

A Rare Case of Migratory Intradural Extramedullary Schwannoma of Dorsal Spine: A Case Report and Review of Literature

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Abstract: *Background:* Mobile or Migratory intradural extramedullary schwannomas are very rare and have been reported more in lumbar spine and very less in dorsal spine and cervical spine. The exact cause of migration of schwannoma arising from nerve sheath of a spinal nerve root is unclear and are mysterious in cervical and dorsal spine. Mobile schwannomas require careful evaluation of their localisation otherwise it leads to unnecessary laminectomies. We report a 16-year-old male who presented with weakness in B/L lower limbs and unable to walk for 4 months, MRI spine revealed an intradural extramedullary lesion at D1 D2 level of spinal cord. Intraoperatively the lesion was not found. On repeat MRI it was found migrated caudally to D3 D4 level. Patient was reoperated and tumor was found at D3 D4 level and was excised. Histopathology confirmed a benign schwannoma. This is the rare occurrence of caudal migration of dorsal intradural extramedullary schwannoma.

Keywords: Schwannoma, Dorsal spine tumour, Rare, Intradural, Extramedullary, Migratory

1. Introduction

Migratory intradural extramedullary lesions are very rare. Most common migratory intradural extramedullary tumors are Schwannoma, ependymoma and neurenteric cyst¹. Schwannoma is the most common nerve sheath tumour constituting about eighty-five percent of nerve sheath tumours, combined with neurofibromas these constitute about 30% of all spinal neoplasms². They occur equally among both sexes and are common in fourth to sixth decade. Nerve sheath tumours are dispersed equally along the various spine segments with no clear predilection to any particular region of spine. Migratory schwannomas are thought to be very rare and sparsely documented in available literature, lumbar spine is the most frequent site of mobile schwannomas. However, there are case reports available in literature depicting their occurrence in cervical and thoracic spine. These migratory tumours source diagnostic as well as surgical hurdles, in worst case scenario the operative finding didn't revealed any tumour at the predetermined level and an extension of laminectomy and durotomy was needed, even that was not successful in all the cases³⁻⁵. Mobile schwannoma are most common in lumbar spine (65%), dorsolumbar junction(15%), dorsal spine(10%), cervical(5%) and cervico dorsal(5%). Direction of movement is rostral (60%), caudal (30%) and 10% to and fro⁶. Here we present a missing dorsal INTRADURAL EXTRAMEDULLARY SCHWANNOMA in a patient intraoperatively which was found to have migrated caudally on repeat MRI. We also summarize review of literature of such cases and preventive measures to avoid unnecessary surgeries at wrong vertebral level and importance of intraoperative imaging.

2. Case Report

A 16year old boy presented with weakness in B/L lower limbs and unable to walk since 4 months. On examination patient was conscious and oriented, paraplegic with sensory deficit below nipple levels with exaggerated lower limb deep tendon reflexes, babinski positive and ankle clonus present.

Patient was admitted, evaluated and was diagnosed with 21 x 14mm intradural extramedullary lesion at D1-D2 level on MRI spine. Provisional diagnosis of schwannoma was made. Pre-operative workup was done and patient underwent D1-D2 laminectomy. Intraoperatively lesion was not found. Intraoperative MRI and USG facility was not available at our institution; surgery was abandoned in view of the same. Post operatively MRI was repeated and it revealed that the tumor had migrated to D3 - D4 Level. Patient was taken for reoperation. Laminectomy was extended to D3-D4 level. After dural opening tumor was found and was excised completely and sent for histopathological examination. Post op period was uneventful. Physiotherapy was continued in post operative period. His motor functions improved. He was able to walk using walker.

3. Discussion

Mobile intraspinal tumors are rare. Most common mobile spinal tumor are schwannoma, ependymoma and neurenteric cyst, schwannoma being the most common¹. The mobile schwannoma of cervical region was first described by Tomimatsu et al in 1974⁷. Schwannomas are nerve sheath tumors that are relatively well marginated with little attachment to the adjacent tissues. In most of the cases, there are no attachments to dura and the lesion is held by the nerve root at its two ends. Tumor movement is most commonly found in lumbar spine because of absence of spinal cord and presence of redundant nerve roots⁸.

