Case study

Recurrent acute pancreatitis due to eosinophilic gastroenteritis

Abstract

Aims: Eosinophilic gastroenteritis (EGE) is a rare disease characterized by focal or diffuse eosinophilic infiltration of the gastrointestinal tract, especially the stomach and duodenum. Clinical presentation is variable.

Presentation of case: We report a patient with EGE presenting with concurrent acute pancreatitis and ascites.. Laboratory findings revealed elevated serum titers lipase, and peripheral blood eosinophil count. An abdominopelvic computed tomography scan showed a normal pancreas, moderate amount of ascites, and duodenal thickening. Biopsies from the duodenum indicated eosinophilic infiltration in the lamina propria. The patient was successfully treated with prednisolone.

Conclusion: Despite its unusual occurrence, EGE may be considered in the differential diagnosis of unexplained acute pancreatitis, especially in a patient with duodenal edema on imaging or peripheral eosinophilia.

Key words:

Eosinophilic gastroenteritis, acute pancreatitis, ascites, eosinophilic infiltration

1. Introduction:

Eosinophilic gastroenteritis (EGE) is a rare disease characterized by eosinophilic infiltration of the gastrointestinal (GI) tract. Eosinophilic cells can accumulate in any part of the GI tract from the esophagus to the rectum; the most commonly affected are the stomach and the duodenum [1,2]. According to the classification by Klein, three different disease patterns of EGE can be discerned: mucosal, submucosal and serosal, although more than one layer may be involved [1]. The clinical manifestations depend on which wall layer and which bowel segments are predominantly involved.

Eosinophilic infiltration of GI tract may be one of the etiologies in the case of recurrent pancreatitis although EGE presenting as pancreatitis is extremely rare. This paper reports a case of EGE causing recurrent acute pancreatitis with concurrent ascitis, and we discuss the clinical characteristics and differential diagnosis.

2. Presentation of case:

A 52-year-old woman was admitted with abdominal pain, nausea and vomiting. There was no history of drug allergy, allergic rhinitis or hepatitis except for diabetis. She had been taking Metformin and sitagliptine for 10 years and had not taken a new medicine recently. She was not consuming alcohol. 4 years ago, the patient was admitted to hospital with acute epigastric pain and vomiting. The diagnosis of acute pancreatitis was made in view of the clinical signs and the elevation of blood lipasemia. Abdominal CT scan showed dilated small bowel loops with normal pancreas. On follow-up, there was no intestinal obstruction, but the intestinal wall was considered edematous. ERCP showed edema and compression of the descending

duodenum and a juxtapapillary diverticulum, but no pathology in the biliary or the pancreatic ducts.

Since this episode, she had had repeated episodes of self limiting crampy abdominal pain and alimentary vomiting.

