

Pericardial Effusion in Celiac Disease

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ABSTRACT

Celiac disease is an autoimmune disorder that affected 1% of all population in United State. Classic manifestations of disease consist of early childhood diarrhea, malabsorption, steatorrhea and growth retardation but disease can affects adult at any age. In adult anemia is a more frequent finding. This patient was a 40-year-old lady with progressive fatigue and lower extremities pitting edema. Iron deficiency anemia and celiac disease were diagnosed on the basis of low serum ferritin, elevated serum level of IgA endomysial and tissue transglutaminase anti-bodies and histologic findings in small bowel biopsies. Pericardial effusion in her evaluation was detected incidentally. Asymptomatic pericardial effusion in this patient was only detectable with imaging. After starting of gluten free diet and iron supplement fatigue, peripheral edema and pericardial effusion on echocardiography decreased. It should be noted that asymptomatic pericardial effusion may be seen in adults with celiac disease.

Keywords: Anemia, celiac disease, pericardial effusion

INTRODUCTION

Celiac disease is an autoimmune disorder triggered by ingestion of gluten. The estimated prevalence of disease is 1%.^[1] In most affected people celiac disease are not diagnosed.^[2] Classic manifestations of disease included early childhood onset of malabsorption, steatorrhea, weight loss and failure to thrive,^[3,4] but it can affect adult at any age without classic childhood manifestations.^[5-8] Anemia is a more frequent symptom at presentation of disease in adult,^[9] that is mainly due to iron deficiency, although nutritional factors deficiency and chronic disease are other causes of anemia.^[10,11]

CASE REPORT

The present case report is about a 40-year-old female patient presented in emergency room on October, 2012, with the complaining of severe fatigue and lower extremities edema beginning 3 months prior to admission. She had no history of frequent diarrhea, weight loss, abdominal pain and discomfort. Her menstrual cycles were normal. Her past medical history was positive for tension type

Case Report

headache and she has a history of acetaminophen use for control of headache. On physical examination, she was pale in appearance. The conjunctiva was pale. Chest, cardiac and abdominal examinations were normal. 3+ pitting edema of the lower limbs was detected. Levels of serum electrolytes, total protein, total and direct bilirubin and uric acid, tests of liver, renal and thyroid function and urine analysis were normal. Positive IgA endomysial (titer: 1/20) and tissue transglutaminase (titer: 140 U/L) antibodies were detected. Other laboratory results are shown in Table 1.

In upper gastro esophageal endoscopy duodenal mucosa appeared atrophic and had a nodular or scalloped appearance. Multiple biopsies were obtained from the duodenal bulb and the second portion of duodenum. In microscopic evaluation of biopsies more than 40 intraepithelial lymphocytes per 100 enterocytes, crypt hyperplasia and villous atrophy were seen that was compatible with celiac disease [Figure 1], (Grade IIIB, according to the Marsh classification).^[12]

In routine chest X-ray, globular enlargement of the heart, suggesting of pericardial effusion was seen [Figure 2]. Cardiac function in echocardiography was normal, but severe pericardial effusion without structural heart disease and right atrium collapse was seen [Figure 3a].

Pericardial effusion in this patient was asymptomatic and only detectable with imaging. In this patient after 2 weeks of starting of Iron supplement with gluten free diet, leg edema and pericardial effusion on echocardiography decreased and symptoms such as malaise and fatigue improved [Figure 3b].

DISCUSSION

The list of conditions associated with celiac disease is extensive. Cardiovascular disease is one of them. Celiac disease has been associated with an increased risk of ischemic heart disease^[13] atrial fibrillation 46,^[14] cardiovascular death,^[15] dilated cardiomyopathy^[16] and autoimmune myocarditis.^[17] Riccabona in a study reported that 50% of children with celiac disease had asymptomatic and limited pericardial effusion that only detectable with instrument.^[18] In these children, a higher value of antiendomysial antibodies and lower amount of iron and selenium were observed.^[18]

