Spinal Metastases from Adenoid Cystic Carcinoma of the Parotid Gland

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To the Editor: The "Parotid Gland", out of all the salivary glands, is the one most frequently affected by neoplasms, accounting for approximately 75% in all cases.^[1] These neoplasms are generally benign, with only 25% of them being malignant forms.^[1] The percentage of distant metastases from salivary gland is relatively low, with their occurrence being associated with high-grade tumors. The "Adenoid Cystic Carcinoma" (ACC) is a tumor of exocrine glands originating primarily from the minor salivary glands of the upper respiratory gland, the major salivary glands, lacrimal gland, bronchus, breast, and intestinal and genital tracts.^[2] Its key characteristic is that it grows slowly; however, peritumoral invasion and perineural infiltration occur in the process. To date, the spinal metastasis from ACC of the parotid gland is exceedingly rare in literature, with only a few cases presenting spinal metastasis of the parotid gland have been documented, thus resulting in consequence of lacking imaging proof. Herein, we described a case of spinal metastases of parotid carcinoma treated with surgical treatment.

A 68-year-old female patient, with a 1-year history of continuous and progressive back pain as well as acute radiculopathy in her bilateral limbs, was presented at the Peking Union Medical College Hospital on April 2017. The patient, having been diagnosed of ACC in her left parotid gland for 3 years, received surgical treatments including a total resection of left parotid gland, transplantation of facial nerve, and transferring of left sternocleidomastoid muscle flap. After being admitted to hospital, magnetic resonance imaging revealed a significant bone destruction, pathological vertebral fractures, and multilevel lumbar spinal cord compressions [Figure 1a and 1b]. She denied a history of injury or any other underlying diseases. The result of general physical examination was remarkable in terms of pressure pain and percussion pain in the spinal region at T12, L1, and L4 level with a limited range of motion and pain in all planes of movement. Routine laboratory studies showed that results were almost within normal range, except that of the tissue polypeptide-specific antigen, which was significantly elevated to 124.23 U/L (normal: <80 U/L), serum neuron-specific enolase elevated to 17.3 ng/ml (normal: <16.3 ng/ml), and cytokeratin 19 fragment

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elevated to 4.7 ng/ml (normal: <3.5 ng/ml). Bone scintigraphy indicated a skeletal abnormality of L1 and L4, with high suspicion of metastases [Figure 1c]. She was administered analgesic therapy for pain-relieving purpose, and later, a circumferential spinal cord decompression, while a stabilization procedure together with cement augmentation was performed [Figure 1d]. The postoperative pathology confirmed a metastatic ACC of spine [Figure 1e–1j]. One week after the operation, the patient's symptoms were relieved significantly. Moreover, visual analog scale score of her back pain improved to 1-2 points compared to the preoperative status, 8-9 points. Postoperatively, the patient declined adjuvant treatment but underwent rehabilitation and was monitored as an outpatient. The postoperative 1-year follow-up visit showed no tumor progression or new symptoms. This was an extremely unique presentation and surgical management of metastatic parotid carcinoma of spine.

ACC is a malignant tumor arising from the major and minor salivary glands, lacrimal glands, upper respiratory tract, breast, or uterine cervix.^[1] Current treatment of this tumor can generally achieve a good short-term prognosis with slow growth that allows prolonged survival. Nevertheless, the long-term prognosis is still poor due to the invasive nature of the tumor and metastases, frequently to the liver, lungs, orbit, and cervical lymph nodes.^[1,2] Metastasis of ACC to the spine is exceedingly rare, and only a few case reports exist in literature.^[3] Histologic examination of the primary tumor, the presence or absence of positive margins, and physical and radiologic studies of anatomical spread are essential for the treatment of spinal metastasis of ACC.^[3,4] We recommend the posterior approach for spinal decompression

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Figure 1: Spinal metastases of parotid carcinoma in a 68-year-old female patient. (a and b) Preoperative sagittal and transverse T2-weighted MRI scan revealed significant bone destruction, pathological vertebral fractures, and multilevel lumbar spinal cord compressions. (c) Bone scintigraphy indicated the skeletal abnormality of L1 and L4, with high suspicion of metastasis. (d) Posteroanterior and lateral X-ray films of the spine revealed instrumentation and cement augmentation were satisfactory. (e) Histopathology confirmed metastatic adenoid cystic carcinomas from parotid gland of spine (H and E, ×200). (f–j) Immunohistochemistry revealed neoplastic cells being CD117, CD5/6, P16, and P63 positive with 2% Ki-67-positive nuclei (×200). MRI: Magnetic resonance imaging.

of the metastatic ACC when the tumor has caused neurological deficits, especially myelopathy and radiculopathy.^[4] Radiation therapy has been suggested as an adjuvant treatment in cases with incomplete excision or with residual disease.^[4,5] In conclusion, we present an extremely unusual occurrence of spinal metastases of parotid carcinoma that was successfully managed by surgical procedure, though not been well documented before. Our focus is to emphasize the importance of considering spinal metastasis of salivary carcinoma as a diagnosis and guiding to the proper management strategy upon treatment.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initial will not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

REFERENCES

- Mannelli G, Cecconi L, Fasolati M, Santoro R, Franchi A, Gallo O, et al. Parotid adenoid cystic carcinoma: Retrospective single institute analysis. Am J Otolaryngol 2017;38:394-400. doi: 10.1016/j. amjoto.2017.03.008.
- Honda K, Tanaka S, Shinohara S, Asato R, Tamaki H, Maetani T, et al. Survival in patients with parotid gland carcinoma – Results of a multi-center study. Am J Otolaryngol 2018;39:65-70. doi: 10.1016/j. amjoto.2017.10.012.
- Liu H, Zhang Y, Zhang T, Li F, Dai C. Correlation between the pathology and clinical presentations in patients with adenoid cystic carcinoma of the external auditory canal. Head Neck 2017;39:2444-9. doi: 10.1002/hed.24915.
- 4. Xu MJ, Wu TJ, van Zante A, El-Sayed IH, Algazi AP, Ryan WR, *et al.* Mortality risk after clinical management of recurrent and metastatic adenoid cystic carcinoma. J Otolaryngol Head Neck Surg 2018;47:28. doi: 10.1186/s40463-018-0273-z.
- Wang Y, Yang L, Li W. Parotid gland as initial metastatic site of nasopharyngeal carcinoma. Chin Med J 2016;129:2265-6. doi: 10.4103/0366-6999.189914.

