SPINAL DUMBBELL EPIDURAL HEMANGIOMA: TWO STAGE/SAME SITTING/SAME POSITION POSTERIOR MICROSURGICAL AND TRANSTHORACIC ENDOSCOPIC RESECTION – CASE REPORT AND REVIEW OF THE LITERATURE

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SUMMARY – Spinal dumbbell tumors are defined by a narrowing at the point where they penetrate the intervertebral foramina or dura mater, assuming an hourglass or dumbbell shape. Dumbbell-shaped spinal hemangiomas are extremely rare. We describe a dumbbell spinal tumor (epidural cavernous hemangioma) resected by a 2-stage single-sitting combined approach. We also conduct a substantial literature review of the subject. We present a case of a 78-year-old male who was found to have a homogeneously enhancing, dumbbell-shaped, intraspinal, extradural tumor mass extending into the left chest cavity. The tumor was resected with a single-sitting 2-stage posterior technique: a microsurgical approach, followed by endoscopic resection via a thoracoscopic approach. There are several reports in the literature on the combined approach for dumbbell tumors of the spinal cord. Our case is the first to describe 2-stage combined surgery in 1 sitting for dumbbell hemangioma with the patient in the lateral decubitus position for the thoracoscopic part of the surgery; and the use of a fat pad, which was applied in the neuroforamen via the posterior route, as a marker for resection during the transthoracic procedure.

Key words: Spine; Dumbbell tumor, epidural; Hemangioma; Microsurgery; Endoscopy; Thoracoscopic approach

Introduction

Spinal dumbbell tumors were first defined by Heuer in 1929 as a group of tumors arising along the spine that are constricted at the point where they penetrate the intervertebral foramina or dura mater, assuming an hourglass or dumbbell shape. Non-schwannoma/non-neurofibroma dumbbell tumors of the spinal cord include 28 different pathological entities, such as vascular lesions (hemangiomas and hemangioblastomas), meningiomas, and various rare lesions described in the literature as single case reports. Hemangiomas are congenital vascular malformations whose pathologies are considered to be hamartomatous malformations. Spinal epidural hemangiomas (SEHs) can be divided into 2 types: epidural hemangiomas of vertebral origin, and primary epidural space hemangiomas. SEHs account for 4% of all spinal epidural tumors, mostly occurring as primary lesions in the vertebral bone. According to a study by Esene et al., there are approximately 100 reported
Table 1. Literature review of dumbbell spinal epidural hemangiomas

