Successful treatment of malignant thymoma with sacrum metastases

A case report and review of literature

Shuzhong Liu, MD, PhD*a, Xi Zhou, MD, PhD*a, An Song, MD, PhDb, Zhen Huo, MD, PhDc, William A. Li, MD, PhDd, Radhika Rastogi, MD, PhDe, Yipeng Wang, MD, PhDe, Yong Liu, MD, PhDe

Abstract

Rationale: Malignant thymoma in the spine is a rare disease without standard curative managements so far. The objective of this article is to report a very rare case of recurrent malignant thymoma with sacrum metastases causing severe lumbosacral pain, which was presented with acute radiculopathy and treated with 2 operations combined with stabilization and cement augmentation. The management of these unique cases is not well-documented.

Patient concerns: A 75-year-old man presented with lumbosacral pain, radiating pain and numbness of the left extremity. The patient underwent thymectomy in 2008, and posterior spinal cord decompression, tumor resection and a stabilization procedure in 2011. Pathologic results confirmed malignant thymomas of the spine. Imaging studies revealed the density of soft tissues, obvious bony destruction in the sacrum, and significant spinal cord obstruction.

Diagnoses: We believe this is a less-documented case of metastatic thymoma of the sacral spine presenting with back pain and radiculopathy, and presenting as a giant solid tumor.

Interventions: The patient underwent osteoplasty via a posterior approach. Pathologic results confirmed malignant thymomas of the sacral spine.

Outcomes: The patient’s neurological deficits improved significantly after the surgery, and the postoperative period was uneventful at the 6-month and 1-year follow-up visit. There were no other complications associated with the operation during the follow-up period.

Lessons: This article emphasizes metastatic thymoma of the spine, although rare, should be part of the differential when the patient presents with back pain and radiculopathy. We recommend the posterior approach for spinal decompression of the metastatic thymoma when the tumor has caused neurological deficits. Osteoplasty by cement augmentation is also a good choice for surgical treatment.

Abbreviations: MRI = magnetic resonance imaging, T1WI = T1-weighted image, T2WI = T2-weighted image, VAS = visual analogue scale.

Keywords: cement augmentation, metastatic spinal thymoma, sacrum, stabilization, surgical treatment

1. Introduction

Thymoma and thymic carcinoma are uncommon epithelial entities, which originate from the thymus gland. To date, the true incidence is not known, but it is estimated to be 0 to 15 cases per 100000 individuals and represents 0.2% to 1.5% of all malignancies.[1] According to the World Health Organization, there are “organotypic” (types A, AB, B1, B2, and B3) and “nonorganotypic” (type C, thymic carcinomas) thymomas.[1,2] Types A, B, B1, and B2 thymomas are benign tumors, while type B3 thymomas are aggressive tumors of intermediate malignancy. Spinal metastases of malignant thymoma (type B3) are extremely rare in literature, only very few patients with spinal metastasis have been documented, thus there is still short of imaging proof. Herein, we are presenting a detailed analysis of a rare case of spinal metastases of thymoma treated with stabilization and cement augmentation.

To the best of our knowledge, this is a rare case of metastatic thymoma to the spine in a man presenting with back pain and radiculopathy. We performed osteoplasty through a posterior approach. In the short term, the patient’s conditions improved significantly post-operatively. After reviewing pertinent literature, we discussed common perioperative considerations in patients with significant metastatic thymomas to the spine and management considerations for these cases.
2. Case report

In May of 2017, a 75-year-old man presented to our hospital, with progressive back pain, radiating pain and numbness of his left lower limb. The patient, having been diagnosed of B3 thymoma for 9 years, received surgical treatments including thymectomy in 2008, and posterior spinal cord decompression, tumor resection and a stabilization procedure due to the spinal metastases of malignant thymoma followed by postoperative radiation in 2011. In the medical journal of his current illness, the patient stated he had been experiencing paroxysmal and severe back pain for approximately 3 months, and he had also experienced a worsening numbness and radiating pain of his left lower limb for approximately 3 months. The pain in his back can reach 6 to 7 points using visual analog scale (VAS) and cannot be alleviated with rest and hot compresses. The patient denied experiencing any other constitutional symptoms. Upon further questioning, he recalled a history of old myocardial infarction since 2003. No pertinent family history was identified, including hypertension and cancer.