In cervico thoracic region, presence of spinal cord itself allows less space for mobility. So, mobile spinal tumors are rare at cervical and thoracic location as compared to lumbar region⁸. Migration of a spinal tumor is a rare phenomenon and etiology is unclear. Disconnection of tumor has been hypothesised to be secondary to increased subarachnoid space around the tumor, stretching of the attached nerve root and CSF pulsations^{9,10}. Possible forces that can drive the movement of schwannomas include postural changes, the thrust of injected radiopaque material during myelography,

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Valsalva effects at micturition and defecation. Factors that contribute to tumor mobility include dilated subarachnoid space and elongation of nerve root from which tumor originates, as well as a pulsatile flow of cerebrospinal fluid¹¹. In a study and literature review by Kim SB, more mobile tumors were found in the cauda equina region and rostral migration was more common than the caudal migration¹. Tumor movement could be attributed to positional adjustments or any procedure which increases intra-abdominal, intrathecal or intra-thoracic pressure^{12,13}. In our case, the tumor was initially located at D1-D2 level on the first MRI and migrated caudally to lie at the level of D3-D4 level as was seen on the repeat post-operative MRI. Possible aetiology for tumor mobility in this location appears to be elongation of nerve roots by tumor weight. Various methods have been used to tackle the problem of migration and prevent unnecessary laminectomies. These include intraoperative MRI, ultrasonography, myelography.

Intraoperative myelography should be done before dural incision. Again, tumor might get displaced because of the force exerted during contrast injection. Still, it is a preferred modality to determine the precise level of laminectomy. Intraoperative MRI can be done only in well-equipped hospitals. However, most health institutions do not have such facilities. Intraoperative USG appears to be the most convenient method to find out the migrated tumor if the tumor is missing at the expected site^{6,14}. The duration between imaging and surgery should be short as possible. In our presented case, the duration between surgery and MRI was 10 days, therefore the possibility of tumor migration prior to surgery cannot be ruled out, hence MRI should be repeated just prior to surgery¹⁴. To the best of our knowledge, till date 6 dorsal (present only in dorsal) migratory schwannoma have been reported^{6,14,15}. We are presenting a rare case report of Dorsal Migratory Schwannoma which has migrated caudally (D1- D2 TO D3 -D4 Level).

All reported cases of mobile spinal schwannoma in literature, degree of migration (Adapted and updated from Kothari 2017¹).

No	Author	Location	Discrepancy	Migration
1	Tomimatsu 1974 [4]	Cervical	2 VB	Caudal
2	Hollin 1978 [5]	Lumbar	3 VB	Rostral
3	Husag 1980 [12]	Lumbar	7 cm	Caudal
4	Pau 1982 [13]	Lumbar	2 VB	Caudal
5	Tavy 1987 [14]	Lumbar	3 VB	Caudal
6	Isu 1989 [15]	Thoracolumbar	1 VB	Caudal
		Lumbar	1 Vb	Caudal
		Lumbar	1/2 VB	Caudal
7	Satoh 1991 [16]	Thoracolumbar	1 Vb	Rostral
8	Namura 1993 [6]	Thoracic	3 VB	Caudal
9	Varughese 1997 [17]	Lumbar	1 VB	Rostral
10	Iizuka 1998 [10]	Cervicothoracic	1 VB	Rostral
11	Friedman 2003 [18]	Lumbar	1 VB	Rostral
		Lumbar	1/2 VB	Rostral
		Lumbar	1/2 VB	Rostral
12	Kim 2005 [7]	Thoracic	1 VB	Caudal
13	Martin-Sanabria 2007 [19]	Lumbar	2.2 cm	Rostral
		Lumbar	2 VB	Caudal
14	Kim 2010 [8]	Lumbar	1 VB	Rostral
		Thoracic	1/2 VB	Rostral
		Lumbar	1	Rostral
15	Sasaki 2011 [20]	Lumbar	1	Rostral
16	Khan 2013 [3]	Thoracic	3 VB	Rostral, caudal
17	Terada 2016 [11]	Cervical	1 VB	Caudal
18	Toscano 2016 [21]	Thoracolumbar	1 VB	Caudal
19	Kothari 2017 ¹	Lumbar	1 VB	Caudal
20	Jia 2019 [9]	Thoracic	2 VB	Rostral
21	Present Case 2019	Thoracic	4 VB	Rostral, caudal

