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Abdominal cocoon syndrome

Rare cause of intestinal obstruction—Case report and systematic review of literature

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Abstract

Background: Abdominal cocoon or sclerosing encapsulating peritonitis is an uncommon condition in which the small bowel is completely or partially encased by a thick fibrotic membrane. Our study presents a case of sclerosing encapsulating peritonitis and conducts a literature review.

Methods: A bibliographic research was conducted. Our research comprised 97 articles. Gender, age, symptoms, diagnostic procedures, and treatment were all included in the database of patient characteristics.

Case presentation: A 51-year-old man complaining of a 2-day history of minor diffuse abdominal pain, loss of appetite, and constipation was presented in emergency department. Physical examination was indicative of intestinal obstruction. Laboratory tests were normal. Diffuse intraperitoneal fluid and dilated small intestinal loops were discovered on computed tomography (CT). An exploratory laparotomy was recommended, in which the sac membrane was removed and adhesiolysis was performed. He was discharged on the tenth postoperative day.

Results: There were 240 cases of abdominal cocoon syndrome in total. In terms of gender, 151 of 240 (62.9%) were male and 89 of 240 (37%) were female. Ages between 20 and 40 are most affected. Symptoms include abdominal pain and obstruction signs. For the diagnosis of abdominal cocoon syndrome, CT may be the gold standard imaging method. The surgical operation was the treatment of choice in the vast majority of cases (96.7%). Only 69 of 239 patients (28.9%) were detected prior to surgery, and CT was applied in these cases.

Conclusion: Abdominal cocoon is a rare condition marked by recurrent episodes of intestinal obstruction. Surgical therapy is the most effective treatment option.

Abbreviations: ACS = abdominal, cocoon syndrome, CT = computed tomography, MRI = magnetic resonance imaging, SEP = sclerosing encapsulating peritonitis, WBC = white blood cells.

Keywords: abdominal cocoon syndrome, acute abdomen, sclerosing encapsulating peritonitis, small bowel obstruction

1. Introduction

Sclerosing encapsulating peritonitis (SEP) or abdominal cocoon syndrome (ACS) is an infrequent cause of intestinal obstruction that is typically detected during laparotomy. It is characterized by a thick fibrocollagenous membrane, partially or completely encasing the small bowel, which can extend to involve other organs. First observed in 1907, Owtschinnikow[1] developed the term peritonitis chronica fibrosa incapsulata to characterize the encasement of the intestines by a fibrous membrane. There are 2 forms of SEP: primary or idiopathic and secondary, originally identified the idiopathic type, also known as abdominal cocoon, in 1978.[2,3] In this study, we report a case of SEP and present an extended systematic review of the international literature on this rare condition.

1.1. Case report

A 51-year-old man presented to our Surgical Emergency Department complaining of a mild diffuse abdominal pain, loss of appetite, and constipation that lasted 2 days. He has no medical history and no previous surgery. He had no allergies or medication use. He reported other 2 episodes of the same symptomatology in the last year. His physical examination revealed sluggish bowel sounds, whereas on palpation, his

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The datasets generated during and/or analyzed during the current study are not publicly available, but are available from the corresponding author on reasonable request

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abdomen was tender on the right side and distended. His laboratory examination was within normal limits (white blood cell = 6.02 K/μL, neutrophil = 51.5%, hemoglobin = 14.6 g/dL, hematocrit = 41.6%, urea = 33 mg/dL, and creatinine = 0.61 mg/dL). In X-ray examination, dilated bowel loops with fluid levels were detected. Contrast-enhanced abdominal tomography (computed tomography [CT]) revealed diffuse intraperitoneal fluid with dilated small bowel loops. The patient has undergone an imaging examination (CT and magnetic resonance imaging [MRI]) few months ago due to his symptomatology advised by a gastroenterologist, which findings were the same as our CT examination (Fig. 1). In abdominal MRI, internal hernia of the small bowel in the omentum was reported, whereas small bowel was found clustered in the right upper quadrant of the peritoneal cavity (Fig. 2). Gastroscopy and colonoscopy 1 month ago had no pathological findings. A nasogastric tube was placed, and an exploratory laparotomy was advised. The surgical findings were a large quantity of ascites, and the small intestine was encapsulated in a layer of the peritoneum (Fig. 3). An adhesiolysis was performed, and the sac membrane was removed. Intestinal resection was not required. Both a granuloma, which was found in the sac and excised and part of the sac, were sent for histopathological examination. In the postoperative course, the patient had clinical signs of ileus that was conservatively managed successfully. He was discharged on tenth postoperative day with normal gastroenteric function. Six months postoperatively, the patient has a normal uneventful life. The histopathological examination reported fibrous connective tissue with collagen reaction and an infracted epiploic appendage and concluded to SEP.

2. Methods

A bibliographic research was performed using PubMed, Scopus, Google Scholar, and MedlinePlus. The search term employed was "abdominal cocoon syndrome" and "sclerosing encapsulating peritonitis." In total, 148 case reports were found, and 43 were excluded as irrelevant. In cross-examination, 8 articles were double references and were also excluded. As a result, 97 articles were included in our study.[3–41] The flow diagram of the study is shown in Figure 4. These articles were carefully studied, and a database with the patients' characteristics was made, which could be available if requested. The database included gender, age, symptoms, diagnostic methods, and treatment. An ethical approval is not required because this study is a review of the existing international literature.

