

A rare nosological entity The perforated solitary cecal diverticulum

Research article



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AIM: To describe three cases of solitary cecal diverticulum, and trying to evaluate the better method of diagnosis and treatment with analysis of the literature.

MATERIAL OF STUDY: Description of three cases of solitary cecal diverticulum's perforation admitted in the Department of General and Oncologic Surgery, Santa Maria della Misericordia Hospital, Perugia, during the period January 2011 – January 2012.

RESULTS: In all patients the clinical presentation was very similar to that of acute appendicitis. Preoperative diagnosis was achieved in one case through abdominal CT scan, other two cases were identified at final pathology. At one year from the treatment all patient are still alive.

DISCUSSION: Cecal diverticulum is a rare condition, often diagnosed either casually or because of inflammatory or perforative complications. The highest incidence is found in Western population. Because of the clinical presentation, very similar to the appendicitis, and the inflammatory reaction involving the colon and its surrounding tissues, the pre- and intra-operative diagnosis are very difficult. The diagnosis is almost always histological. The treatment may vary from simple expectant medical management, carried out with bowel rest, parenteral support and antibiotics as for left-sided diverticulitis, to surgical approach, performed through simple diverticulectomy or by classical right hemicolectomy.

CONCLUSION: Pre-surgical and, also intra-operative, diagnosis of perforated solitary cecal diverticulum is clearly difficult. CT scan represents the gold standard for the differential diagnosis. Right hemicolectomy is an effective and safe approach, allowing accurate control, preventing complications and recurrences, and it represents the optimal management of the disease.

KEY WORDS: Diverticular disease of right colon, Perforation of solitary cecal diverticulum, Right hemicolectomy

Introduction

Diffuse diverticulosis of the right colon is common in Asiatic countries, while solitary diverticulum of the right

colon as well as diverticular disease of the left colon occur more frequently in the West^{1,2}. Solitary cecal diverticulum is a benign but rarely observed condition. Most of times, this condition remains unnoticed and is discovered by accidental diagnosis or as a result of complications as inflammation, bleeding or perforation.

The solitary diverticulum of right colon was described for the first time as an independent nosologic entity by Potier in 1912³. Since then, only approximately 900 cases have been reported in literature, testifying the rarity of such condition.

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Waugh⁴ in 1941 attributed embryological origin to cecal solitary diverticula. According to this theory, during the embryological development of the gut, an outgrowth from the tip of the caecum would occur in the 6th week of gestation, which would atrophy towards the end of the 7th week. This structure has been called “transient appendix” and doesn’t show any relation to the permanent appendix, which differentiates much later.

The solitary cecal diverticulum is often misunderstood. It may be casually diagnosed during a barium enema undertaken for other reasons. Usually asymptomatic it may cause inflammatory, haemorrhagic, or perforative complications thus leading to urgent laparotomy.

Diagnostic dilemma is not easy to resolve because of the clinical presentation overlapping with that of acute appendicitis^{5,6}: fever, abdominal pain mainly localized in right iliac fossa, signs of peritonism/peritonitis, nausea mostly without vomiting, neutrophil leucocytosis.

Intra-operative findings are often unclear, in relation to the remarkable inflammation and to the presence of a mass difficult to distinguish from a neoplastic process. For these reasons is difficult to choose a conservative surgical approach, furthermore considering that final diagnosis is only post-operative, which means histological.

The treatment of right colon complicated solitary diverticula ranges from conservative approach, with bowel rest, appropriate hydro-electrolytic infusion and antibiotic administration, as for left colon diverticular disease, to emergency surgery². The surgical options are diverticulectomy, wedge resection and right hemicolectomy, but, for the above mentioned reasons, the third option is the more frequently adopted.

In our study we considered three cases of perforated solitary diverticulum of right colon, confused with appendicitis. The scope of our study is to illustrate these three cases to demonstrate the difficulty of preoperative diagnosis, and to illustrate the correct management of such kind of patient.

Cases Report

At the Department of General and Oncologic Surgery, *Santa Maria della Misericordia* Hospital, Perugia, from January 2011 to January 2012 three patients with perforated solitary cecal diverticulum have been admitted, all of them suspected for acute appendicitis.

