

Cytomegalovirus enteritis with jejunal perforation in a patient with endometrial adenocarcinoma

Young Jin Jun, Jongmin Sim, Hye In Ahn, Hulin Han, Hyunsung Kim, Kijong Yi, Abdul Rehman, Se Min Jang, Kiseok Jang, Seung Sam Paik

Young Jin Jun, Jongmin Sim, Hye In Ahn, Hulin Han, Hyunsung Kim, Kijong Yi, Abdul Rehman, Se Min Jang, Kiseok Jang, Seung Sam Paik, Department of Pathology, College of Medicine, Hanyang University, Seoul 133-792, South Korea
Author contributions: Jun YJ and Sim J designed the research; Ahn HI and Han H performed the research; Kim H and Yi K contributed new reagents and analytical tools; Rehman A, Jang SM and Jang K analyzed the data; Jun YJ and Paik SS wrote the paper.

Correspondence to: Seung Sam Paik, MD, Department of Pathology, College of Medicine, Hanyang University, 222 Wangsimni-ro, Seongdong-gu, Seoul 133-792, South Korea. sspaik@hanyang.ac.kr

Telephone: +82-2-22908252 Fax: +82-2-22967502

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with a jejunal perforation.

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Core tip: Small bowel involvement with gastrointestinal cytomegalovirus (CMV) infection is very rare. However, CMV enteritis should be included in the differential diagnosis of the ulcerative lesion of a small bowel segment when abdominal pain, vomiting, diarrhea and perforation develop in patients with a history of cancer.

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Abstract

Cytomegalovirus (CMV) infection of the gastrointestinal tract has been reported most frequently in the setting of immunodeficiency. The whole gastrointestinal tract can be affected; however, the small bowel is rarely affected. We report a case of CMV enteritis with jejunal perforation in a 53-year-old woman with a history of chemoradiation therapy for endometrial cancer 8 years previously. At follow-up evaluation, lower abdominal pain, diarrhea and vomiting appeared. Abdominal computed tomography showed intra-abdominal free air in the subphrenic space and porta hepatis. The jejunal segment revealed serosal purulent exudates with a perforation. The resected jejunal segment showed a large geographic ulcerative mucosal lesion. The microscopic findings revealed a diffuse ulcerative mucosal change with a prominent granulation tissue formation and many large atypical vascular endothelial cells and stromal fibroblasts with intranuclear or intracytoplasmic inclusion bodies. These cells were positive for CMV antibody. The final diagnosis was CMV-associated jejunitis

INTRODUCTION

Cytomegalovirus (CMV) infection commonly develops in immunocompromised patients and is a major cause of morbidity and mortality^[1]. Most cases occur in patients with human immunodeficiency virus infection, undergoing cancer chemotherapy, receiving long-term corticosteroid treatment, and organ transplant recipients^[2,3]. It may affect the gastrointestinal tract anywhere from the mouth to the anus. The site most commonly affected is the colon, followed by duodenum, stomach, esophagus and small intestine^[4,5]. Esophagitis, gastritis, duodenitis and enterocolitis are induced by CMV infection in the gastrointestinal tract. However, intestinal perforation is relatively rare^[6]. The most common site of perforation with CMV infection of the gastrointestinal tract is the colon,



Figure 1 Abdominal computed tomography revealed intra-abdominal free air in the right subphrenic space (arrows).

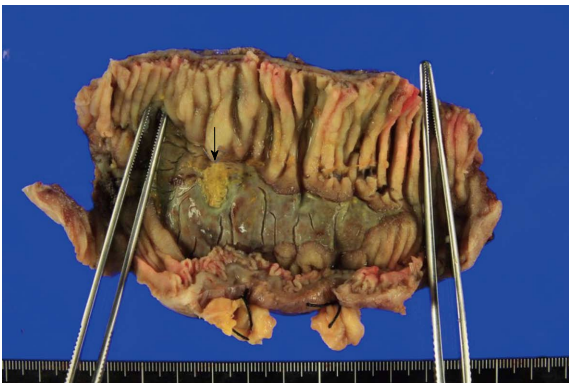


Figure 2 The resected jejunal segment showed a large geographic ulceration with a perforation site (arrow).

followed by the ileum and appendix^[7]. Jejunal perforation due to gastrointestinal CMV infection is extremely rare. Only five cases have been reported in the English literature^[8-12]. Here, we report a case of CMV enteritis with a jejunal perforation in a patient with endometrial adenocarcinoma.

CASE REPORT

A 53-year-old woman with a history of endometrial cancer surgery visited the emergency room with left lower abdominal pain. She had a one week history of diarrhea and vomiting. She had undergone an extended abdominal hysterectomy with bilateral salphingo-oophorectomy and pelvic lymph node dissection for endometrial adenocarcinoma and received chemotherapy and radiation therapy 8 years previously. On physical examination, she complained of abdominal distension and generalized abdominal tenderness with muscle guarding. Clinically, generalized peritonitis was suspected. Simple X-ray and computed tomography of the abdomen demonstrated free intraperitoneal air in the right subphrenic space and porta hepatis (Figure 1). Radiologically, the possibility of intestinal perforation was suspected. She underwent an emergency laparotomy and a perforation was found in a segment of the jejunum with a serosal grayish white exudative covering. The affected

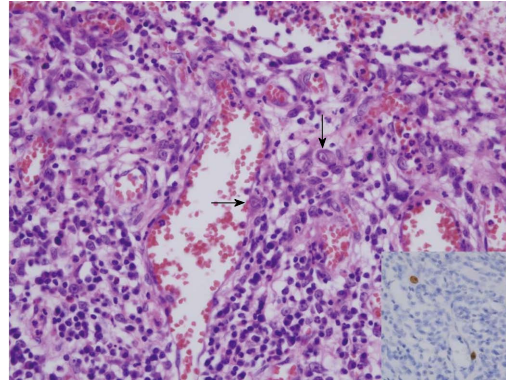


Figure 3 The ulcer bed was composed of granulation tissue with abundant vascular proliferation. Many large atypical endothelial cells and stromal fibroblasts with the formation of intranuclear inclusion bodies were noted (arrows). These cells were positive for cytomegalovirus antibody (Inset).

jejunal segment was resected.