In order to express results, descriptive statistics were used appropriately. Means, medians, and standard deviations were used for continuous variables. The normal distribution of quantitative data has been checked using Kolmogorov-Smirnov test. Nonparametric tests (Mann–Whitney *U* test) have been applied for the nonparametrical distribution data. Crosstab were applied for correlation investigation between nominal values. Excel 2007 (Microsoft) and SPSS 22.0 (IBM, Chicago, Illinois) were employed to statistically analyze the data.

3. Results

In total, 240 cases of ACS were recorded.

Characteristics of ACS were determined concerning gender, age, symptoms, diagnostic methods, treatment, and time of definitive diagnosis. Table 1 summarizes the number of cases fulfilling the criteria in each category.

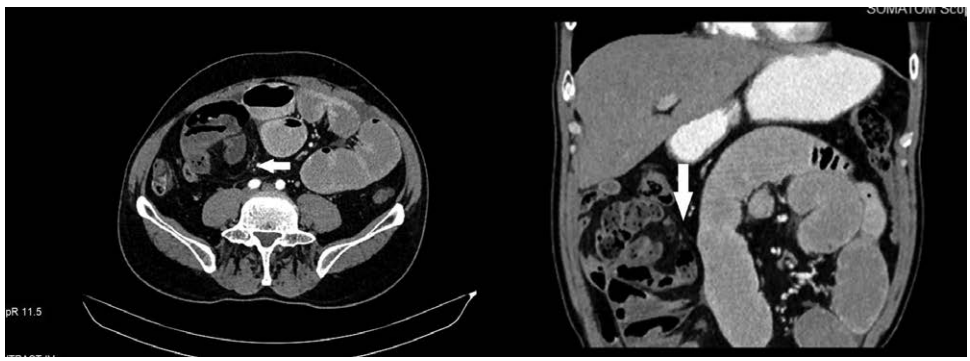

Figure 1. CT imaging dilated small bowels loops, encapsulated by a membrane (arrow) and located in the right quadrant of peritoneal cavity.

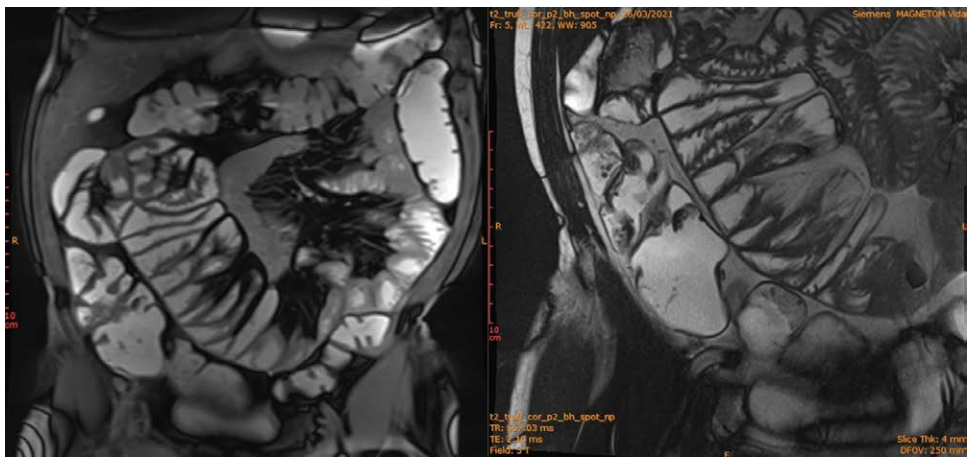

Figure 2. MRI findings, showing a part of small bowel trapped in the right peritoneal cavity.

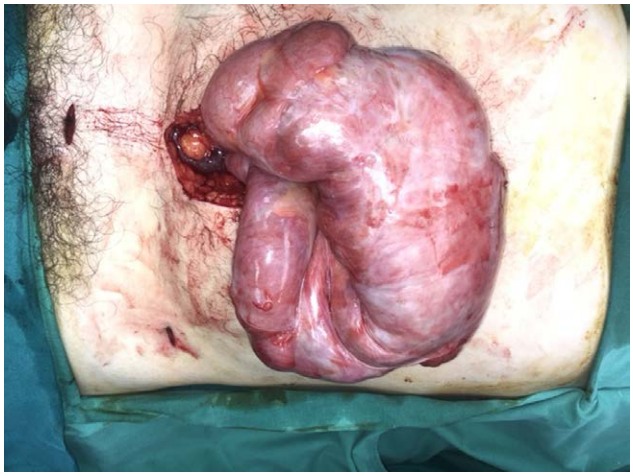

Figure 3. Surgical findings; thin membrane encapsulating small bowel.

Concerning gender, 151 of 240 (62.9%) were male and 89 of 240 (37.1%) were female, with a male-to-female ratio calculated at 1:0.58, suggesting a male predisposition (Fig. 5). Median age of the population was 39 years old, ranging from 7 to 90 years old. As shown in Figure 6, this syndrome is more frequently appeared in ages 20–40 years old. The correlation between age distribution and gender showed statistical significant results, meaning that, in male group, the age was higher than that in female group (U = 2276.5, P < .001).

The symptomatology of the ACS includes abdominal pain and symptoms of obstruction. In the majority of cases, these are chronic symptoms, persisting for several months until an urgent episode leading to the emergency department. From our statistical analysis, data were maintained from 238 patients, from whom 99 of 238 (41.6%) were complaining for diffuse abdominal pain, 134 of 238 (56.3%) has bowel obstruction symptomatology, whereas 5 of 238 (2.1%) were asymptomatic, and the ACS was diagnosed incidentally during laparotomy due to another pathology (Fig. 7).