| Author and year | Age (yrs) /Sex | Location level | Presentation | Surgical technique |
|-----------------|---------------|----------------|--------------|--------------------|
| Rovira et al. 1999 | 51/F | L3/4 | Sciatica, no deficits | Posterior approach (laminectomy) |
| Padovani et al. 1982 | 75/M | T3-6 | Gait disturbances | Posterior approach (laminectomy, partial removal) |
| Franz et al. 1987 | 23/M | T3/4 | Spastic paraplegia | Posterior approach (laminectomy, total removal) |
| Feider et al. 1991 | 50/M | L3/4 | Sciatica, no deficits | Posterior approach (laminectomy, total removal) |
| Morioka et al. 1986 | 50/M | T2/3 | Hypoesthesia below chest | Combined posterior approach (laminectomy) with thoracotomy |
| Fukushima et al. 1987 | 54/M | T7/8 | Spastic paraparesis | Posterior approach (laminectomy, total removal) |
| Haimes et al. 1991 | 46/M | T3/4 | Hypoesthesia of bilateral feet | Combined posterior approach (laminectomy) with thoracotomy |
| Lanotte et al. 1994 | 27/F | T1/2 | Paresthesia of bilateral feet | Posterior approach (arthrolaminectomy) |
| Harrington et al. 1995 | 37/F | L3/4 | Leg numbness and pain | Posterior approach (hemilaminectomy, total removal) |
| Bavbek et al. 1997 | Not specified | C8 | Not specified | Not specified |
| Carlier et al. 2000 | 36/M | Cervical spine | Acute weakness in right upper limb | Posterior approach |
| Saringer et al. 2001 | 56/M | T3/4 | Chronic neuralgia | Posterior approach |
| Badinand et al. 2003 | 40/F | T3/4 | Gait disturbances, sphincter problems | Posterior approach (laminectomy), subtotal removal |
| Hara et al. 2006 | 19/M | T12/L1 | Low back pain | Posterior approach |
| Kang et al. 2006 | 56/M | T3/4 | Chest wall pain | Posterior approach (laminectomy), partial resection |
| Iglesias et al. 2008 | 57/F | C7/T1 | Pain | Posterior approach |
| Doyle et al. 2008 | 57/F | T4/5 | Progressive weakness and altered sensation of the lower limbs | Posterior approach (hemilaminectomy, costotransversectomy), total resection |
| Maeda et al. 2011 | 67/M | T2/3 | Incidental finding | Combined single-stage surgery with thoracotomy and laminectomy with facetectomy |
| Suzuki et al. 2011 | 73/M | T1/2 | Incidental finding | Posterior approach (hemilaminectomy with costotransversectomy) |
| Vassal et al. 2011 | 59/F | T5-7 | Back pain, ataxia, paraparesis | Posterior approach (hemilaminectomy with costotransversectomy) |
| Uchida et al. 2010 | 75/M | T11/12 | Progressive paraparesis | Posterior approach (laminectomy, total removal) |
| Yunoki et al. 2015 | 77/F | L2/3 | Lumbago, L3 Hypoesthesia | Posterior approach (laminectomy, total removal) |
| Garcia-Pallero et al. 2015 | 67/F | T4/5 | Pleural effusion, no deficits | Posterior approach (hemilaminectomy, facetectomy and costotransversectomy), total resection |
cases of pure SEHs. Because of its embryological origin, the most common tumor site is the dorsal or dorsolateral spinal canal where the venous plexus is abundant\(^1\). Most of epidural hemangiomas are the cavernous type; there are fewer than 10 cases of epidural capillary hemangiomas in the literature\(^1,5\). Dumbbell-shaped spinal hemangiomas are extremely rare with only 29 cases in the literature described so far (Table 1)\(^1\).

There are several surgical techniques for the resection of thoracic spine dumbbell spinal tumors. Lately, the combined posterior microsurgical approach has been used to resect the intraspinal part of the tumor, followed by subsequent video-assisted thoracoscopic surgery for the intrathoracic part of the tumor, and has gained popularity\(^2,11,25,26,29\). Other approaches include a posterior-only approach\(^10\), a posterior approach combined with thoracotomy\(^8,21\), an extended lateral cavity approach\(^23\), and a thoracoscopic-only approach\(^19\). Most cases of spinal dumbbell hemangiomas described in the literature so far were resected using a posterior-only approach or a combined approach with thoracotomy.

Herein we describe a case of a rare dumbbell spinal tumor, an epidural cavernous hemangioma, resected by a 2-stage single-sitting combined approach (i.e. a posterior microsurgical approach with resection, followed by thoracoscopic approach and endoscopic resection). In addition, we provide a literature review of this rare lesion, including differential diagnosis of dumbbell spinal tumors and surgical techniques performed for their management, as well as a review of all spinal dumbbell hemangiomas published so far.

