IDCases 26 (2021) e01246

Contents lists available at ScienceDirect

IDCases

journal homepage: www.elsevier.com/locate/idcr

Case report

Passage of intestinal casts—An unusual presentation of Cytomegalovirus enterocolitis

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ARTICLE INFO

Article history:
Received 29 July 2021
Accepted 5 August 2021

Keywords:
Colitis
Cytomegalovirus
Infection
Immunosuppression
Diarrhea

ABSTRACT

Cytomegalovirus (CMV) is one of the leading opportunistic pathogens affecting immunocompromised patients. We report a case of histologically-confirmed extensive CMV enterocolitis in a young woman after receiving rituximab and tocilizumab for the treatment of autoimmune encephalitis. During the antiviral treatment, she spontaneously excreted small intestinal casts per oral and colonic casts per anus. Even though intestinal cast is an extremely unusual condition, CMV infection should be included in the differential diagnosis.

Introduction

Cytomegalovirus (CMV) is a DNA virus and is a member of the Herpesviridae family. Typically, symptomatic CMV-related diseases mostly affect immunocompromised patients (i.e. HIV-infected patients or following solid organ transplantation). Intestinal cast is an extremely rare condition, resulting from extensive enterocolitis. Here, we describe a case of patient who spontaneously passed small intestinal and colonic casts as a consequence of extensive CMV enterocolitis following immunosuppressive treatment for autoimmune encephalitis.

Case report

A 35-year-old woman without remarkable underlying disease was referred to our hospital due to uncontrolled new-onset status epilepticus. She was admitted to the intensive care unit and was subsequently diagnosed with autoimmune encephalitis. She was treated with anti-epileptic drugs and intravenous methylprednisolone. On the fourth week of the admission, she received intravenous rituximab (an anti-CD20 monoclonal antibody) and tocilizumab (an anti-interleukin-6 receptor antibody) due to uncontrolled epilepsy. During the immunosuppressive treatment, she developed hospital-acquired pneumonia and was treated with intravenous carbapenem, colistin, and tigecycline. One week later, she developed high-grade fever, progressive abdominal distension, vomiting, feeding intolerance and watery diarrhea. Stool PCR test for *Clostridium difficile* and stool bacterial culture were negative. Contrast-enhanced CT scan of the abdomen revealed diffuse thickening of small and large bowel walls with surrounding peri-intestinal fat stranding (Fig. 1A). Sigmoidoscopy was performed and showed diffuse markedly edematous colonic mucosa covered with yellowish exudates along the rectum and sigmoid colon (Fig. 1B). Colonic biopsy was performed and showed acute inflammation and ulcerartion with Cytomegalovirus (CMV)-related cytopathic changes (cytomegaly, intranuclear and intracytoplasmic inclusion bodies, and peripheral margination of chromatin) involving stromal cells in granulation tissue (Fig. 1C and D). Specific immunohistochemistry staining with an anti-CMV antibody was positive on colonic biopsy. She was then started on intravenous ganciclovir. During the antiviral treatment, she spontaneously excreted small intestinal casts, a tubular-shaped infarcted bowel wall segment, per oral (Fig. 2A) and colonic casts per anus, measured 40–60 cm in length (Fig. 2B and C). Histological examination of the intestinal casts showed necrotic debris, fibrin and degenerated tissue (Fig. 3A and B). After the antiviral treatment, her conditions were partially improved. Unfortunately, she passed away 2 weeks later due to severe pneumonia with respiratory failure. The autopsy showed extensive CMV infection involving stomach, duodenum, small bowel, colon, lungs, adrenal glands, and lymph nodes.

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http://dx.doi.org/10.1016/j.idcr.2021.e01246

