

Metastatic Spinal Tumor from Benign Pleomorphic Adenoma: Case Report and Literature Review

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Purpose: Pleomorphic adenomas tend to arise in the salivary glands. They are rare and histologically benign but can result in distant metastasis, and their characteristics need further investigation. We report a case of locally recurring benign primary palatal pleomorphic adenoma that resulted in spinal metastases and review the relevant literature.

Case Report: A 58-year-old woman had undergone surgery for a palatal pleomorphic adenoma 22 years earlier and 6 subsequent operations for local recurrences. During follow-up, metastases to multiple organs, including the spine, were diagnosed and 4 CyberKnife treatments were performed. She suffered right flank pain and slight paralysis of the right leg; radiological findings showed a growing metastatic spinal tumor. She underwent removal of a thoracic vertebral tumor and posterolateral fusion. Postoperatively, her symptoms improved. Histopathological analysis indicated a pleomorphic adenoma and no evidence of malignancy. Although there was no local recurrence, 23 months after surgery, a fifth CyberKnife procedure was performed for a growing salivary gland tumor and she is currently being followed up.

Conclusion: We described a rare case of benign pleomorphic adenoma that metastasized to the spine. Long-term follow-up for recurrence and metastasis is required for patients with benign pleomorphic adenoma. (*J Nippon Med Sch* 2023; 90: 121–125)

Key words: metastatic spinal tumor, pleomorphic adenoma

Introduction

Pleomorphic adenomas are the most common benign salivary gland tumors; they account for 60–70% of all salivary gland tumors¹. Approximately 80% of pleomorphic adenomas arise in the parotid gland; the other 20% are found in the submandibular and minor salivary glands¹.

Pleomorphic adenomas occur in children and adults, and the incidence is 1.4 times higher in females². They proliferate slowly, and histologically benign pleomorphic adenomas can elicit distant metastases^{1,3}. Knight et al.² reported that among 81 pleomorphic adenomas, 28 (34.6%) metastasized to bone, 26 (32.1%) to the lungs, and 17

(21.0%) to the cervical lymph nodes. They also observed metastases in the kidney, skin, liver, and brain. The incidence of metastatic spinal tumors is comparatively low, and our literature review found no comprehensive studies of their characteristics.

We describe a rare case of primary benign palatal pleomorphic adenoma that recurred locally despite multiple surgeries and resulted in spinal metastases. We also review the relevant literature.

Case Report

A 58-year-old woman had undergone surgery for primary palatal pleomorphic adenoma 22 years earlier. She

