

## Airway Obstruction Caused by Substernal Thyrotoxic Multinodular Goiter

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**Background:** Substernal thyrotoxic multinodular goiter (MNG) shows signs and symptoms as a result of compression of adjacent organs and thyrotoxicosis. However, acute airway obstruction is rarely caused by substernal thyrotoxic MNG.

**Case report:** We have described a 56-year-old Japanese woman who demonstrated acute airway obstruction because of compression of the airway by substernal thyrotoxic MNG. She had been diagnosed with substernal thyrotoxic MNG 6 years back. However, because she was unwilling to undergo surgery to remove substernal thyrotoxic MNG, she was treated with methimazole. The patient maintained normal thyroid function with this therapy for 6 years. However, after 6 years the patient was admitted to our hospital because of severe dyspnea. Physical examination revealed inspiratory stridor, which indicated an airway obstruction caused by substernal thyrotoxic MNG. Airway intubation and subtotal thyroidectomy were performed. After the surgery, the dyspnea ameliorated. The general condition of the patient remained good 6 months after the surgery.

**Conclusion:** This case clearly demonstrates the need for careful monitoring of substernal thyrotoxic MNG, because it may lead to an airway obstruction.

**Key words:** thyrotoxic multiple nodular goiter, acute airway obstruction

### INTRODUCTION

Multinodular goiter (MNG) is defined as the enlargement of the thyroid gland, in the absence of autoimmune disease, malignancy, or inflammation. Thyroid function is usually normal in MNG, but subclinical or overt hyperthyroidism can evolve due to autonomously hyperfunctioning nodules of the MNG that is termed thyrotoxic MNG. Substernal MNG is widely defined as a goiter in which the thyroid mass has descended the plane of the thoracic inlet or >50% of the thyroid mass is located below the thoracic inlet [1].

It is possible that substernal MNG may cause symptoms by compressing the adjacent structures including the trachea, esophagus, and superior vena cava [1, 2]. The signs and symptoms of thyrotoxicosis are also observed, when the substernal MNG is thyrotoxic. However, acute airway obstruction leading to severe and life threatening asphyxia rarely occurs [1, 3]. In this report, we describe a patient with a substernal thyrotoxic MNG, which caused acute airway obstruction.

### CASE PRESENTATION

A 56-year-old Japanese female was admitted to our hospital because of dyspnea. Six years prior to the admission, she was referred to the hospital because of MNG. Physical examinations showed a palpable large MNG. Ultrasonographic study revealed that the patient's goiter was substernal MNG. Results of thyroid

