Surgical treatment of thoracic dumbbell tumors

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ABSTRACT

Background: Thoracic dumbbell tumors are uncommon neoplasms arising from neurogenic elements of the posterior mediastinum. Surgical removal of these tumors with mediastinal, neuroforaminal and intraspinal components can often be challenging. The purpose of this study is to present our experience of single-stage removal of dumbbell tumors of the posterior mediastinum and to discuss the surgical strategies for such tumors.
Method: A retrospective analysis was performed on 20 patients who underwent surgery for thoracic dumbbell tumors at our department during the period 2008 to 2016. Patient demographics, clinical features, operative reports, and pre- and postoperative images were reviewed.
Result: Complete resection was achieved in all patients, with no postoperative mortality. Surgical excision was performed by laminectomy plus Video-assisted thoracoscopic surgery (VATS) in 10 patients and laminectomy plus thoracotomy in 4 patients. Two patients underwent VATS alone. Supraclavicular and transthoracic approach was performed in 2 patients. Another 2 patients were treated with supra-clavicular approach alone. The mean operative time was 244 min (range 55–370 min), with mean esti-mated blood loss (EBL) of 360 ml (range 50–790 ml). Postoperative complications included one case of Horner's syndrome and one case of cerebrospinal fluid (CSF) leakage. At a mean follow-up of 29 months no patients showed recurrence of the tumor.
Conclusion: Thoracic dumbbell tumors should be evaluated for intraspinal and neuroforaminal involvement. Single-stage posterior laminectomy plus VATS/thoracotomy, VATS/thoracotomy, and supraclavicular alone or combined with transthoracic approach all could be the preferred method for removing these dumbbell tumors with satisfactory outcomes.

Introduction

Thoracic dumbbell tumors are uncommon neoplasms that can arise from neurogenic elements within the posterior mediastinum [1]. Approximately 10–20% of posterior mediastinal neurogenic tumors may extend into the spinal canal through an intervertebral foramen [2,3]. Most of the dumbbell tumors are of benign nerve sheath origin [2]. They often become very large and involve sur-rounding structures causing no clinical symptoms [4].

Complete surgical resection is considered the treatment of choice. Safe removal of these tumors often requires a combined thoracic-neurosurgical approach. There are several approaches for removing the thoracic portion of the tumor, which may be through a thoracotomy [5], thoracoscopy [6], or a posterior extrapleural approach [7]. Sometimes, the supraclavicular approach is needed in cases of tumors at the cervicothoracic junction [8]. For the intra-spinal and foraminal portion of the tumor, the posterior approaches included laminectomy [9], hemilaminectomy [10], costo-transversectomy [11] and ipsilateral facetectomy [3].

In this report, we summarize the clinical course in 20 cases of thoracic dumbbell tumors that were treated with procedures combining thoracotomy/thorascopy, posterior open laminectomy, and supraclavicular approach. The purpose of this study is to pre-sent our experience of single-stage removal of thoracic dumbbell tumors and to discuss the surgical strategies for such tumors.

Patients and methods

Medical records of 20 patients operated in our institution for thoracic dumbbell tumors between 2008 and 2016 were

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retrospectively reviewed. Patient information was collected through thoracic surgery charts, operative reports, pathology reports as well as medical image database. Approval to perform the study was obtained from the medical ethics committee in our hospital. Our indications for surgical resection of thoracic dumbbell tumors have been the suspicion of a posterior mediastinal tumor because of doubt about the diagnosis and the possibility of malignancy.

Surgical procedures

Laminectomy plus VATS

In cases when the tumor extended into intraspinal spaces and was classified as typeIIor III according to the Eden taxonomy [12], a laminectomy was performed before the thoracic procedure. In our institute, the neurosurgeons would be assisting with this part of the resection.