The first patient was a 50-year-old woman, without relevant anamnestic data, who reported the arise of periumbilical abdominal pain, afterwards localized in the right iliac fossa, not associated with nausea nor vomiting, fever or other gastrointestinal disturbances. On physical examination the abdomen was painful but soft and tender at light and deep palpation of right quadrants, with lightly positive Blumberg’s sign. The peristalsis was present and normal, percussion revealed hyper tympanic. Haematological and biochemical exams showed neu-

trophil leucocytosis (12.20×10^3 WBC) (78.9% N). Abdominal X-ray revealed “gaseous distension of some loops of small bowel with air fluid levels”. Ultrasonography showed a situation attributable to inflamed diverticulum. According to the ultrasonography report, in order to dissipate any diagnostic doubt, the patient was submitted to abdominal CT scan which confirmed the presence of inflamed diverticulum of the ascending colon, with initial abscess. According to clinical and radiologic findings, the patient was submitted to urgent laparoscopy, with detection of a mass of about 5 cm in diameter, increased in consistence, partially infiltrating the kidney capsule, but still dissociable, presumably inflammatory, but whose malignant nature couldn’t be excluded. Therefore laparoscopy was converted to laparotomy through a midline xifo-pubic incision, and performing a right hemicolectomy with mechanical ileocolic end-to-side anastomosis, placement of two drainages: one in the paracolic gutter to protect the anastomosis and the other in the pelvis. Post-surgical course was regular. The patient was dismissed in 11th post-operative day. The final pathology evidenced “diverticular disease complicated by abscessualization, perforation and peritoneal reaction”. The patient had a sub-occlusive episode requiring hospitalization about three months after the operation, she was treated conservatively with

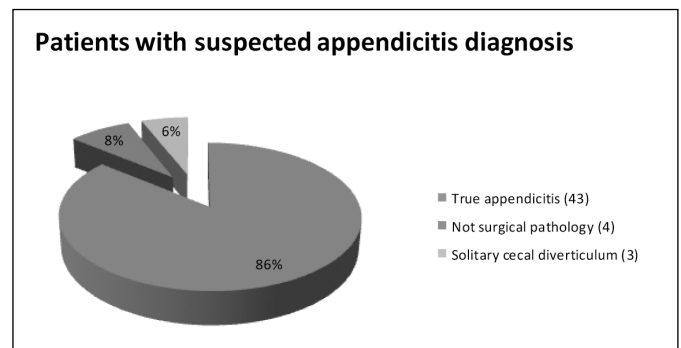


Fig. 1

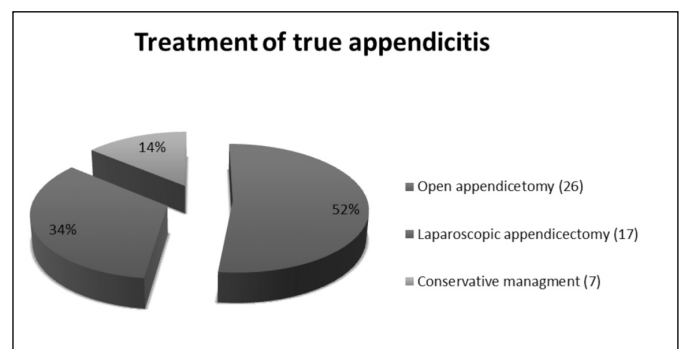


Fig. 2

enemas, vaseline oil and semiliquid diet, then dismissed after six days in good conditions. At the present time she is still in good conditions and the symptomatology hasn't recurred.

