CASE REPORT

Acute respiratory failure in a rapidly enlarging benign cervical goitre

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SUMMARY

Benign goitres have the potential to reach massive sizes if neglected, but most have a protracted course that may or may not present with compressive symptoms. We report the case of a 57-year-old man who presented with a rapidly enlarging nodular goitre resulting in acute respiratory failure. Endotracheal intubation and emergency total thyroidectomy were performed, revealing massive thyroid nodules with minimal intrathoracic extension and tracheal erosion. Despite a course and clinical findings suggestive of malignant disease, histopathology was consistent with a benign multinodular goitre. Several cases of benign goitres necessitating endotracheal intubation have been reported. Airway compromise was attributed to a significant intrathoracic component, or inciting events such as thyroid haemorrhage, pregnancy, radioiodine uptake or major surgery. Obstructive symptoms may not correlate well with objective measures of upper airway obstruction such as radiographs or flow volume loops.

BACKGROUND

The differential diagnoses of massive, rapidly growing anterior neck masses are mainly malignant tumours such as thyroid carcinomas or lymphomas. This is an uncommon case of a patient presenting with a history and clinical findings suggestive of malignancy, but revealed a benign goitre on histological examination. Though rare, the possibility of rapid airway compromise should be included in the factors in deciding surgery in long-standing cases of cervical goitres.

CASE PRESENTATION

A 57-year-old man was transferred to our institution from another local hospital after being admitted for severe respiratory distress associated with a massive goitre. Review of history revealed a 2-year history of nodular goitre, initially grape-sized on either lobe with no hyperthyroid or hypothyroid symptoms. Initial consult was performed in a private clinic where thyroid function tests ordered showed normal results. The patient was advised to undergo fine-needle aspiration biopsy but was lost to follow-up. At around 3 months prior to admission, the patient noted rapid increase in the goitre size but took only local herbal remedies. At 2 weeks from admission, he began having occasional episodes of aspiration during meals, exertional dyspnoea and orthopnoea. Symptoms progressed until 3 h prior to admission, the patient had a severe coughing fit with persistent choking sensation and lost consciousness. He was then brought to a local

hospital, found to be stuporous and cyanotic, and was immediately intubated and placed on mechanical ventilation.

The patient had an unremarkable medical history, and had no family history of thyroid malignancies or goitres.

INVESTIGATIONS

On admission, the patient was still intubated but with stable vital signs and an intact sensorium. Pertinent physical examination findings included a markedly large nodular goitre, with the right lobe larger than the left (10×6 cm), without any cervical or axillary lymphadenopathies; bibasal crackles were heard on chest auscultation. Thyroid function tests (free T4 and thyroid stimulating hormone (TSH)) were normal; a review of the neck and chest CT imaging revealed a small intrathoracic extension of the right thyroid mass (figure 1). Fine needle aspiration biopsies were performed while awaiting surgical intervention, which were non-diagnostic (category I, Bethesda classification).

DIFFERENTIAL DIAGNOSIS

With the aforementioned history and presentation, primary consideration prior to surgery was a

Figure 1 Neck and chest CT scan with intravenous contrast, coronal view, taken preoperatively, showing marked cystic enlargement of the thyroid lobes, more prominent on the right. An endotracheal tube is seen in place with an inflated cuff.

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thyroid malignancy; with the patient's male gender, massive and relatively rapid increase in goitre size and severe airway compromise.

TREATMENT

The patient underwent total thyroidectomy, with thoracocardio-vascular surgeons on standby for possible intrathoracic extension of the surgery.

Intraoperatively, the right thyroid lobe was noted to have been transformed into a large 15×14×10 cm cystic mass with an 8×8×8 cm intrathoracic component or extension. The left thyroid lobe had a 2×2×2 cm cystic component. Aspiration of the right cystic mass was performed to facilitate mobilisation and exposure and obtained ~400 cc of brownish serous fluid. During mobilisation, a 0.5 cm vertical laceration in the dehiscence part of the right tracheal thyroid bed was noted and repaired. The parathyroid glands and recurrent laryngeal nerve of both sides were identified and preserved.

