Clinical Case Report

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Mobile thoracic schwannoma combined with intraosseous schwannomas

A case report

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Abstract

Rationale: Mobile schwannomas have been rarely reported in the lumbar and thoracic spine. These entities are usually intradural extramedullary involving less than 3 vertebrae. Here, we present a rare case of thoracic schwannoma moving over 4 vertebral levels from the primary site combined with intraosseous schwannomas.

Patient concerns: A 64-year-old woman presented with back pain for several months.

Diagnoses: Preoperative computed tomography (CT) and magnetic resonance imaging (MRI) showed 2 intraosseous tumors at the T7 and T8 levels and an intradural extramedullary tumor at the T5-6 levels.

Interventions: The patient underwent a surgical resection of the intraosseous tumors at the T7 and T8 levels, and the tumor at the T5-6 levels was not found. Postoperative MRI showed that the intradural extramedullary tumor had moved to the T3-4 levels. Subsequently, the patient developed gait disturbance and numbness on bilateral lower limbs. During the second operation, we found the tumor at the T1-2 levels. Eventually, the tumor was completely removed.

Outcomes: Histopathological examination showed schwannomas. After a 3-month follow-up, the symptoms were significantly relieved, and there was no clinical or radiological recurrence.

Lessons: The clinicians should be aware of the coincidence of intraosseous schwannomas and mobile schwannoma. Careful preoperative MRIs are essential for early diagnosis of mobile tumors. Intraoperative localization of the mobile tumor is imperative to prevent unnecessary laminotomy.

Abbreviations: CT = computed tomography, MRI = magnetic resonance imaging.

Keywords: case report, intraosseous schwannoma, mobile schwannoma, surgery, thoracic spine

1. Introduction

Spinal mobile tumor was originally reported by Wortzman and Botterell in 1963; the tumor was an ependymoma of the filum terminale and its mobility was due to the unusual laxity of the filum terminale.[1] In the subsequent decades, spinal mobile tumors have been rarely reported, most of which were located in the lumbar spine.[1,2] However, spinal mobile tumors occurring in the cervicothoracic spine were extremely rare.[3,4] Dickson et al[5] reported the first case of intraosseous schwannoma in 1971, and spinal intraosseous schwannomas are rare entities as well.[6] Herein, we present a rare case of thoracic schwannoma moving over 4 vertebral levels from the primary site combined with intraosseous schwannomas. The clinical and radiological profiles and the surgical outcome were presented, and the relevant literature was reviewed.

2. Case report

A 64-year-old woman presented with back pain for several months. Spinal magnetic resonance imaging (MRI) in the local hospital showed space-occupying lesions. On admission, a physical examination showed a slight loss of light touch sensation and thermal hyperalgesia in the lower extremities, and the muscle strength of her bilateral lower extremities was reduced (grade 4/5 according to the British Medical Research Council grading system). MRI showed an intradural extramedullary tumor at the T5-6 levels and 2 intraosseous tumors occupying the vertebral posterior marginal of the T7 and T8 vertebrae (Fig. 1). After administration of gadolinium-diethylene triamine pentaacetic acid (Gd-DTPA), contrast-enhanced T1-weighted images demonstrated that the tumors were heterogeneously enhanced. MRI scanning was performed in the supine position, and no tumor movement or growth was noted compared with the initial scanning.

A surgical resection of tumors was performed via a posterior median approach. After a laminectomy at the T4-8 levels, the intraosseous tumors at the T7-8 levels were completely removed; nevertheless, we did not find a tumor at the T5-6 levels. Then, we expanded the laminectomy to the T3-4 levels and still failed to find a tumor. Considering the potential surgical complications, surgical exploration was terminated. Pathological examination of the

