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ABOUT COVER

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AIMS AND SCOPE

The primary aim of *World Journal of Clinical Cases* (*WJCC*, *World J Clin Cases*) is to provide scholars and readers from various fields of clinical medicine with a platform to publish high-quality clinical research articles and communicate their research findings online.

WJCC mainly publishes articles reporting research results and findings obtained in the field of clinical medicine and covering a wide range of topics, including case control studies, retrospective cohort studies, retrospective studies, clinical trials studies, observational studies, prospective studies, randomized controlled trials, randomized clinical trials, systematic reviews, meta-analysis, and case reports.

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CASE REPORT

Systemic lupus erythematosus presenting with progressive massive ascites and CA-125 elevation indicating Tjalma syndrome? A case report

Jun-Di Wang, Yan-Fei Yang, Xian-Feng Zhang, Jiao Huang

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Abstract

BACKGROUND

Ascites, pleural effusion and raised CA-125 in the absence of malignancy in systemic lupus erythematosus is known as Tjalma syndrome.

CASE SUMMARY

We report a special case of a systemic lupus erythematosus patient presenting with Tjalma syndrome. She presented with ascites and elevated CA-125 in the absence of benign or malignant ovarian tumor and no pleural effusions, which is an unusual presentation for this rare condition.

CONCLUSION

Tjalma syndrome can present with massive ascites alone without pleural or pericardial effusions.

Key Words: Tjalma syndrome; Pseudo–pseudo Meigs' syndrome; Systemic lupus erythematosus; Ascites, CA-125, Case report

Core Tip: We report a special case of a systemic lupus erythematosus patient presenting with pseudo-pseudo Meigs' syndrome. She presented with ascites and elevated CA-125 in the absence of benign or malignant ovarian tumor and no pleural effusions, which is an unusual presentation for this rare condition: Tjalma syndrome can present with massive ascites alone without pleural or pericardial effusions.

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INTRODUCTION

Systemic lupus erythematosus (SLE) is a chronic, autoimmune disease with multiple systemic disorders. Tjalma syndrome, also known as pseudo–pseudo Meigs' syndrome, is a clinical manifestation of SLE that is characterized by ascites, pleural effusions and elevated CA-125 in the absence of benign or malignant ovarian tumor[1]. Massive ascites are rare in SLE patients without any other complications. Herein we report a special case of an SLE patient presenting with Tjalma syndrome. She presented with ascites and elevated CA-125 but no pleural effusions.

CASE PRESENTATION

Chief complaints

A 23-year-old woman presented with nausea, vomiting and distention for 2 wk without abdominal pain, diarrhea, rashes or arthralgia.

History of present illness

The patient had presented herself to an outside hospital 10 d ago where physical examination revealed a distended abdominal wall, while abdominal computed tomography scan revealed massive ascites (Figure 1A). Laboratory examinations at the outside hospital showed markedly elevated CA-125 at 1685 U/mL (0-35 U/mL). Ascitic fluid analyses revealed negative results from Rivalta tests. After diuresis treatment for 7 d, the amount of ascites in the patient was gradually reduced. However, there were no changes in nausea or vomiting.

History of past illness

The patient had a history of immune thrombocytopenia for 2 years and was administered with a long-term maintenance dose of 5 mg/d prednisone.

Personal and family history

The patient denied any family history.

Physical examination

Patient temperature and blood pressure were 37.2 ºC and 123/82 mmHg, respectively, whereas her heart and respiratory rates were 89 beats/min and 20 breaths/min, respectively. No rales were heard in lung auscultation, and her heart beat was regular without murmurs. Her abdomen was distended, shifting dullness was positive, and neither her liver nor spleen were palpable. Physical examination of other parameters did not reveal any abnormalities.

