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CASE REPORT

Intractable postural hypotension resulting from cervical pseudomeningocele after a posterior cervical spinal surgery: a case report

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INTRODUCTION: Postural hypotension (PH) is common in patients with spinal cord injury (SCI), especially those with tetraplegia. To effectively treat PH, identifying and eliminating treatable predisposing factors of PH are prerequisites before applying any interventions.

CASE PRESENTATION: We report a patient with post-acute cervical SCI who suffered from intractable PH resulting from pseudomeningocele causing unfavorable rehabilitation outcomes. A previously healthy 34-year-old man with complete C6 SCI resulting from C6-C7 fracture dislocation developed PH in the first week of the rehabilitation program. No specific predisposing factors including anemia, hyponatremia, and dehydration were identified. Non-pharmacological interventions and pharmacological treatment were administered to the patient without satisfactory result, causing a delay in rehabilitation progression. In the fourth week of rehabilitation program, a mass at the surgical site was detected. A cervical MRI revealed a large fluid collection at the posterior aspect of cervical spines with a size of $7.9 \times 6.8 \times 5.0$ cm. A diagnosis of pseudomeningocele was made and surgical site debridement with closing dura by grafting was immediately conducted. One day after surgery, PH disappeared, and the patient could progress in his rehabilitation program and achieve his short-term goal within three weeks.

CONCLUSION: Pseudomeningocele could be one of the precipitating factors of PH in patients with tetraplegia. Healthcare providers should consider investigating pseudomeningocele in patients who have intractable and unexplainable PH.

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INTRODUCTION

Postural hypotension (PH), or orthostatic hypotension, is one of the most common problems in acute and early post-acute rehabilitation phases of patients with spinal cord injury (SCI), especially those with tetraplegia [1]. PH could delay starting the rehabilitation program, prolong immobilization, and postpone the independency of the patient. Therefore, prevention and treatment of PH in patients with SCI in acute and early post-acute rehabilitation phases are important [2]. Despite using many pharmacological and non-pharmacological interventions [3], however, results of the treatment might not be fully satisfied if the predisposing cause of PH could not be priorly investigated and corrected [4]. Here, we describe a patient with acute SCI resulting in complete tetraplegia, who remained to have severe PH despite full administration of both pharmacological and non-pharmacological interventions, resulting in a delayed progression of rehabilitation programs. However, after the cause of PH had eventually been identified and managed, PH rapidly disappeared, and the patient could promptly attend the post-acute rehabilitation programs. Although it has not been described in previous medical literature, pseudomeningocele could be a cause of PH. In this case report, we aim to describe a patient with PH that was possibly predisposed by cervical pseudomeningocele after spinal surgery, as well as discuss the proposed mechanisms of pseudomeningocele on developing PH and their clinically related points.

CASE PRESENTATION

A previously healthy 34-year-old man was in a motorcycle accident and became weak in all four extremities and loss of sensation. Physical examination at an emergency department demonstrated that muscle strength of biceps and wrist extensors was grade 5 bilaterally whereas no voluntary movement of all key muscles below the bilateral C6 myotomes was detected. There was a complete sensory loss below the C6 dermatome on both sides. Deep tendon reflexes at both lower extremities, as well as bulbocavernosus and anal reflexes, were all absent. The patient was diagnosed with C6 American Spinal Injury Association (ASIA) Impairment Scale (AIS) A SCI.

A radiographic investigation by computed tomography (CT) revealed a right C6-C7 unilateral facet lock with comminuted fracture of left C6-C7 pars interarticularis causing 50% anterolisthesis of C6 over C7 with fracture of anterosuperior C7 vertebral body resulting in spinal cord compression. Emergent magnetic resonance imaging (MRI) of cervical spines showed severe spinal

