Intestinal obstruction due to congenital bands in adults who have never had abdominal surgery

Two case reports and a review of the literature

Ann. Ital. Chir., 2019 90, 6: 524-531
pii: S0003469X19031208

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Intestinal obstruction from congenital bands in adults with never-operated abdomen: An evasive and overlooked pathology. Two case reports and a review of literature

Congenital abdominal bands are a very rare entity and are often silent for a person's entire life. In a very few circumstances, however, they can manifest with clinical symptoms. Diagnosis of these bands is very challenging as the patient has never submitted for abdominal surgery and imaging, laboratory tests and clinical examination can be silent or unspecific. Two patients were admitted to and operated on in our hospital between June 2017 and July 2018. All patients had a bowel obstruction at the time of presentation, presenting no emission of faeces or gas. Conservative therapy was tried in one case, and surgical intervention was required. At surgery, both patients were found to have congenital bands causing the occlusion. The postoperative courses were uneventful.
Small bowel obstruction (SBO) by congenital bands remains a challenging disease for surgeons. Diagnosis of SBO has to be suspected even if radiology does not indicate any relevant features. Laparoscopy should be the preferred approach, but if not feasible, then open surgery can resolve the case.

KEY WORDS: Adhesions, Congenital bands, Intestinal occlusion, Laparoscopy

Introduction

Abdominal adherences are usually secondary to surgery, but in rare cases, they can be found in surgically virgin abdomen. In the latter case, they are generally due to inflammation; for example, in Crohn's disease, abdominal tuberculosis, abdominal abscess, post-radiation sequelae, diverticulitis, malignancies (gynaecologic, prostatic, rectal, and lymphoproliferative diseases), hepatitis or PID (pelvic inflammatory disease) [1,2]. A peculiar and uncommon cause of adherences is the presence of congenital bands of unknown origin.

These adherences usually remain absolutely silent and asymptomatic throughout life; however, in some patients, they can lead to an adhesive disease, usually caused by the entrapping of an intestinal segment into the band [3]. This condition can have a wide spectrum of symptoms, from mild to even life risking conditions. Consequently, the patients can suffer for a long time with mild symptoms that do not permit a correct diagnosis. In mild cases, common diagnostic procedures, such as imaging (US, CT) and laboratory examinations, can yield results in the normal range most of the time. Diagnosis represents difficult for doctors, and patients, who are subjected to inconclusive diagnostic and therapeutic attempts, and frequently became stressed [4].

Pervenuto in Redazione Agosto 2019. Accettato per la pubblicazione Settembre 2019
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The purpose of the present manuscript is to report two cases of abdominal occlusion in patients who were never operated on and who were affected by congenital adhesive bands. A literature review on the topic is also presented.

Case Reports

Case n. 1

A 76-year-old male presented to the emergency department with a two-day history of abdominal pain, nausea, and bowel closed to faeces and gas. His past medical history consisted of angioplasty, peptic ulcer treated with medical therapy, a discectomy following a posterior approach, and consequently untouched abdomen. He denied having previous abdominal surgery.
Physical examination revealed a distended abdomen with tympanic percussion and general abdominal tenderness without any rebound or guarding sign.
Laboratory examinations showed a white blood cell count of 6.60 cells/mcL with 76.80% neutrophilia. None of the test results that are usually increased in bowel infarction were elevated in our patients.
CT scan was performed and showed a mildly distended small bowel (until the distal ileum) and generalized parietal enhancement with related CT signs (Fig. 1). A short "wait and see therapy" for twelve hours was attempted; the patient was in good clinical condition, and there were no signs of needing a short-term surgical procedure. After that period, more than 500 ml of fluid drained through the nasogastric tube; therefore, surgery became mandatory. The patient was submitted to an open procedure, and part of the ileum was found trapped in the band. The intestine was brownish in colour with no signs of vitality and did not change its colour or motility even after band release, waiting for many minutes, and warm band application. Therefore, we decided to perform a small intestine resection, with termino-terminal, ileo-ileal anastomosis. The post-operative period was uneventful, and the patient was discharged after eight days.