The second patient, a male of 37 years was admitted because of continuous right located abdominal pain, together with nausea, light fever (37.4°C), and preserved bowel function. The abdomen was soft, painful on the left side, tender at light and deep palpation on the right side and on the right iliac fossa, with negative Murphy sign and markedly positive Blumberg sign. Peristalsis was present and bowel sounds were not tympanic on auscultation. Haematological and biochemical exams showed neutrophil leucocytosis (14.46×10^3 WBC; 79% N), the remaining parameters were normal. Abdominal sonography was normal. Because of the non-regression of the symptomatology, in the suspicion of an acute appendicitis, urgent surgery was undertaken. Initially, a McBurney incision was carried out and a corpuscular purulent-like liquid was observed in the abdomen. It was difficult to externalize the cecum, at which level a thickening of the posterior wall and conglomerated epiploic fringes were found. Because of the difficulty of an accurate vision of the operating field, a midline laparotomy was made. Then a right hemicolectomy, with mechanical ileocolic end-to-side anastomosis, was performed. The appendiceal serosa appeared lightly erythematous as consensually inflamed. After this, the placement of a drainage in the right paracolic gutter to protect the anastomosis was performed. The post-surgical course was regular. The patient was dismissed the 9th post-operative day, afebrile, with regular bowel function, with a diagnosis of acute abdomen caused by covered caecum perforation and retrocecal abscess. Histological examination revealed diverticular inflammation, with areas of abscessualization and consensual peritonitis. The appendix was normal. At the present time the patient is in good clinical conditions and the symptomatology hasn't recurred.

There weren't post-surgical complications either immediate or at distance.

The third patient, a 51-year-old-woman, affected with anxious-depressive syndrome, was admitted by the ER with a diagnosis of "acute appendicitis". The patient reported the recent arising in the morning of periumbelical pain subsequently localized in the right iliac fossa, associated with fever and nausea without vomiting, and no changes in bowel function. Physical examination revealed soft abdomen but painful and tender at light and deep palpation on the right quadrants, Blumberg positive sign, normal findings on percussion and auscultation. Body temperature was 38.2°C. Haematological profile showed neutrophil leucocytosis (21.05×10^3 WBC; 76% N). Abdominal US evidenced an elongated hypo-anechoic mass in right iliac fossa (diameter 56x18 mm), that was uncompressible, highly suspected for appendicular phlegmon. Thus, the patient underwent urgent exploratory laparoscopy, that evidenced uninjured appendix and presence of corpuscular liquid in the right iliac fossa due to inflammatory fibrinopurulent peritonitis with tenacious adhesions between the last ileal loop and the caecum, therefore was decided to proceed with conversion to midline laparotomy. The exploration of the abdominal cavity confirmed the extended inflammatory process encompassing the caecum and the last ileal loop, with thickened mesentery and consensual lymphadenopathies. After careful peritoneal toilet, an oval mass was evidenced in the anterior wall of the caecum, about 3 cm in dimension, with hard-elastic consistency and no plane of cleavage by the colic wall. Therefore, right hemicolectomy was performed, with restoration of intestinal continuity through ileocolic, manual, side to side, isoperistaltic anastomosis and placement of two drainages, one to protect the anastomosis and the other in the pelvis. The post-surgical course was regular, with patient's discharge in the 9th post-operative day. Histology evidenced, near the ileo-cecal valve, a saccular

TABLE I - *Four variants of colon diverticular disease by Perry and Morson.*

	1	2	3	4
Description	Sigmoid colon diverticulosis with muscle abnormalities	Partial or total colonic diverticulosis without muscle abnormalities	Cecal or ascending colonic solitary diverticulum	Right colon diverticular disease
Colic localization	Left – Sigmoid colon	All the colon	Cecal or ascending colon	Right colon
Population with higher incidence	Caucasian	Caucasian	Caucasian	Asian
Origin of the diverticula	Acquired	Acquired	Congenital	Acquired
Type of diverticula	False	False	True	False
Number of diverticula	Multiple	Multiple	Solitary	Multiple

TABLE II - Description of the three patients analyzed.

	Patient 1	Patient 2	Patient 3
Sex	Female	Male	Female
Age	50	37	51
First diagnosis	Appendicitis	Appendicitis	Appendicitis
Final diagnosis	Solitary cecal diverticulum	Solitary cecal diverticulum	Solitary cecal diverticulum
Diagnostic exam	CT scan	Histology	Histology
Treatment	Right hemicolectomy	Right hemicolectomy	Right hemicolectomy

TABLE III - Results.