The involved vertebral level of the tumor was marked preoperatively on the skin of the patient's back with radiologic guidance. Under general anesthesia, the patient was intubated with a double-lumen endotracheal tube and was placed in the prone position. A vertical midline incision was made to expose the laminae bilaterally at the selected levels, followed by laminectomy of two adjacent laminae. Enough bone should be removed to expose the tumor and its margins, particularly the spinal cord tumor interface and the affected intervertebral foramen where the tumor narrows to pass through. If the tumor was entirely extradural, the nerve root from which the tumor originates was divided to allow mobilization of the tumor. In cases where the tumor had an intradural extension, the dura was opened and its intraspinal component was separated from the spinal cord under the operating microscope. After removing this component of the tumor and sacrificing the entire affected nerve root, we meticulously closed the dura and covered it with fibrin glue to avoid cerebrospinal fluid leak. The remaining tumor with the distal nerve stump was pushed into the chest cavity via the enlarged foramen. The tumor bed and the neural foramen was inspected for hemostasis and completeness of resection. After the posterior incision was closed, the patient was returned to the lateral decubitus position for VATS.

The thoracoscope was introduced through the 7th intercostal space in the midaxillary line and two operative ports were placed under thoracoscopic guidance, one at the 4th intercostal space in the anterioraxillary line and the other at the triangle of auscultation. The pleura covering the junction of the tumor with the chest wall and spine is sharply incised, circumferentially around the tumor, allowing a reasonable edge of pleura to ensure an adequate margin. Appropriate traction of the tumor with sponge-holding forceps allows a combination of sharp and blunt dissection to gradually mobilize and lift the tumor up from adjacent structures. Branches from the intercostal vessels and paravertebral feeding vessels to the tumor were either cauterized or clipped. Once sufficiently mobilized and detached from normal tissues, the paravertebral component of the tumor was freed. The remaining foraminal component of the tumor with the nerve stump, placed during the previous laminectomy, was identified and retracted into the chest, assuring completeness of resection. Monopolar coagulation should be avoided around the neural foramen. Absorbable gelatin sponges for hemostasis should not be packed and left within the foramen. The tumor was extracted using a surgical glove to avoid direct wound contamination. A single chest tube was left in place and the incisions were closed.

VATS

VATS was performed in cases when the tumor had small intraforaminal extension but without intraspinal involvement. The intraforaminal extensions of the tumor can be resected completely through an already enlarged foramen. The paravertebral and the intraforaminal component of the tumor can be excised and extracted out together, and laminectomy was not required.

Thoractomy

For bulky tumors, particularly those tumors that occupied the thoracic inlet and obscured the surrounding thoracic wall, posterolateral thoracotomy was performed through an intercostal space adjacent to the tumor. The remaining surgical procedure was identical with those in the VATS resections.

Supraclavicular and transthoracic approach

Dumbbell tumors of the cervicothoracic junction were treated with the combined supraclavicular and transthoracic approach.

The patient was placed in the supine position with a pillow under the shoulder. A supraclavicular transverse incision was made over the most prominent part of the tumor. When dissecting the cervical component of the tumor, the anterior scalene and sternocleidomastoid muscles were preserved, while the omohyoid muscle was cut. The brachial plexus could be recognised under direct visualization and the tumor could be separated from the nerve fibres. To avoid serious complications, intraoperative stimulation and neuromonitoring was performed throughout the procedure to ensure that no vital nerves were injured. When the deep side of tumor was exposed by blunt dissection, the supraclavicular wound and thoracic cavity were communicated, and sometimes the tumor could be pulled out from the supraclavicular wound. If the tumor was too large to be excised completely through the supraclavicular incision, the cervical component of the tumor was resected firstly and the remaining thoracic component was left for VATS or thoracotomy. After the tumor resection, branches of the brachial plexus were identified and stimulated, confirming its integrity.