On physical examination, the patient showed pressure pain and percussion pain in his sacral region decreased sensation to pin-prick and fine-touch of his left lower limb and exhibited 5-/5 strength in his bilateral lower limbs. Deep tendon reflexes revealed normal for knee-jerk and Achilles tendon reflexes bilaterally. Ataxia, cranial nerves, mini-mental, and the rest of the neurological examination showed no abnormalities. Preoperative hemodynamic and cardiovascular assessments included electrocardiogram, echocardiogram, and chest radiography. Preoperative laboratory assessment was conducted, including routine laboratory tests (electrolytes, liver, and kidney function tests, complete blood count), tumor markers, myocardial enzymogram, and screening for myasthenia gravis. The results of the laboratory studies were almost within normal range. X-rays revealed sacral lesions, with high suspicion of spinal soft tissue tumors (Fig. 1 A, B). Spinal magnetic resonance imaging (MRI) was ordered to visualize the metastatic lesions, assess the stability of the vertebral column, and to aid in the formulation of a surgical approach. MRI of the spine showed the density of soft tissue measuring 10cm × 7.5cm × 5.7cm, obvious bony destruction in the sacrum, and spinal cord compression secondary to the giant mass, with increased metastatic marrow infiltration of the sacrum (Fig. 2 A–I). Tumor infiltrated through the sacrum body into the posterior elements, thus extraosseously spread into the bilateral aspects of the epidural space extending posteriorly, resulting in spinal cord compression (Fig. 3 A, B). The bone scanning revealed high intake in the sacral spine, with high suspicion of spinal metastases (Fig. 4).

Subsequently, osteoplasty with cement augmentation was performed to destroy the functional tumor and stabilize the spine under local anesthesia. In brief, percutaneous vertebroplasty at sacrum was performed according to the original surgical plan. For the posterior approach, we used C-arm for perspective positioning, bilateral vertebral lesions of sacrum were identified as surgical targets, and the bilateral sacral pedicle puncture points were located. Then 2% lidocaine was used for local infiltration anesthesia, and the puncture needle was inserted through the cannula. Under the C-arm fluoroscopy, the vertebral lesion was penetrated through the left pedicle of the sacrum first, and bone cement for vertebroplasty was introduced. Under the perspective, the 10.0mL cement of left side was slowly pushed through the putter, and the biopsy passage was closed. In the same way, the right S1 vertebral pedicle was then punctured to the right S1

Figure 1. (A,B) Preoperative X-rays revealing sacral lesions with high suspicion of spinal metastatic soft tissue tumors.
Figure 2. (A–H) Preoperative sagittal MRI scan revealing the density of soft tissue measuring 10 cm × 7.5 cm × 5.7 cm, obvious bony destruction in the sacrum, and spinal cord compression caused by metastatic malignant thymoma, with increased metastatic marrow infiltration of the sacrum. MRI = magnetic resonance imaging.

Figure 3. (A,B) Preoperative coronal and transverse MRI images showing sacral metastases of B3 thymoma. MRI = magnetic resonance imaging.
vertebral body lesion, and 9.6 mL bone cement was slowly pushed into the right S1 vertebral body lesion through a push rod under fluoroscopy, and the biopsy pathway was closed. Fluoroscopy confirmed the good dispersion of bone cement. The operation was successful and intraoperative bleeding was about 60 mL. Postoperative posteroanterior and lateral radiographs of the spine showed cement augmentation was satisfactory (Fig. 5 A and B). The patient was unwilling to undertake any further treatments and was discharged and monitored on an outpatient basis. The postoperative pathology report confirmed a malignant B3 type thymoma, which was consistent with the primary tumor (Fig. 6 A–D). Pathological analysis was positive for AE1/AE3 indicating epithelial origin from thymus gland. Biopsy samples were negative for chromogranin A, synaptophysin, CD56 (NK-1), TTF-1, CD5, with 30% Ki-67 positive nuclei (Fig. 7 A–F). Consequently, malignant thymoma with sacrum metastases was diagnosed via history taking, laboratory values, imaging results, and pathological studies.

