to prevent short bowel syndrome, which may develop following long and multiple segmental resections, and the associated risks of multiple anastomoses.There was a concern that abdominal pressure would be created if the abdomen was closed without intervention.

Firstly, it was planned to shrink the cysts by cauterization (Video1). After some cysts were cauterized, it was seen that there were also cysts in the mesentery of the small intestine and they could be resected. Cyst resection was performed with Metzenbaum scissors as serosectomy in all cyst areas (Video 2) (resection of cysts with serosectomy without bowel resection in PCI case; In this patient, the cystic lesions were predominantly subserosal, allowing a serosectomy-based excision approach). No full-thickness bowel injury or enteric leakage was observed intraoperatively. A total of 80 cm of small bowel cyst resection was performed with this method. (Video 3; View of the small intestine after resection and specimen) No leakage and septic condition was observed in the post operative period. The patient was discharged uneventfully. Eighteen-month follow-up revealed 2 episodes of ileus. It regressed with conservative treatment. Eighteen months later, no new cyst formation was seen on CT scan. During the patient’s follow-up, a gastroscopic examination was performed due to dyspeptic symptoms, and pyloric stenosis was observed.

Discussion

PCI is characterized by the presence of gas-containing cysts in the intestinal submucosa or subserosa. PCI is more common in middle-aged male patients (45.3 ± 15.6 years) (2.4:1 male/female).[2], [4] PCI has been reported to occur from the esophagus to the rectum. It is most common in the intestines. The large intestine is involved in 46% of cases (sigmoid colon is predominantly involved), the small intestine in 27%, the stomach in 5%, and both the small intestine and large intestine together in around 7%.[8] Rectum and duodenum are rare. In our case, PCI was seen in 4 separate ileal segments 20 cm apart.

A wide variety of reasons for PCI have been presented in the literature. Many articles on etiology are case reports. PCI appears to be an outcome and not a disease in itself.[9] Although unproven, there are three hypotheses for the etiology of PCI, with gas entering the tissue through a defect in the epithelial layer of the intestine. However, gas areas are usually in non-ulcerated areas.[2]

Theories on the pathogenesis of PCI;

-Mechanical theory: Increased intraluminal pressure in the intestine causes mechanical damage and mucosal tearing, leading to migration of gas from the gastrointestinal cavity to the intestinal wall

-Bacterial theory: aerogenic bacteria penetrating the intestinal mucosal barrier ferment in the intestinal wall and produce gas

-Pulmonary theory: emphysema occurs after alveolar rupture due to chronic lung diseases. This causes gas to pass through the aorta and mesenteric blood vessels into the intestinal wall.[10]

In our case, the underlying cause seems to be methamphetamine addiction. Although a direct relationship between methamphetamine abuse and PCI has not been clearly defined, there may be an indirect relationship.[11] The adverse effects of amphetamines on the intestine (vasospasm, ischemic damage, impaired motility, increased intraluminal pressure and inflammation) may contribute to the development of PCI. Amphetamine damage to intestinal tissue may predispose to the development of this rare condition.[12], [13] Among the mechanical causes, many factors such as peptic ulcer, pyloric stenosis and endoscopic trauma have also been proposed.[1] The fact that pyloric stenosis was detected on endoscopy performed because of dyspeptic complaints during the follow-up of our case supports this situation. It is not clear, but we think that ischemic damage and vasospasm due to methamphetamine may cause pyloric stenosis. PCI was also detected in a patient operated for pyloric stenosis in the study of Pülat et al.[1]

Some of the cases present with spontaneous pneumoperitoneum without acute abdomen due to rupture of the cysts. In some cases, an acute abdomen-like presentation may occur due to air retention. In clinical practice, there are many cases in which patients with symptoms of pneumoperitoneum are diagnosed with gastrointestinal perforation and subjected to unnecessary surgical intervention, as in our case. Although it is said that the diagnosis is usually made incidental to imaging, studies and case reports show that the diagnosis is more often made intraoperatively. CT is the most useful modality to diagnose PCI and other intraabdominal pathologies. Cysts usually appear on radiographs as grape cluster-like radiolucent shadows close to the intestinal lumen.[14] If physical examination and imaging findings are suspicious, surgical exploration may be required to confirm the diagnosis of PCI.

Publications about PCI in the literature are mostly case reports and no specific treatment is specified. In asymptomatic and nonspecific symptomatic PCI cases, treatment may be conservative. Hyperbaric oxygen, probiotic preparations, endoscopic treatments such as needle aspiration can be applied.[8]Surgical explorations usually resulted with resection.[2], [4], [7]However, in cases with extensive cystic involvement, resection carries serious risks of mortality and morbidity, such as ‘short bowel syndrome’ and anastomotic leakage.

