CASE REPORT
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Recurrent Painless Thyroiditis in Patients with History of Postpartum Thyroiditis

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It is well known that the long-term prognosis of postpartum thyroiditis (PPT) is excellent except recurrent PPT in subsequent pregnancies and risk of progression to permanent hypothyroidism in some patients. However, the prospective observation of PPT patients who have neither consecutive gestation nor any evidence of hypothyroidism were limited. We describe three patients who have history of PPT and showed repeated painless thyroiditis in the span of more than ten years. The clinical courses of repeated painless thyroiditis were the transient thyrotoxicosis, self-limited, and not related to pregnancy. Based on the clinical courses of our three patients, it is recommended to remember that transient painless thyroiditis could be repeated as a possible long-term course of the patients with history of PPT.

Key Words: Postpartum thyroiditis, Long-term prognosis, Painless thyroiditis

Introduction

Postpartum thyroiditis (PPT) is an autoimmune thyroid disease (AITD) with a self-limited course.[1] Although recurrent PPT could be possible in subsequent pregnancies, it is well known that the long-term prognosis of PPT is excellent, except for the risk of progression to permanent hypothyroidism.[2] Then, what will happen to those patients who have neither consecutive gestation nor any evidence of hypothyroidism? Do they all stay in the life-long, stable euthyroid state after PPT? Here, we introduce three patients who have the history of PPT and showed repeated thyrotoxic phases of painless thyroiditis in the span of more than ten years. Since the beginning of the episodes were hyperthyroid states, this could be confused with Graves' disease. However, the courses were transient painless thyroiditis and were not related to pregnancy. These cases provide not only an important viewpoint of the long-term care for PPT, but also the natural pathologic process of AITD itself.

Case Reports

Case 1

A 39-year-old woman, who delivered her first baby three month ago, visited the outpatient clinic with symptoms of weight loss (−4 kg/3 months), anxiety, mild goiter and insomnia. She had no past history of any thyroid diseases. Based on the history of delivery, hyperthyroid state (Table 1, PPT), decreased radioactive iodine uptake (RAIU) (24 hours RAIU=0.2%) and Tc99m uptake in the thyroid scan (Fig. 1), findings of thyroid ultrasound (coarse and heterogeneous echotexture) (Fig. 2), we made a diagnosis PPT and followed thyroid function test (TFT) without antithyroid drugs. Within one

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Table 1. Data of thyroid function tests of case 1

Date	TSH (0.25–4.0) ulU/ml	FT4 (0.78–1.94) ng/dL	AMA (0–100) IU/ml	ATA (0–70) IU/ml	TSI (0–1.5) IU/L	Descriptions
Sep. 2007: Delivery						
Dec. 2007	0.01	4.95	4.9	120	0.3	PPT
Jan. 2008	5.27	0.5	40	171	0.45	
Sep. 2008	1.98	1.17	215	83	0.3	Euthyroid
15 months after PPT						
Mar. 2009	0.01	1.73	25.5	326	0.5	First episode of painless thyroiditis
Apr. 2009	3.38	0.86	71.2	713	0.5	
Dec. 2009	0.99	1.13	7.6	416	0.3	Euthyroid
26 months after PPT						
Feb. 2010	0.01	3.31				Second episode of painless thyroiditis
Apr. 2010	15.5	0.08				
May 2010	2.94	0.92				Euthyroid
38 months after PPT						
Feb. 2011	0.01	4.32	43.7	970	0.3	Third episode of painless thyroiditis
Mar. 2011	0.67	1.03	51.7	1422	0.3	
Mar. 2012	1.04	1.04	22.7	638	0.2	Euthyroid

AMA: anti microsomal antibody, ATA: anti thyroglobulin antibody, FT4: free thyroxine, PPT: postpartum thyroiditis, TSH: thyroid stimulating hormone, TSI: thyroid stimulating immunoglobulin

year, her symptoms, diffuse goiter and thyroid dysfunction had been normalized (Table 1).

After 15 months, she re-visited our clinic with symptoms of fatigue, weakness and heat intolerance (Table 1, first episode of painless thyroiditis). With non-tender mild goitre, she had a hyperthyroid functional state. She refused to take the thyroid scan and RAIU test. She denied any issues related to recent delivery or a missed abortion. Thyroid function recovered spontaneously with symptomatic management only. After 26 months (Table 1, second episode of painless thyroiditis) and 38 months (Table 1, third episode of painless thyroiditis), she re-visited our clinic with symptoms of weight loss, weakness and mild goitre. A hyperthyroid state in TFT, decreased Tc^{99m} uptake in thyroid scan (Fig. 1) and the thyroid ultrasound findings showed more coarse and heterogeneous echotexture than previous episodes (Fig. 2). We checked her TFT regularly without any medications, and confirmed improvements within several months (Table 1). One year after the third episode of painless thyroiditis, she visited clinic for a follow-up assessment. She had no goitre, no symptoms and the results of TFT were within normal limits (Table 1).