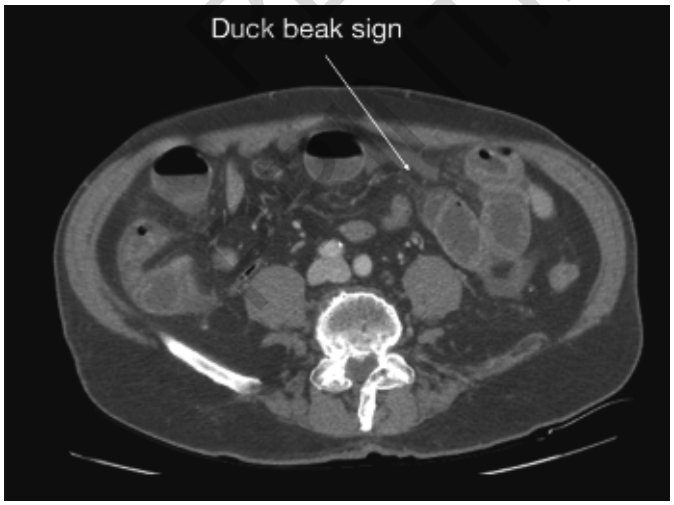


Fig. 1: Abdominal TC. Even if the fat notch, the duck beak, and the whirlpool signs indicate extra-luminal compression or rotation, CT does not provide a direct visualization of the band but only its sign.

