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World Journal of Gastrointestinal Surgery (*World J Gastrointest Surg*, *WJGS*, online ISSN 1948-9366, DOI: 10.4240) is a peer-reviewed open access academic journal that aims to guide clinical practice and improve diagnostic and therapeutic skills of clinicians.

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MINIREVIEWS

Role of stenting in the palliation of gastroesophageal junction cancer: A brief review

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Abstract

Gastroesophageal junction cancer has an increasing incidence in western countries. It is inoperable when first manifested in more than 50% of cases. So, palliation is the only therapeutic option for the advanced disease to relieve dysphagia and its consequences in weakened patients with an estimated mean survival under 6 mo. This article has tried to identify trends focusing on current information about the best palliative treatment, with an emphasis on the role of stenting. Self-expanding stent placement, either metal or plastic, is the main management option. However, this anatomical location creates some particular problems for stent safety and effectiveness which may be overcome by properly designed novel stents. The stents ensure a good quality of life and must be preferred over other alternative methods of loco-regional modalities, *i.e.*, external radiation, laser thermal or photodynamic therapy. Although stent placement is generally a simple, safe and effective method, there are sometimes complications, increasing the morbidity and mortality rate. Bypass operative procedures have now been abandoned as a first choice. The stomach instead of the colon must be used for a bypass operation when it is needed. Chemotherapy, despite the toxicity, and intraluminal radiation (brachytherapy) have a well-defined role.

Key words: Gastroesophageal junction cancer; Palliative therapy; Stent placement; Dysphagia relief; Esophageal carcinoma; Gastric carcinoma

Core tip: The topic is interesting and this manuscript contains the most recent data briefly highlighting it. More than half of the patients with gastroesophageal junction cancer present with inoperable disease at the time of diagnosis so they need palliative treatment to relieve dysphagia and its consequences. Stent placement ensures good quality of life during the short survival time but it has some additional specific problems in this particular location.

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INTRODUCTION

Cancer of the gastroesophageal junction has had an increasing incidence over the past thirty years in western countries and is now the eighth most common malignancy. However, the exact incidence cannot be precisely assessed because it can be allocated as either gastric or esophageal cancer. This confusion is not of particular importance since in both cases the management is the same. Its prognosis is not good, with a 5 year survival less than 20% and even less in younger patients under 35

years old[1,2].

More than half of these patients present with inoperable disease at the time of diagnosis. Hence, they need palliative treatment in order to alleviate dysphagia and its further consequences for the life expectancy, given that the average survival time does not exceed 6 mo[1-4]. Although the kind of palliation must be individualized, generally stent placement is the first choice. There are also other alternatives, such as bypass operation, external radiation, intraluminal radiation (brachytherapy), chemotherapy, laser ablation (thermal Nd: YAG or photodynamic), dilatations, chemical substance injection, mainly ethanol, as well as nutritional support *via* a nasoenteric feeding tube or percutaneous endoscopic gastrostomy[1,4].

STENT PLACEMENT

Endoscopic self-expanding metal stent (SEMS) placement provides a rapid relief of dysphagia and it is currently the most frequently chosen method[3,5,6]. However, according to the relevant researchers, the recurrence of dysphagia varies between 22%-50%. This recurrence may occur due to various reasons, such as tissue growth in the stent space, stent migration or its obstruction by bolus. The incidence of cancerous tissue overgrowth *via* the stent is 26%-36%, while the non-cancerous granulomatous tissue overgrowth is 20%. The former has been estimated to occur after 18 wk and the latter after 22 wk following the stent placement. This tissue overgrowth can be managed either by thermal ablation or the recently introduced self-expanding plastic stent (SEPS) placement, with the advantage of lower cost[3].

The placement of SEMS has also been proposed for relieving dysphagia due to local recurrence after esophagogastrectomy, in which the mean survival time has been limited to 4-6 mo or even less[7].

The application of SEMSs does not require dilatation of the stricture before placement; they are also flexible and ensure a diameter of patent lumen from 16 to 24 mm.

Dysphagia is staged in 5 grades: (0) Ability to swallow normal diet; (1) Ability to swallow part of solid diet; (2) Ability to swallow part of semi-solid diet; (3) Ability to swallow only liquids; and (4) Complete obstruction. The indication for stent placement includes grade 3 and 4 or the presence of tracheobronchial fistula irrespective of the grade[5].

In a recent meta-analysis on published reports including 1027 patients in 16 randomized controlled trials, it was concluded that endoscopic placement of self-expanding stents is the most widely used method for the management of dysphagia in comparison to other alternative methods of loco-regional modalities, *i.e.*, radiation, laser thermal or photodynamic therapy. Despite its high cost, it is a simple and effective method (with minimal invasiveness and discomfort) to ameliorate dysphagia in the vast majority of patients with a mean survival of no more than six months. In addition, its superiority is mainly associated with the fact that, unlike what is commonly observed in alternative methods, there is no need for re-interventions. However, in patients with one year survival, a loco-regional palliation seems better despite the need for further re-intervention. Furthermore, their higher life expectancy is possibly associated with the application of loco-regional treatment[4]. According to the aforementioned meta-analysis, the choice of conventional self-expanding stents *vs* modern anti-reflux stents has been found to be equally effective in relieving reflux since there was no difference between them. There are minimal differences among the various types of stents with regards to the outcome[4].

COMPLICATIONS OF STENTS

Significant differences have been noted between endoscopic and radiographic stent placements regarding the short-term complications. They encompass hemorrhage, pneumonia, exhaustion, heart abnormalities, perforation and sepsis. The morbidity and mortality rate exceeds 45% and 9%, respectively[6].

Due to the high frequency of the manifestation of long-term complications, *i.e.*, stent migration, hemorrhage and protrusion of gastroesophageal junction mucosa, opposition to the use of metal stents has been recently expressed and the individualized design of nitinol stents in special anatomical conditions has been proposed[8]. The placement of SEMSs for palliative management of obstruction due to gastroesophageal junction cancer has often been related to stent migration, as well as with symptoms of gastroesophageal reflux. The stent placement in this particular location has some additional specific problems compared to proximal esophageal cancer and therefore it implies less palliation and a higher rate of complications. Stent migration is more frequent due to the fact that the distal end of the stent protruding freely into the stomach fundus cannot be fixed to the wall. Hemorrhage is even more frequent in such cases. Firstly, the distal end of the stent may corrode the posterior wall of the stomach resulting in ulceration and subsequent bleeding. Secondly, the stent *via* the gastroesophageal junction cannot remain straight due to the angle of his, resulting in high pressure and ulceration with subsequent bleeding. The stent angulations are mainly responsible for the lack of significant improvement in the swallow quality. Moreover, gastroesophageal reflux is particularly common. Novel stents with specific design have certain advantages in overcoming those difficulties of the gastroesophageal junction.

