Acute symptomatic Meckel diverticulum management Our experience on seven consecutive cases



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Seven consecutive cases have been chosen: five males (71,4%) and two females (28,6%). The age ranges over from 13 to 50 years with a 28 years average. Four of them were submitted to emergency surgical intervention for hemorrhage from gastro-enteric tract (57%), two for bowel obstruction (29%) and one for acute appendicitis (14%). In all cases sample was send to histological examination. Two samples showed normal epithelial mucosa. Four of them showed ectopic mucosa inside the diverticulum: three gastric and one pancreatic ectopic mucosa focal areas. The last case showed normal epithelial cells but with ulcerated and hemorrhagic areas. Four samples of patients with hemorrhage from gastro-enteric tissue and the last with normal mucosa but ulcerated and with bleeding areas. In our experience we never speculated that acute symptomatology depended on complicated MD and diagnosis was always done during laparotomy. We think that MD removal is always the correct choice, so that future complications such as neoplasm can be avoided. MD simple resection by Stapler at the base of diverticulum is the correct choice.

KEY WORDS: Bowel obstruction, Enterorrhagia, symptomatic, Meckeldiverticulum

Introduction

Meckel's diverticulum (MD) is the most common congenital anomaly of the gastrointestinal tract, with an incidence on autopsy of 0.3-4%. This condition derives from incomplete obliteration of the omphalo-mesenteric duct. Meckel's diverticulum is a true diverticulum and it is about 3-5 cm long, with all of the layers of the intestinal wall present, and it is located on the antimesenteric side of ileum, about 40-130 cm proximal to the ileocecal valve. Since cells of yolk-sac are pluripotent, in the diverticulum may be present gastric (50%), pancreatic (5%) and less frequently colonic, endometriosis, and hepatobiliary heterotypic mucosa. This kind of tissue, if present, is responsible for complications such as hemorrhage, ulcers and perforation. A person having MD

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through his life has a risk of 4-6% to develop a complication that in 25-50% of cases is going to be a bleeding 1,2 . Hemorrhagic event is almost always resulting in the ulceration of gastric heterotypic mucosa and, in uncommon cases, of pancreatic one 3 .

In this report seven consecutive clinic cases presented in emergency area for acute abdomen successively attributed to complicated MD.

Diagnosis of symptomatic MD had never been done before surgical operation and diverticulum was always discovered during operation.

Patients and Methods

We revalued clinical records of patients discharged from Unit of Urgent and General Surgery of Highly Specialized Hospital "A.O.R.N. Antonio Cardarelli" of Naples with diagnosis of acute pathology associated to complicated MD from 1st January 2011 to 30th November 2012.

Results

Seven consecutive cases have been chosen: five males (71,4%) and two females (28,6%). The age ranges over from 13 to 50 years with a 28 years average. Four of them were hospitalized in our emergency room (57%) and three were transferred from other hospitals (43%). Four of them were submitted to emergency surgical intervention for hemorrhage from gastro-enteric tract (57%), two for bowel obstruction (29%) and one for acute appendicitis (14%).

In five cases MD was resected with Stapler, in two by wedge resection and reconstruction of bowel by hand by end to end anastomosis. In all cases sample was send to histological examination.

Two samples showed normal epithelial mucosa (28,6%). Four of them (57,1%) showed ectopic mucosa inside the diverticulum: three gastric and one pancreatic ectopic mucosa focal areas. The last case (14,2%) showed normal epithelial cells but with ulcerated and hemorrhagic areas. (Table I). Four samples of patients with hemorrhage from gastroenteric tract showed at histological examination: a case of normal mucosa, a case of gastric mucosa areas, one of pancreatic ectopic tissue and the last with normal mucosa but ulcerated and with bleeding areas.

Nobody of patients referred to be submitted to medical therapy with anticoagulants or antiplatelets. Only one reported chronic liver disease.

Colonoscopy, performed upon all patients hospitalized for hemorrhage in the gastro-enteric tract, always excluded the presence of bleeding points in the colon, but was never useful to directly show the bleeding diverticulum. Arteriography was performed only once, but didn't succeed in finding the source of bleeding.