	Patient 1	Patient 2	Patient 3	Average
Hospitalization time	11	9	9	9.6
Removal of bladder catheter	3 days after operation	2 days after operation	4 days after operation	3 days after operation
Removal of nasogastric tube	5 days after operation	2 days after operation	2 days after operation	3 days after operation
Number of drainage	2	1	2	
Removal of drainage	6 and 8 days after operation	5 days after operation	4 and 5 days after operation	5.6 days after operation
Defecation time	5 days after operation	4 days after operation	5 days after operation	4.6 days after operation
Feeding	7 days after operation	5 days after operation	5 days after operation	5.6 days after operation
Complications within 30 days	No one	No one	No one	

evagination of the bowel wall (diameter 5.5 cm), containing fecal material, cecal mucosa erosion and acute transmural inflammation with abscess, peritonitis and consensual peri-appendicitis.

Results

From January 2011 to January 2012, among sixty patients with a suspicion of acute appendicitis twenty-four were males and thirty-six females (mean age 32 years). Forty-three patients have been surgically treated, twenty-six with open technique and seventeen in laparoscopy. Seven have been conservatively managed. Four patients transferred from the ER with suspect appendicitis diagnosis, were not affected by a surgical pathology, but by urinary inflammation or gastroenteritis. The three remaining patients had diverticular perforation misdiagnosed as appendicitis: two females and one male of 51, 50 and 37 years respectively (mean age 46 years). Two out of them three had a rise in body temperature, as two had nausea without vomiting, while the third didn't show neither nausea nor vomiting. All three patients didn't refer about bowel function alterations, in fact peristalsis was present and abdomen was soft even if in those cases with positive Blumberg sign. In all three cases neutrophil leucocytosis was the unique abnormal laboratory value.

Just in one case a pre-surgical diagnosis was achieved, through execution of abdominal CT scan, in the other

two it was defined by histology. Patients were subjected to urgent right hemicolectomy, either due to the impossibility to exclude a neoplastic process, or because of the inflammatory bowel state, contraindicating more restricted resections, in relation to the risk of post-operative complications such as fistulas or anastomotic leakage. Initial laparoscopy was necessarily converted to laparotomy in two cases, and McBurney incision in a median incision due to the need of a wide view and perfect control of the operating field. In all the three cases, the post-surgical course was regular, without adverse events or complications, except for a sub-occlusive episode in one of them.

The total hospitalization time, overlapping the post-surgical hospitalization time because of the urgent surgery was performed at the time of the hospitalization, was respectively of 11, 9, 9 days with an average length of hospitalization of 9.6 days. Bladder catheter and nasogastric tube were always positioned and both removed after an average of three postoperative days. The mean permanence of the drainages was of 5.6 post-operative days. Postoperative return of bowel sounds, gas passage and first spontaneous feces were analyzed, finding an average time to the first postoperative stool of 4.6 day.

The patients gradually restarted to eat, respectively in 7th, 5th, 5th postoperative day, when in all the three cases the patients were canalized, with an average of 5.9 days, beginning with a water diet, then continuing with plain soup, and, after, passing to minced meat and side dish. No immediate or distant complications occurred.

Discussion

Solitary true diverticulum of the right colon is a very rare finding in the clinical practice, with no more than 900 cases described, since Potiers' first description² of perforated caecal diverticulum in 1912. Williams⁷ in 1960 attributed an incidence of 0.1% to right colon solitary diverticula and of 5% to diffuse colic diverticulosis. Left sided diverticular colic disease as well as cecal solitary diverticulum are more common in the Western World², while right sided diverticulosis is rather uncommon in the West but may reach very high frequencies in Asian countries, especially Japan¹.

The reason for this is to be found in the different aetiologies of the various disease forms, with detection of much higher right colon intraluminal pressures in the Eastern populations⁸.

In fact Graham and Ballantyne⁹ found in Western populations that most of right colon diverticula were solitary, with a percentage around 81% of all patients with right colon diverticular disease.

