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Perforation of Meckel's diverticulum in a patient with sclerosing mesenteritis – a case report

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ABSTRACT

Sclerosing mesenteritis is a rare abdominal condition characterized by chronic inflammation and the fibrosis of fat tissue, typically involving the small bowel mesentery. This article presents a case of a 62-year-old patient suffering from sclerosing mesenteritis, whose disease was complicated by perforation of his Meckel's diverticulum. Proper diagnosis of perforation was delayed due to the presence of ascites which masked signs of peritonitis. The patient was operated on and the perforated diverticulum was resected. The post-operative course was complicated by an episode of intraabdominal bleeding, however, the patient survived. The presented case is probably the first description of a perforation of Meckel's diverticulum in a patient suffering from sclerosing mesenteritis.

Keywords: sclerosing mesenteritis; Meckel's diverticulum – complications; bowel perforation.

INTRODUCTION

Sclerosing mesenteritis (also named mesenteric panniculitis, retractile mesenteritis or mesenteric lipodystrophy) is a rare abdominal condition initially described in 1924, with a prevalence rate reported to be less than 1% in the general population [1]. It is characterized by chronic inflammation and fibrosis of fat tissue, typically involving the small bowel mesentery. The aetiology of the disease remains poorly understood but it may be caused by trauma (including surgery), autoimmune reaction, or malignancy. There may be various clinical manifestations which means it is often unlikely to be considered as a diagnosis, however diffuse abdominal pain is the most frequently reported symptom [2, 3]. The clinical course is generally benign with a number of patients diagnosed incidentally on imaging obtained for other reasons. In some patients, the symptoms may be caused by a mass effect on the bowel, resulting in mechanical obstruction or a vascular compromise of the intestine [1]. Treatment of sclerosing mesenteritis is not uniformly determined; it may include immunosuppressive therapy for patients only presenting with pain, or surgical intervention for patients presenting with obstructive symptoms or bowel ischemia.

In this article we present a case of a patient suffering from sclerosing mesenteritis whose disease was complicated by a perforation of Meckel's diverticulum. Both conditions are very rare but their coincidence in 1 patient is almost unheard of.

CASE REPORT

In November 2019, a 62-years-old patient suffering from sclerosing mesenteritis and diabetes mellitus type II was admitted to the Department of Rheumatology due to abdominal pain, ascites and signs and symptoms of sepsis. Diagnosis of sclerosing mesenteritis was made in April 2019 based on an abdominal computed tomography (CT) scan which showed a solid tumour in the mesentery, sized at 8 x 7 x 4 cm and arousing suspicions of desmoid or lymphoma (Figs. 1 and 2). For this reason, the patient was given a diagnostic laparoscopy with a biopsy of the lesion; histopathological examination of the specimens revealed a diagnosis of sclerosing mesenteritis. After establishing a correct diagnosis, an immunosuppressive therapy was commenced with endoxane and methylprednisolone, resulting in a gradual withdrawal of symptoms and partial resolution of the tumour observed on the CT scan. The patient received 6 courses of immunosuppressive treatment, lasting 5 days each and repeated in 1-month intervals.

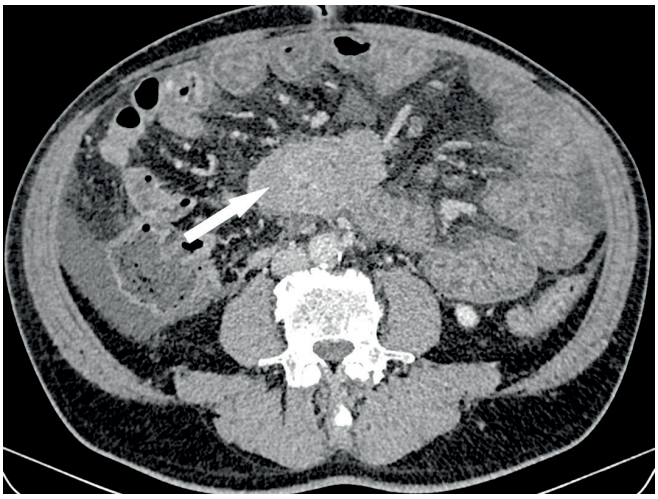

FIGURE 1. Computed tomography scan of the abdomen showing the tumor (marked with a arrow) in the mesentery of the small bowel (coronal view)