The resection of cysts with serosectomy, small bowel resection was not performed and the patient was saved from short bowel and/or multiple anastomoses. In addition, the risk of leakage after resection and anastomosis was prevented. The cysts were blasted with cautery before serosectomy. A similar treatment, endoscopic fenestration, was first performed by Takahashi et al.[15] In our case, endoscopic

fenestration was not appropriate because the cysts were located in the small intestine. It also could not be evaluated endoscopically.

Various studies on small bowel-sparing approaches exist in the literature. However, the serosectomy approach we have described may offer a highly specific and innovative approach to this field.

Park et al. have described a technique involving the peeling of the serosa without entering the intestinal lumen and the repair of the defect, aimed at preventing resection of the rectosigmoid colon in advanced-stage ovarian cancers that have invaded the intestinal wall but are confined solely to the serosal layer. [16] Sugarbaker's method, which involves using scissors to remove small implants located on the small intestine during cytoreduction, follows a similar dissection logic to the method we have described. We believe that if the spontaneous air bubbles beneath the serosa observed during PCI can be induced iatrogenically in patients with implants on the small intestine, implant resections may proceed more successfully. [17]

Conclusion

The serosectomy procedure (Kesmer method) performed in this PCI case appears to be a first in the literature. In previous studies and case reports, patients have either undergone resection or the operation has been concluded without any surgical intervention. Although research in this area is limited, we believe that there may be different variants of PCI with the same radiological and surgical findings, and that some of these could be treated with serosectomy without the need for bowel resection. We also hope that this method may inspire future research into bowel-preserving serosal dissection techniques. (Fig. 4: Illustration of the hypothesis regarding the removal of tumour implants on the serosa of the small intestine by serosectomy. ' İatrogenic subserosal injection hypothesis')

Declarations

Conflict of interest: The authors declare that there is no conflict of interest.

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Ethical statement: Written informed consent was obtained from the patient of this case report.

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Figures



Figure 1

Direct abdominal radiograph; Air fluid levels.

