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SYSTEMATIC REVIEWS

Accurate definition and management of idiopathic sclerosing encapsulating peritonitis

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Abstract

AIM: To review the literature on idiopathic sclerosing encapsulating peritonitis (SEP), also known as abdominal cocoon syndrome.

METHODS: The PubMed, MEDLINE, Google Scholar, and Google databases were searched using specific key words to identify articles related to idiopathic SEP. These key words were "sclerosing encapsulating peritonitis," "idiopathic sclerosing encapsulating peritonitis," "abdominal cocoon," and "abdominal cocoon syndrome." The search included letters to the editor, case reports, review articles, original articles, and meeting presentations published in the English-language literature from January 2000 to May 2014. Articles or abstracts containing adequate information about age, sex, symptom duration, initial diagnosis, radiological tools, and surgical approaches were included in the study. Papers with missing or inadequate data were excluded.

RESULTS: The literature search yielded 73 articles on idiopathic (primary) SEP published in 23 countries. The four countries that published the greatest number of articles were India (*n* = 21), Turkey (*n* = 14), China (*n* = 8) and Nigeria (*n* = 3). The four countries that reported the greatest number of cases were China (*n* = 104; 53.88%), India (*n* = 35; 18.13%), Turkey (*n* = 17; 8.80%) and Nigeria (*n* = 5; 2.59%). The present study included 193 patients. Data on age could be obtained for 184 patients (range: 7-87 years; mean ± SD, 34.7 ± 19.2 years), but were unavailable for nine patients. Of the 184 patients, 122 were male and 62 were female; sex data could not be accessed in the remaining nine patients. Of the 149 patients whose preoperative diagnosis information could be obtained, 65 (43.6%) underwent operations for abdominal cocoon, while the majority of the remaining patients underwent operations for a presumed diagnosis of intestinal obstruction and/or abdominal mass. Management information could be retrieved for 115 patients. Of these, 68 underwent excision + adhesiolysis (one laparoscopic); 24 underwent prophylactic appendectomy in addition to excision + adhesiolysis. Twenty patients underwent various resection and repair techniques along with excision + adhesiolysis. The remaining three patients were managed with antituberculosis therapy (*n* = 2) and immunosuppressive therapy (*n* = 1).

CONCLUSION: Idiopathic SEP is a rare disorder characterized by frequently recurring bouts of intestinal obstruction. Surgical therapy is the gold standard

management strategy.

Key words: Primary; Idiopathic; Intestinal obstruction; Sclerosisis encapsulation peritonitis; Abdominal cocoon syndrome

Core tip: Idiopathic sclerosing encapsulating peritonitis (SEP) is a clinical entity characterized by partial or complete encasement of the small intestines by a thick fibrocollagenous membrane. While some patients with idiopathic SEP are asymptomatic, the majority of affected individuals develop acute, subacute or chronic attacks of gastrointestinal obstruction. Preoperative diagnosis of the disease is quite difficult, and many cases are diagnosed intraoperatively. Nonetheless, recent technological advances in imaging modalities, particularly computed tomography, have made preoperative diagnosis of SEP possible. Surgery remains the best management option for patients with severe signs of intestinal obstruction.

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INTRODUCTION

Sclerosing encapsulating peritonitis (SEP) is a chronic inflammatory process in which the small intestines are encased by a dense fibrocollagenous membrane[1-32]. SEP was first defined nearly 100 years ago, at which time it was termed "peritonitis chronica fibrosa incapsulata"[1,6,7]. The disorder is divided into primary (idiopathic) and secondary forms according to the underlying etiological cause[1-5]. The primary form was termed "abdominal cocoon syndrome" by Foo in 1978[1]. The clinical signs and symptoms of SEP vary with the severity and duration of the disease, underlying causes, and affected person's immunological status. SEP most commonly manifests as recurrent acute, subacute, or chronic episodes of intestinal obstruction[2,4]. However, some cases may also manifest with more uncommon, but life-threatening, complications including enterocutaneous fistula, small intestinal necrosis, and malnutrition. Preoperative diagnosis of SEP is quite difficult, and many cases are diagnosed intraoperatively[4,6]. Fortunately, preoperative diagnosis of SEP has become possible with recent technological advances in imaging modalities, particularly computed tomography (CT)[1,5-11]. Surgery remains the most effective management option for SEP[4], although controversy surrounds the indications, optimal timing, and mode of surgical operation. This is because surgical outcomes are far from satisfactory, and some patients may develop postoperative small intestinal obstruction and new adhesions[4]. The present study reviews and discusses the previously published articles on SEP.

MATERIALS AND METHODS

We reviewed nearly 200 previously published articles on SEP. A serious contradiction was present between selection and classification of cases, because many authors used the term "abdominal cocoon" while actually describing cases of secondary SEP. We therefore aimed to resolve this conflict by establishing a proper definition and classification of SEP before starting the literature review. We divided SEP into primary (idiopathic; abdominal cocoon syndrome) and secondary forms. Patients with no factors explaining SEP after various examinations (history taking, blood tests, radiological imaging, and histopathological tests) performed during the preoperative, perioperative, or postoperative periods were determined to have primary SEP (idiopathic, abdominal cocoon). Patients with SEP that developed as a result of various conditions, including abdominal surgery, abdominal tuberculosis, peritoneal dialysis (PD), ventriculoperitoneal or peritoneovenous shunts, liver transplantation, recurrent peritonitis, beta-blocker treatment (practolol or propranolol), intraperitoneal chemotherapy, intraperitoneal povidone-iodine use, liver cirrhosis, gastrointestinal malignancy, fibrogenic foreign material, systemic lupus erythematosus, or parasitic infection (sometimes leading to granulomatous peritonitis) were determined to have secondary SEP. The main objective of the present study was to perform a brief review of the literature to identify studies on primary SEP (idiopathic; cocoon syndrome) published from January 2000 to May 2014. To achieve this aim, we scanned the PubMed, MEDLINE, Google Scholar, and Google databases for the key words "sclerosing encapsulating peritonitis," "idiopathic sclerosing encapsulating peritonitis," "abdominal cocoon," and "abdominal cocoon syndrome" entered alone or in various combinations. Only articles published in English were included in the scanning process. Cases that met the diagnostic criteria for idiopathic SEP (abdominal cocoon) were included in the review, while cases with features of secondary SEP were excluded. The corresponding authors of some papers were e-mailed several times regarding necessary information about their articles. However, we received no effective responses from the authors of the two largest studies. We created a table with useful information about the reviewed articles, including publication year, country, number of cases, patient age, sex, history, white blood cell count, surgical approach, complications, follow-up duration and other ancillary information.

