

# Thyroid Storm and Incidental Anterior Mediastinal Teratoma: Coincidence or Correlation?

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Teratoma is a rare cause of thyrotoxicosis. Among the different types of teratoma, struma ovarii is the main type which contains thyroid tissue. There is no evidence in the literature that would indicate mediasternal teratoma would also lead to thyrotoxicosis or thyroid storm. Herein we report a 37-year-old woman who suffered from palpitation. Her chest X-ray showed a mass lesion at the left hilum, and chest computed tomography scan yielded a suspicion of pericardial cyst. Thereafter, video-assisted thoracoscopic surgery was performed, and thymic cyst was diagnosed during the operation. However, subsequent pathological studies confirmed a diagnosis of mature cystic teratoma. A thyroid function test demonstrating hyperthyroidism was completed prior to the patient's operation, and thyroid storm was diagnosed by clinical presentation. The patient's symptoms did not improve after the operation until we added beta blocker and anti-thyroid agents. Therefore, was the presence of thyroid storm and anterior mediastinal teratoma coincident or correlative in this case? The special stain of teratoma tissues did not reveal any thyroid tissues. In conclusion, thyroid storm and anterior mediastinal teratoma in our case occurred coincidentally. However, a survey of possible hyperthyroidism in patients with anterior mediastinal tumor before operation is critical to avoid perioperative complications.

**Key Words:** Anterior mediastinal tumor • Hyperthyroidism • Teratoma

## INTRODUCTION

Thyroid storm is a life-threatening disease. Surgical intervention can be very dangerous to patients with thyroid storm if good medical control is not obtained. Without treatment, the mortality rate for thyroid storm ranges from 8%~30%.<sup>1,2</sup> Teratoma is an unusual cause of hyperthyroidism;<sup>3</sup> struma ovarii, a teratoma of the ovary

which contains thyroid tissue, has been reported to be associated with hyperthyroidism. Some literature indicates that testicular teratoma may lead to hyperthyroidism.<sup>4</sup> Here we reported a case involving surgical intervention for an anterior mediastinal mass, and thyroid storm was diagnosed after the operation. The pathological study demonstrated mature cystic teratoma. Fortunately, the patient did not have any complications and was well-controlled by beta-blockers and anti-thyroid drugs. However, the question arises whether thyroid storm and anterior mediastinal teratoma are coincident or correlative in our case?

## CASE REPORT

A 37-year-old female without any systemic disease history presented to our family medicine outpatient department complaining of palpitation and fever up to 37.9 °C for 2 weeks. Initially, upper airway infection was

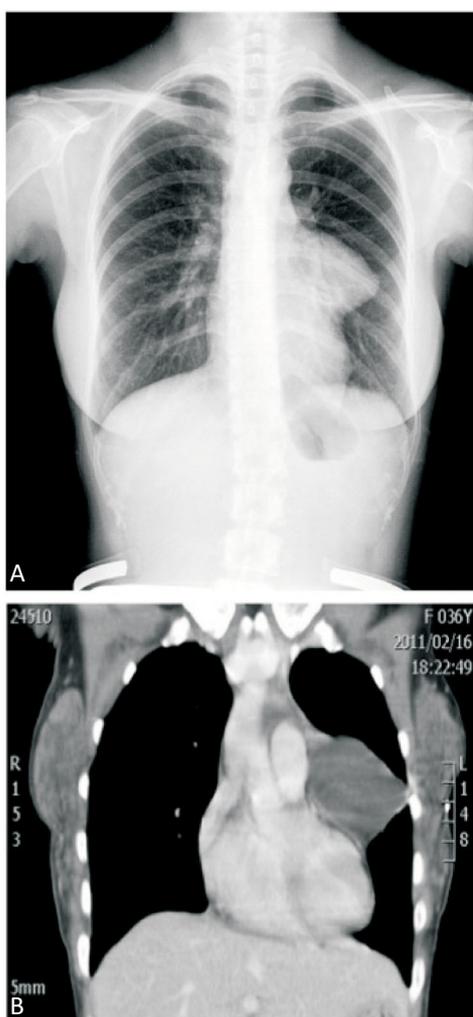
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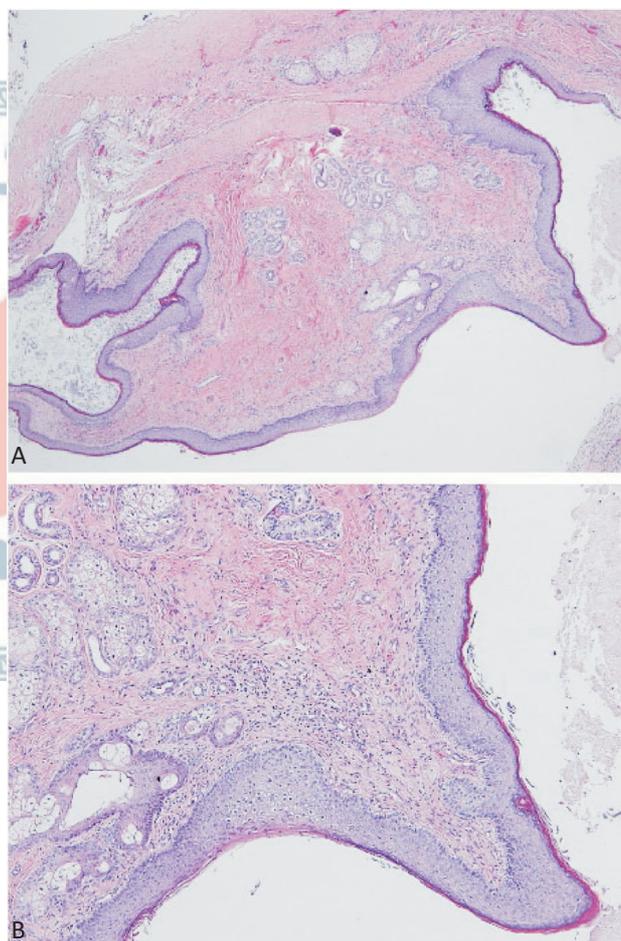
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the first impression of diagnosis. The patient then visited our cardiovascular clinic for further consultation, where she also complained of restlessness, agitation, nausea, vomiting, body weight loss of 2.5 Kg in 3 months, and hand tremor. Physical examination revealed no exophthalmos, neck mass, neck tenderness, paralysis or edema. Application of a 12-lead electrocardiogram showed sinus tachycardia with a heart rate of 123 beats per minute. The patient's chest X-ray displayed a large mass at the left hilum, measuring about 7 cm × 5 cm (Figure 1A). She was transferred to our emergency department for further management. Her body