All widely known imaging methods are applied for the diagnosis of ACS. In our systematic review, in 48 of 219 cases (21.9%), plain radiograph was applied, in 95 of 219 (43.4%) contrast-enhanced tomography (CT), and in 14 of 219 (6.4%) ultrasound examination. In high frequency of cases, plain radiograph followed by CT was applied 60 of 219 (27.4%), while in 2 cases (0.9%), CT combined with MRI was undergone (Fig. 8). The correlation between imaging method and time of diagnosis was explored, and it has been proved that when CT was applied, preoperative diagnosis of ACS was achieved (P < .001). Concluding, CT may be the gold standard imaging technique for the diagnosis of ACS (Fig. 9).

The treatment option followed in the vast majority of cases (231/239—96.7%) was the surgical operation (laparoscopy or laparotomy and adhesiolysis). Conservative treatment with antitubercular drugs or immunosuppressive medication such as colchicine, non-steroidal anti-inflammatory drugs, and steroids was chosen in 4 of 239 (1.7%) cases, while a conversion from conservative (for bowel obstruction cases—bowel rest and nasogastric decompression) to surgical treatment, due to failure of the first one to alleviate the symptomatology, was done in 4 of 239 (1.7%) cases (Fig. 10).

The time of definitive diagnosis of ACS is of great importance. Until now, ACS was suspected only intraoperatively (170/239—70.1%), with the definitive diagnosis done by the histopathological report. Only 69 of 239 (28.9%) cases were diagnosed preoperatively, in whom CT was always applied (Fig. 11).

4.Discussion

4.1.Definition

Idiopathic or secondary SEP or abdominal cocoon is a subtle inflammatory disease presenting as intestinal obstruction.[42] It is believed that SEP is a chronic inflammatory disease with an unclear cause that begins with recurring low-grade or subclinical peritonitis with no distinct abdominal indicators and develops to sclerosis and membrane formation, followed by cocoon formation.[43] This entity is characterized by a thick fibrocollagenous membrane that covers mainly the small intestine and occasionally other intra-abdominal organs such as the liver, stomach, and colon. It can be subdivided into 3 categories based on the degree of encapsulation of the organs: type I—the membrane encases a portion of the intestine; type II—the entire intestine is encased by the membrane; and type III—the entire intestine plus additional organs (e.g., appendix, cecum, ascending colon, ovaries, etc.) are encased by the membrane.[30]

4.2. Epidemiology and causes

Depending on the underlying cause, SEP is classified as either primary (idiopathic) or secondary (acquired). Although the function of cytokines and fibroblasts in the development of peritoneal fibrosis and neoangiogenesis is undeniable, there is no evidence of an underlying cause in primary SEP.[44] Idiopathic SEP is more common in tropical and subtropical regions and has a predisposition of male population with a male-to-female predominance of 2:1.[44] Pathophysiology of secondary SEP is better understood. Plethora of possible causes leading to idiopathic SEP has been mentioned, such as retrograde menstruation, retrograde peritonitis, omentum hypoplasia, and mesenteric vessel defect.[36,41] The overall incidence and prevalence of SEP are unknown due to its rarity and variability of etiologies according to Danford et al.[1] Idiopathic SEP is less prevalent than secondary SE.[23] A local or systemic cause initiates the inflammatory process in the peritoneum in secondary SEP.[44] The inflammation of peritoneum is provoked by many factors including peritoneal dialysis (PD), which is the most common cause, b-blockers, sarcoidosis, tuberculosis, ovarian tecomas, organ transplantation, and cirrhosis.[37]

4.3. Symptomatology

ACS presents as intestinal obstruction, as a result of the intestine's kinking and compression within the encasing membrane. Therefore, its signs and symptoms are typical of ileus, videlicet: acute, subacute or chronic abdominal pain, nausea, anorexia, vomiting, constipation, and weight loss. A past history of similar to the aforementioned symptoms, which resolved spontaneously, also supports the diagnosis. The average duration of symptoms was 3.9 years previous to presentation in the largest case series of idiopathic SEP, with the vast majority of patients malnourished (75%; mean body mass index, 17.5 kg/m^2). Despite the chronic, insidious character of the disease, 29% of patients required "emergency surgery" at the time of presentation, indicating that a considerable percentage of patients show up with more acute obstruction, ischemia, and even perforation.[36] In some cases, palpable mass and ascites can be detected. This modality can remain asymptomatic in the majority of cases.[27–29,31]

4.4. Differential diagnosis

Internal hernia and congenital peritoneal encapsulation are the 2 conditions to consider primarily as differential diagnosis in patients who are suspected for SEP. Internal hernias