### Case Report

A 78-year-old male with no significant medical history presented with progressive bilateral leg weakness and numbness during the previous week. Physical examination revealed deep paraparesis (motor strength 3/5 bilaterally) and inability to walk. Magnetic resonance imaging (MRI) with and without contrast revealed a homogeneously enhancing dumbbell-shaped intraspinal extradural tumor mass extending into the left chest cavity. It had displaced the thoracic spinal cord to the right. The tumor extended from the 3rd thoracic vertebral body to the upper margin of the 5th vertebral body, continuing dumbbell-like through the intervertebral foramen of T4 into the left middle thorax (Fig. 1). The differential diagnosis included neurofi-
broma or schwannoma, as well as other rare dumbbell-shaped tumors of the spine (such as meningiomas and hemangiomas), malignant peripheral nerve sheath tumors, hemangioblastomas, or metastases. Due to paraparesis and compression of the spinal cord, indication for surgery was made.

The patient underwent a single sitting 2-stage surgery, consisting of both posterior microsurgical and transthoracic endoscopic resection of the tumor. For the posterior approach, partial hemilaminectomy of T3-T5 was performed, which revealed a highly vascularized epidural tumor with spinal cord compression extending into the neuroforamen. Gelfoam powder (Pfizer, New York, NY) and bipolar coagulation were used for hemostasis of the tumor. The intraspinal epidural portion of the tumor was completely resected. A fat pad, harvested at the beginning of surgery via a paraumbilical incision, was placed into the foramen as a marker for the extent of resection during the later thoracoscopic procedure. The second stage surgery included video-assisted left thoracoscopic resection of the extraforaminal intrathoracic part of the tumor. For this procedure, the patient was repositioned in the right lateral decubitus position. The fat pad was visualized endoscopically in the neuroforamen at the end of the resection, providing important orientation. Chest tubes were placed at the end of the procedure. The duration of the entire procedure was 4:52 hours. Blood loss was 400 cc. Intraoperative neuromonitoring with somatosensory evoked potentials (SSEPs) and motor evoked potentials (MEPs) was performed and showed no signs of pathological electrophysiological activity.

The patient recovered from surgery and his paraparesis resolved completely. Chest tubes were removed on the sixth day following surgery. Complete resolution of paraparesis to normal lower extremity strength was noted along with the absence of bowel or bladder disturbances. Histopathologic evaluation revealed cavernous hemangioma (Fig. 2). Follow-up MRI with and without contrast showed no residual tumor (Fig. 3).

Discussion

The Eden Classification was the first classification system used for dumbbell tumors, which served as a gold standard for decades. Ninety percent of spinal dumbbell tumors are schwannomas, and up to 33% of schwannomas have a dumbbell form. Other dumbbell-shaped tumors include hemangiomas, meningiomas, malignant peripheral nerve sheath tumors, neurogenic paravertebral tumors with origin from neurogenic elements within the thorax (including neuroblastoma, ganglioneuroblastoma, ganglioneuroma, hemangioblastomas, liposarcomas, lipoblastoma, angiomatosis, angiolipoma, rhabdomyosarcoma, spine extrasosseous chordoma (SEC), mesenchymal chondrosarcoma, soft tissue chondroma, osteochondroma, malignant glomus tumor, malignant solitary fibrous tumor, plasmacytoma, metastasis, Ewing sarcoma, atypical teratoid rhabdoid tumor, lymphoma, lymphangioma, meningeal melanocytoma, small cell malignant tumor, and peripheral primitive neuroectodermal tumor (PNET).

Hemangioma histology

Hemangiomas are benign vascular malformations and are most often found in the skin or soft tissues of
younger patients. They are characterized histologically by nodules of capillary-sized vessels lined by flattened endothelium and often regress spontaneously. Unlike their cutaneous capillary hemangioma counterparts, spinal hemangiomas are solitary lesions and unassociated with systemic or cutaneous hemangiomas or congenital hemangiomatosis. These tumors occur in adults, and require surgery, as they do not regress. They are found most commonly in intradural extramedullary and intramedullary locations of the thoracic spine and cauda region.

