

Life-threatening airway obstruction by Riedel's thyroiditis: a rare presentation and diagnostic dilemma

Maria Leonor Guia Lopes¹, José Pedro Cidade^{2,3}, Clara Cunha¹, Clotilde Limbert¹ and João Sequeira Duarte¹

¹Endocrinology Department, Hospital Egas Moniz, CHLO, Lisbon, Portugal

²Intensive Care Unit 4, Intensive Care Department, Hospital São Francisco Xavier, CHLO, Lisbon, Portugal

³Nova Medical School, Clinical Medicine, New University of Lisbon, Lisbon, Portugal

Correspondence should be addressed to M Guia Lopes: mlolopes@ulslo.min-saude.pt

Summary

Riedel's thyroiditis is the rarest form of thyroiditis, occasionally resulting in rapid thyroid enlargement and potential tracheal obstruction. Here, we detail the case of an 81-year-old woman with a medical history including Hodgkin lymphoma, Hashimoto's thyroiditis, and multinodular goiter. She presented to the emergency room with stridor, cervical swelling, and breathing difficulties for over 2 days. CT scans revealed substantial thyroid enlargement causing significant glottal and tracheal compression, to a minimum tracheal diameter of 7 mm. Due to the severity of the compressive symptoms, orotracheal intubation and mechanical ventilation were deemed necessary. Surprisingly, despite the initial suspicion of malignancy given the rapid growth in the elderly, subsequent cytological and histological evaluations indicated a benign form of invasive fibrous thyroiditis – Riedel's thyroiditis. Although surgical intervention was advised, the patient declined and opted for endobronchial treatment with a prosthetic stent and subsequent treatment with systemic glucocorticoids. Following successful treatment, she was discharged within a week and resumed normal activities without respiratory distress. This case is noteworthy for its rapid benign mass growth, rare emergent presentation, and the patient's advanced age.

Learning points

- The rapid enlargement of the thyroid gland in elderly patients poses a diagnostic challenge, stemming from the higher occurrence of aggressive thyroid carcinomas.
- Despite the clinical presentation, a comprehensive diagnostic workup, including fine-needle aspiration and core-needle biopsy, is crucial for accurately distinguishing between benign and malignant causes of thyroid nodule enlargement.
- This case report illustrates diverse treatment options for Riedel's thyroiditis, and the importance of individualized treatment plans based on the degree of airway obstruction, patient preferences, and response to initial interventions.
- Clinicians should contemplate the inclusion of glucocorticoids in the therapeutic regimen for Riedel's thyroiditis, particularly in cases where surgical intervention is not feasible or declined by the patient.

Background

Airway obstruction (AO) encompasses a range of conditions, including upper AO from the nose or mouth to the larynx, and lower AO affecting the tracheobronchial tree (1). AO can be categorized as intrinsic, stemming from the airway epithelium, or extrinsic, resulting from compression by neighboring cervical and thoracic structures such as organs, vessels, and lymph nodes, or due to factors like foreign bodies, trauma, surgery, or tumors (2). The severity of AO is determined by the extent of anatomical occlusion, potentially compromising ventilation and posing a significant risk of acute respiratory failure and, in some cases, death (3). These acute episodes are considered life-threatening emergencies necessitating immediate assessment and intervention, as prompt restoration of airway patency is the sole effective treatment. Mortality rates in such clinical scenarios have been reported to approach nearly 50% in some series (4).

Riedel's thyroiditis, also known as invasive fibrous thyroiditis, represents the rarest form of thyroiditis with an unclear etiology (5). Typically diagnosed between ages 20 and 50, it involves progressive fibrous parenchymal growth in the thyroid, where the thyroid tissue gradually transforms into fibrous connective tissue, often evolving over months or years (5). In rare instances, rapid enlargement of the thyroid gland may lead to compression of cervical and thoracic structures such as the larynx and trachea, mimicking other conditions such as thyroid lymphoma or anaplastic thyroid cancer (6).