TYPES OF STENTS

Thus, such stents have been designed, including the anti-reflux Z mechanism[9]. There are different types available: (1) Z-stent with the Korean modification (Choo stent) composed of nitinol or the European version of stainless steel, both covered by polyethylene; and (2) Flamingo Wallstent (available only in Europe) composed of cobalt mixture covered by polyurethane specifically designed for gastroesophageal junction with diameter of either 30

mm (distal 20 mm) or 24 mm (distal 16 mm). Some other stent types include Ultraflex composed of a plexus of nitinol wire covered by polyurethane with a diameter of 28 mm (distal 23 mm) or 23 mm (distal 18 mm), Wallstent Ⅱ composed of cobalt mixture covered by silicone with a diameter of 28 mm in both ends (20 mm in the middle) and the latest nitinol stents double layer composition (inner of polyurethane and outer of uncovered nitinol wire preventing stent migration). Reflux is prevented by a glove of polyurethane extended into the stomach[9] or a membrane into the lower end functioning as a valve[10]. Additionally, there are available novel SEPSs (Polyflex) consisting of polyester covered by silicone which have been used with satisfactory results[11].

OTHER MODALITIES

The combination of stricture dilatation by balloon, chemotherapy and/or radiation, with additional metal stent placement has been proposed in some cases[12].

Radiation and intraluminal high dose brachytherapy is indicated in advanced cancer, offering palliation by relieving dysphagia and improving the quality of life[13-17]. Despite the toxicity and its complications, chemotherapy has a place in such condemned patients[18-20].

The cooperation of various medical specialities is mandatory since the use of all available current diagnostic and therapeutic tools improves the outcome[21,22]. Nowadays, in western countries, adenocarcinoma represents two thirds of esophageal cancer. The current modalities of palliative treatment are equally effective in both adenocarcinoma and squamous cell carcinoma, contributing to a satisfactory quality of life[23].

BYPASS OPERATION

With the introduction of modern, safe and effective non-operative alternative interventional methods for the management of dysphagia, the palliative bypass operation has been applied even less, mainly due to its high morbidity and mortality rates. On the other hand, the average survival time of these patients is limited to about 6 mo. The stomach has been preferred to be used for a bypass procedure and when the stomach cannot be used, we use the colon, which historically was first used[24-26]. The gastric conduit was introduced first by Kirschner in 1920 and so the operation was established using his name. However, it should be mentioned that this operation is now considered an obsolete planned procedure although it remains a reasonable choice in cases when unexpected findings appear that exclude any radical operation during the operative exploration[25].

CONCLUSION

In conclusion, palliative treatment of inoperable gastroesophageal junction cancer aims at managing dysphagia. It can now mainly be achieved by interventional stent placement, ensuring good quality of life during the short survival time. The bypass operation must be avoided as a first choice in these severely affected patients. The gastric conduit is preferred instead of colon interposition.

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CASE REPORT

Giant mucinous cystic adenoma with pancreatic atrophy mimicking dorsal agenesis of the pancreas

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Abstract

Mucinous cystic adenoma (MCA) of the pancreas is a rare benign cystic tumor with ovarian-like stroma and lack of communication with the pancreatic ductal system. The ovarian tissue is incorporated from the left gonad within the dorsal pancreas during embryogenesis. Consequently, congenital dorsal agenesis of the pancreas (DAP) cannot be associated with MCA. We report the case of a giant MCA associated with atrophy of the dorsal pancreas mimicking complete DAP. Pancreato-magnetic resonance imaging failed to identify the dorsal pancreas but the absence of diabetes mellitus and compression of the splenic vein with major tributaries rectified the diagnosis of secondary atrophy of the distal pancreas. Unusual proximal location of the cyst in the pancreas may have induced chronic obstruction of both the dorsal pancreatic duct and the splenic vein, with secondary atrophy of the distal pancreas.

Key words: Dorsal agenesis; Pancreas; Cystic tumor; Diabetes; Surgery

Core tip: Mucinous cystic adenoma (MCA) of the pancreas is a benign tumor with ovarian-like tissue located in the body or the tail of the pancreas. We report the first case of atrophy of the distal pancreas secondary to compression by a giant MCA. We raise the question of underlying dorsal agenesis of the pancreas (DAP) but as ovarian-like tissue of MCA comes from the close migration of the left gonad and the dorsal pancreas during embryogenesis MCA can not be associated with true DAP. Finally, the absence of diabetes mellitus, and thrombosis of the splenic vein confirmed the secondary atrophy caused by a mechanism of compression.

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INTRODUCTION

Dorsal agenesis of the pancreas (DAP) is a rare disease that is frequently asymptomatic except when associated with polysplenia syndrome. Etiology remains unclear, but dysgenesis of the dorsal bud during embryogenesis seems to be the most plausible explanation. Confounding diagnoses include pseudo-agenesis of the dorsal pancreas following acute pancreatitis or compression by a tumor[1,2]. In such cases, the mechanism involves pancreatic duct obstruction with atrophy of pancreatic acini replaced by

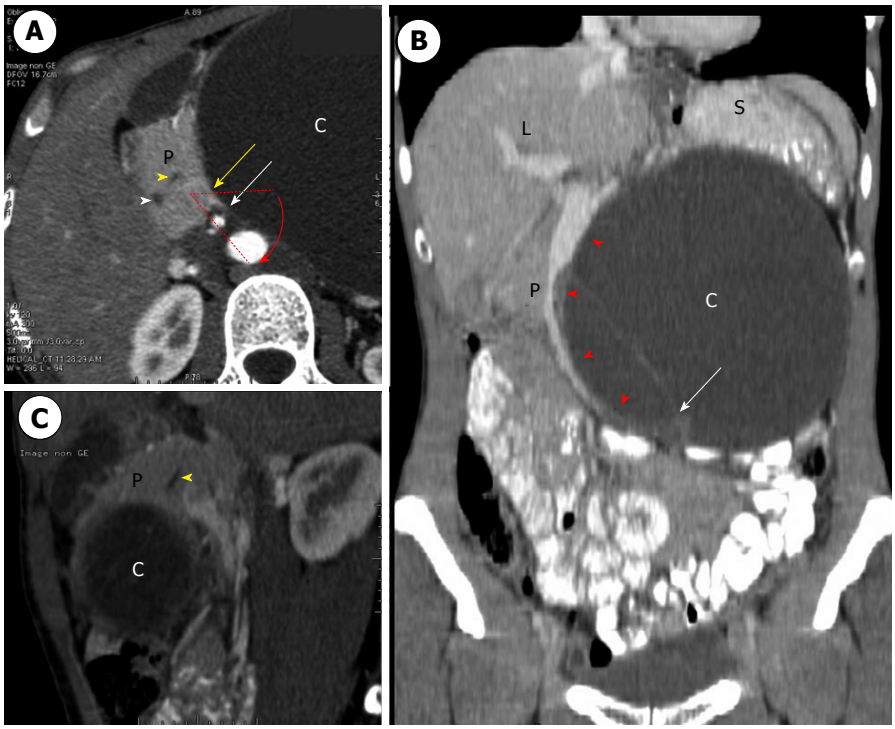

Figure 1 Preoperative contrast-enhanced computed tomography showing a huge cyst with septa developed close to the head of the pancreas, exophytic development to the left and downward, and rotation of the mesenteric axis. A: Axial view showing the head of the P with the intrapancreatic main bile duct (white arrowhead) and ventral pancreatic duct (yellow arrowhead). There is rotation of the mesenteric axis, as shown by the oblique plane made by the superior mesenteric artery (white arrow) and the superior mesenteric vein (yellow arrow); B: Coronal view showing thin septa within the macrocyst (white arrow) and deviation without thrombosis of the mesenterico-portal axis (red arrowhead); C: Sagittal view showing anterior development of the cyst, close to the head of the pancreas (P) and the ventral pancreatic duct (yellow arrowhead). L: Liver; S: Spleen; P: Pancreas; C: Cyst.