One week after the operation, the patient’s muscle strength of lower extremities improved to grade V compared to the preoperative status, and the symptoms were relieved significantly. Moreover, VAS score of his back pain improved to 0 to 1 points compared to the preoperative status, 6 to 7 points. Postoperatively, the patient underwent rehabilitation therapy and was discharged and monitored as an outpatient. The postoperative 6-month and 1-year follow-up visit showed no tumor progression and no new symptoms. There were no other complications associated with the operation during the follow-up period.

3. Discussion
Malignant thymomas are rare tumors that were usually determined by the invasiveness into nearby tissues or distant metastasis. On the basis of the appearance of epithelial cells, the World Health Organization unified classification proposed 3 histological types of thymomas (types A, AB, B1, B2, B3, and C). The incidence of thymomas has been estimated at 0.13 case per 100,000 individuals per year. Malignant thymomas (type B3) are rare tumors which show pathological features of malignancy and highly aggressive biological behavior with extrathoracic metastases to the liver, kidney, and lymph nodes. However, it is extremely rare for type B3 thymomas to metastasize to the spine. Back pain and radiculopathy can often mimic the most common cause of spinal diseases, making timely diagnosis of spinal metastatic thymomas difficult without a high level of suspicion.

In literature, this is a rarity of recurrent metastatic thymomas of the sacrum presenting with back pain and radiculopathy. Clinical studies looking at metastatic thymomas and thymic carcinomas to the spine are lacking due to the extremely low incidence rate. Based on our review of the 33 case reports on PubMed (Table 1), metastatic thymomas of the spine is slightly more common in the thoracic region and are more commonly diagnosed during the fourth and the fifth decades of life for the sporadic form (mean age: 51.5 years; range: 29–79 years).

The location of the spinal lesion determines the neurological deficits, and there is a great deal of variability. Compression of the cervical vertebra often exhibit symptoms of paresthesia and weakness in the upper extremities, while those located in the thoracic and lumbar regions usually show symptoms of low back pain, lower extremity paresthesia, weakness, and dysuria. In our case, the patient sought medical attention after experiencing serious back pain, lower extremity weakness, and numbness. To the best of our knowledge, this is the first reported case of recurrent malignant thymoma with sacrum metastases. Imaging
studies including X-ray, computed tomography (CT), MRI, and bone scan are non-specific, making it difficult to differentiate metastatic spinal thymoma from other common spinal lesions. However, imaging studies play a crucial role in the surgical intervention decision making. Imaging studies can demonstrate consecutive spinal stenosis, spinal cord compression, and pathological vertebral fractures. MRI images from previous case reports demonstrate inhomogenous lesion of spine, isointense on T1-weighted image (T1WI) and hyperintense on T2-weighted image (T2WI), indistinguishable from other metastatic spinal lesions. Heterogeneous enhancement, calcification, and cystic changes are rarely observed.

Our patient’s MRI showed isointense on T1WI and hyperintense lesion on T2WI, which is consistent with previous case reports.

Thymomas (type B3) may become malignant via their metastatic tendency, and the metastases can help us diagnose a malignant type B3 thymoma. Although local spreading occurs rapidly, distant spinal metastasis may occur within a long period, up to 24 years after the diagnosis of thymomas. The “gold-standard” diagnosis of thymoma relies on pathological findings. Histopathologically, metastatic spinal thymomas are characterized by an architecture of nests of tumor cells separated by vascular septa with the cells showing significant nuclear pleomorphism with prominent nucleoli, and large amount of lymphocytes. Generally, metastatic thymomas are commonly immunoreactive for AE1/AE3. A histological examination of our case was positive for AE1/AE3 indicating thymoma from epithelial cells of the thymic gland. Biopsy samples were negative for chromogranin A, synaptophysin, CD56 (NK-1), TTF-1, CD5, with 30% Ki-67 positive nuclei, which confirmed the diagnosis of metastatic type B3 thymoma.