Case 2

The second case showed three painless thyroiditis episodes and mildly fluctuating thyroid functions as the post-PPT course in a 13 years period of follow-up. After the delivery of her first baby, a 37-year-old woman experienced typical symptoms and TFT changes of PPT (Table 2, PPT), even though we could not check the RAIU or Tc^{99m} thyroid scan because of the patient's irrational fear of nuclear materials. Through a short period of a hypothyroid state, her TTF recovered over 8 months (Table 2).

The first fluctuation of thyroid function occurred approximately 22 months later (Table 2, first episode of painless thyroiditis). She complained of dyspnea, fatigue, weakness and mild goitre without tenderness. Her TFT showed hyperthyroidism with increased activity of anti-thyroglobulin antibody. The activity of the anti-TSH receptor antibody was within normal limits. Although we could not check the RAIU because the patient refused any diagnostic processes using isotopes, we thought that the symptoms and finding of thyroid ultrasound (coarse, heterogeneous echotexture) were compatible with transient painless thyroiditis (Fig. 2). Therefore, we simply followed TFT without

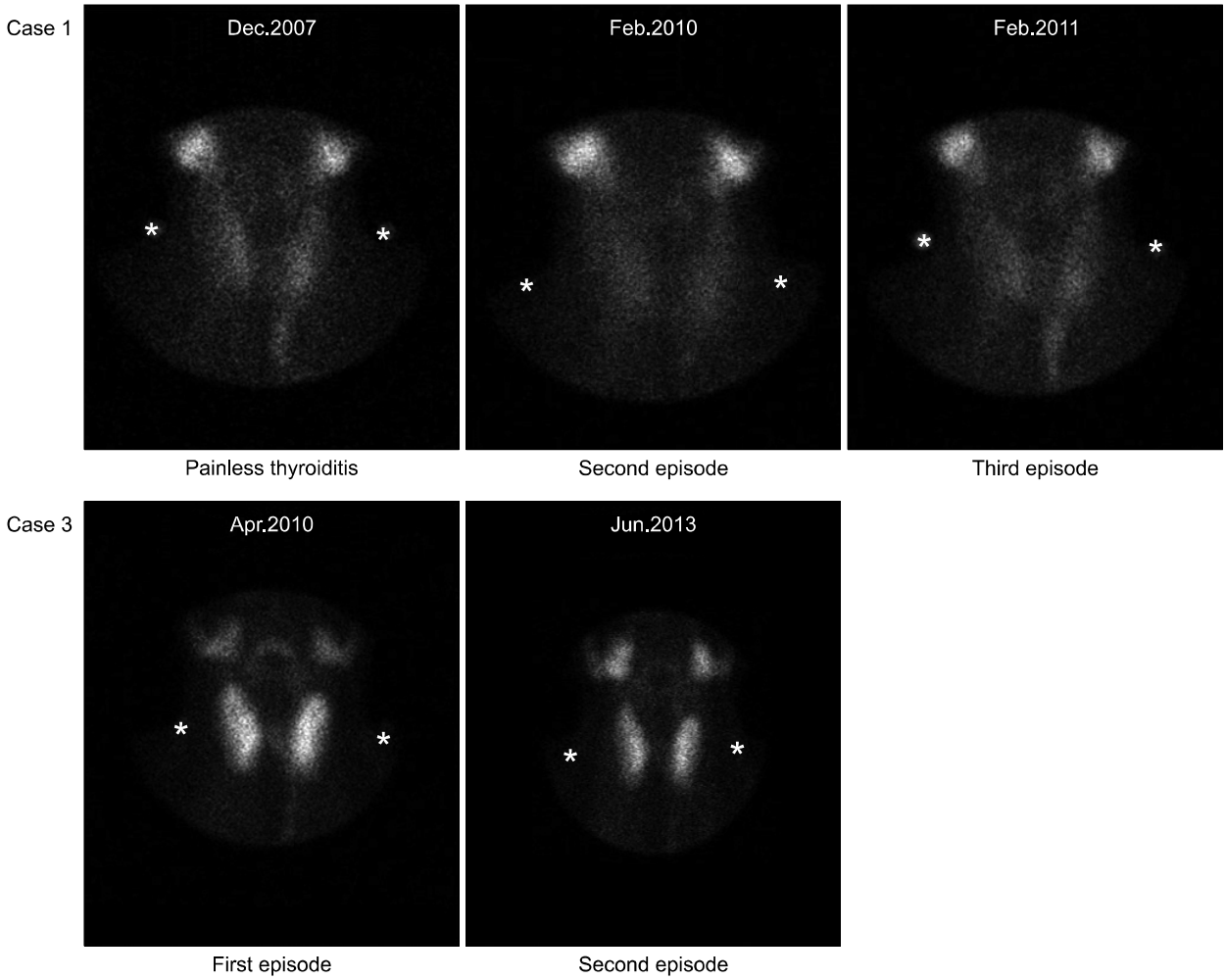

Fig. 1. Thyroid scans of painless thyroiditis episodes in cases 1 and 3.

any medications, and her symptoms and abnormal TFT were progressively normalized within 6 months. An annual health examination program showed that she remained in a euthyroid state for the next two years, a mild subclinical hypothyroid state in the third year and normalized in the fourth year. However, in the next year, she complained again of weakness, weight loss and a mild diffuse goitre (Table 2, second episode of painless thyroiditis) which had a spontaneously recovering course over six months. After a one and half year gap of normal thyroid function, the transient painless thyroiditis had recurred again with symptoms of fatigue and mild diffuse goitre (Table 2, third episode of painless thyroiditis), which spontaneously recovered within 5 months. Since then, her thyroid functions have remained in the normal range.

Case 3

This 41 years old woman bore two children and had recurrent PPT on each pregnancy. About two years after the second PPT, she visited our clinic complaining of fatigue. With a mild hyperthyroid state (Table 3, first episode of painless thyroiditis), her Tc^{99m} thyroid scan showed the decreased uptake mid-lateral margin of the left lobe (Fig. 1) and thyroid ultrasound showed the heterogeneous echotexture (Fig. 2). With the diagnosis of painless thyroiditis, regular checking of her TFT was recommended; however, she dropped. Five years after her second PPT, she revisited with symptoms of general weakness and agitation. Her TFT showed a mild hyperthyroid state (Table 3, second episode of painless thyroiditis), Tc^{99m} thyroid scan showed decreased uptake mainly in the