temperature was normal at the emergency department, with tachycardia and a heart rate of 142 beats per minute, and tachypnea with a respiratory rate of 22 breaths per minute. The patient then underwent contrast-enhanced chest computerized tomography (CT) scan with the suspicion of pericardial cyst (Figure 1B). A chest surgeon thereafter performed video-assisted thoracoscopic surgery (VATS) on the next day. The pathological study (Figure 2) showed a cystic lesion walled by fibrous tissue with necrotic debris, hemosiderin-laden macrophages, cholesterol clefts, chronic inflammation and focal calcification. Epithelial linings including cornified squamous epithelium, ciliated columnar epithelium and simple cuboid to low columnar epithelium (Hematoxylin and eosin stain, 100X). All the above findings make the diagnosis of mature cystic teratoma.



**Figure 1.** (A) The chest X-ray shows a well-defined huge mass at left hilum. (B) Chest computed tomography (CT) scan, sagittal view. The contrast-enhanced chest CT scan shows a 7 × 5 cm well-defined homogenous cystic lesion at left anterior mediastinum. Focal thickening of the cystic wall is noted, and previous inflammatory process can not be excluded. No definite enlarged lymph nodes are noted in the mediastinum.



**Figure 2.** The pathological study. (A) A cystic lesion walled by fibrous tissue with necrotic debris, hemosiderin-laden macrophages, cholesterol clefts, chronic inflammation and focal calcification (Hematoxylin and eosin stain, 40X). (B) Epithelial linings include cornified squamous epithelium, ciliated columnar epithelium and simple cuboid to low columnar epithelium (Hematoxylin and eosin stain, 100X). All the above findings make the diagnosis of mature cystic teratoma.

luminal epithelium, and simple cuboid to low columnar epithelium were also found. Thereafter, a confirmed diagnosis of mature cystic teratoma was made.

The symptoms of fever, palpitation, agitation and hand tremor did not manifestly improve after the operation. The results of thyroid function tests which were obtained before the operation showed thyrotoxicosis with a low thyroid stimulating hormone level of 0.006 uIU/ml and high free-T4 level of 4.2 ng/dl. By using the point system developed by Burch and Wartofsky, this patient was at 55 points.<sup>5</sup> Thyroid storm was highly suspected. After an endocrinology consultation was conducted, beta blocker (propranolol 10 mg twice per day) was used, and her symptoms relieved gradually over the next 3 days. She was discharged without any complications and had regular follow-up in the endocrinology outpatient department. Further study of thyrotoxicosis showed high TSH receptor antibody (60%) and negative anti-thyroglobulin antibody. Graves' disease was diagnosed, and an anti-thyroid agent (methimazole, 30 mg per day) was given. Her thyroid function test returned to normal range 2 months later. For further confirmation of the relationship between thyroid storm and teratoma, we performed a special stain of the teratoma tissues and did not find any thyroid tissues.

