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Case Report

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Laryngeal Airway Obstruction due to Myxedema Hypothyroidism: A Case Report and Review of Literature

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Abstract

This 39-year-old female presented to the emergency room with biphasic stridor. The patient was taken to the operating room for awake fiberoptic intubation. Significant difficulty was encountered due to severe supraglottic edema but she was successfully intubated. She later underwent tracheostomy placement due to failure to wean from the ventilator. During admission thyroid levels were corrected, and on follow up visit in clinic, flexible laryngoscopy revealed complete resolution of supraglottic edema with mild residual subglottic edema.

Keywords: *Hypothyroidism, Endocrinology, Laryngology.*

1. Introduction

Hypothyroidism results in slowing of metabolic processes which can manifest in the form of constipation, weight gain, cold intolerance and lethargy. However, one of the more uncommon and life-threatening complications of untreated hypothyroidism is myxedema coma, which presents with altered mentation, bradycardia, hypotension, hypothermia, and hypoventilation [2]. Myxedema is the result of accumulation of matrix glycosaminoglycans (GAGs) in the interstitial space of tissues with resultant sodium and fluid retention [1].

Laryngeal involvement of myxedema is a rare entity with few cases documented in the literature. Most cases of myxedema involve the supraglottic structures, with only one reported case involving the subglottis. The deposition of the GAGs into laryngeal tissue begins to cause hoarseness as the primary complaint, which may quickly progress to airway obstruction. We present a case of a 39-year-old female who presented with biphasic stridor necessitating emergent intubation in the operating room, who was later discovered to be severe and longstanding hypothyroidism leading to myxedema involving the supraglottic and subglottic structures.

2. Case Presentation

This patient is a 39-year-old female with a significant medical history of hypothyroid goiter treated with radioactive iodine ablation, type II Diabetes, morbid obesity and medication non-compliance. The patient initially presented to the emergency department (ED) 8/26/22 with complaints of shortness of breath and fatigue. After 6mg of Decadron the patient improved and was discharged with instructions to follow up with PCP on 11/1/2022.

She presented again to the emergency department (ED) on 10/31/22 with gradual onset dyspnea for several days that she attributed to a sore throat. She was treated with Guaifenesin and discharged home. The following day she returned to the ED with tachypnea and wheezing. She denied any previous infections, recent fevers, or chills. Imaging of the chest showed no acute findings in the lungs or cardiac abnormalities. Imaging of the neck revealed enlargement of the epiglottis and mucosal thickening of the supraglottic region and aryepiglottic folds causing severe narrowing of the supraglottic airway. Due to initial concern for angioedema, she received a dose of racemic epinephrine and albuterol nebulizer which resulted in minimal improvement. Her oxygen saturation levels stabilized on a 15L nonrebreather mask. We were consulted to perform an awake fiberoptic intubation. Intubation was noted to be difficult due to the degree of supraglottic edema causing near complete airway obstruction.

She had several notable laboratory values with white blood cell count of 9.47 (ref. Range 4.50 - 11.0 K/mcL), a TSH of 65.24 mcIU/mL (ref. Range 0.27 - 4.20 mcIU/mL) and free T4 of <0.1ng/dL (ref. Range 0.7 - 1.7ng/dL). Historical laboratory values showed her TSH 4 months prior to be 113.80 mcIU/mL and seven months prior to 11/1/2022 was only 57.67 mcIU/mL. Patient also reported a long history of 10-15 years of known severe hypothyroidism with non-compliance of her Synthroid and would frequently go several weeks tomonths without taking medications. Severe, long-standing hypothyroidism was also evident with physical examination findings including severe constipation, extensive hair loss, and morbid obesity. Throughout her stay in the intensive care unit she was supplemented with 100mcg of Levothyroxine daily. Endocrinology was consulted and increased her dosage to 200mcg daily with bolus doses as needed. MRSA probe, Blood cultures, Respiratory PCR panel, Rapid Strep, Sputum culture, COVID/Influenza panel, and the rest of the infectious workup were all negative. The patient was treated and remained intubated for 10 days. Despite treatment there was no cuff leak around her endotracheal tube even with coughing and Valsalva. Elective tracheostomy placement was performed on 11/10/2022.