RESULTS

A literature review using the above mentioned inclusion criteria revealed 73 articles on idiopathic (primary) SEP

Table 1 Distribution of articles and number of cases with idiopathic sclerosing encapsulating peritonitis according to countries *n* (%)

Countries	Published articles	Published cases
China	8 (10.95)	104 (53.88)
India	21 (28.76)	35 (18.13)
Turkey	14 (19.17)	17 (8.80)
Nigeria	3 (4.10)	5 (2.59)
Taiwan	2 (2.74)	2 (1.03)
Pakistan	2 (2.74)	2 (1.03)
Qatar	2 (2.74)	3 (1.55)
Saudi Arabia	2 (2.74)	2 (1.03)
Israel	2 (2.74)	3 (1.55)
Iran	2 (2.74)	2 (1.03)
Nepal	2 (2.74)	2 (1.03)
Brazil	2 (2.74)	3 (1.55)
Italy	1 (1.37)	1 (0.51)
United States	1 (1.37)	2 (1.03)
South Korea	1 (1.37)	2 (1.03)
Senegal	1 (1.37)	1 (0.51)
Iraq	1 (1.37)	1 (0.51)
Belgium	1 (1.37)	1 (0.51)
Bangladesh	1 (1.37)	1 (0.51)
Kuwait	1 (1.37)	1 (0.51)
Malaysia	1 (1.37)	1 (0.51)
New Zealand	1 (1.37)	1 (0.51)
Greece	1 (1.37)	1 (0.51)

China has reported the greatest number of cases, while India has published the greatest number of articles.

from 23 countries[2-10,12-31,33-76]. The four countries with the highest numbers of published articles were India (*n* = 21; 28.76%), Turkey (*n* = 14; 19.17%), China (*n* = 8; 10.95%) and Nigeria (*n* = 3; 4.10%). The four countries reporting the highest number of cases were China (*n* = 104; 53.88%), India (*n* = 35; 18.13%), Turkey (*n* = 17; 8.80%) and Nigeria (*n* = 5; 2.59%). Other data related to the article distribution among countries are presented in Table 1. In total, 193 patients were included in this study. Their ages ranged from 7 to 87 years (mean ± SD, 34.7 ± 19.2 years) among 184 patients; this information was unavailable for the remaining 9 patients. Of the 184 patients, 122 were male and 62 were female; no sex data were available for the remaining 9 patients. The symptom duration ranged from 8 h to 210 mo among 174 patients; this information was unavailable for the remaining 19 patients. Of 149 patients with available data on preoperative diagnosis, 65 (43.6%) underwent operations for a presumed diagnosis of abdominal cocoon syndrome, while the majority of the remaining patients underwent operations for an initial diagnosis of intestinal obstruction and/or abdominal mass. Patient management data were available in 115 patients; 68 underwent excision + adhesiolysis, and 24 underwent prophylactic appendectomy in addition to excision + adhesiolysis. Twenty patients underwent various resection and anastomosis techniques in addition to excision + adhesiolysis. Two patients commenced antituberculous therapy without antecedent surgical therapy. Those patients had no signs or symptoms pertaining to tuberculosis. One patient was administered with steroids and immunosuppressive therapy. The demographic and clinical data of the 193 patients included in the present study are summarized in Table 2. Two studies were published from the same institution and used the medical data of the same patient; despite meeting the inclusion criteria for this review, one of these studies was excluded[60,77].

DISCUSSION

Definitions and historical background

The definition of SEP is associated with confusion and lack of information. The concepts of primary and secondary SEP are erroneously used interchangeably in many previously published articles on SEP[11,32]. Thus, we aimed to emphasize the correct use of the definitions of peritoneal encapsulation (PE), abdominal cocoon, idiopathic SEP, and secondary SEP in the present review.

PE was first described by Cleland in 1868[32]. It is a developmental anomaly characterized by the congenital presence of an accessory peritoneal membrane, which is believed to be derived from the yolk sac peritoneum in the early stages of fetal life[10,15,29,32]. This peritoneal membrane is classically found between the mesocolon and omentum, and most of the small intestines lie posterior to this membrane[21,27,39,48,75]. In other words, PE is an anatomical anomaly unrelated to any inflammatory process. PE is typically asymptomatic and incidentally detected during laparotomy performed for other indications[29,32,62,73]. In one patient, we observed anatomical features similar to those of PE during laparotomy performed to treat a gunshot injury (Figure 1).

Unlike PE, SEP is an acquired condition resulting from peritoneal inflammation that may be triggered by various factors[32,38]. While the accessory peritoneal membrane is covered by mesothelium in patients with PE, the membrane that encases the intestines in patients with SEP has a dull, fibrous structure that includes inflammatory cells[33,38,39]. SEP is a clinical entity characterized by partial or complete encasement of the small intestines by a thick fibrocollagenous membrane (Figure 2)[1,4,6,10,17,24]. This membrane often encapsulates the small intestines, but it sometimes also encases other intraperitoneal organs, such as the stomach, liver, and colon[1,6,8,23,55]. This clinical entity was first defined in 1907 by Owtschinnikow, who described encasement of the intestines by a fibrocollagenous membrane[1,50,55]. Considering the morphological and histological properties of the membrane encasing the intestines, Owtschinnikow termed this condition "peritonitis chronica fibrosa incapsulata"[1,16,17,27]. Historically, SEP was classified as primary (idiopathic) or secondary, depending on its underlying cause and the pathogenetic properties of the fibrocollagenous membrane[1,23,42,49]. The idiopathic form of SEP has also been termed "abdominal cocoon syndrome," a term that was first used by Foo in 1978[1]. Abdominal cocoon is categorized into three types

Table 2 Demographic and clinical characteristics of 193 patients with idiopathic sclerosing encapsulating peritonitis