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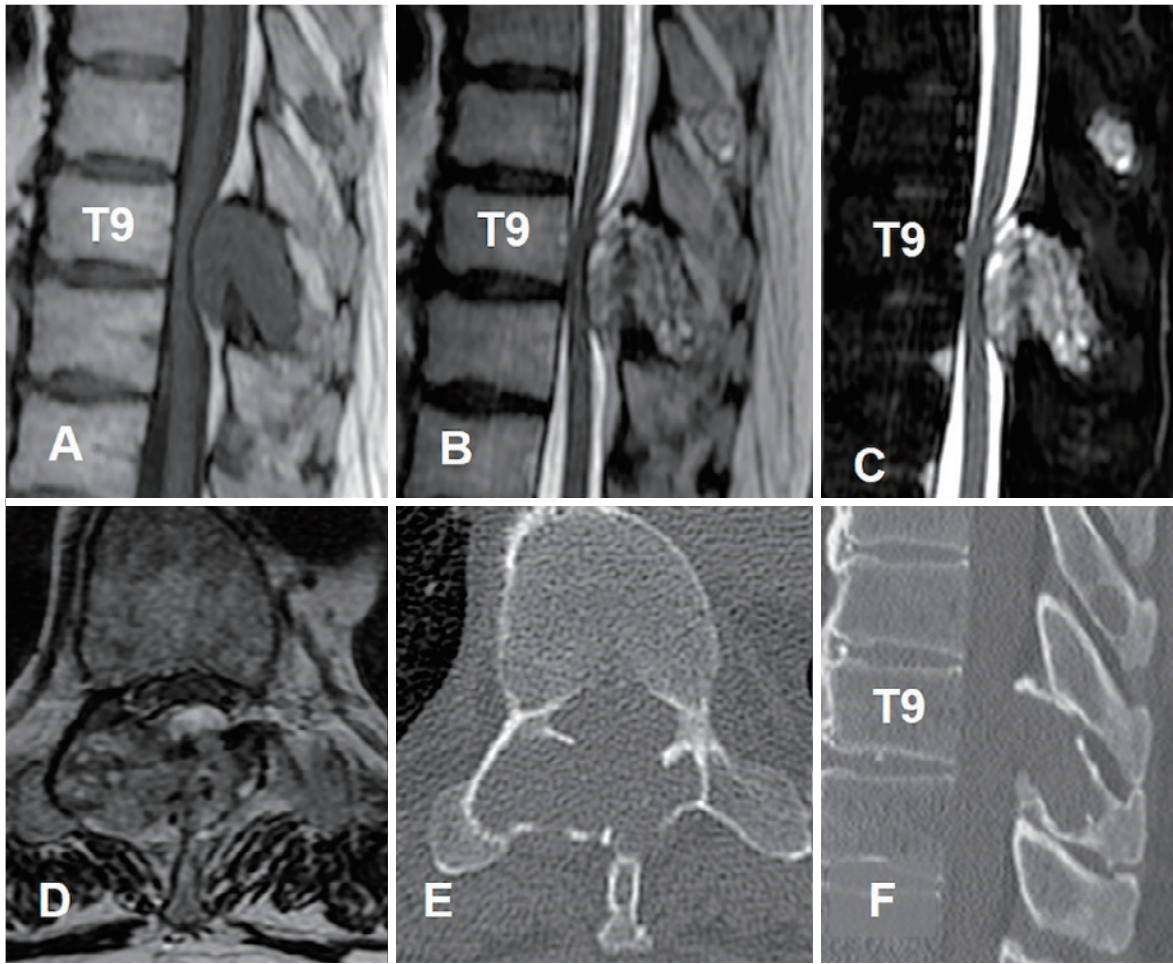


Fig. 1

A-D Thoracic spine MRI scans showing a posterior epidural mass severely compressing the spinal cord. Note the hypointense signal on T1-weighted images (**A**: sagittal view) and heterogeneous hyperintense signal on T2-weighted images (**B**: sagittal view, **D**: axial view) and T2 fat suppression (**C**: sagittal view). Similar signal changes were observed at the 7th spinous process.

E, F Thoracic spine CT scans (**E**: axial view, **F**: sagittal view) revealed bone erosion in the area of abnormal MRI findings.

later developed multiple local recurrences that were treated by 6 more operations. After 18 years of regular follow-up, multiple metastatic lesions, involving the ilium, ribs, spine, and supraclavicular lymph nodes, were detected and treated with 4 CyberKnife procedures. After diagnosis of local recurrence, positron emission tomography (PET) was performed every 6 months. Four months before admission to Chiba Hokuso Hospital, she reported pain in the right flank. We diagnosed growing metastatic thoracic spinal tumor, for which she had previously undergone CyberKnife treatment.

On admission she was conscious and had no neurological symptoms in the upper extremities, and no bladder or rectal disorders. She presented with right flank pain, sensory impairment in the sole of her left foot, and mild paralysis in the right lower extremity (manual mus-

cle test: 5-/5); Brown-Séquard syndrome was diagnosed. Babinski signs and myoclonus of the ankle joint in the right lower extremity were positive. A magnetic resonance imaging (MRI) scan of the thoracic spine revealed a tumor involving the vertebral arch; it severely compressed the spinal cord and involved the pedicle. A similar signal change was observed at the 7th spinous process. Computed tomography (CT) of the thoracic spine showed bone destruction in the abnormal signal area noted on MRI scans (**Fig. 1**).