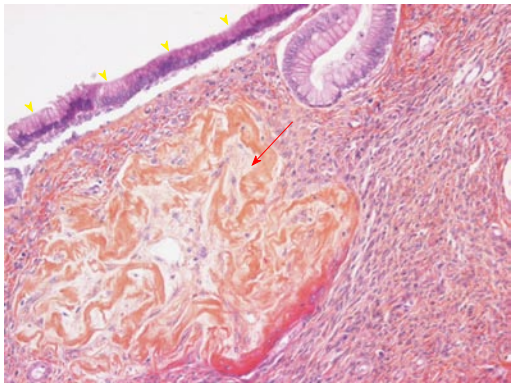

Figure 2 Histologic examination (× 20) showing the cyst lined by tall columnar epithelial cell (yellow arrowheads) with underlying ovarian-type stroma composed of densely packed spindle cells (red arrow).

fat. However, endocrine cells generally still persist and prevent the occurrence of diabetes mellitus. Benign cystic or non-cystic tumors cannot usually induce pancreatic atrophy since invasive contingents are missing. We herein present the first documented case of a giant mucinous cystic adenoma (MCA) of the pancreas responsible for secondary atrophy of the dorsal pancreas and mimicking a complete DAP.

CASE REPORT

A 36-year-old female was referred to a first institution for exploration of an asymptomatic abdominal mass. She had no previous medical or surgical history. Physical examination showed a large painless epigastric mass. Ultrasound (US) showed a well-limited cyst in the epigastric area 15 cm × 10 cm in size with a thick wall, heterogeneous content and peripheral calcifications. Laboratory test results including amylase, lipase and serum glucose levels were within the normal range. The serum tumor markers carcinoembryonic antigen (CEA) and carbohydrate antigen 19-9 (CA19-9) were normal. Contrast-enhanced computed tomography (CT) scan confirmed a well-defined, low-density, 17 cm × 11 cm, unilocular cystic tumor (Figure 1). It seemed to originate from the proximal part of the distal pancreas but the rotation to the left of both the head of the pancreas and the superior mesenteric vessels rendered the exact location of the cyst inconclusive (Figure 1). Thin septa, contrast enhancement and calcifications were also observed. Magnetic resonance imaging (MRI) and endoscopic US-guided fine needle aspiration (EUS-FNA) of the cyst were not performed preoperatively as giant benign MCA was suspected. The patient underwent surgical enucleation of an exophytic 14 cm × 10 cm cystic tumor of the pancreas. In his operative report, the surgeon noted a difficult procedure with accidental intraoperative rupture of the cyst. The postoperative period was uneventful and the patient was discharged on postoperative day 7. The pathological report confirmed a multilocular, thick-walled, 14 cm × 10 cm cyst with intracystic hemorrhage and disruption. Microscopically, the cyst was lined by tall columnar, mucin-containing epithelial cells, surrounded by an ovarian-like stroma (Figure 2). The epithelium was benign and positive for cytokeratins 7 and 19, which is consistent with the diagnosis of pancreatic MCA. Enucleation was complete and conservative as pancreatic parenchyma was absent on the specimen.

An abdominal CT-scan was performed 6 mo after surgery for exploration of abdominal tenderness and showed four low-density homogenous cystic lesions with contrast-enhanced wall in the previous pancreatic

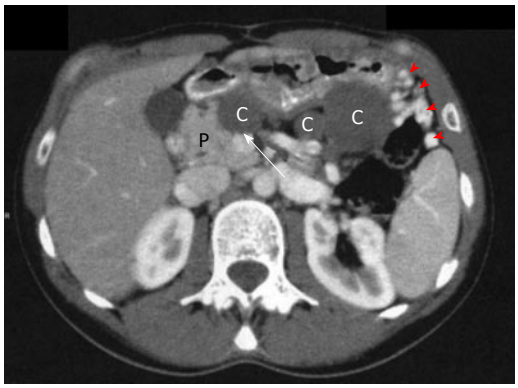

Figure 3 Post-operative contrast-enhanced computed tomography showing three low-density homogenous cystic lesions suggesting secondary dissemination of the resected cyst. Major tributaries were also present around the stomach (red arrowheads). The P has a total distal atrophy as shown by lack of pancreatic tissue behind the mesenterico-portal axis (white arrow). P: Pancreas; C: Cyst.

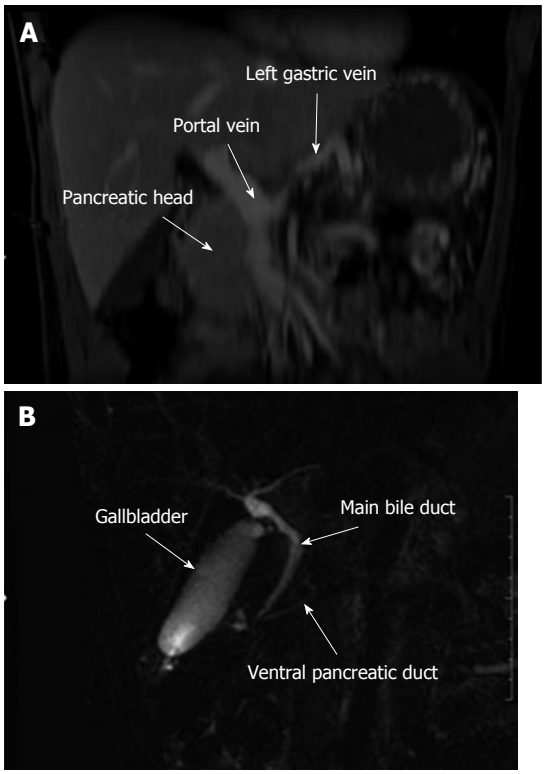

Figure 4 Coronal magnetic resonance imagery showing lack of the body and tail of the pancreas and of the splenic vein (A), magnetic resonance cholangiopancreatography showing the common bile duct joining the ventral pancreatic duct at the posterior part of the head of the pancreas (B). The dorsal pancreatic duct is not visible.

enucleation area (Figure 3). The body and tail of the pancreas were not visible on CT nor on the upper part of the head of the pancreas, suggesting complete DAP. Magnetic resonance cholangiopancreatography (MRCP) showed absence of the body and tail of the pancreas with no accessory pancreatic duct that would confirm the diagnosis of DAP (Figure 4). There was no other pancreatic anomaly and no polysplenia. The patient was then referred to our institution. A second interpretation of the scan pictures showed splenic vein obstruction with major tributaries around the stomach, suggesting segmental portal hypertension (Figure 3). These features were also present on the initial CT. The serum levels of glucose, amylase and lipase were still normal. The suspected diagnoses were either recurrence of the MCA following difficult and incomplete primary resection, as suggested by intraoperative rupture, or multiple pseudocysts due to a latent post-operative pancreatic leak. A second-look laparoscopy was advocated because of abdominal tenderness, risk of recurrence of the MCA and because EUS-FNA failed to distinguish MCA from pancreatic pseudocysts. Laparoscopic exploration showed extra-pancreatic multiple cysts close to the first duodenum at the anterior part of the head of the pancreas, without pancreatic leak. The body and tail of the pancreas were also absent. Intraoperative pathology examination of the cysts confirmed pseudocysts with fat necrosis. Postoperative course was uneventful and the patient was discharged on postoperative day 3. One year after initial resection the patient had no diabetes mellitus, and routine blood parameters, in particular serum glucose level, were normal.