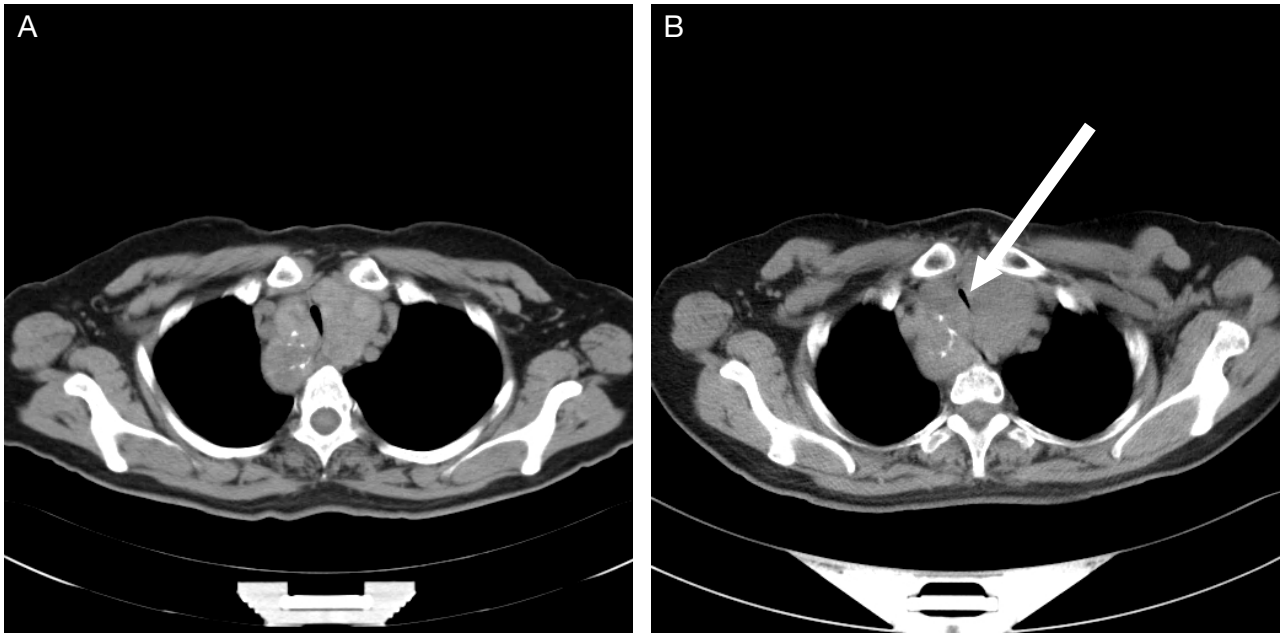
examinations were summarized in Table 1. It showed that thyroid stimulating hormone (TSH) level was completely suppressed (normal range; 0.30-4.00  $\mu$ U/ml), the free triiodothyronine level was 6.23 pg/ml (normal range; 2.50-4.50 pg/ml), and the free thyroxine level was 1.44 ng/dl (normal range; 0.75-1.75 ng/dl). The titers of anti-TSH receptor antibody and thyroid stimulating antibody were 0.0 % (normal range; < 15%) and 128% (normal range; < 180%), respectively. Titers of anti-thyroid peroxidase antibody and anti-thyroglobulin antibody were negative. Computed tomography (CT) scan showed a large MNG that had compressed the trachea (Fig. 1A). Fine needle biopsy of the MNG confirmed benign. A <sup>123</sup>Iodine thyroid uptake imaging study showed an elevated uptake of radioactive iodine in the multiple nodules of the thyroid (Fig. 2). These findings were compatible with substernal thyrotoxic MNG. However, the patient was unwilling to undergo surgery. Therefore, administration of methimazole, 5 mg a day, was started. This administration helped maintain normal thyroid function for 6 years.

One week prior to the admission, she experienced fever and productive cough. Then, the patient was referred to the hospital with progressive dyspnea. On examination, the patient was 160 cm in height and weighed 50 kg. Her blood pressure was 145/95 mmHg, heart rate 100 beats/min, and regular. Respiratory rate was 20 breaths/min. Body temperature was 36.8 °C. Auscultation of the neck revealed significant inspiratory stridor, but auscultation of the chest revealed neither

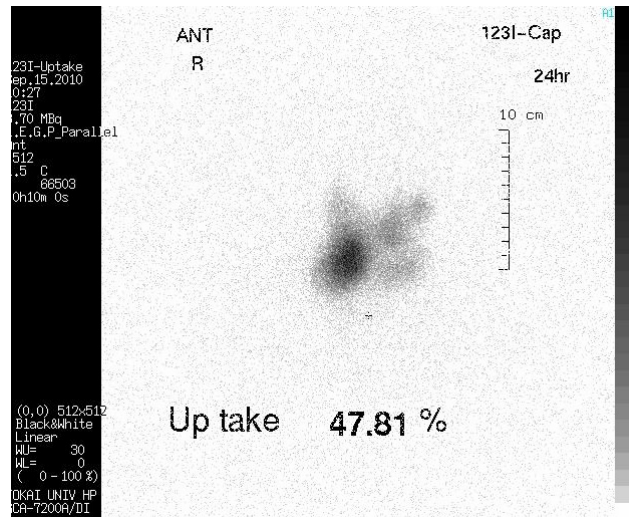
**Table 1**

Thyroid examinations	value	normal range
TSH	< 0.003 $\mu$ U/ml	0.30-4.00
Free T3	4.70 pg/ml	2.50-4.50
Free T4	1.25 ng/dl	0.75-1.75
Anti-TR antibody	0.0 %	< 15
TSAb	128 %	< 180

