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Case Report

A case of eosinophilic pancreatitis in a patient with ulcerative colitis

Koji Toyoda[1], Yasumi Katayama[1,2,*], Ikuhiro Kobori[1], Akihiro Kitahama[1], Yumi Kusano[1], Yuko Ono[3], Shinichi Ban[3] and Masaya Tamano[1]

[1]Department of Gastroenterology, Dokkyo Medical University Saitama Medical Center, Koshigaya City 343-8555, Japan
[2]Endoscopy Center, Dokkyo Medical University Saitama Medical Center, Koshigaya City 343-8555, Japan
[3]Department of Pathology, Dokkyo Medical University Saitama Medical Center, Koshigaya City 343-8555, Japan

*Corresponding author. Department of Gastroenterology and Endoscopy Center, Dokkyo Medical University Saitama Medical Center, 2-1-50 Minamikoshigaya, Koshigaya City, Saitama Prefecture 343-8555, Japan. Tel: +81-48-965-1111; Fax: +81-48-965-1169; E-mail: yasumi@dokkyomed.ac.jp

Abstract

Eosinophilic pancreatitis (EP) is very rare and characterised by infiltration of eosinophils into the pancreatic parenchyma. A 40-year-old man was diagnosed with total-colitis-type ulcerative colitis at the age of 15 years. He was then diagnosed with steroid-dependent ulcerative colitis. He was given golimumab, which resulted in remission. Ten months after beginning golimumab, he was urgently hospitalised with a diagnosis of acute pancreatitis. Hence, endoscopic ultrasound-guided fine needle biopsy was performed to obtain a definitive diagnosis. Pathologically, abundant infiltration of eosinophils was observed in the edematous intralobular stroma of the pancreas. He was diagnosed with EP, and treated with corticosteroids.

INTRODUCTION

The various complications of ulcerative colitis include arthritis and skin symptoms, and, in rare cases, can include eosinophilic disorders. Previous studies have reported eosinophilic esophagitis and myocarditis as complications of ulcerative colitis [1, 2]. Eosinophilic pancreatitis (EP) is extremely rare among eosinophilic diseases, save for one report as a complication of ulcerative colitis [3]. EP is characterised by infiltration of eosinophils into the pancreatic parenchyma. It is often difficult to distinguish from pancreatic cancer and is generally diagnosed after surgery. We report a very rare case of EP that was diagnosed by endoscopic ultrasound-guided fine needle biopsy (EUS-FNB) in a patient with ulcerative colitis.

CASE REPORT

A 40-year-old man was diagnosed with moderate pan-ulcerative colitis at the age of 15 years. Treatment with mesalazine 3600 mg resulted in remission; however, he was referred to our hospital following a gradual worsening of both his symptoms and endoscopic findings. At the first visit, he described tenderness in the lower abdomen and reported 10 episodes of bloody diarrhea daily. Laboratory investigations showed an increased white blood cell count of 11 400 mm^3, C-reactive protein level of 10.77 mg/dl, and anemia with a hemoglobin level of 10.4 g/dL. Colonoscopy revealed pancolonic inflammation with an endoscopic Mayo score of 2.

His symptoms improved with prednisolone 30-mg and mesalazine 4800-mg treatment. Steroids were gradually tapered over 2 months; however, he experienced symptom recurrence and eventually became steroid-dependent. No azathioprine/mercaptopurine was used for this condition. Thirteen weeks after the first visit, the patient was given golimumab 200 mg as an initial induction dose, which resulted in remission. Ten months after beginning golimumab, he had a fever of 39.8°C and severe epigastric pain. Laboratory examination revealed elevated pancreatic enzymes (p-amylase 1324 U/L, lipase 1318 U/L), increased white blood cell count of 14,400 mm^3 with 25.4% eosinophils and normal IgE (166 IU), and a C-reactive protein level of 11.32 mg/dl. His ulcerative colitis condition was in remission. Abdominal computed tomography (CT) showed diffuse swelling of the pancreas (Fig. 1A). He was urgently hospitalised, confirming the diagnosis of acute pancreatitis. He had no history of alcohol use, hypertriglyceridemia, hypercalcemia and gallstone disease. As IgG4 and antinuclear antibody were normal, type 2 autoimmune pancreatitis (AIP) was suspected. Magnetic resonance imaging was contraindicated due to a tattoo. Hence, EUS-FNB using a 22-gage FNA needle (EZ Shot 3 Plus; Olympus, Tokyo, Japan) was performed with three punctures to obtain a definitive diagnosis. There were no abnormal findings in the gastric mucosa. EUS revealed generalised swelling of the pancreas and coarse parenchymal texture. The main pancreatic duct was not dilated (Fig. 2). Pathologically, abundant infiltration of eosinophils was observed in the edematous intralobular stroma of the pancreas. Plasma cells were inconspicuous, IgG-positive cells were scarce and IgG4-positive cells were not found (Fig. 3). No neutrophil infiltration or granulocyte epithelial lesions peculiar to type 2 AIP were observed. Based on these findings, the patient was diagnosed with EP. He was treated with 30 mg of corticosteroids,