Here, we present the case of an 81-year-old woman with acute AO resulting in significant respiratory insufficiency, attributed to compressive Riedel's thyroiditis.

Case presentation

We present the case of an 81-year-old woman with a medical history notable for Hodgkin lymphoma and Hashimoto's thyroiditis, leading to clinical hypothyroidism diagnosed in 2015. Hypothyroidism was managed with a daily dose of 75 µg of levothyroxine, with regular follow-up in the endocrinology outpatient clinics.

The patient presented to the emergency department exhibiting symptoms of stridor, cervical swelling, and respiratory distress persisting for 48 h. Initial clinical assessment revealed severe peripheral desaturation, requiring non-invasive ventilation and high levels of oxygen therapy to marginally restore adequate oxygen saturation. Additionally, a cervical mass exceeding 5 cm in diameter was identified upon palpation, characterized by a bulky and firm texture. Laboratory data at the time of admission to the emergency department are detailed in Table 1.

Table 1 Laboratory data at the admission to the emergency department.

Laboratory test	Result	Reference
Hemoglobin (g/dL)	12.5	12.0–15.0
Mean corpuscular volume (fL)	90.2	80.0–96.1
White blood cells ($\times 10^9/L$)	12.0	4.0–10.0
Platelets ($10^9/L$)	164	150–400
Sodium (mmol/L)	138	136–145
Potassium (mmol/L)	4.1	3.5–5.1
Phosphate (mg/dL)	3.7	2.5–4.5
Magnesium (mg/dL)	1.8	1.7–2.2
Calcium (mg/dL)	9.2	8.6–10.0
Albumin (g/L)	3.7	3.5–5.2
Creatinine (mg/dL)	0.87	0.5–0.9
Glomerular filtration rate (mL/min/1.73 m ²)	68	> 60
Thyroid-stimulating hormone (µUI/mL)	0.290	0.270–4.200
Free thyroxine (T4) (pmol/L)	21.5	12.0–22.0
C-reactive protein (mg/dL)	1.5	< 0.1
Sedimentation rate (mm/h)	36	< 10

Emergency cervical and thoracic CT revealed a substantial increase in thyroid volume, resulting in significant laryngeal and tracheal compression, with a minimum tracheal diameter of 7 mm (Fig. 1). Due to the severity of the compressive symptoms and the risk of AO, orotracheal intubation, and mechanical ventilation were deemed necessary. However, a situation of 'cannot intubate, cannot ventilate' was observed during the intubation attempt, resulting in the need to use a frozen airway intubating introducer to successfully insert a low-diameter tube (5.5 mm). The patient was then admitted to the intensive care unit with the diagnosis of upper AO.

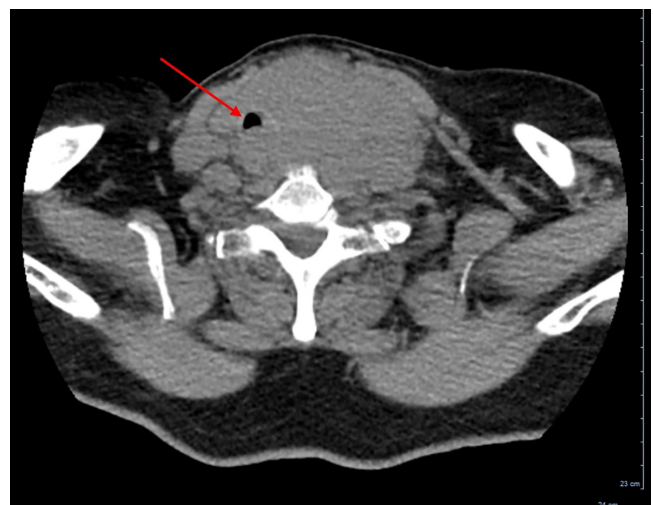
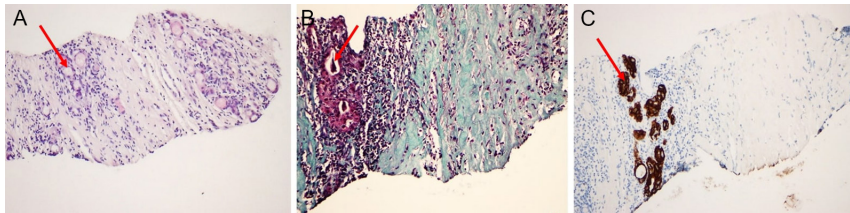