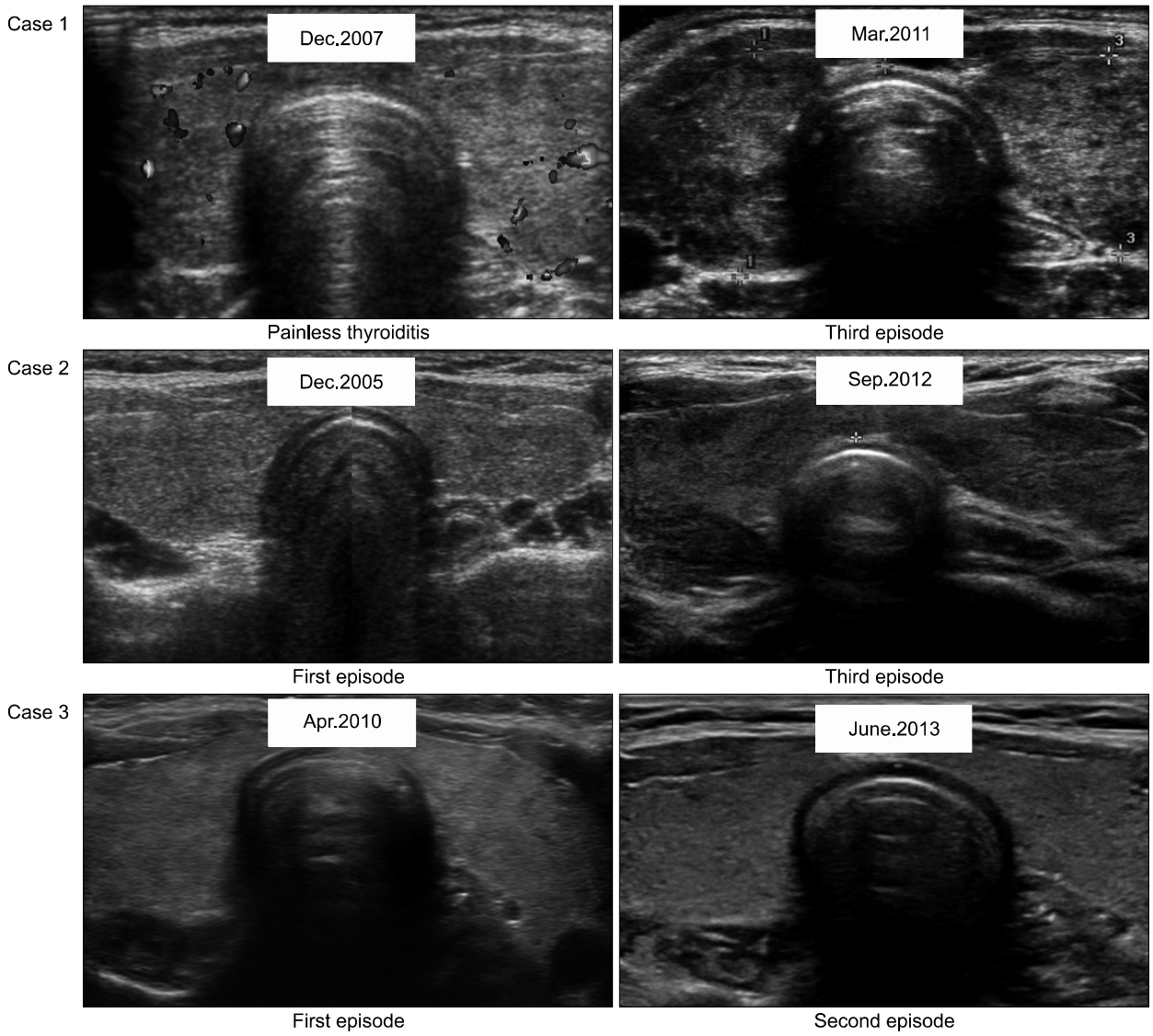

Fig. 2. Thyroid ultrasonographic findings of painless thyroiditis episodes in the three cases.

lateral margin of both lobes and thyroid ultrasound showed no change from the previous one (Fig. 2). Thyroid function was normalized in 4 months without medication. After this second episode of painless thyroiditis, her thyroid functions mildly fluctuated with a subclinical hyperthyroid pattern, then, it normalized spontaneously.

Discussion

The conventional guidelines for PPT management are mainly focused on the evaluation of PPT itself, the possible recurrence of PPT in following pregnancy and the progression to permanent hypothyroidism in some patients.[2,3] However, our three cases provide another option, which is that, some PPT patients could have several episodes of fluctuating thyroid functions as a long-term course which not related to repeated pregnancy. Based on our experiences, we would like to highlight the recurrent painless thyroiditis, as one of important issue of long term care of PPT.

From a clinical aspect, it is worth remembering that patients with a history of PPT could have painless thyroiditis at a certain point of their life. Since most cases would be resolved spontaneously, close observation with reassurance is enough in most cases. In partic-

