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Title:

Spinal cord herniation following resection of cervical spinal neurofibroma with a unique presentation

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ABSTRACT

Background: Spinal Cord herniation (SCH) is a very rare condition. It was first reported in the lumbar spine in 1974. Thereafter cases were reported in the thoracic and cervical spine occurring either spontaneously or following vertebral fracture, nerve root avulsion, and trauma surgery. ***Purpose***: There is only one recorded case of spinal cord herniation following tumor surgery. In this paper we report the second case. ***Design/Setting***: We describe the original surgical procedure, the clinical presentation, the operative repair and the post-operative course. ***Methods***: No funding was required for this case report. ***Results***: The patient was a 56 years old male who presented with spinal cord herniation five years following subtotal excision of a cervical neurofibroma. He presented with right upper monoparesis. ***Conclusion:*** To our knowledge, this presentation was not reported before in the literature.

INTRODUCTION

Post-operative spinal cord herniation (SCH) following resection of spinal tumor is rare. To our knowledge, only one case has been reported in the literature[6]. We report a second case exhibiting unique clinical features which were not reported previously. We describe the original surgical procedure, the clinical presentation, the operative repair and the post-operative course.

CASE REPORT

First Presentation

The patient was a 56 year old right-hand dominant male. He had a past history of left sided Bell's palsy and Dupuytren's contracture of the right palm. His first presentation to our service was in 2007 when an incidental right sided dumbbell neurofibroma at C2/3 level was identified on an MRI of his neck performed to

investigate a painful cervical lymphadenopathy. Clinical examination revealed no neurological deficit.

A C2/3 right hemi-laminectomy and partial facetectomy was performed to expose the lesion. The Dura was opened in the midline and a “T” limb was performed over the nerve root. The tumor was found indenting the Dura and pushing the thecal sac medially. The canalicular component was completely resected. A small residuum was left laterally to avoid injury to the vertebral artery. The Dura was closed in a non-watertight fashion with interrupted 5-0 prolene. No Dural graft was employed.

He had an uneventful recovery and follow up MRI over 4 years revealed a stable tumour remnant.

Second Presentation

He re-presented five years later complaining neck pain and progressive weakness of the right upper limb, more pronounced distally with loss of fine hand movements. There was numbness and a tingling sensation in his right middle and ring fingers and forearm.

Examination revealed global weakness of the right upper limb with power grade three out of five. He was unable to perform fine motor tasks. He had reduced pinprick sensation on the medial aspect of his forearm and tip of his middle and ring fingers.

Radiology

An MRI of his cervical spine revealed a pseudomeningocoele with spinal cord herniation through the operative defect at the level of C2 vertebra. The cord was compressed by the bone edges. The tumour remnant was stable (Appendix 1 figures1 and 2).

Operation

The patient was placed in the prone position with 3-point cranial fixation. Through a posterior cervical approach the pseudomeningocoele was opened. The spinal cord

was found herniating through the dural defect and incarcerated with loss of normal pulsation (Appendix 2 figure 1). The defect was enlarged and the arachnoid adhesions were divided. The cord was reduced back into the dural sac. The Dural defect was sealed with a Dural substitute (Neuro-patch™: polyurethane non-absorbable patch) using interrupted 5-0 prolene sutures (Appendix 2 figure 2).

Follow-up

The patient had an uneventful recovery. He reported immediate improvement in the fine motor movements of his right hand and complete resolution of his neck pain.

DISCUSSION

Spinal cord herniation was first identified by Wortzman and colleagues in the lumbar spine in 1974[13]. Thoracic SCH was described by Aydin et al and Chordia [1,4]. Aydin and colleagues reported spontaneous SCH through two separate adjacent Dural defects in the thoracic region[1]. Belen D and colleagues described SCH following foramen magnum decompression in 2009 [2]. SCH was also reported to occur after trauma including vertebral body fractures [10], following decompressive spinal surgery for trauma [3], and after nerve root avulsion [7,11].

Hosono and colleagues reported the only case of spinal cord herniation following resection of a cervical spinal tumour. Their patient presented with gait disturbance 14 years after cervical laminectomy [6].

The interval between the causative incident and SCH was variable in the literature ranging from 3 to 14 years[6,11]. Post-operative SCH occurred 7 years after foramen magnum decompression[2], and 14 years following cervical laminectomy for tumor resection[6]. Our patient presented in a relatively short time period of 5 years.