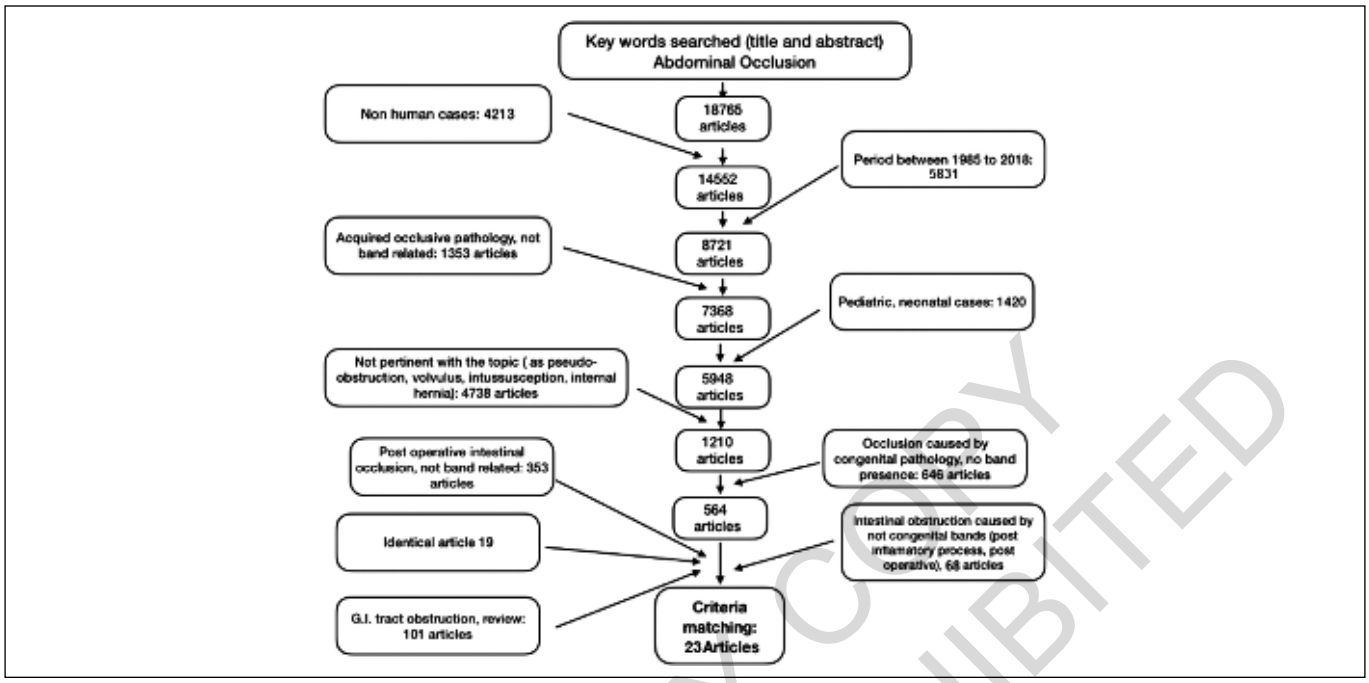
Case n. 2

A 84-year-old female presented to the emergency department with a four-day history of generalized abdominal pain (especially to superficial palpation), a distended and tympanic abdomen, nausea, and a closed bowel. Her past medical history included atrial fibrillation, which was treated with medical therapy. She denied having any previous abdominal surgery. Laboratory tests showed a white blood cell count of 6.90 cells/mcL with 80.3% neutrophilia. CT examination was performed and showed a very large hernia due to rectus muscle diastase, which was initially interpreted as the cause of the occlusion. The hernia sac was filled with proximal ileum, was distended, and had an air-fluid interface; the transition point appeared to be on the right side of the hernia opening. The patient was submitted to open surgery. During the operation, the hernia sac was opened, and a small part of the ileum emerged; this was brownish and strangled by a band not belonging to the hernia and was therefore released. The intestine resumed its natural motility and colour, and no resection was necessary. The post-operative period was uneventful, and the patient was discharged after 6 days.
A literature review was conducted in PubMed by searching for the phrase "Abdominal obstruction". This two-word phrase resulted in a vast number of results but was necessary to avoid excluding papers. Articles from 1985 to 2018 were searched. Articles in English, French and Italian were considered. Patients younger than 18, patients with previous abdominal surgery, and patients with occlusion not caused by a congenital band were excluded from the study.

Discussion

Acute small bowel obstruction (SBO) due to adhesions is a common surgical emergency. In an autopsy study of 752 cadavers, adhesions were found in 67 per cent of the subjects who had undergone laparotomy and in 28 per cent of those who had not [5].
One of the most frequent causes of SBO in patients with a virgin abdomen is congenital banding due to remnants of the omphalomesenteric duct, vitelline duct, and/or artery and/or vein. Other frequent causes are Meckel's diverticulum, remnant urachus, or Ladd's band;

TABLE I - *Flowchart of the process used to screen the literature.*



[3,6,7,8] these overlooked structures are frequent in children but are very rare in adults. An excessive development of the peritoneal folds during the process of embryogenesis is commonly recognized the underlying "primum movens".

Up to 15% of cases of Meckel's diverticulum and vitello-intestinal duct and/or artery remnants are present at the same time, although they can also develop independently of each other. Although accurate dissection and haemostasis is always advised during surgery, here, it is more important than ever because if the remnants originate in the duct, its dissection is absolutely not harmful; conversely, if the artery remnant is not accurately dissected and cauterized, this can cause low but elusive bleeding, sometimes even requiring re-operation due to significant blood loss in the drainage [9].

Two theories have also been formulated to explain SBO in patients who have never been operated on: the "ventral mesentery theory" [8], where an abnormal development of the mesentery can promote band formation, and "the intrauterine mesothelioma trauma theory" [10]; in a clever and promising study on the role of the gene therapy to prevent abdominal adhesion, trauma was shown to play a central role in adhesion formation, resulting in the latter theory. Bands may especially arise from structures with a wide surface; primarily the epiploon, peritoneum and mesentery; at other times, the digestive tract or omphalomesenteric remnants are involved.

All parts of the gastrointestinal tract can be involved, including the organs, peritoneum and/or intestine. Congenital bands cause up to 2% of all small bowel and colon occlusions, 6% of small bowel occlusions (caused by bands), and 12% of SBO and colon occlusion in patients with virgin abdomen without parietal herniation. However, only a small proportion of patients with idiopathic adhesions are symptomatic, in fact, approximately 9% of patients with SBO have undergone no previous abdominal surgery [11].

Very often, patients live their whole life without any symptoms. When symptoms appear, the onset is sometimes slow (as a cyclic pain complicating a known and long-term abdominal complaint), but usually the condition occurs suddenly, rendering treatment research a priority. The principal mechanism is sub-occlusion or plain occlusion, especially of the small intestine (SBO), the most frequently involved part of the gastrointestinal tract. The time range between symptom onset and hospital admission is between 3 and 72 hours [12]. Typical presentations include abdominal pain that is transformed within 6-12 hours to a defence reaction and diffuse pain in the whole abdomen, with possible complete bowel obstruction, which is associated with enteric vomit and swollen abdomen. The "beak sign" on CT is a well-known indicator of SBO. However, it only represents the configuration consisting of a dilated proximal intestine and transition point. Although the fat notch sign indicates extraluminal compression of the bowel by an adhesive band, CT does not provide a direct visualization of the adhesive bands [13].