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Consent for publication: Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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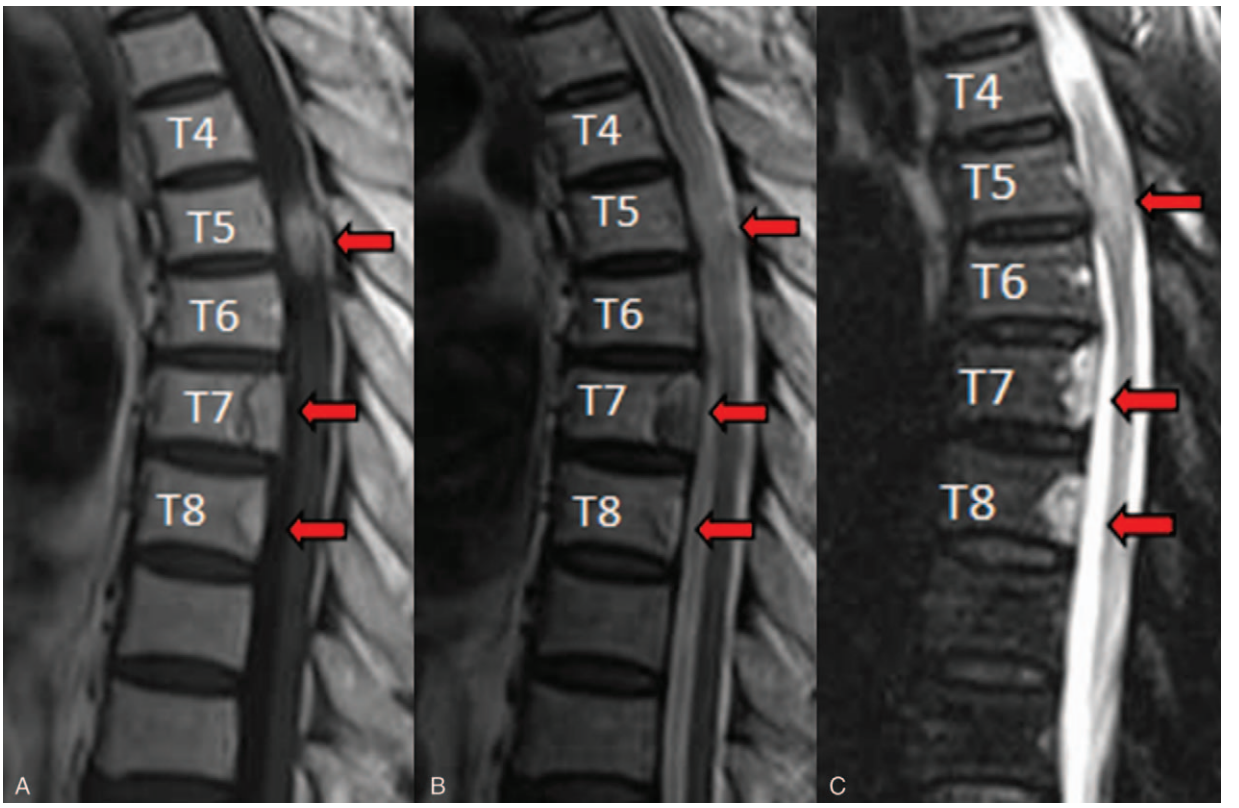

Figure 1. Serial preoperative magnetic resonance imaging. (A) T1-weighted imaging; (B) T2-weighted imaging; (C) Gd-DTPA contrast imaging) showed an intradural extramedullary tumor (red arrow) at the T5-6 levels and 2 intraosseous tumors at the T7-8 levels (red arrow).

