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Case Report

Thymoma dissemination through the thoracic intervertebral foramen: Pleural recurrence resulting in spinal cord compression

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Abstract

Background: Spinal dissemination of thymic tumors is rare but should be considered in the differential diagnosis of thoracic dumbbell-shaped lesions and/or vertebral tumors, irrespective of the time since the initial diagnosis.

Case Description: A 63-year-old man, with a history of invasive type AB thymoma treated 21 years ago, newly presented to the hospital with a dumbbell-shaped T8-T9 lesion compressing the spinal cord. A review of the literature showed only 16 previous cases of thymic tumors with thoracic spine involvement. Here, we report the lengthiest interval between the initial tumor diagnosis and the detection of spinal involvement, that was secondary to a pleural recurrence from his thymoma. The patient did well following successful excision of the intraspinal mass which had encased the T8 nerve root.

Conclusion: Spinal dissemination of thymic tumors can occur due to vertebral metastasis or to extension of a pleural recurrence through the intervertebral foramen. Definitive treatment for spinal lesions should be considered to provide adequate cord decompression.

Key Words: Thymoma, thymic tumor, spinal cord compression, pleural recurrence, spinal metastases

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INTRODUCTION

Thymomas are infrequently encountered. They are slow-growing and typically benign mediastinal tumors originating from epithelial cells of the thymus. The most aggressive tumor variant are thymic carcinomas, which are found on rare occasions.[10,13] Less than 10% of thymic tumors show pleural dissemination;[11] and extension through the intervertebral foramen from pleural nodules is an exceptional event.[9,11] If extrathoracic metastases develop, they generally spread to lymph nodes, the liver, and soft tissue/skeletal muscles.[15] Involvement of the thoracic spine by thymic tumors is very rare and typically involves the vertebral bodies.[1,4] Here, we describe a thoracic spine dissemination of a pleural recurrence from thymoma through the intervertebral foramen, resulting in spinal cord compression, 21 years after initial diagnosis. Fortunately, it was successfully managed with secondary definitive surgery.

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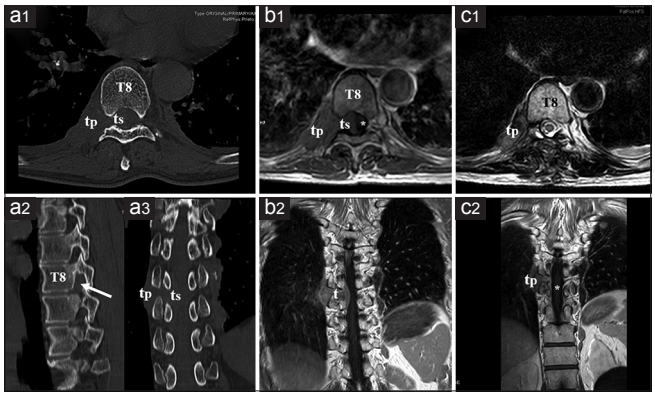

Figure 1: Preoperative and postoperative neuroradiological studies. (a) Preoperative computed tomography: axial (a_1), sagittal (a_2), and coronal (a_3) scans showing an irregular tumor with pleural (tp) and spinal (ts) components. Note that the vertebral foramen is slightly enlarged (arrow) and the adjacent bone structures are not infiltrated. T8: Eighth thoracic vertebrae. (b) Preoperative MRI: axial (b_1) and coronal (b_2) MRI scans demonstrating a dumbbell-shaped mass along the course of the right eighth nerve root. The spinal cord (asterisk) is displaced to the left. (c) Postoperative MRI shows an adequate decompression of the spinal cord (asterisk) and persistence of the pleural tumor component (tp)