DISCUSSION

To the best of our knowledge, there are no documented reports of pancreatic cyst-including benign MCA-associated with congenital or secondary atrophy of the distal pancreas. MCA is a rare benign cystic tumor characterized by an ovarian stroma underlying the epithelium of the cyst. Differential diagnosis includes other benign cystic lesions such as serous cystic neoplasm, intraductal papillary mucinous neoplasm and post-pancreatitis pseudocysts[3]. Clinical presentation (female sex, location in the distal pancreas and no history of pancreatitis) and paraclinical investigations (MRI and EUS-FNA showing no pancreatic duct communication, wall calcifications and high level of intra-cystic CA19-9 and CEA) are suggestive of MCA[4]. Prophylactic resection is warranted as malignant transformation can occur in 6%-27% of cases[5,6]. The origin of ovarian stroma remains unclear. It has been suggested to derive from ectopic tissue within the pancreas incorporated throughout close migration of the left primordial gonad and dorsal pancreatic bud during embryogenesis[7-9], which would explain the predilection of MCA for the body-tail region of the pancreas. Consequently, the association of complete DAP with MCA is theoretically not possible.

DAP is a congenital agenesis of the pancreas that can be partial or complete. It is a rare event since only 54 cases have been reported in the literature[10]. The pancreas develops from ventral and dorsal endodermal buds during embryogenesis. The ventral bud gives rise to the major part of the head and uncinate process, which drains through the duct of Wirsung (*i.e.*, the main pancreatic duct). The dorsal bud forms the upper part of the head, body and tail of the pancreas and drains through the duct of Santorini (*i.e.*, the accessory pancreatic duct). Each

bud develops a tree-like ductal system and, during growth and rotation of the gut in the seventh week of gestation, the two buds fuse and form the main pancreatic gland. Exocrine secretion is consistent in both dorsal and ventral pancreas, whereas insulin-secreting cells of the islets of Langerhans are located predominantly in the dorsal pancreas[11]. Rarely, DAP is complete with lack of structures originating from the dorsal pancreas - such as minor papilla, accessory pancreatic duct, body and tail[12]. When DAP is partial, which is most frequently the case, the minor papilla with a remnant accessory pancreatic duct and the body of the pancreas usually persist[13]. Confounding diagnosis is secondary atrophy of the distal pancreas due to chronic obstruction of the pancreatic duct. In this case, atrophy involves predominantly the exocrine tissue while endocrine cells are still present and prevent the occurrence of diabetes mellitus[1,2].

In our patient, despite arguments for congenital DAP, compression of the main pancreatic duct by the giant cyst with secondary atrophy of the distal pancreas was the most probable hypothesis. As discussed above, the association of complete DAP with MCA is not possible. Although there was no accessory pancreatic duct on MR-CP, the unusual proximal location within the dorsal pancreas of the MCA could have induced atrophy of the distal pancreas with no or undetectable remnant accessory pancreatic duct. This is consistent with atrophy of the splenic vein and collateral vascularization developed from the gastric veins, usually absent in congenital DAP[10,13]. Atrophy may have been worsened by intraoperative injury in what was described as a difficult procedure, as shown by the presence of a postoperative pseudocyst close to the head of the pancreas (Figure 3). Another argument for secondary atrophy is the absence of diabetes mellitus, since congenital DAP involves both endocrine and exocrine secretions with diabetes mellitus in around 40% of cases[2].

To the best of our knowledge, there is no published report of the effects of MCA on the distal pancreas. There are at least two reasons for this[14,15]. First, observations usually focus on the size, symptoms and management of the MCA, with little or no information about the distal pancreas. Second, MCAs are located in the distal position and usually spare the proximal pancreatic parenchyma. The pathological report usually insists on the features of the MCA, but not in the distal pancreas, and whether it is atrophied or absent. There have been reports of DAP associated with non-invasive tumors[16,17]. In these cases, the diagnosis of DAP was based on atrophy of the distal pancreas but in no instance it was possible to differentiate congenital agenesis from secondary atrophy. The unusual occurrence of congenital DAP makes the association with tumor very unlikely and we suggest that, as in our case, most DAPs associated with huge tumors are the result of secondary atrophy. Furthermore, tumors located within the dorsal pancreas cannot be associated with DAP given that complete agenesis of an organ cannot lead to the development of a tumor because neoplastic transformation can not occur from cells that do not exist.

Our observation is a reminder that the management of huge benign tumors is problematic. Preoperative imaging must be rigorous to detect congenital or acquired anomalies of the pancreas, and to describe pancreatic ductal anatomy. Resection must be conservative as often as possible, to avoid injury of the ductal system and secondary occurrence of pancreatic fistula or pseudocysts. Non-visualization of distal pancreas can be the consequence of long term compression of the main pancreatic duct. However, islets cells may still be present and accidental resection of the atrophic pancreas can lead to secondary diabetes mellitus. Thus, we recommend addressing these patients to tertiary centers for adequate preoperative evaluation and surgical management.

COMMENTS

Case characteristics

A 36-year-old female presented with asymptomatic abdominal mass.

Clinical diagnosis

Painless huge epigastric mass with no digestive repercussion.

Differential diagnosis

Gastric tumor, liver tumor, liver cyst, pancreatic tumor.

Laboratory diagnosis

WBC 9.80 k/μL, HGB 14.0 mg/dL, glucose 7 mmol/L, lipase 80 U/L, CRP 2.9 mg/L. Carbohydrate antigen 19-9 and carcinoembryonic antigen were within normal limits.

Imaging diagnosis

Computed tomography showed a huge mucinous cystic adenoma (MCA) that originated from the head or the body of the pancreas, with thrombosis of the splenic vein and complete atrophy of the pancreas distal to the cyst mimicking dorsal agenesis of the pancreas (DAP).

Pathological diagnosis

Specimen showed a cyst lined by tall columnar epithelial cells surrounded by an ovarian-like stroma, positive for cytokeratins 7 and 19, consistent with the diagnosis of MCA.