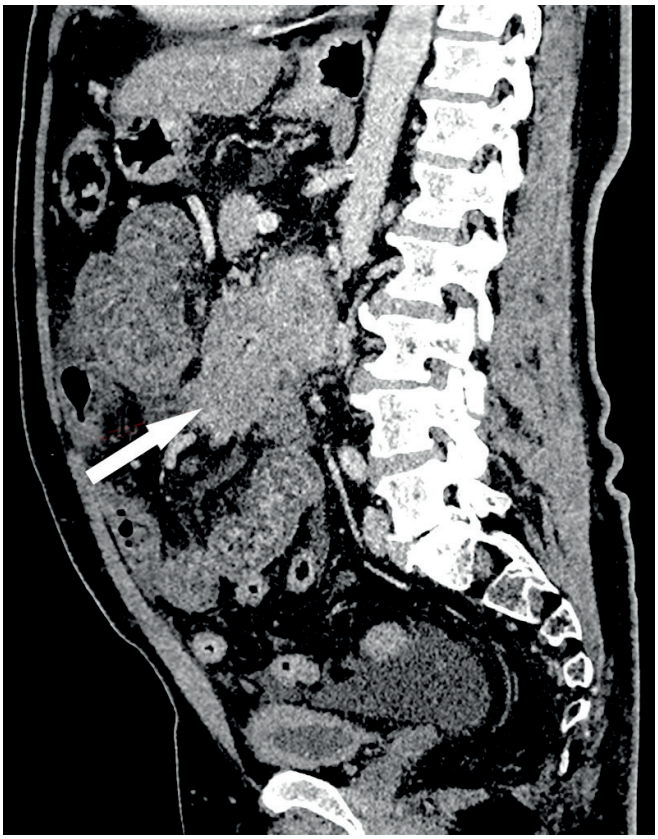

FIGURE 2. Computed tomography scan of the abdomen showing the tumor (marked with a arrow) in the mesentery of the small bowel (saggital view)

At the point of admission to the hospital in November 2019, the patient suffered from diffuse abdominal pain, weakness and a fever of 39°C which had lasted for 2 days. Biochemical tests showed an elevated C-reactive protein (CRP) concentration of 340 mg/L and procalcitonin of 8.5 mg/L, suggesting sepsis. The plain abdominal X-ray was normal and the ultrasonography only showed a moderate volume of ascites. A blood culture revealed *Escherichia coli* and *Klebsiella pneumoniae* which confirmed diagnosis of sepsis. An intensive, aimed antibiotic therapy was commenced resulting in clinical improvement and a significant reduction of inflammatory parameters. Unfortunately, 16 days after admission, the condition of the patient suddenly deteriorated: abdominal pain reoccurred and the volume of ascites increased. This was followed by an elevation of inflammatory parameters (CRP 310 mg/L and procalcitonin 4.5 mg/L). A laparocentesis was performed and 1800 mL of turbid fluid was evacuated from the abdominal cavity which was sent for bacterial culturing. An abdominal X-ray showed the presence of gas in the abdominal cavity, suggesting perforation of the digestive tract. The patient was sent for an emergency laparotomy.

Intraoperative findings

The abdominal cavity was opened via midline incision and a large volume (8 L) of turbid fluid was evacuated. Loops of small intestine were swollen and matted in 1 mass; the mesentery was hard and stiff. Further exploration revealed a perforated Meckel diverticulum at the top of the intestinal mass which had adhered to the anterior abdominal wall. Two undigested pills were found and evacuated from the lumen of the diverticulum. The surface of the small intestine and abdominal wall were coated by a layer of fibrin, suggesting a neglected perforation (occurring at least several days earlier). A meticulous debridement of the abdominal cavity was conducted by means of cleaning the infected fibrin from the intestine and abdominal wall. Meckel's diverticulum was resected, and the bowel was closed by 2 layers of stitches. A stapler was not used due to the inflammation of adjacent tissue. Three drains were left in the abdominal cavity before closure. Cultures from the fluid obtained at laparotomy revealed *Enterococcus faecium* and *Candida glabrata* (fungus) and aimed antibiotic therapy was commenced as appropriate.

Postoperative course

Parenteral feeding was commenced after the operation followed by a light oral feeding from the 3rd postoperative day. The condition of the patient gradually improved, but leakage of 0.5–1.0 L of turbid and blood-stained fluid persisted. A week after the operation, one of the drains was retrieved from the abdominal cavity; several hours thereafter, a leakage of dark, blood-stained fluid with clots was observed from a drain-hole. By the next day, 700 mL of this fluid had been evacuated and the condition of the patient deteriorated: he felt weak and blood tests revealed anaemia (haemoglobin = 7.4 mg%, E = 2.6 T/L, Ht = 28%). A transfusion of 4 units of red-cell concentrate was given and a re-operation was conducted. During the 2nd laparotomy, approx. 500 mL of blood clots were evacuated from the abdominal cavity – mostly from the right hypogastrium. No apparent source of bleeding was found in the intestine nor in the mesentery, but a leakage of a small volume of blood persisted from the surface. Therefore, a decision of leaving dressings in the abdominal cavity (packing), mostly in the right hypogastrium, was made before closure of the abdomen. The day after the 2nd laparotomy was performed, blood leakage was not observed and dressings were evacuated. Fluid obtained during the 2nd laparotomy was cultured and revealed *Klebsiella pneumoniae* ESBL. The postoperative course was uneventful after reoperation, the patient gradually improved and on the 16th day after the 1st operation he was transferred back to the Department of Rheumatology.

