https://doi.org/10.64142/jsmcr.3.3.73

Journal of Surgery and Medical Case Reports

ISSN: 3117-3187

JSMCR-26-73

Subglottic Laryngeal Amyloidosis Presenting as Acute Respiratory Distress: A Case Report

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Received date: Oct 24, 2026; **Accepted Date:** Nov 07, 2026; **Published date:** Nov 24, 2026

Citation: Hafidi S, Elouahab S, BijouW, Rouadi S, Abada R, et al. (2026) Subglottic Laryngeal Amyloidosis Presenting as Acute Respiratory Distress: A Case Report. J Surg Med Case Rep Vol.3 No.3: 073.

Abstract

Background: Laryngeal amyloidosis is a rare benign condition defined by extracellular deposition of amyloid proteins in the laryngeal tissues. It accounts for less than 1% of benign laryngeal tumors. The disease course is typically insidious, dominated by progressive dysphonia that may evolve toward life-threatening airway obstruction when the diagnosis is delayed.

Case Report: We report the case of a 53-year-old man admitted to the emergency department with acute respiratory distress following a one-year history of progressive dysphonia. Flexible laryngoscopy revealed a characteristic "adidas stripes" appearance with marked subglottic filling. CT scan confirmed significant narrowing of the laryngeal lumen extending from the glottis to the subglottis. Direct laryngoscopy with biopsy was performed. Congo red staining demonstrated amyloid deposits with apple-green birefringence under polarized light, confirmed by immunohistochemistry. Systemic workup was negative, establishing the diagnosis of primary localized laryngeal amyloidosis. An emergency tracheotomy was performed, followed by endoscopic CO_2 laser excision. The patient was successfully decannulated.

Conclusion: This case underlines the importance of early laryngoscopic evaluation of prolonged dysphonia. Emergency tracheotomy secures the airway in acute settings. Endoscopic CO_2 laser excision is the treatment of choice, and long-term endoscopic follow-up is mandatory given the risk of local recurrence.

Keywords: Laryngeal amyloidosis; Dysphonia; Acute airway obstruction; Congo red; CO_2 laser; Tracheotomy

Introduction

Amyloidosis is a group of disorders defined by the abnormal extracellular deposition of misfolded proteins in a characteristic fibrillar structure. These deposits can accumulate in virtually any organ. Laryngeal involvement is uncommon. It accounts for less than 1% of benign laryngeal tumors and represents only 0.2% of all laryngeal lesions [1,2].

The condition predominantly affects adults between the fourth and sixth decades of life, with a slight male predominance [3,4]. The subglottis and the laryngeal ventricles are the most frequently involved anatomical sites, followed by the supraglottis and glottis [1,5].

Laryngeal amyloidosis is almost always a primary localized disease. Progression to systemic amyloidosis is rare but must be excluded in every patient [6,7]. The clinical course is typically slow and insidious. Patients often present with months or years of progressive hoarseness before seeking medical attention [4,8].

The diagnosis is histological. Tissue biopsy with Congo red staining, showing apple-green birefringence under polarized light, is the gold standard [5,9]. Immunohistochemical analysis complements histology by helping to characterize the amyloid subtype.

Treatment relies on surgical excision, most commonly performed endoscopically using the CO_2 laser. Local recurrence is a known challenge and requires prolonged endoscopic surveillance [1,10,11].

We report a case of primary localized subglottic amyloidosis that presented as an acute respiratory emergency, successfully managed by emergency tracheotomy followed by CO_2 laser excision.

Case Report

A 53-year-old man with no significant past medical history was admitted to our emergency department with acute respiratory distress. He reported a one-year history of progressive dysphonia that he had not previously investigated.

On arrival, the patient was in significant respiratory distress. He presented with inspiratory stridor, suprasternal retraction, and reduced oxygen saturation. Initial stabilization measures were promptly initiated, including high-flow oxygen therapy and patient positioning.

Once the patient was stabilized, flexible laryngoscopy was performed. It revealed a

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https://doi.org/10.64142/jsmcr.3.3.73

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ISSN: 3117-3187

characteristic endoscopic pattern described as “adidas stripes”: Whitish submucosal deposits arranged in longitudinal stripes, with marked filling of the subglottic region **(Figure 1)**. This appearance is highly suggestive of amyloid deposits within the submucosal layer.