After the supraclavicular incision was closed, the patient was placed in the lateral decubitus position. Thoracotomy was performed through the third or the fourth intercostal space, allowing better visualization of the intrathoracic component of the tumor. We performed intracapsular enucleation of the tumor in order to avoid vascular and nerve injury. The capsule of the tumor was opened with an electrical hook or harmonic scalpel, and the mass was enucleated. The remaining tumor capsule was then carefully dissected and removed, and the involved nerve root was ligated and resected.

Postoperative care

The chest tube was usually removed on postoperative day 1–3. If the dura was opened intraoperatively, the patient was immobilised for 3 days and suction on the chest drain should be avoided to reduce the risk of CSF leakage. The patient may be discharged the following day after chest drain removal.

Results

Patient characteristics

The patient characteristics are shown in Table 1. There were 8 men and 12 women ranging in age from 18 to 67 years (mean, 43 years). Six of 20 patients (30%) had clinical symptoms.

Table 1
Patient characteristics.

Case no.	Age/ Sex	Symptom	Tumor location	Tumor Size (cm)	Eden Taxonomy	Operation Approach	Pathology	Follow-Up(mon)
1	49/F	Back pain	L, T3-T4	7	III	VATS plus laminectomy	Schwannoma	15
2	46/M	None	L, T5-T6	5	III	VATS plus laminectomy	Schwannoma	35
3	18/F	Upper extremity numbness and discomfort	R, C7-T1	7.3	IV	Supraclavicular approach plus thoracotomy	Schwannoma	28
4	42/M	None	L, T2-T3	3.7	IV	VATS	Schwannoma	16
5	47/F	None	R, T3-T4	4.8	IV	VATS	Schwannoma	6
6	58/F	None	L, T3-T4	6.5	II	VATS plus laminectomy	Schwannoma	41
7	54/F	None	R, T10-T11	6	II	VATS plus laminectomy	Schwannoma	27
8	31/F	None	R, T1-T2	4.7	II	VATS plus laminectomy	Schwannoma	53
9	39/F	None	L, T2-T3	3.5	II	VATS plus laminectomy	Schwannoma	28
10	30/F	None	R, T2-T3	4.2	III	VATS plus laminectomy	Schwannoma	12
11	42/M	None	R, T3-T4	5	III	VATS plus laminectomy	Schwannoma	12
12	37/M	Horner's Syndrome	R, T2-T3	11	IV	Supraclavicular approach plus thoracotomy	Ganglioneuroma	24
13	65/M	None	R, T5-T6	4.8	III	VATS plus laminectomy	Schwannoma	30
14	43/M	Upper extremity numbness and discomfort	R, C7-T1	9.5	IV	Supraclavicular approach	Schwannoma	27
15	58/F	None	R, T1-T2	8	III	Thoracotomy plus laminectomy	Schwannoma	31
16	67/F	None	R, C7-T1	4	IV	Supraclavicular approach	Neurofibroma	36
17	31/F	Back pain	L, T2-T3	8.5	III	Thoracotomy plus laminectomy	Schwannoma	60
18	29/F	None	R, T2-T3	8	III	Thoracotomy plus laminectomy	Schwannoma	24
19	20/M	chest tightness	R, T3-T4	12	III	Thoracotomy plus laminectomy	Schwannoma	42
20	47/M	None	L, T5-T6	3.5	III	VATS plus laminectomy	Schwannoma	25

Radiographic findings

The lesion was right-sided in 13 patients and left-sided in 7 patients (Table 1). Sixteen patients had tumors at the thoracic tract, and 4 had tumors at the cervicothoracic junction. All 20 patients had intraforaminal extension of the tumor and 14 of them had intraspinal involvement. According to Eden classification, 2 patients were classified as type II, 12 patients as type III and 6 patients as type IV.