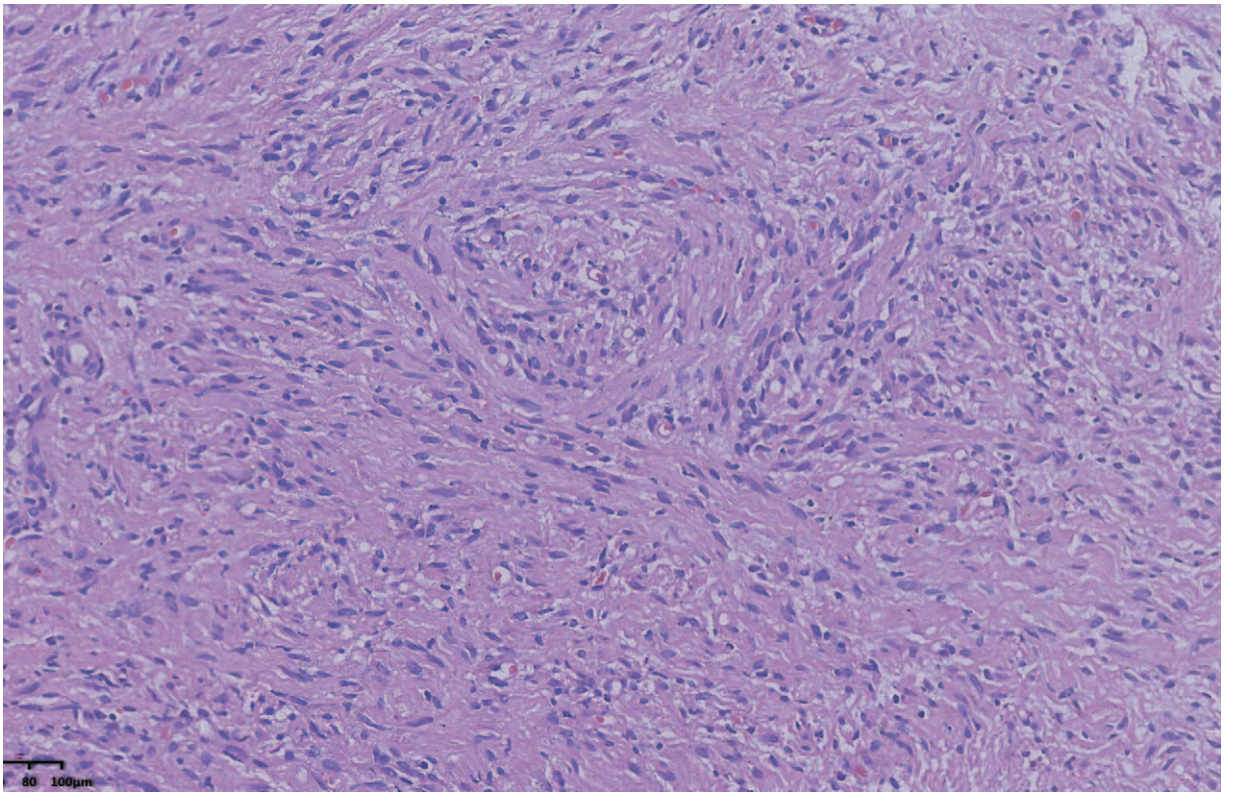

Figure 2. Pathological examination of the intraosseous tumors at the T7-8 levels showed schwannomas (The original magnification was 200×.).

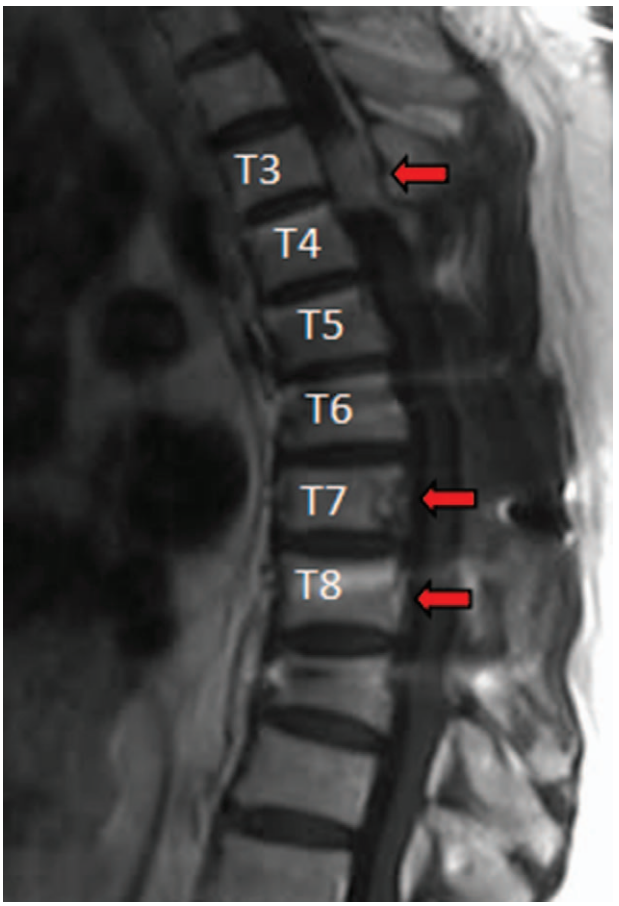

Figure 3. Postoperative magnetic resonance imaging showed that the intraosseous tumors had been removed while the intradural extramedullary tumor (red arrow) had moved from the T5-6 levels to the T3-4 levels.

Figure 5. The tumor (1.7 cm×0.8 cm) was completely resected.

intraosseous tumors at the T7-8 levels showed schwannomas(Fig. 2). Postoperatively, repeated MRI showed that the intradural extramedullary tumor had moved to the T3-4 levels (Fig. 3).

