

Case Report

Non-alcoholic chronic calcific pancreatitis in a patient of celiac disease: a rare presentation

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ABSTRACT

Celiac disease is a common malabsorptive disorder in the Indian subcontinent and autoimmune dysfunction of thyroid and pancreas is frequently encountered along with. Chronic Calcific pancreatitis is a unique entity commonly seen in alcoholics but very rare in a patient of celiac disease. This case report includes the interesting constellation of calcific pancreatitis with celiac disease in a young adult male patient known case of insulin dependent diabetes and hypothyroidism. We believe it to be the only case report from north India. A 32 year old Indian male patient known case of Diabetes and hypothyroidism presented with features of malabsorption and was diagnosed with Celiac disease and calcific pancreatitis on imaging. The symptoms and insulin requirement also improved with the treatment of Celiac disease. Although a common involvement of pancreas in celiac disease, calcific pancreatitis is a rare finding and improvement of both the insulin requirement and malabsorptive symptoms with the treatment of celiac disease and pancreatitis vice-a-versa.

Keywords: Calcific pancreatitis, Celiac disease, Diabetes mellitus

BACKGROUND

Celiac disease (CD) or Gluten sensitive enteropathy is an immune-mediated enteropathy triggered by the ingestion of gluten in genetically susceptible individuals. It is one of the most common lifelong disorders on a worldwide basis. Celiac disease (CD) is characterized by both intestinal and extraintestinal manifestations. Chronic diarrhoea, abdominal distension, and anemia are frequent findings. It is quite frequent finding in North Indian patients with weight loss and chronic steatorrhea.¹ Chronic pancreatitis along with exocrine pancreatic insufficiency is also commonly seen in these patients.^{2,3} But advanced pancreatic parenchymal atrophy and calcification in a patient of celiac disease is a rare manifestation.

Exocrine insufficiency and diabetes have been seen uncommonly in patients with protein calorie malnutrition due to various causes.^{4,5} Pancreatic calcification is most commonly attributed to heavy alcohol intake and recurrent attacks of pancreatitis. It is also seen in patients of protein malnutrition in Indian subcontinent.⁶ H J Freeman and colleagues described one such finding of chronic calcific pancreatitis in celiac patient.⁷

The present case report describes a diabetic male with pancreatic exocrine insufficiency and calcification. Persisting weight loss and diarrhoea led to further investigations and, eventually, a diagnosis of celiac disease was made. In non-alcoholic patient pancreatic calcification may be the presenting feature of occult

celiac disease reflecting impaired absorption and longstanding severe malnutrition.

CASE REPORT

A 32 year old diabetic and hypothyroid male Indian patient on treatment from outpatient department was referred to emergency services for uncontrolled blood sugars and chronic diarrhoea and significant weight loss. He had a past history of hospitalisation for abdominal pain which was non pancreatic in origin. He had no past history of abdominal trauma, gall stones, jaundice or intake of cassava. No family history of diabetes mellitus, hypothyroidism or pancreatic disease.

On initial blood investigations his blood sugar values were very high on OHA and Insulin therapy but without any features of ketoacidosis or hyperglycaemic hyperosmolality. He also did not have any features of micro or macrovascular complications evaluated by nephrologist and ophthalmologist.

On examination he was emaciated with weight and BMI of 46 kg and 14.79 g/m² respectively. He did not have any pallor, clubbing or lymphedema and was nonicteric. Systemic examination was fairly normal with no features of chronic infection or any prolonged illness.

His investigation revealed mild anemia with Hemoglobin of 11 g/dl and raised MCV values of 104 nl. Transaminases were raised up to five times and serum alkaline phosphatase was 954 IU/l. Serum protein and albumin were low. Fecal Elastase levels were 88ug/g of stool.

He had severe hypovitaminosis D and also reduced Vitamin B12 levels. Surprisingly Iron profile was within normal limits despite being a case of celiac disease. Anti TTG levels were 32.3 U/ml. Titres of Anti TPO antibodies were also raised. Hepatitis virus serology, antinuclear and antimitochondrial antibodies were negative. Serum ferritin, ceruloplasmin, amylase and lipase Urinalysis and 24 h urine protein determination were also normal.

Initial chest and abdominal X-rays and CECT chest were essentially normal. CECT abdomen revealed Pancreatic head calcification and atrophy of rest of pancreas and non visualisation of main duct. There were also porto-systemic collaterals along the greater curvature of stomach and dilated portal vein and attenuated splenic vein (Figure 1).