Ref.	Year	Country	Case number	Age (yr)	Sex	Duration symptom	Preoperative diagnosis	Radiologic tools	Surgical approach
Rasihashemi *et al*[2]	2014	Iran	1	25	M	2 mo	Int Obst	X-ray + Barium + CT	E + A
Nanwadekar *et al*[3]	2014	India	1	17	F	4 d	Int Obst	X-ray + US + Endosc.	E + A
Li *et al*[4]	2014	China	65	39 (14-79)	M: 57 F: 8	3.9 ± 6.7 yr	ACS: 31 NS: 34	NS	NS
Jovani *et al*[5]	2014	Italy	1	44	M	60 mo	ACS	US + CT + MR	NS
Akbulut *et al*[6]	2014	Turkey	1	87	M	3 mo	Int Obst + perforation	X-ray + US	E + A + resection + ileostomy
Sreevathsa *et al*[7]	2013	India	3	43	M	12 mo	ACS	X-ray + CT	E + A
				13	F	12 mo	Int Obst	X-ray	Ileocecal resection
				14	F	6 mo	Int Obst (Subacute)	X-ray	Ileocecal resection
Singh *et al*[8]	2013	India	9	NS	NS	NS	NS: 9	NS	NS
Shah *et al*[9]	2013	India	1	14	F	6 mo	ACS	Barium + CT	E + A
Serter *et al*[10]	2013	Turkey	2	32	M	2 d	Int Hernia	X-Ray + CT	E + A
				49	M	1 wk	ACS	CT	E + A
Rahmati *et al*[12]	2013	Iran	1	50	M	3 mo	ACS	US + CT + Endosc.	E + A
Patel *et al*[13]	2013	India	1	45	M	6 mo	Int Obst	X-ray + CT	E + A + ileal resection
Ozkan *et al*[14]	2013	Turkey	1	48	M	1 wk	ACS	X-ray + CT	E + A
Hu *et al*[15]	2013	China	1	29	F	Asympt.	Infertility	US	E + A + suturing (iatrogenic ileal injury)
Gupta *et al*[16]	2013	India	1	40	M	NS	ACS	X-ray + US + CT	E + A
Gadhire *et al*[17]	2013	India	1	35	M	1 mo	ACS	X-ray + US + CT	E + A
Awe[18]	2013	Nigeria	1	18	F	3 d	Int Obst	X-ray	E + A
Al Thani *et al*[19]	2013	Qatar	1	41	M	7 mo	Int Obst (subacute)	CT	E + A
Thakur *et al*[20]	2012	India	1	14	F	6 mo	Abd Mass	US	E + A
Taylor *et al*[21]	2012	N Zealand	1	42	M	3 d	Int Obst	X-ray + CT	E + A + appendectomy
Solak *et al*[22]	2012	Turkey	1	58	M	24 mo	ACS (previously operated)	X-ray + US + CT	Steroid + mycophenolate mofetil
Shakya *et al*[23]	2012	Nepal	1	20	M	12 mo	Int Obst	X-ray	E + A + Ileostomy (iatrogenic ileal injury)
Ndiaye *et al*[24]	2012	Senegal	1	15	F	2 mo	ACS	Barium + CT	E + A + Suturing (iatrogenic ileal injury)
Meshikhes *et al*[25]	2012	Saudi Arabia	1	45	M	6 mo	Int Obst + Abd mass	CT	E + A + appendecectomy
Malik *et al*[26]	2012	Pakistan	1	24	F	60 mo	Int Obst	X-ray	E + A
Kumar *et al*[27]	2012	India	2	18	F	24 mo	ACS ?	Barium + US + CT	Antitubercular therapy
				14	F	NS	ACS ?	CT + US	Antitubercular therapy
Kayastha *et al*[28]	2012	Pakistan	1	13	F	2 mo	Acute appendicitis	US	E + A
Kaur *et al*[29]	2012	India	2	43	M	180 mo	ACS	X-ray + US + CT	E + A
				17	F	4 mo	ACS	X-ray + US + CT	E + A
Araujo Filho *et al*[30]	2012	Brazil	1	36	M	10 d	ACS	US + CT	E + A
Chatura *et al*[31]	2012	India	1	14	F	NS	Int Obst + Abd mass	US	E + A + ileocolectomy
Yeniay *et al*[33]	2011	Turkey	2	26	F	2 d	Int Obst	X-ray + CT	E + A
				71	M	3 mo	Int Obst	X-ray + CT	E + A
Kirshtein *et al*[34]	2011	Israel	1	82	M	4 d	Int Obst	X-Ray + gastrografin	E + A
Jayant *et al*[35]	2011	India	1	16	F	NS	Int Obst	CT	E + A
Gupta *et al*[36]	2011	Nepal	1	42	M	4 mo	ACS	X-ray + US + CT	E + A
Ertem *et al*[37]	2011	Turkey	1	29	M	2 d	Int Obst	X-ray + US + CT	E + A - laparoscopic
Da Luz *et al*[38]	2011	Brazil	2	30	M	NS	Int Obst + Int Hernia	X-ray + barium	E + A + laparostomy
				32	M	6 mo	Int Obst + Chron?	X-ray + barium	E + A
Bassiouny *et al*[39]	2011	Qatar	2	7	M	48 mo	Int Obst + Abd mass	X-ray	E + A
				12	F	48 mo	Int Obst	X-ray + US	E + A
Wang *et al*[40]	2010	China	1	48	M	3 mo	ACS	CT	E + A + appendectomy
Tombak *et al*[41]	2011	Turkey	1	36	M	1 mo	ACS	CT	E + A
Naik *et al*[42]	2010	India	1	70	M	48 mo	Int Obst	X-ray + US + CT + Endosc.	E + A