Six months later, the patient developed progressive clumsiness in the lower extremities. Eight months after the first operation, she was readmitted to our hospital. A physical examination showed the weakness in her bilateral lower extremities was aggravated. The muscle strength of her hip flexors and knee extensors was grade 2/5. The bilateral knee jerk reflexes were exaggerated, and she had severe numbness below the T4 dermatome. After a dural incision at the T2-3 levels, we failed to find the tumor (Fig. 4A and B). Then we incised the dura mater longitudinally at the T1-2 levels, and a yellowish tumor was discovered underneath the nerve root and the dentate ligament (Fig. 4C). The tumor was well demarcated, and there was no structural adhesion to the spinal cord or the dura mater. The

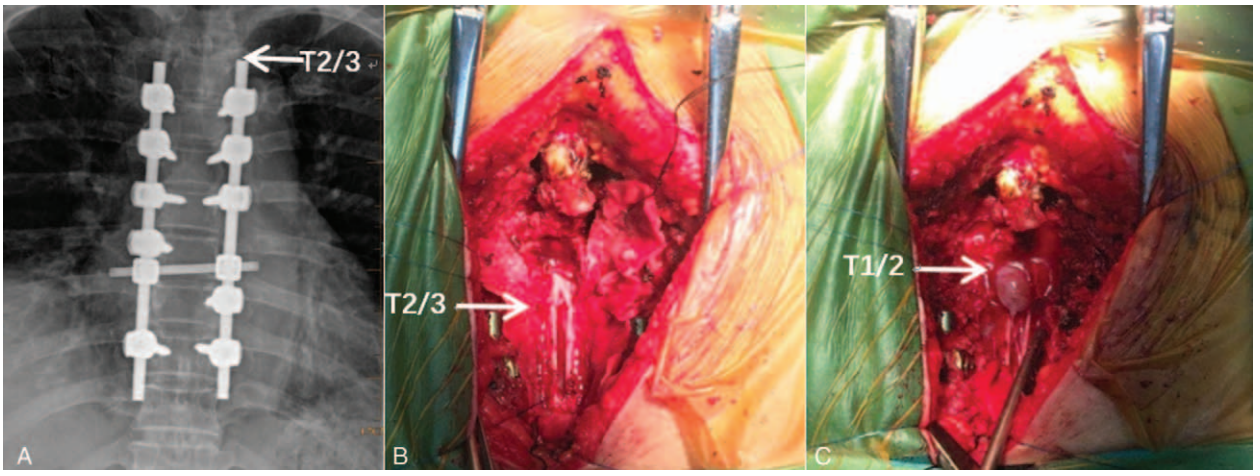

Figure 4. (A and B) After a dural incision at the T2-3 levels (white arrow), we failed to find the tumor. (C) After the dura at the T1-2 levels was incised longitudinally (white arrow), the tumor was found.