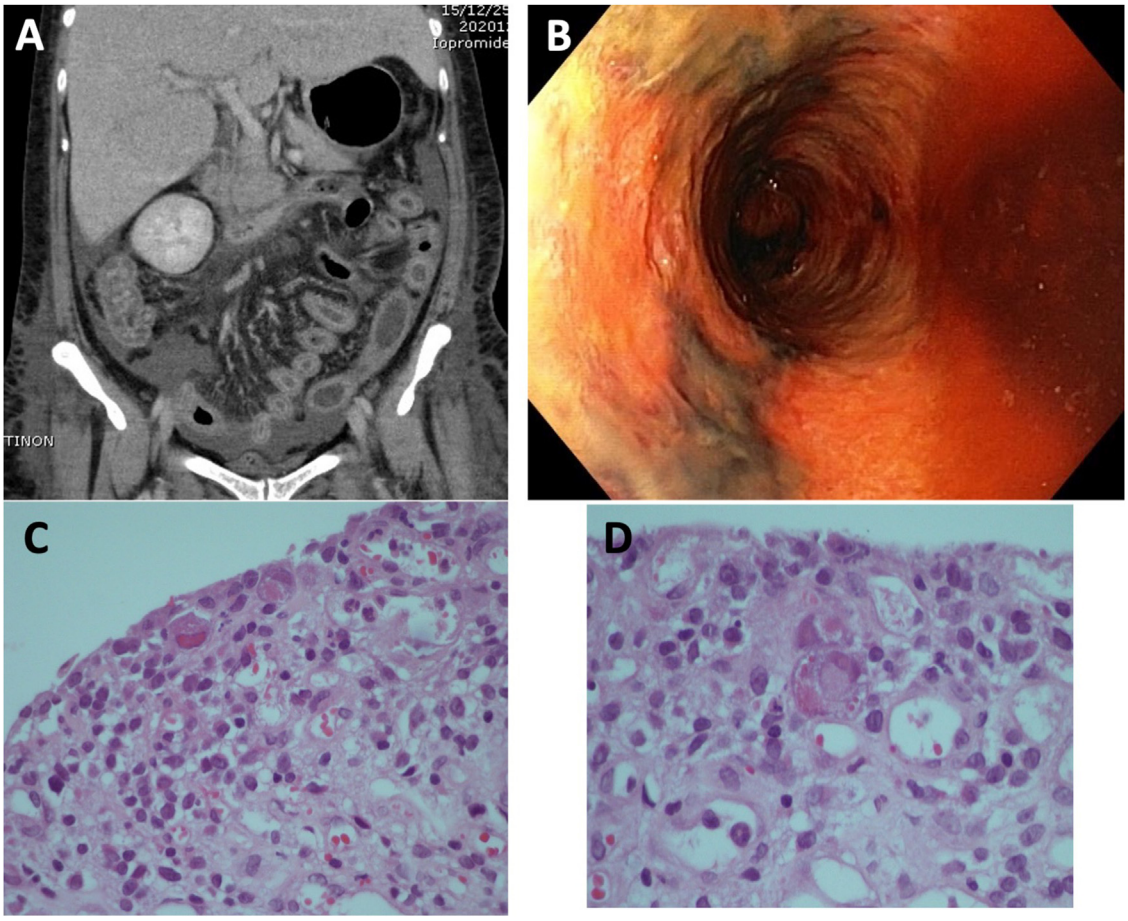

Fig. 1. Contrast-enhanced CT scan of the abdomen showed diffuse thickening of small and large bowel walls with surrounding peri-intestinal fat stranding (Fig. 1A). Sigmoidoscopy showed diffuse markedly edematous mucosa covered by yellowish exudates along the rectum and sigmoid colon (1B). Colonic biopsy, hematoxylin-eosin staining, showed acute inflammation and ulceration with CMV-related cytopathic changes (cytomegaly, intranuclear and intracytoplasmic inclusion bodies, and peripheral margination of chromatin) involving stromal cells in granulation tissue (Fig. 1C and D).

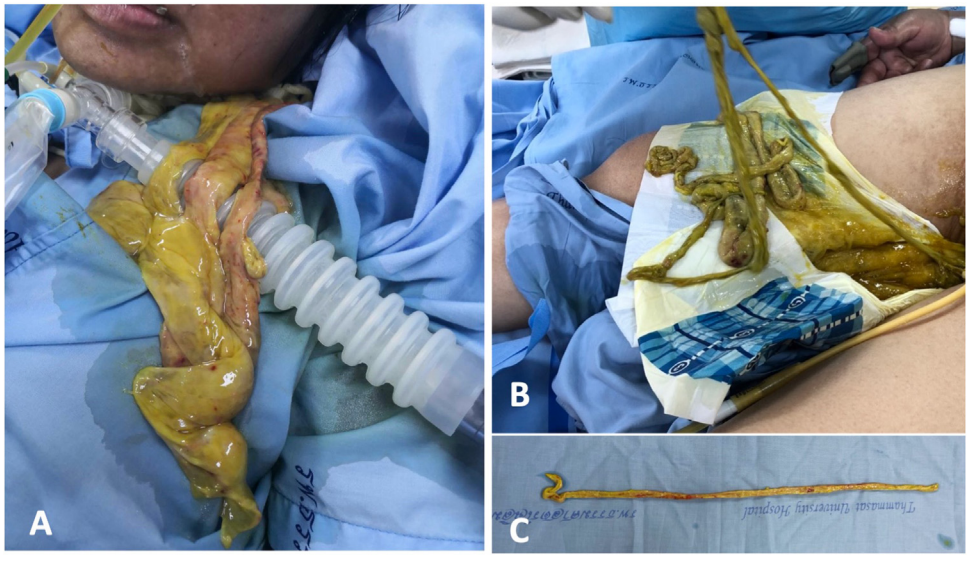

Fig. 2. The spontaneously-excreted small intestinal cast (Fig. 2A) and colonic cast (Fig. 2B and C).

