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ABOUT COVER

Yosuke Tsuji, MD, PhD, Assistant Professor, Department of Gastroenterology, Graduate School of Medicine, The University of Tokyo, 7-3-1, Hongo, Bunkyo-ku, Tokyo 113-8655, Japan. ytsuji-tky@umin.ac.jp

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CASE REPORT

Vedolizumab in Crohn's disease with rectal fistulas and presacral abscess: A case report

Heng Yeh, Chia-Jung Kuo, Ren-Chin Wu, Chien-Ming Chen, Wen-Sy Tsai, Ming-Yao Su, Cheng-Tang Chiu, Puo-Hsien Le

ORCID number: Heng Yeh 0000-0002-3908-6927; Chia-Jung Kuo 0000-0003-2845-6248: Ren-Chin Wu 0000-0003-1439-0874; Chien-Ming Chen 0000-0001-7829-1504; Wen-Sy Tsai 0000-0001-6873-2863; Ming-Yao Su 0000-0002-5229-3074; Cheng-Tang Chiu 0000-0002-7347-8052; Puo-Hsien Le 0000-0002-1100-5371.

Author contributions: Yeh H wrote and revised the manuscript; Kuo CJ acquired the data; Wu RC interpreted the pathological result; Chen CM analyzed the radiological images; Tsai WS, Su MY and Chiu CT analyzed the data; Le PH decided the treatment plan, wrote and revised the manuscript.

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Heng Yeh, School of Medicine, Chang Gung University, Taoyuan City 333323, Taiwan

Chia-Jung Kuo, Cheng-Tang Chiu, Puo-Hsien Le, Department of Gastroenterology and Hepatology, Chang Gung Memorial Hospital, Linkou Branch, Taoyuan 333, Taiwan

Chia-Jung Kuo, Ming-Yao Su, Cheng-Tang Chiu, Puo-Hsien Le, Taiwan Association of the Study of Small Intestine Disease, Taoyuan 333, Taiwan

Ren-Chin Wu, Department of Pathology, Chang Gung Memorial Hospital, Linkou Branch, Taoyuan 333, Taiwan

Chien-Ming Chen, Department of Medical Imaging and Intervention, Chang Gung Memorial Hospital, Linkou Branch, Taoyuan 333, Taiwan

Wen-Sy Tsai, Department of Colon and Rectal Surgery, Chang Gung Memorial Hospital, Linkou Branch, Taoyuan 333, Taiwan

Ming-Yao Su, Department of Gastroenterology and Hepatology, New Taipei City Municipal Tucheng Hospital, New Taipei City 236017, Taiwan

Puo-Hsien Le, Liver Research Center, Chang Gung Memorial Hospital, Linkou Branch, Taoyuan 333, Taiwan

Corresponding author: Puo-Hsien Le, MD, Assistant Professor, Department of Gastroenterology and Hepatology, Chang Gung Memorial Hospital, Linkou Branch, No. 5 Fuxing Street, Guishan District, Taoyuan 333, Taiwan. puohsien@gmail.com

Abstract

BACKGROUND

Fistula and intraabdominal abscess are common complications of Crohn's disease (CD), but complex rectal fistula with abscess formation is rare. Tumor necrosis factor antagonists combined with percutaneous drainage or surgical intervention is optimal treatment for fistulizing CD with intraabdominal abscess. There is no study showing the efficacy of vedolizumab in such complicated condition.

CASE SUMMARY

A 47-year-old man has decompensated liver cirrhosis, Child B. He suffered from abdominal pain, bloody diarrhea, fever, and body weight loss. CD with rectoprostatic fistula, rectopresacral fistula, presacral abscess and cyto-

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megalovirus (CMV) infection were noted. He received antibiotics, anti-viral therapy, transverse colostomy and vedolizumab treatment. Six months later, he had deep remission and complete fistula tracts closure.

CONCLUSION

Early vedolizumab and stool diversion are effective and safe in treating CD with complex rectal fistula with abscess formation.

Key Words: Vedolizumab; Crohn's disease; Rectoprostatic fistula; Rectal presacral fistula; Presacral abscess; Case report

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Core Tip: Fistulas and intraabdominal abscess are common complications of Crohn's disease (CD) and tumor necrosis factor antagonists combined with percutaneous drainage or surgical intervention is the optimal treatment for fistulizing CD with intraabdominal abscess. However, no previous study has reported the efficacy of vedolizumab in this complicated situation. This 47-year-old male presented with CD with complex fistulas and presacral abscess. He received vedolizumab with transverse colostomy and the follow-up sigmoidoscopy 6 mo later showed mucosal healing without any visible fistula tracts. We think early vedolizumab treatment with stool diversion are effective and safe in treating CD with complex fistulas and abscess formation.