Operative management

All patients underwent single-stage tumor removal (Table 1). Surgical excision was performed by laminectomy plus VATS in 10 patients and laminectomy plus thoracotomy in 4 patients (70%). Two of them with intradural involvement required intradural dissection during laminectomy and the other two have extradural portions. Supraclavicular approach plus thoracotomy was performed in 2 patients with tumors at the cervicothoracic junction. In another 4 patients, the dumbbell tumor had a small intrathoracic component, and VATS or supraclavicular approach was sufficient. The mean operative time was 244 min (range 55–370min), with mean EBL of 360 ml (range 50–790 ml) (Table 2). Mean chest drain duration was 3 days (range 1–4 days) and the mean hospital stay was 4 days (range 2–10 days) (Table 2).

Outcome

Complete resection was achieved in all patients, with no postoperative mortality (Table 2). None of the patients undergoing VATS required conversion to open thoracotomy. Two patients showed minor postoperative complications. One patient had the tumor excised from cervicalthoracic junction producing aggravation of preexisting Horner's syndrome, which resolved in one year. In one patient with dura opening, CSF leakage persisted for 10 days postoperatively.

Pathology

Pathological examination confirmed the masses to be benign neurogenic tumors in all cases, with mean size of 6.4 cm (range 3.5–12 cm) (Table 1). Eighteen cases were schwannomas, one case was neurofibroma, and one case was ganglioneuroma.

Follow-up

The mean follow-up period for all cases was 29 months (range 6–60 months) and there were no cases of recurrence or spinal instability (Table 2). Preoperative back pain resolved postoperatively for 2 affected patients. Horner's syndrome and upper extremity discomfort in patient 3, 12 and 14 improved postoperatively and completely resolved within 1 years.

Illustrative case 1: patient no. 1

A 49-year-old woman presented to our department with a history of backache for more than 3 months. A MRI and CT scan showed a dumbbell-shaped lesion with the intraspinal and extraspinal parts connected through a widened left intervertebral foramen at T3-T4 level (Fig. 1a and b).

The patient underwent a combined neurosurgical and thoracoscopic approach. Laminectomy of T3 permitted access for removal of the extradural tumor. The extraspinal tumor was resected completely with thoracoscopic procedures (Fig. 1c). The posterior mediastinal tumor bed is inspected after resection (Fig. 1d). Pathologic examination confirmed a schwannoma. Postoperative MRI confirmed complete tumor removal (Fig. 1e). At follow-up back pain was completely resolved.
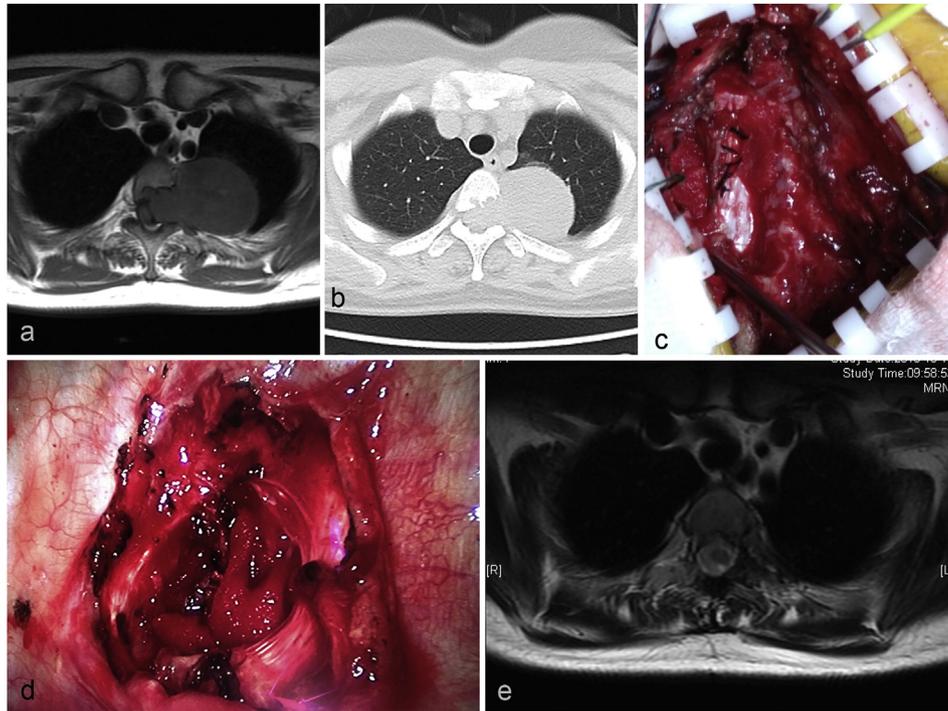
Table 2
Surgical outcomes of the patients of the dumbbell tumor (n = 20).