Table	1:	Laboratory	data
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Variable	Reference range	Results
Red blood cell (millions/µl)	4.5-5.9	3.34
Hematocrit (%)	41.5-50.5	28.3
Hemoglobin (g/dl)	14-17.5	6.9
MCV (fl)	80-100	73.8
MCH (pg)	27.5-33.2	18.6
MCHC (g/dl)	33-35.2	25.2
White-cell count (per mm ³)	4400-11000	4600
Differential count (%)		
Neutrophils	50-70	45
Lymphocytes	20-40	55
Serum iron (µg/dl)	40-120	38
Ca (mg/dl)	8.6-10.3	8.6
P (mg/dl)	2.6-4.5	3.3
Albumin (g/dl)	3.4-4.8	3.5
K (mEq/l)	3.8-5	3.5
TIBC (mg/dl)	250-450	330
Ferritin (ng/ml)	22-322	11
Retic count %	0.5-1.5	1.2
LDH (U/l)	100-480	478
IgA endomysialAb (IF)	<1/10	1/20
IgA tissue TG Ab (U/l)	<12	140
TSH (µU/l)	0.3-5	2.8
T4 (µg/dl)	4-12	10.1
ANA (Elisa)	< 0.8	0.25
PT (second)	12-14.7	14.7
PTT (second)	28-40	32
INR	1-1.2	1.2
Cholesterol (mg/dl)	60-200	84
LDL (mg/dl)	<130	49
HDL (mg/dl)	35-80	24
Triglyceride (mg/dl)	46-200	75

MCV=Mean corpuscular volume, MCH=Mean corpuscular hemoglobin, MCHC=Mean corpuscular hemoglobin concentration, TIBC=Total iron binding capacity, LDH=Lactate dehydrogenase, ANA=Antinuclear antibody, PTT=Partial thromboplastin time, PT=Prothrombin time, INR=International normalized ratio, LDL=Low density lipoprotein, HDL=High density lipoprotein, TG=Transglutaminase

Inourpatient diagnosis of celiac disease was based on serologic and histologic findings. Pericardial effusion in our patient was not symptomatic and it was an incidental finding. Anemia and leg edema were the major findings in the patient. Anemia work-up revealed iron deficiency anemia. Anemia is the most common presentation of celiac disease in adults and it may be the only presentation of disease and iron deficiency is the major cause of

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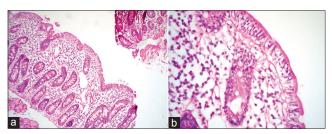


Figure 1: Histological sections show intraepithelial lymphocytosis, crypt hyperplasia and villous atrophy (AX100, BX400)

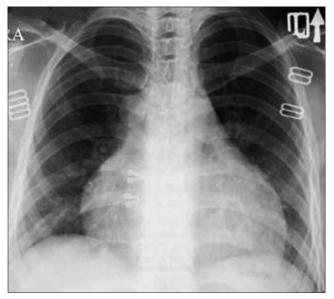


Figure 2: Globular enlargement of heart

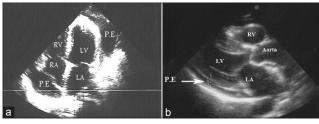


Figure 3: Views of 2 D-echocardiogram. (a) Showed severe pericardial effusion in posterior of right atrium and lateral wall before treatment. (b) Showed mild pericardial effusion in posterior wall after treatment. RA: Right atrium, RV: Right ventricle, LA: Left atrium, LV: Left ventricle, PE: Pericardial effusion

anemia.^[19] Interestingly celiac disease is a common cause of iron deficiency anemia.^[20,21]

In our patient cardiac, hepatic and renal function were normal and serum albumin level were normal. In the subject except severe anemia we couldn't find any other cause of leg pitting edema. Despite normal cardiac, liver and renal function, chronic severe anemia could induce peripheral edema that respond to correction of anemia.^[22]

Nutritional therapy is the only acceptable treatment of celiac disease and consist of a gluten free diet and nutritional supplements such as iron, calcium and vitamins.^[23] Pericardial effusion in this case report reminded the unusual and extra intestinal manifestations associated with celiac disease. It is usually asymptomatic and responds to treatment with gluten free diet and doesn't require invasive evaluation.

