

CASE REPORT

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A rare cause of polyserositis: Delayed diagnosis of Sheehan's syndrome

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Abstract

Polyserositis is a challenging differential diagnosis. We present a case of severe serositis in a 51-year-old Caucasian woman complicated by pericardial, peritoneal and pleural exudates in the setting of panhypopituitarism and no other obvious etiology. The patient had obtundation, pretibial edema, anemia, hyponatremia, and high levels of CA 125. Low levels of TSH, free T3 and free T4 suggested central hypothyroidism. Decreased levels of morning cortisol, growth hormone, and gonadotropins indicated panhypopituitarism. The patient reported cessation of menses after her last delivery approximately 20 years ago. To our knowledge, this is the first reported case of panhypopituitarism presenting with polyserositis.

Keywords: Hypopituitarism, serositis, myxedema

Introduction

Polyserositis is defined as general inflammation of serous membranes associated with serous effusion due to many causes. The causes of polyserositis were presented in Table 1 [1-8]. In our knowledge, this is the first published case of tissue edema (pretibial) and multiple body cavity effusions (ascites, pleural and pericardial effusions) due to panhypopituitarism.

Table 1. Some causes of polyserositis

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|----|---|
| A. | Inflammatory disorders: Familial Mediterranean Fever |
| B. | Autoimmune diseases: Systemic lupus erythematosus, polyglandular autoimmune syndrome type II |
| C. | Immunological conditions: Graft-versus-host disease, adult-onset Still's disease, Castleman's disease, 13-valent pneumococcal conjugate vaccine |
| D. | Infections: Tuberculosis, Mycoplasma pneumoniae, Burkholderia pseudomallei, Neisseria meningitidis |
| E. | Drugs: Amiodarone, Clozapine, Ramipril, methotrexate, praziquantel |
| F. | Benign neoplasms: Mucinous cystadenoma of appendix |
| G. | Chronic myelomonocytic leukemia, Waldenström's macroglobulinemia, Nasal-type T/natural killer (NK)-cell lymphoma |
| H. | Hypothyroidism |

Case Report

A 51 year old female, postmenopausal, nonsmoker, nonalcoholic presented to our outpatient department in September 2017 with a history of dyspnea and generalized weakness. A physical examination showed pallor, pretibial edema and doughy skin. Her face is dull and expressionless. Leukocytes were 9000/mm³, hemoglobin 9.7 g/dl, platelets 328.000/μl. Serum urea 15 mg/dl, creatinine 0.53 mg/dl, sodium 129 mol/L, potassium 3.4 mmol/L, albumin 2.74 g/dl, total protein 5.27 g/dl, LDH 251 U/L, 24 hour urine protein level was 7 mg/day. Other biochemical tests were within the normal range. PO₂ 73 mmHg, PCO₂ 39 mmHg, pH 7.51, HCO₃ 31 mmol/L in arterial blood gas analyses. A chest CT-scan showed left upper lobe consolidation (Fig.1), and bilateral pleural effusion, and pericardial effusion (Fig.2). She treated with ampicillin + sulbactam (4x1 gr) and clarithromycin (2x 500 mg) for the diagnosis of pneumonia. No endobronchial lesion observed in fiberoptic bronchoscopy. There were not acid-fast bacilli in bronchial lavage fluid.

Serum carcinoembryonic antigen (CEA), CA 19-9 and CA 15-3 levels were within normal ranges whereas CA 125 levels were elevated at 69.2 U/ml (normal < 21 U/ml). She had ascites but not an ovarian mass in pelvic ultrasound examination. Echocardiography revealed pericardial effusion measuring 11 mm but left ventricular functions were within the normal range. Pleural fluid glucose was 81 mg/dl, LDH 119 U/L, albumin 1.91 g/dl, protein 3.21 g/dl. No acid-fast bacilli observed in pleural effusion. Bacteriological, mycological and mycobacteriological cultures of pleural fluid were free of agent pathogen. Cytological examination of pleural

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effusion showed no signs of malignant cells.