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INTRODUCTION

Fistula affects up to 50% Crohn's disease (CD) patients within 20 years of initial diagnosis[1,2]. It includes perianal, rectovaginal, enterocutaneous and internal fistula, but no rectoprostatic fistula or rectopresacral fistula were reported. The treatment of fistula usually requires a combination of medical and surgical approach[3]. As far as medical treatment is concerned, tumor necrosis factor (TNF) antagonists are the most effective to treat fistulizing CD^[4]. However, some studies also noted the beneficial effect of vedolizumab for fistulizing CD[5,6]. We presented the patient of CD, complicated with rectoprostatic fistula, rectopresacral fistula and presacral abscess. He had complete fistula closure and deep remission after transverse colostomy and vedolizumab treatment for six months.

CASE PRESENTATION

Chief complaints

Low abdominal pain and intermittent bloody stool for 6 mo.

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History of present illness

A 47-year-old man has decompensated liver cirrhosis, Child B, hepatitis C virus (HCV) and alcoholism related, complicated with hypoalbuminemia, hyperbilirubinemia, coagulopathy, thrombocytopenia, splenomegaly and esophageal varices, Form 1. He complained low abdominal pain and intermittent bloody stool for 6 mo. Appendicitis was diagnosed in local hospital, and he received appendectomy on 16th November 2019. However, he suffered from progressive low abdominal pain, bloody stool, dizziness, and intermittent fever up to 38 °C for four days. He also mentioned body weight loss 20 kg within one year. He was brought to our emergent department.

History of past illness

Decompensated liver cirrhosis, Child B, hepatitis C virus infection and alcoholism.

Personal and family history

Appendicitis was diagnosed in local hospital, and he received appendectomy on 16th November 2019.

Laboratory examinations

Lab data revealed hemoglobin 10.1 g/dL, platelet 66000/μL, white blood cell 7700/μL, segment 68.7%, lymphocyte 23.7%, international normalized ratio (INR) 1.5, aspartate aminotransferase (AST) 55 U/L, alanine aminotransferase (ALT) 95 U/L, total bilirubin 2.1 mg/dL, albumin 2.38 g/dL and CRP 22.81 mg/L. Cytomegalovirus (CMV) immunoglobulin (Ig) M, CMV DNA, Epstein-Barr virus (EBV)-VCA IgM, EBV DNA, human immunodeficiency virus (HIV) antibody (Ab), amebic Ab, Clostridium difficile toxin, culture for Salmonella, Shigella and Campylobacter were all negative. Positive CMV IgG, EBV-VCA IgG, stool pus cell and occult blood were noted.

Imaging examinations

Colonoscopy showed terminal ileal shallow ulcer (Figure 1A) and multiple complex rectal fistula tracts (Figure 1B and C) on 10th December 2019. Magnetic resonance imaging (MRI) noted decompensated liver cirrhosis with ascites (Figure 2A), rectoprostatic fistula (Figure 2B), rectopresacral fistula (Figure 2C) and presacral abscess (Figure 2D) on 21th December 2019. Pathology revealed acute on chronic inflammation with granulation tissue, compactable with CD (Figure 3A). Besides, there was positive result of CMV immunohistochemistry (IHC) staining (Figure 3B), which was performed with a monoclonal antibody directed against the CMV pp65 antigen (NovocastraTM lyophilized mouse monoclonal antibody; Leica Microsystems, Wetzlar, Germany).

FINAL DIAGNOSIS

He was diagnosed to have CD with CMV infection based on endoscopy and pathological findings. The CD activity index (CDAI) was 526 points and Harvey-Bradshaw index (HBI) was 22 points. He also had rectoprostatic fistula, rectopresacral fistula and presacral abscess diagnosed by MRI findings.

TREATMENT

He refused percutaneous abscess fine needle aspiration, and we kept Tazocin for abscess treatment for 27 d. Because of positive CMV IHC staining result, he also received intravenous ganciclovir for 17 d and then valganciclovir oral treatment for two months. Transverse colostomy was performed for stool diversion on 25th December 2019. He couldn't tolerant azathioprine due to pancytopenia and vedolizumab (300 mg) was prescript since 22th January 2020.

OUTCOME AND FOLLOW-UP

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Follow-up sigmoidoscopy showed mucosal healing without any fistula tract (Figure 1D) on 9th July 2020. The pathologist reported minimal inflammatory activity (Figure 3C). Lower gastrointestinal series mentioned no more fistula tract (Figure 4) on 21th July 2020. There was no more rectoprostatic or rectopresacral fistula (Figure 5A) and presacral abscess (Figure 5B) in MRI. After vedolizumab treatment for 6 mo, the CDAI was 42 points and HBI score was 0 point. His body weight also increased 20 kg, back to the same level before the episode.