Variable	Measurement (mean ± SD)
Operation time	244.3 ± 92.0 min
Estimated blood loss	359.5 ± 211.9 min
Postoperative drain duration (n = 18)	2.8 ± 1.1 d
Postoperative hospital stay	4.1 ± 1.9 d
Follow-up	28.6 ± 13.6 month

Fig. 1. **a** Preoperative MRI showed the intraspinal and extraspinal soft tissue mass at T3-T4. Spinal cord was compressed and displaced by the tumor. **b** CT scan revealed enlargement of left intervertebral foramen. **c** Intraoperative photograph of the intraspinal operative field after the resection. **d** Intraoperative photograph of the tumor bed following resection. **e** MRI of the thoracic spine of the same patient at 15-monthes follow-up after surgery showed no remnant tumor and laminectomy state.

Case 2: patient no. 16

A 67-year-old woman presented with a swelling on the right side of the neck. Physical examination showed a palpable mass approximately 5 cm in diameter in his right supraclavicular region. A MRI and CT scan showed a well encapsulated tumor localized at the right cervicothoracic junction extending to the intervertebral foramen at C7-T1 level (Fig. 2a and b).

The patient underwent a supraclavicular approach to remove the tumor. The supraclavicular incision allowed an exposure of the tumor and surrounding nerves and vessels (Fig. 2c). The tumor had small intraforaminal extension that was resected completely
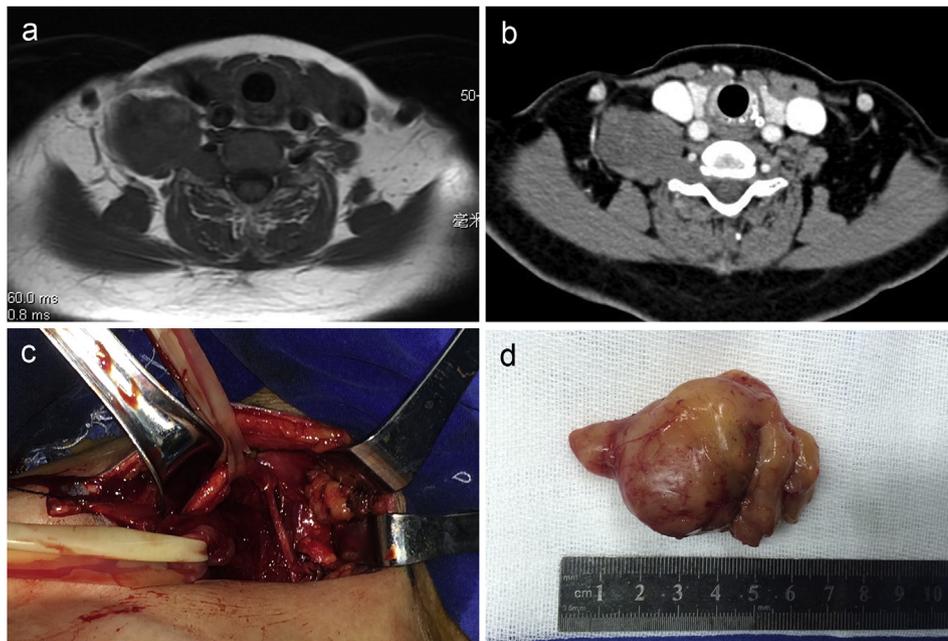

