Critical airway compression caused by a large mediastinal tumour with spontaneous haemorrhage

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Abstract
We report the case of a 77-year-old woman presenting with out-of-hospital cardiac arrest, which was then interpreted as an acute, life-threatening critical airway compression by a huge mediastinal tumour without appropriate diagnosis. Emergency extracorporeal membrane oxygenation was cannulated for sufficient respiratory support after spontaneous circulation was regained. After the multidisciplinary team, involving thoracic surgeons, discussed the resectability of the mediastinal tumour, the patient underwent successful resection of the mediastinal tumour through a median sternotomy. The pathological report demonstrated an intrathoracic goitre with spontaneous haemorrhage and haematoma formation, and the patient was discharged with favourable respiratory and neurological outcomes.

Introduction
Critical airway obstruction is a dreaded complication of a large mediastinal mass, especially for patients presenting with acute collapse without significant past history or subjective airway-related symptoms before the episode. The pathogenesis of such a mediastinal mass could be caused by a heterogeneous group of mediastinal pathologies, encompassing surgical or non-surgical management. Spontaneous haemorrhage of substernal goitre resulting in tracheal compression is rare, and the mechanism causing it is still unclear. Some clear precipitating factors have been reported, such as taking anticoagulation medicine, trauma, coincident upper respiratory tract infection [1], and actions that increase intravascular pressure, like coughing, choking on food, straining at defecation, and the valsalva manoeuvre [2]. Thoracic surgeons should be aware of this critical scenario and thoroughly differentiate this from other surgical or non-surgical mediastinal masses.

Case Report
A 77-year-old woman with a known history of thyroid goitre without regular follow up was found lying on the floor of her dining room after choking on a candy. Her daughter-in-law performed cardiopulmonary resuscitation promptly. The emergency medical services providers arrived quickly, and she was brought to our emergency room (ER) at a territory medical centre. Her Glasgow Coma Scale was E1V1M1, and she had no spontaneous breathing and heart beats initially. The advanced cardiac life support protocol was started immediately. She received endotracheal intubation and mechanical ventilation. A portable chest X-ray showed a huge mass lesion at the mediastinum with mass effect, which compressed the trachea and resulted in severe airway deviation (Fig. 1A).

Tracing her medical history according to her family members, she did not complain of any respiratory symptoms from her daily activities. Further chest computed tomography (CT) demonstrated a huge heterogeneous mass at the mediastinum, suspected to have originated from the left

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lobe of the thyroid gland, with a mass effect with compression on the tracheobronchus and oesophagus with mixed density and small contrast pooling, indicative to be spontaneous haemorrhage and haematoma formation (Fig. 1B, C). Although spontaneous circulation returned after cardiopulmonary resuscitation at ER, low tidal volume, high airway pressure, and persistent hypoxemia persisted even with 100% oxygen supplied. Venovenous-extracorporeal membrane oxygenation (VV-ECMO) was then cannulated. The patient was then transferred to our respiratory intensive care unit (RICU) for further care. Laboratory data obtained at ER and RICU showed results in the normal range for the thyroid function test, but only elevated lactate dehydrogenase (LDH) level (420 U/L; range 131–250) was noted. At the RICU, tidal volume was low, and intermittent Ambu-bagging was still needed. Considering the possibility of tumour resection and to relieve the airway compression, without preoperative tissue proof, we performed a median sternotomy for tumour excision on the following day after improvement of conscious level (E2VtM3). Intraoperative findings revealed a heterogeneous huge mass, measuring 13.5 × 6.0 × 4.0 cm, with extension along the left thyroid goitre and the left trachea until the left main bronchus and extending over the subcarinal area. The pathological report confirmed the diagnosis of nodular goitre with cystic degeneration, with central haemorrhage and a large amount of blood clots (Fig. 2). The patient tolerated the operation well and was weaned off both the VV-ECMO and the mechanical ventilator successfully. She was then discharged uneventfully with favourable respiratory and neurological outcomes.

**Discussion**

As members of a multidisciplinary team for the management of a large mediastinal mass with critical airway compression, thoracic surgeons play a crucial role in handling such a critical scenario, including airway maintenance (stent implantation or extracorporeal circulation cannulation), obtaining abundant tissue amounts for precise pathological diagnosis through a surgical biopsy if fine-needle biopsy fails (perhaps through an anterior mediastinotomy;...
Chamberlain procedure or video-assisted thoracoscopic surgery), especially for an unresectable tumour before further chemotherapy or radiotherapy, or even to make a proper decision to perform direct surgical removal of such a huge mass to release external airway compression for a resectable tumour in our presented case.

Spontaneous haemorrhage of benign thyroid gland resulting in tracheal compression is well reported in the literature, but most are reported in cervical goitres [2]. When this occurs, patients may need an emergency intubation to maintain the patency of the airway. Gauger et al. reported three (0.11%) patients needing emergency intubation before being transferred to the operating room in a series of 2592 patients scheduled for thyroidectomy [3]. When these scenarios occur, clinicians usually easily misdiagnose due to patients’ previous thyroid goitre histories and related respiratory symptomatic complaints.

On the other hand, substernal goitre itself is reported as a risk factor for developing acute, critical airway compression [4]. Interestingly, in the literature, we found only one case report that demonstrated spontaneous haemorrhage of substernal goitre. Kokatnur and colleagues reported a 73-year-old female with a history of arterial fibrillation on warfarin therapy, whose international normalized ratio was supra-therapeutic at 3, presented with worsening respiratory distress that warranted emergency intubation due to haemorrhage in the thyroid gland with substernal extension [5]. However, in our case, our patient has no underlying disease that may cause platelet dysfunction or coagulopathy, and she was not on any anticoagulant. Spontaneous haematoma developed after choking on a candy, which might be caused by high pressure to the goitre veins, resulting in extravasation of blood into the goitre and in a rapidly expanding haematoma [2], and sequentially followed by inducing acute airway obstruction and out-of-hospital cardiac arrest, which is extremely rare. Luckily, ECMO cannulation temporarily supported the respiratory function to obtain more time to clarify the aetiology of the mediastinal mass and to decide on the appropriate further treatment.

Tracing the history of our patient at out-patient departments after the operation, we found that she was diagnosed with asymptomatic substernal goitre (Fig. 1D: not available during hospitalization) when she was 67 years old. Observation only without surgical removal was recommended by her family doctor at that time. In our opinion, substernal goitres typically grow slowly; therefore, symptoms are generally not clinically apparent until the goitre compresses the airway or other adjacent structures. Therefore, according to the case we presented here and the literature review, we advocate that substernal goitre, especially for those extending below the level of the brachiocephalic vein, should be considered a risk factor for developing acute, critical airway compression, especially in women in their 50s or 60s [4]. Early surgical removal should be recommended if possible when patients’ medical conditions match the description.

In conclusion, although acute, life-threatening presentation of airway compression by a spontaneous haemorrhage substernal goitre is rare, thoracic specialists should be aware of this critical scenario and thoroughly differentiate this from other surgical or non-surgical mediastinal masses. The ECMO for respiratory support followed by aggressive surgical decompression could be life-saving, with favourable outcomes. In addition, surgical resection of the substernal goitres should be recommended after early recognition if the patient’s medical conditions are appropriate.

Disclosure Statement
Appropriate written informed consent was obtained for publication of this case report and accompanying images.

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