Table 2. Data of thyroid function tests of case 2

Date	TSH (0.25–4.0) ulU/ml	FT4 (0.78–1.94) ng/dL	AMA (0–100) IU/ml	ATA (0–70) IU/ml	TSI (0–1.5) IU/L	Descriptions
Nov. 2003: Delivery						
Feb. 2004	0.03	3.65	22.3	454.3	0.5	PPT
Apr. 2004	17.79	0.77	32.3	772.9	0.3	
Oct. 2004	2.5	1.18	64.1	471.1	0.3	Euthyroid
22 months after PPT						
Dec. 2005	0.01	3.20	33.4	1298	0.3	First episode of painless thyroiditis
Feb. 2006	0.01	2.12	109.3	1196	0.3	
Aug. 2006	2.72	1.22	11.1	639.6	0.3	Euthyroid
Oct. 2008	4.33	1.23				Subclinical hypothyroid state
Dec. 2009	3.06	1.22				Euthyroid
6 years after PPT						
Apr. 2010	0.07	3.15	102.3	1344.9	0.2	Second episode of painless thyroiditis
Oct. 2010	2.87	1.13	14.9	384.1	0.1	Euthyroid
8 years after PPT						
Sep. 2012	0.02	3.81	47.2	422.4	0.1	Third episode of painless thyroiditis
Jan. 2013	2.80	1.11				Euthyroid
Sep. 2015	1.83	1.12				Euthyroid
Jun. 2017	2.10	1.14	20.8	855.5	0.1	Euthyroid

AMA: anti microsomal antibody, ATA: anti thyroglobulin antibody, FT4: free thyroxine, PPT: postpartum thyroiditis, TSH: thyroid stimulating hormone, TSI: thyroid stimulating immunoglobulin