Lee *et al*[43]	2010	Taiwan	1	57	F		ACS	X-ray + US + CT	E + A
Gurleyik *et al*[44]	2010	Turkey	1	30	M	36 mo	Int Obst	X-ray + US + CT	E + A
Al Saied *et al*[45]	2010	Saudi Arabia	1	24	M	36 mo	ACS	X-ray + CT	E + A
Yang *et al*[46]	2009	China	1	43	M	NS	NS	X-ray + Endosc.	Resection (?)
Yang *et al*[47]	2009	China	6	43.7 (39-48)	M: 4 F: 2	3-60 mo	Int Obst: 5 ACS: 1	X-ray + CT	E + A: 5 E + A + jejunal resection: 1
Wu *et al*[48]	2009	Taiwan	1	80	M	24 mo	Int Obst	X-ray + US + CT	E + A
Wei *et al*[49]	2009	China	24	34 (15-57)	M: 9 F: 15	3 d-216 mo	ACS: 4 Int Obst/mass: 20	X-ay + barium + US + CT	E + A + appendectomy: 17 E + A + enterotomy: 2 E + A + cecofixation: 2 E + A: 3
Tasdelen *et al*[50]	2009	Turkey	1	85	F	3 d	Int Obst + Int Hernia	X-ray + CT	E + A + jejunoileal resection with anastomosis
Reynders *et al*[51]	2009	Belgium	1	40	M	36 mo	Int Obst	X-ray + CT	E + A + Meckel's resection + appendectomy
Mohanty *et al*[52]	2009	India	1	15	F	24 mo	ACS	X-ray + US + CT	E + A
Kumar *et al*[53]	2009	India	3	45	M	24 mo	ACS	X-ray + CT	E + A
				63	M	216 mo	ACS	X-ray + US + CT	E + A
				16	F	10 h	ACS	X-ray + US + CT	E + A
Ibrahim *et al*[54]	2009	Nigeria	1	14	M	72 h	Int Obst	X-ray	E + A + appendectomy
Choudhury *et al*[55]	2009	Bangladesh	1	15	F	12 mo	Appendiceal mass	US	Partial ileocolic resection with anastomosis
Zheng *et al*[56]	2008	China	1	69	M	1 d	ACS	X-ray + US + CT	E + A + ileal resection with anastomosis
Bas *et al*[57]	2008	Turkey	1	42	M	5 mo	Int Obst	X-ray + CT	E + A
Singh *et al*[58]	2008	India	1	38	M	12 mo	Int Obst	X-ray + US	E + A
Xu *et al*[59]	2007	China	5	41	F	4 mo	Int Obst	X-ray + CT + Endosc.	E + A
				49	F	120 mo	Int Obst	X-ray + CT + Endosc.	E + A
				21	M	36 mo	Int Obst	X-ray + CT + Endosc.	E + A
				41	M	1 mo	Int Obst	X-ray + CT + Endosc.	Adhesiolysis + jejunal resection with anastomosis
				36	M	2 wk	Int Obst	X-ray + CT + Endosc.	E + A
Demir *et al*[60]	2007	Turkey	1	38	M	6-7 mo	ACS	CT	E + A
Cai *et al*[61]	2007	United States	2	38	M	2 d	Int Obst	X-ray	E + A
				45	M	8 h	Int Obst	CT	E + A
Basu *et al*[62]	2007	India	1	47	M	3 mo	Abd mass	X-ray + US + barium	E + A
Al-Ibrahim *et al*[63]	2007	Kuwait	1	33	M	1 mo	Int Obst	X-ray + US + CT	E + A
Serafimidis *et al*[64]	2006	Greece	1	56	M	48 mo	Int Obst	X-ray + US + CT + Endosc.	E + A
Rokade *et al*[65]	2006	India	1	26	F	12 mo	ACS (previously operated)	US + CT	E + A
Pillai *et al*[66]	2006	India	1	13	F	NS	ACS	X-ray + US + CT	E + A
Akca *et al*[67]	2006	Turkey	1	57	M	75 d	Int Hernia + mesenteritis	US + CT + Colonosc.	NS
Yucel *et al*[68]	2004	Turkey	2	15	F	NS	Int Obst	X-ray + CT	E + A
				38	M	72 mo	Int Obst	X-ray + CT	E + A
Hur *et al*[69]	2004	South Korea	2	34	F	120 mo	Int Obst	X-ray + barium + US + CT	NS
				47	M	NS	Int Obst	X-ray + barium + CT	NS
Vijayaraghavan *et al*[70]	2003	India	1	12	F	3 mo	ACS + Int Hernia	US	E + A
Ranganathan *et al*[71]	2003	Malaysia	1	25	F	3 mo	Large ovarian mass + ascites	US + CT	E + A
Hasan[72]	2002	Iraq	1	20	F	NS	Acute abdomen	Pregnant patient	E + A
Hamaloglu *et al*[73]	2002	Turkey	1	38	M	12 mo	Int Obst	X-ray + Barium + US	E + A
Okobia *et al*[74]	2001	Nigeria	3	18	F	5 mo	Pelvic collection	US	E + A + appendectomy
				12	F	1 wk	Mesenteric cyst	X-ray + US	E + A + appendectomy
				10	F	2 mo	Ovarian Tm + Burkitt's Tm + uterine mass	X-ray + Urography	E + A + appendectomy

Mordehai *et al*[75]	2001	Israel	2	14	F	1 mo	Int Obst	X-ray + US + CT	E + A
				15	F	6 mo	Int Obst	X-ray + US	E + A
Kumar *et al*[76]	2000	India	1	12	F	24 h	Int Obst	X-ray + US	E + A

CT: Computed tomography; US: Ultrasonography; X-Ray: Plain X-ray abdominal radiography; Endosc: Gastrointestinal endoscopy; Int Obst: Intestinal obstruction; ACS: Abdominal cocoon syndrome; Abd mass: Abdominal mass; Int Hernia: Internal herniation; NS: Non-stated; E + A: Excision + adhesiolysis.

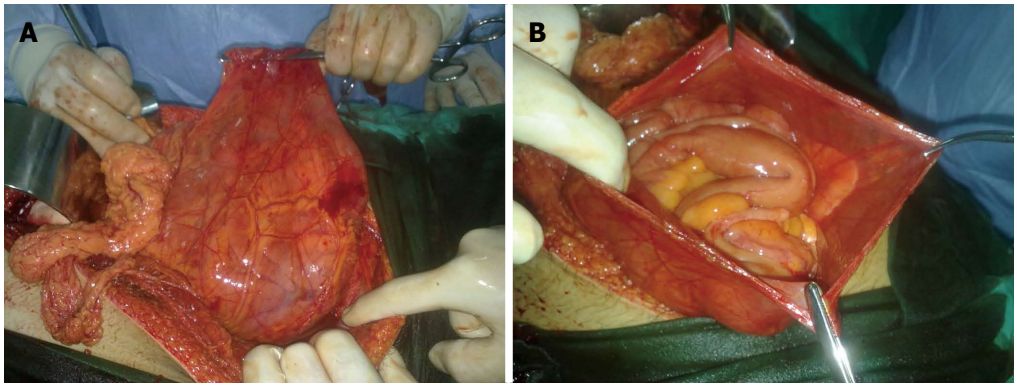

Figure 1 Bowel encased in a membranous sac suggestive of peritoneal encapsulation. A: The overall appearance of the membranous sac is shown. All intestines are localized behind the accessory peritoneal membrane; B: The appearance of the opened membranous sac is shown.

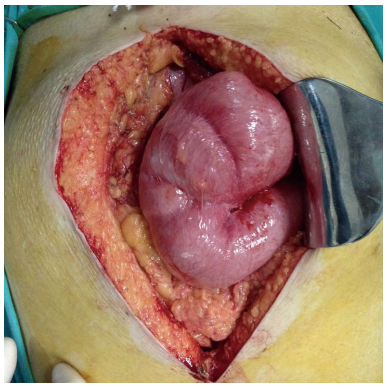

Figure 2 Intraoperative photograph showing the encapsulated small bowel (dense, cocoon-like fibrous membrane).

according to the extent of the encasing membrane that covers the intestine. Encasement of part of the intestine by a fibrocollagenous membrane is called type 1 cocoon syndrome. Complete coverage of the intestine by the membrane is called type 2 abdominal cocoon syndrome. Type 3 cocoon syndrome refers to encasement of the whole intestine, as well as other intra-abdominal organs, such as the appendix, cecum, ascending colon, and ovaries[1,49].