In general, the management of patients with an acute episode of adhesive SBO remains controversial.[11] In 1992, Cox et al. [14] concluded that non-operative treatment with intestinal decompression using gastrointestinal tubes was safe for patients with adhesive SBO but

Table II - *All cases reported in the literature*

N°	Year	Author	P	S	A	Preospedalization symptoms	Occlusion/Bands location	Surgical Technique
1	2016	Yang[3]	1	F	75	Abdominal pain	Ileum, between mesentery-sigmoid colon	LPR with BR
			1	F	75	Abdominal pain	Terminal ileum, between ileal mesentery and terminal ileum	LPR with SR
			1	M	25	Abdominal pain	Ileum, between ileal mesentery and terminal ileum	VLS with BR
			1	M	55	Abdominal pain	Ileum, between terminal ileum and sigmoid colon	VLS with BR
			1	M	65	Abdominal pain	Ileum, between ileal mesentery and distal ileum	LPR with SR
2	2016	Nicolas[21]	1	M	33	Epigastric pain	Two band: one upper jejunum and one Treitz angle	VLS with BR
			1	M	18	Crampy abdominal pain	Band jejunum-jejunum	VLS with BR
			1	M	19	Diffuse abdominal pain	Two band: one mid jejunum and one terminal ileum	LPR with BR
3	2016	Abdelwahed[22]	1	M	56	Abdominal pain, bilious vomiting	Ileum, between gallbladder and transverse mesocolon	VLS with BR
4	2016	Pembroke[23]	1	M	57	Central abdominal pain and vomiting	Terminal ileum, two bands crossing appendix and ileum	VLS with BR
5	2016	Wettasingh[24]	1	M	69	Vomiting	Fibrous band between the Meckel's diverticulum and anterior abdominal wall	LPR with BR
6	2015	Tenreiro[25]	1	M	18	Abdominal pain, nausea, bilious vomiting	Umbilicus and the tip of the Mechel's diverticulum around which the bowel twisted.	LPR with SR
7	2015	Aranovich[26]	1	F	27	Abdominal pain, nausea, vomitig	Thick fibrous band between the root of mesentery and transverse colon causing positional anomalies	LPR with BR
8	2015	Vaziri[27]	1	M	62	Colichy, peri-umbilical addominal pain, nausea, vomiting, nogas or defecation	Band between umbilicus and urinary bladder	VLS convert LPR with BR
			1	M	42	Colicky right-sided abdominal pain, nausea, no gas.	Jejunum, band between umbilicus and bladder	LPR with BR
9	2012	Sozen[28]	1	M	22	Abdominal pain,	terminal ileum , between ascending colon and ileum	LPR with BR
			1	M	18	Abdominal pain,	terminal ileum , between ascending colon and ileum	LPR with BR
			1	F	33	Abdominal pain,	terminal ileum , between ascending colon and ileum	LPR with BR
			1	M	54	Abdominal pain,	terminal ileum , between ascending colon and ileum	LPR with BR and SR
			1	M	44	Abdominal pain,	jejunum, inter-jejunum	LPR with BR
			1	M	72	Abdominal pain,	jejunum, inter-jejunum	LPR with BR
			1	M	38	Abdominal pain,	terminal ileum, between treitz's ligament and ileum	LPR with BR
			1	M	27	Abdominal pain,	ileum, between umbilicus and ileum and Mecker's diverticulum	LPR with BR
			1	M	32	Abdominal pain,	ileum, between umbilicus and ileum and Mecker's diverticulum	LPR with BR
			1	F	66	Abdominal pain,	Ladd's band (cecum-retroperitoneum)	LPR with BR

(*segue*)

(*continua*)