TC-scan, to which the two patients hospitalized for bowel obstruction were submitted, never allowed us to directly visualize the MD, even though it showed the indirect radiological findings of bowel obstruction (liquid in abdomen, distension of intestine etc.)

CASE A (F.V.)

24-years-old female presents to emergency room for abdominal aches appeared a few hours before, followed by enterorrhagia. In the medical history was absence of previous illnesses and no recent administration of drugs potentially injurious for mucosa of gastroenteric tract. She's submitted to first-level instrumental diagnostic procedure (chest rx-graphy, abdomen rx-graphy, and ultrasound abdomen scan) that show no notable pathology. Blood tests show significant anemia with 5.6 g/dL as level of hemoglobin, without alteration of coagulation factors. She's transfused with concentrated red corpuscles and submitted to both gastroscopy and colonoscopy. Gastrosocopy didn't show hemorrhagic areas in the upper gastrointestinal tract, while colonoscopy, with exploration of terminal ileum tract, excluded bleeding areas in the colon, and described blood presence in it.

The patient is submitted to abdomen high resolution CT-scan with contrast medium and CT angiography to research the hemorrhagic pathology. Examination doesn't show the bleeding point and for this reason we performed an urgent exploratory laparotomy. During procedure we found an MD with active bleeding that is

TABLE	I
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Case	Sex	Age	Cause of surgical interventation	Epithelial features of MD referted by istological examination
A	F	24	hemorrhage	Focal areas of pancreatic tissue
В	М	25	Bowel obstruction	Normal enteric type
С	М	13	Bowel obstruction	Focal areas of gastric mucos
D	F	18	hemorrhage	Focal areas of gastric mucosa
Е	М	50	hemorrhage	Normal but ulcerated and presenting hemorrhagic areas
F	М	18	hemorrhage	Normal enteric type
G	М	48	appendicitis	Focal areas of gastric mucosa

resected with a GIA 60 stapler and vicryl over lock. Histological examination showed ectopic small areas of pancreas into the resected diverticulum.

CASE B (R.F.)

25-years-old male presents to emergency room with acute abdomen. Patient referred that he had already been feeling abdomen pains since a little time, and that for this reason he had been submitted to colecistectomy for stones, because it had been thought that abdominal pains resulted by them. During abdominal examination, pain appeared in the lower abdominal area with Murphy's point painless. She's submitted to first-level instrumental diagnostic procedure and blood tests without significant alterations. Abdomen RX-graphy didn't show any disease while abdominal ultrasound scan showed a little effusion in abdominal cavity. Suspecting this was a complication depending by previous surgical operation, we decided to submit the patient to a CT-scan with contrast medium. It showed the presence of liquid distributed in the whole abdominal cavity with a tract of invaginated ileum to which associated an upper ileum gaseous distension. Therefore the patient is submitted to explorative laparotomy that allows us to find an ileal invagination caused by the MD. On its tip a solid tumefaction is found and considered a neoplasm. We performed wedge-resection of bowel with end-to-end handmade reconstruction. The report of histological examination asserts that "ileal tract showed ischemic alterations. The MD results chronically inflamed. Epithelial cells are normally enteric. The neoplasm on the tip results to be constituted by cells of myofibroblastic proliferative type.

According to previous illnesses and operative results we can speculate that patient had several episodes of abdominal pain because of various occluding invaginations of bowel and that stones in the gallbladder were wrongly considered as the reason for the abdominal pain.

Case C (R.G.)

13 years old boy presents to emergency room for acute abdomen appeared about ten hours before. There are no significant diseases in his medical history. During examination abdomen hurts above all in the right side, but there aren't any symptoms of peritonitis. Patient did not referred any alterations of regular bowel activity. He was submitted to first-level diagnostic instrumental diagnostic procedure and to urgent blood tests. RX-graphy showed bowel gaseous distension but not fluid levels. Ultrasound abdominal scan didn't evidence significant illnesses. Blood tests revealed a little increase of WBC (12,6 x 10³/µL). We decided to submit patient to antibiotic therapy and then to diagnostic deepening with abdominal CT-scan with contrast medium 4 hours after hospitalization. Examination showed an effusion in the

pelvis, invaginated ileum and extension of invaginated bowel, together with several lymph node enlargements in mesentery. According to the CT-scan report we decided to submit the patient to explorative laparotomy. Surgical find is an ileal volvulus that is derotated, with an ileum tract presenting an MD. The aforesaid tract kept an ecchymosis and for this reason we decided to resect the affected bowel area with the MD by removing them "en block. Histological examination shows areas of gastric mucosa with erosive chronic phlogosis. Mucosa presented alterations by angyodisplasia.