Laboratory examinations

The following is the patient's laboratory examination results: White blood cell count, 6.8 × 10^9/L; neutrophil%, 77.9%; hemoglobin, 100 g/L; platelet, 130 × 10^9/L; total protein, 58 g/L; albumin, 31.6 g/L; d-dimer, 1910 μg/L; and ferritin, 37.7 μg/L. The 24-h urine protein quantitate was 74 mg/24 h. Antinuclear antibody 1:100 (+), Anti-Sjogren's syndrome A antigen antibody (+), C3 0.46 g/L (0.79–1.52) and C4 0.11 g/L (0.12–0.36). Lymphocyte subset findings were: Total lymphocytes, 500 × 10^6/L; T-cell lymphocytes (CD3), 226 × 10^6/L; B-cell lymphocytes (CD19), 265.9 × 10^6/L; T-helper lymphocytes (CD4), 54.4 × 10^6/L; natural killer cells (CD16/56), 5.6 × 10^6/L; and CD4/CD8, 0.71. In addition, the tumor marker (CA-125) was 439.9 U/mL, whereas other tumor markers, including AFP, CEA, NSE, CA153, CA199 and β-HCG were normal. Moreover, erythrocyte sedimentation rate, C-reactive protein, ANCA,

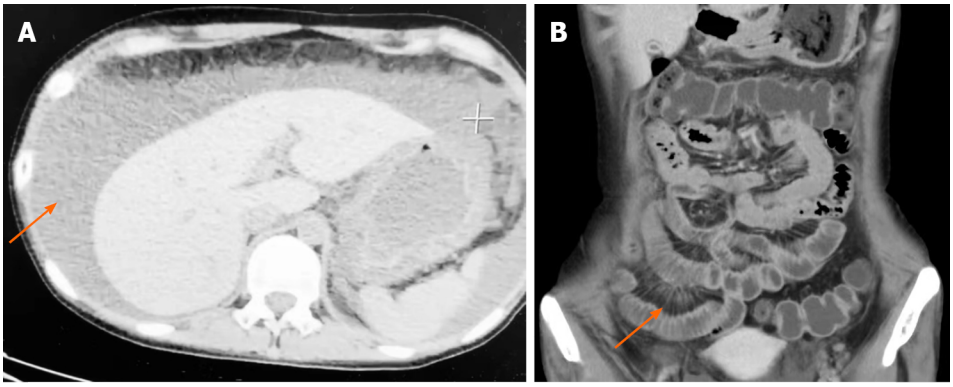

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Figure 1 Computed tomography scan. A: Abdominal computed tomography scan revealed massive ascites (orange arrow); B: Small bowel enhanced computed tomography revealed that the number of mesenteric vessels was increased. Mesenteric vessels were engorged and exhibited a "comb sign" (orange arrow) appearance.

index of autoimmune liver diseases, immunoglobulin G4, hepatitis B surface antigen and HIV were all found to be within normal ranges. T-SPOT showed negative results.

Imaging examinations

Small bowel enhanced computed tomography revealed a swollen gastric wall. Small bowel wall and colon wall were slightly thickened with abnormal bowel enhancement. The number of mesenteric vessels was increased, and mesenteric vessels were engorged exhibiting a "comb sign" appearance (Figure 1B). Enhanced magnetic resonance imaging scans of the pelvic tumor showed bilateral ovaries with enlarged multifocal cystic lesions; thus, endometriotic cysts were considered. Abdominal ultrasound showed abdominal effusions, while portal ultrasound observations were normal. Ultrasonic examinations did not reveal any pleural or pericardial effusions. Gastrointestinal endoscopy revealed diffuse edema of gastric and colon walls.

FINAL DIAGNOSIS

Tjalma syndrome, protein-losing enteropathy and lupus cystitis.

TREATMENT

The patient was treated with 20 mg intravenous methylprednisolone and 0.2 g hydroxychloroquine *per* day.