Currently, physical examination was unremarkable except for mild epigastric tenderness. Laboratory investigations indicated a white blood cell count of 7800/mm3 with high percentage of eosinophils (19.6%). The serum biochemistry showed an aspartate aminotransferase of 48 IU/L, alanine aminotransferase of 30 IU/L, lipase of 678.71 U/L, and triglycerides of 69 mg/dL. Serum immunoglobulin E (IgE) and immunoglobulin G4 (IgG4) levels were in the normal range. Antinuclear antibody was negative. Abdominopelvic computed tomography (CT) was performed under the diagnosis of acute pancreatitis. It revealed diffuse wall thickening in the duodenum at the 2nd and 3rd portions, mild edema of small bowel loops and a moderate amount of ascites. However, there was no evidence of gallstones, abnormality of the pancreas or peripancreatic fluid collection. We diagnosed her as Balthazar grade A of acute pancreatitis based on the elevated lipase and normal pancreas on CT. Esophagogastroduodenoscopy (EGD) was performed to examine the duodenal wall thickening found on CT. EGD findings showed mild erythematous mucosal change but the duodenum was unremarkable. Bowel rest and supportive care including intravenous fluids and acid-reducing agents were initiated. The level lipase decreased to the normal range on the following day and her abdominal pain gradually decreased within a week. However, mild epigastric pain persisted and subsequent laboratory examination on the 7th hospital day showed that blood eosinophils had increased to 35, 8% and the eosinophil count was 3.5×103/μL (normal, 0.04–0.5×103/μL). Stool examination for parasites was negative. Allergen skin prick test was also negative. At that time, we thought of the possibility of EGE because her GI symptoms continued even after treatment and the eosinophilia was more aggravated without any other obvious cause. So we decided to perform EGD and colonoscopy again to take a biopsy from the GI tract. The 2nd look-EGD revealed aggravated mucosal erythema of the antrum and on the 2nd portion of the duodenum. Colonoscopy revealed erythematous mucosal changes of the terminal ileum. Multiple biopsies were obtained from the normal mucosa to the erythematous lesions on the stomach, duodenum and ileum. Histological examination indicated chronic inflammation with increased eosinophils in the lamina propria from the duodenal erythematous lesion (Fig. 1). The patient was treated with 40 mg of prednisolone for 1 month. Her symptoms improved immediately and the eosinophil count normalized within 2 weeks. Prednisolone was tapered over 8 weeks. The follow-up EGD after 12 months showed normal antral and duodenal mucosa with the eosinophil count normalized.

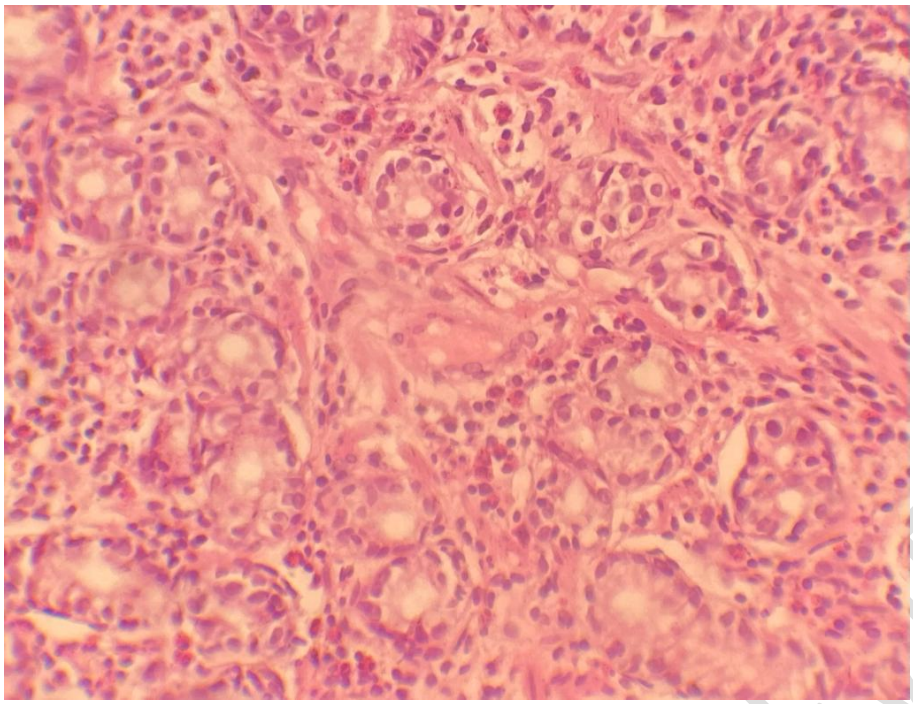

Figure 1: Histopathology of duodenal mucosa showing dense eosinophilic infiltrate of the lamina propria. (Hematoxylin-eosin stain. Magnification x10)

