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Aggravated Hashimoto Thyroiditis and Chronic Inflammatory Demyelinating Polyradiculoneuropathy in Postpartum Period: Case Report

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Abstract

Chronic inflammatory demyelinating polyradiculoneuropathy (CIDP) is an autoimmune disease characterized with multifocal demyelination in peripheral nerves. Hashimoto thyroiditis is autoimmune disease of thyroid gland. We reported a patient having these diseases together in the postpartum period. Twenty-three year-old female patient presented with progressive weakness in lower extremities and walking disability for three months. Symmetric weakness (4/5) in both flexor and extensors of ankle and toes, decreased deep tendon reflexes, decreased vibration and position sence and unresponsive plantar reflex in lower extremities were found in neurological examination. We found elevated TSH, AntiTPO and ATG levels, decreased T3 and T4 levels in serum, albuminocytological dissociation in cerebrospinal fluid (CSF) and conduction blocks in peripheric nerves in electrophysiological examination. Patient was diagnosed as Hashimoto thyroiditis and CIDP according to these findings, and she was treated with L-thyroxine and intravenous methylprednisolone. Clinical findings improved, but electrophysiological findings didn't improved at second month.

Keywords

Chronic Inflammatory Demyelinating Polyradiculoneuropathy, Hashimoto Thyroiditis, Postpartum Hashimoto Thyroiditis, Postpartum Chronic Inflammatory Demyelinating Polyradiculoneuropathy

Introduction

Chronic inflammatory demyelinating polyradiculoneuropathy (CIDP), is an autoimmune disease characterized with multifocal demyelination in peripheral nerves [1]. It's pathogenesis has not been understood yet [2]. CIDP is characterized with relapsing or progressive weakness which affects proximally muscles dominantly, sensory loss, hyporeflexia or areflexia, demyelination findings in electrophysiological studies (prolonged distale motor latency, prolonged F-wave latency, decreased motor nerve conduction velocity, conduction block, temporal dispersion), albuminocytologic dissociation in cerebrospinal fluid (CSF), demyelination and remyelination findings in peripheral nerve biopsy [3]. Most of CIDP patients can be diagnosed with clinical and electrophysiological findings; nerve biopsy is less often required [4]. Hashimoto thyroiditis is an autoimmune disease of thyroid gland, and it is the most frequent cause of hypothyroidism. Hashimoto thyroiditis can progress in postpartum period due to disappearance of placenta's immunosupressive effect [5,6].

The appearance of CIDP and Hashimoto thyroiditis together is very rare. There are a few case reports about this autoimmune complex. But, there is no case report about appearance of these two diseases in the postpartum period. Here, we reported a patient with aggravated Hashimoto thyroiditis with CIDP in postpartum period.

Case

Twenty-three-year-old female patient presented with increasing weakness in lower extremities and walking disability last three months. She had ascending numbness in lower limbs, which recovered in a week four months ago. She had two pregnancies in medical history, and she gave a birth five months ago. There was a period of hypothyroidism after her first pregnancy four years prior to this, recovered without any treatment. In examination, we found symmetrical paraparesis with the weakness of flexors and extensors of thigh and knee (4/5), flexors and extensors of toe (4/5), decreased patellar and achilles reflexes, unresponsive plantar reflex, and decreased vibration and position sense in lower extremities. We found thyroid stimulating hormone (TSH) > 100 uIU/ml (0.35–4.94), freeT3 2.32 pmol/l (2.6–5.7) freeT4 < 5.15 pmol/l (9–20), antithyroglobulin (ATG) 321.12 IU/ml (<4.11) and antithyroid peroxidase (anti-TPO) > 1000 IU/ml (< 5.61). Chronic thyroiditis was spotted in thyroid ultrasonography. Hemograme, serum glucose, electrolytes, liver and kidney functions, protein, albumin, and creatine kinase were normal. The patient was diagnosed with Hashimoto thyroiditis based on these laboratory findings.