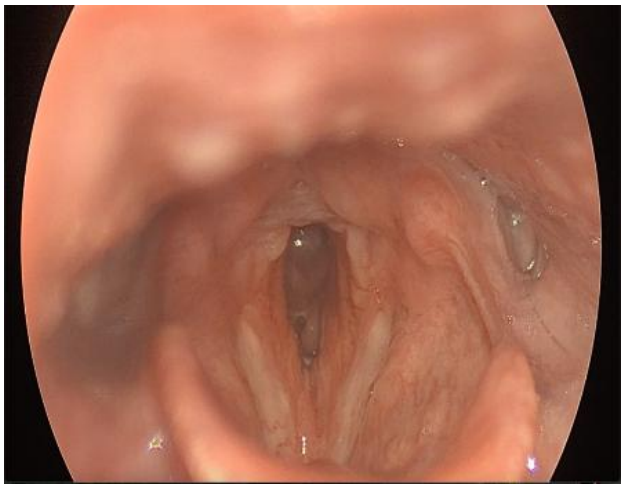


Figure 1: Flexible laryngoscopy showing the characteristic “adidas stripes” image with filling of the subglottic region.

A cervical and thoracic CT scan was obtained. It demonstrated significant circumferential soft tissue thickening with a marked reduction of the laryngeal lumen extending from the glottis to the subglottis **(Figure 2)**.

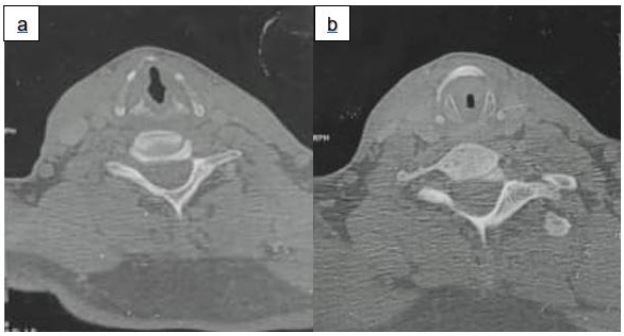


Figure 2: Cervical CT Scan pictures. a) axial section showing reduction of the laryngeal lumen of the glottis; b) Axial section showing reduction of the laryngeal lumen of the subglottis.

Despite initial medical management, the patient’s dyspnea worsened progressively. Given the critical degree of airway obstruction, an emergency tracheotomy was performed to secure the airway. The procedure was uneventful, and the patient’s respiratory status improved immediately.

After stabilization, the patient was taken to the operating room for direct laryngoscopy under general anesthesia. The procedure confirmed the endoscopic findings. Multiple biopsies were taken.

Histological analysis showed acellular eosinophilic amorphous deposits in the submucosa. Congo red staining was positive, with characteristic apple-green birefringence under polarized light microscopy, confirming the diagnosis of amyloidosis. Immunohistochemical (IHC) analysis further confirmed the amyloid nature of the deposits. Light chain immunohistochemistry (kappa and lambda) was performed to attempt subtyping.

A complete systemic evaluation was performed to exclude secondary or systemic amyloidosis. All results were within normal limits. The diagnosis of primary localized laryngeal amyloidosis was established.

After a multidisciplinary discussion, endoscopic CO_2 laser excision of the amyloid deposits was planned. The procedure was performed under general anesthesia with jet ventilation. The CO_2 laser was used to precisely excise the subglottic deposits while preserving the surrounding laryngeal structures. Hemostasis was satisfactory.

The postoperative course was uneventful. Endoscopic control confirmed adequate clearance of the obstructive deposits and restoration of an adequate airway lumen. The patient was successfully decannulated and discharged in good condition. Regular endoscopic follow-up was scheduled every six months given the known risk of local recurrence.

Discussion

Laryngeal amyloidosis is a rare condition. It accounts for less than 1% of benign laryngeal tumors in most reported series [1,2]. The exact prevalence is difficult to estimate due to the small number of published cases. The literature is largely composed of case reports and small retrospective series.

The subglottis is one of the most commonly affected sites. Symptoms depend on the location and extent of the deposits. Hoarseness is the most frequent presenting symptom, followed by dyspnea and stridor [3,4]. In a large series of 118 patients, Wu X, Zhang J, Wei C, et al. confirmed that hoarseness was the predominant symptom in the majority of cases [1].