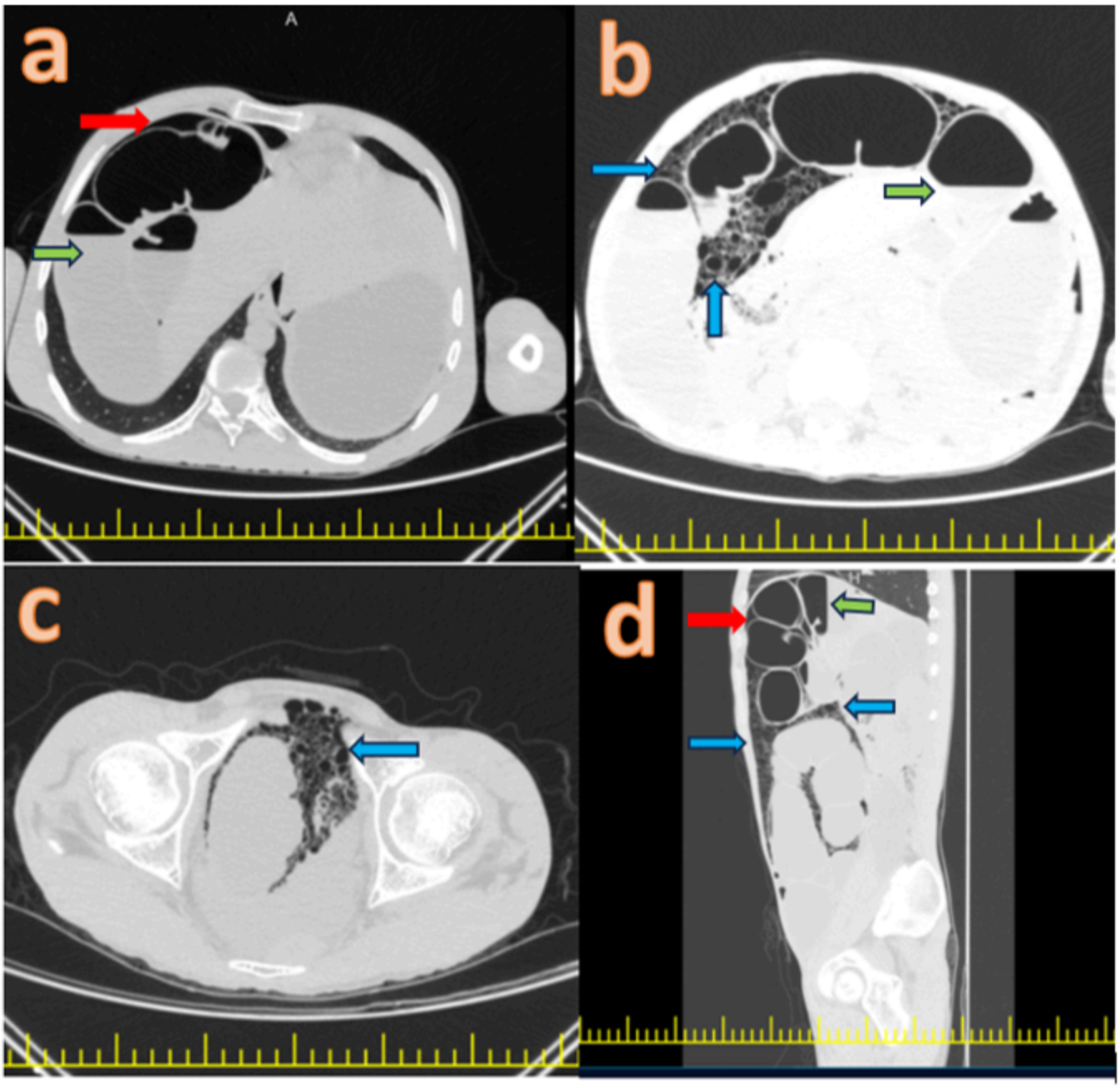


Figure 2

CT images (a,b,c,d,); Red arrows: intra-abdominal free air, Blue arrows: PCI appearing like septa, Green arrows: Air fluid levels.

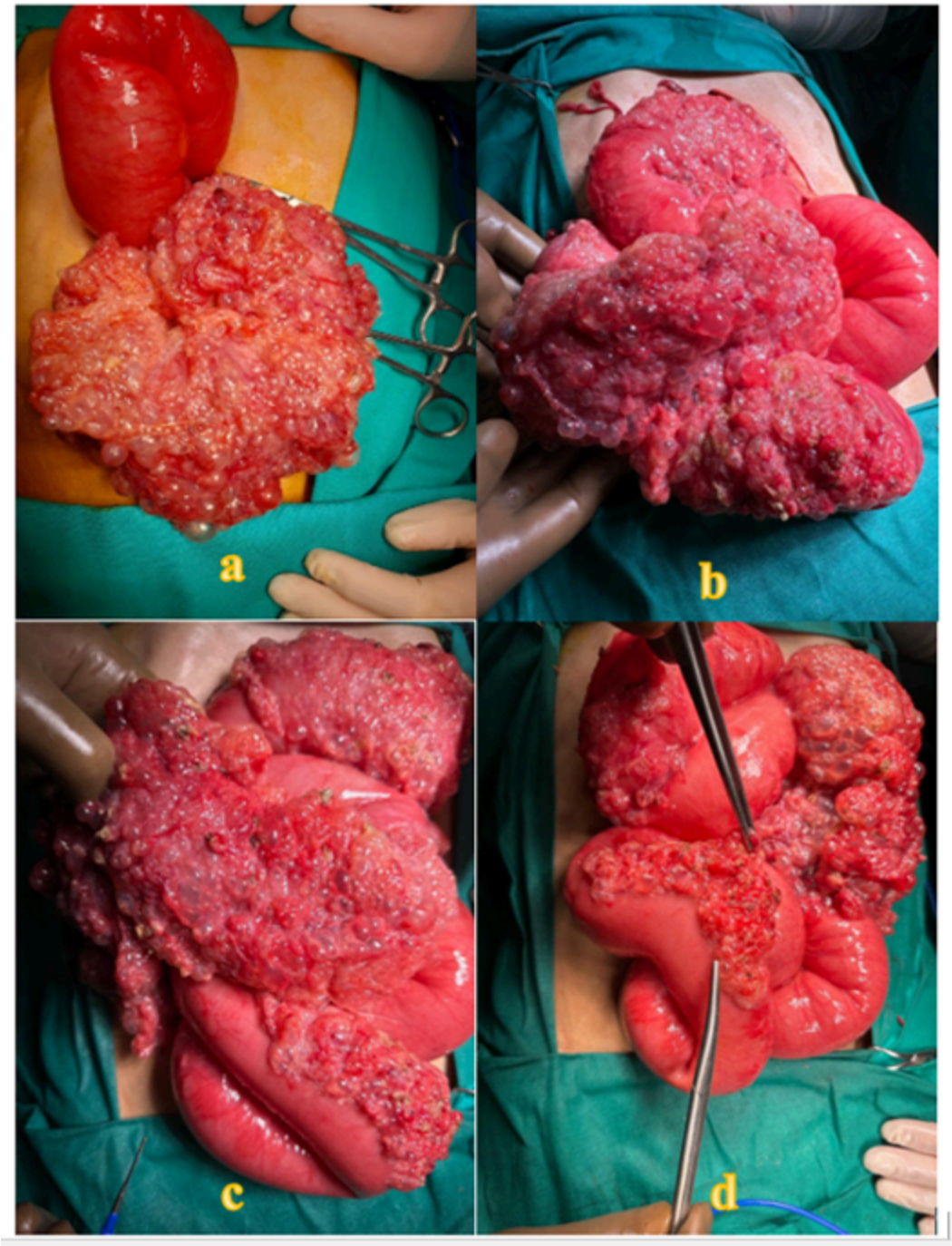


Figure 3

PCI seen on the small intestine during laparotomy (a,b,c,d,)

