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Case Report

Case Report of Chronic Pancreatitis in aPatient with Celiac Disease and Extra-Intestinal Manifestations of Celiac Disease: Report of a Case and a Literature Review

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Abstract

Celiac disease is characterized by both inflammation of the small intestine and villous atrophy. It is triggered by exposure to dietary gluten in genetically susceptible individuals. Celiac diseases frequently manifest themselves in a diverse variety of extra intestinal symptoms which are well-documented. There is now evidence of the association of pancreaticobiliary diseases in patients with celiac disease, and it has been reported that celiac disease is associated with an increased risk of pancreatitis and pancreatic insufficiency. In the last 30 years, celiac cases exhibiting extra-intestinal symptoms were not well understood and no clear association with pancreaticobiliary diseases had yet been established. A young female with chronic pancreatitis was referred to our clinic for endoscopic therapy. Her past medical history was of an acute recurrent pancreatitis of a couple of years' duration, and had been diagnosed with celiac disease. Endoscopic ultrasound revealed severe chronic pancreatitis, with total villous atrophy and flat-shaped duodenal mucosa. The patient was then treated with a combination of gluten-free diet and pancreatic enzymes, with this approach patient showed a dramatic improvement in her symptoms and soon started tolerating the GFD. The patient is free of abdominal pain and has had significant weight gain. In this brief review we reported this and present a literature review.

Introduction:

Celiac disease has been found to occur in genetically susceptible patients, and is closely associated with the genes that code HLA-II antigens, mainly the DQ2 and DQ8 classes. Patients usually present with common gastro-intestinal symptoms, including abdominal pain, diarrhea and weight loss. The disease is common in Europe and the United States, but only sporadic reports of it are found in East Asia, including China.

Celiac disease (CD) has been shown to be associated with a variety of extra-intestinal manifestations involving most organs and systems, and can include pancreatic diseases. A good number of new cases of celiac disease (CD) are diagnosed in adults, and particularly in patients with extra-intestinal symptoms.

Pancreatic abnormalities are uncommon extra-intestinal manifestations of celiac disease. The pancreatic diseases associated with celiac disease are autoimmune pancreatitis,

pancreatitis, pancreatitis and pancreatic insufficiency. In a good number of reported cases of pancreatic diseases associated with celiac disease, however, chronic pancreatitis is only reported in a few such cases. Patients with celiac disease with established chronic pancreatitis as an extra-intestinal symptom are often difficult to manage with standard gluten free diet.

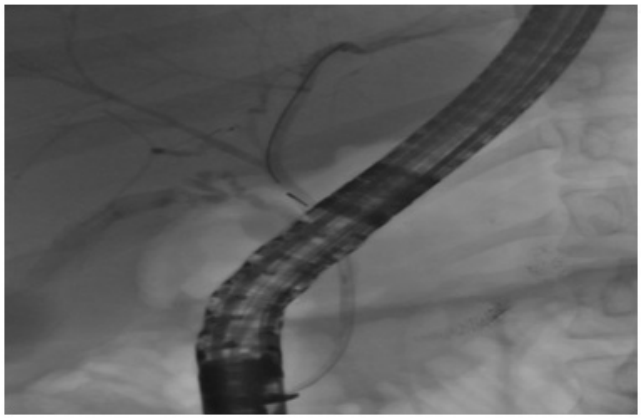

Figure 1: ERCP showed a normal biliary tree.

Case scenario:

A 15 year old female patient was referred as a case of severe chronic pancreatitis of several years' duration, with a history of chronic abdominal pain without any history of anorexia to our outpatient clinic of King Fahad Medical City, a tertiary care center in Riyadh, Saudi Arabia. The abdominal pain was diffused, was aggravated by fatty food and not radiating to the back. There was positive history of diarrhea and weight loss. She denied any history of trauma or any offending medications. In addition, there was no family history of acute recurrent or chronic pancreatitis. Over a period of two years she had lost more than 15 kilograms in weight despite having a normal appetite and the use of pancreatic enzymes. With these symptoms she had been managed in a local hospital, where imaging, including a CT scan had been performed, which had revealed chronic calcified pancreatitis. On examination the patient was conscious, with a low body mass index of 16kg/M^2. She had stable vitals, her sclera were pale and abdominal examination revealed nothing remarkable. Other systemic examinations were all normal. On evaluation, the patient was shown to have Hemoglobin levels of 8.7gm/dL with low MCV (59) and KCH (16) and high platelet counts of 754. The vitamin D 25-OH was 30 nmol/L. A mild elevation of Amylase level of 150 and lipase level of 94 IgG and IgA and IgM were normal. Tests on renal, liver and blood sugar functions yielded normal results. After initial fluid resuscitation, computed tomography of the abdomen and MRI of the pancreas showed pancreatic atrophy with dilated side branches. Endoscopic ultrasound was then performed, which showed severe chronic pancreatitis based on the Rosemont criteria (hyperechoic foci lobularity, cystic lesions, dilated and irregular main pancreatic duct) but no mass lesion was seen [1]. The patient was taken for endoscopic examination during which duodenal mucosa appeared flat mucosa and scalloping shape. Multiple biopsies were taken and the pathology results showed total villous atrophy and intraepitheliallymphocytosis, with positive serology (AntigliadinIgG II was 124, Antigliadin IgA II was 135, Endomysial IgA was 287 and tissue transglutaminase IgA was 263). The patient was put on a gluten free diet and started on a recommended dose of pancreatic enzymes. With excellent nutritional support, the patient tolerated GFD well. On follow up she had shown a progressive increase in her weight and was symptom-free.

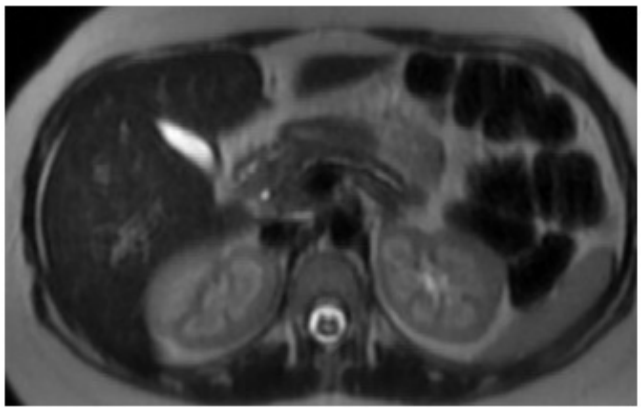

Figure 2: MRI: pancreas showed a feature of chronic pancreatitis.

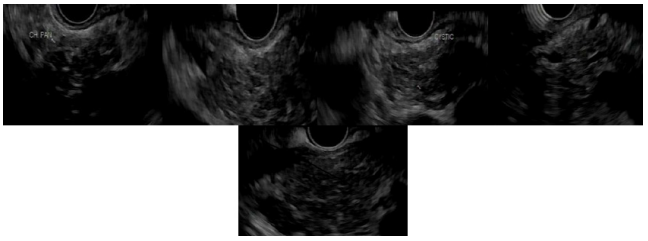

Figure 3: EUS images showed a feature of severe chronic pancreatitis.

Discussion:

Celiac disease (CD) is commonly encountered in our daily clinical practice. The recognition of the complex clinical picture of the disease and understanding the genetic, environmental, and immunologic components as a background to the pathophysiology of the disease helps the physician to diagnose celiac disease even if the gastrointestinal symptoms are lacking. The increase in incidence implies that a prevalence of 1.0% is possible [2]. A retrospective study of 779 patients with celiac disease observed that the clinical profile of celiac disease has changed over time, with an increased rate of non-classical symptoms, including extra-intestinal manifestations and subclinical indications [3]. The same study reported that the extra-intestinal manifestations included osteopenia/osteoporosis (52%), anemia (34%), cryptogenic hypertransaminasemia (29%) and recurrent miscarriages (12%) [4]. Different neurological symptoms associated with celiac disease including depression, ataxia and encephalopathy associated with celiac disease were also reported [5]. The available data suggest that screening for CD in patients with affective and/or psychiatric symptoms may be useful, since these disorders could be the expression