In electrophysiological examination, we found decreased motor conduction velocities (MCV) and conduction blocks(CB) in median, ulnar, peroneal, and posterior tibial nerves; prolonged distal motor latencies (DML) in posterior tibial nerves; prolonged F-wave latencies in median nerves; absent F-waves in ulnar and posterior tibial nerves; multiple A-waves in posterior tibial nerves. Bilateral median nerve sensory conduction velocities (SCV) and sensory action potentials (SAP) were normal. Ulnar and sural nerve SAPs were absent (Table

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Table 1: Electrophysiological findings at onset

Motor Conduction Studies					
Nerve	**Stimulation Point**	**Record Point**	**Latency (ms)**	**Velocity (m/s)**	**Amplitude (mV)**
Right Median	Wrist	APB	2.8		10.3
	Elbow		16.6	16.5	4.4
	Axilla		24.9	17	2.5
Left Median	Wrist	APB	3.0		8.6
	Elbow		16.8	18.3	3.5
	Axilla		24.2	21.2	2.2
Right Ulnar	Wrist	ADM	2.5		7.1
	Elbow		21.4	12	1.6
	Axilla		34.8	6	1.2
Left Ulnar	Wrist	ADM	2.7		7.5
	Elbow		20.5	11.3	2.2
	Axilla		33.1	6.7	1.8
Right Peroneal	Ankle	EDB	5.1		3.9
	Head of fibula		46.7	0.7	0.8
Left Peroneal	Ankle	EDB	5.0		3.0
	Head of fibula		44.9	1.0	0.6
Right Tibial	Ankle	AHL	9.9		4.2
	Poplitea		46.4	1.1	1.4
Left Tibial	Ankle	AHL	9.5		4.0
	Poplitea		45.5	0.8	1.2
Sensory Conduction Studies					
Nerve	**Stimulation Point**	**Record Point**	**Latency (ms)**	**Velocity (m/s)**	**Amplitude (μV)**
Right Median	Wrist	2nd Finger	2.52	47.6	8
Left Median	Wrist	2nd Finger	2.45	45.2	9
Right Ulnar	Wrist	5th Finger	Unelicitable		
Left Ulnar	Wrist	5th Finger	Unelicitable		
Right Sural	Low Cruris	Lat. Malleol	Unelicitable		
Left Sural	Low Cruris	Lat. Malleol	Unelicitable		

ms: milisecond; m/s: metre/second; mV: milivolt; APB: Abductor Pollicis Brevis; ADM: Abductor Digiti Minimi; EDB: Extensor Digitorum Brevis; AHL: Abductor Hallucis Longus; μV: microvolt.

Table 2: Electrophysiological findings at second month

Motor Conduction Studies					
Nerve	**Stimulation Point**	**Record Point**	**Latency (ms)**	**Velocity (m/s)**	**Amplitude (mV)**
Right Median	Wrist	APB	3		10
	Elbow		10.3	31	9
Left Median	Wrist	APB	3.2		8
	Elbow		10.2	30	8
Right Ulnar	Wrist	ADM	3		4.6
	Elbow		11.1	22	1.4
	Axilla		14.0	31	2.0
Left Ulnar	Wrist	ADM	2.8		4.2
	Elbow		10.5	21.5	1.5
	Axilla		13.7	30.3	2.2
Right Peroneal	Ankle	EDB	7.5		2.0
	Head of fibula		20.2	21.2	0.3
Left Peroneal	Ankle	EDB	7.7		1.8
	Head of fibula		20.5	23.8	0.2
Right Tibial	Ankle	AHL	6.4		4.6
	Poplitea		26.1	19.3	0.7
Left Tibial	Ankle	AHL	6.6		4.4
	Poplitea		25.7	21	0.5
Sensory Conduction Studies					
Nerve	**Stimulation Point**	**Record Point**	**Latency (ms)**	**Velocity (m/s)**	**Amplitude (μV)**
Right Median	Wrist	2nd Finger	Unelicitable		
Left Median	Wrist	2nd Finger	Unelicitable		
Right Ulnar	Wrist	5th Finger	Unelicitable		
Left Ulnar	Wrist	5th Finger	Unelicitable		
Right Sural	Low Cruris	Lat. Malleol	Unelicitable		
Left Sural	Low Cruris	Lat. Malleol	Unelicitable		

ms: milisecond; m/s: metre/second; mV: milivolt; APB: Abductor Pollicis Brevis; ADM: Abductor Digiti Minimi; EDB: Extensor Digitorum Brevis; AHL: Abductor Hallucis Longus; μV: microvolt.