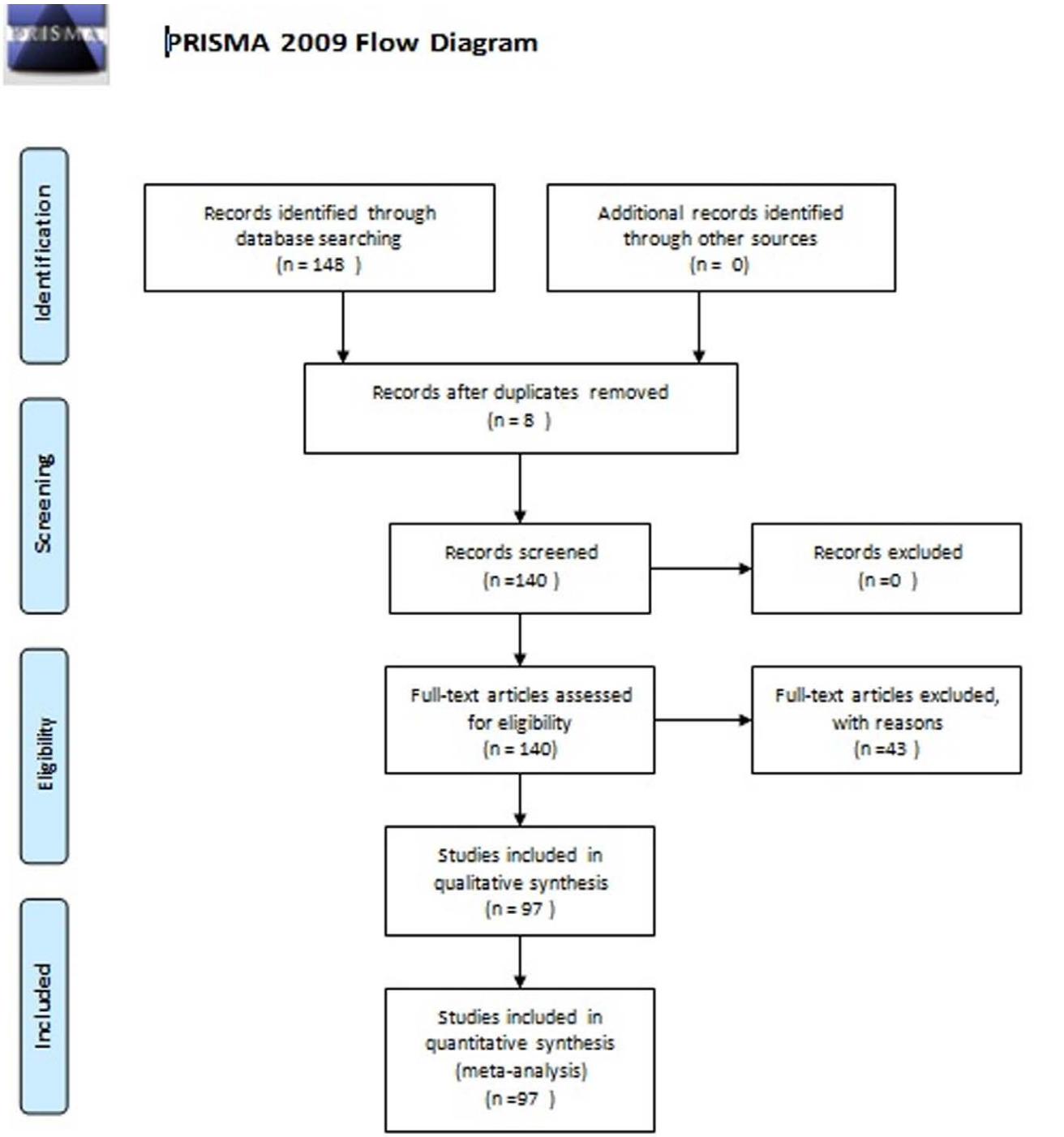

Figure 4. Flow diagram of systematic review.

Table 1

Cases of ACS fulfilling the criteria.

Categories	No of Cases	Percentage
Age	239/240	99.6
Gender	240/240	100
Symptoms	238/240	99.2
Imaging method	219/240	91.3
Treatment	239/240	99.6
Time of diagnosis	239/240	99.6

have CT findings that are similar to those seen in abdominal cocoon. In patients with internal hernias, however, no membrane-like sac can be detected.[34] Congenital peritoneal encapsulation is a developmental defect defined by a thin additional peritoneal sac enveloping the small bowel and is usually asymptomatic, whereas abdominal cocoon is characterized by a thick fibrous membrane and causes intestinal obstruction.[45] Voluminous invagination, intestinal malrotation, and other causes of peritoneal adhesion are less prevalent disorders to consider as differential diagnoses. In cases of tuberculosis-prone areas, tuberculous peritonitis should be ruled out definitively.[2,40]

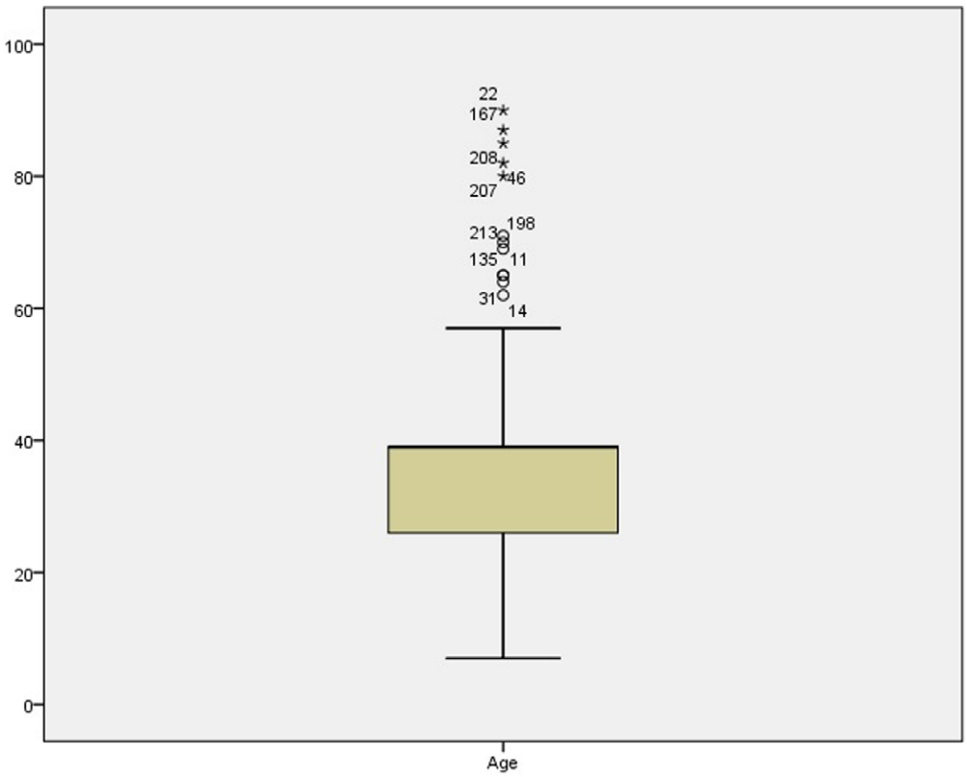

Figure 5. Age distribution.