The patient was seen in the outpatient clinic for follow up and was not able to achieve voicing or airflow around her tracheostomy tube. She was taken to surgery on 12/21/22 for direct laryngoscopy and to down-size her tracheostomy tube. At the time of surgery, she was noted to still have complete obstruction of the subglottis by soft, pitting edema of the tissues (Fig. 1, Fig. 2).

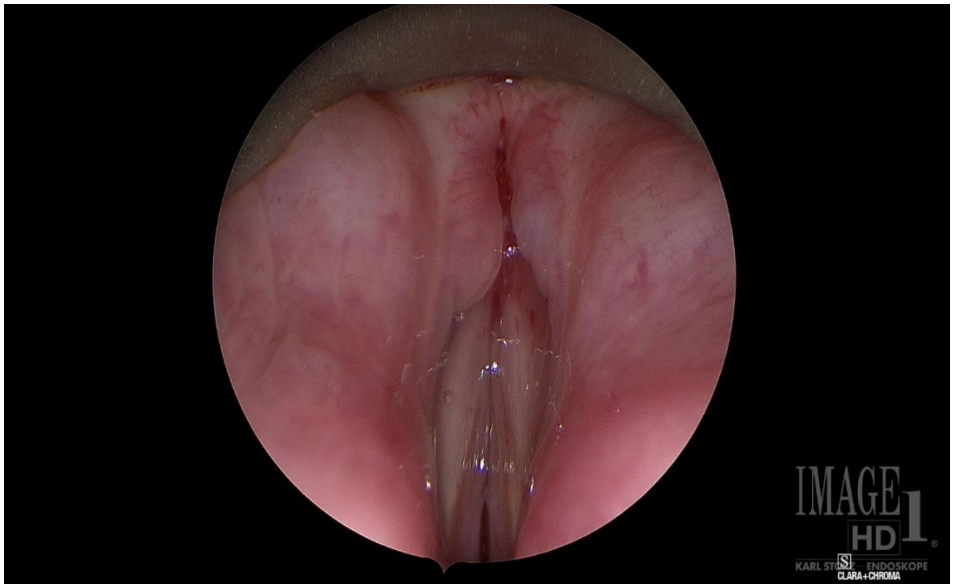

Figure 1: View of supraglottis and glottis.

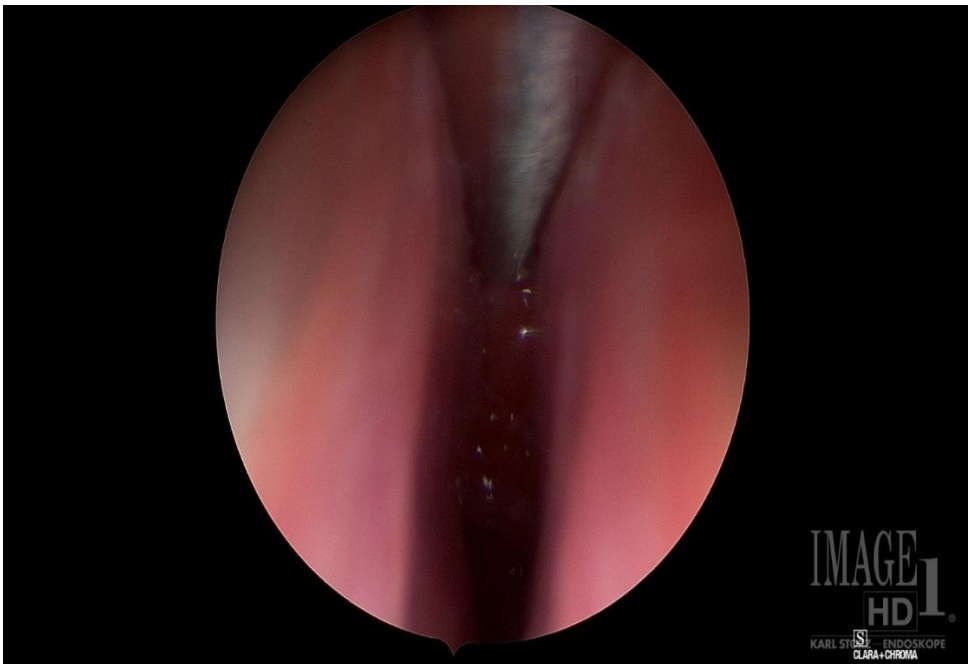

Figure 2: View of subglottis directly below true vocal cords down to the level of tracheostomy tube.

