Remote cerebellar hemorrhage following thoracic spinal surgery of an intradural extramedullary tumor: a case report

Masazumi Suzuki¹, Takashi Kobayashi¹, Naohisa Miyakoshi², Eiji Abe¹, Toshiki Abe¹ and Yoichi Shimada²

Abstract

Introduction: Remote cerebellar hemorrhage is a rare complication of spinal surgery. Although loss of cerebrospinal fluid seems to play an important role in the pathogenesis of this complication, the detailed mechanism of remote cerebellar hemorrhage after spinal surgery remains unclear. We report the case of a patient with remote cerebellar hemorrhage following thoracic spinal surgery of an intradural extramedullary tumor and discuss this entity with reference to the literature.

Case presentation: A 57-year-old Japanese woman presented to our hospital with back pain, dysuria, and numbness of both legs. A neurological examination was performed, and imaging was performed with ordinary radiography, magnetic resonance imaging, and computed tomography. Her magnetic resonance imaging scan showed an intradural extramedullary tumor at the T3 level. A tumor resection and T1-T5 pedicle screw fixation were performed. Twelve hours after spinal surgery, she complained of unexpected dizziness, nausea, and vomiting. A total of 850mL of serosanguineous fluid had been drained at that time, and drainage was stopped. An urgent brain computed tomography scan showed a cerebellar hemorrhage. She was treated conservatively, and was able to leave hospital six weeks after the initial operation, without any neurological deficits except for slight ataxia.

Conclusions: Remote cerebellar hemorrhage has to be suspected when unexpected neurological signs occur after spinal surgery. If an excessive amount of cerebrospinal fluid drains from the drainage tube after spinal surgery, drainage should be stopped.

Keywords: Postoperative complication, Intradural extramedullary tumor, Remote cerebellar hemorrhage, Spinal surgery, Thoracic spine, Neurological signs

Introduction

Remote cerebellar hemorrhage (RCH) following spinal surgery is a rare complication [1-25]. Although loss of cerebrospinal fluid (CSF) plays an important role in the pathogenesis of this complication [1-25], the detailed mechanism of RCH after spinal surgery remains unclear. Here, we present a case of RCH after thoracic spinal surgery for an intradural extramedullary tumor, along with a review of previously reported cases and a discussion of the mechanism of RCH.

Case presentation

A 57-year-old Japanese woman, with no past medical history, presented to our institution with a one-year history of abdominal pain, a two-month history of back pain, numbness of both her legs, and a one-month history of dysuria. She initially reported abdominal pain and underwent extensive gastroenterological evaluation at another hospital, including an esophagogastroduodenoscopy, which was unremarkable.

Her physical examination revealed no motor weakness and normal tendon reflexes. She felt hypoesthesia below the umbilicus. Magnetic resonance imaging (MRI) results demonstrated a large intradural extramedullary mass at the T3 level that was compressing her spinal cord from the ventral side (Figure 1).

* Correspondence: masazumi.suzuki0512@gmail.com
¹Department of Orthopedic Surgery, Akita Kousei Medical Center, 1-1-1 Iijima, Nishifukuro, Akita 011-0948, Japan
Full list of author information is available at the end of the article

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The intradural extramedullary tumor was resected through a laminectomy of T2-T4 and a facetectomy of T2-T3 and T3-T4 in the prone position under transcra-
nial motor-evoked potential (MEP) monitoring. As the
 tumor was completely covered by her spinal cord, it was
surgically removed by rotation of the spinal cord using
tenting of the dentate ligament. After tumor resection,
the dura that adhered to the tumor was cauterized. A
watertight repair of the dura was performed, using fibrin
 glue to avoid CSF leakage. A T1-T5 pedicle screw fix-
ation was performed (Figure 2). Abnormal MEP signals
were observed on her left leg during and after the tumor
resection. A subfascial drain was put in place, with
negative pressure. After she woke the motor power was
weakened to grade three to four in her left knee and
ankle. The total operating time was 4 hours 39 minutes,
and the amount of bleeding was 108g. The histological
diagnosis of the tumor was a meningioma.