Thyroid stimulating hormone (TSH, 0.96 mIU/ml), free triiodothyronine (T3, <0.26 pg/ml), free thyroxine (T4, 0.14 ng/dl), anti thyroperoxydase (<5 IU/ml), anti thyroglobulin (346.3 IU/ml), Adrenocorticotropin (ACTH, 16.4 pg/ml), Luteinizing hormone (LH, 0.52 mIU/ml), Follicle-stimulating hormone (FSH, 3.4 mIU/ml), growth hormone (< 0.05 mg/L), prolactin 2.8 ng/ml (N:4.8-23.3), morning cortisol levels 2.68 µg/dl were analyzed. Rheumatoid factor, anti-ds DNA, ANA, ANCA, anti-mitochondrial antibodies, anti-smooth muscle antibody, anti-gliadin, anti-endomysium antibodies, and islet cell antibodies were negative.



Figure 1. A chest CT-scan showing left upper lobe consolidation

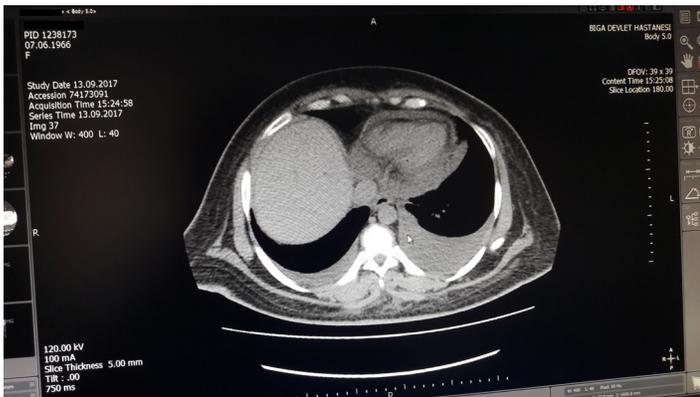


Figure 2. Pericardial effusion and bilateral pleural effusion

The story of the patient and the hormone results were sufficient to make a diagnosis of panhypopituitarism due to Sheehan's syndrome. She treated with hydrocortisone 10mg/day, and levothyroxin 100 µg/day. She responded well to hormone replacement therapy.

Discussion

The accumulation of proteinaceous fluid in vital body cavities of persons diagnosed with hypothyroidism is widely documented, the most common sites being the pericardial, pleural, and peritoneal cavities. These complications may be attributed to increased capillary permeability and subsequent leakage of fluid high in protein into the interstitial space, impaired lymphatic drainage, and a greater retention of salt and water [9]. Hypothyroidism is associated with reduced glomerular filtration rate. Reduced glomerular filtration rate can directly decrease free water excretion by diminishing water delivery to the diluting segments

[10]. Because the serous effusions due to hypothyroidism have generally high levels of protein, the underlying mechanism is probably increased vascular permeability. The extravascular accumulation of albumin can be detected after 2-3 months of hypothyroidism [9]. The precise mechanism of effusion formation in patients with myxedema is unclear but Vascular Endothelial Growth Factor increases capillary permeability, and the levels of this factor are elevated in patients with hypothyroidism and return to normal with thyroid replacement therapy [11].

Low serum levels of thyroid hormones, cortisol, and gonadotrophin (FSH, LH) indicated panhypopituitarism. Hyponatremia is an important presenting feature of pituitary disease and a strong indicator of life-threatening steroid deficiency. Old age and severity of hypopituitarism are major risk factors for hyponatremia [12]. Our patient had hyponatremia and anemia. Pituitary hormones modulate the production of erythropoietin in the kidney [13]. On the other hand, the cause of the patient's decompensation was probably pneumonia. Hyponatremia may be due to pneumonia.

Although increased levels of CA 125 with ascites suggested of an ovarian tumor in the patient, our investigations excluded an abdominal neoplastic disease. On the other hand, both tuberculosis [14] and hypothyroidism [15], as a cause of polyserositis, can be associated with high CA 125 levels.

Conclusion

Although the causes of panhypopituitarism might be due to tumoral, immunological, genetical, iatrogenic, traumatic or infectious, the patient reported amenorrhea after the last parturition 20 years ago. This finding strongly suggested Sheehan syndrome. In conclusion, primary hypothyroidism should be kept in mind in patients with exudates.

Competing interests

The authors declare that they have no competing interest.

Financial Disclosure

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