Fig. 2. **a** MRI showed a soft tissue mass at the thoracic apex extended into the vertebral foramina at C7-T1. **b** CT scan revealed a soft tissue mass in the right paravertebral region with small extension into the vertebral foramen. **c** Intraoperative photograph of the supraclavicular operative field after the resection. The brachial plexus and the subclavian artery were freed out with rubber bands pulling them open. **d** Extracted specimen.

through an already widened foramen (Fig. 2d). The patient had an uneventful recovery. Pathologic examination revealed a benign schwannoma.

Discussion

The term "dumbbell tumor" refers to posterior mediastinal tumors that connect two or more separate regions, such as intradural, epidural, intraforaminal and paravertebral spaces [12]. These tumors typically originate from spinal nerve roots or the sympathetic chain and are located in the paravertebral sulcus. Malignancy is rare in adults and 90% are benign nerve sheath tumors such as schwannomas and neurofibromas [13]. Between 30% and 40% of thoracic dumbell tumors are asymptomatic at the time of diagnosis [2,5,14]. With growth, they can produce symptoms by local compression of adjacent tissue, bone erosion, and spinal canal involvement [15]. Occasionally, apical tumors may result in Horner's syndrome from the sympathetic ganglion involvement or focal neurological deficits of the brachial plexus [4,16]. In our series, 6 (30%) patients had clinical symptoms at presentation, and 5 of them had complaints of neurologic symptoms.

Thoracic dumbbell tumors are often detected incidentally on imaging. A high index of suspicion is required to identify the intraforaminal involvement in all posterior mediastinal tumors [17]. CT scans may miss the neuroforaminal involvement in a minority of patients [17]. MRI is indicated whenever the tumor is continous with a neural foramen, there is a widened intervertebral foramen or erosion of a vertebral body or pedicle [18]. Neither CT nor MRI scan can accurately differentiate between benign and malignant tumors [19]. The definitive diagnosis is depended on histopathologic examination.

The diagnosis of a dumbbell posterior mediastinal tumor is enough to warrant surgical treatment. It is crucial to fully assess the presence of and degree of intraspinal extension prior to planning the surgical procedure. Unawareness of an intraspinal extension may lead to major complications intraoperatively due to undue traction on the spinal cord [2]. Unknowingly cutting across the narrow foraminal tumor may result in incomplete resection, leaving an intraspinal component. In addition, potential undetected hemorrhage in the foramen or spinal canal, which can result in devastating neurologic sequelae, is difficult to control from a thoracic approach [2,5]. Therefore, a multidisciplinary team combined of thoracic surgeons and neurosurgeons should collaborate together to determine the appropriate approach for individual patients.

Dumbbell tumors are classified into 4 types depending on their locations in the following different spaces: intradural, extradural, intraforaminal, and paravertebral. They are as follows: Type I is intradural and extradural; type II is intradural, extradural, foraminal, and paravertebral; type III is extradural, foraminal, and paravertebral; and Type IV is foraminal and paravertebral [12]. Purely intradural tumors (type I) can be removed with standard open laminectomy, or hemilaminectomy [10]. For large tumors with intraspinal and extraspinal involvement (type II or III), recommended treatment has long been through a single-stage posterior approach by laminectomy, hemilaminectomy with partial costotransversectomy [11], costotransversectomy with extension to a posterolateral thoracotomy [20], or through a combined posterior and transthoracic approach performed either in one stage or in two stages [21]. Perioperative and long-term results of resection of tumors with intraspinal involvement are very good regardless of which techniques are used. In our series, 14 dumbbell tumors (type II or III) were removed with a combined laminectomy and thoracotomy/VATS in a single stage. Advocates of the combined approach claim that this approach avoids traction on the spinal