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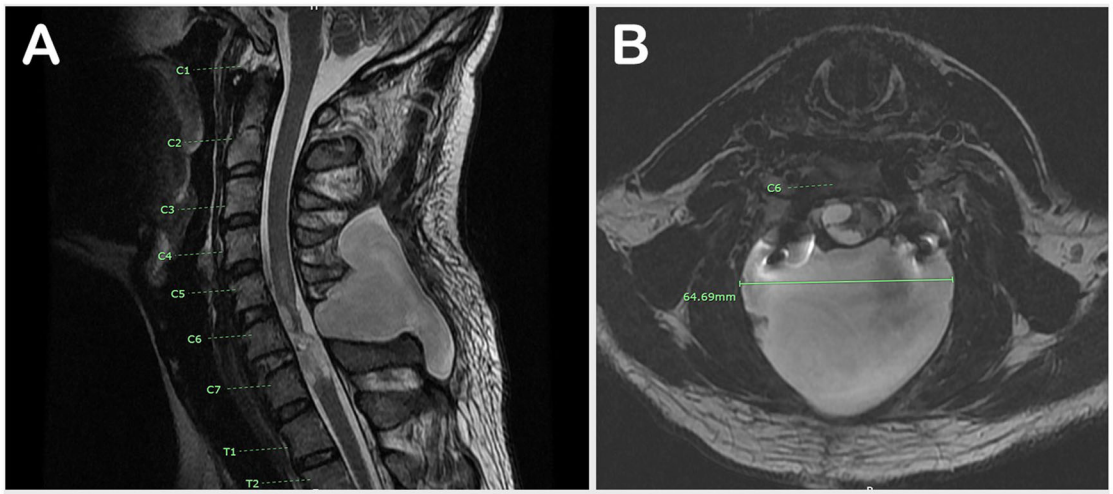

Fig. 1 A cervical MRI reveals a large fluid collection at the posterior aspect of cervical spines in a man with tetraplegia and intractable postural hypotension. A T2, sagittal section; **B** T2, axial section.

cord compression at C6-C7 level with spinal cord edema at C5-C7 level and hemorrhagic spinal cord contusion at C5-C6 level. The patient underwent an open reduction C6-C7 facet joint with decompressive laminectomy C6 with pedicular screw fixation at C6-C7 two days after the onset. However, he was transferred to a post-acute rehabilitation ward three weeks after SCI since his post-operative condition was complicated by fever and urinary tract infection which necessitated a complete course of intravenous antibiotics before the transfer. No rehabilitation program except range of motion exercise of all extremities was applied in the acute SCI phase.

At the rehabilitation facility, the results of the neurological examination were not changed. The patient still had the C6 AIS A SCI. A six-week intensive rehabilitation program for optimizing C6 level functions was anticipated. However, the program could not be advanced due to symptomatic PH. On the first day of rehabilitation, the patient's blood pressure was 120/80 mmHg in the supine position but dropped to 80/40 mmHg in 30 degrees head-up position. Despite the appliance of all available non-pharmacological interventions, including abdominal binding, elastic bandaging at both lower extremities, and titrating head up as tolerated for the entire first two weeks, he still had a severe drop in blood pressure of 85/45 mmHg at 30 degrees head up from 120/80 mmHg in supine position simultaneously with having symptoms of dizziness and blurred vision. On the third week of rehabilitation, a sodium chloride tablet three times daily was prescribed. Laboratory investigation demonstrated no anemia, no hyponatremia (140 mmol/l), and no abnormalities of thyroid function test nor liver enzymes. Chest radiograph and electrocardiogram were within normal limits.

Since PH persisted, a pharmacological intervention using Fludrocortisone of 0.1 mg per day was initiated in the fourth week of post-acute rehabilitation. Despite adding pharmacological treatment, symptomatic PH was not alleviated. Apart from dizziness and blurry vision, the patient experienced a subtle loss of consciousness or blacking out a couple of times when changed to head up at 45 degrees from a supine position where blood pressure was 110/50 mmHg. However, blood pressure at the time of blacking out was not recorded since he was emergently positioned supine after reporting the symptoms.

On the other hand, the patient continuously reported posterior neck pain around the surgical scar after the time of surgery. The pain was symptomatically treated with NSAIDs and paracetamol. It was subsequently observed that the surgical scar was somewhat soft and loose. Although it is not mandatory in our guideline, a post-operative MRI was requested and revealed a large fluid collection at the posterior aspect of cervical spines; deep back muscles, and spinous process, size $7.9 \times 6.8 \times 5.0$ cm (Fig. 1). The previously attended orthopedic surgeon was consulted, resulting in an immediate reoperation in the 8th week after surgery or the 5th week after transfer to the rehabilitation ward. Pseudomeningocele caused by a non-approximated dura tear was found intraoperatively. Surgical site debridement with closing dura by synthetic dura graft, fat graft, and tissue fibrin was carried out.