## DISCUSSION

Anterior mediastinal mass can be divided into two groups by pathology: neoplasm and non-neoplasm. Neoplasm includes thymoma, thymic carcinoma, thymic carcinoid, thymolipoma, germ cell tumors, and parathyroid adenoma. The non-neoplasm group includes thymic cyst, lymphangioma, and intrathoracic goiter.<sup>1,3</sup> Among anterior mediastinal masses, thymoma is the most common tumor. Germ cell tumor accounts for 10-15% of adult anterior mediastinal tumor, and teratoma is the most common form of mediastinal germ cell tumor. Radiologically, a benign teratoma is a well-defined mass,<sup>6,7</sup> and it can be round or lobulated. Additionally, up to 26% of teratoma has calcification. Thymic cyst is a rare cause of anterior mediastinal mass, and it only accounts for 3%.<sup>6,7</sup> The typical manifestation of thymic cyst is a well-circumscribed homogenous cyst, located at the anterior superior mediastinum.

Thyroid storm is known as a critical illness. Although the incidence of patients with thyrotoxicosis developing thyroid storm is low, thyroid storm itself has a high mortality rate. There are many causes of thyrotoxicosis. The most common cause is Graves' disease, and it also the main cause of thyroid storm in patient with thyrotoxicosis.<sup>2</sup> Other causes of thyroid storm include toxic adenoma, thyrotropin-secreting pituitary adenoma, thyroid carcinoma and trophoblastic disease. Several reports in the literature had reported teratoma causing hyperthyroidism. Teratoma, which may consist of thyroid tissue, can also be the cause of hyperthyroidism. Struma ovarii, an ovarian teratoma, has been acknowledged as a rare cause of thyroid storm. Testicular teratoma also has been reported to be associated with thyrotoxicosis. However, there is no existing evidence supporting the proposition that anterior mediastinal teratoma also causes hyperthyroidism. But the possibility cannot be completely excluded.

The risk is also elevated if a patient that has thyroid storm is undergoing surgery. Mortality rate up to 8%-30% is reported before.<sup>1,2</sup> According to the guidelines of American Thyroid Association, patients with hyperthyroidism have to be rendered euthyroid by drugs, such as methimazole and beta-adrenergic blockade, before the operation.<sup>2</sup> If the surgery is emergent, oral cholecystographic agent and corticosteroid are also needed.

Due to the high mortality and complication rate of thyroid storm, hyperthyroidism should be under firm control before the operation is done. In our case, the cardiologist, radiologist and chest surgeon all focused on the chest tumor and the CT scan did not favor thyroid disease. Therefore, the surgery was performed even before the diagnosis of thyroid storm. Fortunately, the surgery was done uneventfully. The pathological studies finally confirmed the diagnosis of mature cystic teratoma. Are thyroid storm and anterior mediastinal teratoma coincident or correlative in our case? We did perform a special stain of the teratoma tissues, yet did not find any thyroid tissues. Therefore, thyroid storm and anterior mediastinal teratoma should be just a coincidence.

We have presented this case to serve as a reminder for physicians managing such patients with an anterior mediastinal tumor. For those patients undergoing sur-

gery, the physicians should keep our example in mind for evaluating any possibilities of hyperthyroidism. Thyroid function and the symptoms of thyroid storm also should be under careful medical control prior to the operation.

## REFERENCES

1. Sarlis NJ, Gourgiotis L. Thyroid emergencies. *Rev Endocr Metab Disord* 2003;4:129-36.
2. Nayak B, Burman K. Thyrotoxicosis and thyroid storm. *Endocrinol Metab Clin North Am* 2006;35:663-86i.
3. Mitra ES, Niederkohr RD, Rodriguez C, et al. Uncommon causes of thyrotoxicosis. *J Nucl Med* 2008;49:265-78.
4. Stromberg P, Ratcliffe JG, Trotter J, et al. Teratoma of testis and hyperthyroidism. *Scott Med J* 1981;26:346-7.
5. Burch HB, Wartofsky L. Life-threatening thyrotoxicosis. Thyroid storm. *Endocrinol Metab Clin North Am* 1993;22:263-77.
6. Duwe BV, Sterman DH, Musani AI. Tumors of the mediastinum. *Chest* 2005;128:2893-909.
7. Strollo DC, Rosado de Christenson ML, Jett JR. Primary mediastinal tumors. Part 1: tumors of the anterior mediastinum. *Chest* 1997;112:511-22.