of an organic disease rather than primary psychiatric illnesses [6]. Ghozzi M et al described in their study of 211 patients with articular manifestation that five patients were positive of endomysial antibodies (EmA) (2.37%). Those five patients were respectively showing some gastro-intestinal symptoms and other extra-intestinal manifestations of celiac disease [7]. Liver abnormalities in a patient with celiac disease were also reported, the most frequent of which are primary sclerosing cholangitis (PSC) and primary biliary cirrhosis [8,9]. There are conflicting data supporting dental symptoms in celiac disease, however, the author suggested that CD should be screened for in a patient with systematic dental enamel defects, even in the absence of gastro-intestinal symptoms [10]. However, some of extra- intestinal manifestations are rare, including the pancreatic disorders. It has been demonstrated in a retrospective study of 143,746 patients in the Swedish Patient Register that patients with celiac disease have an almost 3-fold increase in risk of developing pancreatitis, compared with the general population [11,12]. Masoodi I et al reported the first case of auto-immune pancreatitis in a celiac patient [13]. The author suggested that celiac disease should be considered in the etiological work-up of patients with unexplained pancreatitis [14]. The pediatric literature has also reported similar cases [15,16]. In these studies, the index case had a diagnosis of chronic pancreatitis and later on celiac disease was confirmed. Despite a completed work-up search for another diagnosis of chronic pancreatitis, the result was negative. Fecal elastase 1-determination in celiac may be useful in detecting and curing exocrine pancreatic insufficiency, especially in patients with a new diagnosis of celiac disease or in those with the refractory diarrhea that was described in this review [17]. However, cases of celiac disease with extra-intestinal symptoms are always challenging to diagnose without high clinical indices. Historically there are few reported cases of pancreatic disorders in celiac disease, and one of the reported cases, the case of a young woman with chronic calcific pancreatitis in whom non-response to treatment was due to celiac disease, is described [18]. Later, many reports followed, describing similar cases with variety of pancreatic diseases [19-24] Patel RS et al described in a retrospective study of 169 patients with sphincter of odd dysfunction and recurrent pancreatitis that 12 of those patients were diagnosed with Celiac disease (7.1%). This study suggested that Celiac disease should be considered in the etiology of papillary stenosis or idiopathic recurrent pancreatitis [25]. One theory behind the association of pancreatitis in celiac patient is the presence of pancreatitis-associated protein (PAP), which leads the authors to observe that the active phase of the disease was accompanied by an increased serum concentration of PAP [26]. The disease is not restricted to or specific to certain age group, and a retrospective study of 7 patients showed that the celiac disease is under-diagnosed in elderly patients, and that it can be presented with a wide spectrum clinical picture including a common gastro-intestinal and extra-intestinal manifestations [27]. Health care providers should understand all clinical practice in the treatment of celiac disease, because this is a common condition with diverse manifestations that can be effectively diagnosed and easily treated for the prevention of both acute and long-term complications. Patients should follow a strictly gluten-free diet for life to prevent reoccurrence [28]. The extra-intestinal symptoms of associated illnesses may appear both at the time of diagnosis and throughout the evolution of the disease, and an adequate adherence to a GFD may slow down their evolution, especially if implemented during an early stage [29]. Overall, autoimmune diseases occur more frequently in those with celiac disease than in the general population. To conclude, the present case brings to the fore the need for full systemic examination of the symptoms of celiac disease and the effective management of the celiac to treat the extra-intestinal symptoms of the disease.

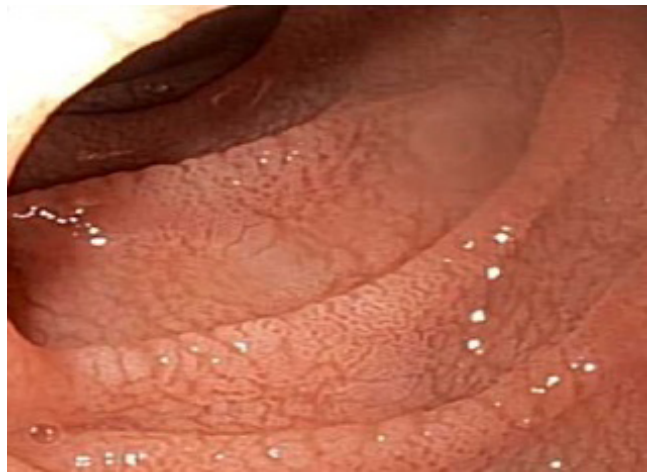

Figure 4: Upper Endoscopy showed a flat scalloping duodenal mucosa.

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