The resected jejunal segment measured 10 cm in length and 7 cm in circumference. The outer surface showed a perforation site with serosal purulent exudates. The mucosal surface of the jejunal segment revealed a diffuse geographic ulcerative lesion which measured 9.5 cm × 3.5 cm in size. The ulcerative lesion showed an irregular, dirty mucosal surface and a perforation site was noted (Figure 2). Microscopically, the jejunal wall showed a diffuse ulceration with exuberant granulation tissue formation and heavy inflammatory cell infiltration. Many large atypical vascular endothelial cells and stromal fibroblasts with intranuclear or intracytoplasmic inclusion bodies were found in the granulation tissue area (Figure 3). The features of vasculitis were combined. The immunohistochemical staining using monoclonal anti-CMV antibody revealed many positive nuclear reactions of large atypical cells with or without intranuclear inclusion bodies (Figure 3, inset).

DISCUSSION

In this report, we have described a rare case of CMV enteritis with a jejunal perforation in a patient with a history of endometrial cancer surgery and chemoradiation therapy. To the best of our knowledge, only five cases of CMV enteritis with a jejunal perforation have been reported^[8-12]. The reported five cases are summarized in Table 1. Four cases were male and one case was female. The mean age was 42.4 years (range: 28 to 60 years). The clinical presentations were lower abdominal pain, diarrhea, fever, nausea, loss of appetite, intermittent epigastralgia and emesis. The underlying diseases were acquired immunodeficiency syndrome (AIDS) in three patients, adult T-cell leukemia-lymphoma in one patient and no underlying disease in one patient. Our case was a 53-year-old woman with a clinical presentation of left lower abdominal pain, diarrhea and vomiting and a history of endometrial adenocarcinoma.

Cytomegalovirus infection is a well-known opportunistic viral infection that frequently occurs in im-

Table 1 Summary of reported five cases of cytomegalovirus enteritis with jejunal perforation.

Cases	Sex	Age	Perforation sites	Clinical presentation	Underlying disease	Ref.
1	M	50	Jejunum	Lower abdominal pain, nausea, emesis, diarrhea	AIDS	[7]
2	M	34	Jejunum and ileum	Loss of appetite, intermittent epigastralgia	Adult T-cell leukemia-lymphoma	[8]
3	M	28	Jejunum and ileum	Chronic diarrhea, fever, abdominal pain	AIDS	[9]
4	M	40	Jejunum	Lower abdominal pain, fever	AIDS	[10]
5	F	60	Jejunum	Diarrhea, fever	None	[11]
Our case	F	53	Jejunum	Left lower abdominal pain, diarrhea, vomiting	Endometrial adenocarcinoma	

M: Male; F: Female; AIDS: Acquired immunodeficiency syndrome; Ref.: Reference number.

munocompromised patients, including patients with AIDS, those who have received bone marrow or organ transplants, those who received long-term corticosteroid treatment and those who have received chemotherapy or radiation therapy^[2,6]. CMV is a member of the herpes viral group and is a DNA virus. More than 90% of healthy adults are seropositive for CMV^[3,6]. Pulmonary infection is the most frequently recognized type of CMV infection. However, CMV infection of the gastrointestinal tract is also common^[13]. It can affect the gastrointestinal tract anywhere from the mouth to the anus. In CMV infection of the gastrointestinal tract, the site most commonly affected is the colon (47%), followed by the duodenum (21.7%), stomach (17.4%), esophagus (8.7%) and small bowel (4.3%)^[4]. Intestinal perforation as a complication of gastrointestinal CMV infection is a rare finding. The most common site of perforation is the colon (53%), followed by the ileum (40%) and appendix (7%)^[7]. The jejunum, as a perforation site in gastrointestinal CMV infection, is an extremely rare location. In our case, the patient presented with a gastrointestinal CMV infection manifested as jejunitis which led to a jejunal perforation.

CMV-associated enteritis appears to be related in part to up-regulation of the production of local pro-inflammatory cytokines, potentially by altering resident intestinal macrophages to express human immunodeficiency virus proteins^[14]. The CMV-induced ulceration is thought to involve ischemic mucosal injury secondary to the infection of the vascular endothelial cells^[5,15]. CMV may infect various gastrointestinal cells. The most commonly affected cells are the vascular endothelial cells and stromal fibroblasts. CMV infection in the vascular endothelial cells leads to abnormal cellular swelling and enlargement, vascular luminal compromise, fibrin thrombus formation, local vasculitis and tissue damage in the intestinal segment supplied by the affected vessels^[3,15]. Finally, intestinal ulceration is developed in the infected intestinal segment. Our case showed a large geographic ulceration with exuberant granulation tissue formation. CMV infection is frequently found in the vascular endothelial cells and stromal fibroblasts, which is supportive of the reported pathophysiology of CMV-induced ulceration.

In conclusion, the small bowel is a rare site of involvement by gastrointestinal CMV infection and CMV enteritis with jejunal perforation is extremely rare. However, CMV enteritis should be considered as a possible di-

agnosis in the ulcerative lesion of a small bowel segment when abdominal pain, vomiting, diarrhea and perforation develop in patients with a history of endometrial cancer.

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