Case Report

Graves' Disease Associated with Intersterno-costoclavicular Ossification and Pustulosis palmoplantaris

—A Case Report and Review of the Literature—

Takaaki Iwasaki, Hiroshige Ozeki, Manabu Narimiya and Yukihide Isogai

It was shown that intersterno-costoclavicular ossification often appears together with pustulosis palmoplantaris by Sonozaki et al., 1980. We report here a patient of Graves' disease associated with intersterno-costoclavicular ossification and pustulosis palmoplantaris, and discuss the relationship with these conditions.

Key Words: Intersterno-costoclavicular ossification, Pustulosis palmoplantaris, Graves' disease, TSH receptor antibody, HLA-typing, Autoimmuno disease.

Evidence for altered cell-mediated immunity has recently been demonstrated in Graves' disease. Current thinking on the pathogenesis of Graves' disease involves the immediate antibody that binds to the TSH receptor on thyroid cell membranes. Intersterno-costoclavicular ossification and pustulosis palmoplantaris are also considered to belong to the category of immunological disease.1, 2)

The present paper reports a twenty-five-year-old woman suffering from Graves' disease in conjunction with intersterno-costoclavicular ossification (ISCO) and pustulosis palmoplantaris (PPP).

CASE PRESENTATION

A twenty-five-year-old woman who complained chiefly palpitation and anterior chest pain, was admitted to the Higashitochigi National Sanatorium Hospital on May 16, 1984. The patient was first seen at this hospital in connection with noticeable palpitation and general fatigue. For the one-year period following the conclusion of propylthiouracil administration, the patient remained in good health. She then suffered a relapse of Graves' disease in 1980 simultaneous with an increased joint pain on shrugging her shoulders. The administration of propylthiouracil for one year restored her health and improved her arthralgia.

On May, 1984, the patient afflicted with a recurrence of palpitation and anterior chest pain which led her to this hospital again.

PHYSICAL EXAMINATION ON ADMISSION

Upon admission, the patient's temperature was 36.5°C by axilla, her pulse was 102/min and regular, having quick rise and fall. Her blood pressure was 102/62 mmHg. In addition, her weight was 40 kg, which was 4 kg less than the average expected weight. Moderate exophthalmos was noted bilaterally and her eyelids did not close completely on attempted lid closure. The thyroid gland was enlarged without tenderness and was softly palpable. The right lobe of the thyroid gland was 5 x 2 cm and the left lobe was 6 x 3 cm. The tenderness was apparent in the sterno-clavicular...
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Fig. 1. Many small pustules on palms and soles.

joint region. A grade-2 systolic murmur was audible at the base of the heart. The abdomen was flat and soft. Neurological examination was normal. There were many small pustules on the palms and soles (Fig. 1).

Finally, there was nothing remarkable concerning the patient’s family history.

LABORATORY EXAMINATION

Urinalysis values were as follows: specific gravity, 1.028; protein, negative; glucose, negative; sediment, normal.

Hematological values were as follows: Hematocrit, 37%; hemoglobin, 12.6 g/dl; white blood cell count, 9300/mm; platelets, 157,000/mm; sedimentation rate, 26 mm in/hour; LE cell test, negative. The blood chemical analysis findings were as follows: glutamic oxaloacetic transaminase, 21 mU/ml; glutamic purvic transaminase, 30 mU/ml; lactic dehydrogenase 313 mU/ml; total bilirubin, 1.2 mg/ml; cholesterol, 97 mg/dl; urea nitrogen, 20 mg/dl; albumin 3.1 g/dl;

Fig. 2. Tc-99m scinti scan shows “hot” lesion of the sterno-clavicular joints and thyroid glands.
globulin 1.6 g/dl; creatine phosphokinase, 51 IU/l.
The serologic test results were as follows: C-reactive protein, negative; rheumatoid factor, negative; serologic test for syphilis, negative; thyroid test, negative; microscope test, 1:320. The thyroid function test values were as follows: triiodothyronine (RIA), 3.2 ng/ml; free triiodothyronine, above 15.0 pg/ml; thyroxine (RIA), 24.2 μg/dl; free thyroxine, 5.5 ng/dl; thyroid stimulating hormone, below 1.3 μU/ml; thyrotropin binding inhibitor immunoglobulin (THII), 63.4% positive. The HL-A typing findings were A locus, A2; B locus, BW 46; C locus, CW1; DR locus, DR2.
Pustule cultures were negative. Chest X-ray film revealed slight ossification at the sternocostoclavicular joints. The tomography of the sternum and sternocostoclavicular joints, thyroid gland and peripherals of the extremities (Fig. 2).

COURSE IN THE HOSPITAL

Propylthiouracil therapy in 300 mg/day started on admission. Two months after the treatment, the patient was clinically in euthyroid and the sternocostoclavicular joint pain diminished. Pustulations of the extremities did not demonstrate good clinical restoration in the early stage of local corticosteroid therapy, but they improved significantly along with the recovery of Graves' disease.

DISCUSSION

Long acting thyroid stimulator (LATS) was found in the serum of Graves' disease patients by Adams and Purves in 1956. Since then, several thyroid stimulating immunoglobulins have been demonstrated one by one. Although underlying nature of these factors, their numbers and their relationships are not clear, some reports suggest that they are one or more autonomic antibodies against some components of the thyroid plasma membrane, perhaps the TSH receptor itself. Graves' disease has therefore come to be regarded as one of the receptor disease.

ISCO is a newly described rheumatic disease which is given a firm basis by Sonozaki et al. in 1974. The disease is characterized by symmetrical ossifications between the clavicle and the first rib. The most characteristic feature of this disease is an anterior chest pain, especially joint pain between the clavicle and the first rib with accompanying tenderness. Patients with ISCO complain of severe joint pain when shrugging their shoulders. X-ray findings are negative in the early phase of the disease. Abnormal ossifications and/or erosions at the costoclavicular ligament become more apparent as the disease progresses. In the most advanced stage, the clavicle and the first rib fuse with each other.

Recent reports have shown that approximately 80% of ISCO patients have PPP, whereas about 10% of PPP patients have ISCO. These complication rates are more than coincidental. In addition, in some PPP patients, the resection of the ossificated lesion has been reported to improve the PPP. PPP and ISCO are closely related. Therefore, Sonozaki et al. proposed the term "pustulotic arthro-ostitis" to describe this condition.

A few case reports exist, however, describing "pustulotic arthro-ostitis" patients as having hyperthyroidism. This complication is not rare, but is seem to be overlooked due to little awareness of its presence.

In the view of Graves' disease, a high incident rate of dermopathy is usually observed in LATS positive Graves' disease patients. Endo indicated many patients exhibiting localized pretibial myxedema had positive TSH receptor antibodies in their clinical course. This patient showed a high titer of TBII. It is necessary to look out the dermopathy of the Graves' disease patient when TBII is detected particularly.

ISCO, PPP and hyperthyroidism also traced the same clinical course in this patient. In addition, the 99m-Tc isotope scan showed a marked abnormal accumulation in thyroid gland, intersterno-costoclavicular joints and peripheral extremities at the same time. Therefore, some particular autoimmune-mechanism is thought to have a role in this case.
REFERENCES