N°	Year	Author	P	S	A	Preospedalization symptoms	Occlusion/Bands location	Surgical Technique
10	2013	Jalil O[29]	1	M	64	Colicky pain in the right lower quadrant, abdominal distension, bloting, pain following meals.	Ileum, between the distal ileal mesentery and the anterior abdominal wall near the umbilicus	VLS with BR
11	2012	Attaallah[30]	1	M	30	Abdominal pain, vomiting, no defecation	Jejunum, between mesentery-jejunum	LPR with BR
12	2011	Alevl[31]	1	M	23	Abdominal pain, fever, nausea, vomiting, no gas or defecation	Patent vitelline duct (between umbilicus-distal ileum)	LPR with SR
13	2011	Cartanese[32]	1	M	42	Lower quadrant and suprapubic pain, vomiting without gas or feces	Fibrous band connecting the tip of the diverticulum to the mesentery	LPR with BR
14	2010	Güner[33]	1	M	42	Abdominal pain, nausea, vomiting, no gas or defecation	Ileum, band between umbilicus and distal ileum	LPR with BR
15	2009	Zapardiel[34]	1	F	35	Abdominal pain, vomiting, 27th week of gestation.	Meckel's diverticulum	LPR with SR
16	2008	Dimitrios[35]	1	M	20	Epigastric pain, vomiting.	Jejunum, between mesentery-jejunum	LPR with BR
17	2008	Sharma[36]	1	M	24	No reported	Band between gangrenous Meckel's diverticulum and the mesentery	LPR with SR
18	2007	Markogiannakis[37]	1	F	20	Colicky abdominal pain, vomiting, no gas or defecation, abdominal distension.	Persistent omphalomesenteric duct (between umbilicus-distal ileum)	LPR with BR
19	2007	Hinkle[38]	1	M	22	Crampy abdominal pain.	Ladd's bands between right colon and right parietal peritoneum ????	LPR with SR (right colectomy)
20	2005	Wu[39]	1	M	33	Epigastric pain, vomiting	Ileum, between omentum and ileum's mesentery	VLS with BR
21	2003	Habib[20]	16	8M 8F	59	Abdominal pain	Epiploon-mesentery 6 cases, Inter Omentum 4 cases, Epiploon-mesentery 2 cases, Epiploon and anterior peritoneum 1 case, Mesentery-colon 1 case, Intermesentery 1 case, Mesentery and posterior peritoneum 1 case, Ileo-colon 1 case, Ileo and anterior peritoneum 1 case	7 cases LPR with BR 5 cases LPR with SR 4 cases no specified
22	2003	Amendolara[40]	1	F	84	Abdominal pain, nausea, vomiting, no gas or defecation, abdominal distension.	Persistent omphalomesenteric duct (between umbilicus-Meckel diverticulum)	LPR with SR
			1	M	20	Right lower quadrant	Persistent omphalomesenteric duct (between umbilicus-Meckel diverticulum)	VLS convert LPR with BR and SR
23	2001	Prall[41]	1	M	31	Periumbilican abdominal pain, vomiting	Meckel's diverticulum adhesed to the mesentery underlying	LPR with BR diverticulum section
24		Present cases	1	M	76	Abdominal pain, nausea, no gas or defecation	Middle ileum, between mesentery - jejunum	LPR with BR and SR
			1	F	84	Abdominal pain, distended and tympanic abdomen, nausea, no gas or defecation	Middle ileum, between mesentery - jejunum	LPR with BR

Legend: P: patient; S: sex; A: age; M: male; F: female; LPR: laparotomy; VLS: videolaparoscopy; BR: band releas; SR: segmental resection.