Case D (D.A.)

18-years-old female patient. In medical history she didn't refer significant pathologies or ingestion of anticoagulants or antiplatelets.

Because of hematochezia followed by lower gastrointestinal bleeding and medical collapse she had been hospitalized at another medical center. During hospitalization she had been submitted to various instrumental examinations (EGDS, CLS, AngioTC) which hadn't showed the reason of symptomatology and then the number of patient's blood red cells had decreased so much that they had been forced to transfuse her with a blood unit. So transfer to our hospital had been decided, where we can perform capsule endoscopy on the bowel. When she arrived, the patient didn't show any shock signs, without active lower gastrointestinal bleeding. She was submitted to colonoscopy, that showed blood coagula inside the bowel, and once again to abdomen AngioTC

that didn't show active bleeding points. The patient was then submitted to capsule endoscopy of bowel and at the same time we requested for TC99 scintigraphy. The examination with endoscopic capsule didn't reveal bleeding sources and, while we were waiting for scintigraphy, suddenly massive hemorrhage appeared and caused serious decrease of red blood cells, so we had to transfuse 4 blood units and then decided to perform urgent explorative laparotomy on the patient. Operative findings consisted in active bleeding from MD, resected with Stapler. Histological report stated "4,5 cm MD with ectopic gastric mucosa".

CASE E (R.P.)

50-years-old male patient, affected by cirrhosis. No ingestion of anticoagulants or antiplatelets. In medical history previous hospitalizations for lower gastrointestinal bleeding. He was transferred from another medical centre for massive lower gastrointestinal bleeding. After his arrival at our hospital, blood tests was satisfying: Hgb (=9.6g/dl), PTL (=193 x 10^{9} /L) and INR (=1.10). At the previous hospital he had already been submitted to colonoscopy which showed normal colic mucosa and bleeding above the ileocecal valve. AngioTC had also

been performed but it had not revealed the true reason of bleeding. Because of this, we decided to perform arteriography but it resulted negative too as regards of bleeding sources. We decided to perform explorative laparotomy, showing an MD that was resected with Stapler. Histological examination stated "3 cm MD with 2 cm area of ulcerated mucosa located 3 cm away from edge of resection. Enteric epithelium with eroded and ulcerated areas, serous inflammation, edema and venous congestion".

CASE F (R.D.)

18-years-old male patient. In medical history he didn't refer significant pathologies or ingestion of anticoagulants or antiplatelets. He was transferred from another medical centre, where he had already been submitted to gastroscopy and colonoscopy. After his arrival, blood tests were satisfying (Hgb = 8.1g/dl; PTL = $237 \times 10^9/L$ and INR =1.10) and so we first decided to perform angiotac on the patient but it didn't show the source of bleeding. Explorative laparotomy revealed an MD that was resected. Histological examination affirmed "MD with nonspecific chronic phlogosis, enteric mucosa".

CASE G (V.N.)

48-years-old male patient. In his medical history no significant pathologies or ingestion of antiplatelets or anticoagulants. He gets to our emergency room for abdominal pain. He's submitted to abdominal echography that shows an hypoechoic area depending on abscess in the right iliac fossa. WBC have increased (13,480 x $10^{3}/\mu$ L) so we decide for explorative laparotomy because we are suspecting acute appendicitis. Exploration shows an inflammatory mass consisting of appendix, caecum and one ileal tract with MD. Patient is submitted to appendicectomy, resection of MD and drainage of abscess. On seventh day after the intervention the absence of canalization recommends the performance of abdominal TC scan that shows dehiscence of colic stump. For this reason we perform a right hemicolectomy.

