

# Adenoid cystic carcinoma of the larynx presenting with unusual subglottic mass: Case report



Takashi Kashiwagi, Hiroaki Kanaya\*, Wataru Konno, Kazutaka Goto, Hideki Hirabayashi, Shin-ichi Haruna

Department of Otorhinolaryngology – Head and Neck Surgery, Dokkyo Medical University, 880 Kitakobayashi, Mibu, Tochigi 321-0293, Japan<sup>1</sup>

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## ABSTRACT

A 33-year-old woman presented with an unusual subglottic bulging mass accompanied by prolonged cough and wheeze. Laryngeal endoscopy revealed a bilateral, symmetrical mass immediately below the vocal cords with marked airway obstruction. Chronic subglottic laryngitis with inflammation or another condition such as amyloidosis was initially suspected. Cervicothoracic computed tomography revealed an obvious reduction of laryngeal caliber caused by an engulfing mass extending from just under the vocal cords to the cricoid ring, which was associated with thyroid, arytenoid, and cricoid cartilage destruction. Histopathological diagnosis of a biopsy specimen collected via a tracheotomy revealed that the lesion was a cT4aN0M0 adenoid cystic carcinoma (ACC) originating from the laryngeal minor salivary glands. The patient was treated by total laryngectomy with elective bilateral neck dissection under general anesthesia. Gross inspection of resected tissue confirmed yellowish-white, solid tumor mainly circumferentially encompassing the lumina of the cricoid ring. The histopathological findings confirmed typical ACC accompanied by a predominant cribriform appearance with no evidence of lymph node metastasis. The patient remains well and free of recurrence or metastasis. We herein describe laryngeal ACC and discuss radiological images and the surgical pathology.

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## 1. Introduction

Adenoid cystic carcinoma (ACC) of the larynx is a distinct and extremely rare malignant neoplasm [1–5] that was thought to arise from minor salivary glands in the laryngeal submucosa. An ACC of the larynx was reported as a rare tumor during the 1960s, but the nomenclature of this tumor was confused with that of other neoplasms such as cystic basal cell carcinoma or pleomorphic adenoma with a cribriform appearance [2]. The incidence of ACC originating from the larynx comprises <1% of all laryngeal tumors [1,3]. Laryngeal ACC usually affects

subglottal subsites [3–6] and occurs as submucosal mass, and patients tend to present with symptoms of long duration because of slow growth and progression of the mass [4–8]. Moreover, early subglottic tumors present with non-specific airway symptoms characterized by cough and wheezing that are often misdiagnosed as asthma [9,10] or as occasionally mimicking amyloidosis [11]. We describe a patient with laryngeal ACC presenting with a large subglottic bulging mass and compare detailed radiological and surgical/pathological findings of this extremely rare disease.

## 2. Case report

A general physician diagnosed asthma in a 33-year-old female non-smoker who was allergic to house dust and

\* Corresponding author.

E-mail address: [h-kanaya@dokkyomed.ac.jp](mailto:h-kanaya@dokkyomed.ac.jp) (H. Kanaya).

<sup>1</sup> Tel.: +81 282 87 2164; fax: +81 282 86 5928.

presented with prolonged cough and wheezing accompanied by gradually worsening hoarseness over a period of 10 months. Since anterior neck tightness also developed during the past two months, the patient was referred to an otolaryngology clinic for medical assessment. Subglottic stenosis was identified and she was referred to our department. Her symptoms had fluctuated, but steadily progressed.

Laryngeal endoscopy revealed a bilateral and symmetrical bulging mass with an apparently intact mucosal surface immediately below the vocal cords accompanied by obvious airway obstruction (Fig. 1A and B). The bilateral vocal cords were laterally biased, with mobility limited due to oppression of the mass. Chronic subglottic laryngitis of unknown cause or a degenerative disease such as amyloidosis was suspected at this point. The findings of fiberscopy confused the diagnosis because we had never encountered such a lesion. Cervicothoracic computed tomography (CT) revealed an obviously reduced laryngeal caliber caused mainly by lesion engulfment from below the vocal cords to the cricoid ring that had destroyed the thyroid, bilateral arytenoid, and cricoid cartilage (Fig. 1C and D). The mass had poorly enhanced areas in contrast to the thyroid gland. These findings indicated a differential diagnosis of a malignant tumor arising from the subglottic region despite the absence of cervical lymphadenopathy.