Etiology

SEP is considered to be primary (idiopathic) or secondary, depending on its underlying cause[1-10]. No underlying cause can be demonstrated in primary SEP, although the role of cytokines and fibroblasts in development of peritoneal fibrosis and neoangiogenesis is indisputable[40,58]. Idiopathic SEP classically presents in young adolescent girls in tropical and subtropical countries such as China, Malaysia, Singapore, Pakistan, India, Nigeria, Kenya, Saudi Arabia, and South Africa, although adult cases of idiopathic SEP in temperate zones have also been reported[1,4,7,22,23,58,61]. The present study showed that idiopathic SEP is twice as common in men than in women. Our findings on the geographical distribution of SEP coincide with those in the previously published literature. Indeed, nearly all cases presented herein occurred in tropical or subtropical regions of the world.

Many hypotheses regarding the etiology of idiopathic SEP have been proposed[55,59,64]. Some of these hypotheses involve retrograde menstruation with a superimposed viral infection, retrograde peritonitis *via* the fallopian tubes, and cell-mediated immunological tissue damage secondary to gynecological infection[1,4,7,13,23,28,36,39]. However, SEP also develops in men, premenopausal women, and children, reducing support for these theories[1,4,7,28,61]. In total, 66 of 89 patients included in the largest two studies on idiopathic SEP in the literature to date were male[4,49]. Some authors have argued that the fibrous membrane that encases the intestines is a result of a developmental disorder, citing vascular anomalies and omental hypoplasia as the basis of their hypothesis[1,49,59].

Secondary SEP is more common than idiopathic SEP[22,45,52]. In secondary SEP, a local or systemic factor triggers the inflammatory process in the peritoneum[52]. PD is the most common cause of secondary SEP[1]. In other words, secondary SEP is the leading cause of the most severe complications of PD. This is because once secondary SEP has developed, the ultrafiltration capacity of the peritoneal surface decreases and the risk of intestinal obstruction increases[1]. Studies have shown a direct relationship between prolonged PD and

Table 3 Classification of sclerosing encapsulating peritonitis according to underlying cause

Primary (idiopathic) sclerosing encapsulating peritonitis
Ⅰ Adolescent form
Ⅱ Adult form
Secondary sclerosing encapsulating peritonitis
Ⅰ Systemically induced by
Beta adrenergic blocking agents
Practolol
Timolol
Propanolol
Other drugs
Methotrexate
Protein S deficiency
Exposure to asbestosis
Ⅱ Induced by possible local and/or systemic irritants
Peritoneal dialysis
Abdominal trauma
Abdominal surgery
Liver transplantation
Peritoneovenous shunt
Ventriculoperitoneal shunt
Peritoneal sarcoidosis
Liver cirrhosis
Peritoneal tuberculosis
Sarcoidosis
Familial mediterranean fever
Systemic lupus erythematosus
Gastrointestinal malignancy
Intraperitoneal chemotherapy
Fibrogenic foreign body
Endometriosis
Dermoid cyst rupture
Luteinized ovarian thecomas
Cytomegalovirus peritonitis
Recurrent peritonitis
Granulomatous peritonitis related with parasitic infestation

the development of secondary SEP[1,11]. Considering the number of patients undergoing PD worldwide, the importance of the relationship between PD and secondary SEP needs to be better understood. Abdominal tuberculosis continues to be a major public health issue and an important etiological agent of secondary SEP in underdeveloped countries[8]. Among the less frequent causes of secondary SEP are a history of abdominal surgery, autoimmune disorders, some drugs, peritoneal shunts, and recurrent episodes of peritonitis[1,4,7,10,17,28,32,34]. The classification and potential etiological factors of SEP are listed in detail in Table 3.

Clinical presentation

Idiopathic SEP is an uncommon entity, and a great majority of physicians either never encounter patients with this condition or miss the diagnosis even when they do. Achieving a correct preoperative diagnosis in affected patients is extremely difficult and requires a high index of clinical suspicion[1,4,25,38,51]. Recent advances in radiological modalities have allowed physicians to achieve a correct preoperative diagnosis of SEP in affected patients[59,71,77]. Nevertheless, preoperative diagnosis remains a clinical challenge because most patients with SEP present to emergency departments with signs and symptoms of intestinal obstruction, and many emergency departments lack advanced radiological equipment and adequate staff, and patients with this syndrome usually undergo operations on an urgent basis[38]. In one large case series, 52.3% to 100.0% of admitted patients were diagnosed during surgery and 16.7% to 48.7% were diagnosed during their preoperative examinations[4,8,50]. While some patients with SEP are asymptomatic, most affected individuals develop acute, subacute, or chronic attacks of gastrointestinal obstruction (incomplete or complete); nausea; vomiting; anorexia; appetite loss; weight loss; and malnutrition[1,4,8,10,11,26]. Although rare, a painless, soft abdominal mass can be palpated in some patients[1,4,8,29,30,76]. Additionally, abdominal ascites and distention are detectable in some patients with severe disease. Ascites may be massive enough to induce suspicion of underlying hepatic disease. Primary SEP may be considered in patients presenting with recurrent attacks of abdominal pain who are free of any disease explaining such attacks[1]. Gastrointestinal perforation is quite rare in patients with SEP; of all reported cases of SEP, only two (one secondary to tuberculosis and the other idiopathic) were associated with spontaneous perforation[6].

Diagnostic approaches

The diagnosis of SEP is often made by a combination of the medical history, a high clinical index of suspicion, various biochemical parameters, and radiological findings[18,23,26]. The patient's medical history (tuberculosis, PD, systemic lupus erythematosus, previous abdominal operations, *etc.*) usually provides important clues regarding the etiology of secondary SEP. The most commonly used radiological techniques are abdominal X-rays, small intestinal barium studies, ultrasonography, abdominal CT, and occasionally contrast-enhanced magnetic resonance (MR) imaging[5,6,28,30]. Abdominal X-rays may show diffuse air-fluid levels and dilated small intestinal loops[1,3,29,35]. However, X-ray findings are not specific to idiopathic SEP; rather, they are common to many conditions characterized by intestinal obstruction[22]. In patients with SEP, small intestinal barium studies show the intestinal loops that are accumulated and conglomerated at the center of the abdomen (Figure 3A)[1,9,24,29,76]. This appearance is termed the cauliflower sign or accordion pattern and is a clue for the diagnosis of SEP[9,24,29,64,66,69]. A prolonged transit time may also aid in the diagnosis (Figure 3B)[1,23,24,38]. However, barium studies may not be possible in patients with prominent signs of intestinal obstruction. Abdominal ultrasonography may show dilated bowel segments encased by a dense fibrous membrane[1,44] or free abdominal fluid and a thickened peritoneal layer[22,29,68]. Contrast-enhanced CT is the most helpful imaging modality for the diagnosis of abdominal cocoon[3,29,36]. The characteristic sign on CT is the appearance of small bowel segments that are conglomerated at the