The tracheostomy tube was left in place and the patient was returned to endocrinology for continued treatment of her hypothyroidism. By her clinic exam on 2/17/23 she was able to speak with her tracheostomy tube occluded and tolerated a Passey Muir Valve for extended periods of time. On 3/8/23 the patient was admitted to the hospital for direct laryngoscopy, an overnight capping trial and possible decannulation. On direct laryngoscopy there was improvement in the subglottic edema but with persistent partial obstruction. The decision was made to keep the 6-0 proximal XLT Shiley tracheostomy tube in place and decannulation plans were delayed. The patient was kept in the hospital over night and was successful with a capping trial. The next morning the patient was discharged home with her tracheostomy in place. On 3/14/23 she returned to the ED due to a coughing spell that dislodged the tracheostomy tube. Several attempts were made to replace the tracheostomy tube back into the trachea however attempts were unsuccessful due to stomal granulation tissue and stenosis of the stoma. The ptient was deemed stable with tracheostomy tube removed, subsequently admitted for observation for two days, and discharged in stable condition decannulated. During the most recent follow up on 3/21/23 the patient continued to do well, stoma healing well, and has been compliant with all medications.

3. Discussion

In this case, the patient presented with symptoms indicative of acute upper airway obstruction. Initial imaging of the soft tissues of the neck revealed mucosal thickening and stenosis throughout the larynx. After securing the airway extensive workup was negative for angioedema, infectious causes, and malignancy. These are findings that have been shared across all case reports of supraglottic laryngeal myxedema. To date, only one case has been reported demonstrating subglottic involvement such as our patient which was a pediatric patient with congenital hypothyroidism [9]. This highlights the difficulty in diagnosing laryngeal myxedema as thyroid levels are not often included in the standard workup for laryngeal edema. Laryngeal myxedema is a diagnosis of exclusion that can be considered if a patient presents with airway compromise and workup is negative apart from significant hypothyroidism.

Profound hypothyroidism impacts the respiratory system through several mechanisms, including weakening of inspiratory muscles, depression of the ventilatory drive in response to hypoxia and hypercapnia, and obstruction of the airway from macroglossia [4]. Laryngeal myxedema is a rare cause of upper airway obstruction in patients with hypothyroidism. The exact mechanism of laryngeal myxedema has not been well described. However, there appears to be a role for thyroid hormones in the larynx, as receptors have been identified within the tissues of the larynx, including the cartilage, salivary glands, and the connective tissue of the lamina propria [5].

Literature review has shown seven cases of laryngeal swelling, assumed to be secondary to laryngeal myxedema from hypothyroidism (Table 1).

Table 1: Summary of Cases of Laryngeal Myxedema in Literature, including the Patient in the Present Study.

Reference	Age	Sex	TFTs	Laryngeal Subsite	Setting	Intubation	Tracheostomy	Treatment	Follow-up laryngoscopy
Erwin [6]	48	M	TSH: 42.4 T4: 20 mmol/L T3: 0.7 nmol/L	supraglottic	Medication noncompliance	Y	Y	levothyroxine	Persistent edema after 2 weeks
Batniji [7]	69	M	TSH: elevated FT4: 0.4ng/dL	supraglottic	newly diagnosed hypothyroidism	Y	Y	levothyroxine	Edema resolved by 2 weeks
Uzunpinar [8]	59	F	not available	supraglottic	newly diagnosed hypothyroidism	Y	Y	levothyroxine, glucocorticoids	Edema resolved by 4 weeks
Salgado [3]	63	M	TSH: 53.28 mIU/L FT4: 0.3ng/dL TT3: 57 ng/dL TPO-Ab: >600 IU/mL	supraglottic	newly diagnosed hypothyroidism	Y	Y	levothyroxine, glucocorticoids	not done
Salgado [3]	70	M	TSH: 70.6 mcIU/mL	supraglottic	medication noncompliance	Y	Y	levothyroxine,	