Treatment

The patient was treated by enucleation of the cyst, but recurrence of multiple cysts six months later led to second-look laparoscopy, which showed pseudocysts resulting from the initial surgery.

Related reports

Atrophy of the dorsal pancreas is usually observed in invasive tumors or in chronic pancreatitis but not in non-invasive benign tumors.

Term explanation

DAP is defined as embryological agency of the dorsal pancreatic bud resulting in lack of development of the superior part of the head, body and tail of the pancreas.

Experiences and lessons

Right-sided pancreatic MCA can lead to atrophy of the distal pancreas but can not be associated with DAP as MCAs usually originate from the dorsal pancreas.

Peer review

Splenic vein thrombosis and absence of diabetes mellitus are good markers of secondary atrophy of the pancreas.

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CASE REPORT

Giant Meckel's diverticulum: An exceptional cause of intestinal obstruction

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Abstract

Meckel's diverticulum (MD) results from incomplete involution of the proximal portion of the vitelline (also known as the omphalomesenteric) duct during weeks 5-7 of foetal development. Although MD is the most commonly diagnosed congenital gastrointestinal anomaly, it is estimated to affect only 2% of the population worldwide. Most cases are asymptomatic, and diagnosis is often made following investigation of unexplained gastrointestinal bleeding, perforation, inflammation or obstruction that prompt clinic presentation. While MD range in size from 1-10 cm, cases of giant MD (≥ 5 cm) are relatively rare and associated with more severe forms of the complications, especially for obstruction. Herein, we report a case of giant MD with secondary small bowel obstruction in an adult male that was successfully managed by surgical resection and anastomosis created with endoscopic stapler device (80 mm, endo-GIA stapler). Patient was discharged on postoperative day 6 without any complications. Histopathologic examination indicated Meckel's diverticulitis without gastric or pancreatic metaplasia.

Key words: Meckel's diverticulum; Giant Meckel's diverticulum; Intestinal obstruction; Small bowel

Core tip: The most commonly diagnosed congenital anomaly of the gastrointestinal tract is Meckel's diverticulum (MD), which occurs upon failure of the omphalomesenteric duct to regress and involute. MD can remain asymptomatic, and cases are generally diagnosed incidentally or upon investigation of unexplained gastrointestinal bleeding, perforation, inflammation, or obstruction for both paediatric and adult cases. It is estimated that as little as 4% of cases manifest complications, and obstruction is the most common presenting symptom in adults. In this case study, we report a case of giant MD with secondary small bowel obstruction in an adult male that was successfully managed by surgical resection and anastomosis created with endoscopic stapler.

Akbulut S, Yagmur Y. Giant Meckel's diverticulum: An exceptional cause of intestinal obstruction. *World J Gastrointest Surg* 2014; 6(3): 47-50 Available from: URL: http://www.wjgnet.com/1948-9366/full/v6/i3/47.htm DOI: http://dx.doi.org/10.4240/wjgs.v6.i3.47

INTRODUCTION

The most commonly diagnosed congenital anomaly of the gastrointestinal tract is Meckel's diverticulum (MD), which occurs upon failure of the vitelline (also known as the omphalomesenteric) duct to regress and involute[1-3]. Accumulated experience with surgical treatment of MD (using both open and laparoscopic procedures) has led to the clinical "rule of 2" for symptomatic cases, whereby the anatomical deformity (with estimated prevalence in 2% of the population) is most frequently located 2 feet from the ileocaecal junction and is 2 inches long[2]. MD can remain asymptomatic, and cases are generally diag-

nosed incidentally or upon investigation of unexplained gastrointestinal bleeding, perforation, inflammation, or obstruction for both paediatric and adult cases[1].

It is estimated that as little as 4% of cases manifest complications, and obstruction is the most common presenting symptom in adults[1]. There is evidence that severity of symptoms correlates with MD size. Ninety percent of the reported MDs are between 1 and 10 cm, the average size being 3 cm. MDs ⩾ 5 cm are classified as giant MD, are relatively rare, and may be more prone to complications[1]. Here, we report a case of giant MD which was diagnosed in an adult male with small bowel obstruction and successfully managed by resection.

CASE REPORT

A 23-year-old male patient presented at the Emergency Department with a complaint of abdominal pain, nausea, and vomiting that had persisted for 5 d and increased in severity over the last 24 h. The patient reported no faecal or gas discharge during the previous 48 h. History taking upon admission revealed that the patient had visited hospitals frequently for many years with similar gastrointestinal complaints as well as bloating. The patient's abdomen was remarkably distended and initial clinical assessment indicated hypovolemia. Physical examination revealed significant bowel sounds and substantial abdominal rebound pain, both more robust in the periumbilical area. Laboratory testing showed increased white blood cell count ($11.8 \times 10^3/\mu L$; normal range: 4.1×10^3-11.2×10^3), haemoglobin (17.0 g/dL; 12.5-16.0), haematocrit (49.6%; 37.0-47.0) and creatinine (1.4 mg/dL; 0.4-1.2), but normal blood urea nitrogen (27 mg/dL; 10-50). Abdominal X-ray indicated remarkably high air-fluid levels (Figure 1).

An emergency laparotomy was performed and revealed oedema throughout the entire small bowel, dilation of small bowel segments, and a giant MD (27 cm long and 6 cm wide) on the antimesenteric border of the small bowel at 80 cm proximal to the ileocaecal valve (Figure 2). The diverticulum's tip was strongly adhered to the parietal peritoneum of the abdominal wall at the site of the pelvis, having been pushed up against this site due to the MD's excessively large size and high-volume intestinal content. No other obstruction was observed in the gastrointestinal tract. Resection of the small bowel was performed with a linear stapler and an ileoileal anastomosis was generated using a 80 mm endo-GIA stapler (Figure 3). The resection was completed without incident, and the patient was discharged on post-operative day 6 without any complications. Pathology findings indicated diverticulitis without gastric or pancreatic metaplasia.

DISCUSSION

MD is a true diverticulum, comprising all three layers of the small intestine. Compared to the overall incidence of 0.14%-4.50% (estimated by autopsy findings and retrospective studies)[4], giant MD are rare[5]. The largest giant

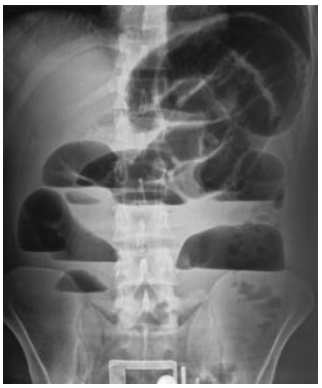

Figure 1 Abdominal X-ray radiography showing air-fluid levels representative of intestinal obstruction.

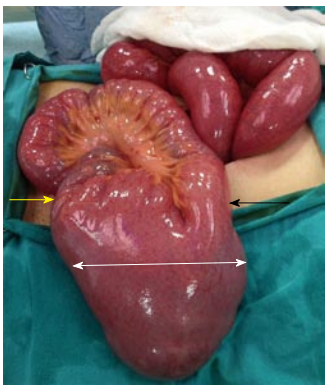

Figure 2 Giant Meckel's diverticulum causing gastrointestinal obstruction. White arrow: Diameter of Meckel's diverticulum; Black arrow: Proximal ileal segment; Yellow arrow: Distal ileal segment.