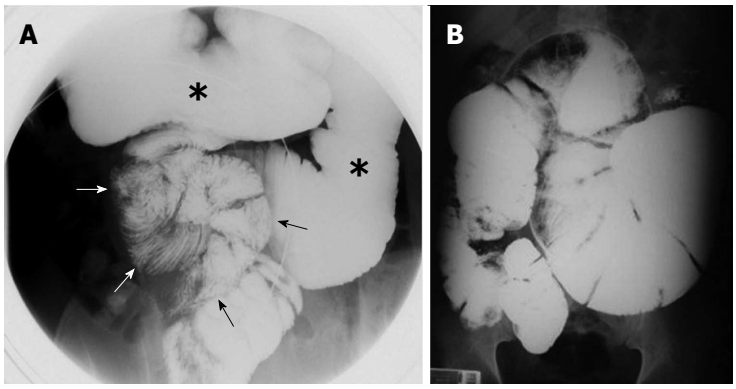

Figure 3 Small bowel transit. Procubitus image with localized compression. Liquid distension of the gastroduodenum (asterisks) and adhesion of the small intestinal loops (arrows) are persistent despite localized compression, producing a "cauliflower" appearance[24]; B: Upper gastrointestinal images reveal dilatation of the duodenum and jejunal loops, delayed bowel transit, and failure of the oral contrast to pass distally[38].

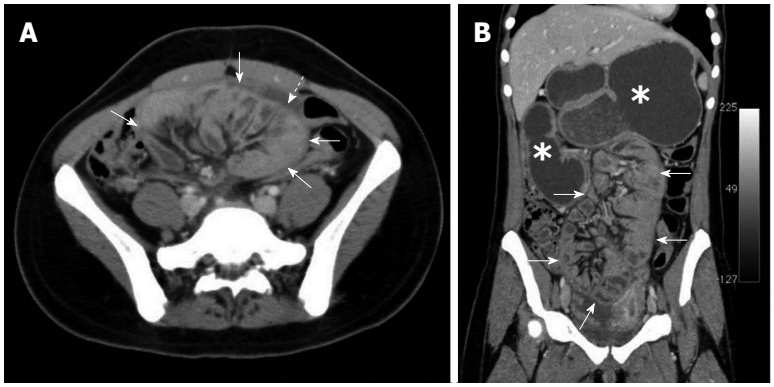

Figure 4 Contrast-enhanced abdominal computed tomography[24]. Small intestinal loops are encased in a sac of thick peritoneal membrane (continuous arrows) with a small volume of peritoneal liquid effusion (discontinuous arrow). Gastroduodenal distension is also present (asterisks). A: Axial slice; B: Multiplanar coronal reconstruction.

midline and encased by a dense capsule with a contrast-free periphery (Figure 4)[1,4,14,19,35,36]. CT may also show intestinal obstruction, ascites, localized fluid collections, peritoneal or mesenteric thickening, mural or peritoneal calcifications, and lymphadenopathy[1,60]. Multidetector CT technology has greater accuracy because it allows for multiplanar (axial, sagittal, and coronal) reconstruction. It thus provides valuable information about the severity and level of intestinal obstruction[22,40,41]. Multiplanar reformatted images provided by multidetector CT are very helpful for both exclusion of other potential causes of intestinal obstruction and planning of the surgical operation[22,29,37,41,69]. To the best of our knowledge, only one report to date has described the use of contrast-enhanced MR imaging in a patient with idiopathic SEP. Jovani *et al*[5] performed MR enterography of their patients and compared MR images with CT images after oral administration of 1.5 L of polyethylene glycol and intravenous administration of gadolinium. The authors concluded that MR-acquired images were similar to or even better than CT-acquired images in patients with SEP (Figure 5). In summary, contrast-enhanced CT (multidetector CT with multiplanar reformation) is the most helpful radiological tool for confirming the diagnosis, planning therapy, and avoiding unnecessary resection in patients with SEP.

Differential diagnosis

Most patients with symptomatic SEP present to an emergency department or general surgery clinic with recurrent acute, subacute, or chronic episodes of gastrointestinal obstruction[8,34]. Postoperative adhesions are detectable in approximately 60% to 80% of patients who present with small intestinal obstruction, while unusual conditions are diagnosed in about 6% of affected individuals[1,26,31,34,36,43,50]. Idiopathic SEP is one of the more unusual conditions that lead to intestinal obstruction[36,52,53]. Internal herniation and congenital PE are the two pathological conditions that should be primarily considered as differential diagnoses in such patients[10,16,29,43,70]. Less common conditions to be considered as differential diagnoses are voluminous invagination, intestinal malrotation, secondary peritonitis, and other causes of peritoneal adhesion[1,10,60]. Tuberculous peritonitis should be definitively excluded in patients who live in tuberculosis-prevalent regions[17,23]. Tuberculosis is so common in some regions that antituberculosis therapy is empirically administered to some patients with intestinal obstruction[23,25,27]. The medical history of the patient (*e.g.*, pulmonary or genital tuberculosis), adenosine