without signs of strangulation. In addition, this type of management did not lead to an exaggerated delay in the surgical treatment of patients with small bowel strangulation [14]. Consequently, patients with clear signs and symptoms of partial or complete SBO but without fever, leukocytosis, tachycardia, metabolic acidosis and continuous pain or peritonitis can be safely treated with non-operative management. Moreover, the administration of water soluble contrast (Gastrografin) in patients with partial SBO can improve bowel function and reduce length of stay and is therefore both therapeutic and diagnostic [13]. The failure rate of conservative management is between 20% and 30%, and this is represented by a lack of clinical improvement or signs of strangulation [15,16]. As a result, if ileus persists for more than 3 days and the drainage volume on day 3 is > 500 ml, surgery is recommended[17]. Laparoscopy has also changed the diagnostic steps before surgery. Indeed, many patients can not only obtain a diagnosis but also be simultaneously cured. Moreover, surgical sequelae are absolutely inferior, making the procedure easily accepted by the patients and aiding in diagnosis. When operative treatment is required, surgeons can choose between laparoscopy and open surgery. Laparoscopic adhesiolysis for small bowel obstruction has several advantages: less postoperative pain, faster return of intestinal function, shorter hospital stay, reduced recovery time, more rapid return to full activity, fewer wound complications, and decreased postoperative adhesion formation [18]. Although laparoscopic adhesiolysis has many positive aspects, the surgeon's ability to perform advanced laparoscopy should first be considered [17]. Then, adequate patient selection is fundamental [18]. Inclusion criteria for laparoscopic management are as follows: mild abdominal distention allowing adequate visualization, a proximal obstruction, a partial obstruction, and an anticipated single-band obstruction (indirect CT signs of its presence) [15]. In cases of extensive adhesions, distended small bowel, free air and devascularized bowel, laparotomy or early conversion from laparoscopy to an open approach are advised [17,19].
In addition, if an immediate surgical intervention is performed, almost 50% of patients have advanced necrosis of one or more intestinal segments, making resection mandatory [20].
Suspecting adhesions in adults without any history of previous surgery or abdominal trauma is often challenging[21]. This is the main reason why these pathologies often receive delayed treatment, and diagnosis is usually erified intra-operatively. Laparoscopy is a very valuable tool for this purpose.
Table I shows a flowchart of the process used to screen the literature. In the period studied, only twenty-three papers matching the criteria for the present study were found[3,20-41]. Seventeen of these were single case reports. The wider series included 16 and 10 cases.
The total number of cases reported was 57, including 40 males (70.2%) and 17 females (29.8 %). The mean age was 31.8 years (range, 18 to 84 years). The most common location was the middle ileum. In four cases (7.0%) the surgical procedure was not reported. In the remnant 53 cases (93.0%) the most common surgical approach was laparotomy (43 cases; 81.1% of the total). The laparoscopic approach accounted only for 10 cases (18.9%) of the total. Two cases (3.8%) were converted from laparoscopic to laparotomy. Band releas was necessary in 37 cases (69.8%). Bowel segmental resection was reported in 13 cases (24.5%). Band release and bowel segmental resection was done in 3 cases (5.7%). Meckel's diverticulum resection was necessary in 1 case (1.9%), and right colectomy was done in 1 case (1.9%). Details regarding all patients reported in the literature are summarized in Table II.

Conclusion

Abdominal occlusion in surgically virgin abdomen caused by congenital bands is very rare.
Based on our experience with the two cases reported here and the literature review, the diagnosis should be suspected even if CT scans and laboratory tests are not mandatory for surgery. Clinical evaluation is an important step for surgeons, especially when the 500 ml fluid limit is met. If this amount is gathered within 24 hours, it incontrovertibly shows the necessity of an urgent surgical procedure. Laparoscopy is currently a fundamental diagnostic and therapeutic tool. If the surgical approach is mandatory, laparoscopy has to be at least attempted; however, if this is not feasible, open surgery remains a valid and resolutive option.

Riassunto

Le aderenze addominali congenite sono molto rare, solitamente asintomatiche tali da passare misconosciute per tutta la vita. In rari casi, tuttavia, possono manifestarsi con sintomi clinici. La diagnosi è molto impegnativa poiché il paziente non riporta in anamnesi interventi o esami radiologici addominali pregressi. Inoltre gli esami ematochimici e l'esame clinico possono essere non indicativi o non specifici.
Due pazienti sono stati ricoverati e operati presso la nostra unità operativa complessa di Chirurgia Generale tra giugno 2017 e luglio 2018. Entrambi i pazienti presentavano un quadro di occlusione intestinale al momento del ricovero con alvo chiuso a feci e gas. In un caso è stata tentata la terapia conservativa. Entrambi i pazienti sono stati sottoposti a trattamento chirurgico che ha evidenziato la presenza di una briglia aderenziale congenita, causa dell'occlusione. Il decorso post operatorio è stato regolare in assenza di complicanze.
L'ostruzione dell'intestino tenue (SBO) causata da briglie aderenziali congenite è una patologia rara rappresentante una sfida per il chirurgo. La diagnosi di SBO

deve essere sospettata, anche quando gli esami radiologici non evidenziano alcuna patologica responsabile del quadro occlusivo intestinale in pazienti che non sono mai stati sottoposti a chirurgia addominale. La laparoscopia va utilizzata come primo approccio, riservandosi comunque la possibilità della chirurgia.

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