Currently, no treatment guidelines exist because of the variability in treatment modalities and reported outcomes. Surgical resection is the mainstay of treatment for thymic tumors and spinal metastatic lesions. We recommend surgical management of the spinal metastatic tumor when the tumor has caused neurological deficits, spinal cord compression, or destruction of spinal stability. The best treatment for metastatic spinal thymomas causing acute partial paralysis and lower back pain is posterior decompression, tumor resection, and internal fixation. This protocol accomplishes 2 objectives: it alleviates the neurological deficits by decompressing the stenosis and at the same time provides histopathological specimens for diagnosis, which is valuable in cases where the patient presents with atypical clinical and radiological findings. The highly vascular nature of the tumor and its potential for infiltration makes total resection difficult, thus recurrence is likely. The survival benefit of resection of spinal metastases is still unproven. However, such a procedure does have the benefit aiming at controlling residual tumor and is recommended for most patients. This survival benefit of reducing the tumor burden, decompressing the spinal stenosis to alleviate radicul-
Figure 6. Pathologic histology of primary type B3 thymoma. (A, B) Microphotography showing significant nuclear pleomorphism with prominent nucleoli (H&E, original magnification 100× and 200×). (C) CK19 immunostaining is strongly positive in the epithelial cells. (D) Ki-67 immunostaining shows 15% Ki-67 positive cells. Ki-67 staining is localized in the tumor nuclei.

Figure 7. Pathologic histology of spinal metastases. (A–C) Microphotography showing characteristic nests of tumor cells separated by vascular septa (Zellballen) with cells showing significant nuclear pleomorphism with prominent nucleoli (H&E, original magnification 100×, 200×, and 200×). (D) AE1/AE3 immunostaining is strongly positive in the epithelial cells. (E) CDS immunostaining shows positive staining in the tumor cells. (F) Ki-67 immunostaining shows 30% Ki-67 positive cells. Ki-67 staining is localized in the tumor nuclei.
## Table 1
Clinical review of 33 previously published metastatic thymomas and thymic carcinomas of spine.