OUTCOME AND FOLLOW-UP

His intraoperative course was unremarkable. He was weaned off mechanical ventilation and extubated on the third postoperative day. Laryngeal videostroboscopy was performed, which revealed bilateral vocal cord paresis, with granulation tissue over the true cords and oedematous false cords. The patient was discharged on the eight postoperative day after completion of treatment for pneumonia, and was primed on the possibility of radioiodine ablation therapy once the biopsy confirmed a malignant tumour. Surprisingly, histopathology of the thyroid masses was consistent with a multinodular colloid goitre involving both lobes (figure 2).

On follow-up, the patient had complete resolution of compressive symptoms and hoarseness (figure 3). He was started on thyroid hormone replacement with levothyroxine at 1.6 µg/kg/day, and was advised regular TSH monitoring for titration of levothyroxine dose. Repeat neck ultrasonography after 2 months showed no remnant thyroid tissue and no cervical lymphadenopathies; while repeat videostroboscopy revealed fully mobile vocal cords with resolution of oedema.

DISCUSSION

Goitres are among the most common diagnoses in patients consulting at our institution's otorhinolaryngology and endocrinology outpatient clinics. Prevalence rates among populations vary widely probably due to differences in iodine sufficiency,

genetics and even the method of diagnosis used. A systematic review by Korwal *et al*¹ summarises the total goitre prevalence among several regions at 15.8% in 2004, ranging from 4.7% in the Americas to as high as 37.3% in the Eastern Mediterranean. Local Philippine data obtained from a nationwide survey by Carlos-Raboca *et al*² among 7227 non-pregnant adults show a prevalence rate of 8.9% based on WHO-ICCD criteria by palpation.

In such patients with goitres, several factors suggestive of a malignancy have been identified,³ including men, rapidly enlarging nodule size and severe compressive symptoms which were present in this particular case. In a review by Kitamura *et al*⁴ of 161 fatal cases of thyroid malignancy, respiratory insufficiency was the most common fatal condition occurring in 46 (43%) of those with specific causes of death. This was attributed to extensive pulmonary metastases, pneumonia, laryngeal and tracheal stenosis, and recurrent nerve palsy due to tumour invasion. Airway obstruction, described separately as being due to vocal cord oedema, occurred in 70 (44%) of the total number of mortalities. Fourteen were described as having died of asphyxia. Another case-matched control study on 56 patients focusing on fatal papillary thyroid carcinoma in the Mayo Clinic⁵ identified pulmonary and local causes as the top causes of death in this cohort (37% and 36% of all mortalities, respectively).

On the other hand, for obstructive symptoms in benign goitres, Shaha *et al*⁶ reported that 28% of patients with benign thyroid disease presented with tracheo-oesophageal compression (112 out of 300 cases). Among this number, only 20 (0.67%) were described as presenting with acute airway problems requiring emergency intervention, attributed to either severe luminal compression, intrathyroidal haemorrhage or upper respiratory infection. In a retrospective review of 813 cases of total thyroidectomy in India by Agarwal *et al*,⁷ data analysis included a comparison between malignant and benign goitres with regard to respiratory distress. They noted a significantly higher rate of respiratory distress in malignant goitres among those weighing ≤400 g (5.3% vs 1.9%, $p=0.013$), and a slight but insignificantly increased rate among those over 400 g (40% vs 20%, $p=0.15$). They also compared patients with severe respiratory distress, though the criteria for this qualification were not described in the methodology. For severe respiratory distress, rates were similar between malignant and benign goitres; both

Figure 2 Biopsy specimen of the thyroid lobes, ×100 total magnification, showing the thyroid follicles lined by a single layer of cells with eosinophilic cytoplasm.

Figure 3 Photograph of the patient's neck, anterior view, taken 2 months postoperative, showing a healing transverse neck scar with no palpable neck masses or lymphadenopathies.

for those ≤ 400 g (15.8% vs 13.3%, $p=0.39$) and those >400 g (53.3% vs 46.7%, $p=0.76$).

