CASE REPORT
Endocrinology, Nutrition & Metabolism

JKMS

DOI: 10.3346/jkms.2011.26.4.580 • J Korean Med Sci 2011; 26: 580-582

A Case of Sheehan's Syndrome that Manifested as Bilateral Ptosis

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Received: 15 September 2010
Accepted: 8 December 2010

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Hypothyroidism can cause a variety of signs and symptoms of the neuromuscular system. However, ptosis in a patient with hypothyroidism is very rare. We report here on a case of central hypothyroidism that was due to Sheehan's syndrome and it manifested as bilateral ptosis in a 51-yr-old woman. She complained of exertional dyspnea and weakness. About 25-yr ago, she had a history of severe postpartum vaginal bleeding. The laboratory studies demonstrated hypopituitarism with secondary hypothyroidism. The ptosis was improved by replacement of thyroid hormone. Hypothyroidism should be considered in the differential diagnosis of patients who manifest with ptosis and that prompt replacement of hormone can lead to a complete recovery.

Key Words: Hypothyroidism; Hypopituitarism; Ptosis

INTORDUCTION

Ptosis may be due to myogenic, neurogenic, aponeurotic, mechanical or traumatic causes. The most frequent disorder causing myogenic ptosis is myasthenia gravis (1). Hypothyroidism can cause a variety of signs and symptoms of the neuromuscular system (2). However, in the absence of other obvious etiologies, ptosis in a patient with hypothyroidism is very rare (3-5). We report here on a case of central hypothyroidism that was due to Sheehan's syndrome and the patient manifested with bilateral ptosis, and the patient was initially suspected as having myasthenia gravis.

CASE DESCRIPTION

The patient was a 51-yr-old woman with swelling of the extremities and bilateral ptosis on March 8, 2010. She complained of exertional dyspnea and weakness. She married at the age of 25. After that time, she developed drooping of her eyes, with the left eye drooping more than the right eye. Her symptoms persisted and they had gradually increased during about the last 10 yr. About 8 yr ago, she underwent a ptosis repair operation at a local clinic, but her symptoms were not improved. She denied a history of loss of consciousness, headache, dysphagia, dysarthria and myalgia. There was no family history of similar symptoms and no history of trauma. There was no previous history of diabetes and hypertension.

On examination, she was determined to be 151 cm in height and 48 kg in weight. The initial vital signs were a blood pressure of 140/80 mmHg, a pulse rate of 68 beats/min and a respiratory rate of 20/min. She was lethargic and had facial puffiness and no goiter. The lung and heart examinations were unremarkable. All the extremities showed pitting. She had marked bilateral ptosis, while the external ocular movements were normal (Fig. 1A). The other cranial nerve examinations were unremarkable.

The laboratory findings were a total leucocyte count 4.75 × 10^3/μL with 61.6% polymorphs, a hemoglobin level of 9.0 g/dL, the random blood glucose was 124 mg/dL, the serum sodium was 130 mM/L, the potassium was 3.7 mM/L, the blood urea nitrogen (BUN) was 13 mg/dL, the creatinine was 0.8 mg/dL, the aspartate transaminase (AST) was 81 IU/L, the alanine transaminase (ALT) was 29 IU/L, the total bilirubin was 0.93 mg/dL, the creatine kinase (CK) was 1,195 IU/L (normal reference: 20-180 IU/L), the lactate dehydrogenase (LDH) was 517 IU/L (normal reference: 101-218 IU/L), the total cholesterol was 256 mg/dL, the triglyceride was 121 mg/dL, the high density lipoprotein cholesterol (HDL-C) was 40 mg/dL and the low density lipoprotein cholesterol (LDL-C) was 196 mg/dL. The urinary analysis was negative for blood and protein with using a dipstick. As the clinical findings suggested hypothyroidism and myasthenia gravis, a thyroid function test and acetylcholine receptor binding antibody test were done. The serum T3 was 0.195 ng/mL (normal reference: 0.86-2.02 ng/mL), the free T4 was 0.08 ng/dL (normal reference: 0.93-1.705 ng/dL), the thyroid-stimulating hormone (TSH) was 2.08 μIU/mL (normal reference: 0.27-4.2 μIU/mL), the antimicrosomal antibody was 17 IU/mL (normal reference: 0-34 IU/mL), the antithyroglobulin antibody was 20.1 IU/mL (normal reference: 0-114 IU/mL) and the acetylcholine receptor binding antibody was negative.