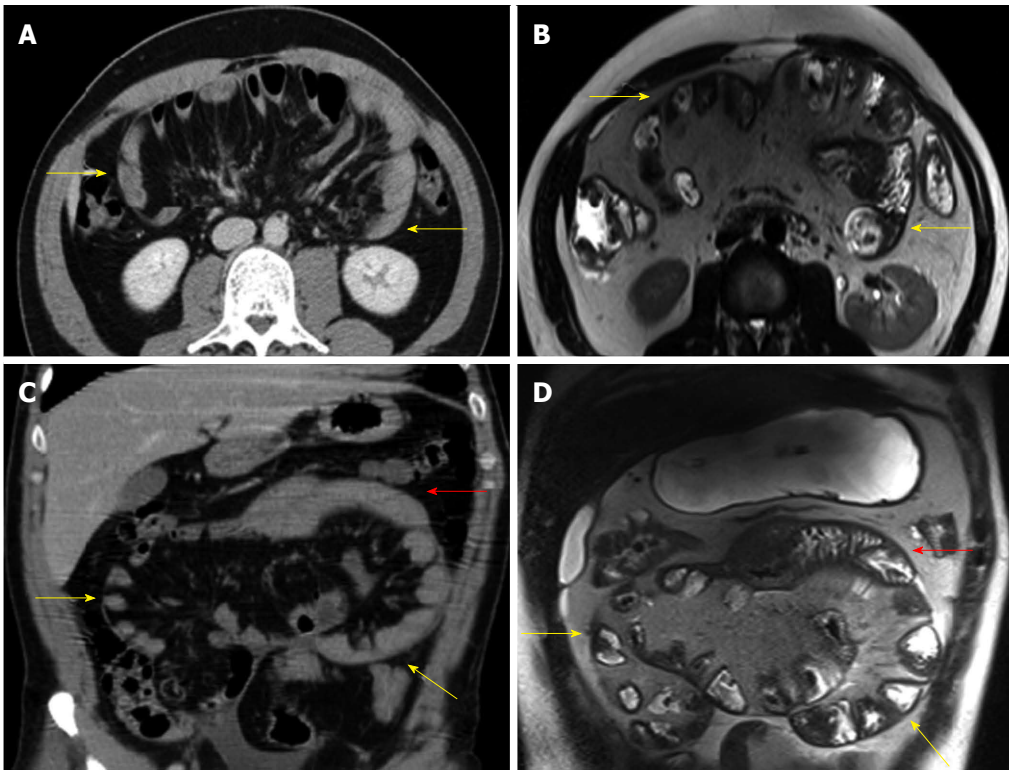

Figure 5 Comparison of diagnostic features on computed tomography and magnetic resonance images[5]. A: Computed tomography scan in the axial plane showing a subtotal conglomeration of small bowel loops coiled in a concertina-like fashion and encased by a thick membrane (yellow arrows); B: T2-weighted magnetic resonance imaging sequence in the axial plane showing bowel loops aggregated in a festoon-like shape and encased by a thick membrane (yellow arrows); C: Computed tomography scan in the coronal plane showing the conglomeration of small bowls loops (yellow arrows); a few free loops are present in the upper quadrant (red arrow); D: T2-weighted magnetic resonance imaging sequence in the coronal plane showing the same conglomerated small bowel loops (yellow arrows) and a few free bowel loops (red arrow).

deaminase level in ascitic fluid, culture of sputum and ascitic fluid, and erythrocyte sedimentation rate should be evaluated to avoid erroneous administration of clinical therapies[17,65]. Laparoscopic or open surgical biopsy of the peritoneum may be performed to rule in a diagnosis of SEP[25]. An accurate preoperative diagnosis is vital for both accurate treatment planning and prognosis prediction[1,4]. The surgeon may avoid complications more effectively when he or she knows what to expect during laparotomy[1,4,10,22,43]. However, reaching a preoperative diagnosis for many patients is a challenging task, despite the performance of an extensive preoperative radiological and clinical workup; the correct diagnosis can only be achieved by intraoperative observation and histopathological examination[36,47,57,65].

Histopathological features of SEP

The peritoneum of patients with SEP is characterized histopathologically by fibroconnective tissue proliferation, inflammatory infiltration, and dilated lymphatics. No evidence of foreign body granulomas, giant cells, or birefringent material is present. The term "sclerosing" refers to the progressive formation of sheets of dense collagenous tissue, the term "encapsulating" describes the sheath of new fibrous tissue that covers and constricts the small bowel and restricts its motility, and the term "peritonitis" implies an ongoing inflammatory process and the presence of a mononuclear inflammatory infiltrate within the new fibrosing tissue[1,41]. Although not pathognomonic, these findings are useful for the diagnosis of SEP when combined with characteristic surgical findings.

Management

There is no evidence-based consensus regarding the optimal treatment approach in patients with idiopathic SEP[8], because 97.7% of the papers on idiopathic SEP to date are case reports (1 to 6 cases). Administration of conservative treatment for as long as possible is the best approach in patients with mild abdominal symptoms. In such patients, bowel rest, nasogastric decompression, and nutritional support (enteral or parenteral) are the most appropriate treatment options[22,78]. Appetite loss, malnutrition, and weight loss are the most common symptoms in patients with idiopathic SEP[4,78]. This is because recurrent bouts of intestinal obstruction, nausea, and vomiting limit patients' oral intake, leading to weight loss and malnutrition. Li *et al*[4] showed that preoperative nutritional support is a statistically significant independent factor for preventing postoperative complications. Based on the results of their study, the authors recommended enteral nutritional support in patients who are able to eat and parenteral nutritional support in those unable to eat. Studies have indicated that enteral or parenteral

nutritional support is key to avoiding complications and malnutrition, as well as to guarantee satisfaction among patients who undergo either medical or surgical management[4,78]. Patients with symptoms resistant to conservative therapy may be treated with drug therapies comprising tamoxifen, steroids, colchicine, azathioprine and mycophenolate mofetil[1,22,38,47,79]. Corticosteroids are thought to inhibit collagen synthesis and maturation by suppressing the inflammatory process within the peritoneal membrane. They also completely eliminate the thickened membrane[78,79]. Tamoxifen is a selective estrogen receptor modulator that inhibits fibroblastic production of transforming growth factor beta, a probiotic cytokine. This drug is therefore commonly used to treat certain fibrosclerotic disorders, such as retroperitoneal fibrosis and Riedel's thyroiditis[1,26,78,79]. Many articles have described the use of tamoxifen in patients with SEP[1,26,78]. Colchicine inhibits mRNA expression of transforming growth factor beta, thereby exhibiting an anti-inflammatory action. It has a low side effect profile and cost, but a strong antifibrogenic effect[22]. Cornelis *et al*[79] reported that corticosteroids and tamoxifen are useful in preventing and/or treating SEP. However, the authors concluded that data on other agents are quite limited. Many previous studies have evaluated anti-inflammatory/antifibrogenic medical therapy in patients with SEP undergoing PD[38]. However, there are almost no data, apart from a few case reports, on the use of such medications in patients with idiopathic SEP[78,79]. Solak *et al*[22] reported the successful use of a steroid+mycophenolate mofetil in a patient with recurrent symptoms after a surgical operation for idiopathic SEP. Malik *et al*[26] similarly administered postoperative steroids. Based on the aforementioned study data, we can conclude that medical therapy may be of benefit in patients with type Ⅱ and Ⅲ cocoon syndrome in whom adequate excision + adhesiolysis cannot be achieved or in patients with recurrent postoperative symptoms.