Twelve hours after surgery, she developed nausea and
confusion, and her clinical status deteriorated with loss
of consciousness (Glasgow Coma Scale score of seven).
A total of 850mL serosanguineous fluid had been
drained at that time, and drainage was stopped. An
emergency brain computed tomography (CT) scan dem-
onstrated an acute cerebellar hemorrhage in the superior
folia of the cerebellar hemispheres (Figure 3). An MRI

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**Figure 1** Preoperative magnetic resonance images. Sagittal T1-weighted (a) and T2-weighted (b) magnetic resonance images of the thoracic
spine, demonstrating an intradural extramedullary mass anterior to the spinal cord at the T3 level. The mass was iso-intense on T1-weighted
imaging and T2-weighted imaging. Axial T1-weighted (c) and T2-weighted (d) magnetic resonance images show that the tumor seemed to be
completely covered by the spinal cord.
scan demonstrated a herniation of the cerebellar tonsils (Figure 4a, b). She was treated conservatively with anti-edema and antihypertensive drugs, and her clinical status improved gradually. After removal of the drain, there was no CSF leakage. The results of her follow-up CT scan performed one week later showed that her hematoma and brain edema were decreased. Twelve days later, the results of her follow-up MRI scan showed ascent of the cerebellum to the normal position (Figure 4c, d). At six weeks after surgery, she had slight ataxia and was discharged with a cane. At her one-year follow-up assessment, she had a normal neurological examination except for hypoesthesia of the right leg, and there was no CSF collection visible on her MRI scan.

Discussion
Our case report has two characteristics. First, this case of thoracic meningioma that was located anterior to the spinal cord presented with a one-year history of undiagnosed abdominal pain. Lyons et al. [26] reported a similar case presenting as chronic abdominal pain. Second, although the tumor (which completely covered the spinal cord) was totally removed with a posterior surgical approach, our patient had some left lower extremity weakness postoperatively that improved gradually. A total resection of intradural extramedullary tumors located anterior to the spinal cord can be performed using an isolated posterior approach, with rotation of the spinal cord and tenting of the dentate ligament [27,28].
RCHs are rare and dramatic complications can follow spinal surgery. Prevention is important, because RCHs sometimes follow a fatal course. Sporadic cases have been published since the first description by Chadduck [4]. At the time of writing, 32 cases of RCH after spinal surgery have been reported in the English-language literature (Table 1). Including the present case, the 33 cases consisted of 23 women and 10 men, with an age range of 36 to 85 years (mean: 60.9 years). Initial surgery was performed at the lumbar spine in 21 cases, thoracic spine in six, cervical spine in five, and thoracolumbar spine in one. A dural tear during surgery was present in 26 cases, but was not noticed in seven cases. The neurological symptoms were detected between 0 and 192 hours (mean: 45.7 hours) after surgery. A total of 16 RCHs were resolved with conservative treatment, but three patients died or developed serious paresis [1-3,5,8,10,11,15,19,21-23]. However, in severe cases, emergency surgical intervention with ventricular drainage or posterior fossa craniotomy was needed. Cranial surgery was performed in 14 patients, nine of whom improved, and five died or had serious paresis after surgery [4,6,7,9,11-14,17,18,20,24,25].

RCH occurs in patients with a dural tear and CSF leakage, whether occult or not. It is thus believed that perioperative and/or postoperative CSF losses, leading to cranial hypotension, represent the main contributing factor in RCH [1,8,12]. The exact pathophysiology of RCH is still controversial. It is suggested that transient stretching and occlusion of superior cerebellar veins, resulting from downward cerebellar displacement under conditions of intracranial hypotension, may lead to cerebellar hemorrhagic infarction [8,20]. It is also suggested that cerebellar sag can directly cause tearing and
bleeding of superior cerebellar veins [8]. Pallud et al. [20] hypothesized that RCH results primarily from superior cerebellar venous stretching and tearing, and that cerebellar infarction and swelling occur secondarily. The loss of CSF should be restricted and controlled, because intracranial hypotension may be the initial cause of RCH. Closed wound suction drainage is recommended for spinal surgery, because a postoperative drain theoretically reduces the risk of infection and/or wound breakdown by decompressing the site of postoperative hematoma formation. However, if too much serosanguineous fluid drains postoperatively, stopping drainage or removing the drainage tube should be considered to prevent intracranial hypotension. Removal of the drain restores the normal CSF flow dynamics, allowing the cerebellum to resume its normal position [1]. Friedman et al. [8] described a 56-year-old woman with postoperative RCH whose headache resolved when suction drainage of the wound was discontinued. Thus, considering our case and the published literature, we suggest stopping drainage