A literature search for detailed case reports of acute airway obstruction in benign nodular goitres revealed only a handful of cases, using the search terms 'airway obstruction', 'respiratory failure', 'benign goitre' and 'nodular non-toxic goitre' in Pubmed.

Warren *et al*⁸ in 1979 described three cases of life-threatening respiratory failure in large goitres. All cases were elderly patients; one expired prior to surgery.

Sajja *et al*⁹ reported a case of intrathyroidal haemorrhage causing respiratory failure shortly after cardiac valve replacement surgery in a 60-year-old woman.

Sharma *et al*¹⁰ also reported acute respiratory failure in a 64-year-old woman with a long-standing goitre. This required emergency total thyroidectomy and histopathology revealed a benign multinodular goitre with minimal intrathoracic extension but no compression of the retrosternal trachea.

Testini *et al*¹¹ described six cases of goitres presenting with acute airway obstruction out of a total of 919 patients who underwent thyroidectomy. However, only two of these (0.2%) were confirmed to be benign by histology, one of which was noted to have had massive parenchymal haemorrhage.

Abraham *et al*¹² reported five cases of acute airway obstruction out of 1115 patients with benign goitres, resulting in a prevalence of 0.6%. These cases were all women in the elderly age range with chronic comorbidities, and estimated gland weights ranging from 90 to 435 g.

Several studies have looked into possible predictors for airway obstruction in patients with goitres. Pradeep *et al*¹³ studied 71 patients consulting for asymptomatic goitres with mean estimated gland weights of 126.4 and 150.3 g for women and men, respectively. Preoperative pulmonary function tests (PFTs) were performed showing significantly lower vital capacity, FVC and FEV1 than predicted; despite the lack of reported symptoms. Menon *et al*¹⁴ compared 18 patients with symptomatic goitres to 38 cases without symptoms; using clinical parameters, radiological assessment with CT and PFTs. They found no significant correlation between obstructive symptoms, airway obstruction measured by PFTs, and tracheal narrowing measured by CT scans. Daykin *et al*¹⁵ also studied 153 patients with goitres, and showed upper airway obstruction by flow volume loops in 33% of the cohort. When comparing those with obstructive symptoms to those without, they found no significant difference between the two groups in terms of clinical history and type of goitre. However, they noted a significantly higher number of patients with airway obstruction in those showing tracheal deviation (63% vs 44%) and tracheal compression (23% vs 5%) in plain radiographs of the thoracic inlet.

This case illustrates that though severe compressive symptoms in goitres strongly suggest the presence of a malignant process, this can still occur—though uncommonly—in benign goitres. Contributory factors in this case may have been the goitre's relatively rapid enlargement, presence of an intrathoracic extension and perhaps an undocumented respiratory infection prior to airway compromise. Definitive histopathological confirmation should be obtained prior to final prognostication and to guide long-term management. As for patients with long-standing cervical goitres, clinical history including obstructive symptoms may not correlate well with more objective measures of upper airway obstruction including flow volume loops and radiographic assessment (radiograph or CT scan). Prospective trials may be needed to determine the role of routine assessment of upper airway obstruction in patients with chronic benign goitres.

Learning points

- ▶ Clinical factors in a cervical goitre suggestive of a malignancy include men, rapidly enlarging nodule size and severe compressive symptoms.
- ▶ Respiratory insufficiency is a common specific cause of death in malignant goitres attributed to extensive pulmonary metastases, pneumonia, laryngeal and tracheal stenosis and recurrent nerve palsy due to tumour invasion.
- ▶ Benign goitres may rarely present with acute airway problems requiring emergency intervention, with an occurrence of 0.67% in a case review.
- ▶ Upper airway obstructive symptoms and clinical history may not correlate well with objective measures of airway obstruction (pulmonary function tests or radiological assessment).

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Contributors The case was admitted under the primary service of otorhinolaryngology with RG as senior resident in charge; and comanaged by CJG of the endocrinology service with CA-C as supervising consultant. All the authors have significantly contributed to the patient's care and to the writing of this manuscript to assume authorship.

Competing interests None.

Patient consent Obtained.

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