After surgical removal of the spinal tumor, we performed posterolateral fusion. With the patient in prone position, we removed the spinous processes from the 7th to the 9th thoracic vertebrae and some of the muscle near the tumor where tumor invasion was suspected. The epidural tumor on the 9th thoracic level in the spinal ca-

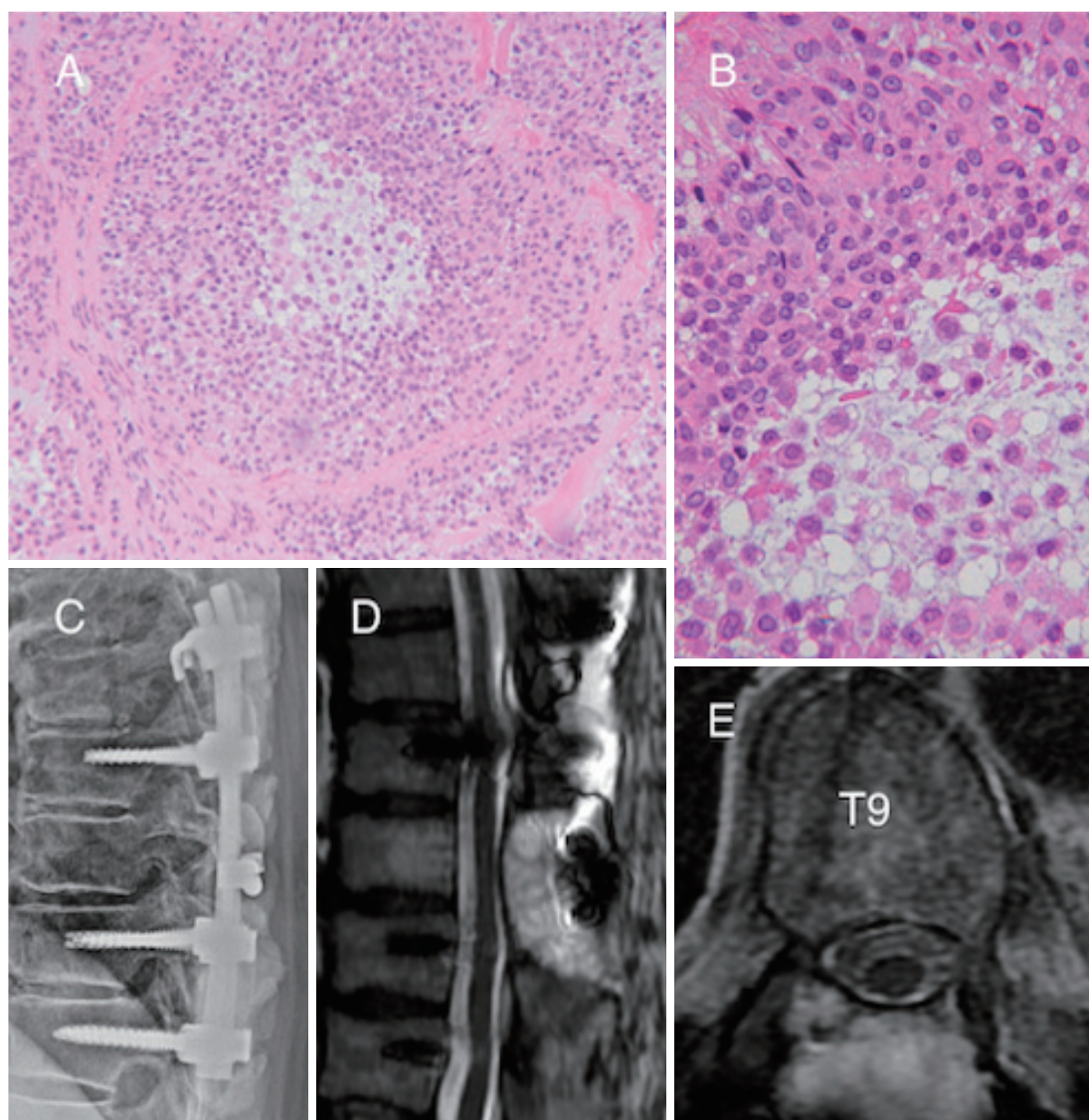


Fig. 2 Pathological findings for the surgical spinal tumor specimen.