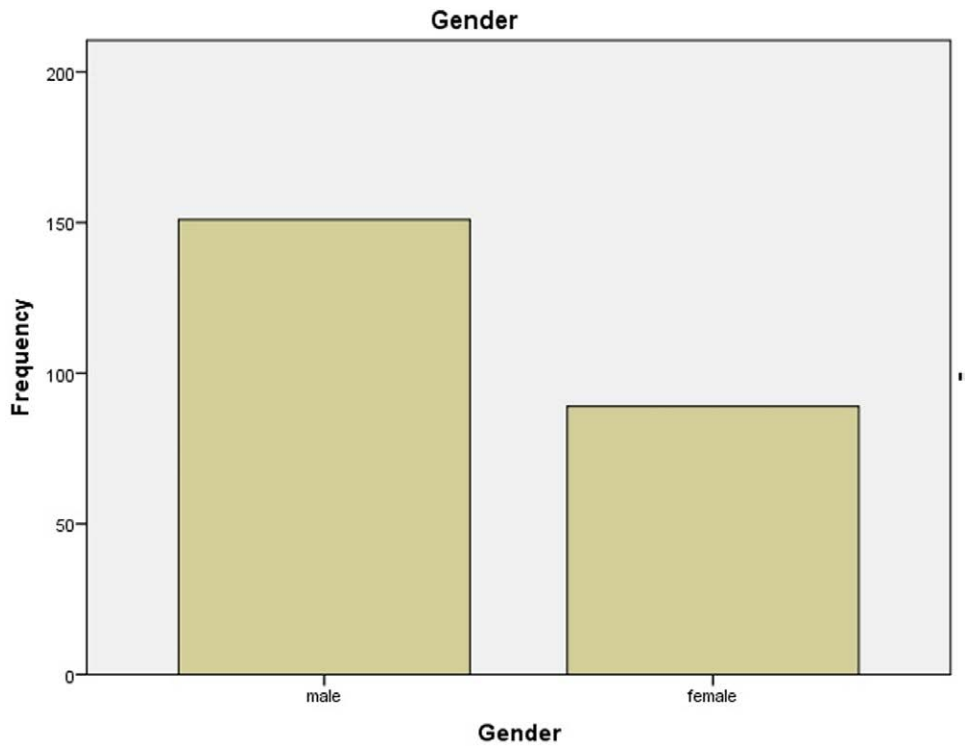

Figure 6. Bar chart of gender frequencies.

4.5. Diagnosis

Idiopathic SEP is a rare illness, and most doctors never see individuals who have it or overlook the diagnosis when they do. It is extremely difficult to make an accurate preoperative diagnosis in affected patients, and it necessitates a high index of clinical suspicion.[2] Because of the lack of specificity in the early clinical manifestations, preoperative diagnosis of SEP is difficult.[46] A high index of clinical suspicion is mandatory. Apart from patient's history and clinical presentation, imaging techniques play an important role in diagnosis of SEP. Diffuse

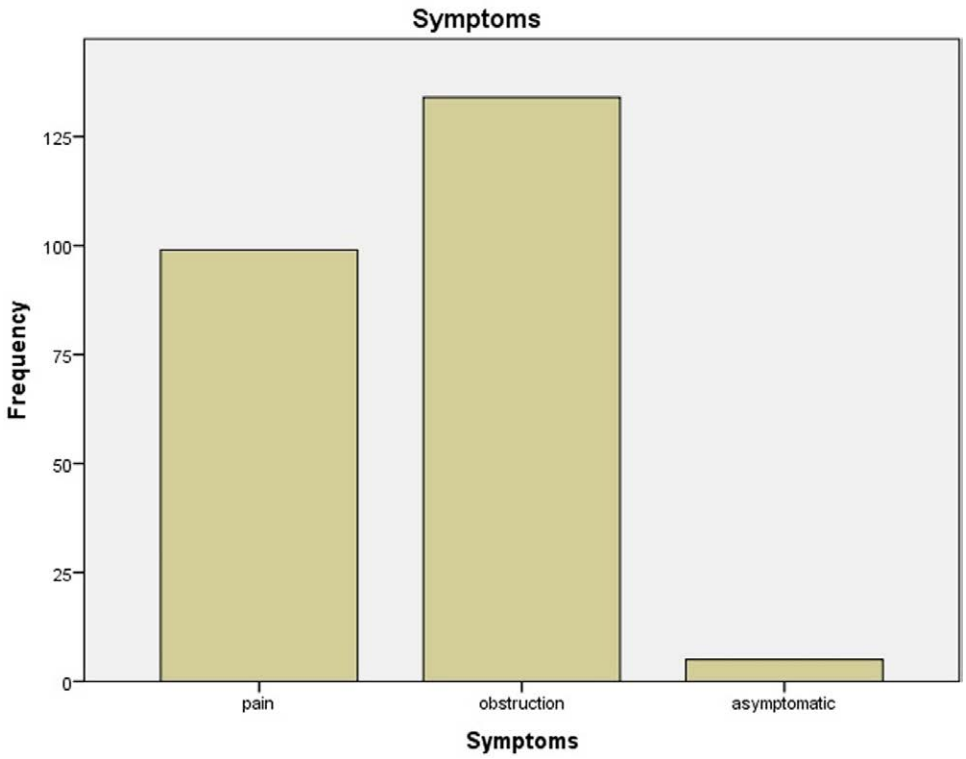

Figure 7. Bar chart of symptoms frequencies.

