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C3 Vertebral Metastases From Tongue Adenoid Cystic Carcinoma

A Rare Case Report

Helin Feng, MD, Jin Wang, MM, Peng Guo, MM, Jianfa Xu, MM, and Jiangan Feng, MD

Abstract: We report a rare case involving a patient with C3 vertebral body metastasis secondary to adenoid cystic carcinoma of the tongue.

Five years after local resection of the primary tumor, magnetic resonance imaging showed a metastasis located in the left posterior border of the C3 vertebral body. Additionally, multiple pulmonary metastases were identified by computed tomography. Based on these findings, the patient underwent C2–3, C3–4 discectomy; C3 corpectomy; and titanium mesh fusion with a Zephir plate. The diagnosis was confirmed by the pathology findings. During 6 months of follow-up, the patient showed improvement and return of function of the cervical vertebrae, with no serious complications.

Because of the scarcity of cases of vertebral metastases from tumors of the tongue in the literature, we have reported this case to add to the available evidence regarding this rarely encountered condition.

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Abbreviations: ACC = adenoid cystic carcinoma, MRI = magnetic resonance imaging, SCC = squamous cell carcinoma.

INTRODUCTION

Tongue cancer is the most common type of oral cancer¹; however, most malignant tongue tumors are squamous cell carcinoma (SCC).² Other common locations of oral SCCs include the salivary glands,³ followed by the hard palate and the base of the tongue.⁴ On the other hand, tongue adenoid cystic carcinoma (ACC) is rarely seen in the clinic.⁵ Further, while this rare malignancy has been reported to frequently metastasize to the lung and bones,⁶ we here describe a very rare case involving a patient who presented with a left posterior tongue lesion with metastatic spread to the C3 vertebrae. The patient was successfully treated with surgery and radiotherapy.

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Received: March 17, 2015; revised: June 2, 2015; accepted: June 16, 2015. From the Department of Orthopedics, The Fourth Affiliated Hospital of Hebei Medical University, Shijiazhuang, Hebei, P.R. China (HF, JW, PG, JX, JF).

Correspondence: Jiangan Feng, The Fourth Affiliated Hospital of Hebei Medical University, Shijiazhuang, Hebei, P.R. China (e-mail: fengjiangan0311@126.com).

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CASE REPORT

In 2009, a 52-year-old woman was referred to our hospital with an asymptomatic volume growth on her left posterior tongue. Examination of a biopsy specimen showed cancer cells. Accordingly, she was diagnosed with tongue cancer and underwent local resection. The tumor was diagnosed as tongue ACC based on the pathology findings (Figure 1). The patient received interstitial iodine-125 implantation postoperatively. Five years later, in June 2014, she started to experience paralysis in both hands and reported pain in her neck. X-ray imaging showed that the C3 posterior edge density was slightly decreased (Figure 2A and B). Further, T1-weighted magnetic resonance imaging (MRI) demonstrated a subtle decrease in signal in the C3 vertebral body (Figure 2C and D). The findings of each imaging modality showed that the left posterior border of the C3 vertebral body was invaded, and that multiple pulmonary metastases were present (Figure 3). Thus, we performed anterior C2–3, C3–4 discectomy; C3 vertebral resection; and titanium mesh fusion with a Zephir plate. The operation was successful, and the intraoperative blood loss was approximately 200 mL. Histopathological analysis demonstrated a neoplasm that was formed of microcystic spaces surrounded by atypical hyperchromatic epithelial cells invading the adjacent muscular tissue. The pathology findings of the C3 vertebral body lesion revealed ACC (Figure 4). After 2 weeks of adjuvant chemoradiation therapy, the patient remained in good health. Two months later, magnetic resonance images and computed tomography showed no recurrence (Figure 5). At the 6-month follow-up, the patient was alive and the function of the cervical vertebrae was improved. The patient provided written informed consent for the publication of these case details and the consent procedure was approved by the Human Ethics and