Spinal epidural hemangiomas account for 4% of all spinal epidural tumors, mostly occurring as a primary lesion of the vertebral bone, and can be divided into 2 types: epidural hemangiomas of vertebral origin and primary epidural-space hemangiomas. The majority of extradural cavernous hemangiomas extend from a vertebral hemangioma into the spinal canal with purely extradural locations. They represent only 1%-2% of all reported spinal hemangiomas. According to a study by Esene et al., there are approximately 100 reported cases of pure SEHs. Most epidural hemangiomas are of the cavernous type. There are fewer than 10 cases of epidural capillary hemangiomas in the literature. Only extradural cases with spinal cord compression have been described, as well as cavernous hemangiomas with intradural and extradural growth. There is one case report of juxtaposition of intradural schwannoma and extradural cavernous hemangioma of the thoracic spine.

Histologically, hemangiomas can be divided into capillary and cavernous types, depending on the dominant vessel size under microscopy. Generally, the cavernous type presents with a large number of sinusoidal channels in collagenous tissue, whereas capillary hemangiomas present capsulated lesions characterized by lobules of thin irregular capillary-sized vessels lined by endothelial cells, which are separated by septa of fibrous connective tissue. In addition, the cavernous type of hemangioma often presents with acute symptomatology, mainly related to intratumoral bleeding. On the contrary, capillary hemangiomas usually present in a chronic and progressive manner because of mass effect and nerve root irritation. Table 1 provides an overview of the cases of dumbbell SEHs described in the literature thus far. Most of the described cases were cavernous hemangiomas; there are only 5 capillary hemangiomas reported.
More than half of all reported dumbbell tumors were completely restricted to the extradural space, although preoperative MRI suggested the presence of intradural/extradural tumors in some cases. A wide variety of unusual lesions that can cause neural foraminal widening need to be taken into consideration, including the following: (1) neoplastic lesions, such as benign/malignant peripheral nerve sheath tumors (PNSTs), solitary bone plasmacytoma (SBP), chondroid chordoma, superior sulcus tumor; and (2) metastatic and non-neoplastic lesions, such as infectious process (tuberculosis, hydatid cyst), aneurysmal bone cyst (ABC), synovial cyst, traumatic pseudomeningocele, arachnoid cyst or vertebral artery tortuosity.

Vascular lesions are particularly important in differential diagnosis as they can present with bleeding. It is also important to consider vascular tumors during surgical planning. It is often impossible to differentiate between dumbbell-shaped schwannomas, meningiomas, or vascular lesions. Cavernous hemangiomas should be included in the differential diagnosis of dumbbell-shaped spinal tumors when the intervertebral foramina is not highly dilated, and a non-enhanced nerve root is identified in the tumor.

Magnetic resonance imaging usually yields no definitive findings; both neurogenic tumors and hemangiomas are isointense-to-hypointense on T1 images, and homogeneously hyperintense on T2 images. As a general rule, a diagnosis of schwannoma should be made when a spinal intradural extramedullary tumor shows hyperintensity on T2-weighted images or intense enhancement without dural tail sign; otherwise, a meningioma diagnosis is more probable. One characteristic of spinal meningiomas is the so-called ‘ginkgo leaf sign’ seen on axial post contrast T1 imaging (the leaf represents a distorted spinal cord pushed to one side of the theca by the meningioma, and the stem is seen as a non-enhancing ‘streak’ representing the stretched dentate ligament). One of the signs most indicative of hemangioblastoma is the presence of blood-flow voids on T2-weighted MRI.

Cavernous SEH has a different MRI appearance due to the presence or absence of degenerative phenomena, and the presence of hemosiderin pigments. As in cerebral locations, mixed signal intensity in all MRI sequences might be indicative of cavernous hemangioma. The most common MRI features of cavernous SEHs are solid hypervascular masses with lobular contour. A rim of low T2 signal intensity could be present due to deposition of hemosiderin; fibrotic capsule and epidural fat are relatively common findings, too. Additional MRI characteristics for making a differential diagnosis are solid hypervascularity, T1 hyperintensity, or multisegmental involvement. The MRI presentation of SEHs with isointense signal on T1-weighted imaging and hyperintense signal on T2-weighted imaging with homogeneously strong enhancement has been described as a ‘sign of wafting silk’. The widening of the intervertebral neural foramen and erosion of the adjacent bones are not uncommon. Typical computed tomography (CT) findings for hemangioma are reported to be lobulation, heterogeneous enhancement with contrast media, multiple ring-like calcifications, and the presence of an intact intervertebral foramen when the tumor extends to the spinal canal; however, these findings are not always observed.

