Subglottic Metastasis of Adenoid Cystic Carcinoma from an Anterior Tongue Primary

Abstract

A 69-year-old lady, whose clinical course included previous pulmonary and cutaneous metastases, presented six years subsequent to primary excision and radiotherapy for left anterior tongue adenoid cystic carcinoma. Computed tomography neck, chest, and abdomen performed for pulmonary metastatic monitoring, incidentally detected a subglottic soft tissue swelling. Microlaryngoscopy and carbon dioxide laser excision completely removed this subglottic polyposidal mass. Histology confirmed respiratory-type mucosa invaded and ulcerated by adenoid cystic carcinoma with perineural invasion. Adenoid cystic carcinoma is locally invasive and only accounts for 1% of all head and neck malignancies. Hematogenous spread to lung, bone and liver are common, conversely regional lymph node metastases are rare. Despite this, due to the proximity of the primary and metastasis, the complex lymphatic connection between these sites may play a crucial role in this case.

Keywords: Adenoid cystic carcinoma; Tongue; Metastasis; Larynx subglottic

Introduction

Adenoid cystic carcinoma (ACC) is a well-documented malignant tumor of the major and minor salivary glands that can also originate from other areas of the aero-digestive tract, including the larynx and trachea, and accounts for 1% of all head and neck malignancies. ACC is locally invasive, with radical surgical resection and postoperative radiotherapy regarded as the treatment of choice. Although regional lymph node metastases are rare, hematogenous spread to lung, bone and liver are common [1]. This report describes a unique case of a delayed subglottic metastasis of ACC of the left anterior tongue subsequent to surgical resection and radiotherapy.

Case Report

A 69-year-old lady presented with a constant burning sensation in her tongue of three months’ duration. She was found to have an extremely nodular left tongue, especially at the tip and lateral surface, which was exquisitely tender upon palpation. Diagnosis was uncertain, thus MRI was performed, demonstrating a 2.2 cm by 1.7 cm lobulated left-side tongue mass of high signal intensity on short T1-inversion recovery (STIR) images (Figure 1). It did not extend into the floor of the mouth and there was no abnormal adenopathy. A biopsy was performed, providing a histological diagnosis of ACC. Staging CT chest and abdomen with contrast showed a non-specific solitary 9.5 cm pulmonary nodule peripherally at the left lung base with no mediastinal or hilar adenopathy. It was elected in the head and neck cancer multidisciplinary team meeting that she have local excision of the tumor. A partial glossectomy with resection of tumor was performed utilizing a carbon dioxide laser (10 watts continuous). During the procedure the ducts were avoided and preserved.

The specimen revealed tongue mucosa and underlying muscle invaded by a grade 2 ACC, 3.5 cm in maximum extent by microscopy (but only 1.7 cm by macroscopic inspection). This discrepancy between macroscopic and microscopic size is most likely due to the highly infiltrative nature of the carcinoma, whereby thin invasive spikes emanate beyond the edge of the main carcinoma mass, often following nerves (Figure 2). The ACC involved the deep margin focally including via extensive perineural
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Figure 2: Histopathology of the Resected Primary Tumor in the Tongue.

A follow-up PET scan was undertaken following scalp lesion excision, showing widespread metastatic disease in bone (primarily lumbar spine) and lung, and MRI highlighted a right fundal lesion (orbit), as well as right occipital and cerebellar metastases. These were treated with palliative radiotherapy (20 Gy in 5 fractions). Repeat CT neck, thorax, and abdomen was subsequently performed, demonstrating a mild reduction of metastatic disease within the occipital lobe, cerebellum and right orbit. However, a new finding of a polypoidal soft tissue thickening in the trachea at the level of the cricoid was also noted (Figure 3). The otolaryngologically asymptomatic patient therefore received clinical review in the head and neck clinic, where flexible nasal endoscopic evaluation of the larynx revealed an obvious polypoidal swelling in the right subglottis. A laryngotracheal lesion had not been encountered in previous imaging studies, nor during previous procedures or clinical examinations, thus were the clinical features consistent with metastatic ACC.

Microlaryngoscopy and laser resection of the mass was performed using carbon dioxide laser (10 watts-1 mm superpulsed), by which time over 6 years had passed since primary tumor resection. The subglottic lesion was completely excised and the histology report described the tissue as respiratory-type mucosa invaded and ulcerated by ACC with perineural invasion. On postoperative clinic evaluation, the patient reported no voice complaints and no tumor remnant was visible on flexible nasal laryngoscopic evaluation of the larynx. Ten months following microlaryngoscopic laser resection, the patient was clinically stable following completion of widespread palliative radiotherapy, with no new metastases detected, and without otolaryngological symptoms. Discussion Adenoid cystic carcinoma most commonly arises in the parotid, submandibular and minor salivary glands [1]. However, it can also occur in other sites, including the tongue which accounts for 4.4% of cases [2]. This therefore establishes the uncommon nature of the left-sided ACC tongue mass in this case.

Prognosis depends on the histological grade of the carcinoma with grade 1 tumor having a 15-year survival of about 39%, for grade 2 the 15-year survival is about 26%, and for grade 3 it is about 5% [3]. The poor prognosis of ACC is also linked to failure to control distant disease [1]. This is evident in this case, since despite active treatment of the primary tumor, numerous distant metastases occurred. Among this distant disease, the finding of a subglottic metastasis from the tongue was a highly unusual discovery. Furthermore, the metastasis appeared 6 years after primary excision of the tumor. A database search of PubMed, Embase and Cochrane Library was therefore performed with MeSH terms 'metastatic carcinoma; larynx; subglottic AND tongue; adenoid cystic carcinoma; metastasis' to identify any previously published cases. The literature search revealed no cases associated with metastasis to the subglottis from the tongue.

Metastatic tumors to the larynx are rare and account for 0.1 to 0.4% of all laryngeal tumors [4]. The most common primary sites are cutaneous melanoma and renal cell carcinoma, with colorectal adenocarcinoma metastasis increasingly reported. The signs and symptoms of metastatic laryngeal lesions are similar to those of primary laryngeal malignancies, including hoarseness, stridor and dyspnea. Tracheostomy can be performed in those with respiratory distress where the tumor is compromising their airway, but in our case, the patient was asymptomatic allowing this to be avoided [4]. The mechanism of spread from tongue to subglottis...
However, single collecting vessels drain the posterior floor of the mouth to the oropharynx, which in turn is connected to the superficial and deep lymphatic networks of the laryngeal mucosa in an uninterrupted manner. The supraglottis has a high density of lymph collectors and may communicate with the subglottis [9]. This complex system could explain the rare spread of ACC from tongue to the subglottis.

Conclusion

To the best of our knowledge this is the first documented case of a subglottic metastasis from a tongue cancer. The mechanism by which this has occurred is uncertain but there are various routes by which this could happen. The lymphatic supply from the tongue to the larynx may play a crucial role due to the proximity of the two regions, yet due to the patient’s multiple metastatic deposits, a hematogenous means is also implicated.

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References