The clinical features of the condition were summarized by Watters MR and colleagues in his review of SCH [12]. The clinical syndromes of SCH included unilateral

pyramidal signs, worsening paraparesis, and progressive Brown-Sequard syndrome [12]. Burres KP and Conley FK described quadriparesis in their patient with cervical SCH [3].

Our patient presented with a right upper limb monoparesis. There were no other long tract signs. To our knowledge, this has not been reported. We offer no explanation for these findings

Magnetic resonance imaging (MRI) was the diagnostic modality of choice in all reports of SCH. It showed pseudomeningocoele with herniation of the cord into the cyst [1,12,14]. High intensity signal areas in the T2-weighted images of the herniated cord suggestive of cord damage were found in SCH following tumor resection [6]. Other abnormalities reported were apparent syrinx formation in a post-traumatic case which was not found later during surgery, and vertebral body and nuclear trail sign [12].

The aetiology of post-operative SCH is thought to be arachnoid adhesions causing spinal cord tethering around the operative Dural defect [2,6]. Belen further concluded in his report that CSF circulatory disturbances due to arachnoid scarring facilitated herniation of the cord through the Dural defect [2].

In resecting dumbbell tumors it has been the senior author's practice to open the Dura in the midline and make a" T "incision over the tumor as necessary. Closure of the Dura has been performed with interrupted sutures approximating the edges in a non-water tight fashion. This is the first time the author has encountered a SCH using this technique. On the balance of probability had a Dural graft been employed this complication would not have occurred.

The principals of management of SCH are: (1) adequate exposure, (2) untethering and reducing the spinal cord in an atraumatic fashion, and (3) repair of the defect. Both Hosona and Belen described untethering of the spinal cord microsurgically by lysing the adhesions around the defect, then reducing it back in the thecal sac and closing the defect with a Duraplasty [2,6]. Aydin and colleagues reported a similar technique in managing their case of spontaneous thoracic cord herniation in two areas;

they connected the two defects, reduced the spinal cord, and reinforced the single big defect with a fascial graft before closing it with interrupted stitches [1].

CONCLUSION

SCH is a rare condition that can occur spontaneously or following spinal surgery requiring a durotomy. It presents in a delayed and variable fashion. The proposed pathology of the condition is spinal cord tethering around the Dural defect resulting in localized CSF flow abnormalities. The principles of operative management are untethering and reduction of the spinal cord and repair of the Dural defect.

REFERENCES

1. Aydin AL, Sasani M, Erhan B, et al. Idiopathic spinal cord herniation at two separate zones of the thoracic spine: the first reported case and literature review. *Spine J*;11:e9-e14.
2. Belen D, Er U, Gurses L, et al. Delayed pseudomyelomeningocele: a rare complication after foramen magnum decompression for Chiari malformation. *Surg Neurol* 2009;71:357-61, discussion 61.
3. Burres KP, Conley FK. Progressive neurological dysfunction secondary to postoperative cervical pseudomeningocele in a C-4 quadriplegic. Case report. *J Neurosurg* 1978;48:289-91.
4. Choradia M. Idiopathic spinal cord herniation. *Neurol India*;60:342-3.
5. DaSilva VR, Al-Gahtany M, Midha R, et al. Upper thoracic spinal cord herniation after traumatic nerve root avulsion. Case report and review of the literature. *J Neurosurg* 2003;99:306-9.
6. Hosono N, Yonenobu K, Ono K. Postoperative cervical pseudomeningocele with herniation of the spinal cord. *Spine (Phila Pa 1976)* 1995;20:2147-50.

7. Ijiri K, Hida K, Yano S, et al. Traumatic spinal-cord herniation associated with pseudomeningocele after lower-thoracic nerve-root avulsion. *Spinal Cord* 2009;47:829-31.
8. Kumar R, Taha J, Greiner AL. Herniation of the spinal cord. Case report. *J Neurosurg* 1995;82:131-6.
9. Lee ST, Lui TN, Jeng CM. Spinal cord herniation after stabbing injury. *Br J Neurosurg* 1997;11:84-6.
10. Sachdev VP, Huang YP, Shah CP, et al. Posttraumatic pseudomeningomyelocele (enlarging fracture?) in a vertebral body. Case report. *J Neurosurg* 1981;54:545-9.
11. Tanaka M, Ikuma H, Nakanishi K, et al. Spinal cord herniation into pseudomeningocele after traumatic nerve root avulsion: case report and review of the literature. *Eur Spine J* 2008;17 Suppl 2:S263-6.
12. Watters MR, Stears JC, Osborn AG, et al. Transdural spinal cord herniation: imaging and clinical spectra. *AJNR Am J Neuroradiol* 1998;19:1337-44.
13. Wortzman G, Tasker RR, Rewcastle NB, et al. Spontaneous incarcerated herniation of the spinal cord into a vertebral body: a unique cause of paraplegia. Case report. *J Neurosurg* 1974;41:631-5.
14. Yokota H, Yokoyama K, Noguchi H, et al. Spinal cord herniation into associated pseudomeningocele after brachial plexus avulsion injury: case report. *Neurosurgery* 2007;60:E205; discussion E.

