International Journal of Otolaryngology and Head & Neck Surgery, 2026, 15(2), 153-157
https://www.scirp.org/journal/ijohns
ISSN Online: 2168-5460
ISSN Print: 2168-5452

Adenoid Cystic Carcinoma of the Larynx: Prolonged Course and Late Pulmonary Metastases: A Case Report

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How to cite this paper: Ndiaye, N., Diallo, M.M.R., Dieye, A., Rachide, V. and Ndiaye, M. (2026) Adenoid Cystic Carcinoma of the Larynx: Prolonged Course and Late Pulmonary Metastases: A Case Report. *International Journal of Otolaryngology and Head & Neck Surgery*, **15**, 153-157.
https://doi.org/10.4236/ijohns.2026.152014

Received: January 5, 2026
Accepted: February 28, 2026
Published: March 3, 2026

Abstract

Introduction: Adenoid cystic carcinoma (ACC) of the larynx is an extremely rare tumor, representing less than 1% of all laryngeal malignancies. Its slow progression, perineural invasion, and tendency for late distant metastases, especially pulmonary, pose significant diagnostic and therapeutic challenges. **Case Report:** We describe the case of a 24-year-old woman with chronic dysphonia and progressive laryngeal dyspnea. Histopathologic examination revealed a subglottic adenoid cystic carcinoma. She underwent total laryngectomy with bilateral functional neck dissection and total thyroidectomy. Postoperative recovery was uneventful, and no adjuvant therapy was administered. Fourteen years later, she presented with bilateral pulmonary metastases, revealed by persistent dry cough. Histologic subtype: cribriform pattern identified, without solid component. **Discussion:** Laryngeal ACC typically follows a protracted clinical course with frequent local recurrence and delayed metastases. Surgical resection remains the mainstay of treatment, while adjuvant radiotherapy is indicated for cases with positive margins or aggressive histologic patterns. The solid histologic subtype is the most significant prognostic factor, surpassing the AJCC stage in predicting outcomes. **Conclusion:** This case highlights the importance of lifelong surveillance for patients with laryngeal ACC, even after apparently curative surgery, due to the high risk of late recurrence and metastasis.

Keywords

Adenoid Cystic Carcinoma, Larynx, Pulmonary Metastases, Laryngectomy, Rare Tumor, Long-Term Follow-Up

1. Introduction

Adenoid cystic carcinoma (ACC) of the larynx is a rare malignant tumor, representing less than 1% of laryngeal cancers and approximately 10% of salivary gland malignancies [1] [2]. It typically originates from minor or major salivary glands and predominantly involves the subglottic region, followed by the supraglottic and glottic areas [1] [3]-[5].

The tumor is known for its slow progression, frequent perineural invasion, high recurrence rates, and delayed distant metastases [1] [4] [6]. Early symptoms—such as dysphonia, dyspnea, or dysphagia—are non-specific, frequently leading to delayed diagnosis and advanced disease at presentation [3] [5] [7] [8].

Histologically, ACC may present in three patterns: cribriform, tubular, and solid, with the solid subtype associated with poorer prognosis [2] [4] [9]. Lymphatic spread is rare, making the role of prophylactic neck dissection controversial [1] [3] [7].

Surgical excision with negative margins remains the cornerstone of management. Total laryngectomy is often required for extensive lesions, while conservative surgery may be feasible for limited disease [1] [3]. Adjuvant radiotherapy is recommended in cases of positive margins, perineural invasion, or high-grade histologic features [1] [4] [7].

2. Case Report

2.1. Initial Presentation

A 24-year-old woman, homemaker, residing in Louga (Senegal), with no significant past medical history, was admitted on March 4, 2011, for chronic dysphonia evolving over three years, associated with progressive laryngeal dyspnea for one month.

Panendoscopy performed on March 8, 2011, revealed a large right subglottic mass obstructing the laryngeal lumen. Biopsy and histopathological analysis confirmed adenoid cystic carcinoma of the larynx.

2.2. Initial Surgical Management and Follow-Up

She underwent partial excision of the laryngeal lesion via median pharyngotomy on April 24, 2014. The patient was subsequently lost to follow-up between 2014 and 2016.

2.3. Recurrence During Pregnancy and Definitive Surgery

In August 2016, at 35 weeks and 3 days of gestation, the patient presented with severe laryngeal dyspnea. Examination revealed a soft subhyoid swelling with intact skin and no cervical lymphadenopathy. Subisthmic tracheostomy was performed.

A cesarean section on August 14, 2016, resulted in the delivery of a healthy newborn. One month later, on September 14, 2016, total laryngectomy with bilateral functional neck dissection and total thyroidectomy was performed. Histolog-

ical examination confirmed laryngeal ACC.

2.4. Postoperative Course

Postoperative complications included an orostome, which healed completely after 34 days of honey-based dressing. The nasogastric tube was removed on September 18, 2016, and complete decannulation was achieved on July 9, 2018.

Chest CT on August 3, 2017, revealed bilateral pulmonary nodules consistent with metastases, as well as a cystic lesion involving the 11th thoracic vertebra.

The patient received six cycles of adjuvant chemotherapy without radiotherapy.