Histological examination stated "4x2 cm MD with enteric and focal gastric mucosa. Acute appendix".

Discussion

Comparative anatomy tells us that in other living species, above all in birds, MD is a normal find ⁴. In human species it is conversely considered a congenital anomaly depending on incomplete obliteration of omphalo-mesenteric duct. During embryonic life yolk-sac communicates with developing bowel thanks to omphalo-mesenteric duct itself, and in 5th week of pregnancy the latter starts to close and to detach by fetus. If the tip of the duct doesn't close, an MD is produced ⁴. MD is not the only

anomaly deriving from incomplete obliteration of omphalo-mesenteric duct; in fact enteric fistulas (enteric cysts or fibrotic bands connecting bowel with umbilicus) can also be born ^{5,6}. Though it is considered an embryonic life development anomaly, we can find in medical literature reports talking about familiarity of this disease. In 1955 Michel described four patients belonging to the same family: two sisters of 27 and 29-years old, and two children of the older one of 7 and 2 and half-years-old, everyone submitted to surgical operation for acute abdomen by complicated MD.

In the majority of cases MD is located on the antimesenteric side of ileum and rarely on the mesenteric one ⁸. It is generally situated 45-60 cm from ileocecal valve (6), but it can sometime also be 180 cm far from it ⁴. Yamagouchi in his report on 600 cases asserts that average distance of MD from ileocecal valve changes according to the age: about 35 cm in children under 2 years old, 46 cm between 3 and 6 years old and 67 cm in adults ⁹.

MD can be between 1 and 10 cm long, and its basis is usually 1-4 cm wide. In medical literature we can find reports about MDs whose bases were until 1 meter wide⁵ or of gigantic size^{10,11}.

Blood provision of diverticulum is guaranteed by vessels that derive from superior mesenteric artery, go through ileal wall and run under the serosa, or by vessels running in a little mesentery ¹⁰ that reaches the tip.

DM has histologically the features of a diverticulum having all normal layers of enteric wall, and presents in the order, from internal side to external: mucosa, submucosa, muscular and serous layer ¹⁰. MD is born from yolk-sac cells, that are pluripotent, so we can find inside the diverticulum ectopic tissue ⁶. Symptomatic diverticula in high rate are featured by it. We discover most frequently ectopic gastric tissue, described in percentages from 16 to 32%^{10,12}. Less often, from 3 to 5% of cases, presence of pancreatic tissue is described ^{10,12}. Some authors also tell about the finding of biliaric, colic or endometrial tissue ⁶. Incidence of MD in global population is attested between 0.3% and 3% ^{4,6,12,13}. Rojhan's autoptic study ¹⁰ reports an higher rate, with presence of MD in 6% of cases.

MD is more frequent in males than in females with a ratio of 2-3 to ^{1,4,5,12}, except for Robin's report ¹⁴, that reports the same ratio for both sexes. The most frequent clinical sign of MD is bleeding in children and enteric obstruction in adults. The presence of pancreatic or gastric ectopic tissue is considered as the main reason of the bleeding. In Michel's series ⁷ on 5 patients presenting hemorrhage 4 had ectopic gastric tissue in MD and only one pancreatic tissue. Hemorrhage depends on acid secretion from ectopic gastric mucosa or on alkaline ones from ectopic pancreatic mucosa, that cause inflammation, ulceration and bleeding from near ileal mucosa ³. MD can present as Littrè's hernia through inguinal or crural canal ¹⁵ or originate abdominal neoplasm ^{16,17}. Preoperative diagnosis of symptomatic MD can be very

difficult since clinical features and imaging often don't allow to distinguish into different diseases. Besides, particularly large bleeding or acute abdomen don't allow to follow the regular preoperative diagnostic process in emergency area.

If bleeding is present, colonoscopy can only say us if in terminal tract of ileum is there some blood, but it can't get to the area in which diverticulum is located.

Furthermore capsule endoscopy or double-balloon enteroscopy can allow us to see the bleeding cause even though medical experience in this branch is still restricted. TC-99 m scintigraphy is useful in patient with bleeding to point out ectopic gastric mucosa in MD.