Unlike asymptomatic/mildly symptomatic patients, those with severe signs of intestinal obstruction or who have been intraoperatively diagnosed with SEP may have several surgical options. Partial membrane excision + adhesiolysis, resection + anastomosis, resection + anastomosis + protective enterostomy, and explorative laparotomy may be used alone or in combination, depending on the patient-related factors involved[1,2,12,20,68,74]. In patients with idiopathic SEP, the most suitable procedure includes peeling the membrane off of the intestinal surface and excising the dense adhesions between the intestinal loops[4,8,75]. Membrane excision + adhesiolysis should be applied to all encased intestinal segments when there are no other contraindications for this procedure. The risk of recurrence is quite low when the membrane on the intestinal surface can be totally excised[4]. Instilling an antiadhesive substance with between the intestinal loops before closing the abdomen may prevent the development of postoperative adhesive small bowel obstruction[25,49]. Whether administration of an antifibrogenic/anti-inflammatory agent during the postoperative period is beneficial in patients in whom the membrane that encapsulates the intestinal loops cannot be completely excised is debatable. To avoid complications, such as anastomosis leakage and short bowel syndrome, in patients with idiopathic SEP, bowel resection is indicated only when necrosis has developed[1,2,4,8,63]. Resection is usually unnecessary, and, when performed without a solid indication, may increase patient morbidity and mortality[1,4,26].

The most common postoperative complications are early postoperative small bowel obstruction (EPSBO), intra-abdominal infection, enterocutaneous fistula, short bowel syndrome, and bowel perforation[4,25,34,45,56]. EPSBO usually develops within 30 d postoperatively in patients who have undergone extensive adhesiolysis and excision[56]. EPSBO is secondary to excessive manipulation of the intestinal loops, prolonged operation times, and intestinal edema[4,17,56]. It is a temporary form of intestinal obstruction that usually has no sequelae after treatment with bowel rest and total parenteral nutrition[4,17,56]. Some authors have recommended the performance of small bowel intubation through the orifice of the appendix in patients with type Ⅱ and Ⅲ cocoon syndrome to reduce the risk of developing postoperative EPBSO[4,49]. Li *et al*[4] reported that EPSBO (P = 0.0001) and adhesive intestinal obstructions (P = 0.005) were less common in SEP patients undergoing intestinal intubation. The same authors also reported that they administered nutritional support combined with somatostatin and, when necessary, low-dose steroids in patients with EPSBO[4,56]. Such a treatment approach both reduces intestinal edema and minimizes bacterial translocation caused by stasis. Spontaneous development of enterocutaneous fistulas and perforation are rare, and only one such case has been reported to date; this case was characterized by idiopathic SEP-induced spontaneous perforation[6]. Postoperative fistula and perforation, on the other hand, are secondary to iatrogenic injury or anastomosis leakage. Long-term outcomes are quite impressive in patients who have undergone appropriate membrane excision + adhesiolysis[4,8,34].

Laparoscopy is not part of the standard surgical approach in patients with SEP. A limited number of case reports have described successful laparoscopic membrane excision and adhesiolysis[37]. An advantage of laparoscopy is that it can be used for both diagnostic and therapeutic purposes in patients with an unclear diagnosis after appropriate testing (Figure 6)[17,25]. However, Hu *et al*[15] reported that when they attempted laparoscopic exploration in one patient, the trocar directly entered the bowel because of the presence of adhesions. According to both our personal experience and impressions gained from the literature, it is best to first insert the trocar into the abdomen *via* the open technique when laparoscopy is planned for treatment of intestinal obstruction or intra-abdominal space-occupying lesions[36]. This rule also applies to patients with peritoneal fibrosis secondary to SEP or other causes, as well as to patients with a history

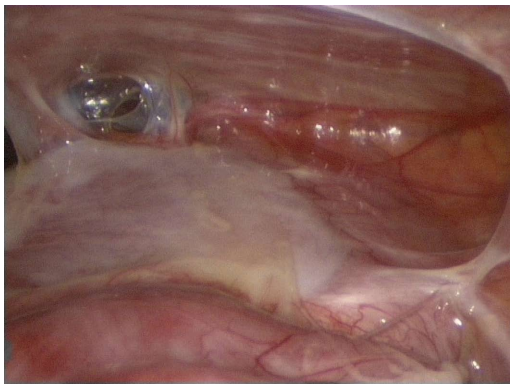

Figure 6 Laparoscopic view of the entire bowel segment encased with a fibrocollagenous membrane[37].

of abdominal surgery. Moreover, it is vital that the laparoscopic procedure is performed by an experienced operator to avoid iatrogenic bowel perforation[15,37].

In conclusion, idiopathic SEP is a clinical entity of unknown cause that is characterized by encasement of the intestines by a fibrocollagenous cocoon-like membrane. Most affected patients present to emergency departments with frequently recurring signs and symptoms of intestinal obstruction. Although recent advances in CT devices that allow for multiplanar imaging have enabled preoperative diagnosis of SEP, most cases are still incidentally diagnosed during laparotomy. Surgery remains the gold standard treatment for symptomatic idiopathic SEP. The most commonly used surgical method is membrane excision coupled with adhesiolysis. Minimally invasive management strategies help to avoid complications. Bowel rest, nasogastric decompression, and nutritional support may provide successful outcomes in asymptomatic or minimally symptomatic patients. Although various immunosuppressive, anti-inflammatory, and antifibrogenic agents reportedly provide satisfactory results in patients with secondary SEP, data on their use in patients with idiopathic SEP are limited. How those medications affect patients with idiopathic SEP remains unclear.

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COMMENTS

Background

Sclerosing encapsulating peritonitis (SEP) is a chronic inflammatory process in which the small intestines are encased by a dense fibrocollagenous membrane. SEP was first described in 1907 by Owtschinnikow, who termed this condition "peritonitis chronica fibrosa incapsulata." SEP is characterized as either primary (idiopathic) or secondary, according to its underlying cause.

Innovations and breakthroughs

The primary aim of this review was to screen the literature on idiopathic SEP, also known as abdominal cocoon syndrome. To the best of our knowledge, no studies on the use of correct terminology regarding SEP, primary SEP, and secondary SEP have been performed.

Terminology

SEP is characterized by a thick, grayish-white fibrocollagenous membrane that partially or totally encases the small bowel and that can extend to involve other organs. Patients with no factors explaining the condition are considered to have primary SEP, while patients with SEP that has developed as a result of various surgical or medical conditions are considered to have secondary SEP. Based on the extent of the encasing membrane that covers the intestine, SEP is categorized into three types. Encasement of part of the intestine by a fibrocollagenous membrane is called type 1 SEP. Complete coverage of the intestine by the membrane is called type 2 SEP. Type 3 SEP refers to encasement of the whole intestine as well as other intra-abdominal organs such as the appendix, cecum, ascending colon, and ovaries.

Peer review

The study is interesting, in which authors review the literature on idiopathic SEP, also known as abdominal cocoon syndrome. The results are interesting and suggest that idiopathic SEP is a rare disorder characterized by frequently recurring bouts of intestinal obstruction.

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