Case Report

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Polycystic Ovary Syndrome Combined with Type II Polyglandular Autoimmune Syndrome

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ABSTRACT

A 30-year-old woman with a 20-year history of type 1 diabetes referred to our clinic with the complaints of facial acne, hirsutism, and irregular menstrual cycles (hirsutism was identified in physical examination). On biochemical analysis, high serum levels of anti-TPO and DHEA-S were detected. Based on ultrasonographic findings indicative of thyroiditis and positive anti-TPO test, the diagnosis of Hashimoto's thyroiditis was made. A diagnosis of Polycystic Ovary Syndrome (PCOS) was confirmed later according to the Rotterdam criteria (revised 2003). The patient received metformin and insulin for 3 months and her menstrual cycles became regular.

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Introduction

Polycystic ovary syndrome (PCOS) is one of the most common endocrine disorders. It affects 5-10% of women in the reproductive age and about 50-70% of these women suffer from infertility due to anovulatory cycles (1,2). Clinical presentations of this syndrome include irregular menstrual cycles, hyperandrogenism, insulin resistance, and obesity. Clinical features of Polyglandular Autoimmune Syndrome (PAS) type 2 include existence of two or more of the disorders including Addison's disease, Hashimoto's thyroiditis, type 1 diabetes, premature ovarian failure, and celiac disease.

The combination of these two disorders is a rare condition and has been reported rarely (3). The aim of current study was to present coincidence of PCOS and autoimmune polyglandular syndrome type II simultaneously in a patient.

Case Report

A 30-year-old woman with a 20-year history of type-1 diabetes, referred to our endocrine clinic with the complaint of irregular menstrual cycles, acne, and hirsutism. She had been hospitalized twice with a diagnosis of diabetic

ketoacidosis previously (at age 10 and 15 years). Her family history was unremarkable. Her blood sugar was poorly controlled despite receiving multiple daily injections (MDI) of insulin. Clinical examination showed stable vital signs and a blood pressure of 120/80 mmHg without any sign of hypotension. Her BMI and waist circumference were 23 kg/m^{2}and 78 cm, respectively and she had no complaint about weight loss or loss of appetite.

On physical examination and ultrasonography, thyroid was rigid and large with hypoechoic thyroid lobes and some heterogeneity (Figure 1). The Frriman-Gallwey score was calculated as 9. Results of serum biochemical analysis was as follows: FBS; 180 mg/dl, BS 2hpp: 346 mg/dl and HbA1c: 7.9%. Accordingly, the daily insulin dose was adjusted to 50 IU/d long acting and rapid acting insulin 4 times a day. Metformin 500 mg three times a day was also started.

Serum TSH and fasting blood cortisol was measured to check the function of the thyroid and adrenal gland. In immunological tests, positive ANA and high anti-TPO antibody level was reported. Based on the high level of anti-TPO antibody and ultrasonographic findings, Hashimoto's thyroiditis was diagnosed despite the fact that all thyroid function tests were normal (4). A negative anti TTG test rejected the possibility of celiac disease, and normal prolactin and 17-hydroxyprogesterone levels ruled out prolactinemia and adrenal congenital hyperplasia, respectively. Cushing syndrome and androgen tumors were also ruled out by normal androstendione, testosterone, and urine free cortisol (in a 24-hour urine collection) (Table 1). Considering hyperandrogenism (high DHEA-S), irregular anovulatory menstrual cycles, and polycystic ovaries was reported in ultrasonography (>12 small follicles in both ovaries), a diagnosis of PCOS was made for the patient according to the Rotterdam criteria (revised 2003).

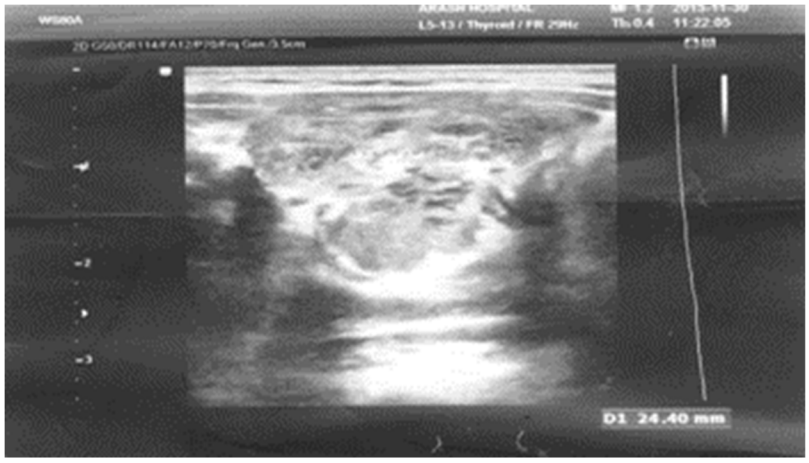

Figure 1. Thyroid ultrasonography

Table 1. Laboratory findings in the patients

	Patient	Normal Range
ANA	1/80	Up to 1/80
Anti TPO (IU/ml)	526	Up to 34
TSH (mUI/L)	0.9	0.2- 4.5
T4 (ng/dl)	9	5- 12.5
T3 (ng/dl)	180	70- 200
Serum cortisol (µg/dl)	11	4.5-24
Anti TTG (U/ml)	5.2	<12
Prolactin (ng/ml)	20.6	6- 30
17(OH) P (ng/ml)	0.4	0.15- 1.1
DHEA.S (µg/dl)	351	99- 340
Testosterone (ng/ml)	0.47	0.06- 0.8
Urine free cortisol (µg/24h)	26	13.7- 75
Androstenedione (ng/ml)	1.8	0.3- 2.4

Discussion

Several studies have demonstrated a positive relationship between PCOS and autoimmune diseases (5-6). One study by Luborsky et al. rejected any association and even suggested a protective role for PCOS against autoimmune diseases (7). Type 1 diabetes and Hashimoto's thyroiditis are two well-known autoimmune diseases that have been discussed in details in the literature (2, 8, 9). Moreover, Kachuei et al. reported significantly higher level of anti-thyroid antibodies in patients with PCOS (p=0.04) in comparison with control group (8).

The case we presented here was diagnosed with Hashimoto's thyroiditis based on thyroid enlargement detected on physical examination and sonography and high levels of anti-TPO antibodies.

In 2010, Ganie et al. reported a significantly higher incidence of PCOS (47%) in patients with chronic lymphocytic thyroiditis (CLT) in comparison with the control group (4%) (10).

Considering the inhibitory effects of progesterone on the immune system and the low levels of progesterone in PCOS patients, it may be suggested that over-stimulation of the immune system in patients with PCOS results in the secretion of certain autoantibodies (11). In contrast to the aforementioned findings, some studies have demonstrated that considering its protective function, high levels of androgen play a protective role in PCOS cases and prevent the development of autoimmune diseases (7, 12). Findings of different studies have demonstrated that type 1 diabetes and hyperinsulinemia are closely related to each other. Insulin in vena cava plays a key role in testosterone and estrogen metabolism by preventing the liver from the synthesis of SHBG through increasing androgen secretion from theca cell and increasing the levels of free testosterone (13).

Considering the contradictory results of the previous investigations regarding the association between PCOS and autoimmune disorders, the present study demonstrated a possible association between PCOS and type II autoimmune polyglandular syndrome. Based on the findings of the current study, investigations for autoimmune thyroiditis and other autoimmune disorders are recommended in patients with PCOS. However, more in-depth studies are warranted to shed light on the impact of autoimmune diseases on the development of PCOS.

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