AORTITIS SYNDROME AND HYPERTHYROIDISM —A CASE REPORT AND REVIEW OF THE JAPANESE LITERATURE—

YOSHIHIRO SATO¹⁾, MASARO KAJI¹⁾, SHUNJIRO KAGIYAMA²⁾
AND KENICHI NAGASAWA²⁾

*The First Department of Internal Medicine, Kurume University,
School of Medicine, Kurume, 830¹⁾ and Department of Internal
Medicine, Saga Prefectural Hospital, Saga, 840²⁾, Japan*

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A patient with aortitis syndrome and hyperthyroidism was reported. A review of the literature revealed 14 other cases with aortitis syndrome and autoimmune thyroid diseases in Japan. These cases may add to other indirect evidence that aortitis syndrome may be caused by an autoimmune mechanism.

INTRODUCTION

Aortitis syndrome, observed most frequently in young oriental women, is an occlusive polyarteritis mainly of the branches of aortic arch (Ueda et al, 1965). For this condition various names, such as Takayasu's arteritis, pulseless disease, aortic arch syndrome, and young female arteritis, have been given. Ueda et al (1976) proposed the term "aortitis syndrome", which we will use in the present report.

We have recently seen a patient with aortitis syndrome and hyperthyroidism. This is of interest in relation to the etiology of these disorders. Although the etiology of aortitis syndrome is still unknown, many authors suggested the disease may be included in the group of autoimmune disease. On the other hand, the evidence for an autoimmune basis in hyperthyroidism is considerable (Werner et al. 1972). It is not uncommon for more than one autoimmune disease to occur an individual concurrently, but a few papers referred to this association.

REPORT OF A CASE

A 32-year-old man was admitted to the hospital in December 12, 1978 because of palpitation, general malaise, and weight loss, with six months' duration. There was no past history of fever, visual disturbance, dizziness, and unconsciousness attack.

Physical examination revealed an alert, slightly under-nourished individual. The left radial pulse was normal in tension, but no pulsation was felt in the right branchial and radial arteries. Blood pressure was 110 systolic, 80 diastolic in the right arm, and 190 systolic, 80 diastolic in the left. Temperature was normal. The cardiac rhythm was regular with a rate of 108. The hands were warm and moist, and tremor of the fingers was present. The patient had exophthalmus. The thyroid was diffusely enlarged. Auscultation revealed a bruit maximal at the right neck, indicating stenosis of the right common carotid artery. This bruit was transmitted to the base of the heart. There was also noted a bruit in the left upper quadrant of the abdomen.

Ophthalmologic examination revealed nothing peculiar in both fundi. Deep tendon reflexes were exaggerated.

Examination of the blood revealed a hematocrit 48%, a red-cell count of 5.7 million, and a white-cell count of 6100, with 49% polymorphonuclear leukocyte, 6% monocyte, 1% eosinocyte, and 44% lymphocyte. Total serum protein was 7.6 mg., total cholesterol 132 mg., blood urea nitrogen 16 mg., and creatinine 0.8 mg. per 100 ml. Serum sodium was 143, potassium 4.3, and chloride 104 mEq. per liter. Serum immunoelectrophoresis showed an albumin 61.0%, α_1 -globulin 3.0%, α_2 -globulin 8.3%, β -globulin 10.2% and γ -globulin 17.2%. Erythrocyte sedimentation rate was 2mm/1hr. Tuberculin reaction was ++ positive. A serologic test for syphilis and CRP were negative. The basal metabolic rate was +29%. Serum thyroxine

level was 17.5 μ g. per 100 ml, and triiodothyronine was 4.4 ng. per ml. Serum thyrotropin response to thyrotropin-releasing hormone was subnormal. Plasma renin activity in basal state was 3.5 ng. per ml. per hour. Titer of antimicrosomal antibody was 1:6400, but antithyroglobulin antibody, rheumatoid factor, antinuclear antibody, and anti-aorta antibody were all negative.

A retrograde thoracic aortogram revealed a marked stenosis of the left subclavian artery, narrowing of the stem of the right common carotid artery, and irregular outline of both the aortic arch and the descending aorta (Fig. 1). A subsequent abdominal aortogram revealed narrowing of the abdominal aorta at the level of the first and second lumbar vertebrae, a marked stenosis of the left renal artery,

Fig. 1. Thoracic aortogram showing stenosis of the left subclavian artery, narrowing of the stem of the right common carotid artery, and irregular outline of both the aortic arch and the descending aorta.