| Authors            | Year | Age (y) | Sex | Symptoms and Signs             | Thymoma Type (WHO) | Spinal Metastases Location | Intratrual or Extraparital | Incomplete Paraplegy or Paralysis | Myasthenia Gravis | Years since Initial Diagnosis | Resection of Primary Lesion | Surgical Treatments | Adjuvant Treatment | Postop Complications and Outcome |
|--------------------|------|---------|-----|---------------------------------|--------------------|---------------------------|--------------------------|----------------------------------|-------------------|-----------------------------|----------------------|-----------------------|----------------------|----------------------------------|
| Posner et al.      | 1977 | 32, M   |     | Midback pain and radiated down legs and into toes; constipation; mild paraparesis | Lymphoid and epithelial thymoma | Th3 to Th2 | Extradural | No | 1 | Thymectomy | None | Radiation and chemotherapy | Not reported |
| Alemata et al.     | 1993 | -       | BLE paralyis | Staging of bilateral paraparesis and urinary incontinence | Staging M0 | Thoracic | Extradural | | No | 1 | No | Not reported | Exploratory throracotomy | Radiation and chemotherapy | Patient died of paraneoplastic postoperatively |
| Selvaraj et al.    | 1999 | 73, M   |     | Neck pain; BLE paresthesias; gait unsteadiness | Mixed cell thymoma | C3 | Extradural | No | Yes | 10 | Thymectomy | Excisional biopsy at C3 | Radiation | Initial resolution of symptoms; ocular myasthenia gravis developed | MB improvement of symptoms; died of respiratory failure shortly after surgery |
| Alafaci et al.     | 1999 | 33, F   |     | BLE weakness; BLE paresthesias; BLE hyporeflexia | Mixed invasive thymoma, epithelial predominance, low differentiation, clusters of lymphocytes | T4 | Extradural | No | No | 1 | Thymectomy | T4 laminectomy with tumor decompression | None | In the postoperative period, the patient had near complete relief of her axial and radicular pain |
| Hentschel et al.   | 2004 | 60, F   |     | Axial thoracic spinal pain with a severe radicular component | C | T7 | Extradural | No | No | 2 | Thymectomy | Percutaneous vertebroplasty with cement augmentation | Radiation and chemotherapy | In the postoperative period, the patient had near complete relief of her axial and radicular pain |
| Posner et al.      | 2004 | 30, F   |     | Back pain; RLE pain | Predominantly lymphocytic malignant thymoma | T11-T12, L5 | Extradural | No | No | 8 | Not reported | None | Radiation and chemotherapy | Patient died 6 years after presenting with back pain |
| Oguri et al.       | 2004 | 64, F   |     | Hoarseness; dysphagia | C, squamous cell | C3-C4 | Extradural | No | No | 0 | CT-guided biopsy | No surgical intervention for spinal involvement | Chemotherapy | Multi-site recurrence with rapid enlargement of tumor |
| Forin et al.       | 2005 | 45, M   |     | Progressive back pain; diminished sensation in bilateral toes | Dense, fibrous, cellular epithelium with mature lymphocytes | T11-T12 | Extradural | No | Yes | 12 | Thymectomy | T1-T2 laminectomy and partial corpectomy, GTR, pedicle screw fixation | Radiation and chemotherapy | Initial improvement with ability to ambulate; disease free 9 months postoperatively |
| Toba et al.        | 2009 | 29, F   |     | Back pain | C | T10-T11 | Intratradular | No | Yes | 4 | Thymectomy | T1-2 laminotomy with tumor decompression | Radiation and chemotherapy | No sign of further recurrence for 15 months |
| Hu et al.          | 2011 | 67, M   |     | Monoparesis of leg | Carcinoid tumor of the thymus | T3, T8, L5 | Extradural | Yes | No | 16 | Not reported | Tumor resection via laminectomies T2-3, T8-9 and L4-5 | None | Died 1 year later |
| Liu et al.         | 2011 | 57, M   |     | Progressive cervico-dorsal pain and decreased sensation in both hands; decreased grip strength | Poorly differentiated squamous TC | C4-T1 | Extradural | No | No | 0 | Not reported | C5-7 laminectomies, tumor resection, and C7-C6 posterolateral internal fixation and fusion | Radiation and chemotherapy | Symptoms resolved; initially nuceeved well, died from respiratory failure 5 months after surgery |
| Madlansay et al.   | 2012 | 66, M   |     | Chest symptoms | B3 | L1 | Extradural | No | No | 2 | Not reported | Not reported | Not reported | Not reported | Not reported |
| Madlansay et al.   | 2012 | 46, M   |     | Ostealgia, chest symptoms of systemic symptoms | B1-B2 | L1-L3 | Extradural | No | No | 0 | Not reported | Not reported | Not reported | Not reported | Not reported |
| Hong et al.        | 2012 | 31, M   |     | Chest symptoms | B3 | T10 | Extradural | No | No | 0 | Not reported | Neurally total resection preserving the nerve roots was performed via a partial L4 and L5 hemilaminectomy | Radiation and chemotherapy | Neurally total resection preserving the nerve roots was performed via a partial L4 and L5 hemilaminectomy |
| Hong et al.        | 2013 | 62, F   |     | Segmental thoracic pain | C, well-differentiated thymic carcinoma | T9-T10 | Extradural | No | No | 13 | 6 surgical procedures for resection of a thymic carcinoma during a period of 13 years | A gross-total tumor resection was performed via a costotransversectomy and facetectomy | Radiation and chemotherapy | Neurally total resection preserving the nerve roots was performed via a partial L4 and L5 hemilaminectomy |
| Jee et al.         | 2014 | 61, M   |     | Paraparesis | C | T4-T5 | Extradural | No | No | 0 | Not reported | Decompression, tumor resection, and pedicle screw fixation | Radiation or chemotherapy | Died 3 years after surgery |
| Jee et al.         | 2014 | 42, M   |     | Paraplegia | C | T3-T5 | Extradural | Yes | No | 2 | Not reported | Decompression, tumor resection, and pedicle screw fixation | Radiation or chemotherapy | Recurrence of paraparesis, died < 1 year postoperatively |