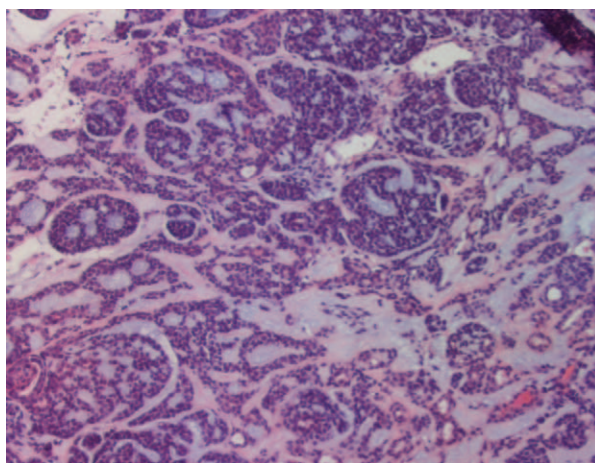


FIGURE 1. Pathology findings resulted in a diagnosis of the tumor as tongue adenoid cystic carcinoma.

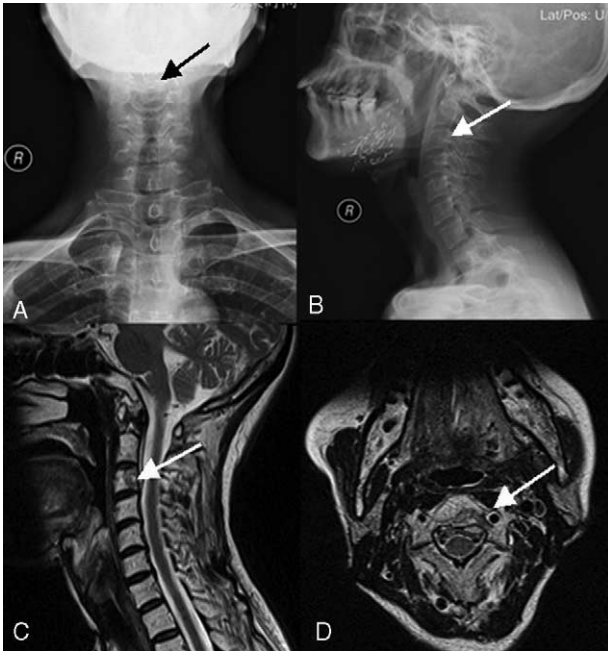


FIGURE 2. X-ray imaging (A and B) showed that the C3 posterior edge density was slightly decreased (arrow). T1-weighted magnetic resonance images without contrast enhancement (C, sagittal; D, axial) demonstrated a subtle decrease in signal in the C3 vertebral body (arrow).

Research Ethics committees of the Fourth Hospital of Hebei Medical University.

DISCUSSION

In 1859, Billroth first coined the term “cylindroma” to describe the histological standard of 4 salivary gland tumors. This term was widely used until 1953, when Foote and Frazell redesignated these lesions as “adenoid cystic carcinomas.”⁷⁻⁹ ACC can originate from both the minor and major salivary glands,¹⁰ and a recent study suggested that 4.4% of minor salivary gland tumors are located on the tongue.³ These tumors

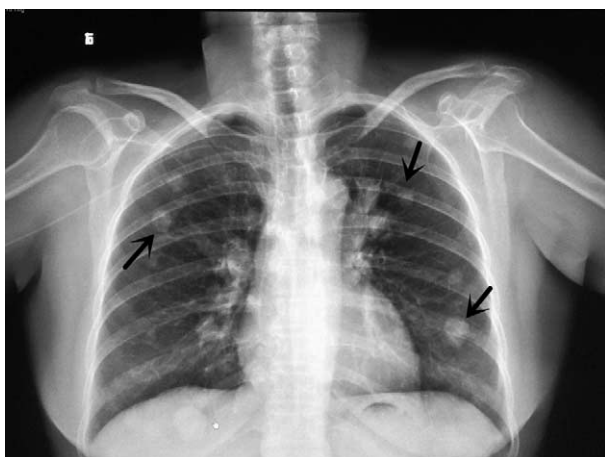


FIGURE 3. Chest radiographs revealed multiple pulmonary metastases (arrow).