Fig. 2. Abdominal aortogram showing irregular outline and narrowing of the abdominal aorta. Note stenosis of the left renal artery.

and irregular outline of the upper half of the abdominal aorta (Fig. 2).

Thus, he was diagnosed as aortitis syndrome and hyperthyroidism and the diagnosis of aortitis syndrome was made from the arteriographic findings although there were no such findings as increase the erythrocyte sedimentation rate, positive CRP and anti-aorta antibody.

DISCUSSION

Not merely autoimmune thyroid diseases (Graves' disease and Hashimoto's thyroiditis) but also aortitis syndrome sometimes are associated with such a autoimmune disease as rheumatoid arthritis or systemic lupus erythematosus. However, the association of aortitis syndrome and autoimmune thy-

roid disease has been reported in only 14 cases previously (Tezuka et al., 1965; Takasu et al., 1962; Ueda et al., 1965; Fujiwara et al., 1969; Eguchi et al., 1972; Kuniyoshi et al., 1973; Kishii et al., 1974; Kito et al., 1974; Yoshimaru et al., 1976; Takagi et al., 1978). As shown in Table, all patients were female, and the age ranged from 20 to 54 years. Five patients had histological findings compatible with Hashimoto's thyroiditis, and five had clinical and laboratory findings which are suggestive of Graves' disease. One patient reported by Kuniyoshi et al. (1973) had systemic lupus erythematosus. Takagi et al. (1978) reported that three cases (13 %) out of 23 patients of aortitis syndrome were associated with autoimmune thyroid diseases and suggested that autoimmune thyroid disease may be a common complication of aortitis syndrome. On the

TABLE
Reported cases of aortitis syndrome associated with autoimmune thyroid diseases

No.	Authors	Year	Age	Sex	Clinical diagnosis	Histological diagnosis	Thyroid antibody
1	Tezuka	1965	26	F	Hyperthyroidism		
2	Takasu et al.	1962	29	F	Hyperthyroidism		
3	Fujiwara et al.	1967	29	F	Myxoedema	Hashimoto's thyroiditis	anti-TG* positive
4	Kuniyoshi et al.	1973	45	F	Myxoedema, SLE		anti-TG* 1 : 10 ⁵
5	Kishii	1974	45	F	Hyperthyroidism		
6	Kito et al.	1974	38	F	Myxoedema	Hashimoto's thyroidism	anti-M** 1 : 10 ⁴
7	Yoshimaru et al.	1976	38	F	Hyperthyroidism		anti-M** positive
8	Takagi et al.	1978	42	F	Euthyroid Graves'	Hashimoto's thyroiditis	anti-TG* 1 : 160 ²
9	Takagi et al.	1978	20	F	Hypothyroidism	Hashimoto's thyroiditis	anti-TG* 1 : 1600
10	Takagi et al.	1978	54	F	Hyperthyroidism	Hashimoto's thyroiditis	anti-TG* 1 : 400

* Anti-thyroglobulin antibody

** Anti-microsomal antibody

other hand, Thomas and Croft (1974) reported five (8.5%) out of 59 patients with giant cell arteritis had a history of thyrotoxicosis and suggested that giant cell arteritis is caused by an abnormal immune mechanism. Although aortitis syndrome differs clinically from giant cell arteritis, there is no clear histological difference between them (Nasu et al, 1968; Ito et al, 1978). Therefore, such association of autoimmune thyroid disease and aortitis syndrome or giant cell arteritis evokes considerable interest and rises question about the possible common immunological mechanisms of these diseases of artery.

The evidence for an autoimmune basis for hyperthyroidism is considerable (Werner et al, 1972). On the other hand, as to the etiology of aortitis syndrome, the most widely recognized opinion is that an autoimmune process is working in its origin (Sandring and Welin, 1961; Judge et al, 1962; McKusich, 1965; Riehl and Brown, 1965). The association of aortitis syndrome and hyperthyroidism may indicate a common origin as autoimmune disorders.