(continued)
| Authors                  | Year | Age (y), Sex | Symptoms and Signs                                                                 | Thymoma Type (WHO) | Spinal Metastases Location | Intradural or Extradural | Incomplete Paralysis or Paralysis | Myasthenia Gravis | Years since Initial Diagnosis | Resection of Primary Lesion | Surgical Treatments                                                                 | Adjunct Treatment | Radiosurgery or chemotheraphy Postoperative Complications and Outcome |
|-------------------------|------|--------------|------------------------------------------------------------------------------------|--------------------|----------------------------|--------------------------|-----------------------------------|-------------------|-----------------------------|-----------------------------|--------------------------------------------------------------------------------|-----------------|---------------------------------------------------------------------------------|
| Marotta et al[2]        | 2014 | 46, M        | Left coincocordalgia and a reduction of strength of the left arm                    | C                  | C5-C7                      | Intradural               | No                                | No                | 24                          | Thymectomy                  | Intradural component radiation or chemotherapy                               |                  | Died 2 weeks after surgery                                                      |
| Sasaki et al[16]        | 2015 | 50, F        | Back pain; LLE weakness and numbness                                               | C, squamous cell    | T3                         | Intradural               | Yes                               | No                | 0                           | Thymectomy                  | T3 laminectomy with tumor resection Radiotherapy and chemotherapy             |                  | Improvement of symptoms                                                        |
| Zhao et al[5]           | 2016 | 47, F        | Back pain and lower extremity weakness, hypotension, urine incontinence;           | B2, type B1, locally B3 | T9-T11                     | Intradural               | Yes                               | Yes               | 8                           | Thymectomy                  | Posterior decompression, tumor resection with bone cement reconstruction and internal fixation |                  | Muscle strength and urinary function recovered gradually                      |
| Low et al[11]           | 2016 | 71, M        | Back pain; transient radiculopathy                                                | Low-grade mucoepidermoid tumor | T8-T9                      | Intradural               | No                                | No                | 12                          | None                        | None                                                                          |                  | Returned to normal 2 years postoperatively                                    |
| Shivapathasundaram et al | 2016 | 44, M        | Back pain, gait abnormality; bowel/bladder symptoms; neurologic claudication;     | B3                 | L4-L5                      | Intradural               | No                                | No                | 5                           | L3-L4 laminectomy with GTR of epidural disease | Radiation and chemotherapy |                  | Died 9 months after surgery                                                   |
| Jazi et al[7]           | 2017 | 44, M        | Double vision and cervical paraspinal weakness; unable to lift his head;           | B3                 | C7                         | Not reported             | No                                | Yes               | 10                          | Mediastinotomy               | Steroids, pyridostigmine, and five sessions of plasmapheresis; Radiation and chemotherapy |                  | Symptoms controlled at 3-year follow-up                                        |
| Kim et al[7]            | 2017 | 71, F        | Weakness of the left wrist and grasp                                              | C                  | C6-T1, intervertebral foramen | Intradural               | No                                | No                | 7                           | Surgical resection            | A nearly total resection preserving nerve roots was performed via a total C6-T1 laminectomy |                  | Disease free at 6 month follow-up                                              |
| Achey et al[5]          | 2018 | 63, M        | Acute urinary incontinence; low back pain; right lower extremity radiculopathy   | C                  | T8, L5-S1                  | Intradural               | No                                | No                | 8                           | L5-S1 decompression and GTR of the metastatic lesion at L5, along with stabilization | Radiation        | Remained neurologically stable, symptoms of lower back pain and radiculopathy improved |
|                        | 2018 | 38, F        | Left flank pain radiating to the lower abdomen and groin and rapidly progressive    | B3 with cytologic atypia, mitoses and infiltrative growth concerning for type C thymic carcinoma | T12-L1                   | Intradural               | No                                | No                | 2                           | Thymectomy                  | T12-L1 decompression with facetectomy, T11-L2 posterior instrumentation fusion; GTR of T12-L1 tumor | Chemotherapy     | Neurologically stable                                                         |
|                        | 2018 | 44, F        | Rapidly progressive myelopathy; neck pain                                         | C                  | C7                         | Intradural               | No                                | No                | 4                           | Mediastinotomy               | Anterior C7 vertebrectomy for decompression and tumor resection, with the placement of a cage graft and plating from C5 to C7 |                  | Remained neurologically stable, Died 4 months after surgery                   |

BUE= bilateral upper extremity; LLE= left lower extremity; BLE= bilateral lower extremities; RLE= right lower extremity; GTR= gross total resection; STR= subtotal resection.
of resection, Masaoka stage, and Mueller determinants of long-term survival in thymoma are completeness spine. To date, studies have revealed that the most important chemoradiotherapy, although such adjuvant therapy is not tumors are often treated by surgery combined with adjuvant classi –

Spinal cord compression.