DISCUSSION

The presented case is interesting because of the simultaneous occurrence of 2 very uncommon conditions: sclerosing mesenteritis and perforated Meckel's diverticulum. The authors are not aware of any similar cases existing in the literature. A *post-hoc* analysis of the clinical course suggests that the direct cause of sepsis at presentation of the patient was Meckel's diverticulitis. In normal conditions, this does not cause sepsis but mild to moderate abdominal disease, however, our patient's immunity was compromised by ongoing chemotherapy. This was enough to develop serious complications in the course of Meckel's diverticulitis.

An antibiotic therapy commenced at admission and resulted in partial clinical improvement but did not prevent perforation of the inflamed diverticulum which probably occurred several days prior to making a confident diagnosis. This relatively mild clinical course of perforation was caused by a leakage of a small volume of intestinal fluid into the abdominal cavity which was filled with ascites; this resulted in the dissolution of the irritating substance and subsequently masked the peritoneal signs which are normally, at perforation of the bowel, strongly expressed (guarding, rigidity, rebound tenderness).

LITERATURE REVIEW

Several reports presenting single cases of surgically treated sclerosing mesenteritis are available in the literature. Corado et al., reported a case of a middle-aged male patient presenting with signs and symptoms of a mechanical obstruction in the small bowel. The patient had no previous laparotomy or history of chronic abdominal disease. As the obstruction was absolute, he was given surgery; during laparotomy, a stenosis of the distal ileum with mesenteric thickening was found requiring resection of the involved intestine. In the postoperative period, symptoms of mechanical obstruction were not resolved and re-laparotomy was necessary on the 4th postoperative day. This revealed a highly aggravated fibrosing process in the mesentery with a mechanical compression on the remaining bowel. For this reason, an ileo-colic resection was performed followed by end-to-end anastomosis. After obtaining the histological diagnosis, immunotherapy was commenced with clinical improvement [4].

Gupta et al. presented a case of a 68-year-old male with a history of mild abdominal pain lasting 2 months and associated with low-grade fever and intermittent constipation. During investigations, an abdominal CT scan revealed a mesenteric tumour which was suspected of malignancy; no biopsy was performed and the patient was given surgery. During laparotomy a tumour in the mesentery was radically excised followed by a resection of the corresponding bowel. The post-operative course was uneventful. Histopathology showed a poorly defined lesion comprised of nodular, poorly defined proliferation of bland looking fibroblast like cells, suggestive of sclerosing mesenteritis. The patient was prepared for further immunotherapy [5].

Butt et al. reported a case of a 60-year-old male with a history of abdominal pain, distention, anorexia and weight loss for several weeks. During investigation, a CT scan revealed a mesenteric tumour with surrounding stranding and poorly defined borders. The patient underwent an exploratory laparotomy and complete resection of the tumour, without any compromise to the vascularity of the corresponding bowel. The post-operative course was uneventful. Histopathological examination was relatively complicated but, it eventually revealed fibro-adipose tissue with lymphoid hyperplasia, vague nodular collections of foamy histiocytes with giant cell reaction, marked chronic inflammation, fat necrosis and prominent sclerosis/fibrosis leading to a final diagnosis of IgG4-related sclerosing mesenteritis. The patient did not require any other therapy and was disease free at the 6-month follow up [6].

Ueno et al. reported a case of an 82-year-old female presenting with appetite loss, weight loss and epigastric pain. A CT scan of the abdomen and pelvis showed increased density of the mesentery adjacent to the small bowel and enlarged lymph nodes suggesting sclerosing mesenteritis. Malignancies, such as lymphoma or leukaemia, were also considered. Unlike in previously reported cases, the CT and US-guided needle biopsy were performed with a finer than normal needle, which allowed a confident histological diagnosis of sclerosing mesenteritis. The patient avoided unnecessary surgery but was treated with corticosteroids resulting in gradual resolution of symptoms and radiographic findings [7].

Sharma et al. presented a systematic review of the literature on sclerosing mesenteritis. The authors found reports of 192 cases of the condition, predominantly in males (69% male vs. 31% female), with a mean age of 55 years. In 55 patients (29%) some possible predisposing factors were identified, including abdominal surgery or trauma, history of malignancy or autoimmune disease. Most of the patients (n = 164.85%) underwent surgical abdominal exploration prior to diagnosis. In almost half (n = 80) of these patients, resection of the involved bowel and mesentery was necessary, however, in the other half, adequate specimens for histopathological examination were obtained. Approximately $^1/_3$ (n = 67) of patients required medical treatment with the majority receiving steroids. The most common complications in the course of the disease included mechanical obstruction and ischemia of the small intestine (n = 20). There were a total of 14 deaths, the majority of which were secondary to disease related complications [3].

The presented case is, to date, the 1st report on perforation of Meckel's diverticulum in patients suffering from sclerosing mesenteritis. Due to the simultaneous occurrence of these pathologies, the course of the disease was more serious than if perforation would occur in a healthy person. This also contributed to the development of postoperative complications and the delay in making a proper diagnosis.

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