Surgical technique

Most of the dumbbell spinal hemangiomas described in the literature were resected using a posterior approach.
microsurgical approach with laminectomy or hemilaminectomy, and with or without facetectomy and costotransversectomy (Table 1). A single-stage removal of thoracic dumbbell tumors from a posterior approach with costotransversectomy only, and with subsequent instrumentation has been described for schwannomas, neurofibromas, and ganglioneuromas. For resection of the extraspinal component, simple enucleation without thoracotomy can be performed. Although the extraspinal component, including the aorta, cannot be fully seen, it has been argued that enucleation is a safer method since ablation with aorta is unnecessary. A posterior approach may necessitate additional stabilization – its main disadvantage – and reduced visibility of the extraforaminal part of the tumor and adjacent vessels, including the aorta.

Our report describes dumbbell hemangioma resection using a combined approach: unilateral hemilaminectomy with microsurgical resection of the intraspinal part of the tumor, and thoracoscopic resection of the extraforaminal left thoracic tumor extension. Herein, we describe for the first time the following: (1) a 2-stage combined surgery in a single sitting for dumbbell hemangioma with the patient in the lateral decubitus position for the first thoracoscopic part of the surgery; and (2) the use of a fat pad, which was applied in the neuroforamen via the posterior route, which served as a marker for resection during the second transthoracic procedure.

Combined 2-stage single session surgery with a posterior microsurgical and thoracoscopic approach

The combined posterior microsurgical and endoscopic approach for benign mediastinal dumbbell tumors was first described in a case series by Vallieres et al. in 1995. So far, there are several reports in the literature on the combined approach for dumbbell tumors of the spinal cord, but only 1 case report using the combined approach for spinal epidural dumbbell hemangioma. In most reports, the pathology included schwannomas and neurofibromas, and there were rare cases of meningioma, granular cell tumor, and ganglioneuroma. In one patient with neurofibromatosis, Chen et al. describe a combined resection of 2 dumbbell tumors – a meningioma and schwannoma at C1/2 and T5/6, respectively.

The posterior approach is restricted to a vertical midline incision centered over the tumor and a transthoracic transpleural approach, which requires removal of fewer facet joints, transverse processes, and ribs. For these reasons, several authors have hypothesized that segmental stability may be less compromised with a combined approach than with a purely posterior approach coupled with costotransversectomy.

The combined approach has several disadvantages, though. It is difficult to precisely use thoracoscopic instruments with unexpected bleeding or other emergencies. Postoperative chest tubes may have potential complications, such as postoperative pain, pulmonary dysfunction, and infection. For most video-assisted thoracic surgery, patients are usually placed in the lateral decubitus position with safe resection of posterior mediastinal dumbbell tumors using video-assisted thoracoscopy. However, the only case report of dumbbell spinal hemangioma resected via this combined approach included turning the patient in lateral decubitus position.