The 5-year survival rate of patients with distant metastasis of thymoma varied widely between 13.3% and 81%

Surgical extent, cement volume, and complication was cement leakage into the canal and subsequent this approach still needs to be con

The 5-year survival rate of patients with distant metastasis of thymoma varied widely between 13.3% and 81%

Thymomas are generally radiosensitive and chemosensitive

4. Conclusion

Although uncommon, metastatic malignant thymoma of the spine should be part of the differential when the patient presents with neurological deficits and has a medical history of thymoma. We recommend the posterior approach for spinal decompression of the metastatic tumor when the tumor has caused neurological deficits. Vertebroplasty by cement augmentation may be a proper treatment option for patients with spinal metastatic thymoma who cannot undergo appropriate surgery or decline open surgery. Systemic chemotherapy and radiotherapy should be considered in a multimodality protocol if tumor recurrence is expected. Thymomas are generally radiosensitive and chemosensitive tumors, with high response rates. Thus, higher-staged tumors are often treated by surgery combined with adjuvant chemoradiotherapy, although such adjuvant therapy is not standardized due to its rarity. Moreover, little is known about the natural history and prognosis of metastatic thymoma to the spine. To date, studies have revealed that the most important determinants of long-term survival in thymoma are completeness of resection, Masaoka stage, and Mueller–Hermelin histologic classification. The 5-year survival rate of patients with distant metastasis of thymoma varied widely between 13.3% and 81% after multimodality treatment, including surgical resection of primary tumor, pleurectomy, chemotherapy, and radiotherapy. A multidisciplinary approach is required to improve patients’ long-term outcomes. In conclusion, we present an extremely unusual occurrence of spinal metastasis of malignant thymoma that successfully managed by surgical procedure, percutaneous vertebroplasty with cement, which has not been previously well reported. Our focus is to emphasize the importance of considering spinal metastasis of malignant thymomas as a diagnosis and guiding the proper management strategy upon treatment.

Author contributions

Conceptualization: Shuzhong Liu, An Song, Yong Liu.

Methods: Shuzhong Liu, Xi Zhou, An Song, Yong Liu.

Investigation: Shuzhong Liu, Xi Zhou, An Song, Yong Liu.

Resources: Shuzhong Liu, Xi Zhou, An Song, Zhen Huo, Yipeng Wang, Yong Liu.

Writing – original draft: Shuzhong Liu, Xi Zhou, An Song, William A. Li, Radhika Rastogi.

Writing – review and editing: Shuzhong Liu, Xi Zhou, William A. Li, Radhika Rastogi, Yipeng Wang, Yong Liu.

