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Case Report

AN INTRIGUING CAUSE OF CHRONIC DIARRHEA – PROTEIN-LOSING ENTEROPATHY AS THE PRESENTING MANIFESTATION OF SYSTEMIC LUPUS ERYTHEMATOSUS

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ABSTRACT

Systemic lupus erythematosus (SLE) is a systemic autoimmune inflammatory disease. It presents with a myriad of clinical manifestations affecting every other organ system in the human body. The gastrointestinal (GI) system is one of the most commonly affected systems and it involves any part of the GI tract with an incidence ranging from 1.3% to 27.5%. GI manifestations as a presenting feature of SLE are unusual. Chronic diarrhea due to protein-losing enteropathy as the presenting manifestation of SLE is very rare, and a diagnosis of SLE should be kept in mind when other systemic manifestations are absent. Hence, we report a 29-year-old female with chronic diarrhea as the main manifestation of SLE.

Keywords: Systemic lupus, Protein-losing enteropathy, Diarrhea, Bowel wall edema.

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INTRODUCTION

Systemic lupus erythematosus (SLE) is a systemic autoimmune inflammatory disease. It presents with myriad of clinical manifestations affecting every other organ system in the human body. It is very common in female population presenting with varied clinical manifestations in about 90% [1]. The gastrointestinal (GI) system is one of the most commonly affected systems and it involves any part of the GI tract with an incidence ranging from 1.3% to 27.5% [2,3]. However, most GI manifestations are mainly due to adverse reactions from drugs used therapeutically and infections. The symptoms related to the disease *per se* are very common as compared to other organ involvements such as lupus nephritis. Diarrhea in patients with SLE generally can be due to parasitic or bacterial intestinal infections and because of chronic pancreatitis or drugs such as azathioprine [4,5]. Diarrhea due to rare complications of SLE includes protein-losing enteropathy, lupus enteritis, intestinal pseudo-obstruction, eosinophilic enteritis, and celiac disease [6]. Here, we report a 29-year-old female with chronic diarrhea as the main manifestation of SLE.

CASE REPORT

Informed consent was taken from patient. A 29-year-old female presented with complaints of diarrhea for 2 months. Diarrhea episodes were 8-10 times/days, watery, large-volume not associated with blood or mucus. Occasional episodes of vomiting were present. She had weight loss of 15 kg in the last 2 months. There was no history of fever, rash, or arthralgia. On examination, vitals were stable. Systemic examination was unremarkable, except for mild periumbilical tenderness. Initial blood investigations showed normal blood count (Table 1) and blood urea and creatinine levels were also normal (Table 2). Stool microscopy was negative for white blood cell and parasites. Liver function showed hypoalbuminemia (2.6 g/dL) and transaminases were elevated to two times the upper limit of normal (Table 2). Urine spot protein was negative. In view of hypoproteinemia and negative proteinuria, contrast-enhanced computed tomography (CECT) abdomen was done and CT findings revealed diffuse bowel wall edema with small capacity urinary bladder and mild bilateral hydroureteronephrosis (Fig. 1). The possibility of vasculitis or a collagen disorder was suspected. Upper GI endoscopy was normal. Antinuclear antibody serology was strongly positive with anti-ds-DNA positivity detected by indirect immunofluorescence.

She was initiated on injectable methyl prednisolone and changed over to oral prednisolone. Patient's symptom showed marked improvement. Two weeks later, she presented with pedal edema and facial puffiness. Evaluation showed nephrotic range proteinuria. Renal biopsy was suggestive of lupus nephritis. She was treated with pulse cyclophosphamide, on which she went into remission.

DISCUSSION

SLE is an autoimmune inflammatory disease. It is usually characterized by autoantibodies and immune complex formation. Every system and organ can be affected by SLE. The disease is predominant in women as compared to men with a reported female:male ratio of 10:1.1. In 1895, physician William Osler described the GI complications of SLE and how it mimics other abdominal conditions and completely masks the other organ complications associated with SLE [6]. GI complications can cause myriad of symptoms across the GI tract. Patients usually experience diarrhea, anorexia, nausea, or vomiting. GI symptoms are common in SLE patients and are secondary to side effects of drugs used for treatment and due to viral or bacterial infections. It is not as common as lupus nephritis. GI involvement in SLE is highly important as mortality rates are high if patients are not treated promptly. Lupus mesenteric vasculitis is the most common cause, followed by protein-losing enteropathy, intestinal pseudo-obstruction, acute pancreatitis, and other rare complications such as celiac disease and inflammatory bowel diseases. Specific autoantibody associated with SLE-related gastroenteropathy has not been identified. Abdominal computed tomography scans are very helpful in diagnosing. Good therapeutic responses to corticosteroids and immunosuppressive agents are usually seen. Supportive measures include bowel rest, nutritional support, antibiotics, and prokinetic medications. It helps in recovering the function of bowel and improving therapeutic outcome. Protein-losing gastroenteropathy (PLGE) is characterized by edema and severe hypoalbuminemia secondary to excessive loss of from the bowel. It is clinically indistinguishable from nephrotic syndrome. It is very uncommon in SLE. So far, only 60 patients with SLE-related