Secondary hypothyroidism was suspected. We performed a careful history taking and other pituitary hormone evaluations. At the age of 26 the patient delivered her daughter and she had

pISSN 1011-8934
eISSN 1598-6357

a history of massive postpartum vaginal bleeding. Thereafter she not resumed menses. The basal levels of other hormones were a serum cortisol of 4.03 μg/dL, the adrenocorticotropic hormone (ACTH) was 29.64 pg/mL, the growth hormone (GH) was 0.06 ng/mL, the IGF-1 was 25 ng/mL (normal reference: 71-263 ng/mL), the prolactin was 1.59 ng/mL, the luteinizing hormone (LH) was 1.49 IU/L, the follicle-stimulating hormone (FSH) was 4.91 IU/L, the E2 was 17.29 pg/mL and the testosterone was 0.02 ng/mL. The combined pituitary stimulation test, including the insulin tolerance test, the thyrotropin-releasing hormone (TRH) stimulation test and the gonadotropin-releasing hormone (GnRH) stimulation test showed panhypopituitarism (Table 1). Brain magnetic resonance imaging (MRI) showed a finding of an empty sella turcica and there was no evidence of an intracranial mass, hemorrhage and aneurysm (Fig. 2).

Nerve conduction study (NCS) and electromyography (EMG) of the limbs reveled normal results with the exception of incidentally detected carpal tunnel syndrome. Needle EMG of the orbicularis oculi showed no evidence of dysfunction of the neuromuscular junction.

We diagnosed her as having Sheehan's syndrome, bilateral ptosis and subclinical myopathy caused by secondary hypothyroidism. Replacement with prednisolone of 10 mg/day and thyroxine (T4) 100 μg/day was started. The dose of prednisolone was decreased to 5 mg/day after 4 weeks. At 3 months follow-up, she had become euthyroid with normalized muscle enzymes. She recovered from her presenting symptoms and the bilateral ptosis was much improved (Fig. 1B).

DISCUSSION

Sheehan's syndrome occurs as a result of ischemic pituitary necrosis due to severe postpartum hemorrhage. Although a small percentage of patients with Sheehan's syndrome may have an abrupt onset of severe hypopituitarism immediately after delivery, most patients have mild disease and they go undiagnosed for a long time and they are treated inappropriately (6). In this current patient, chronic fatigue and weakness had persisted, but it had gone undiagnosed for about 20 yr.

Initially, her symptoms such as ptosis, progressive dyspnea and weakness were suspicious for myasthenia gravis. Myasthenia gravis is an acquired neuromuscular disorder that is characterized by fatigability and fluctuating weakness of the skeletal muscles, and especially eye muscle weakness. The weakness is

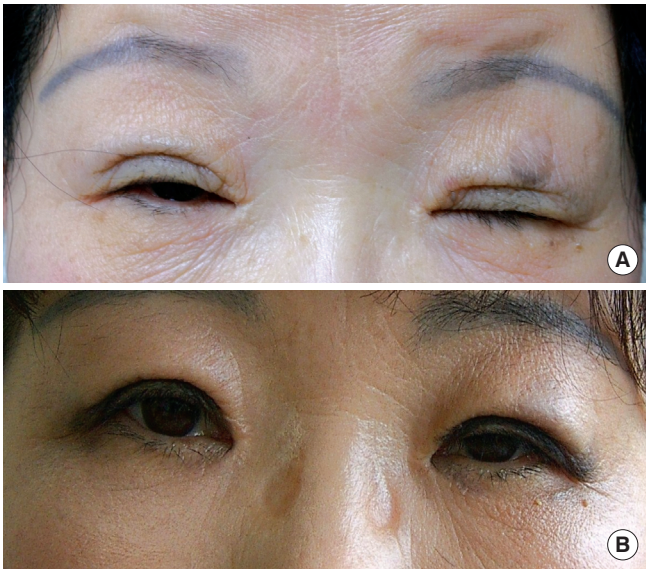

Fig. 1. Bilateral ptosis before (**A**) and 3 months after (**B**) replacement of thyroid hormone.