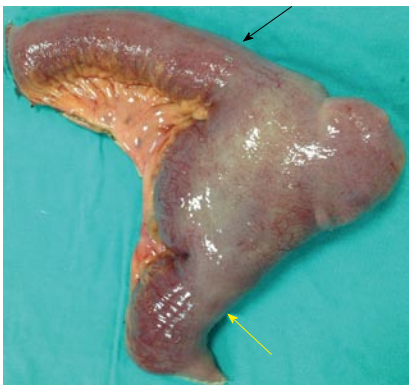

Figure 3 Giant Meckel's diverticulum view after resection. Black arrow: Proximal ileal segment; Yellow arrow: Distal ileal segment.

MDs reported have been > 100 cm long[6], 96 cm long[7], 85 cm long[8], and 66 cm long[9,10].

In adult cases of MD, obstruction is the most frequently reporting presenting symptom[11-14] and can be caused by either the diverticulum's attachment to the umbilicus, abdominal wall or other viscera by a fibrous band or by interference due to the mobility of an unattached diverticulum[11]. Though first hypothesized in 1902[13], these potential reasons for MD-caused intestinal obstruc-

tion remain the features by which MD cases are classified. The obstructions associated with a free or unattached diverticulum, or having only one attachment to the intestine, represent first MD type, and obstructions associated with an attached diverticulum, including through its terminal ligament, to the abdominal wall or intestinal viscus, represent the second type. Between these two types, the former is much rarer.

When the congenital malformation occurs, the free diverticulum forms a volvulus with a loop, twisting the gut structure. Adhesions commonly form between the two arms of the twist, making an obstruction. Subsequent inflammation of the diverticulum further promotes constriction of the bowel. Furthermore, an unattached, distended diverticulum may cause movement of the looped intestine so that a kink forms in the intestine at the attachment point of the diverticulum; this event could lead to an obstruction without any concomitant structural changes in the intestinal wall. Persistence of such kinking may ultimately cause necrosis of the involved and proximal gut tissues. Other potential aetiologies of MD-related intestinal obstructions exist. For example, the obstruction may be caused by twisting of the bowel along its long axis at the point of the diverticulum's origin, by chronic inflammation of the diverticulum and its adjacent bowel, or by inversion of the mucous membrane alone, or of the entire diverticulum, with or without invagination.

Several case reports of MD-related obstructions have described strangulation caused by an adherent diverticulum. Many causes of such an event have been proposed. First, the adherent diverticulum itself may act as a constricting band, such as an adventitious band or a peritoneal adhesion. Second, the adherent diverticulum may have resulted from looping and twisting of the gut in upon itself, forming a volvulus. Third, a volvulus of the attached diverticulum may itself represent a physical obstruction of the intestine. Finally, the diverticular band may become tensely drawn under certain conditions[13].

In a review of 402 patients with MD, 16.9% of the patients were found to have demonstrated symptoms that are considered clinical references for diverticulum[14], with obstruction of the small intestine, and inflammation and bleeding of the lower gastrointestinal tract accounting for 90% of those presenting symptoms. In another study of 34 MD cases, the most common complications were intestinal obstruction (37%), intussusception (14%), inflammation, and rectal bleeding (12%); interestingly, intussusception and volvulus were associated with those patients having intestinal obstruction[15].

For the current case of giant MD, the diverticulum was large in diameter, long in length, and adherent (causing a small bowel obstruction). The structural features of a MD provide clues to the type of complications it may cause. For example, diverticulitis and torsion are common complications observed with long MDs that have a narrow base, while short MDs that have a stumpy base are more often associated with intussusception[16]. Thus, an elongated variant with a narrow neck is more likely to result in torsion, whereas a short, wide-base diverticula may promote foreign body entrapment.

Cullen *et al*[17] studied the outcomes of diverticulectomy surgical management of MD-related complications and determined that the operative mortality and morbidity rates were 2% and 12%, respectively, and that the cumulative risk of long-term post-operative complications was 7%; in contrast, analysis of patients receiving incidental diverticulectomy showed that the operative mortality, morbidity, and risk of long-term post-operative complications were lower (1%, 2%, and 2%, respectively). It is generally recommended that MD discovered incidentally during operation should be removed, regardless of the patient's age.

In conclusion, this report describes a very rare form of acute small bowel obstruction secondary to giant MD encircling the terminal ileum, providing novel insights into this condition and describing its successful management by surgical resection.

COMMENTS

Case characteristics

Clinical symptoms include abdominal pain, nausea, vomiting, and no faecal or gas discharge.

Clinical diagnosis

Acute abdomen, mechanical small bowel obstructions.

Differential diagnosis

Intestinal malrotation, congenital anomalous bands, tumor obstruction.

Laboratory diagnosis

Laboratory tests showed a leukocytosis (11800/μL; 4100-11200), haemoglobin (17.0 g/dL; 12.5-16.0), haematocrit (49.6%; 37.0-47.0) and creatinine (1.4 mg/dL; 0.4-1.2).

Imaging diagnosis

An abdominal X-ray radiography indicated remarkably high air-fluid levels.

Pathological diagnosis

Pathology findings indicated Meckel's diverticulitis without gastric or pancreatic metaplasia.

Treatment

Limited ileal resection and end-to-end anastomosis created with stapler device.

Term explanation

Meckel's diverticulum (MD), a remnant of the vitelline duct that normally disappears at the end of the seventh week of gestation, is the most common congenital abnormality of the small intestine. It arises from the antimesenteric border of the terminal ileum as a true diverticulum that contains all layers of the intestinal wall.

Peer review

This is a well written case report on a farly common subject. It is well know that MD can couse intestinal obstruction and can reach fairly large dimensions, depending on the duration of the sub-occlusive symptoms.

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CASE REPORT

Coexistence of abdominal cocoon, intestinal perforation and incarcerated Meckel's diverticulum in an inguinal hernia: A troublesome condition

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Abstract

Sclerosing encapsulating peritonitis (SEP) is a rare disease entity, in which the small intestine becomes encased and mechanically obstructed by a dense, fibrotic membrane. The disorder is characterized as either primary (idiopathic) or secondary to other causes. The idiopathic cases of SEP, which lack any identifiable etiology according to clinical, radiological and histopathological findings, are also reported under the designation of abdominal cocoon syndrome. The most frequent presenting symptoms of all SEP cases are nausea, vomiting, abdominal distention and inability to defecate, all of which are associated with the underlying intestinal obstruction. Persistent untreated SEP may advance to intestinal perforation, representing a life-threatening condition. However, preoperative diagnosis remains a particular clinical challenge, and most diagnoses are confirmed only when the typical fibrous membrane encasing the small intestine is discovered by laparotomy. Here, we report the clinical presentation of an 87-year-old male with signs of intestinal obstruction and the ultimate diagnosis of concurrent abdominal cocoon, right incarcerated Meckel's diverticulum, and gastrointestinal perforation in laparotomy.