REFERENCES

- 1. Dubé C, Rostom A, Sy R, Cranney A, Saloojee N, Garritty C, *et al.* The prevalence of celiac disease in average-risk and at-risk Western European populations: A systematic review. Gastroenterology 2005;128 4 Suppl 1:S57-67.
- 2. West J, Logan RF, Hill PG, Lloyd A, Lewis S, Hubbard R, *et al.* Seroprevalence, correlates, and characteristics of undetected coeliac disease in England. Gut 2003;52:960-5.
- Andersen DH. Celiac syndrome; the relationship of celiac disease, starch intolerance, and steatorrhea. J Pediatr 1947;30:564-82.
- 4. Wilson R. The coeliac syndrome with adolescent rickets. Ir J Med Sci 1951;301:39-42.
- Mann JG, Brown WR, Kern F Jr. The subtle and variable clinical expressions of gluten-induced enteropathy (adult celiac disease, nontropical sprue). An analysis of twenty-one consecutive cases. Am J Med 1970;48:357-66.
- Logan RF, Tucker G, Rifkind EA, Heading RC, Ferguson A. Changes in clinical features of coeliac disease in adults in Edinburgh and the Lothians 1960-79. Br Med J (Clin Res Ed) 1983;286:95-7.
- Mäki M, Collin P. Coeliac disease. Lancet 1997;349:1755-9.
- 8. Farrell RJ, Kelly CP. Celiac sprue. N Engl J Med 2002;346:180-8.
- Rodrigo-Sáez L, Fuentes-Álvarez D, Pérez-Martínez I, Alvarez-Mieres N, Niño-García P, de-Francisco-García R, *et al.* Differences between pediatric and adult celiac disease. Rev Esp Enferm Dig 2011;103:238-44.
- Harper JW, Holleran SF, Ramakrishnan R, Bhagat G, Green PH. Anemia in celiac disease is multifactorial in etiology. Am J Hematol 2007;82:996-1000.
- 11. Bergamaschi G, Markopoulos K, Albertini R, Di Sabatino A, Biagi F, Ciccocioppo R, *et al.* Anemia of chronic disease and defective erythropoietin production in patients with celiac disease. Haematologica 2008;93:1785-91.

- 12. Oberhuber G, Granditsch G, Vogelsang H. The histopathology of coeliac disease: Time for a standardized report scheme for pathologists. Eur J Gastroenterol Hepatol 1999;11:1185-94.
- Ludvigsson JF, James S, Askling J, Stenestrand U, Ingelsson E. Nationwide cohort study of risk of ischemic heart disease in patients with celiac disease. Circulation 2011;123:483-90.
- Emilsson L, Smith JG, West J, Melander O, Ludvigsson JF. Increased risk of atrial fibrillation in patients with coeliac disease: A nationwide cohort study. Eur Heart J 2011;32:2430-7.
- 15. Ludvigsson JF, Montgomery SM, Ekbom A, Brandt L, Granath F. Small-intestinal histopathology and mortality risk in celiac disease. JAMA 2009;302:1171-8.
- Emilsson L, Andersson B, Elfström P, Green PH, Ludvigsson JF. Risk of idiopathic dilated cardiomyopathy in 29 000 patients with celiac disease. J Am Heart Assoc 2012;1:e001594.
- Frustaci A, Cuoco L, Chimenti C, Pieroni M, Fioravanti G, Gentiloni N, *et al.* Celiac disease associated with autoimmune myocarditis. Circulation 2002;105:2611-8.
- 18. Riccabona M, Rossipal E. Pericardial effusion in celiac

disease: An incidental finding?. Wien Klin Wochenschr 2000;112:27-31.

- Lo W, Sano K, Lebwohl B, Diamond B, Green PH. Changing presentation of adult celiac disease. Dig Dis Sci 2003;48:395-8.
- Ackerman Z, Eliakim R, Stalnikowicz R, Rachmilewitz D. Role of small bowel biopsy in the endoscopic evaluation of adults with iron deficiency anemia. Am J Gastroenterol 1996;91:2099-102.
- 21. Unsworth DJ, Lock FJ, Harvey RF. Iron-deficiency anaemia in premenopausal women. Lancet 1999;353:1100.
- 22. Anand IS, Chandrashekhar Y, Ferrari R, Poole-Wilson PA, Harris PC. Pathogenesis of oedema in chronic severe anaemia: Studies of body water and sodium, renal function, haemodynamic variables, and plasma hormones. Br Heart J 1993;70:357-62.
- Hopman EG, le Cessie S, von Blomberg BM, Mearin ML. Nutritional management of the gluten-free diet in young people with celiac disease in the Netherlands. J Pediatr Gastroenterol Nutr 2006;43:102-8.

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