Table 1: Laboratory investigations

Hemoglobin	12.1 g%
Neutrophils	69%
Lymphocytes	16%
Eosinophils	5%
Monocytes	10%
Packed cell volume	37%
WBC count	$6.04\times10^3/mm^3$
Platelet count	$224\times10^3/mm^3$
ESR	8 mm/hrs

WBC: White blood cell, ESR: Erythrocyte sedimentation rate

Table 2: Laboratory investigations

Albumin - serum	2.6 g/dL
Globulin - serum	4.1 g/dL
Alkaline phosphatase - serum/plasma	132 U/L
Alanine transaminase - serum/plasma	65 U/L
Aspartate transaminase - serum	62 U/L
GGTP	38 U/L
Glucose - serum/plasma (random)	90 mg/dL
Creatinine - serum/plasma	0.7 mg/dL
Urea - serum/plasma	33 mg/dL
Sodium - serum/plasma	135 mEq/L

GGTP: Gamma glutamyl transpeptidase

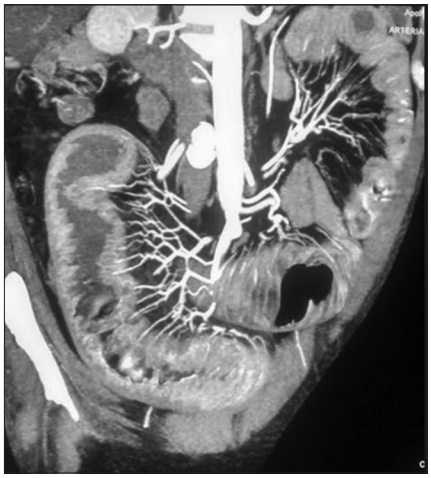

Fig. 1: Contrast-enhanced computed tomography (CT) of abdomen showing diffuse bowel wall edema with small capacity urinary bladder and mild bilateral hydroureteronephrosis

PLGE have been reported in the literature. It is predominant among the Asians and whether genetics or environmental factors play a role in it remains to be elucidated. PLGE has a female predominance and can affect any age group. The clinical manifestations of PLGE develop before the diagnosis of SLE. Gornisiewicz *et al*. [7] found PLGE is usually associated with multiorgan system involvement in SLE. Mucosal ulceration, non-necrotizing mesentericor intestinal blood vessel vasculitis, increase in capillary permeability caused by intravascular activation and conversion of complement, cytokine-mediated (such as tumor necrosis factor-α and interleukin-6) or complement-mediated vascular or mucosal damage, and intestinal lymphangiectasia have been postulated as the pathogenic mechanisms [8-10]. In our case, patient had diarrhea, vomiting, and weight loss for 2 months without a history of fever, arthralgia. Hypoalbuminemia was noted. Other causes of diarrhea were ruled out and CECT abdomen showed edematous bowel loops. Upper GI endoscopy was also normal. Patient was positive for anti-ds antibody. Hence, a diagnosis of protein-losing enteropathy due to SLE was made and the treatment was initiated. Patient was started on intravenous methyl prednisolone and changed over to oral form; there was a marked improvement in the condition of patient, and after 2 weeks, patient presented with pedal edema and facial puffiness. Laboratory evaluation revealed nephrotic range proteinuria and renal biopsy confirmed lupus nephritis. The patient was successfully treated with cyclophosphamide.

CONCLUSION

SLE is one of the most common connective tissue disorders in females. GI manifestations as a presenting feature of SLE are unusual. Chronic diarrhea due to PLE as the presenting manifestation of SLE is very rare, and a diagnosis of SLE should be kept in mind when other systemic manifestations are absent. The case highlights the importance of suspecting uncommon presentations of common diseases whenever there is a dilemma in the arriving at a plausible and valid diagnosis.

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