Discussion

Intestinal cast, a spontaneously excreted, full-thickness, infarcted bowel segment, is an extremely rare condition. The majority of previously reported cases were spontaneous per anal passage of colonic cast and, more rarely, small intestinal cast [1–3]. A spontaneous excretion of small intestinal cast per oral has never been reported. The main pathogenesis of intestinal cast is

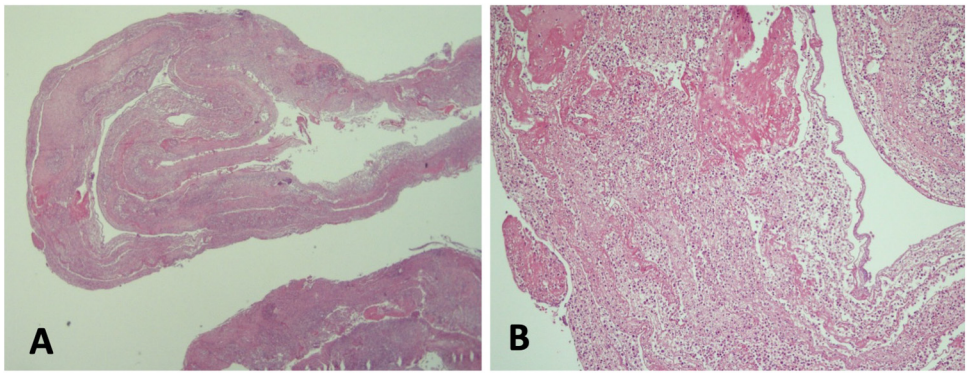

Fig. 3. Histological examination of the intestinal casts, hematoxylin-eosin staining, showed necrotic debris, fibrin and degenerated tissue (Fig. 3A and B).

attributed to extensive enterocolitis. Currently, there are only few etiologies reported: ischemic colitis [1,3,4], acute graft-versus-host disease [5], or in immunocompromised patients, such as neutropenic sepsis secondary to chemotherapy [2]. In addition, intestinal cast has been reported following major abdominal surgery, such as colorectal cancer surgery or abdominal aortic aneurysm repair with ligation of the inferior mesenteric artery or feeding vessels of the bowel [6]. It has been proposed that intestinal cast develop as a consequence of severely compromised bowel circulation leading to an excretion of totally necrotic colonic mucosa. Regarding the management, some patients responded well to conservative treatment; however, surgical intervention may be required in those with treatment failure [3].

To our knowledge, spontaneous passage of intestinal cast as a complication of extensive CMV-related enterocolitis has never been reported. Here, we describe a case of young woman who developed histologically-confirmed multiorgan CMV-related disease after receiving immunosuppressive treatment. In this patient, the development of small intestinal and colonic casts are likely caused by a total denudation of the mucosa from the deeper layers of the intestinal wall as a result of extensive mucosal inflammation. From the pathophysiological standpoint, the pathogenic organisms likely invade gut mucosa, and subsequently result in vascular endothelial cell injury and markedly compromised intestinal blood supply. Notably, several studies have previously reported cases of CMV-associated ischemic colitis in both immunocompromised and immunocompetent patients [7,8]. Another interesting finding in our patient is that spontaneous per oral excretion of small intestinal casts has never been reported. We propose that the presentation of this unique finding is likely explained by our patient had concomitant severe small intestinal ileus as she also presented with abdominal distension and vomiting, thus expelling the small intestinal casts by her mouth.

Learning objectives

We report a case of patient who developed small intestinal and colonic casts as a complication of extensive CMV enterocolitis following immunosuppressive therapy. Even though is finding is unusual, CMV infection should be included in the differential diagnosis of the passage of intestinal cast.

Declaration of Competing Interest

The authors report no declarations of interest.

Sources of funding

No funding.

Ethical approval

N/A.

Consent

Written inform consent was obtained from the patient's next-of-kin for publication of this case report and accompanying images.

Author contribution

All authors wrote the first draft of the manuscript.
Sith Siramolpiwat corrected the final draft of the manuscript.

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