DISCUSSION

Fistulizing CD results in not only high morbidity but also impairs health-related quality of life^[4]. Biologics combined with surgical intervention seems to be the best

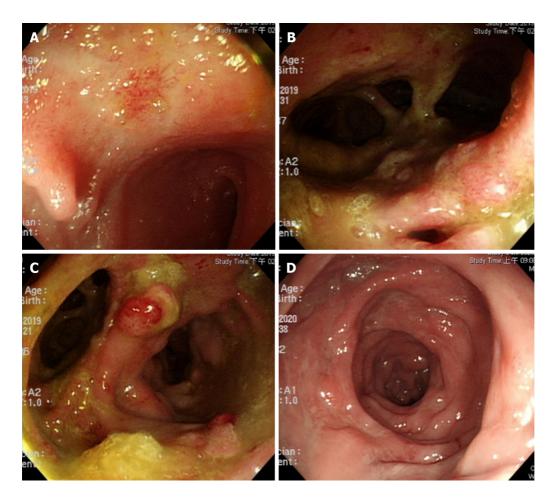


Figure 1 Endoscopic findings. A: Terminal ileal shallow ulcer at diagnosis; B and C: Multiple rectal fistula tracts with inflammation; D: Mucosal healing without fistula tracts six months after vedolizumab treatment, seven months after diagnosis.

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solution. Although Infliximab has the strongest evidence in fistulizing CD treatment^[7,8], vedolizumab also showed its efficacy in some studies^[5,6]. However, vedolizumab has better safety profiles (less severe adverse events and infections) in real world studies[9,10].

Intra-abdominal abscess occurs in up to 20% of patients with CD[11,12]. Adequate percutaneous drainage combined with early adalimumab treatment achieves up to 74% successful rate^[13]. In this case, it was difficult to drain the presacral abscess and patient refused, too. Therefore, we chose vedolizumab with transverse colostomy in treating the complex rectal fistula and presacral abscess without abscess drainage.

This patient received vedolizumab treatment one month after the diagnosis. Earlier initiation of biological treatment shortly after diagnosis (less than one year) in patients with moderately to severely active CD improved the long-term clinical outcomes^[14]. Besides, early stool diversion with transverse colostomy and early anti-viral treatment for CMV infection were crucial to achieve the good outcome in this case.

CONCLUSION

Vedolizumab with loop transverse colostomy was effective in treating CD with complex rectal fistulas with presacral abscess. Besides, early biological and anti-CMV treatments also lead to the favorable outcome.

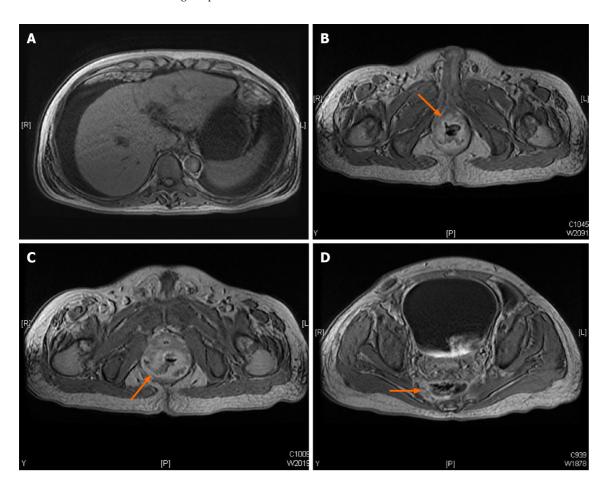


Figure 2 Magnetic resonance imaging at diagnosis. A: Liver cirrhosis with ascites; B: Rectoprostatic fistula; C: Rectopresacral fistula; D: Presacral abscess.

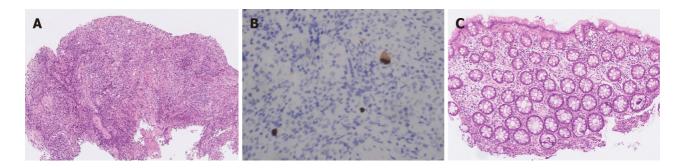


Figure 3 Pathology. A: Ulcer with acute on chronic inflammation and granulation tissue at diagnosis; B: Pathological presentations of cytomegalovirus (CMV) infection, immunohistochemistry stain (20 × objective) was performed with 1:200 diluted Novocastra™ lyophilized mouse monoclonal antibody against CMV pp65 antigen and showed strong focal CMV immunoreactivity with brownish areas; C: Minimal inflammatory cells infiltration six months after vedolizumab treatment, seven months after diagnosis.



Figure 4 Lower gastrointestinal series showed no more rectal fistula tract.

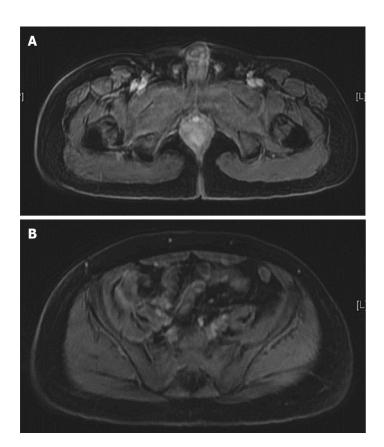


Figure 5 Magnetic resonance imaging seven months after diagnosis. A: No more rectal fistula tract; B: No more presacral abscess.

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