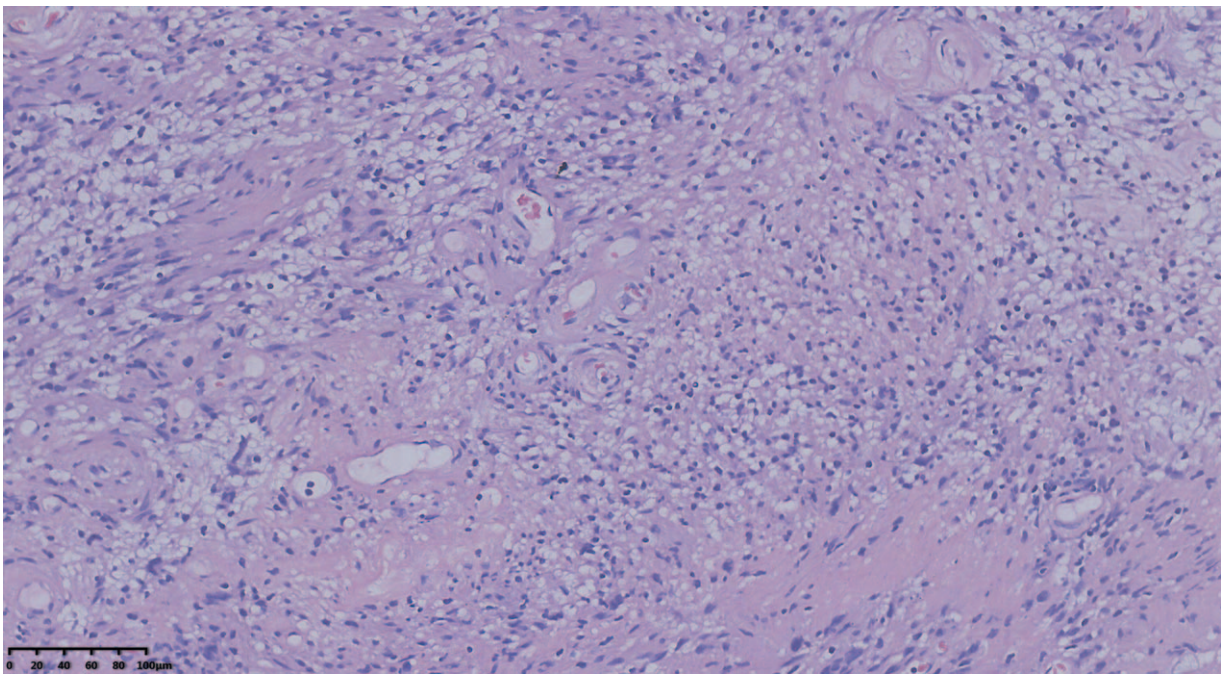

Figure 6. Pathological examination of the extramedullary intradural tumor at the T1-2 levels showed a schwannoma (The original magnification was 200×).

tumor was completely removed (Fig. 5). Histopathological examination showed a schwannoma (Fig. 6). Three days postoperatively, the gait disturbance and numbness of the bilateral lower limbs were relieved. The muscle strength of the hip flexors and knee extensors was improved to grade 3/5 on the right side and grade 4/5 on the left side. After a 3-month follow-up, there was no clinical or radiological recurrence.

3. Discussion

There are 3 mainstream hypotheses regarding the pathogenesis of intraosseous schwannomas: the bone is eroded by an extraosseous tumor, the tumor arises within the nerve canal and grows in a dumbbell-shaped configuration leading to enlargement of the canal, and the tumor arises primarily within the bone.[7] In this case, we found a part of the tumor was extraosseous, which supported the second hypothesis.

Mobile tumors occurring in the spinal canal are extremely rare. In previous literature, neurological deterioration related to tumor mobility has only been reported in 3 cases, and the predisposing factors included intrathecal contrast injection,[3] trauma,[2] postural change,[8] Valsalva effects at micturition and defecation,[3,8,9] pulsatile flow of the cerebrospinal fluid,[4] and the laminectomy procedure.[3,9] Tumor mobility is multifactorial, including the pulsatile flow of the cerebrospinal fluid, elongation of the nerve root from which the tumor arises, and dilated subarachnoid space. Some scholars proposed that the elongation of the nerve root by tension resulting from the tumor weight would contribute to mobility of an attached tumor.[10,11] Moreover, extension of the lumbar spine and hips can cause a considerable laxity of the nerve roots.[12] It appears that intradural pulsatile flow of the cerebrospinal fluid and gravity of the tumor may induce an elongation of the nerve root, which contributes to tumor mobility as well.[3,9,13] A spinal cord deformity induced by the extramedullary tumor can contribute to the dilatation of subarachnoid space, which may be another cause of tumor mobility.[13] As the subarachnoid space in the cervicothoracic spine is narrower and the nerve roots are shorter than their counterparts in the lumbar spine, the mobile tumors in the cervicothoracic spine are exceedingly rare. Some clinicians attributed the tumor shifting to the thrust of injected radiopaque material during myelography.[14,15] At present, myelography has been replaced by MRI for preoperative evaluation. In our case, there was no intrathecal injection of contrast material.

During the first surgery, we expanded the laminectomy to the T3-4 levels but still missed the tumor. However, the subsequent MRI revealed that the tumor was at the T3-4 levels. Postural changes may explain this discrepancy. In addition, the dural suture at the T3-4 levels blocked the backward movement of the tumor to the T5-6 levels, which can explain the different locations on the preoperative and postoperative MRIs. Furthermore, we found that the nerve root ventral to the tumor was markedly elongated, with pulsatile flow of the cerebrospinal fluid. We speculate that the extensive mobility of schwannoma in the thoracic spine may result from multiple factors including the elongation of the nerve root, postural change, pulsatile flow of the cerebrospinal fluid, and the dural suture.