| Table 1 Clinical parameters and outcomes in previous reports of remote cerebellar hemorrhage |
|------------------------------|----------------|----------------|-----------|----------------|----------------|----------------|
| Author (year) | Surgery | Location | Age, sex | Onset | Dural tear | Treatment | Results |
| Chadduck (1981) [4] | laminectomy | CS | 59, M | 2 days | present | surgery | improved |
| Mikawa et al. (1994) [17] | C1/2 fusion, durotomy | CS | 75, M | 1 day | present | surgery | died |
| Andrews and Koci (1995) [1] | scoliosis correction | LS | 36, M | 36 hours | unknown | conservative | quadripareisis |
| Friedman et al. (2002) [8] | posterior thoracic disc herniation removal | PSF | 43, M | 12 hours | present | conservative | improved |
| Thomas et al. (2002) [22] | IETR | TLS | 38, F | 5 days | present | conservative | improved |
| Farag et al. (2005) [7] | PSF | LS | 43, F | 36 hours | present | surgery | improved |
| Karaeminogullari et al. (2005) [12] | PSF | LS | 73, F | 2 days | present | surgery | improved |
| Nakazawa et al. (2005) [19] | IETR | CS | 74, F | perioperative | present | conservative | improved |
| Konya et al. (2006) [15] | PSF | LS | 48, F | 12 hours | present | conservative | improved |
| Calisandeller et al. (2007) [2] | PSF | LS | 67, F | 8 days | present | conservative | improved |
| Comips et al. (2007) [5] | thoracoscopic microdiscectomy | TS | 48, F | 3 days | unknown | conservative | died |
| Hashidate et al. (2008) [9] | vertebral tumor resection | TS | 85, F | 40 hours | unknown | surgery | improved |
| Cevik et al. (2009) [3] | laminectomy | LS | 79, F | 3 days | unknown | conservative | improved |
| Enel et al. (2009) [6] | PSF | LS | 51, F | 30 hours | present | surgery | died |
| Khong and Jerry Day (2009) [14] | PSF | LS | 70, F | 36 hours | present | surgery | improved |
| Morofuji et al. (2009) [18] | laminectomy | TS | 51, M | 18 hours | present | surgery | improved |
| Pallud et al. (2009) [20] | laminectomy | LS | 73, F | 3 days | present | surgery | improved |
| Ullivieri et al. (2009) [23] | microdiscectomy | LS | 53, M | 2 hours | present | conservative | improved |
| Yang et al. (2011) [24] | PSF | LS | 56, F | 21 hours | unknown | surgery | ataxia and aphasia |
| Hempelmann and Mater (2012) [10] | IETR | TS | 61, F | 7 days | present | conservative | improved |
| | PSF | LS | 69, F | 2 days | present | conservative | improved |
| | PSF | LS | 62, F | 1 day | present | conservative | improved |
| Khalatbari et al. (2012) [13] | discectomy | LS | 53, M | 8 hours | present | surgery | improved |
| | laminectomy | LS | 75, M | perioperative | present | surgery | died |
| Lee et al. (2012) [16] | PSF | LS | 63, F | 6 hours | present | conservative | improved |
| Takahashi et al. (2012) [21] | laminoplasty | CS | 69, F | 15 hours | unknown | conservative | improved |
| Kaloostian et al. (2013) [11] | PSF | CS | 45, M | perioperative | present | conservative | improved |
| | PSF | LS | 64, F | 2 days | present | conservative | brain dead |
| | PSF | LS | 81, F | 1 day | present | surgery | died |
| Yoo et al. (2013) [25] | intradural disc surgery | LS | 66, M | 2 days | present | surgery | improved |

CS, Cervical spine; F, Female; IETR, Intradural extramedullary tumor resection; LS, Lumbar spine; M, Male; PSF, Posterior spinal fusion; TS, Thoracic spine.
when RCH is suspected based on the patient’s complaints, including nausea and headache, and/or if an excessive amount of serosanguineous fluid has been drained postoperatively. This complication can be prevented by observing the amount of drainage fluid. If an excessive amount of fluid is drained, drainage should be stopped or converted to a gravity drain instead of a suction drain.

Conclusions
RCH is a rare postoperative complication of spinal surgery. RCH must be suspected when intracranial symptoms or unexpected neurological signs occur after spinal surgery. If an excessive amount of serosanguineous fluid is found coming from the drainage tube postoperatively, drainage should be stopped.

Consent
Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Abbreviations
CT: Computed tomography; CSF: Cerebrospinal fluid; CT: Computed tomography; MEP: Motor-evoked potential; MRI: Magnetic resonance imaging; RCH: Remote cerebellar hemorrhage.

Competing interests
The authors declare that they have no competing interests.

Authors’ contributions
Surgery was performed by TK. MS, TK, and NM were the major contributors in writing the manuscript. TA, TA, and YS supervised the whole work. All authors read and approved the final manuscript.

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Author details
1Department of Orthopedic Surgery, Akita Kousei Medical Center, 1-1-1 Iijima, Nishiharu, Akita 011-0948, Japan. 2Department of Orthopedic Surgery, Akita University Graduate School of Medicine, 1-1-1 Hondo, Akita 010-8543, Japan.

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