OUTCOME AND FOLLOW-UP

There was a subsequent improvement in nausea and vomiting during her hospital stay while her ascites were reduced. However, she later presented with violent vomiting, and 7 d after admission, she was vomiting moderate amounts of a coffee-like liquid. Then, the patient started presenting with yellow watery diarrhea. Ultrasonographic examinations and abdominal computed tomography scans showed bilateral hydronephrosis and hydroureter in addition to bladder wall thickening and small abdominal effusions. The fecal occult blood test was positive, and stool cultures revealed an infection of *Clostridium difficile*. Tests for *Clostridium difficile* toxins A and B were positive. Then, she was treated with 80 mg intravenous methylprednisolone twice daily and oral vancomycin for 10 d, which resulted in symptomatic improvement and the absence of any pathogens from her repeat stool microbiological investigations. Prior to discharge, her renal ultrasound was normal and CA-125 was 21.8 U/mL. The patient was discharged from the hospital with 12 mg oral prednisolone and 0.2 g hydroxychloroquine. At follow up 1 mo later, there was no vomiting or diarrhea.

Table 1 Reported cases of pseudo-pseudo Meigs syndrome

Year	Ref.	Gender	Age	Naive SLE	CA125[1]	Ascites	Pleural effusion	Pericardial effusion	Nausea/ vomiting	Dyspnea	Hypoproteinemia	Proteinuria	ANA	dsDNA	SSA	Low complement	Leukopenia	Anemia	Thrombocytopenia	APS	Treatment	Outcome
2005	Tjalma [13]	Female	38	Yes	887	+	+			+	+							+	+	+	MP + AZA	Remission
2005	Schmitt *et al*[1]	Female	33	Yes	2287	+	+			+	+	+	+	+		+	+	+	+	+	MP + MMF + HCQ	Remission
2008	Ural *et al* [14]	Female	38	Yes	1229	+	+			+			+	+	+	+					MP + HCQ	Remission
2011	Bes and Soy[15]	Female	47	Yes	233	+	+	+	+	+			+	+		+	+	+			MP + HCQ	Remission
2012	Dalvi *et al* [3]	Female	56	No	70.1	+	+		+				+	+		+	+				MP + MMF	Remission
2013	Bes *et al* [16]	Female	42	Yes	91.3	+	+	+	+	+	+	+	+	+		+			+		MP + CYC + AZA	Remission
2013	Lee *et al* [17]	Female	29	Yes	345	+	+	+	+	+	+	+	+	+		+	+	+	+	+	MP + HCQ	Remission
2013	Lee *et al* [17]	Female	54	No	344.9	+	+	+			+	+	+	+		+	+		+		MP + MMF	Remission
2016	Cheah *et al*[7]	Female	34	No	1613.8	+	+				+		+			+		+			MP + HCQ	Remission
2016	McVorran *et al*[18]	Female	40	Yes	307	+	+	+		+			+	+		+					MP	Remission
2019	Torres Jiménez *et al*[19]	Female	14	Yes	59	+	+	+		+	+		+	+		+		+	+		MP + CYC + MMF + RTX	Remission
2018	Zampeli *et al*[20]	Female	40	No	85	+	+	+		+	+		+	+	+	+	+	+			MP + CYC + MMF	Remission
2019	Awad *et al*[21]	Female	43	No	80	+	+				+	+	+	+		+		+			MP + MMF + HCQ	Remission
2019	Tansir *et al*[22]	Female	22	Yes	2025	+	+	+			+	+	+	+		+		+	+	+	MP + CYC + HCQ + AZA	Remission
2019	Li *et al*[23]	Female	24	No	949	+	+	+		+	+		+			+	+				MP + MMF	Remission

2019	Ahmed *et al*[24]	Female	44	Yes	227	+	+			+		+		+	+	+	+							MP + AZA	Remission
2019	Gao *et al* [6]	Female	44	Yes	360.8	+	+				+	+	+	+	+	+	+		+	+				MP + HCQ + LEF	Remission
2021	Quintero-Muñoz *et al*[25]	Female	33	No	187	+	+			+	+	+	+	+	+	+	+		+				+	MP + MMF + HCQ + CYC	Death
2021	Meena *et al*[26]	Female	23	No	230.5	+	+				+	+		+	+		+			+			+	MP + HCQ + AZA	Remission
2022	Karadeniz *et al*[27]	Female	33	No	476	+	+	+		+	+	+	+	+	+		+							MP + MMF + HCQ	Remission
2022	Current case	Female	23	Yes	1685	+				+		+		+		+	+				+			MP + HCQ	Remission

[1]U/mL.