Key words: Sclerosing encapsulating peritonitis; Cocoon syndrome; Perforation; Meckel's diverticulum

Core tip: Abdominal cocoon syndrome, also known as idiopathic sclerosing encapsulating peritonitis, is a rare disease entity, in which the small intestine becomes encased and mechanically obstructed by a dense, fibrotic membrane. While some patients with cocoon syndrome remain asymptomatic, the majority experience gastrointestinal symptoms, including recurrent attacks of acute, sub-acute or chronic gastrointestinal obstruction, weight loss, loss of appetite, and development of a palpable abdominal mass. Herein, we describe an elderly patient who presented with signs of intestinal obstruction and who was diagnosed with concurrent abdominal cocoon, right incarcerated Meckel's diverticulum, and gastrointestinal perforation by exploratory laparotomy.

Akbulut S, Yagmur Y, Babur M. Coexistence of abdominal cocoon, intestinal perforation and incarcerated Meckel's diverticulum in an inguinal hernia: A troublesome condition. *World J Gastrointest Surg* 2014; 6(3): 51-54 Available from: URL: http://www.wjgnet.com/1948-9366/full/v6/i3/51.htm DOI: http://dx.doi.org/10.4240/wjgs.v6.i3.51

INTRODUCTION

Sclerosing encapsulating peritonitis (SEP) is a rare structural abnormality whereby the small intestine becomes encased (or cocooned) by a dense fibrocollagenous

membrane that causes intestinal obstruction[1,2]. First described in 1907 as "peritonitis chronica fibrosa incapsula" by Owtschinnikow[1,3-7], subsequent case reports have described the disease condition as primary (idiopathic) or secondary, depending on the underlying etiological causes. The idiopathic form is also reported under the alternative descriptive name of "abdominal cocoon syndrome"[1,3,4,8,9].

The most common clinical signs and symptoms of SEP include nausea, vomiting, abdominal distention and inability to defecate, all of which are indicative of gastrointestinal obstruction. Rare cases of SEP manifest the severe complication of perforation. The relatively nonspecific nature of the common symptomology makes preoperative diagnosis a clinical challenge, and many cases are only diagnosed during laparotomy[3,6]. In this case report, we describe an elderly patient who presented with signs of intestinal obstruction and who was diagnosed with concurrent abdominal cocoon, right incarcerated Meckel's diverticulum, and gastrointestinal perforation by exploratory laparotomy.

CASE REPORT

An 87-year-old male patient presented to our emergency department upon referral from an outside center for management of severe abdominal pain, nausea, vomiting and inability to defecate for 3 d. The patient's medical history was generally unremarkable, with hypertension and bronchial asthma the only major afflictions. Physical examination findings were distended abdomen, sunken eyes, dry mucosa, and vital signs consistent with hypovolemia and sepsis (blood pressure: 100/60 mmHg; pulse: 90 beats per minute; body temperature: 38.1 ℃). In addition, severe rebound tenderness was observed in all abdominal quadrants, with the most severe being in the right lower quadrant. Findings from laboratory tests were white blood cell count of 13900/μL (neutrophils: 83.2%), hemoglobin level of 11.4 g/dL, blood urea nitrogen level of 60 mg/dL, and creatinine level of 1.5 mg/dL. Abdominal imaging examination revealed diffuse air-fluid levels (by X-ray; Figure 1) and free fluid in the pelvis and right lower quadrant, as well as dilatation and edema in all intestinal segments (by ultrasonography, United States). Exploratory laparotomy was performed according to the initial diagnosis of perforation, mechanical bowel obstruction due to tumor, and mesenteric vascular disease. All intestinal segments from 60 cm distal to the Treitz ligament to the ileocecal valve were found to be dilated and encapsulated (Figure 2). Meckel's diverticulum, at the 60 cm proximal to ileocecal valve, was incarcerated into the right inguinal canal. This clinicopathologic condition is also referred to as Littre hernia (Figure 3). Some intestinal segments at 100 cm proximal to the ileocecal valve were conglomerated and adhered to the retroperitoneum. In addition, a 1 cm perforation immediately proximal to the conglomerated segment and an abscess located posterior to the conglomerated segment were detected. The

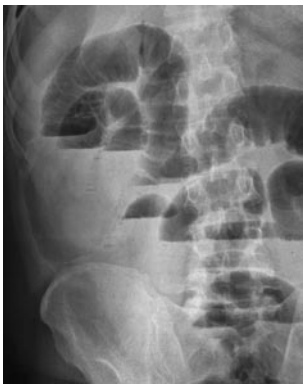

Figure 1 X-ray plain abdominal imaging (posteroanterior) revealed intestinal obstruction with marked small bowel air-fluid levels.

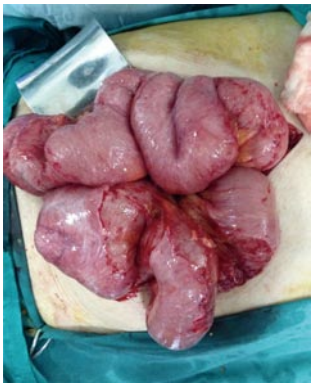

Figure 2 Intraoperative photographs taken along the midline incision and showing the encapsulated small bowel segments with a dense fibrous layer.

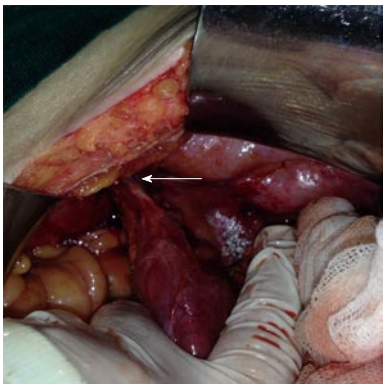

Figure 3 Intraoperative photographs showing the Meckel's diverticulum incarcerated within the right inguinal canal (arrow).

intra-operative management was initiated by performing decapsulation and adhesiolysis of all encapsulated segments, and followed by freeing of the incarcerated intestinal segments and closing of the mouth of the hernia sac. Then, the conglomerated and perforated ileal segment was resected and the abdominal cavity was irrigated. A side-to-side ileoileal anastomosis was constructed using an Endo-GIA stapler (Covidien, Dublin, Ireland), and a loop ileostomy was constructed at 40 cm proximal

to the anastomosis site. Post-operative recovery was uncomplicated and the patient was discharged to home at 12 d after the surgery. The histopathologic examination of the excised peritoneal capsule showed proliferation of the fibroconnective tissue with signs of a non-specific inflammatory reaction. Diagnosis of SEP was established based on the intraoperative view, results from histopathologic examination, and findings from other clinical and biochemical analyses.