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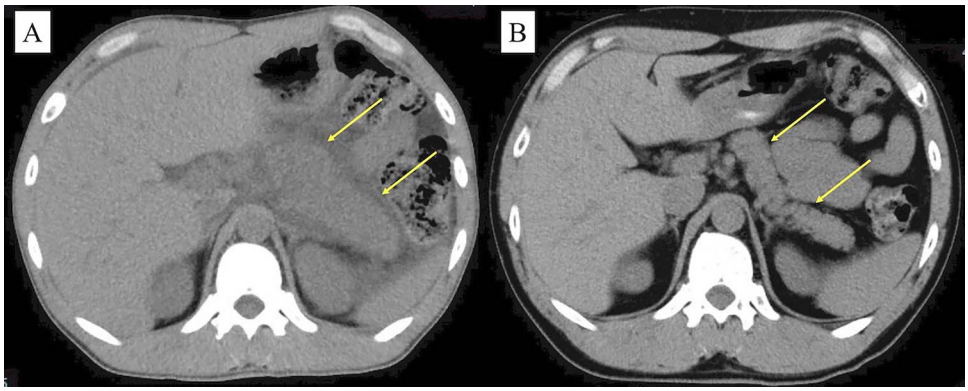

Figure 1. (**A**) Abdominal CT obtained at admission shows diffuse swelling of the pancreas (arrow). (**B**) Abdominal CT shows improvement of the diffuse swelling of the pancreas after corticosteroids therapy (arrow).

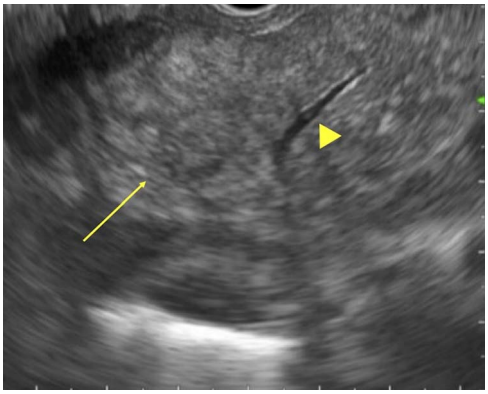

Figure 2. Endoscopic ultrasonography shows diffuse swelling of the pancreas and coarse parenchymal texture (arrow). There is no dilation of the main pancreatic duct. (arrowhead).

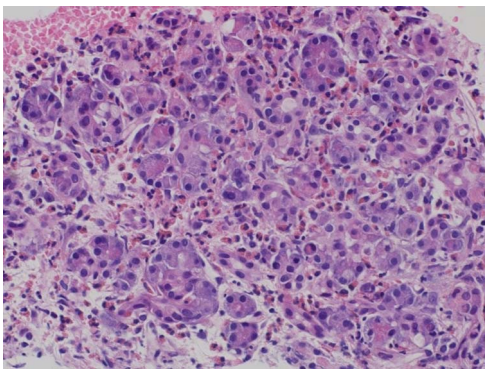

Figure 3. Marked infiltration of eosinophils is seen in the edematous intralobular stroma of the pancreas (hematoxylin and eosin, 40×).

which improved his symptoms and the CT findings (Fig. 1B). His corticosteroid dose was gradually reduced. He continued with golimumab treatment, with no recurrence of EP or ulcerative colitis for 2 years.

DISCUSSION

EP is a rare disease that often takes the form of chronic pancreatitis but can also present as acute pancreatitis [4]. Furthermore, to our knowledge, there has only been one other case describing EP in the setting of ulcerative colitis [3]. In EP, eosinophils infiltrate the pancreatic tissue, which, in many cases, increases eosinophils in peripheral blood. In the present case, eosinophils were 25.4% of white blood cells. The involvement of eosinophilic gastroenteritis and allergic diseases have been suggested as potential causes of EP, although these findings were not observed in our case. In addition, similar to IgG in AIP, IgE is important for the development of EP [5]. However, the IgE level was normal in our patient. A pancreatic cancer-like mass forms in many cases of EP [6], and the diagnosis is generally confirmed by pathological examination after surgery. In the present case, there was diffuse swelling of the pancreas similar to AIP and no tumor formation was observed. There are rare reports of EP that exhibit diffuse swelling as well as mass formation [7]. To avoid excessive invasiveness, EUS-FNB should be actively considered for diagnosis.

There are several previous reports of elevated eosinophils in peripheral blood inpatients using anti-tumor necrosis factor alpha (TNF-α) agents. The present patient was also receiving golimumab, which is an anti-TNF-α agent, and developed EP about 10 months after commencing golimumab therapy. However, as our patient improved without stopping golimumab, the role of the anti-TNF-α agent in the development of EP is unclear. In this case, continued anti-TNF-α agent had no effect on the pancreas. Anti-TNF-α agent and EP do not seem to be related. In addition, no study has reported the involvement of anti-TNF-α agent in the development of EP. Interestingly, there is one reported case of drug-induced pancreatitis with eosinophilia in a patient treated with vedolizumab [8].

In summary, we report a rare case of EP complicated with ulcerative colitis. As a large-scale research on this condition is difficult, it is very important to continue to accumulate reports of such cases.

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CONFLICT OF INTEREST STATEMENT

None declared.

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Not applicable.

CONSENT

The patients provided written, informed consent for the publication of this report.

GUARANTOR

Yasumi Katayama MD, PhD.

PRIOR PRESENTATION

None.

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