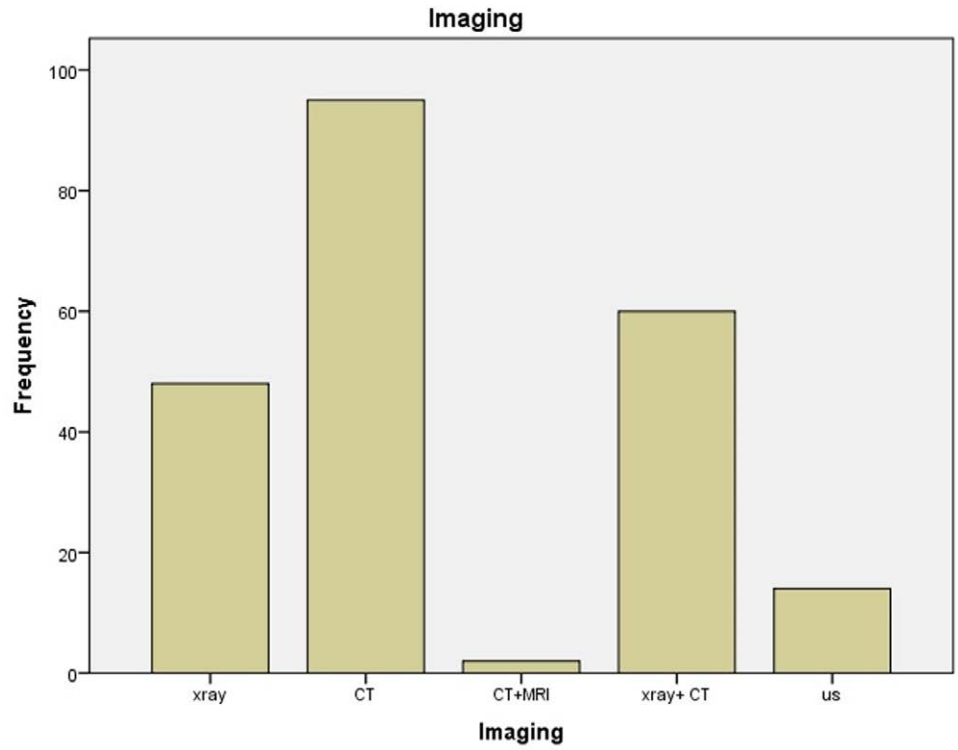

Figure 8. Bar chart of imaging method frequencies. CT = computed tomography, MRI = magnetic resonance imaging, US = ultrasound.

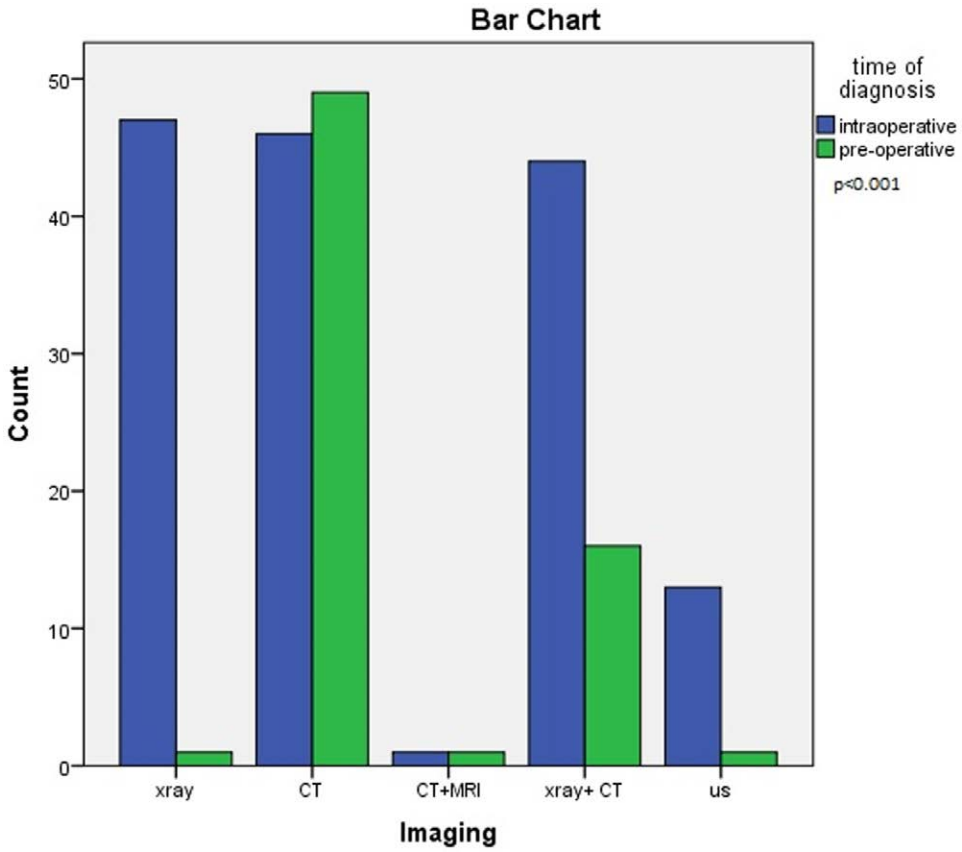

Figure 9. Correlation of imaging method and time of ACS diagnosis ($P < 0.001$). CT = computed tomography, MRI = magnetic resonance imaging, US = ultrasound.