Figure 1

Cervical CT scan on admission to the emergency department. The red arrow indicates the minimal tracheal lumen of 7 mm.

**Figure 2**

Core-needle biopsy histopathological exam showing residual thyroid follicles devoid of malignant nuclear features (red arrows), surrounded by lymphocytes encased in abundant hypocellular fibrosis with few maintained vessels. (A) Hematoxylin and eosin; (B) Masson trichrome stain; and (C) cytokeratin AE1/AE3 immunostaining.

secondary to rapid thyroid growth. Mechanical ventilation proceeded smoothly with high compliance and low resistance values upon lung mechanic evaluation.

Investigation

Given the patient's age, exuberant clinical presentation, and medical history, suspicion for malignancy, including anaplastic thyroid carcinoma, thyroid lymphoma, or metastasis to the thyroid from another primary tumor, was high. Fine-needle aspiration (FNA) supported by ultrasound was performed on a section of the thyroid parenchyma described as multinodular, confluent, markedly hypoechogenic, and lacking vascularization. Contrary to initial clinical impressions of possible malignancy, cytology revealed the presence of CD3+, CD20+, and CD68+ cells (inflammatory pattern) without cytopathological signs of malignancy. To accurately confirm benignity, a core-needle thyroid biopsy was performed. Histological findings revealed a benign pattern characterized by residual clusters of thyroid follicles devoid of nuclear atypia or oncocyctic metaplasia, surrounded by abundant lymphocytes encased in hypocellular fibrosis with few maintained vessels (Fig. 2), consistent with the diagnosis of Riedel's thyroiditis.

Treatment

Considering the severity of AO, surgical intervention was proposed, but the patient declined. Endotracheal treatment with a prosthetic stent was performed to restore tracheal lumen diameter, ensuring safe extubation and resolution of compressive symptoms. Additionally, the patient underwent treatment with systemic glucocorticoids (40 mg of prednisolone for 3 months), followed by a slow tapering.

Outcome and follow-up

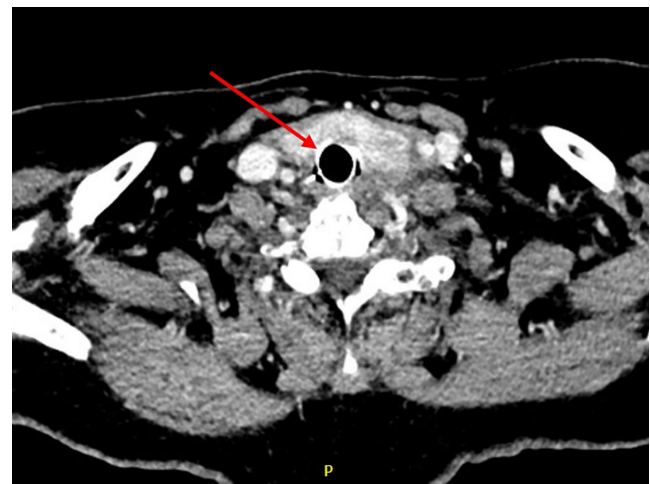
The patient was discharged within a week of admission and resumed daily activities with full autonomy, without relapse of respiratory distress. Six months post endoscopic procedure, the patient remained free of new compressive complaints, with no changes in thyroid function (Fig. 3).