As regards the aetiology, an embryologic origin was attributed to the solitary cecal diverticulum, in contrast to diverticulosis of left and right colon that would be acquired. Already Waugh⁴ in 1941, and later other Authors¹⁰ brought back the solitary cecal diverticulum to an embryological origin.

According to this theory, an outgrowth from the tip of the caecum occurs in the embryological development of the gut during the first six weeks and atrophies towards the end of the seventh. This structure has been referred to as the "transient appendix" and it is probably an independent structure not connected with the permanent appendix vermiformis, which is differentiated later.

Solitary cecal diverticula are therefore congenital and true, thus consisting of all layers of the colic wall, including muscle layer, as opposed to false diverticula, characteristic of diffuse acquired diverticulosis, in which only mucosa and submucosa herniate through the weak points of colic wall.

Single true diverticular disease equally interests both sex, affecting younger patients than diffuse diverticulosis⁵ but older if compared to appendicopathies, even though its incidence does not increase with age¹¹.

The correct diagnosis of solitary cecal diverticulum is rarely made¹², sometimes as an accidental finding of an x-ray contrast performed for other reasons, or as a consequence of its complications.

In the last case, the diagnostic difficulties should be also attributed to a clinical presentation, which is totally similar to that of the acute appendicitis⁵ with fever, neutrophil leucocytosis, signs of localized peritonitis, pain and sometimes a palpable mass in the right iliac fossa. In presence of the aforementioned clinical signs, diagnosis is obviously much easier in appendectomized patients.

Secondly we should pay attention to the fact that the onset of pain in the case of diverticular perforation is

more rapid and the symptoms of pain last longer; generally it doesn't start in the middle of the quadrants moving afterwards towards the right iliac fossa, but it starts directly at this level¹³.

Among the presenting symptoms, nausea and, in particular, vomiting are rarer than in acute appendicitis^{14;15}, besides, it's not uncommon, at abdominal physical examination, to palpate an hard-elastic mass in right iliac fossa compatible with the ongoing inflammatory process⁶, but not allowing to exclude an abscessualized colon cancer.

Concerning preoperative diagnosis, if patient's clinical conditions allow it, a double-contrast barium enema can be performed in the suspicion of covered perforation, or a CT scanning, mostly useful in resolving doubts among appendicitis, diverticular disease and neoplastic process¹⁶. Right colon diverticular perforation necessarily arises in the differential diagnosis with several other morbid conditions⁶, such as angiodysplasia¹⁷, solitary cecal ulcer, first manifestations of Crohn's disease, right tubo-ovarian disease, right kidney and ureter disease, cecal perforation by foreign body, or even abnormal irradiation of gallbladder pain.

Generalized peritonitis, recurrence of symptoms before the final diagnosis and intestinal obstruction are rare manifestations of right colon diverticular disease and of single congenital perforated diverticula.

Intraoperative diagnosis is also complex, as often burdened by advanced peritonitis, that does not allow an easy identification of the triggering cause. It is difficult to distinguish whether a hard-elastic mass adherent to surrounding structures and with no clear cleavage plane from them, might be either inflammatory or neoplastic in nature. Indeed, the surrounding structures, omentum and lymph nodes are involved in both cases.

When a neoplasia is suspected it is useless to proceed to an intraoperative biopsy, because, even in the case of cancer-abscess, the perilesional tissue may be reactive in nature and therefore not diagnostic.

Easier is intraoperatively the exclusion of appendicitis, appendix is indeed almost always free of macroscopic disease, or at most appears slightly erythematous, as if by consensual inflammatory process. Intraoperatively gynaecological pathologies or presence of foreign bodies should also be sought and excluded.

Therapeutic approach to solitary right colon diverticular perforation may vary from conservative treatment, based on bowel rest, fluids and antibiotics as for left colon diverticulitis, to surgery^{2;5}, which can consist of simple prophylactic appendectomy with abdominal cavity toilet, diverticulectomy, segmental resection of the colic tract affected by diverticulitis, up to right hemicolectomy.