Table 2. Summary of Published Cases of Migratory Schwannoma with Probable Cause of Migration

Study	Location	Discrepancy	Probable Cause of Migration
Tomimatsu et al., 1974 ²	Cervical	2 vertebrae	CSF dynamics
Hollin et al. ¹⁵	Cauda equina	3 vertebrae	Traction during positioning
Husag et al., 1978 ¹⁵	Cauda equina	7 cm	Redundant nerve root
Pau et al., 1982 ¹⁹	Cauda equina	2 vertebrae	Redundant nerve root
Tavy et al., 1987 ¹⁶	Cauda equina	3 vertebrae	Change in posture
Isu et al., 1989 ⁶	Dorso Lumbar Lumbar	1 vertebra 1 vertebra One half vertebral body	Change in posture
Satoh et al., 1991 ²⁰	Dorso Lumbar	1 vertebra	Redundant nerve roots
Namura et al., 1993 ¹³	Dorsal	5 vertebrae	Positive pressure ventilation during the laminectomy and dilated subarachnoid space
Varughese and Mazagri, 1997 ²¹	Lumbar Lumbar	1 vertebra 1 vertebra	Redundant nerve root
Iizuka et al., 1998 ²²	Cervico dorsal	1 vertebra	Traction on nerves because of flexion of neck in prone position, dilated subarachnoid space, and laminectomy procedure
Friedman et al., 2003 ¹⁴	Lumbar Lumbar Lumbar	1 vertebra One half vertebral body One half vertebral body	Redundant nerve root
Marin-Sanabria et al., 2007 ¹²	Lumbar Lumbar	2.2 cm 2 vertebrae	Strained during micturition (Valsalva maneuver)
Kim et al., 2010 ¹	Lumbar Dorsal Lumbar	1 vertebra One half vertebral body 1 vertebra	Intrathecal pressure or spine posture and dilated subarachnoid space
Khan et al., 2013 ²³	Dorsal	3 vertebrae	Redundant nerve root and dilated subarachnoid space
Terada et al., 2016 ⁹	Cervical	1 vertebra	CSF dynamics and dilated subarachnoid space
Toscano et al., 2016 ⁷	Dorso lumbar	1 vertebra	Redundant nerve root
Naik et al., 2016 ¹⁷	Cervico dorsal	2 vertebrae	Elongation of nerve roots by tumor weight, spinal cord deformity and enlargement of subarachnoid space
Kothari et al., 2017 ⁵	Lumbar	1 vertebra	Redundant nerve root
Lee et al., 2017 ¹⁰	Lumbar	One half vertebral body	CSF dynamics
Kasam et al., 2017 ²⁴	Lumbar	1 vertebra	CSF dynamics, dilated subarachnoid space, change in posture, and redundant nerve root
Current study	Dorsal	2 vertebrae	CSF dynamics, dilated subarachnoid space, and redundant nerve root

Mobile Schwannoma of spine; published cases in literature.

S. No	Author	Location	Discrepancy	Migration
1.	Tomimatsu et al. ²¹	Cervical	2 vertebrae	Caudal
2.	Hollin et al. ¹⁹	Cauda equina	3 vertebrae	Rostral
3.	Husag et al. ¹⁵	Cauda equina	7 cm	Caudal
4.	Pau et al. ⁹	Cauda equina	2 vertebrae	Caudal
5.	Tavy et al. ²⁰	Cauda equina	3 vertebrae	Caudal
6.	Isu et al. ¹⁴	Thoracolumbar Lumbar Lumbar	1 vertebrae 1 vertebrae 1/2 vertebrae	Caudal Caudal Caudal
7.	Satoh et al. ¹⁷	Thoraco-lumbar	1 vertebrae	Rostral
8.	Namura et al. ¹²	Thoracic	3 vertebrae	Caudal
9.	Varughese and Mazagri ³	Lumbar Lumbar	1 vertebrae 1 vertebrae	Rostral Rostral
10.	Iizuka et al. ²²	Cervico thoracic	1 vertebrae	Caudal
11.	Friedman et al. ¹³	Lumbar Lumbar Lumbar	1 vertebrae 1/2 vertebrae 1/2 vertebrae	Rostral Rostral Rostral
12.	Marin-Sanabria et al. ⁴	Lumbar Lumbar Lumbar	2.2 cm 2 vertebrae 1 vertebrae	Rostral Caudal Rostral
13.	Kim et al. ⁵	Lumbar Lumbar Thoracic	1/2 vertebrae 1 vertebrae 1/2 vertebrae	Rostral Rostral Rostral
14.	Khan et al. ²	Thoracic	3 vertebrae	Rostral
15.	Terada et al. ¹¹	Cervical	1 vertebrae	Caudal
16.	Toscano et al. ¹⁸	Thoracolumbar	1 vertebrae	Caudal
17.	Kothari et al. (present study)	Lumbar	1 vertebrae	Caudal