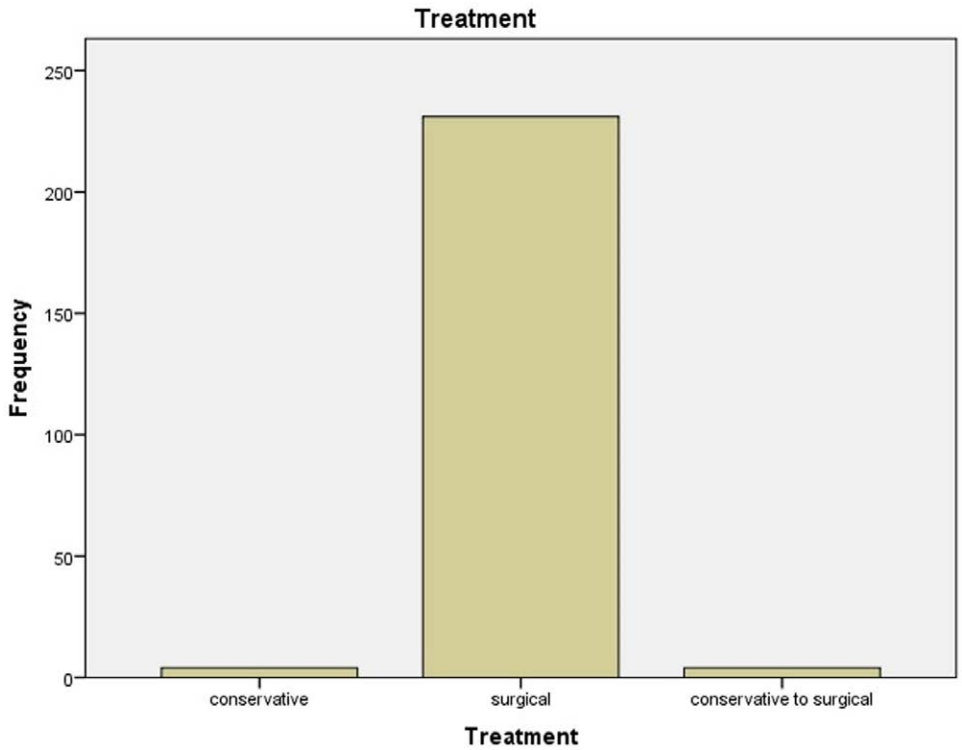

Figure 10. Bar chart of treatment frequencies.

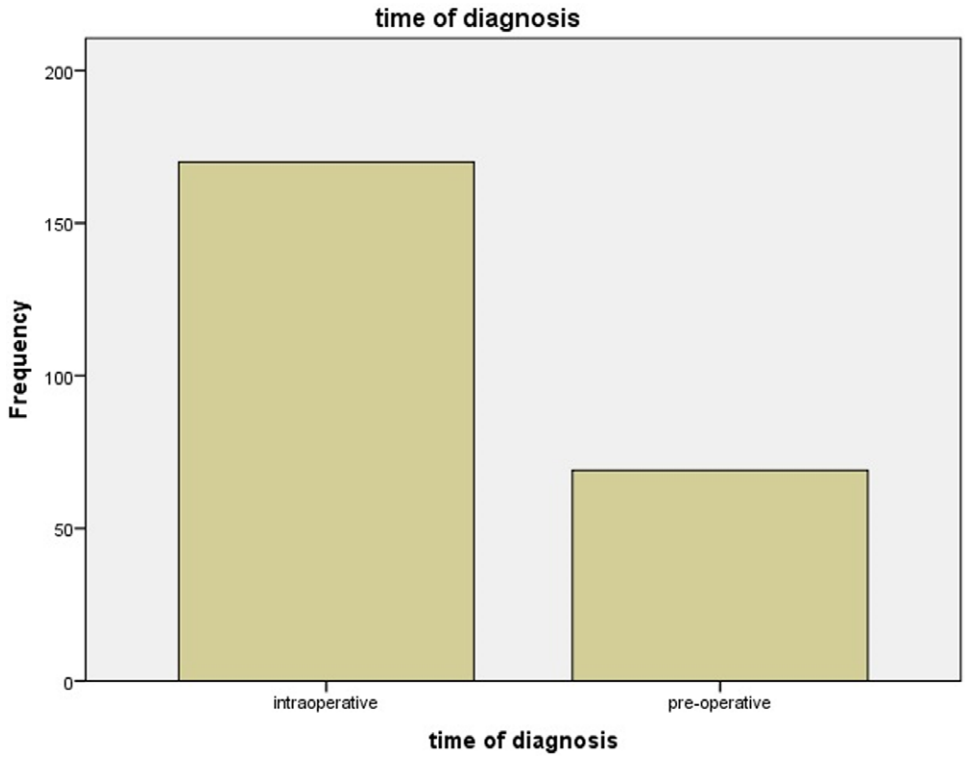

Figure 11. Bar chart of time of diagnosis frequencies.