Table. Bibliometric Summary of Spinal Cord Herniation in year of publication order

Journal & Year	Author(s)	Type of article	Case
1975 American Journal of Neuroradiology	Wotzman G et al	Case Report	Spontaneous herniation
1978 Journal of neurosurgery	Burres KP et al	Case Report	Post-operative SCH after trauma surgery
1981 Journal of Neurosurgery	Sachdev VP et al	Case Report	Post-traumatic SCH
1981 Journal of Neurosurgery	Masuzawa H et al	Case Report	Spontaneous (congenital) SCH
1986 Neurological Surgery	Mizuno J et al	Case Report	Post-operative SCH
1987 American Journal of Neuroradiology	Dunn V et al	Case Report	Post-traumatic SCH
1991 Neurosurgery	Tronnier VM et al	Case Report	Spontaneous SCH
1993 Spine Journal	Nakazawa H et al	Case Report	Spontanoeus SCH
1995 Spine Journal	Hosono N et al	Case Report	Post-operative SCH after tumor resection
1995 Journal of Neurosurgery	Kumar R et al	Case Report	Spontaneous SCH
1995 Journal Neurosurgery	Borges LF et al	Case Report	Spontaneous SCH
1996 Neuroradiology	Urbach H et al	Case Report	Post-traumatic SCH
1996 Neuroradiology	Miura T et al	Case Report	Spontaneous SCH
1996 Neuroradiology	Hausmann ON et al	Case Report	Spontaneous SCH
1997 British Journal of Neurosurgery	Lee ST et al	Case Report	Post-traumatic SCH
1997 European Radiology	Uchino A et al	Case Report (2 Cases)	Spontaneous SCH
1998 Journal of Neuroradiology	Watters G et al	Case Series (5 Cases)	Post-traumatic SCH (3 cases) and Spontaneous SCH (2 cases)
2003 Neurosurgery	DaSilva VR et al	Case Report	Post-traumatic SCH
2007	Yokota H et al	Case Report	Post-traumatic

Neurosurgery			SCH
2008 European Journal of Spine Surgery	Tanaka K et al	Case Report	Post-traumatic SCH
2009 Surgical Neurology	Belen D et al	Case Report	Post-operative SCH following foramen magnum decompression
2009 Spinal Cord	Ijiri K et al	Case Report	Post-Traumatic SCH
2011 Spine Journal	Aydin Al et al	Case Report	Spontaneous SCH