During the surgery, all pharmacological measures were discontinued. On the first day after surgery, he could progress to sitting upright without symptoms or PH. He was then transferred back to the rehabilitation facility and continued the post-acute rehabilitation program. With the application of an abdominal binder and elastic stocking, he could progress to sitting at 70 degrees upright with 50 degrees of leg drop without PH in two weeks. By the third week post-surgery, the patient could sit upright at 90 degrees for two hours with a blood pressure of 100/60 mmHg without symptoms. He was discharged home to continue other parts of the rehabilitation program in his domicile as he wished.

DISCUSSION

Despite not being previously described, we assume that PH in this patient was, at least partly, resulted from pseudomeningocele since the PH suddenly disappeared after the definite treatment of pseudomeningocele, which is compatible with a counterfactual explanation [5]. The main pitfall in this case is a delayed diagnosis and treatment of pseudomeningocele due to misleading clinical features of PH, a condition that has not been described as a sign of pseudomeningocele. However, we should have paid more attention to physical examination, including inspecting and palpating, at the area adjacent to the surgical site, especially in patients who have ongoing or new onset pain at surgical site or radicular pain, as well as unexplainable PH. This pitfall might be due to the fact that the patient was transferred to the rehabilitation ward after suture removal, making the attending rehabilitation physicians, who have not been familiar with surgical wound care, neglect observing the postsurgical complications resulting in the delayed diagnosis of pseudomeningocele.

PH is common in people with SCI with a prevalence of 73.6%, and 58.9% of them have symptoms [1]. It is a debilitating condition impacting morbidity and rehabilitation outcomes [3]. Previously proposed mechanisms of postural hypotension

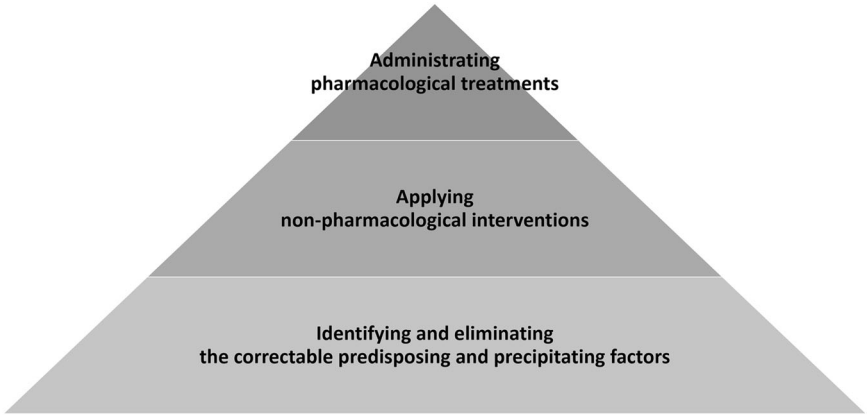

Fig. 2 The treatment paradigm of postural hypotension starting with addressing the factors, non-pharmacological, then pharmacological interventions. First, clinicians should identify and try to eliminate the correctable predisposing and precipitating factors. If the postural hypotension still persists, non-pharmacological interventions should applied. If the postural hypotension still presents, pharmacological interventions should be additionally administrated.