TSH, thyroid stimulating hormone; T3, triiodothyronine; T4, thyroxine; anti-TR antibody, anti-TSH receptor antibody; TSAb, thyroid stimulating antibody



**Fig. 1** A: CT scan showing the large substernal multinodular goiter compressing and deviating the trachea and narrowing of tracheal lumen.  
 B: Current CT scan showing substernal multinodular goiter that is causing severe increase in tracheal pressure and deviation, compared with the CT findings 6 years back. White arrow indicates the pressured trachea.



**Fig. 2** A  $^{123}$ I-iodine thyroid uptake imaging study showing elevated uptake of radioactive iodine in multiple nodules of the thyroid.

**Table 2**

Complete blood cell counts	value	normal range
White blood cell	8500/ml	4000-8000
Red blood cell	$554 \times 10^4/\mu\text{l}$	380-480
Hemoglobin	16.4 g/dl	11.5-15.5
Hematocrit	49.5 %	34.0-42.0
MCV	89.4 fl	84.0-99.0
MCH	29.6 pg	27.0-32.0
MCHC	33.1 %	32.0-36.0
Platelets	$33.8 \times 10^4/\mu\text{l}$	14.0-40.0

MCV, mean corpuscular volume; MCH mean corpuscular hemoglobin, MCHC, mean corpuscular hemoglobin concentration

Blood chemistry	value	normal range
Albumin	4.7 g/dl	3.9-4.8
Aspartate aminotransferase	21 U/l	< 30
Alanine aminotransferase	15 U/l	< 35
Creatinine	0.50 mg/dl	0.50-0.80
Glucose	148 mg/dl	70-109
Triglyceride	58 mg/dl	50-150
Total cholesterol	270 mg/dl	140-220
Sodium	141 mEq/l	136-145
Potassium	4.1 mEq/l	3.5-4.8
Chloride	102 mEq/l	98-108
C-reactive protein	< 0.09 mg/dl	< 0.3

Arterial blood gas analysis (ambient air)	value	normal range
pH	7.380	7.35-7.45
PaO <sub>2</sub>	98 Torr	75-100
PaCO <sub>2</sub>	42.6 Torr	35-45
HCO <sub>3</sub> <sup>-</sup>	24.6 mEq/l	21-28
SaO <sub>2</sub>	97.9 %	95-99

wheeze nor crackles.

Results of complete blood cell counts, blood chemistry and arterial blood gas analysis were summarized in table 2. Complete blood cell counts revealed elevated levels of red blood cell ( $554 \times 10^4/\mu\text{l}$ ), hemoglobin (16.4 g/dl), and hematocrit (49.5%). Blood chemistry examinations revealed elevated levels of plasma glucose (148 mg/dl) and total cholesterol (270 mg/dl). Arterial blood gas analysis showed pH of 7.380, PaO<sub>2</sub> of 98 Torr, PaCO<sub>2</sub> of 42.6 Torr, HCO<sub>3</sub><sup>-</sup> of 24.6 mEq/l, and SaO<sub>2</sub> of 97.9 % at the ambient air.

CT scan showed the presence of a substernal MNG, which had caused an increase in the tracheal pressure and deviated the trachea. These findings appeared to be more severe than that noted 6 years back (Fig. 1B). The patient was suspected to have asphyxia due to acute airway obstruction; therefore, she was immediately transferred to the intensive care unit. She required airway intubation and ventilation to relieve the asphyxia. The patient underwent a subtotal thyroidectomy on the 10th day of hospitalization. After the surgery, the respiratory condition of the patient remarkably improved. The patient was discharged on 17th day of hospitalization. The patient had no signs and symptoms of airway obstruction recurrence for 6 months after the surgery. She continued to receive the administration of