cord during manipulation of the intrathoracic component and allows a water-tight dural closure to be performed using the neurosurgical microscope [22,23]. Furthermore, the segmental stability may be less compromised [23,24]. VATS has gradually gained acceptance as a safe and reliable minimal access alternative to thoracotomy for the management of posterior mediastinal tumors [1,25]. The open approach may be more appropriate for cases where the tumor is larger, suspicious of malignancy, located at the costophrenic angle or thoracic apex [1,26]. In our cases, a combined approach was successfully performed from T1/2 to T10/T11 in 14 patients. In patients with tumor having neural foraminal extension but without intraspinal involvement (type IV), tumors can be easily removed through transthoracic or supraclavicular approach without any need for laminectomy [23]. In our cases, 4 tumors were successfully excised through this method.

Occasionally dumbell tumors located at the thoracic apex extend to the cervical region. These tumors have always posed particular difficulties to surgeons because of their close association with the large vessels and nerves at the apex of the chest, along with their inaccessibility through lateral thoracotomy [27]. Sakuraba et al. consider that tumors located no lower than T3 can be resected via the cervical incision instead of entering the visceral pleura [28]. Yamaguchi and Akashi described the surgical excision of superior mediastinal tumor by means of a preliminary VATS to dissect the intrathoracic component of the tumor followed by a supraclavicular incision for the complete resection [8,29]. In our cases, a supraclavicular approach or combined with transthoracic approach was successfully performed to remove the tumor at the cervicothoracic junction in 4 patients. The supraclavicular approach afforded an excellent exposure of cervical structures, allowing direct dissection of the superior portion of the thoracic inlet tumors safely. We believe that this approach was conducive to minimize the severity of neurologic injury, thus maximize preservation of the functions of the important nerves.

Complete removal of neurogenic dumbbell tumors has been reported to be successful in almost all cases [11,17]. Long-term results after single-stage resection for dumbbell tumors are excellent with few complications reported [11,17]. The excessive, forceful extraction of tumors could lead to intratumoral bleeding and cord compression. Unrecognized injury to the stellate ganglia and brachial plexus during dissection will cause Horner's syndrome and focal neurological deficits respectively. Unrepaired perforation of the duramater will result in leakage of CSF and may require reoperation. Overall complications in our patients were minor.

One advantage of the single stage combined approach is that initially placing the patient in a prone position permit the use of the neurosurgical operating microscope, and avoid spinal cord traction injury. Second, performing laminectomy and spinal dissection prior to VATS provides a secondary advantage of obtaining tumor mobility, which can simplify the subsequent thoracic surgery. There are several disadvantage to this procedure. First, it need to reposition the patient. Second, it leaves the patient a postoperative chest tube, which is which is required after transthoracic procedures. Finally, this approach may lead to an unnecessary posterior laminectomy on tumors thought to have intraspinal components preoperatively have turned out to be extraspinal.

There are several limitations to our study. First, this is a retrospective observational data study. This study inherent had selection bias. Second, the sample size in this study was small considering the rarity of dumbbell tumors. Third, the follow-up period was relatively short and continued evaluation is needed.

Conclusion

The surgical approach to thoracic dumbbell tumors should be

tailored to the location and extent of intraspinal/neuroforaminal involvement. Most dumbbell tumors can be completely resected via a single-stage posterior laminectomy plus VATS/thoracotomy. Tumors with limited neuroforaminal involvement can be removed via the transthoracic approach alone. A supraclavicular approach or combined with transthoracic approach can be used in tumors located at the cervicothoracic junction.

Disclosure-conflict of interest

Dayu Huang, Xiaofeng Chen make substantial contributions to conception and design, and/or acquisition of data, and/or analysis and interpretation of data; Qinyun Ma, Shaohua Wang and Huijun Zhang participate in drafting the article or revising it critically for important intellectual content; and Dayu Huang give final approval of the version to be published.

Declarations of interest

None.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.ejso.2018.10.536.

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