The majority of authors¹¹ suggest an aggressive approach, by performing in most cases a classic right hemicolectomy, in order to resect all macroscopically affected tissue. At least three different reasons motivate us to support this surgical attitude. First, most of surgical inter-

ventions are performed in emergency, condition in which it is not always possible to exclude malignancy^{9,11}, even for unavailability of intraoperative biopsies. Therefore the execution of a right hemicolectomy with lymphadenectomy appears justified in these cases, as it is an oncologically correct surgical procedure. Another motivation is the high rate of symptoms recurrence if the cause, namely the diverticulum, is left in place^{2,4,18}, and this is especially true for solitary right colon diverticula, which mainly affect Western populations. A third reason is the prolongation of hospital stay when a conservative strategy is undertaken, because of the need to extend the antibiotic treatment³.

On the other side, a conservative approach is preferable in the case of right sided diverticular disease, which is typical of Asian populations⁴, when a certain diagnosis of uncomplicated diverticulosis is established.

Over the years, right hemicolectomy has become a safe procedure even when performed in emergency, realizing the anastomosis in a recent site of inflammation, without colic preparation. Morbidity and mortality are negligible, the mortality rate defined by Lane is around 1.4%¹¹.

According to the literature the pathologies concerning the solitary diverticula of cecum and ascending colon, though very rare, should always be considered in the differential diagnosis of acute appendicitis^{2,6}.

The surgeon should know how to manage and deal with it, even if the pre and intraoperative diagnosis are complex due to the inflammatory status of the colon and surrounding tissues⁵.

Right hemicolectomy is the treatment of choice for the above-mentioned reasons. The patients we treated came to our attention with a diagnosis of suspected acute appendicitis and all of them underwent a right hemicolectomy. Diagnosis is mainly histological, then posthumous. In one of our cases, diagnosis was preoperative, while in the other two it was revealed at histologic tissue analysis. The post-surgical course was regular, without medium and long-term complications, the average length of hospital stay did not exceed 10 days. At the present time all of them enjoy good health conditions, and symptomatology did not recur.

Conclusions

There is a need for the surgeon in the West, to become familiar with right colon diverticular disease, especially concerning the solitary diverticulum. This rare pathology should always be taken into account in the differential diagnosis of right iliac fossa pain, and when suspected investigated through abdominal CT scan, which is often decisive. Intraoperative diagnosis is often complicated by concurrent peritonitis, and emergency surgery should not always allow distinguishing diverticulitis from malignancy, thus leading to perform right hemicolectomy. Furthermore, such procedure is effective to defini-

tively treat the pathology and burdened by low morbidity and mortality. Our three patients, hospitalized with misdiagnosis of acute appendicitis, underwent right hemicolectomy with regular postoperative course and only one of them presented a sub-occlusive episode at distance, conservatively treated. The hospitalization time doesn't exceed ten days and all of three patients restart to eat in no more than 6 days. Therefore, we recommend to carefully consider this disease, to attempt in its suspicion preoperative diagnosis also through abdominal CT, and to perform right hemicolectomy in emergency.

Riassunto

Dal nostro studio emerge la difficoltà di diagnosi preoperatoria nel caso di diverticolo solitario del ceco perforato, anche se nel dubbio la TC addome è risultata essere l'esame dirimente per la diagnosi. Altrettanto complessa è la stessa diagnosi intraoperatoria. Per tale motivo al momento dell'atto chirurgico si è preferito essere aggressivi, e condurre una emicolectomia destra, non potendo escludere la patologia neoplastica o la sicurezza nell'eseguire la semplice diverticulectomia, dato lo stato peritonitico in cui ci si è ritrovati ad operare. Tale approccio, come anche testimoniato da altri studi sull'argomento, si è dimostrato efficace, sicuro ed è risultato una soluzione definitiva della patologia diverticolare, la quale ha mostrato un alto tasso di complicanze e di recidive nei pazienti in cui l'approccio è stato più conservativo.

Competing interests

The Authors state that none of the authors involved in the manuscript preparation has any conflicts of interest regarding the manuscript itself, neither financial nor moral conflicts. Furthermore, none of the authors received support in the form of grants, equipment, and/or pharmaceutical items.

Authors' contributions

All authors contributed equally to this work, read and approved the final manuscript.

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