1). In CSF examination, we spotted protein 72.8 mg/dl and glucose 70 mg/dl (serumglucose = 95 mg/dl). Albuminocytologic dissociation was found in CSF examination as there were no cells in the CSF. According to EFNS 2010 criterias [3], patient was diagnosed as CIDP. We treated patient with 1 g/day intravenous methylprednisolone (IVMP) for seven days, and 100 μg/day L-thyroxine. Muscle strengths and gait were recovered on the fourth day of treatment.

After two months from IVMP treatment, the patient's symptom

lessened partially. Paresis on ankle flexors and extensors recovered. But paresis in the toe flexors and extensors continued (-5/5). Thyroid functions were normal. We found demyelination findings in electrophysiological studies (Table 2). We followed the patient with 100 µg/day L-thyroxine treatment.

Discussion

Hashimoto thyroiditis and CIDP are both autoimmune diseases. The appearance of these two diseases, at same time, is extremely rare. CIDP diagnoses depend on clinical findings, electrophysiological findings, and albuminocytologic dissociation in CSF examination [3]. Our patient had relapsing, remitting clinical evaluations with symmetric weakness, loss of vibration and position sensations, and hyporeflexia over two months. In the electrophysiological studies, we found demyelinating findings as reduction in motor conduction velocities (70%) in lower limb nerves, conduction blocks in all nerves and loss of F-wave in four nerves. Albuminocytologic dissociation was found in CSF. These clinical, electrophysiological, and laboratory findings support the CIDP. We excluded Guillain-Barre Syndrome based on relapsing and remitting clinical results over two months; multifocal motor neuropathy due to symmetric findings; paraproteinemias due to normal serum protein and albumin levels; hereditary polyneuropathies due to electrophysiological findings and no family history.

Hypothyroidism and high levels of ATG and anti-TPO levels support the Hashimoto thyroiditis. Our patient had hypothyroidism, which recovered without any treatment, four years ago. After second pregnancy, Hashimoto thyroiditis aggravated. Progression of Hashimoto thyroiditis and appearance of CIDP in postpartum period suggest that these diseases may occur with same autoimmune mechanisms.

Hashimoto thyroiditis is an autoimmune disease characterized by degeneration following lymphocyte infiltration and fibrosis [7]. ATG and anti-TPO levels are high in most postpartum patients [7]. ATG and anti-TPO levels may decrease due to the placenta's immune suppressive effect during pregnancy. After loss of placenta's immune suppression, ATG and anti-TPO levels may increase and this may cause the progression of Hashimoto thyroiditis [5]. Our patient's ATG and anti-TPO levels were very high in postpartum period.

CIDP may appear due to cellular or humoral mechanisms, including immunoglobulins, complement, lymphocyte, and macrophages [1,8]. Despite researchs, antigens that evoke an immune response have not been found. Experimental studies suggest that possible targets may be glycolipids and myelin proteins, but antibodies against this structures were found in low numbers of patients [9].

In a study of 61 CIDP patients, McCombe et al. found that relapse rate may increase during pregnancy [10]. Kawada et al reported a CIDP patient started in pregnancy and recovered after breedeing [11]. Vital et al. reported two patients with severe axonal polyneuropathy in postpartum period [12]. Zeeman et al. reported an acute inflammatory demyelinating polyneuropathy in pregnancy [13]. Meenakshi-Sundaram et al. reported a patient with Guillian-Barre Syndorme which occured in pregnancy and relapsed in postpartum period [14]. But we didn't find any CIDP patient appearred in postpartum period in literature.

Togetherness of CIDP and Hashimoto Thyroiditis is extremely rare. Bairactaris et al. reported a patient with CIDP and Hashimoto throiditis [15]. Raghavendra et al. reported patient with CIDP, Hashimoto thyroiditis and nephropathy [16]. Toscano et al reported a patient with multifocal motor neuropathy and asympthomatic Hashimoto thyroiditis [17]. Reisin et al. reported a patient with multifocal motor neuropathy, Hashimoto thyroiditis and type-1 diabetes [18]. We didn't find any patient occurred in postpartum period.

The pathophysiology of this autoimmune complex is unclear. Complex and various mechanisms, including the cell mediated and antibody mediated responses, may be effective in this autoimmune complex [4]. This autoimmune complex may occur due to structural homology and epitope sharing of the thyroglobulin peptide fragments with myelin related proteins [16]. More studies are required to understand the relationship between these two diseases.

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