Table 3. Data of thyroid function tests of case 3

Date	TSH (0.25–4.0) ulU/ml	FT4 (0.78–1.94) ng/dL	AMA (0–100) IU/ml	ATA (0–70) IU/ml	TSI (0–1.5) IU/L	Descriptions
Sep. 2002: Delivery (first baby)						
Apr. 2003	0.01	1.34	4.0	80.2	0.3	First PPT
May 2003	0.03	3.98				
Mar. 2008: Delivery (second baby)						
Jul. 2008	0.01	2.14	14.25	254.8	0.3	Second PPT
Sep. 2008	20.42	0.69	21.82	307.7	0.3	
Dec. 2008	4.88	0.8	7.41	210.1	0.3	
Jun. 2009	1.12	1.19	60.35	175.5	0.3	Euthyroid
2 years after second PPT						
Apr. 2010	0.03	2.12	7.5	726.1	0.3	First episode of painless thyroiditis
Jan. 2013	2.1	1.21				Euthyroid
5 years after second PPT						
Jun. 2013	0.01	2.43	10.0	10.2	0.3	Second episode of painless thyroiditis
Jul. 2013	0.01	1.58	7.4	15.5	0.1	
Oct. 2013	2.9	1.36	10.0	78	0.1	Euthyroid
May 2015	2.32	1.34	18.2	149	0.1	
Apr. 2016	0.12	1.53	1.3	147.3	0.1	Subclinical hyperthyroid state
Oct. 2017	2.62	1.15				Euthyroid

AMA: anti microsomal antibody, ATA: anti thyroglobulin antibody, FT4: free thyroxine, PPT: postpartum thyroiditis, TSH: thyroid stimulating hormone, TSI: thyroid stimulating immunoglobulin

ular, hasty initiation of antithyroid drugs in the hyperthyroid state should be performed carefully. Actually, in the early phases of painless thyroiditis episodes, two of our three cases (cases 2 and 3) were recommended the treatment with antithyroid drugs in primary clinics under a diagnosis of Graves' disease.

However, they did not take it as their personal decision and seek second opinion. All of them were satisfied with confirming the temporal course based on regular thyroid function assays instead of starting medication which might be unnecessary or aggravated the courses of the disease.

There is an opinion that autoimmune thyroid diseases should be understood to be on a mobile spectrum rather than as separate static disease entities.[4] Following these lines of thoughts, PPT might not be a separate event, but a changing status of thyroid autoimmunity on the spectrum, like a wave. After PPT, some patients have a calming-down course of their wave, which never increases again, some have a downhill wave which slowly progress to a permanent hypothyroid state; while others (as in the three cases discussed here) have temporally unstable thyroid status as a long-term course, like a fluctuating, wave. Although the detailed mechanisms of this instability are still unclear, we think the PPT could be understood not a solitary, one-off disease entity, but rather a part of long-standing chronic autoimmune disease.

Recurrence of sporadic painless thyroiditis itself is not unusual,[5-7] however, cases of recurrent transient thyroiditis after PPT are rare.[8,9] The previously reported cases had relatively short follow-up period (less than 5 years) and past history of autoimmune thyroid diseases.[8,9] On contrast, our cases were followed more than 10 years (except case 1) and all the patients did not have any past history of thyroid diseases or goiter. It suggests PPT of our patients were the first pathologic event of their thyroid, and repeated episodes of painless thyroiditis after PPT were triggered by PPT itself, not the pre-existing thyroid diseases.

Even though the case reports of transient thyroiditis after PPT are rare, we believe it may not be that rare. There is a possibility that many cases would be just ignored due to the mild, vague symptoms related to transient thyroid dysfunction. According to a follow-up study of postpartum thyroiditis, more than half of the patients had various forms of thyroid diseases within three years of PPT.[10] Approximately half of patient with PPT will eventually develop hypothyroidism after the first PPT.[11,12] Assuming the incidence of transient thyroiditis after PPT is not as rare as once believed, we wonder about the clinical characteristics belonging to this category. By comparing the similarities of our cases, unique points are as follows; they had their first child at an older age (over 35 years), the courses of their PPT lasted for a relatively long period (1 year) and their first transient thyroiditis happened mainly in the winter and spring. The seasonal correlation in the development of autoimmune thyroid disease has been introduced in previous studies and is of particular interest.[9,13] This suggests that the immune instability could be related to environmental or demographic factors. For a proper explanation, it will be worth attempting to collect these cases for comparable study.

Although the reported incidences vary depending on the countries of interest,[14] PPT is one of the most common autoimmune thyroid diseases in women. Thus, not only the management of PPT itself, but also the comprehensive information about long-term course of PPT is an important clinical issue. We expect that the experiences of our 3 cases could broaden the understanding of the natural course of PPT and appropriate management in various clinical settings.

Acknowledgments

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