air-fluid levels and dilated small intestine loops may be seen on abdominal X-rays.[38] Small intestines barium transit reveals collected and conglomerated bowel loops in the center of the abdomen, suggestive of the so called "cauliflower sign."[32] Additionally, prolonged transit time encourages the diagnosis.[33] Abdominal ultrasonography may reveal dilated bowel segments surrounded in a dense fibrous membrane, as well as free abdominal fluid and a thicker peritoneal layer.[38,39] On CT, which is the most useful tool in confirming the diagnosis, the appearance of small intestine segments that are conglomerated at the midline and surrounded by a thick capsule with a contrast-free periphery is a distinctive sign.[2] Peritoneal thickening is normally assessed subjectively, and no ideal threshold has been established, albeit a thickness larger than 2 mm appears to be a suitable cutoff.[47] Other radiographic characteristics comprise loculated ascites, increased mesenteric fat density, and localized or diffuse peritoneal calcification.[48] Whereas complex loculations may indicate intra-abdominal bleeding, they should raise concerns about perforation or sepsis, particularly if they contain gas.[49]

In regard to predicting the presence of SEP, no relevant biomarker has been discovered.[50] SEP laboratory findings are nonspecific and linked to infection, malnutrition, and inflammation.[51] Inflammatory cytokine values in dialysate in SEP patients were found to be greater in comparison to PD controls up to years before clinical development of SEP.[52]

The histologic features of SEP are nonspecific and can be confused with those of simple peritoneal sclerosis or infectious peritonitis.[53] Podoplanin, a transmembrane glycoprotein present in peritoneal mesothelial cells that binds inflammatory cytokines, aids in the distinction between EPS and peritoneal sclerosis and peritonitis.[54,55] However, in histopathological examination, which gives the definitive diagnosis, the peritoneum exhibits fibroconnective tissue growth, inflammatory infiltrates, and dilated lymphatics histologically, with no evidence of foreign body granulomas, giant cells, or birefringent material.[44]

4.6. Therapeutic options

4.6.1. Conservative treatment and drugs. In regard to therapeutic options, individuals with mild symptoms should be treated conservatively. This includes bowel rest, nasogastric decompression, and either enteral or parenteral nutritional support.[35] Nutritional disorders affect a large proportion of people with recurring abdominal complaints; hence, addressing them is an important part of the treatment. Improving these patients' nutritional health is critical because it may improve their response to conservative treatment or prevent surgical complications like infection and fistulae.[43] Patients who do not respond to conservative treatment may be treated with drugs such as tamoxifen, steroids, colchicine, azathioprine, and mycophenolic acid.[56] Tamoxifen plus steroids may be effective in the prevention and/or treatment of SEP, according to human studies. The pharmacological method of action of corticosteroids on SEP is uncertain. However, it is possible that it is due to both the anti-inflammatory and immunosuppressive effects of the drug. The concept of using tamoxifen to treat SEP in its early stages is particularly intriguing. Patients with long-term PD who develop ultrafiltration failure with a high transport status but who do not have the characteristic imaging signs as seen in SEP could benefit from this.[56] Additionally, individuals who experience persistent postoperative symptoms may benefit from the usage of these medications.[44]

4.6.2. Surgical intervention. Surgery for SEP is only recommended in patients who have failed conservative, medicinal therapy and, if possible, in centers with competence in such operations, due to the time-consuming, risky, and technical complexity of surgical techniques for SEP. Surgical techniques range from those aimed at curing a specific complication, such as enterolysis (ablation of fibrous tissue and lysis of adhesions), to those intended to address a particular complication, including limited lysis of adhesions or ablation of perforated or ischemic bowel.[1] Patients with severe evidence of intestinal obstruction

or who have been identified with SEP intraoperatively should undergo a laparotomy or laparoscopy. Membrane excision with adhesiolysis is the general rule, and in cases of gut injury, resection plus anastomosis with or without a protective enterostomy is the treatment of choice.[43] Peritoneal deterioration, which has been reported to worsen with the duration of PD, particularly in cases that have lasted longer than 10 years, is a crucial factor in predicting postsurgical outcomes. The capsules are weakly delineated from the intestinal wall in such individuals, and inaccurate enterolysis can easily lead to intestinal perforation. Peritoneal calcification surrounding capillaries may increase the chance of perforation on excision.[57] If the membrane on the intestinal surface can be completely removed, the chance of recurrence is quite low.[58] Placing antiadhesive compounds between the bowel loops before closing the abdomen after complete excision of the membrane may lower the likelihood of postoperative adhesions; however, the benefit of these substances for patients with partially excised membranes remains questionable.[44] In cases of secondary SEP, therapy of the underlying cause is crucial for their improvement. Thus, in idiopathic SEP cases, the diagnosis and treatment are based on surgical operation and histopathological examination of peritoneum.[44] In cases of SEP, intestinal rehabilitation program and intestinal transplantation have been applied in order to rescue patients from total parenteral nutrition complications.[59]

5.Conclusion

ACS is an inflammatory disease, characterized by a thick fibrocollagenous membrane encapsulating the small bowel. It is a rare cause of small bowel obstruction and is divided in idiopathic and secondary syndrome. It should be taken into consideration in differential diagnosis of small bowel obstruction especially in cases of repeated episodes of bowel obstruction. The gold standard for the diagnosis of SEP is laparoscopy or laparotomy, although CT has gained ground in preoperative diagnosis of SEP. Surgical treatment seems to be the effective therapeutic option in cases of conservative treatment's failure or in severe cases of SEP.

Author contributions

Angeliki Chorti, Stavros Panidis, Antonios Michalopoulos, and Daniel Paramythiotis are the surgical team that handled the case. Angeliki Cheva is the pathologist who made the diagnosis. Dimitrios Konstantinidis is a 4th year medical student dedicated to general surgery who made the literature review. Angeliki Chorti was responsible for the statistical analysis of this study.

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