4. Conclusion

We report a rare presentation of migratory dorsal intradural extramedullary schwannoma with caudal migration. Migratory schwannoma should be suspected in case of missing lesion during surgery. Intraoperative MRI, ultrasonography facility should be made available in hospitals. Preventive measures should be taken to avoid tumor migration. Purpose of this report is to remind about the possibility of migration of intradural extramedullary tumor when there is negative exploration in the expected area of surgery.

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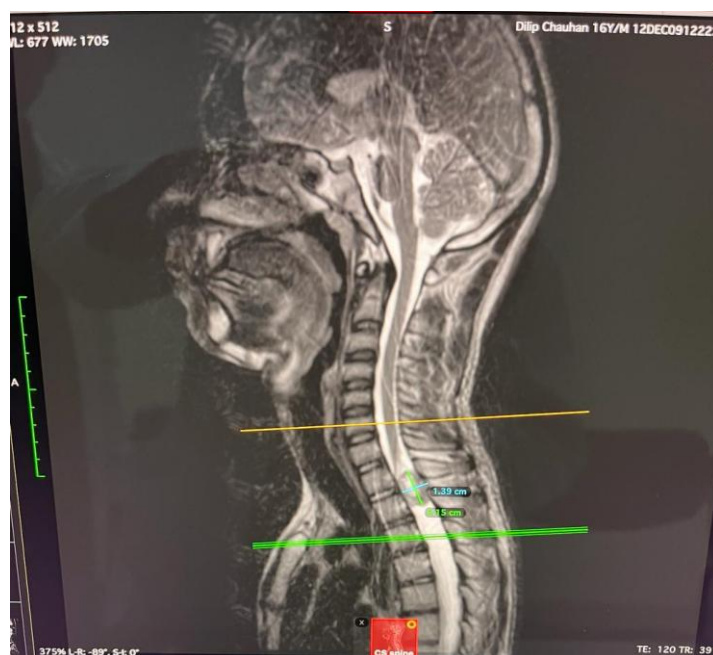


Figure 1: First MRI Scan Tumor at D1 D2 Level.

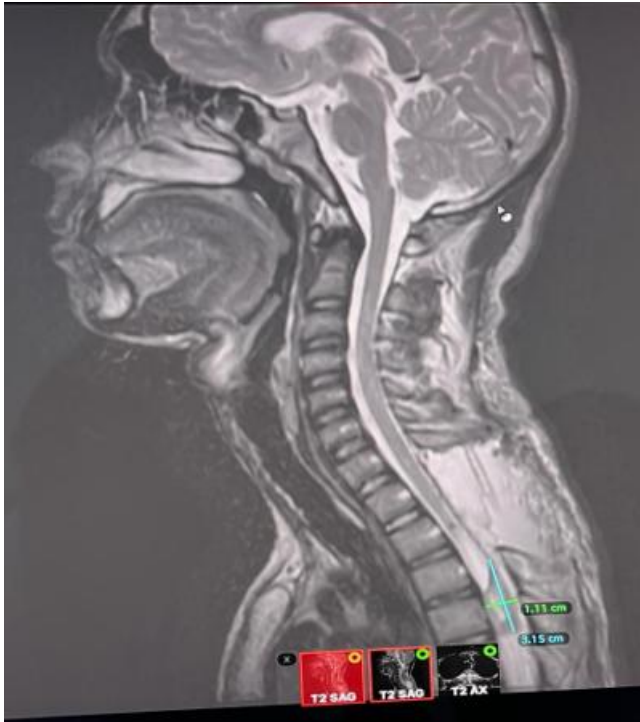


Figure 2: Repeat MRI Scan Tumor Migration to D3 D4 Level



Figure 4 : Intra Operative Picture



Figure 3: Comparison of Pre op and Post op scan.