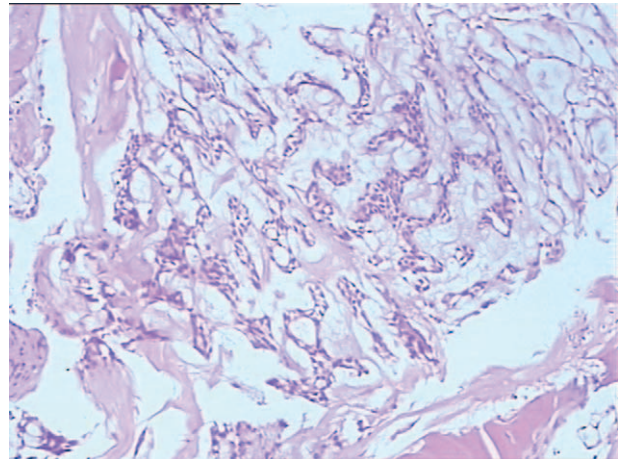


FIGURE 4. Microphotograph, showing the formation of microcystic spaces surrounded by hyperchromatic cells (hematoxylin and eosin stain).

generally progress slowly and tend to show wide perineural invasion into the adjacent nerves and distant metastasis. The primary tumor generally metastasizes via the lymphatics¹¹ and rarely metastasizes via the blood. Carlson and Ord¹² reported the cases of 4 patients with 13 involved vertebrae, all of whom presented with symptoms such as severe pain, hypercalcemic somnolence, and flaccid paralysis of the lower extremities as part of cauda equina syndrome. Accordingly, the authors advocated that such patients, especially those with bone pain, should undergo radiographic examinations to identify distant metastases. Mendes et al¹³ described 3 patients who presented with symptoms and signs of nerve root compression affecting the upper limbs secondary to tongue cancer metastasis. Further, in

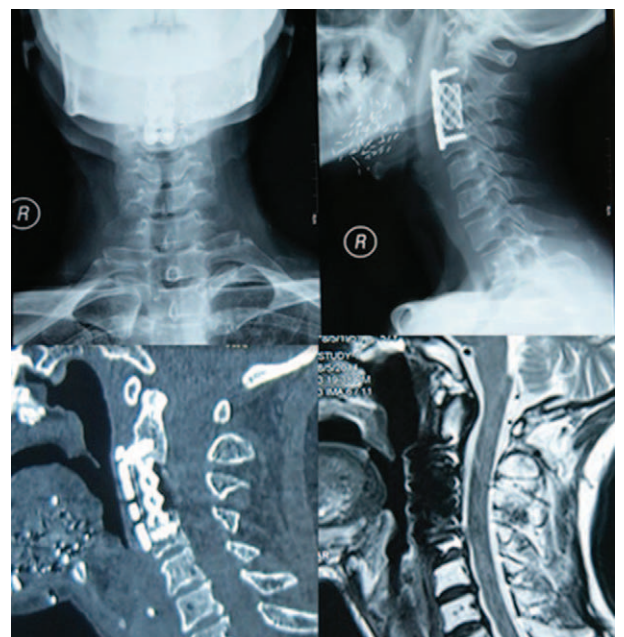


FIGURE 5. The patient was followed-up with magnetic resonance imaging and computed tomography scans, which did not show any detectable recurrence at 2 months.

2007, Lee et al¹⁴ reported 2 cases in which signs of spinal cord compression developed due to tongue tumor metastasis to the lumbar vertebrae, and the authors suggested that oral cancer may cause vertebral metastasis. In a more recent report, Tornwall et al¹⁵ described a case of spinal cord metastases from oral SCC that started with pain in the lower limbs and spine. However, the present case differed from previously reported cases in several respects aspects: including the facts that it was involved tongue ACC rather than SCC, showed metastasis to the C3 vertebrae, and that the patient achieved significant improvements in quality of life using the management strategies that have been described above. In resemblance with the previously reported cases, our case indicated that tongue cancer has a tendency toward distant metastasis, and clinicians should hence be aware of the possibility of spine metastasis in any patient presenting with tongue cancer.

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