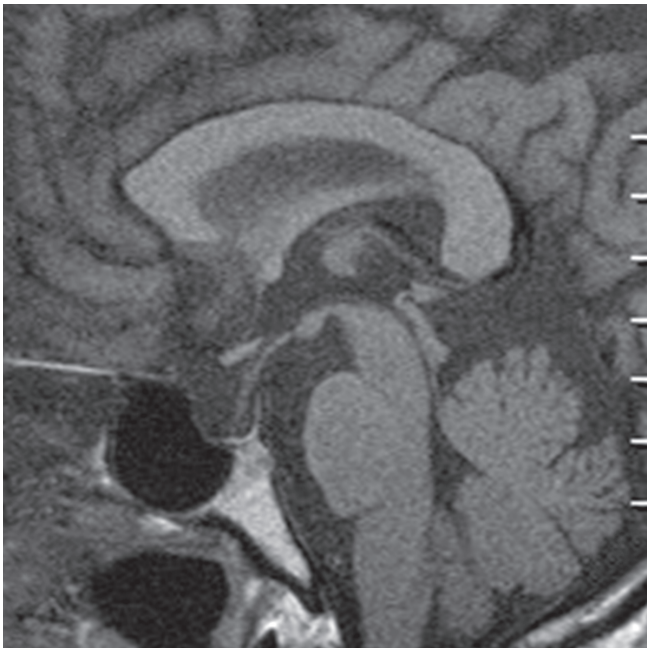

Fig. 2. The sagittal noncontrast T1 weighted magnetic resonance image of the brain shows an enlarged sella turcica and a nonvisualized gland.

Table 1. Results of the combined pituitary stimulation test

Time (min)	Glucose (mg/dL)	GH (ng/mL)	Cortisol (μg/dL)	Prolactin (ng/mL)	TSH (μU/mL)	LH (IU/L)	FSH (IU/L)
0	82	0.06	4.03	1.59	2.08	1.49	4.91
30	39	0.07	4.32	2.02	2.66	2.56	5.27
60	202	0.16	4.44	1.59	2.65	3.04	4.96
90	108	0.14	4.42	1.55	2.66	3.51	5.67
120	62	0.06	3.87	1.40	2.6	3.22	5.60

GH, growth hormon; TSH, thyoid-stimulating hormone; LH, luteinizing hormone; FSH, follicle-stimulating hormone.

caused by circulating antibodies that block the acetylcholine receptors at the postsynaptic neuromuscular junction, and this inhibits the stimulative effect of the neurotransmitter acetylcholine (7). In most cases, the first noticeable symptom is weakness of the eye muscles. Therefore, the presence of ptosis necessitates the exclusion of underlying ocular myasthenia gravis (4). In this patient, the acetylcholine receptor binding antibody was negative and needle EMG of the orbicularis oculi showed no evidence of dysfunction of the neuromuscular junctions. Also, the ptosis was improved by replacement of thyroid hormone. Therefore, myasthenia was adequately excluded.

Ptosis has been previously reported in patients with primary hypothyroidism, but there have been only a few case reports (3-5). In patients with hypothyroidism, myopathy, mononeuropathy and diffuse peripheral polyneuropathy may encountered with various incidences (8). Carpal tunnel syndrome is the most frequently observed in hypothyroidism. Polyneuropathy and myopathy are also well known (8). Patients can complain of muscle and joint pain, cramps, fatigue and weakness. However, involvement of a cranial nerve in a patient with hypothyroidism is very rare. Although neuromuscular complications are well recognized in thyroid disorders, their pathophysiology remains unclear. Generally, these abnormalities are secondary to mucopolysaccharide infiltration of various peripheral organs (5). Cho et al. (9) described a case of pituitary apoplexy with Sheehan's syndrome and isolated third cranial nerve palsy. In their case, direct mechanical compression of the third cranial nerve or the vascular supply to the nerve resulted in a sudden onset of isolated third cranial nerve palsy. The patient's ptosis was improved by surgical decompression. However, our patient had long standing secondary hypothyroidism and the ptosis had an insidious onset. The patient's ptosis was improved by replacement of hormone.

Moderate myopathy frequently develops in patients with hypothyroidism. This myopathy is most often manifested as myalgia, muscle stiffness, cramps and sometimes elevated plasma levels of creatinine phosphokinase. Infrequently, rhabdomyolysis can also develop in patients with hypothyroidism (10). Our patient had no specific symptoms of myopathy, but she had elevated levels of muscle enzyme. After replacement of hormone, the muscle enzymes were normalized.

Adrenal insufficiency is can produce generalized muscle weakness, muscle cramping and fatigue. However, except for generalized weakness, musculoskeletal manifestations have rarely been described in adrenal insufficiency (11) and which is thought to be due to the electrolyte abnormalities in primary adrenal insufficiency (12). Our patients had secondary adrenal insufficiency and mild hyponatremia. Therefore we thought that our patient's musculoskeletal manifestations with ptosis is due to mainly central hypothyroidism.

We report here on an unusual case of ptosis associated with central hypothyroidism due to Sheehan's syndrome. It is concluded that hypothyroidism should be considered in the differential diagnosis of patients who manifest with ptosis and that prompt replacement of hormone can lead to a complete recovery.

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DOI: 10.3346/jkms.2011.26.4.580