2.5. Long-Term Evolution and Late Metastases

On September 20, 2025—14 years after initial diagnosis—the patient presented with persistent dry cough and mild exertional dyspnea. Examination revealed a functional tracheostoma and good general condition (WHO stage I). Figure 1 shows the patent and functional tracheostoma.

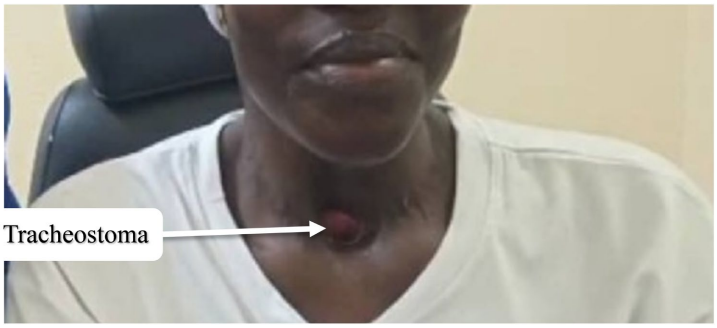

Figure 1. Patent and functional tracheostoma (September 20, 2025).

The chest X-ray revealed right pulmonary metastases, as presented in Figure 2, confirming secondary dissemination of the adenoid cystic carcinoma.

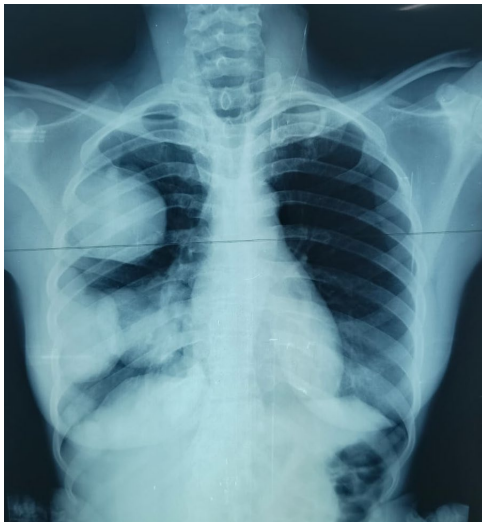

Figure 2. Chest X-ray showing right-sided pulmonary metastases, (September 20, 2025).

The chest X-ray revealed right pulmonary metastases, confirming secondary dissemination of the adenoid cystic carcinoma.

Cervico-thoraco-abdominopelvic CT confirmed multiple bilateral pulmonary nodules without local recurrence or other metastases. Multiple bilateral pulmonary metastases are demonstrated in Figure 3.

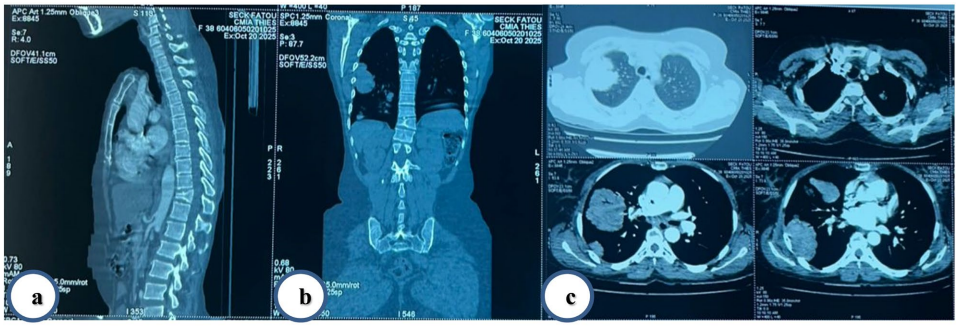

Figure 3. CT scan (October 20, 2025) (a) Sagittal, (b) Coronal, and (c) Axial sections showing multiple bilateral pulmonary metastases.

Cervico-thoraco-abdominopelvic CT confirmed multiple bilateral pulmonary nodules without local recurrence or other metastases.

The multidisciplinary tumor board recommended conservative management with close follow-up and symptomatic treatment.

3. Discussion

Laryngeal ACC is an uncommon malignancy, accounting for less than 1% of all laryngeal tumors [1] [2] [7]. The subglottic area is most frequently involved [1] [3] [5].

Histologically, the solid subtype is associated with worse prognosis [2] [4] [9]. Perineural invasion represents an unfavorable prognostic factor [1] [3] [6].

Surgery is the primary treatment modality. Total laryngectomy is often required for extensive subglottic tumors [1] [3]. Adjuvant radiotherapy remains controversial. In the present case, radiotherapy was not administered due to negative margins and absence of aggressive histologic features.

Given the potential for very late metastases—sometimes decades after treatment—lifelong surveillance is mandatory [1] [5] [7].

4. Conclusion

Adenoid cystic carcinoma of the larynx is a rare malignancy with prognosis primarily determined by histologic grade. Surgery remains the cornerstone of treatment. Because of its indolent but persistent nature, long-term, often lifelong follow-up is crucial.

Informed Consent

Written informed consent was obtained from the patient.

Conflicts of Interest

The authors declare no conflicts of interest.

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