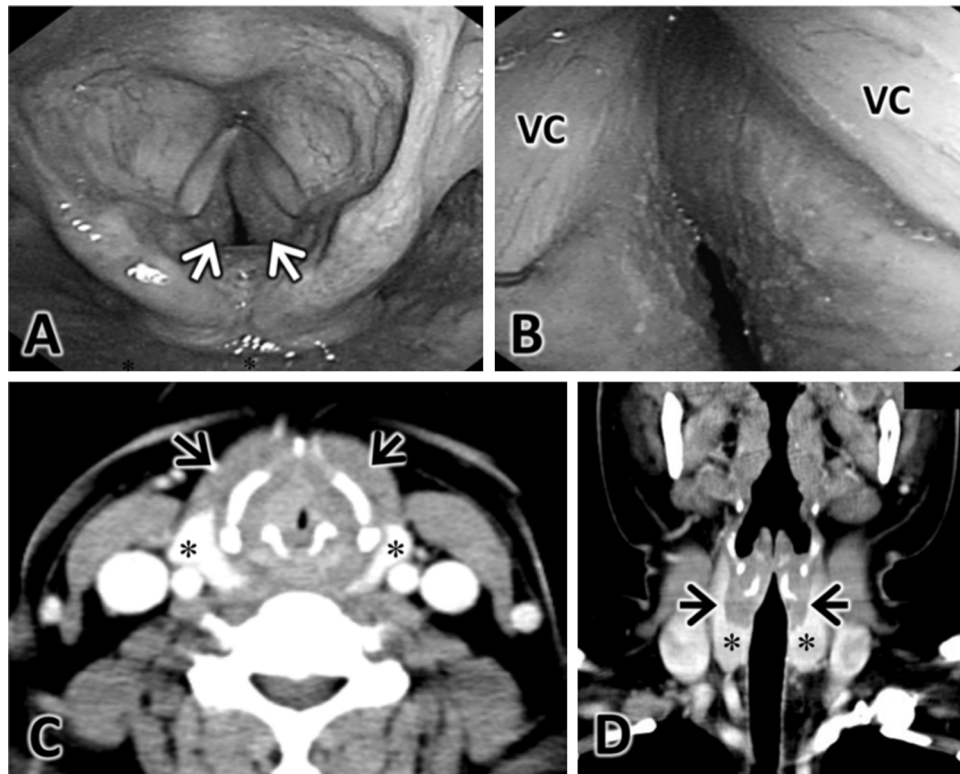
The histopathological diagnosis of a biopsy obtained from the lesion via a tracheotomy to secure the respiratory passage was ACC that appeared to originate from the laryngeal minor salivary glands. We generally searched for metastatic lesions

using positron emission tomography with CT and the mass was clinically staged as cT4aN0M0 ACC according to the UICC TNM classification.

Total laryngectomy (Fig. 2A) with elective bilateral neck dissection of levels II–III–IV proceeded under general anesthesia. Surgical radicality was improved by en-bloc resection of the thyroid gland. Gross observation of the resected tissue confirmed a yellowish-white solid tumor that circumferentially involved mainly the lumina of the cricoid ring (Fig. 2B and C). The tumor was covered with intact mucosa (Fig. 2D) and spread outside the cricoid cartilage via the annular ligament between the cricoid and the first tracheal cartilage (Fig. 2E), and directly invaded the thyroid gland. The surgical margins were tumor-free and histopathological assessment under higher magnification revealed typical ACC with a predominant cribriform appearance with less than 30% of solid component (Fig. 2F and G). Metastases, perineural invasion, lymphangio-invasion, and vascular invasion were undetectable by microscopy including 71 dissected lymph nodes. The tumor was finally diagnosed as stage IV laryngeal ACC, pT4aN0M0. The patient has remained free of local recurrence or distant metastasis for two years.

### 3. Discussion

Although ACC derived from the larynx is extremely rare [1,3–11], a few case series have been described. Moukarbel et al. [4] reviewed 15 patients with laryngeal ACC and



**Fig. 1.** Laryngeal endoscopy and computed tomography findings of bulging mass. (A) Laryngeal endoscopy shows bilateral and symmetrical bulging mass (arrows) just below vocal cords (VC) that were fixed laterally due to oppression by mass. (B) Closer view of mass. (C and D) Cervicothoracic CT images show subglottic mass surrounding vocal cords extending to cricoid ring with associated destruction of thyroid, bilateral arytenoid, and cricoid cartilage (arrows). Cervical lymphadenopathy is absent. \*Thyroid gland.

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