Discussion

Extrinsic glottal and tracheal AO are rare causes of respiratory insufficiency in the elderly, considering the higher prevalence of other conditions such as cardiogenic pulmonary edema, lobar pneumonia, pulmonary embolism, chronic obstructive pulmonary disease, and asthma (7). Furthermore, previous studies have highlighted that improper management of acute respiratory failure in the elderly can lead to a twofold increase in mortality (7).

In this clinical case, we present an 81-year-old woman experiencing acute respiratory insufficiency due to extrinsic AO resulting from the rapid onset of Riedel's thyroiditis, necessitating urgent restoration of airflow through invasive mechanical ventilation. Emergent intubation facilitated a secure intervention for acute respiratory insufficiency, enabling accurate diagnosis establishment and safe implementation of therapeutic measures.

CT findings, in this case, revealed severe airway compression attributed to sudden thyroid gland enlargement, compounded by the patient's advanced age, raising significant suspicion of malignancy.

**Figure 3**

Cervical CT scan after 6 months of endotracheal prosthesis placement and glucocorticoid therapy, showing sustained airway patency (red arrow).

The literature consistently associates rapid thyroid enlargement with malignancy, with studies suggesting that a linear growth rate exceeding 2 mm per year strongly predicts malignancy (8). Moreover, in the elderly, especially with rapid thyroid tissue growth, there is an increased incidence of less differentiated carcinomas such as anaplastic carcinomas and thyroid lymphoma (9).

Contrary to expectations, both FNA and core needle biopsy results indicated the presence of an avascular benign inflammatory histological pattern with abundant hypocellular fibrosis, consistent with Riedel's thyroiditis. Riedel's thyroiditis, also known as invasive fibrous thyroiditis, is the rarest form of thyroiditis, with an unclear etiopathogenesis. Usually manifesting between the second and fourth decades of life, this condition entails gradual enlargement of the thyroid gland over a period of months or years, as the follicular thyroid tissue is gradually replaced by fibrous tissue (6). In exceptional cases, like the one presented, rapid thyroid glandular volume expansion may cause glottal and tracheal obstruction, mimicking malignancy.

Riedel's thyroiditis poses a rare etiology, and guidelines on treatment strategies are lacking. Treatment usually depends on the degree of compression and symptom severity. In severe cases, surgical intervention is indicated to relieve obstruction (5). Endoscopic procedures like tracheal and bronchial stent placement may also resolve airway compression. Some adjunctive therapies have been proposed, albeit with limited evidence due to the condition's rarity. Glucocorticoid therapy has been shown to reduce thyroid size, softening of thyroid parenchyma, symptom alleviation, and slow disease progression (10). In specific cases, tamoxifen has exhibited significant anti-proliferative effects on fibroblast proliferation. Other drugs like rituximab may serve as alternative treatments for refractory Riedel's thyroiditis after glucocorticoid and tamoxifen trials.

In this case, due to extensive glottal and tracheal obstruction and the emergent clinical scenario, surgical thyroid parenchyma resection was proposed. However, the patient declined this option, and an endotracheal stent combined with glucocorticoid therapy resulted in a favorable clinical outcome. This case aligns with others suggesting favorable outcomes with glucocorticoid use, advocating for their consideration as adjunctive therapy in life-saving approaches for acute dyspnea and significantly compromised airway conditions (5, 10).

Declaration of interest

The authors report that there is no conflict of interest that could be perceived as prejudicing the impartiality of the study reported.

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Patient consent

Written informed consent for the publication of their clinical details and clinical images was obtained from the patient.

Author contribution statement

Conceptualization: MLGL; data collection: MLGL, CC, CL, JSD; data analysis: MLGL and JPC; writing – original draft: MLGL and JPC; writing (review and editing): MLGL, JPC, CC. All authors read and approved the final version of the manuscript.

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