- A, B** The diagnosis was pleomorphic adenoma. Small tumor cells formed a glandular tubule structure of the cord behind the myxoid stroma. Neither cellular dysmorphism nor tumor emboli were observed in the honeycomb-like blood vessels of the tissue (**A**: magnification $\times 100$, **B**: magnification $\times 400$)
- C-E** Postoperative X-ray image obtained 23 months after surgery (**C**: lateral view) showing posterolateral fusion from the 7th to the 11th thoracic vertebrae. Postoperative thoracic MRI scans of the 9th thoracic vertebral level performed 10 months after surgery (**D**: T2-WI, sagittal view, **E**: T2-WI, axial view) showed no further evidence of tumor recurrence at the surgical site.

nal bled easily and adhered to the dura; under a microscope it was dissected from the normal dura mater. Because the tumor invading the right pedicle of the 9th thoracic vertebra was highly hemorrhagic it was only partially resected. We completed posterolateral fusion of the 7th to 11th thoracic vertebrae (**Fig. 2**). The duration of surgery was 6.25 hours; blood loss was 50 mL.

Analysis of a surgical specimen of the spinal tumor yielded a diagnosis of pleomorphic adenoma; there were no findings suggestive of malignancy (**Fig. 2**). Postoperatively, her right flank pain and lower-extremity symp-

toms improved. There was no local tumor recurrence at the surgical site at 23 months postoperatively (**Fig. 2**). However, a fifth CyberKnife procedure was performed to address a growing salivary gland tumor. The patient is currently undergoing regular follow-up and her quality of life (QOL) has been preserved.

Discussion

Spinal metastasis from benign primary pleomorphic adenomas is rare: to date, only 17 cases have been reported in the literature and ours is the 18th case²⁻¹⁴. The male:fe-

male ratio was 1:1.6, and the mean interval from tumor detection to metastasis was 13.1 years (range 0-33 years). The mean patient age at the time of metastasis identification was 57.6 years (range 25-77 years). The most common site of metastasis in the 18 cases was the thoracic spine (8 cases; 44.4%), followed by the sacrum (5 cases; 27.8%), lumbar spine (4 cases; 22.2%), and cervical spine (1 case; 5.6%). Spinal metastases were identified during examinations for low back pain (5 cases; 27.8%), by PET or whole-body CT studies (4 cases; 22.2%) of patient complaints of leg paralysis (4 cases; 22.2%), after death (2 cases; 11.1%), and after patient complaints of bladder or rectal disorders and neck pain (1 case; 5.6% each)²⁻¹⁴.

Metastasis of pleomorphic adenoma to multiple sites is associated with a poor prognosis. Our literature search identified only 1 report of long-term follow-up of a patient with a metastasizing pleomorphic salivary adenoma¹⁵. In a previous study² of 81 metastatic pleomorphic adenomas, 13 (16.0%) resulted in metastasis at more than 3 sites, and 3 of these metastases were fatal within 3 years of diagnosis. Nouraei et al.¹⁵ reported that patients with metastases that developed within 10 years of an initial pleomorphic adenoma diagnosis had significantly worse outcomes than patients whose metastases were detected more than 10 years post-diagnosis. Their review of 42 pleomorphic adenomas revealed that 34 (81%) recurred locally at least once before detection of distant metastases. Patients with local recurrence or longstanding pleomorphic adenoma are more likely to develop distant metastases; thus, such metastases should be ruled out by systemic examinations with CT and/or PET¹⁶. We conducted FDG-PET studies at 6 months after diagnosis of local recurrence and addressed early systemic metastases, thus maintaining good QOL.

Among 17 previously reported cases of metastatic spinal tumor²⁻¹⁴, 8 (47.1%) involved 3 or more sites, 2 were fatal within 3 years of their detection, and 5 involved patients who were not followed for longer than 2 years. Although our patient had metastases at multiple sites, the interval from first detection to diagnosis of distant metastasis was 18 years. During that period she underwent repeat operations and CyberKnife treatments and was able to pursue her daily activities independently. We suggest that patients with metastases from primary pleomorphic adenomas should receive CyberKnife treatments as necessary for local control of the disease.

Conclusion

We described a rare case of benign pleomorphic adenoma that metastasized to the spine. Such cases require

long-term follow-up to ensure early detection of tumor recurrence and metastasis. Although Cyberknife procedures may help control metastatic or recurrent lesions, surgical intervention should be considered in cases of symptomatic lesion enlargement.

Conflict of Interest: None.

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