ANA: Antinuclear antibodies; dsDNA: Anti-double-stranded DNA antibodies; SSA: Anti-Sjögren's syndrome-related antigen A; APS: Antiphospholipid syndrome; MP: Methylprednisolone; AZA: Azathioprine; MMF: Mycophenolate mofetil; HCQ: Hydroxychloroquine; CYC: Cyclophosphamide; RTX: Rituximab; LEF: Leflunomide.

DISCUSSION

CA-125 is a biomarker for gynecological malignancy. Clinically, CA-125 can be elevated by various benign diseases. Elevated CA-125 levels in SLE patients are attributed to mesothelial cell activation. In SLE patients, elevated serum CA-125 levels are independently associated with serositis[2,3]. Pleural and pericardial effusions are common among SLE patients. However, massive ascites are rare in SLE patients without any other complications[4]. Ascites in SLE are attributed to nephrotic syndrome, constrictive pericarditis, lupus peritonitis, protein-losing enteropathy or Budd–Chiari syndrome. A rapid onset of massive ascites can be an initial manifestation of SLE[5].

Our patient presented with painless massive ascites coexisting with low complement and hypoproteinemia. However, she did not show any overt proteinuria, and heart ultrasound as well as hepatic hilum ultrasound were normal. Therefore, lupus peritonitis, nephrotic syndrome, constrictive pericarditis and Budd–Chiari syndrome were ruled out. We postulated that hypoproteinemia was due to protein-losing enteropathy, resulting in intestinal damage caused by SLE (diarrhea, bowel wall edema and mesenteric vasculitis), consistent with previous studies[6,7]. However, 99m-labeled human serum albumin is required for definite diagnosis[8], which is not available at our hospital.

Lupus cystitis is a rare complication of SLE that generally presents with lower urinary tract symptoms and gastrointestinal symptoms, such as vomiting, nausea and abdominal pain[9,10]. Ultrasonographic examination of the patient showed bilateral hydronephrosis and hydroureter in addition to bladder wall thickening, which conforms to manifestations of lupus cystitis. Yuan *et al*[11] reported that lupus mesenteric vasculitis and lupus cystitis concurrently occurred in 22.7% of patients,

thus lupus cystitis should be suspected in SLE patients, especially those with lower urinary tract and gastrointestinal symptoms.

We summarized the clinical features of previous 20 cases of Tjalma syndrome and current cases (Table 1). All patients were female, and their mean age was 36.5 ± 10.7 (mean ± SD) years. A decrease in serum C3 and C4 levels was reported in all Tjalma syndrome patients, which was attributed to complement consumption caused by complement system activation[12]. The patient was clinically diagnosed with SLE with elevated CA-125, but there were no benign or malignant tumors. A review of previous studies revealed ascites and pleural effusions in all cases, but only 10 patients presented with pericardial effusions. Although there were no pleural effusions, just as pericardial effusions were not found in some previous cases, the clinical features of this case fit the Tjalma syndrome, which can be a specific finding. Tjalma syndrome can present with massive ascites alone without pleural or pericardial effusions, which requires further clinical attention. Generally, Tjalma syndrome has good prognostic outcomes after administration of methylprednisolone and immunosuppressants, with resolution of ascites and pleural effusions and normalization of CA-125.

CONCLUSION

In conclusion, massive ascites with increased CA-125 do not always indicate the presence of malignancy, especially in patients with SLE. Although rare, Tjalma syndrome has been increasingly reported in recent years. Therefore, there is a need for increased awareness of this condition.

FOOTNOTES

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