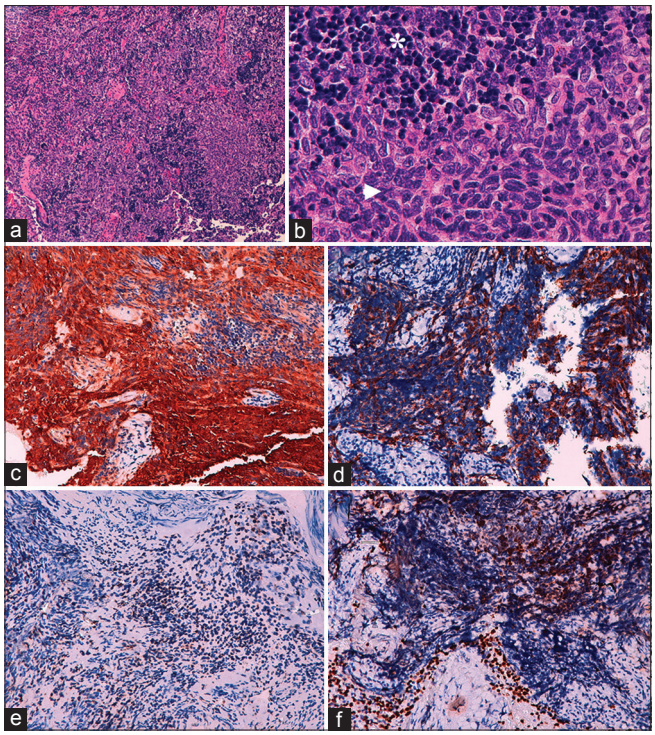

Figure 3: Photomicrographs of the surgical specimen. Hematoxylin–eosinstain ((a) 10×, (b) 40×). The neoplasm consisted of a mixture of poligonal (oval) and spindle-shaped epithelial cells (arrowhead), small-to-medium sized, arranged in nests, and surrounded or intermingled with nonneoplastic small mature-appearing lymphocytes (asterisk). The epithelial cells show vesicular oval nuclei with nucleoli and pale cytoplasms. Immunohistochemistry stains (c–f): The epithelial nature of the polygonal and spindle-shaped cells was supported by their immunostaining for low- and high-molecular weight cytokeratin markers CKAE1-AE3 (c) and CK5-6 (d). The nonneoplastic, immature lymphoid population intermingled with the epithelial cells showed positivity for CD3 (e) and TdT (f)

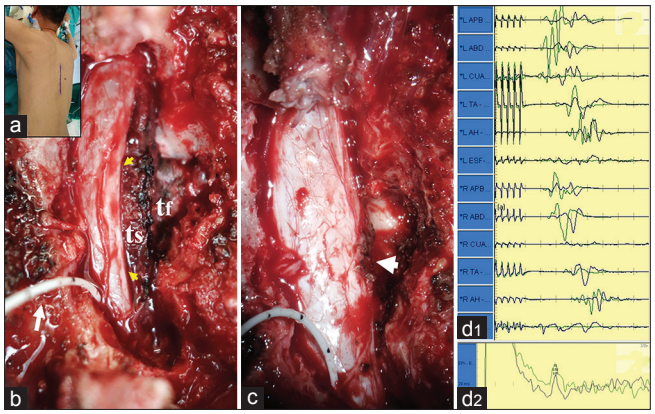

Figure 2: Intraoperative photographs and neurophysiologic monitoring. (a) The patient is placed in a prone position. A vertical midline incision was made centered at the T8 level. (b) Following a one-level laminectomy and extensive foraminotomy, the intraspinal (ts) and foraminal (tf) components of the tumor were exposed. Observe the notable spinal cord compression (yellow arrows). White arrow points to the D-wave cable placed in the epidural space below the tumor. (c) The spinal cord has been decompressed. Two nonabsorbable stitches (arrowhead) were placed at the anatomical area of the T8's right exiting nerve root, where a small dural hole was found after tumor removal. Motor evoked potentials to transcranial electrical stimulation (d_1) and D-wave (d_2) monitoring. No changes were observed between basal recordings (green waves) and those measured at the end of tumor removal (black waves)

CASE DESCRIPTION

A 63-year-old male presented with dorsal pain of a 3 month duration. He had undergone gross-total resection of an invasive mixed AB thymoma (Masaoka-Koga II–III) 21 years ago, followed by local radiotherapy (52 Gy). After staying disease free for 12 years, he developed three pleural nodules; subsequent biopsy demonstrated pleural recurrent thymoma that was then treated with gamma knife radiosurgery (13 × 300 cGy in each lesion). The pleural lesions remained stable for the next 10 years. The patient was referred to our institution at the time radiographic spinal cord compression at T8-T9 level was discovered in addition to pleural thickening involving both hemithoraces [Figure 1a]. Examination did not evidence any neurological deficits.

MR findings

The magnetic resonance imaging showed an irregular, dumbbell-shaped lesion at the T8–T9 level with an intraspinal component occupying 60% of the canal, displacing the cord toward the left [Figure 1b].

Surgery

The spinal and foraminal components of the tumor were removed through a one-level laminectomy with right-sided foraminotomy and medial T8–T9 facetectomy [Figure 2a]. The right T8 root was encased by an epidural solid tumor mass [Figure 2b]. Following gross total resection, a small dural hole was repaired at the right-sided T8 exiting nerve root [Figure 2c].

Intraoperatively, neurophysiologic recordings remained stable [Figure 2d]. The patient's postoperative course was uneventful. Postoperative MRI confirmed complete removal of the spinal tumor component [Figure 1c]. He declined postoperative adjuvant chemotherapy. Six months postoperatively, he remained neurologically intact without pain.

Histologic Diagnosis

The histological analysis revealed an admixture of neoplastic epithelial cells and nonneoplastic small lymphocytes. The morphological and immunostaining features of the tumor corresponded to metastatic thymoma [Figure 3].