Other combined approaches for resection of spinal dumbbell tumors in the thoracic spine

The combined approach with posterior microsurgical resection via laminectomy followed by thoracotomy has been performed for spinal dumbbell meningiomas. Other authors have favored thoracotomy prior to laminectomy for the resection of dumbbell-shaped tumors of the mediastinum, especially with marked vascularity, such as that seen with hemangiomas because the initial thoracotomy facilitates subsequent laminectomy procedures, enables ligation of the tumor arteries, and precludes extensive bleeding in spinal canal. Image-guided thoracoscopic-only resection of dumbbell mediastinal tumors with a small intraspinal portion have been previously described. Uehara et al. describe a case of dumbbell Ewing sarcoma where transthoracic biopsy was performed and the tumor was consequently treated with chemotherapy. Another report by Yamaguchi et al. describes combined thoracoscopic and supraclavicular approach for a cervico-mediastinal neurogenic tumor with an intraforaminal portion at C7/T1. Hakuba et al. performed a transspondiscal approach to dumbbell tumors of the cervical spinal canal, which included anterior discectomy and ipsilateral uncectomy, and removal of the posterolateral corners and posterior transverse ridges of the
upper and lower vertebral bodies at the level of the tumor.

Resection of thoracic and lumbar spinal schwannomas can be performed using the unilateral posterior approach only. The lateral extracavitary approach (LECA) can be used for resection of dumbbell tumors of the thoracolumbar spine, especially when extensive or difficult and paraspinal exposure is required. Vecil et al. report multi-level rib resections with extended lateral parascapular approach for giant thoracic schwannoma and laminectomy necessitating posterior spinal stabilization.

Alternative treatments

When totally removed, these tumors do not recur during follow-up periods of at least 5 years. Even when complete removal is accomplished, some symptoms may persist, possibly due to scarring around the dural sac or the involved nerve root but this does not necessarily indicate lesion recurrence. However, in cases of subtotal resection, radiation therapy has been advised. Endovascular embolization has been recently used to remove a hemangioma, successfully minimizing blood loss during the operation. However, this management approach carries the risk of spinal infarction. There is also a report on image-guided stereotactic radiosurgery of epidural hemangiomas with hypofractionated dose of 32 Gy in 4 fractions in a patient having undergone open decompression and biopsy; follow-up showed gradual reduction of the tumor mass.

Conclusion

Vascular dumbbell lesions of the spinal cord, such as hemangiomas, are rare and may be clinically or radiologically indistinguishable from other lesions, but they should be considered when making a differential diagnosis. The combined single-sitting, 2-stage posterior microsurgical approach via hemilaminectomy followed by thoracoscopic resection is a safe, time-saving, stable, and conformable surgical treatment option for these lesions, resulting in excellent clinical outcome with complete resolution of deep paraparesis.

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Spinalni tumori pješčanog sata definirani su suženjem na mjestu gdje prodiru u intervertebralni foramen ili dura mater, uz pretpostavku da imaju oblik pješčanog sata ili bućice. Spinalni hemangiomi u obliku bućice su iznimno rijetki. Opisuje se spinalni tumor pješčanog sata (epiduralni kavernozni hemangiom) reseciran dvostupanjskim kombiniranim pristupom, uz opsežan pregled literature. Prikazuje se slučaj 78-godišnjeg muškarca za kojeg je utvrđeno da ima homogeno pojačanu intraspinalnu, ekstraduralnu masu u obliku bućice koja se širi u lijevu prsnu šupljinu. Tumor je reseciran jednokratnom 2-stupanjskom posteriornom tehnikom: mikrokirurški pristup, nakon čega slijedi endoskopska resekcija putem torakoskopskog pristupa. U literaturi postoji nekoliko izvješća o kombiniranom pristupu za tumore bućice kralježnične moždine. Naš je slučaj prvi koji opisuje 2-stupanjsku kombiniranu kirurgiju u 1 sjedinju za hemangiom pješčanog sata u bolesnika u lateralnom dekubitusnom položaju za torakoskopsku dio operacije; i uporabu masnog jastučića koji je primijenjen u neuroforamenu preko stražnjeg puta, kao marker za resekciju tijekom transtorakalnog postupka.

Ključne riječi: Kralježnica; Tumor pješčanog sata, epiduralni; Hemangiom; Mikrokirurška; Endoskopska; Torakoskopski pristup