3. Discussion:

EGE is a rare disease of unknown etiology and is defined as a GI disorder of undetermined cause characterized by infiltration of eosinophils in the GI tract [2]. EGE presenting with acute pancreatitis was first reported in 1973 [3]. Thereafter, several papers on EGE associated with pancreatitis have been reported [4] .The diagnosis of EGE may be difficult and requires a high index of suspicion since EGE has a wide spectrum of clinical presentations. The patient in the current case presented with abdominal pain, nausea, and elevated levels of lipase. She also had peripheral blood eosinophilia at presentation. However, it was overlooked carelessly and she was initially diagnosed as having acute pancreatitis. EGE should be considered in any patient with GI symptoms associated with peripheral eosinophilia because it is associated with peripheral blood eosinophilia in nearly 30% to 80% of the cases [1]. The definite diagnosis of EGE is established by demonstrating eosinophilic infiltration on endoscopic, laparoscopic or laparotomic biopsies. Endoscopic findings in EGE may vary from normal mucosa to mild erythema, thickened mucosal folds, nodularity, and frank ulceration [5]. This patient was finally diagnosed with EGE according to the pathologic findings from the duodenal erythematous mucosal lesion. Establishing the diagnosis also requires exclusion of other causes of eosinophilic gut infiltration, such as food allergy, drug idiosyncrasy, parasitic/ helminthic infestation, connective tissue disease, vasculitis, malignancy, Crohn's disease, and non-tropical sprue, etc [2, 6]. Because autoimmune pancreatitis (AIP) is also associated with peripheral eosinophilia, with a prevalence of 28% [7], it should be differentiated from the current case. AIP can be diagnosed by typical CT findings such as diffuse enlargement with homogeneous attenuation and the peripheral rim of a hypoattenuation, and elevated serum IgG4 [8]. However, our patient exhibited a normal pancreas on CT and normal serum IgG4.

According to the classification by Klein, three different disease patterns of EGE can be discerned: mucosal, submucosal and serosal, although more than one layer may be involved. The less common form of EGE is the subserosal disease, defined by the presence of

eosinophilic infiltration of the gut and eosinophilic ascites [1]. The clinical manifestations depend on which wall layer and which bowel segments are predominantly involved. Thus, mucosal disease may result in anemia, protein-losing enteropathy, and malabsorption, whereas muscle layer infiltration typically causes luminal obstruction [1, 2, 9]. The rarest form is serosal disease with eosinophilic ascites [1, 2]. However, many of the clinical manifestations are non-specific, such as nausea, vomiting, crampy abdominal pain, and diarrhea, thus making a diagnosis may be difficult [1, 2]. We thought our case might be the subserosal type, although we could not identify eosinophilic ascites.

One of the important clues for the diagnosis of EGE in this patient was the duodenal wall thickening on CT. It is believed that eosinophilic infiltration can cause edema, fibrosis and distortion in the ampulla and periampullary duodenum, leading to pancreatitis [10], but there was no evidence of papillitis in this patient. Not only our case but also most reported cases of EGE presenting with acute pancreatitis exhibited duodenal edema or thickening of mucosal folds on imaging studies [10, 11]. Therefore EGE should be considered in patients with acute or recurrent idiopathic pancreatitis who have duodenal wall thickening on imaging study. During endoscopy in a patient with unexplained pancreatitis, the duodenum should be carefully examined and multiple biopsies should be taken from normal mucosa as well as from the abnormal mucosal lesion.

Steroids remain the mainstay of therapy in patients of EGE, although no controlled trials are available. Recently, some alternatives to steroids, such as sodium cromoglycate, montelukast, suplatast tosilate, and ketotifen, have been used [2]. We successfully treated this patient with steroids initially. the short and medium term development is satisfactory. there are no more abdominal pain attacks and the blood eosinophil level is normal.

4. Conclusion:

Eosinophilic gastroenteritis can be considered as causes of recurrent acute pancreatitis without obvious causes found. Edema and duodenal thickening caused by eosinophilic infiltration is the main pathogenesis of this entity. The treatment based on steroids remains very effective in the medium and long term.

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