DISCUSSION

In the literature, we could only find 16 previous cases of thymic tumors involving the thoracic spine [Table 1]. These included metastasis to the bony elements of the vertebrae (10 out of 16 cases)[2-4,7,15] and growth of pleural nodules into the spinal canal through the intervertebral foramen (6 cases).[1,6,8,12,14] The latter lesions should be considered in the differential diagnosis of thoracic dumbbell-shaped lesions. Regarding the histological type, despite representing less than 10% of thymic tumors, three quarters of reported cases with thoracic spine involvement corresponded to thymic carcinomas.[13]

Table 1: Disseminated thymic tumors to the thoracic spine

Author (Reference)	Age, sex	Interval from initial diagnosis to spinal involvement (years)	WHO type[§] of thymic tumor	Masaoka-Koga stage[†] at initial diagnosis	Spinal level (compartments involved)	Treatment of spinal tumor	Outcome after spinal surgery
Alafaci *et al*.[2]	33, F	1	Thymoma	NA	T4 (NA)	Partial removal	Death 2 months later
Farin *et al*.[4]	45, M	12	Thymoma	NA	T11-T12 (VB + posterior + extradural)	Subtotal removal + RT	Disease free at 9 months
Gamboa *et al*.[5]	77, F	1.25	Thymoma (A)	I	T2-T3 (intradural)	RT	Death 2 months later
Toba *et al*.[14]	29, F	3.75	Thymoma (B3)	IVa	T10-T11 (PV + foramen: Eden IV)	Total removal + RT	Disease free at 15 months
Vladislav *et al*.[15]	31, M	0*	Thymoma (B3)	IVb	T10 (NA)	NA	NA
Hong *et al*.[6]	62, F	13	Thymic carcinoma	NA	T9-T10 (PV + foramen + extradural: Eden III)	Total removal	Death 2 years later
Jee *et al*.[7]	61, M	0*	Thymic carcinoma	NA	T4-T5 (extradural)	NA	Death 2.7 years later
	42, M	2	Thymic carcinoma	NA	T3-T5 (extradural)	NA	Death 1.1 years later
	36, M	5	Thymic carcinoma	NA	T2-T4 (extradural)	NA	Death 16 days later
	57, M	5	Thymic carcinoma	NA	T4-T6 (extradural)	NA	Death 3.7 months later
	59, F	0*	Thymic carcinoma	NA	T5 (extradural)	NA	Death 7.6 months later
	54, F	14 (Thymoma) 2 (Thymic carcinoma)	Thymoma (B2) → Thymic carcinoma	NA	T11 (VB + extradural)	Partial removal + RT	Death 8.4 months later
Sasaki *et al*.[12]	50, F	0*	Thymic carcinoma	IVa	T2-T3 (PV + foramen + extradural: Eden III)	Partial + RT + ChT	Alive 1.5 years later
Low *et al*.[8]	79, M	12	Thymic carcinoma	NA	T8-T9 (PV + foramen: Eden IV)	Biopsy + RT	Alive 1 year later
Alekseyev *et al*.[3]	29, M	0*	Thymic carcinoma	IVb	T10-L1 (VB + extradural)	Biopsy + RT	NA
Achey *et al*.[1]	38, F	2	Thymic carcinoma	NA	T12-L1 (PV + foramen + extradural: Eden III)	Total removal + ChT	NA
Present case	63, M	21	Thymoma (AB)	II-III	T8-T9 (PV + foramen + extradural: Eden III)	Partial removal	Alive 6 months later and in good general condition

ChT=Chemotherapy; F=female; M=male; NA=not available; PV=paravertebral; Posterior=posterior elements; RT=radiotherapy; VB=vertebral body; 0* = spinal involvement at initial diagnosis. Eden classification of dumbbell-shaped tumors: type III=extradural and paravertebral; type IV=foraminal and paravertebral. [†]Masaoka-Koga staging (tumor invasion and dissemination): I=completely encapsulated; II=macroscopic invasion into surrounding fatty tissue or mediastinal pleural, or microscopic invasion beyond the thymic capsule; III=macroscopic invasion into neighboring organs; IVa=pleural or pericardial dissemination; IVb=lymphogenous or hematogenous metastasis. [§]World Health Organization type (histological appearance of the neoplastic epithelial cells): A, AB, B1, B2, B3, and carcinoma

There is a lack of consensus regarding the optimal management of thymic tumors in advanced stages and specifically how to treat the rare cases that exhibit spinal disseminations.[13] Recent studies suggest a longer survival after complete resection of pleural recurrences.[11] In the present case, resection of the intrathoracic nodules was not carried out owing pleural adhesions caused by previous radiotherapy treatment and the lack of respiratory symptoms. The intraspinal dissemination 21 years after the original thymic surgery, however, warranted aggressive surgical extirpation.

CONCLUSION

Spinal dissemination from thymic tumors is rare and should be included in the differential diagnosis of thoracic dumbbell-shaped lesions and vertebral tumors. Definitive treatment and surgery should be based on the status of the disease and the patient's clinical condition. As these are slow-growing mediastinal lesions, early decompression of the spinal cord may avoid the subsequent onset of progressive neurological dysfunction.

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Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest
There are no conflicts of interest.

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