Multiple primaries of head and neck: Adeno cystic carcinoma of the larynx with follicular carcinoma of the thyroid gland
Case Report

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Abstract
Adeno cystic carcinoma of the larynx is very rare and concomitantly with the follicular thyroid carcinoma is not reported in the literature. This case was presented to our centre because of the stridor and small anterior neck mass. Evaluation of the case showed a subglottic mass with an enhanced right thyroid mass on the CT scan. The treatment was thyroidectomy followed by total laryngectomy and bilateral neck dissection. The histopathological study of the subglottic mass showed adeno cystic carcinoma and of the thyroid mass was follicular carcinoma. The adeno cystic carcinoma of the larynx is usually asymptomatic and it has the opportunity to invade deeply before the diagnosis. Proper evaluation of the head and neck is mandatory in any laryngeal lesion.

Keywords: Adeno cystic carcinoma, larynx, follicular carcinoma

Introduction
Adeno cystic carcinoma of the larynx is very rare; the subglottis in contrast with other laryngeal malignancies is the commonest site. Women and men are equally affected by adeno cystic carcinoma1,2. Some report a slight female predisposition, and their peak incidence is in the fifth and sixth decades of life3. Adeno cystic carcinomas account for less than 1% of all malignant tumors in the larynx, only about 120 cases have been reported in the literature until now4. Follicular thyroid carcinoma histopathologically is a differentiated tumor and is the second most common cancer of the thyroid after papillary carcinoma. The incidence of follicular carcinoma is high in areas of endemic goiter possibly due to thyroid stimulating hormone stimulation5. The occurrence of occult synchronous thyroid tumors in patients with laryngeal tumors has previously been reported6. There are reported cases of synchronous thyroid lesions with squamous cell carcinoma of the larynx7. The aim of reporting this case is that it is unusual and to our knowledge there has been no reported case of adeno cystic carcinoma of the larynx with follicular thyroid carcinoma.

Case Presentation:
A 55 year old female presented to our otorhinolaryngology clinic at Rizgari Teaching Hospital, Erbil, Iraq with symptoms of dyspnea and stridor on March 2006. The patient decided to seek medical treatment outside Iraq. A CT scan of the neck demonstrated a subglottic mass and a right thyroid mass, Figure 1. She underwent total thyroidectomy on May 2006, abroad, Figure 2 a. The subglottic mass was not addressed at that stage. She received postoperative radioactive iodine treatment according to the histopathological result from the specimen of the thyroidectomy.

Figure 1: CT scan at April 2006 showing laryngeal and thyroid lesions (before thyroidectomy)

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In January of 2008 she had an emergency tracheostomy for stridor at another institution and she was referred to our medical center for further management. Physical examination of the head and neck area revealed the neck with a tracheostomy tube and a healed thyroidectomy incision, with no evidence of adenopathy. A fiberoptic laryngeal examination showed a subglottic mass located anteriorly with extension into the upper tracheal wall. A CT scan of the neck revealed a laryngeal tumor involving the anterior and lateral subglottis with extension into the trachea, Figure 2 a,b. The patient underwent direct laryngoscopy and biopsy of the subglottic mass. Histopathology analysis showed evidence of adenoid cystic carcinoma, which is a neoplastic tissue composed of epithelial cells which are arranged in glandular pattern surrounded by loss stroma stained metachromatically by methylene blue, Figure 3.

**Figure 2 a:** Subglottic mass with intact anterior wall of the larynx (after thyroidectomy)

**Figure 2, b:** The mass in the lumen of the upper trachea anteriorly

**Figure 3:** Adenoid cystic carcinoma (subglottis). H&E X400
Total laryngectomy with bilateral selective neck dissection was performed on the patient, (levels IV, V&VI), because during the operation there was remnant of thyroid tissue. Histopathology showed negative margins around the primary (subglottic) tumor, Figure 4a.

The specimens after total laryngectomy and neck dissection were sent for histopathological examination which revealed evidence of adenocystic carcinoma. The specimen of left neck dissection showed follicular carcinoma with glandular differentiation while the specimen from right neck dissection showed metastatic follicular carcinoma of thyroid origin, Figure 5.

There were no postoperative complications. The patient stayed 10 days at hospital. The course was uneventful and the patient was discharged on 10$^{th}$ postoperative day tolerating oral diet. She didn’t receive further radiotherapy. The patient is currently alive with no evidence of any disease and was seen on follow up consultation. The postoperative protocol was as follows: Monthly evaluated in otolaryngology department for the first six months & then every six month for two years, after that every year for five years. Later she will be evaluated every five years forever.

Figure 4a: Specimen of the larynx(subglottic mass)

Figure 4b: bilateral neck dissection

Figure 5: Follicular carcinoma of thyroid origin H&E X400

Figure 6: the patient after three years of her treatment
Discussion

Adeno cystic carcinoma usually occurs as a largely asymptomatic, nonulcerated submucosal mass. As a result, diagnosis is often delayed and, in the larynx, subglottic tumors have the opportunity to invade deeply before they are diagnosed. Preoperative histopathological analysis is essential because the symptoms do not differ greatly from squamous cell carcinoma. Subglottic tumors typically present with shortness of breath, dyspnea on exertion, and may be accompanied by hoarseness. Patient may present with a neck mass. In majority of patients symptoms were present for more than six months. On endoscopy, patients typically have a submucosal mass with normal overlying respiratory epithelium. Similarly, our patient presented with persistence hoarseness and decrease effort tolerance. The histopathological pattern of adenoid cystic carcinoma is classified into three distinct subtypes: cribriform, which is the most common; tubular, which has the best prognosis; and solid, which carries the worst prognosis. Local recurrences and distant metastases (especially to the lung) are common and sometimes arise years after the primary tumor has been diagnosed and treated. Another very important factor with adenocystic carcinoma is the tendency for perineural invasion. Therefore, these patients require long-term follow-up (more than 5 years). Most authors agree that the treatment of choice is wide-margin local excision, partial or total laryngectomy, depending on the location and size of the tumor. In the absence of neck metastasis, elective neck dissection is unnecessary. Radical neck dissection is indicated for patients who have clinically or histopathologically confirmed nodal metastases. The role of radiotherapy is still open to debate. These tumors have been shown to be radiosensitive but usually not radioresistant. Therefore, radiotherapy alone usually has little role in treatment.

Conclusion

This is an unusual and very rare case of adenocystic carcinoma with follicular carcinoma of the thyroid gland, which has not been reported previously. Proper preoperative evaluation of the thyroid especially the thyroid is mandatory in every case of carcinoma of the larynx.

References