References

[1] Liu T, Qu G, Tian Y. Thymic carcinoma with primary spine metastasis. J Clin Neurosci 2011;18:840–2.
[2] Marotta N, Mancarella C, Colistra D, et al. First description of cervical intrathoracic thymoma metastasis. World J Clin Cases 2015;3:946–50.
[3] Kim JY, Lee YS, Kang DH, et al. Epidural metastasis in malignant thymoma mimicking epidural abscess: case report and literature review. Korean J Spine 2017;14:162–5.
[4] Hong B, Nakamura M, Hartmann C, et al. Delayed distant spinal metastasis in thymomas. Spine (Phila Pa 1976) 2013;38:E1709–13.
[5] Achey RL, Lee BS, Sundar S, et al. Rare thymoma metastases to the spine: case reports and review of the literature. World Neurosurg 2018; 110:423–31.
[6] Low HM, Wong CF, H’ng MW. Thymic carcinoma presenting with an unusual and delayed metastasis to the neural foramen, mimicking thoracic spinal radiculopathy. Med J Malaysia 2016;71:368–9.
[7] Jazi HH, Harmon DM, Tran T, et al. Malignant thymoma with metastasis associated with paraneoplastic myasthenia gravis. Proc (Bayl Univ Med Cent) 2017;30:330–2.
[8] Konovolov NA, Nazarenko AG, Ayuskin DS, et al. Comprehensive assessment of the outcomes of surgical treatment of patients with metastatic spinal cord injuries. Zh Vopr Neirokhir Im N N Burdenko 2015;79:34–44.
[9] Shuvapathasundram G, Sammons V, Bazina R, et al. Metastatic thymoma presenting as spontaneous epidural lumbar haematoma. Eur Spine J 2016;25(suppl 1):33–7.
[10] Jee TK, Lee SH, Kim HJ, et al. Spinal metastasis of thymic carcinoma as a rare manifestation: a summary of 7 consecutive cases. Korean J Spine 2014;11:157–61.
[11] Devic P, Choumert A, Vukusic S, et al. Myopathic camptocormia associated with myasthenia gravis. Clin Neurol Neurosurg 2013;115: 1488–9.
[12] Ahn S, Lee JH, Ha SY, et al. Clinicopathological analysis of 21 thymic neuroendocrine tumors. Korean J Pathol 2012;46:221–5.
[13] Posner JB, Howeson J, Cvitkovic E. “Disappearing” spinal cord compression: oncolytic effect of glucocorticoids (and other chemotherapeutic agents) on epidural metastases. Ann Neurol 1977;2:409–13.
[14] Žeppe P, Varone V, Cozzolino I, et al. Fine needle cytology and flow cytometry of ectopic cervical thymoma: a case report. Acta Cytol 2010;54(suppl 5):998–1002.
[15] Dutta R, Kumar A, Julka PK, et al. Thymic neuroendocrine tumour (carcinoid): clinicopathological features of four patients with different presentation. Interact Cardiovasc Thorac Surg 2010;11:732–6.
[16] Sasaki S, Fukushima T, Maruyama Y, et al. Two cases of thymic carcinoma initially presenting as bone metastasis: a clinical report and the usefulness of CDS immunohistochemistry for assessing bone lesions. Intern Med 2015;54:1781–5.
[17] Toba H, Kondo K, Takizawa H, et al. Recurrent thymoma with a pleural dissemination invading the intervertebral foramen. Eur J Cardiothorac Surg 2009;35:917–9.
[18] Nagel SJ, Hughes G, Ugowke KT, et al. Spinal carcinoid metastasis with dural invasion. World Neurosurg 2011;76:478.e7–11.
[19] Farin A, Aryan HE, Abshire BB, et al. Thymoma metastatic to the extradural spine. J Clin Neurosci 2009;15:2824–7.
[20] Ogurtz A, Achiume H, Kato D, et al. Efficacy of doxetaxel as a second-line chemotherapy for thymic carcinoma. Chemotherapy 2004;50:279–82.
[21] Hentschel SJ, Rhines LD, Shah HN, et al. Percutaneous vertebroplasty in vertebra plana secondary to metastasis. J Spinal Disord Tech 2004; 17:554–7.
[22] Selvaraj N, Chad DA, Smith TW, et al. Myasthenic crisis after resection of an isolated metastatic thymoma of the cervical spine. J Clin Neuromuscul Dis 1999;1:11–3.
[23] McLennan MK. Case report 637: Malignant epithelial thymoma with osteoplastic metastases. Skeletal Radiol 1991;20:141–4.
[24] Zhao L, Zhou X, Li Z, et al. Bone metastasis of malignant thymomas associated with peripheral T-cell lymphocytosis. BMC Surg 2016;16:58.
[25] Alafaci C, Salpietro FM, Grasso G, et al. Spinal cord compression by a metastasizing thymoma. Acta Neurochir (Wien) 1999;141:215–6.
[26] Akamatsu H, Tsukuura T, Matsunaga H, et al. Study of combination therapy for thymoma: a case of stage IV which presented as total spinal block caused by epidural metastasis and which preoperative combination therapy was effective for minimizing the tumor. Kyobu Geka 1993;46:1156–60.
[27] Marandino F, Zoccali C, Salducca N, et al. Ectopic primary type A thymoma located in two thoracic vertebrae: a case report. BMC Cancer 2010;10:322.
[28] Vladislav T, Jain RK, Alvarez R, et al. Extrathoracic metastases of thymic origin: a review of 35 cases. Mod Pathol 2012;25:370–7.
[29] Liu S, Song A, Zhou X, et al. Malignant pheochromocytoma with multiple vertebral metastases causing acute incomplete paralysis during pregnancy: Literature review with one case report. Medicine (Baltimore) 2017;96:e8535.
[30] Liu S, Zhou X, Song A, et al. Successful treatment of Gorham-Stout syndrome in the spine by vertebroplasty with cement augmentation: a case report and literature review. Medicine (Baltimore) 2018;97:e11555.
[31] Liu S, Zhou X, Song A, et al. Successful treatment of malignant pheochromocytoma with sacrum metastases: A case report. Medicine (Baltimore) 2018;97:e12184.