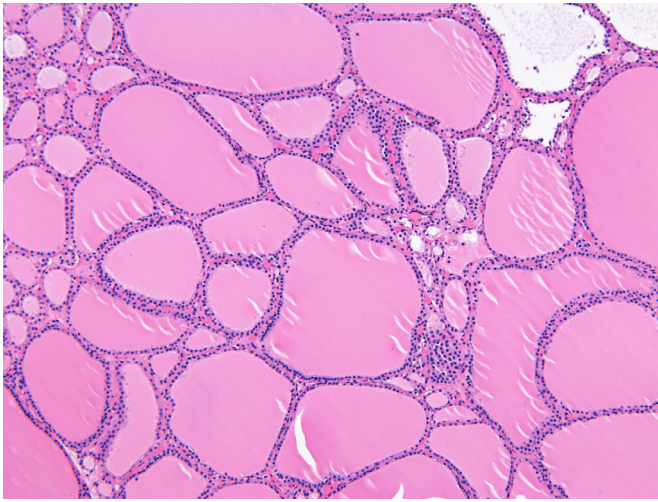
methimazole, 5 mg a day, and showed normal thyroid function.

Hematoxylin and eosin staining of the biopsy tissue showed morphological changes including hyperplasia, colloid accumulation, and nodule formation in the thyroid (Fig. 3). No malignancy was observed. These features were compatible with MNG [4].

## DISCUSSION

The clinical presentation of patients with MNG is variable and depends on the size, location, and functional status of the thyroid. Hyperthyroidism is present in up to 25% of these patients [5]. The thyroid may extend into the thoracic cavity resulting in obstruction or compression of the structures within the cavity. Nonetheless, the incidence of MNG causing airway obstruction is rare [1, 3]. It was suggested that MNG may rarely present with acute airway obstruction requiring emergency intervention, with an occurrence of 0.67 % in a case review [6, 7].

The causes of acute airway obstruction include upper respiratory tract infections resulting in edema and retention of secretions, sudden intrathyroidal hemorrhage, and tracheal stenosis or collapse [8-10]. In the patient, according to the history including fever and productive cough for one week, a respiratory infection



**Fig. 3** Hematoxylin and eosin staining showing a variety of morphological changes including hyperplasia, colloid accumulation, and nodule formation of the thyroid. These features are compatible with MNG. Original magnification  $\times 100$

may have caused acute airway obstruction.

Medical treatment of substernal MNG, including thyroxine or  $^{131}\text{I}$ -iodine administration to decrease the volume of MNG, is not particularly useful [1]. Surgery is the treatment of choice for symptomatic and thyrotoxic MNG [3]. But our patient was asymptomatic and unwilling to undergo surgery. Thus, administration of methimazole continued to maintain normal level of thyroid hormone. While there is no consensus regarding the indications for thyroidectomy in treating MNG, substernal MNG may be treated surgically if it is symptomatic and does not respond to medical treatment, to prevent potential airway obstruction [3]. In cases of airway obstruction, both partial and total thyroidectomy relieved the signs and symptoms [3, 5, 11]. The most common surgical complications associated with thyroidectomy are injury of the recurrent laryngeal nerve, transient or persistent hypocalcemia, and hematoma [1, 3]. Therefore, a subtotal thyroidectomy was performed in our patient to prevent the surgical complications.

The most prominent epidemiologic factor for development of thyrotoxic MNG is iodine deficiency [12, 13]. Japan is a country without iodine fortification because Japanese diet includes regular intake of iodine-rich food such as seaweeds and kelp [14]. Thus, it was unclear that the cause of development of thyrotoxic MNG in the patient.

In conclusion, we describe a patient with substernal thyrotoxic MNG who presented with airway obstruction. This case clearly demonstrates the need for careful monitoring of substernal thyrotoxic MNG, because it may lead to acute airway obstruction.

#### DISCLOSURE

The authors declare that there is no conflict of interests regarding the publication of this article.

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