Sensibility of this procedure is 60%¹. Finally, with recent introduction of computed 64 slice CT-scan, with minimal thickness of 0.5 mm and 0.4 seconds as rotation time, we can acquire high resolution images to find sources of bleeding in gastro-enteric tract with high sensibility and specificity². Unfortunately, to discover an active bleeding of gastrointestinal tract with this procedure it is necessary that there is an hemorrhage with a blood flow of 0.4 mL/min while examination is performed 4.

DMs are clinically distinguishes into symptomatic and asymptomatic ones. We qualify as symptomatic diverticula complicated by a pathology requiring to perform surgical operation, and asymptomatic the ones identified during a surgical intervention executed for another disease and that are not the main reason for the surgical pathology (incidentaloma).

Opinions about the possible removal of incidentalomas are different and often dissimilar.

A casuistry recently published about 1476 patients hospitalized from 1950 to 2002 in Mayo Clinic ¹² showed that average age (±DS) of the ones presenting symptomatic MD is 31±23.6 years old (median 27 years). Negative prognostic factors for complication of MD include age lower than 50, male sex, MD longer than 2 centimeters, and presence of ectopic tissue. Another fact deriving from Park's report is the percentage of postoperative complications, that is 13% in symptomatic patients and 20% in asymptomatic ones (1 decease). In all cases postoperative complications, death included, weren't attributed to surgical intervention.

A report on 58 patients with symptomatic MD (average age 23 years, range <1-82 years) and 87 with asymptomatic one (average age 37 years, range 4-86) pointed out a 7% percentage of long-term complications (e.g. adherence syndrome) in symptomatic patients (1 decease) and only a 2% rate in asymptomatic ones ¹⁹. Though the author thinks that there's no increase of risk to develop DM complications associated to patient's age rise, and that the risk to develop complications owing to symptomatic DM is the 6,4% for both sexes, he always recommends the removal of incidentalomas exactly to avoid the appearance of complications ¹⁴.

According to Thirunavukarasu ¹³, that examined 163 cases of MD, diverticulum is an anomaly risking to devel-

op a cancer. He reports an average yearly incidence of neoplasia originated by the diverticulum equal to 1,44% on ten millions of inhabitants, with an increase five times more in last 5 decades, considering a period going from 1973 to 2006. Besides, the risk to develop cancer on MD, than in every other ileal site, increases according to age and reaches its higher peak when the patient is about 70 more. Moreover, since tumor develops, the most frequent histological type results to be the carcinoid, with a percentage equal to 77% of all cancers originated from MD. Therefore he always advises the removal of incidentalomas.

In Zani's retrospective report ¹⁷ 244 patients affected by MD were evaluated. He reports a death rate in patients with asymptomatic MD equal to 0,01%. The number of surgical interventions for diverticular complications significantly decreases after pediatric age and the frequency of complications for removal of asymptomatic diverticula is clearly higher than in patients whose incidentalomas aren't removed. Since in lifetime of patients with not removed asymptomatic MDs there isn't a significant rate of complications, resection of them isn't recommended. In fact he thinks that at least 758 patients would need the resection of the incidentaloma to prevent a decease caused by complications associated with MD itself

In 2006 some authors ¹⁴, basing on literary dates, identified the ones seeming the main risk factors for onset of postoperative complications ¹⁶. Then they purposed a riskscore considering several factors: male sex, age lower than 45, diverticulum ± 2 cm long, and presence of fibrous band. The risk is calculated assigning score to every factor in relation to their presence or absence, until a maximum of 10 as total score. It is so calculated: male sex=3, female one=1; age <45=2, >45=1; length of diverticulum higher than 2 centimeters=2, lower=1; presence of fibrous band=3, absence=0. If total score of this calculation is \geq 6 removal of MD is always recommended.

In medical literature there's no uniformity of opinions neither about the kind of surgical technique to use to remove the MD. Considering that most of symptomatology depends on the presence of ectopic tissue inside the diverticulum, and that the scope of surgical intervention is to remove not only the diverticulum, but always the total ectopic tissue, some author thinks that diverticulectomy with stapler is not enough.