REFERENCES

- EGUCHI, K., FUJIWARA, C., OSHIBUCHI, R., MORISAKI, S. and HASHIBA, K. (1972). Clinical evaluation of Takayasu's aortitis. *J. Jpn. Soc. Intern. Med.* **61**, 1218. (in Japanese).
- FUJIWARA, C., YANO, S. and OSHIBUCHI, R. (1967). A case of Takayasu's arteritis observed in the course of Hashimoto's disease. *Nagasaki Igaku Zasshi*, **42**, 867. (in Japanese).
- ITO, I. (1978). Giant cell arteritis. *Rinshoi*, **4**, 1145. (in Japanese).
- JUDGE, R. D., CURRIER, R. D., GRACIE, W. A. and FIGLEY, M. M. (1962). Takayasu's arteritis and the aortic arch syndrome. *Am. J. Med.* **32**, 379.
- KISHII, T., YAMANAKA, K., SAKURAI, H., FUNAYAMA, M., UEDA, A. and OTSUKA, A. (1974). Aortitis syndrome associated with hyperthyroidism. *J. Jpn. Soc. Intern. Med.* **63**, 584. (in Japanese).
- KITO, C., UENO, T., KURODA, M., TAKEDA, R. and TAKEGOSHI, Y. (1974). A case of aortitis syndrome complicating myxoedema. *Naika*, **34**, 876. (in Japanese).
- KUNIYOSHI, N., SUGITA, Y., TAKEZAWA, H., SHIMOJO, H., SHIINA, Y., SHIMODA, S., SUGISHITA, T., FUKUDA, H., KAN, K., SHICHIJO, K. and ISOMURA, J. (1973). A case of aortitis syndrome associated with systemic lupus erythematosus and hypothyroidism. *Naika*, **31**, 176. (in Japanese).
- McKUSICH, V. A. (1965). A form of vascular disease relatively frequent in the Orient. *Am. Heart J.* **63**, 57.
- NASU, T. (1968). Pathology and pathogenesis of truncoarteritis productiva obliterans. *Nippon Rinsho*, **26**, 3292. (in Japanese).
- RIEHL, J. L. and BROWN, W. J. (1965). Takayasu's arteritis — an autoimmune disease. *Arch. Neurol. (Chic.)* **12**, 92.
- SANDRING, H. and WELIN, G. (1961). Aortic arch syndrome with special reference to rheumatoid arteritis. *Acta Med. Scand.* **170**, 1.
- TAKAGI, Y., OWAKI, T., KATO, H., YANAGIHARA, F., OKUMACHI, F., YOSHIKAWA, J., MORI, T. and TAKAYAMA, H. (1978). Three cases of aortitis syndrome associated with autoimmune thyroid diseases. *Naika*, **42**, 1047. (in Japanese).
- TAKASU, T., NAGATAKI, S., MIURA, Y. and KURAMOTO, K. (1962). Aortic arch syndrome associated with hypertension due to unilateral renal lesion — A clinical case report. *Jpn. Heart. J.* **3**, 183.
- TEZUKA, T. (1956). A case of pulseless disease associated with Graves' disease. *Shinshu Igaku Zasshi*, **5**, 280. (in Japanese).
- THOMAS, R. D. and CROFT, D. N. (1974). Thyrotoxicosis and giant-cell arteritis. *Br. Med. J.* **25**, 408.
- UEDA, H., ITO, I. and SAITO, Y. (1965). Aortitis syndrome. *Naika*, **15**, 239. (in Japanese).
- UEDA, H., SAITO, Y., ITO, I., YAMAGUCHI, H., SUGIURA, M. and MOROOKA, S. (1967). Immunological studies of aortitis syndrome. *Jpn. Heart. J.* **8**, 4.

WERNER, S. C., WEGELIUS, O., FIERER, J. A. and HSU, K. C. (1972). Immunoglobulins (E, M, G) and complement in the connective tissues of the thyroid in Graves's disease. New Engl. J. Med. 287, 421.

YOSHIMARU, R., MATSUO, T., INOUE, J., KATAOKA, K., TAKANO, T. and NAKAGAWA, M. (1976). A case of aortitis syndrome associated with hyperthyroidism. J. Jpn. Soc. Intern. Med. 65, 804. (in Japanese).