Our patient had experienced progressive hoarseness for one year without consulting a physician. This delay is commonly reported and reflects the slow, insidious nature of the disease. It also underlines a critical clinical message: Progressive dysphonia should never be dismissed without a proper laryngoscopic evaluation.

The endoscopic appearance of laryngeal amyloidosis is variable. Deposits can appear as submucosal plaques, nodules, polypoid masses, or diffuse circumferential thickening [5,9]. In our case, the “adidas stripes” pattern whitish longitudinal

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https://doi.org/10.64142/jsmcr.3.3.73

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ISSN: 3117-3187

submucosal deposits was a distinctive and visually striking finding that immediately raised the suspicion of amyloidosis.

CT scan is an essential diagnostic tool. It provides accurate assessment of the extent of the deposits, the degree of airway narrowing, and the absence of features suggesting malignancy such as cartilage erosion or lymph node enlargement [5,12]. In our patient, the CT scan confirmed the glotto-subglottic distribution of the lesion and guided the surgical strategy.

The diagnosis of amyloidosis requires histological confirmation. Tissue biopsy remains the gold standard. On hematoxylin-eosin staining, amyloid deposits appear as acellular eosinophilic amorphous material in the submucosa. Congo red staining is the definitive step: Amyloid deposits show a salmon-pink color on light microscopy and a pathognomonic apple-green birefringence under polarized light [5,6,9]. Immunohistochemical analysis provides additional information. It helps to characterize the amyloid subtype and to exclude an AA (serum amyloid A) pattern, which would suggest an underlying chronic inflammatory disease. Light chain staining (kappa/lambda) may be variably sensitive, and complementary laboratory testing is therefore necessary [6,7].

Every patient with laryngeal amyloidosis must undergo a thorough systemic evaluation to exclude systemic amyloidosis or an underlying plasma cell dyscrasia [6,7]. Bartels H, Dikkers FG, Van Der Wal JE, et al. showed that systemic involvement, although uncommon, can be identified in a subset of patients with apparently localized laryngeal disease [7].

Recommended investigations include serum and urine protein electrophoresis, serum free light chains, renal function tests, cardiac evaluation, and abdominal fat pad aspiration when indicated. In our patient, the complete workup was negative, confirming the diagnosis of primary localized amyloidosis, which represents the majority of reported cases and carries a favorable prognosis compared to systemic forms [4,6].

Emergency airway management takes priority in patients presenting with acute respiratory distress. In our case, tracheotomy was necessary given the critical degree of airway narrowing. Tracheotomy and airway stenting have been reported in the literature as salvage procedures for patients with severe subglottic or tracheobronchial obstruction [9,10]. Endoscopic CO_2 laser excision is the treatment of choice for localized laryngeal amyloidosis. It allows precise resection of the deposits with good hemostasis and minimal damage to adjacent structures [3,6,10]. Wu X, Zhang J, Wei C, et al. compared cold microforceps and CO_2 laser in 118 patients and found both techniques to be effective, with comparable recurrence rates [1]. Piazza C, Cavaliere S, Foccoli P, et al. described successful endoscopic management in a series of 32 patients with laryngo-tracheobronchial involvement [3]. A systematic review by Galluzzi F, Garavello W confirmed that endoscopic surgery remains the most widely used and effective treatment modality [11]. In our patient, a single CO_2 laser session was sufficient to achieve satisfactory debulking and allow decannulation.

Local recurrence is a recognized challenge in the management of laryngeal amyloidosis. Wu X, et al. reported a recurrence rate of approximately 25% in a large cohort, with a mean time to recurrence of 16.5 months [1]. In another series, 30 out of 104 patients required revision surgery [12]. Subglottic involvement has been identified as an independent risk factor for local recurrence [13]. Long-term endoscopic follow-up is therefore mandatory, especially during the first four to six years after initial treatment [2,11].

Conclusion

In conclusion, laryngeal amyloidosis is a rare but serious cause of airway obstruction. Prolonged dysphonia must be properly investigated with laryngoscopy. Emergency tracheotomy is the first-line measure when respiratory distress is present. Histological confirmation with Congo red staining and immunohistochemistry is essential, along with a complete systemic workup to rule out systemic disease. Endoscopic CO_2 laser excision is safe and effective, and regular long-term surveillance is critical given the significant risk of local recurrence.

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ISSN: 3117-3187

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