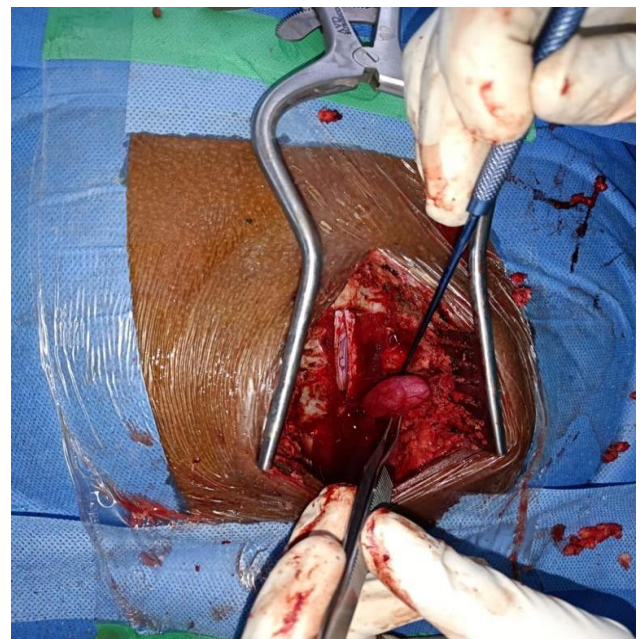


Figure 5: Intra operative picture

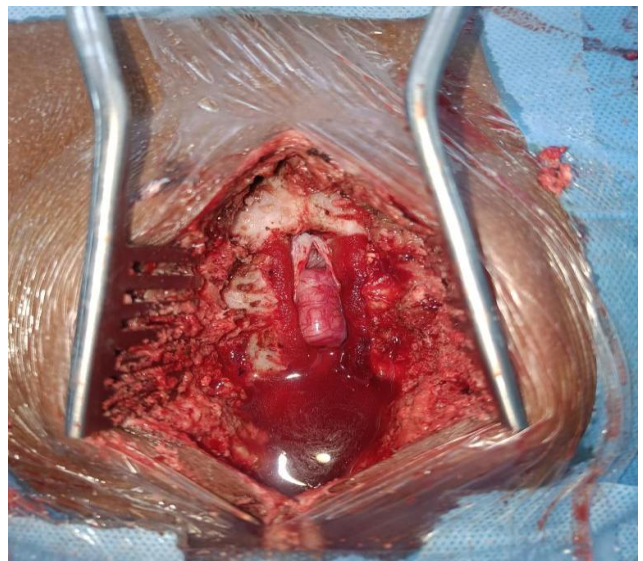


Figure 6: Intra operative picture



Figure 7: Tumor specimen



Figure 8: Tumor specimen (2.5cm length)