SCH: Spinal Cord Herniation

Appendix 1: MRI Figures

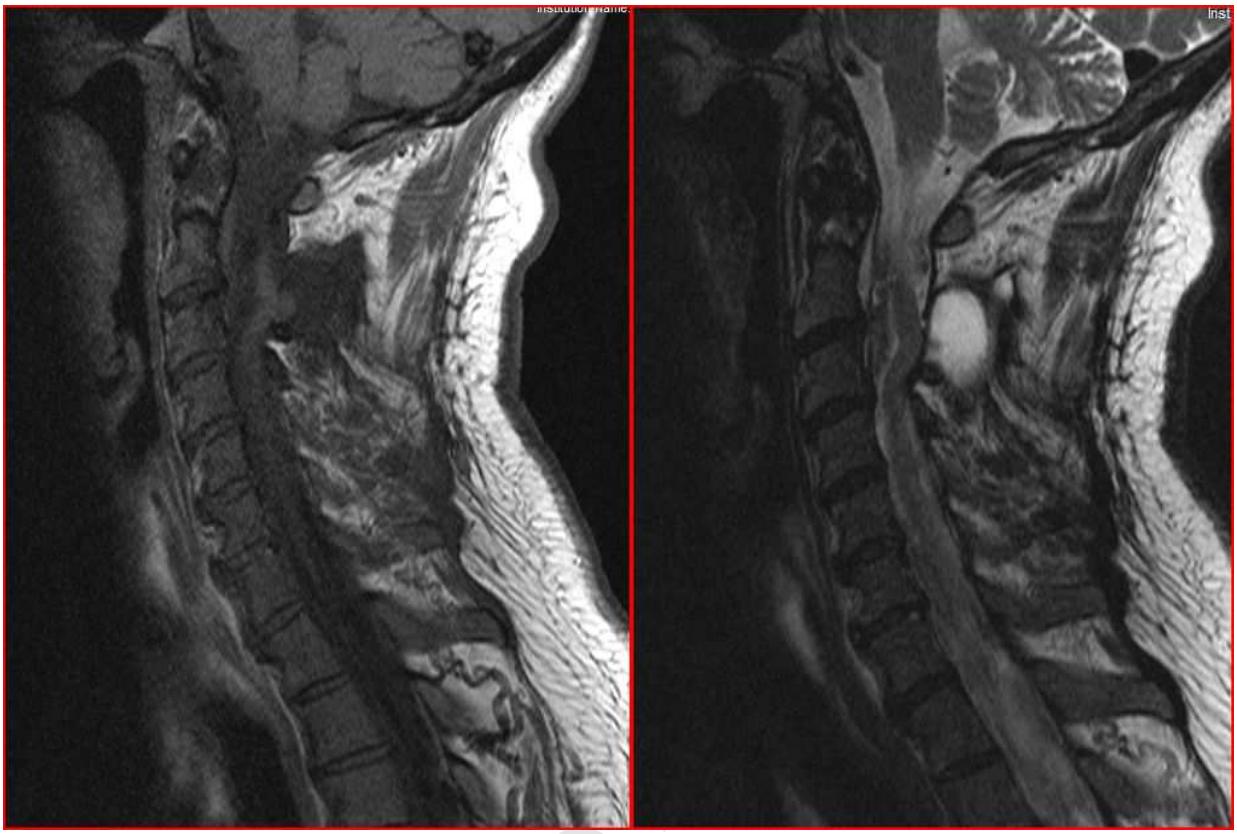

Figure 1. Sagittal View MRI T2 (left) and T1 (right) of the cervical spine.