			FT4: <0.1ng/dL					liothyronine, glucocorticoids	Edema resolved by 2 weeks
Iftikhar [4]	59	F	TSH: 14.3 IU/L	supraglottic	medication noncompliance	Y	Y	levothyroxine, glucocorticoids	not done
			FT4: <0.25 mg/dL						
Levi [9]	Day 2	M	TSH: >100 mIU/L	subglottic	congenital hypothyroidism	Y	N	levothyroxine, glucocorticoids	Improvement in edema noted by day 18 of life
Case	39	F	TSH: 113.8 mcIU/mL	subglottic	Medication noncompliance	Y	Y	levothyroxine	Persistent edema at 4 months
			FT4: <0.1 ng/dL						

Six of the cases were in adults ages 48 to 70 presenting with supraglottic myxedema, and one case was in an infant presenting with subglottic myxedema. The most common presentation was respiratory distress with stridor, with other presentations including weakness, hoarseness, and macroglossia [3, 6-8]. All of the adult patients, including our patient, required intubation with subsequent tracheostomy. Of note, in the case of the neonate, tracheostomy was deferred and CPAP was utilized, given the compressible nature of the subglottic swelling on laryngoscopy and laboratory confirmation of congenital hypothyroidism [9]. All seven cases saw resolution of laryngeal myxedema within 4 weeks after starting levothyroxine. Our case, however, is the only reported adult patient with longstanding and severe hypothyroidism (Previous TSH levels reaching over 100) and subglottic involvement which may have contributed to improvement not beinnoted until 50 days and resolution by 133 days.

One of the challenges with management of laryngeal myxedema is recognizing its presentation. In the setting of known hypothyroidism and medication noncompliance, there should be high suspicion for laryngeal myxedema in the patient presenting with respiratory distress and stridor. However, as seen in the cases in Table 1, laryngeal myxedema can occur in patients with undiagnosed hypothyroidism or even congenital hypothyroidism. In these cases, evaluation of thyroid function and laryngoscopy (edema has been consistently reported to be compressible in nature) can be helpful to distinguish this diagnosis from other etiologies, such as allergic reactions or respiratory infections.

Upon recognition of this diagnosis, airway management and prompt initiation of levothyroxine are warranted. Intubation and tracheostomy were utilized in every adult case of laryngeal myxedema. Case by case consideration in the neonate should be utilized, as CPAP alone was successful in the case described [9]. Both oral and intravenous levothyroxine have been used in treatment. However, one should consider the possibility of bowel edema in the severely hypothyroid patient, which is why some practitioners recommended intravenous levothyroxine over oral levothyroxine to ensure adequate drug delivery [3]. Furthermore, Iftikhar et al. noted in their review that patients treated with intravenous levothyroxine were able to be weaned from the ventilator sooner than those who received oral levothyroxine [4]. One case utilized intravenous liothyronine in the setting of worsening laryngeal edema, but typically this therapy is reserved for the setting of severe hypothyroidism and myxedema coma [3]. The role of glucocorticoids in resolution of laryngeal myxedema is not well understood. Glucocorticoids are traditionally used to treat laryngeal edema secondary to infection, surgical edema, allergic reactions, and peri-extubation [4]. However, two of the seven cases reviewed resolved with levothyroxine alone.

4. Conclusion

Laryngeal myxedema is a rare but life-threatening complication of hypothyroidism. It should be suspected in patients presenting with respiratory distress and stridor with a history of hypothyroidism and in patients who are not responding to traditional therapies for airway edema. This case, coupled with literature review, suggests laryngeal myxedema is a slowly reversible entity with replacement of thyroid hormone. This case also suggests subglottic myxedema, along with severe and long-standing hypothyroidism, may further delay resolution compared to myxedema confined to the supraglottis caused by more transient hypothyroidism.

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Conflicts of interest
None declared

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Consent
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