Preoperative evaluation of mobile schwannomas is essential to prevent extended laminotomy.[3,16,17] When the movement of the tumor is beyond 2 or more vertebral levels, the surgical localization may be challenging and comprehensive application of multiple modalities (such as intraoperative myelography, ultrasonography, and MRI) can be helpful.[2,3,13,15,17–19]

4. Conclusion

We reported an extremely rare case of mobile schwannoma combined with intraosseous schwannomas in the thoracic spine. The clinicians should be aware of both intraosseous schwannomas and mobile schwannomas attaching to the nerve roots. Careful preoperative MRIs are essential for early diagnosis of mobile tumors. Intraoperative accurate localization of the mobile

tumor should be highlighted in order to avoid unnecessary laminotomy.

Author contributions

Data curation: Yang Huang, Tao Chen.
Funding acquisition: Tao Chen.
Validation: Junhui Guan.
Writing – original draft: Shunjie Jia.
Writing – review & editing: Shunjie Jia, Wenbiao Zheng, Jianwei Ruan.

References

[1] Wortzman G, Botterell EH. A mobile ependymoma of the filum terminale. J Neurosurg 1963;20:164–6.
[2] Sasaki M, Aoki M, Yoshimine T. Mobile schwannoma of the cauda equina incarcerated following caudal migration after trauma—case report. Neurol Med Chir 2011;51:710–2.
[3] Namura S, Hanakita J, Suwa H, et al. Thoracic mobile neurinoma. Case report. J Neurosurg 1993;79:277–9.
[4] Terada Y, Toda H, Yokote A, et al. A mobile schwannoma of the cervical spinal cord: case report and review of the literature. Neurosurgery 2016;78:0000000000000975.
[5] Dickson JH, Waltz TA, Fechner RE. Intraosseous neurilemoma of the third lumbar vertebra. J Bone Joint Surg Am 1971;53:349–55.
[6] Song D, Chen Z, Li Z. Lumbar intraosseous schwannoma: case report and review of the literature. Turk Neurosurg 2014;24:982–6.
[7] Gordon EJ. Solitary intraosseous neurilemmoma of the tibia: review of intraosseous neurilemmoma and neurofibroma. Clin Orthop Relat Res 1976;117:271–82.
[8] Kim SB, Kim HS, Jang JS, et al. Mobility of intradural extramedullary schwannoma at spine: report of three cases with literature review. J Korean Neurosurg Soc 2010;47:64–7.
[9] Marin-Sanabria EA, Sih IM, Tan KK, et al. Mobile cauda equina schwannomas. Singapore Med J 2007;48:e53–56.
[10] Hollin SA, Drapkin AJ, Wancier J, et al. Mobile schwannoma of the cauda equina. Case report. J Neurosurg 1978;48:135–7.
[11] Pau A, Orunesu G, Sehrbundt Viale E, et al. Mobile neurinoma of the cauda equina. Case report. Acta Neurochir 1982;60:115–7.
[12] Ehni G, Moiel RH, Bragg TG. The "redundant" or "knotted" nerve root: a clue to spondylotic cauda equina radiculopathy. Case report. J Neurosurg 1970;32:252–4.
[13] Iizuka H, Iida T, Akai T. Mobile neurinoma of the cervicothoracic junction. Surg Neurol 1998;50:492–3.
[14] Caracalos A. Elusive cauda equina tumor. J Neurosurg 1987;67:952.
[15] Tavy DL, Kuiters RR, Koster PA, et al. Elusive tumor of the cauda equina. Case report. J Neurosurg 1987;66:131–3.
[16] Isu T, Iwasaki Y, Akino M, et al. Mobile schwannoma of the cauda equina diagnosed by magnetic resonance imaging. Neurosurgery 1989;25:968–71.
[17] Khan RA, Rahman A, Bhandari PB, et al. Double migration of a schwannoma of thoracic spine. BMJ Case Rep 2013;23:2012–008182.
[18] Friedman JA, Atkinson JL, Lane JI. Migration of an intraspinal schwannoma documented by intraoperative ultrasound: case report. Surg Neurol 2000;54:455–7.
[19] Friedman JA, Wetjen NM, Atkinson JL. Utility of intraoperative ultrasound for tumors of the cauda equina. Spine 1976;28:288–90.