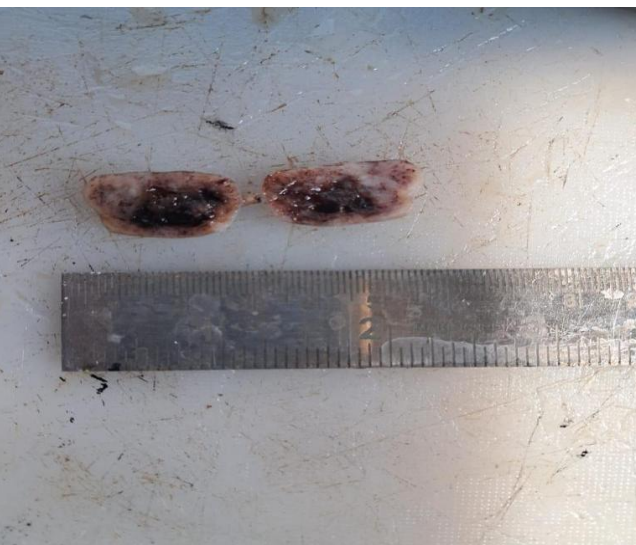


Figure 9: Tumor Cut section

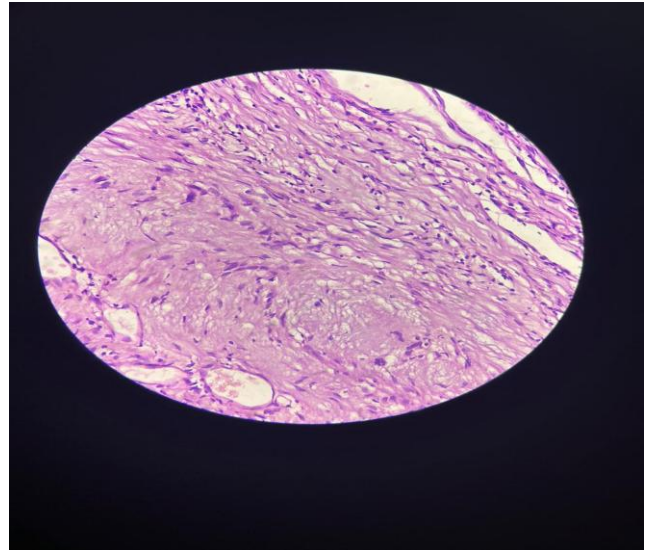


Figure 10: Microscopy appearance: Spindle cells arranged in fascicles with hypocellular and hypercellular areas. Antoni A & B type pattern showing verocay bodies.

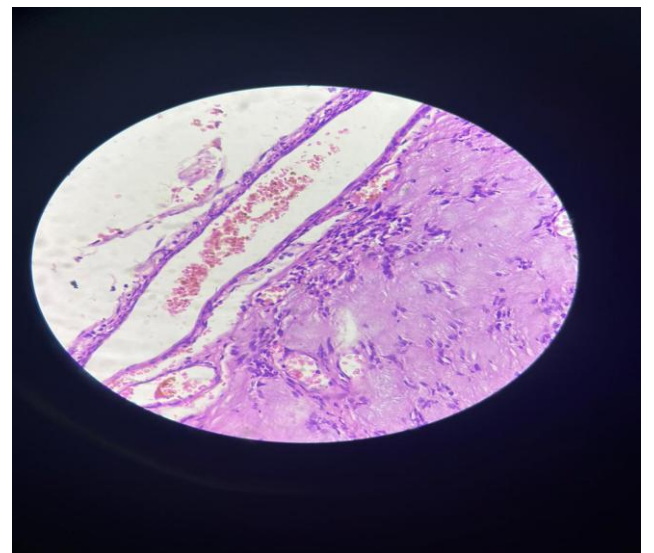


Figure 11: Microscopy findings: Blood vessels are thickened, ectatic with hyalinisation

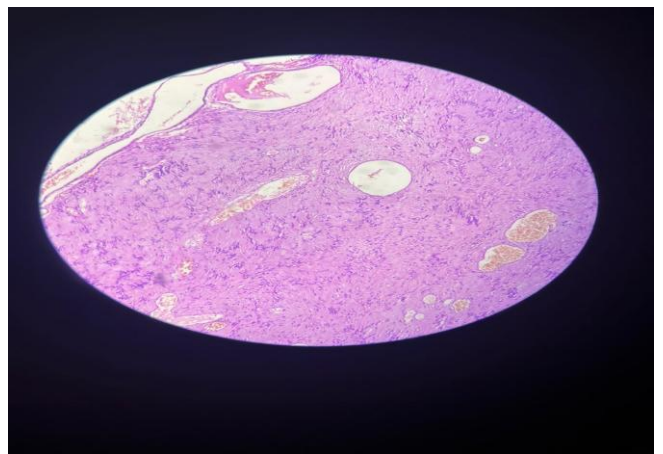


Figure 12: Microscopy findings: Nuclear palisading seen with presence of ill- defined verocay bodies, spindle cells arranged as fascicles, Schwannoma.