Extradural Spinal Metastasis of Adenoid Cystic Carcinoma (ACC): A Case Report

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ABSTRACT

Adenoid cystic carcinoma (ACC) is a rare malignant tumour of the major salivary glands. It accounts for 10-15% of all salivary gland tumours and 1% of all head and neck tumours. Surgical resection followed by radiation is the choice of treatment for ACC. However, late loco-regional recurrence and metastasis is often seen emphasizing the importance of long-term follow-up.

We report an unusual case of extradural metastasis of ACC in the dorsal spine. The primary submandibular gland tumour was resected 11 years back. A recurrence had been detected two years prior to the occurrence of spinal metastasis. Surgical decompression was done which was followed by palliative radiotherapy. Patient is symptomatically better, ambulant and on regular follow-up.

Keywords: Adenoid cystic carcinoma, Compressive syndrome, Extradural spinal metastasis, Perineural invasion

CASE REPORT

History

A 45-year-old male presented to the neurosurgery out patient department with diffuse low back pain radiating to the right girdle since two months and inability to walk since two days. The pain was continuous in nature, exaggerated on activity and was persistent during the night with relief on taking analgesics. He had no associated co-morbid illness and or habituations. There was no history of trauma or tuberculosis.

Patient gave a past history of primary submandibular gland tumour resection 11 years back followed by radiation therapy. The diagnosis of ACC was rendered on the excised specimen (as per outside histopathology reports). He was advised an FDG-PET (Positron emission testing) scan, however differed the same. Nine years later, he developed a recurrent nodule at the previous scar and was detected to have recurrent disease on fine needle aspiration cytology. He underwent a wide local excision of the lesion with modified radical neck dissection (done at KMC, Manipal). The final histopathology was reported as ACC with negative margins and nodal metastasis (6 out of 12 isolated were positive for metastasis). He deferred palliative radiation therapy and was kept on regular follow-up.

Investigation

Magnetic resonance imaging (MRI) of the dorso-lumbar spine showed an extradural lesion at the level of D11 vertebra, hypointense on T1, hyperintense on T2 with contrast enhancement. The lesion measured 2.4 x 1.7 x1.5 cm with involvement of the right pedicle, articular pillar and lamina. The cord appeared displaced to the left with subtle T2w hyperintensity suggesting cord edema. Another enhancing T1 isointense and T2w hyperintense nodule measuring 3 x 2.7cm was seen in the postero-basal aspect of right lung suggestive of metastasis. Fatty marrow changes from C2 to C6 vertebral bodies were also noted indicating radiotherapy related changes [Table/Fig-1].

Operative and Postoperative Course

The patient underwent D10-D11 laminectomy with surgical excision of the lesion. Histopathological diagnosis of metastatic adenoid cystic carcinoma was preferred [Table/Fig-2]. There was immediate

Examination

On clinical examination patient had mild spastic gait (Ashworth grade 4) [1] with bilateral lower limb spasticity. Power was 5/5 in both lower limbs. There was sensory loss below L1 level on the right side, and below L3 on the left side with gross involvement of the posterior column with no involvement of bowel or bladder. Bilateral knee and ankle jerk were brisk (3+) [1]. Mouth opening was restricted with Grade II trismus, intraoral examination revealing a densely fibrosed buccal mucosa [2] on the irradiated side. Rest of the oral cavity examination was unremarkable. He had a keloid formation over the operative site.

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[Table/Fig-1]: Magnetic resonance image of the dorsolumbar spine – sagittal (a-d) and axial sections (g-i) showing a lesion, hypointense on T1, hyperintense on T2 with contrast enhancement at the level of D11 vertebra. Fatty marrow changes from C2 to C6 vertebral bodies (saggital cuts) (e) indicating post radiation changes.
and significant symptomatic relief following the surgery. The patient was ambulant on the first postoperative day and was discharged on the 8th postoperative day. Palliative systemic chemotherapy and spinal radiation therapy was given and the patient is neurologically better on follow-up after eight months of spinal surgery.

DISCUSSION

ACC is a rare tumour, which is difficult to treat. It occurs in the major and minor salivary glands of the skin and upper aero-digestive tract. Loco-regional recurrence and metastatic spread is well known. The common sites of metastases are lungs, bone, liver and brain. Metastasis to the spinal vertebrae is extremely rare though perineural invasion in margin positive surgical resections have been noted in literature [3-6]. Typically, low back or neck pain is the commonest presentation of spinal metastasis, followed by weakness, sensory loss, and bladder dysfunction [7].

ACC in the head and neck region is treated with surgical resection along with preoperative or postoperative irradiation. This reduces loco-regional recurrence and increases disease free survival rates [9,9]. Metastasis to the lymph nodes is rare and hence neck dissection is usually not warranted [10].

Histologically ACC has three different variants: glandular (cribriform), tubular and solid. Mitotic figures are generally scarce in cribriform and adenoid pattern with luminal mucoid material (H&E, X200). In the present case, ACC demonstrates its striking propensity for spinal metastasis, followed by new onset, progressive low back pain, mandates prompt radiological investigation in the form of an MRI of the affected vertebral segment and early surgical decompression/excision of the lesion.

CONCLUSION

To conclude, early clinical suspicion in a young patient with proven primary in the salivary gland with sudden onset, progressive low back pain, mandates prompt radiological investigation in the form of an MRI of the affected vertebral segment and early surgical decompression/excision of the lesion.

Our case demonstrates the recurrence and intraspinal metastasis, after nine years of diagnosis, of an ACC of the submandibular gland. The extradural location of the metastatic thoracic vertebral deposit mandates its rarity and anecdotal value.

REFERENCES