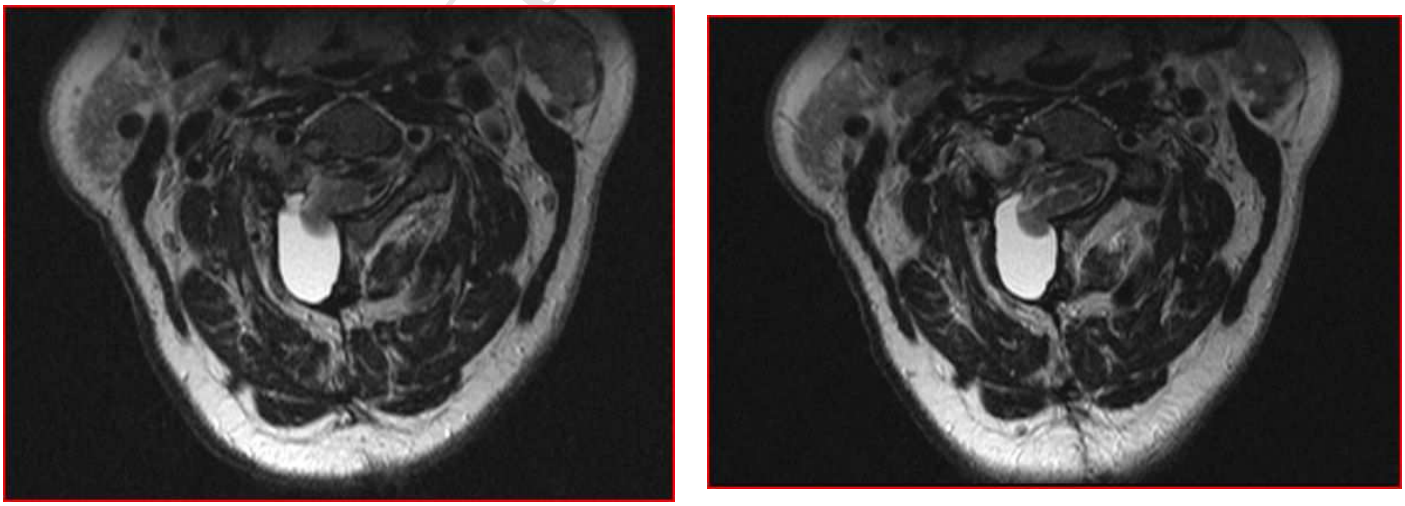

Figure 2. Axial T2 MRI views of cervical spine across C2-3 level

Appendix 2: Intra-Operative Images

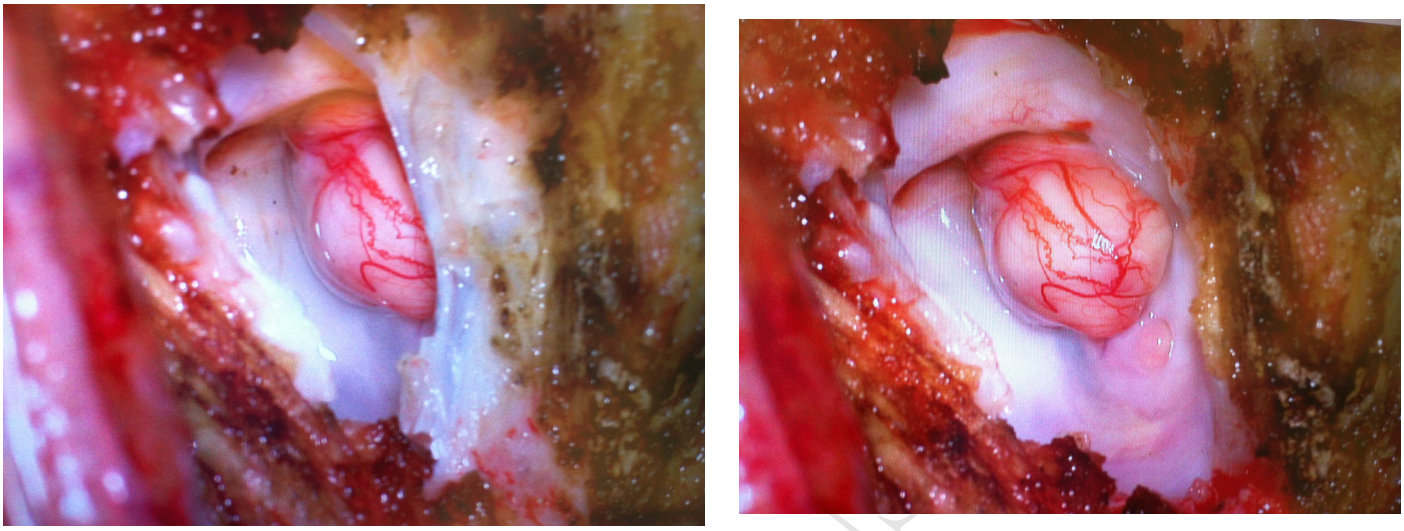

Figure 1. Pseudomeningeocele opened, showing spinal cord herniating through the defect.

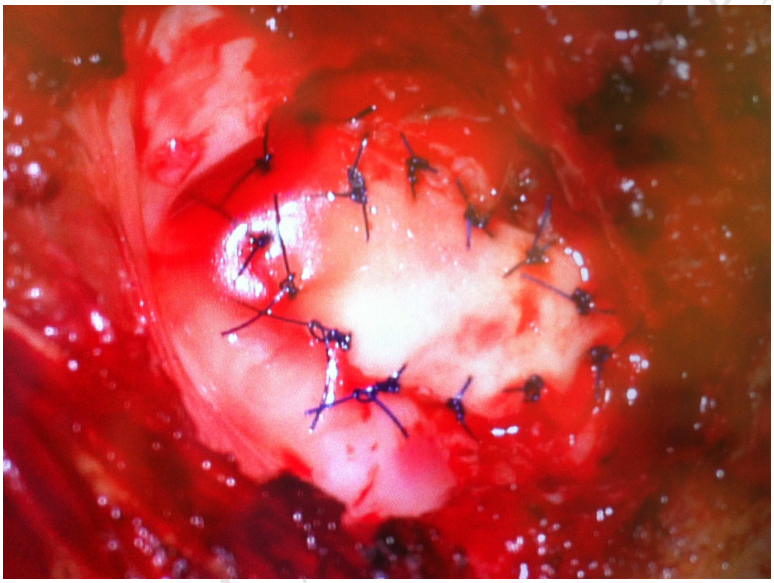

Figure 2. Duraplasty after reduction of the spinal cord. The dural substitute (Neuro-patch™) stitched in place using interrupted 5-0 prolene sutures.