DISCUSSION

SEP is a disease entity characterized by partial or complete encasement of small intestine by a thick fibrotic membrane[1,5]. This fibrocollagenous membrane may extend to encase other proximal organs as well, such as stomach, liver and large intestine; in some forms of the disease, this extension of the encasing membrane can cause segregation of the intraperitoneal organs (as if they were extraperitoneal)[8]. Although epidemiological studies of SEP have not fully elucidated the precise etiologic profile of primary/idiopathic or secondary SEP[4], they have revealed a trend in incidence involving young females living in temperate geographic zones. It has been speculated that infections of the Fallopian tubes or retrograde menstruation may be related to disease onset and progression[7]. In addition, incidence of SEP is not infrequent in patients with ambulatory peritoneal dialysis, suggesting that this condition may represent an etiology of secondary SEP[6,10]. Other cases of SEP have been reported in patients with abdominal tuberculosis, sarcoidosis, gastrointestinal malignancies, systemic lupus erythematosus, familial Mediterranean fever, with fibrogenic foreign materials, undergoing beta-blocker (practolol) therapy[4,7,9-12], fitted with ventriculoperitoneal and peritoneovenous shunts, recipients of orthotopic liver transplantation, and suffering from recurrent peritonitis attacks[6]. The current patient, described herein, had no chronic disease history and normal test results from biochemical (erythrocyte sedimentation rate) and microbiological (blood and peritoneal fluid culture, and PPD skin test) assays. Therefore, we considered that this case was likely primary SEP.

While some SEP patients remain asymptomatic, the majority experience gastrointestinal symptoms, including recurrent attacks of acute, sub-acute or chronic gastrointestinal obstruction, weight loss, loss of appetite, and a palpable abdominal mass[1,3,6,8]. Additionally, some of the SEP patients with severe abdominal distention also have ascites[3]. Gastrointestinal perforation is a relatively rare complication of SEP. To our knowledge only one case of SEP-related perforation has been reported, and the etiology was evidenced as tuberculosis[12]. The current case herein also had an ileal perforation, but the absence of any clear etiologic factor (primary SEP) makes this case unique in the literature.

SEP is suspected according to the gastrointestinal clinical signs coupled with suggestive findings from physical examination, biochemical testing, and radiological analyses [*i.e.,* X-ray, barium studies, United States, computed tomography (CT)]. Unfortunately, preoperative diagnosis of SEP remains a challenge and intraoperative findings and histopathological data are required for a definitive diagnosis[9]. Malignancy (particularly colorectal tumor) is the first differential diagnosis considered for individuals with advanced age, unremarkable medical history, physical symptoms of intestinal obstruction, and abdominal X-ray detection of air-fluid levels[1,2,6]. Furthermore, ulcerative perforation, septic peritonitis, and tuberculotic peritonitis encapsulans should be considered in the differential diagnosis.

Barium-enhanced imaging is not always possible in patients with marked intestinal obstruction. Abdominal US can demonstrate a dilated intestinal segment, free fluid accumulation, and the status of the peritoneal membrane (when sufficiently thickened)[4]. Abdominal United States examination of the current case showed free fluid in the pelvis and right lower quadrant, as well as dilatation and edema in all intestinal segments. Nonetheless, CT is considered the best imaging modality for diagnostic purposes, as it can show thickened peritoneum and mesentery as well as capsulated intestinal loops[13]. The typical CT finding for SEP is intestinal loops conglomerated at the midline and encased by a dense mantle without contrast uptake[5,6,13]. Since an urgent laparotomy had to be performed on our patient to address the severe distention, we did not have the opportunity to carry out a CT scan.

The typical finding of SEP is a bobbin-like appearance of the intestine, which results from the partial or whole encasement of intestinal segments by the thick, dense membrane[9]. Upon histopathological examination, the encapsulating membrane appears as a thickened and inflamed vascular fibrocollagenous tissue, with infiltrating lymphocytes and plasma cells[5]. The efficacy and safety of surgical treatment for cases that have a surgical indication and are confirmed by laparotomy remain unknown. Review of the literature suggests that many surgeons favor a minimally invasive approach to treating SEP[6], possibly because the sclerotic membranes on intestinal surfaces (and also between segments) can be easily removed in mild cases without marked intestinal obstruction. However, it is almost impossible to remove the sclerotic membrane successfully in patients with complete intestinal obstruction.

Clinical suspicion and early diagnosis of SEP are crucial to disease and treatment outcome[7]; overly aggressive surgical intervention in SEP cases with severe adhesions may cause multiple perforations. In summary, the basic approach of surgical treatment should include freeing of adhesions, total excision of the membrane, or partial intestinal resection when necessary[9]. It has been reported that the morbidity and mortality rates are higher in patients undergoing intestinal resection[9].

Entirely by coincidence, several clinical conditions were present simultaneously in the case presented herein. The clinical picture of this patient was made even worse

by the collective presence of intestinal perforation, a focus of interloop abscess extending to retroperitoneum, and a Meckel's diverticulum incarcerated into the right inguinal canal. Fortunately, the patient experienced an uncomplicated recovery following the surgical treatment, at least partially due to the well-managed clinical care that was given.

In conclusion, SEP is a rare disease entity causing intestinal obstruction. Preoperative diagnosis is considerably difficult to make, and the majority of previously reported cases have been diagnosed incidentally during laparotomy[11]. Some imaging methods may help clinicians to make the diagnosis of suspected cases. Surgery is an important treatment modality for SEP, but the dissections must be made carefully to free the small intestine and resect the affected tissues and to allow a complete cure[3,6].

COMMENTS

Case characteristics

Clinical symptoms included severe abdominal pain, nausea, vomiting and inability to defecate.

Clinical diagnosis

Mechanical small bowel obstruction.

Differential diagnosis

Gastrointestinal malignancy, perforation, herniation, and mesenteric vascular disease.

Laboratory diagnosis

Laboratory analysis showed leukocytosis (neutrophils: 83.2%) and elevated levels of renal function markers.

Imaging diagnosis

Abdominal X-ray examination showed diffuse air-fluid levels and abdominal ultrasonography revealed free fluid in the lower right quadrant and pelvis, as well as dilatation and edema in all intestinal segments.

Pathological diagnosis

Histopathologic examination of the excised peritoneal capsule showed proliferation of fibroconnective tissue with signs of a non-specific inflammatory reaction. The diagnosis of abdominal cocoon syndrome was established according to the intraoperative view and by ruling-out any other causes.

Treatment

The operative treatment included decapsulation, adhesiolysis, partial small bowel resection, side-to-side ileoileal anastomosis (using an endoscopic stapler), and a loop ileostomy (opened at 40 cm proximal to the anastomosis site).

Related reports

Early diagnosis of abdominal cocoon syndrome is crucial to disease and treatment outcome; overly aggressive surgical intervention in cocoon cases may cause severe adhesions. The basic approach of surgical treatment should include freeing of adhesions, total excision of the membrane, or partial intestinal resection when necessary, as in our case.

Term explanation

Sclerosing encapsulating peritonitis, also known abdominal cocoon syndrome, is a rare structural abnormality whereby the small intestine becomes encased (or cocooned) by a dense fibrocollagenous membrane that causes intestinal obstruction.

Peer review

This paper demonstrates coexistence of abdominal cocoon, intestinal perforation and incarcerated Meckel's diverticulum in an inguinal hernia, and considered to be well written.

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