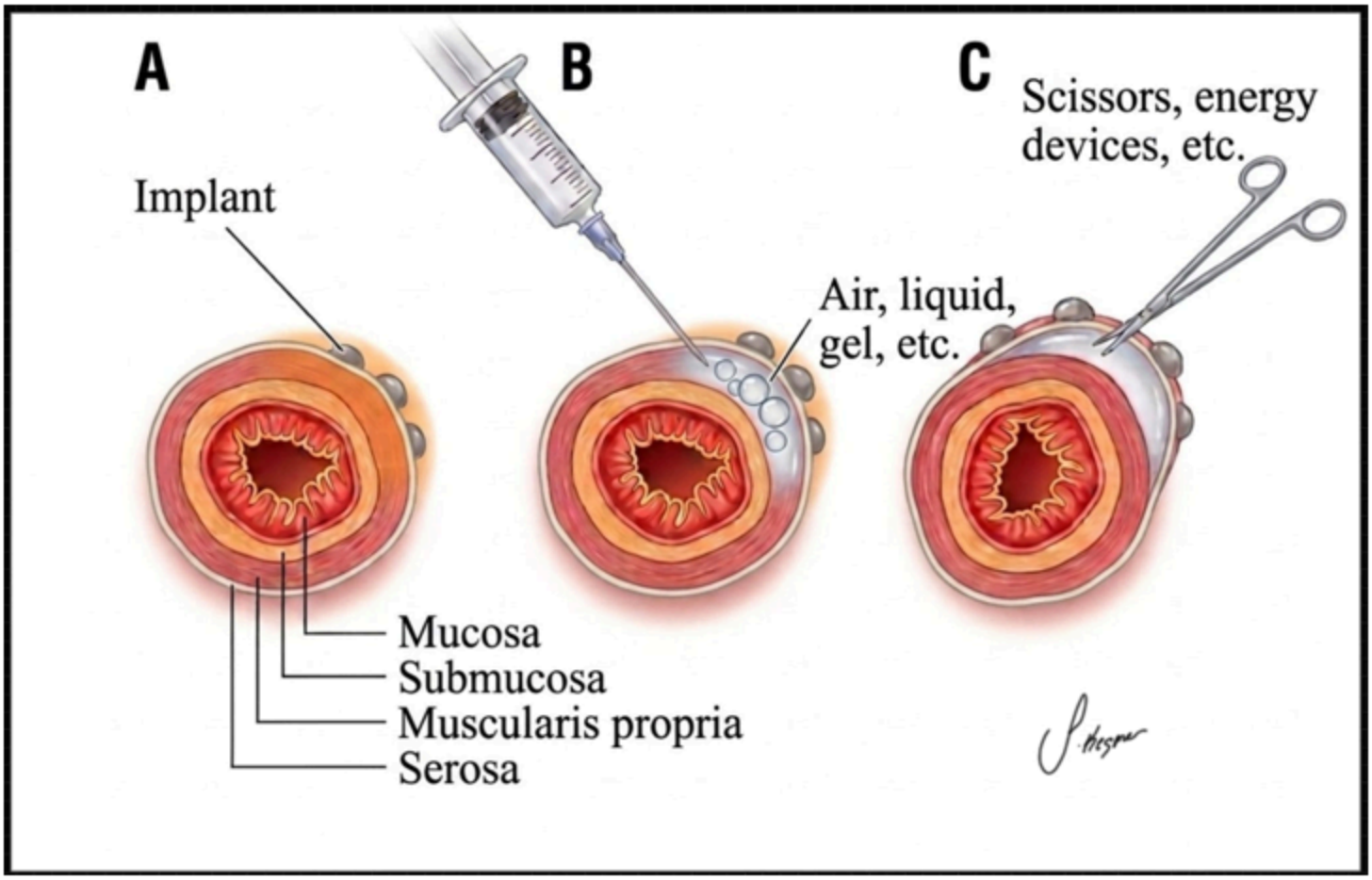


Figure 4

Illustration of the hypothesis regarding the removal of tumour implants on the serosa of the small intestine by serosectomy. A: A loop of small intestine containing implants B: The injection of a substance (e.g. air, fluid, gel, etc.) into the subserosal space to elevate the serosa C: Dissection of the exposed serosa from the subserosal space using a surgical instrument (e.g. scissors, energy devices or a new device to be developed for dissection).

Supplementary Files

This is a list of supplementary files associated with this preprint. Click to download.

- Video1.avi
- video2.avi
- video3.mp4