In Varcoe's retrospective study ²⁰ 77 patients having MD submitted to surgical intervention from 1993 to 2003 were evaluated. Morphologic features of diverticulum were considered according to height-to-diameter ratio (HDR). MDs with HDR \geq 2 were defined "long", and the ones having HDR lower than 2 "short". The morphology was then correlated with the presence of heterotypic gastric mucosa. In 25 cases on 77 (17 symptomatic and 8 asymptomatic - 5 with HDR higher than 2) MD contained ectopic gastric tissue. The 5 cases with HDR higher than 2 showed ectopic gastric tissue on the

tip or inside the diverticulum without involving the base. On the remaining 20 patients with HDR lower than 2, 12 (60%) showed ectopic gastric mucosa inside the base of diverticulum. According to these results, Varcoe does not recommend simple diverticulectomy in presence of a short diverticulum. This conduct is accordant with medical literary reports because almost all authors advice wedge-resection with anastomosis of residual enteric stumps to be sure of removing all ectopic tissue ^{4,6,12,14}. Diverticulectomy is also recommended when the diverticulum is located inside an hernia, before the reconstruction of the parietal defect ¹⁷. Finally, though everyone consider it the best technique, intracorporeal laparoscopic removal could sometimes be not enough to perform a correct diverticulectomy. In fact, in case of laparoscopic removal of large MD with stapler, since direct manual palpation of lesion isn't possible, we risk to leave in the site ectopic tissue with possible negative consequences through the time. To avoid this, assisted laparoscopic resection is considered more safe because it allows to extract the bowel and to touch the diverticulum with a wider abscission of tissue ^{6,21}.

Conclusions

Medical literature does not establish a uniform behavior of conduct and the management of this patients often depends on experience of surgical team that manage this patients. In our experience we never speculated that acute symptomatology depended on complicated MD and diagnosis was always done during laparotomy. In fact, neither colonoscopy and TC scan were able to reveal the diverticulum. Colonoscopy can give us an indirect indication of possible bleeding sources from ileum when it excludes any hemorrhagic colic sources. Our experience is neither significant about relation between ectopic tissue inside the diverticulum and active bleeding: we can only report that histological examination showed in a case ectopic pancreatic tissue, in two cases ectopic gastric tissue areas and in the last one normal enteric tissue on the four patients submitted to surgical intervention for hemorrhage.

As regards the possible removal of diverticula accidentally found during laparotomy performed for another pathology, we think that removal is always the correct choice, so that future complications such as neoplasm can be avoided. MD simple resection by Stapler at the base of diverticulum is the correct choice.

Riassunto

La diagnosi di patologia associata DM complicato può essere molto difficile in quanto le caratteristiche cliniche e l'imaging spesso non sono dirimenti tra differenti condizioni morbose. Sono state rivalutate le cartelle dei pazienti dimessi con diagnosi di patologia acuta associata a diverticolo di Meckel complicato dal 1 Gennaio 2011 al 30 Novembre 2012.

Sono stati identificati sette casi consecutivi: cinque maschi (71,4%) e due femmine (28,6%). Il range di età va da 13 a 50 aa con una media di 28 aa. Quattro sono stati sottoposti ad intervento in regime di urgenza vera per emorragia digestiva (57%) e due per occlusione intestinale (29%). Uno per sospetta appendicite (14%).

In tutti i casi il diverticolo di Meckel resecato è stato inviato all'esame istologico. Due campioni presentavano linea epiteliale normale di tipo enterico. Quattro presentavano aree di mucosa ectopica all' interno del diverticolo : tre casi di aree focali di mucosa gastrica e uno di mucosa pancreatica ectopica. L' ultimo caso presentava si linea epiteliale normale ma essa era ulcerata e con aree di emorragia.

Nella nostra esperienza in nessun caso è stata ipotizzato pre operatoriamente che il quadro acuto dipendesse da un DM complicato e la diagnosi è sempre stata effettuata in corso di intervento chirurgico. Noi riteniamo che l'asportazione del DM sia sempre l'opzione di scelta, con lo scopo di evitare complicanze future anche di tipo neoplastico e la resezione semplice con stapler alla base del diverticolo è l'opzione chirurgica di scelta.

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