Endoscopy showed normal visualisation of stomach, esophageal and duodenal mucosa. Multiple biopsies were taken from duodenal bulb and distal part whose examination revealed changes in mucosal villi and crypt hyperplasia consistent with celiac disease (Figure 2).

The patient was treated with a gluten-free diet and, pancreatic enzyme supplement capsules and Pantoprazole. The vitamin and micronutrient deficiencies were corrected with injectable supplements. Diabetes managed with basal and bolus insulin regimens. Patient showed marked improvement in symptoms and gained weight of 5 kg in subsequent 2 weeks.



Figure 1: Calcification of pancreatic head and body with atrophic ducts.

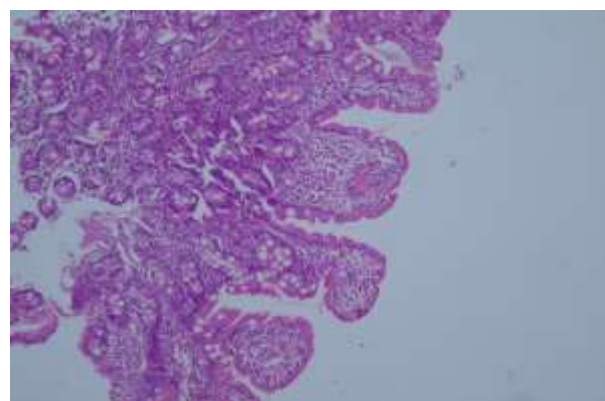


Figure 2: Duodenal biopsy showing typical mucosal abnormalities of celiac disease.

DISCUSSION

The patient discussed in this report presented initially with marked weight loss and steatorrhea and hypoalbuminemia. Celiac disease was found to be the culprit behind the symptoms along with Chronic pancreatitis and pancreatic calcification. The patient responded to pancreatic enzymes and Gluten free diet. For steatorrhea, it was believed that severe pancreatic insufficiency was responsible.⁸ This report further lays stress on the need to establish the cause for impaired nutrient absorption and weight loss in patients with steatorrhea. In celiac disease, it has long been known that occult pancreatic disease may lead to an incomplete clinical response despite a strict gluten-free diet.⁹

Pancreatic exocrine insufficiency in patients with Celiac disease may lead to steatorrhea through a number of possible mechanisms. Impaired pancreatic function may result in impaired digestion and absorption of critical nutrients along with malnutrition.⁹ Some studies have estimated that up to 20% or more of celiac patients have defective pancreatic function.¹⁰ Most frequent cause that is appreciated - impaired release of pancreatic stimulating hormones from diseased proximal small intestine because of mucosal endocrine cell loss.⁸⁻¹¹ Loss of enteric endocrine cells, including secretin cells in proximal small intestinal biopsies from seven celiac patients compared with five nonceliac controls have been demonstrated in Previous studies.¹² Controlled diet studies in celiac disease have suggested impaired cholecystokinin-pancreozymin secretion leading to a reduction in pancreatic exocrine cell stimulation and intraluminal dilution of pancreatic lipase rather than lipase deficiency contributes to steatorrhea.¹³ Secondly, amino acids deficiency may result, in part, from impaired small intestinal transport of amino acids leading to a diminution in the precursors available for pancreatic enzyme synthesis.¹⁰⁻¹⁴ Protein malnutrition and micronutrient deficiency per se may result in structural changes in the pancreas like acinar cell atrophy and pancreatic fibrosis.⁵⁻⁹⁻¹⁴

Chronic alcohol use is the leading cause of pancreatitis with calcification as well as of exocrine failure, the present report emphasizes that other causes, including celiac disease with severe malabsorption and protein deficiency should also be explored. Previous reports describe of a woman of Russian origin with celiac disease and chronic calcific pancreatitis but no information was provided on alcohol consumption.¹⁵ In a report from India tropical calcific pancreatitis was associated with celiac disease in a nonalcoholic patient.¹⁶

Thus in this scenario young male patient who did not consume alcohol and had no past or familial history of pancreatic disease; developed calcific pancreatitis, possibly due to his protein deficient state. Chronic steatorrhea and weight loss along with diabetes was the major presenting manifestation. Presence of unrecognized and, presumably, longstanding, severe celiac disease was the turning point in diagnosis making. As seen in this patient, even with calcific pancreatitis a poor clinical response to pancreatic enzyme supplements, should lead to re-evaluation and exclusion of a possible superimposed cause such as celiac disease for malabsorption and steatorrhea. Prospective studies, including detailed investigations on the effects of celiac disease on altered parenchymal structure and function are needed. We hereby suggest that celiac disease may have contributed to the development of calcification of the pancreas.

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