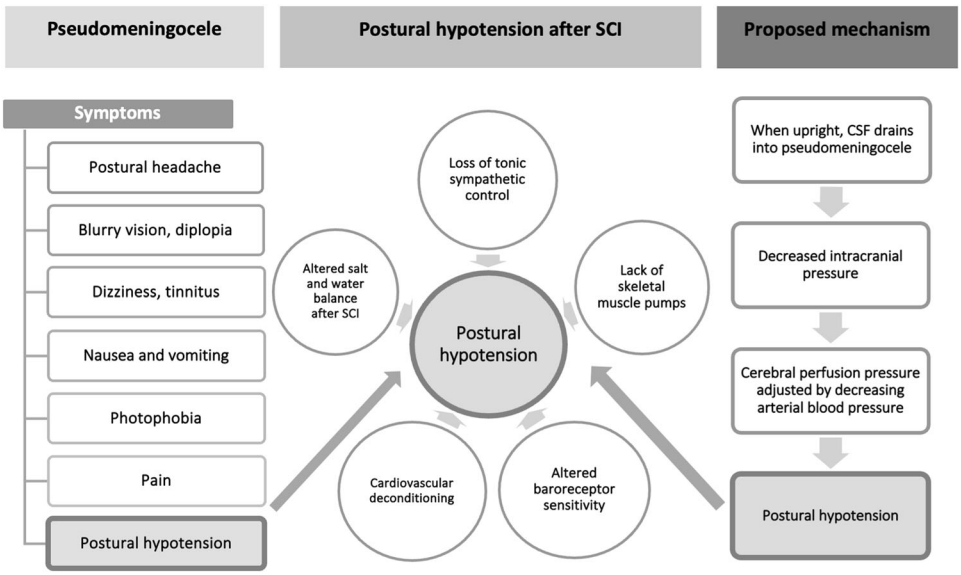

Fig. 3 The proposed mechanisms of pseudomeningocele-induced postural hypotension in this patient. SCI spinal cord injury, CSF cerebrospinal fluid.

included loss of tonic sympathetic control [6], morphologic changes in sympathetic neurons [7], altered salt and water balance from abnormal renin-angiotensin-aldosterone activity [8], low plasma catecholamine levels [9], cardiovascular deconditioning [10], motor deficits leading to lack of skeletal muscle pumping activity [11], and altered baroreceptor sensitivity [12]. However, the mostly described pathophysiologic mechanism of PH is multifactorial [2] and related to the body function impairments after SCI, which are more common and more severe in tetraplegia when compared with paraplegia [2]. Although all previously mentioned mechanisms are injury-related and non-modifiable after SCI, PH is also induced by some non-injury-related, correctable predisposing factors such as dehydration, anemia, and hyponatremia [3].

To eliminate PH, healthcare providers should first identify and remove the correctable predisposing factors such as dehydration, anemia, and hyponatremia, as well as avoid precipitating factors such as unprepared, sudden head-up postural change, straining, high environmental temperature, large meals, alcohol, and drugs with vasodepressor properties [13]. PH treatments would not be satisfied unless these factors are comprehensively identified and eliminated. If these factors are absent or uncorrectable, non-pharmacological interventions, including progressive head-up tilting, small frequent meals, high salt intake, water ingestion, elastic stockings, abdominal binders, and functional electrical stimulation (FES) at lower extremity muscles should be initially applied [3]. If non-pharmacological interventions are not effective, pharmacological treatments, including midodrine, fludrocortisone, ergotamine, ephedrine, L-threo-3,4-dihydroxyphenylserine (L-DOPS, droxidopa), and nitro-L-arginine methyl ester (L-NAME) [3], should be administrated according to the patient's conditions and the healthcare provider's clinical judgment. Figure 2 summarizes the treatment paradigm of PH.

Despite consecutively using all strategies, including predisposing and precipitating factors removal, non-pharmacological interventions, and pharmacological treatment, this patient still had intractable PH. Incidentally, we found another complication, which was pseudomeningocele. Pseudomeningocele from incidental durotomy has been found to occur in 1% to 17% of spinal surgery cases [14]. It could be the culprit of numerous symptoms from palpable mass at the area adjacent to the surgical site, postural headache, blurry vision, dizziness, nausea

and vomiting, diplopia, photophobia, tinnitus, back pain, limb pain and radicular symptoms, and weakness [14]. Intraoperative tracheal compression and expiratory obstruction have been reported as a result of acutely severe obstruction from a large pseudomeningocele [15]. Noteworthy, pseudomeningoceles more than 8 cm in size are described as giant pseudomeningoceles and those more than 5 cm as large pseudomeningoceles [16]. MRI is an investigation of choice since it could differentiate pseudomeningocele from the more common post-operative hematoma [17]. Despite being possibly treated by conservative treatment, especially in small and asymptomatic cases [18], large or very large size, located at the cervical level, or neurologically symptomatic pseudomeningocele should be surgically managed by dura repair in addition to the standard decompression of the extradural mass [14].

To all our knowledge, PH has not been reported as one of the symptoms of pseudomeningocele. The proposed mechanism of pseudomeningocele-induced PH in this patient was related to cerebral perfusion pressure hemostasis. When the patient was in a lying down position, despite the presence of pseudomeningocele, cerebrospinal fluid (CSF) would not be significantly drained into the potential space at the tissues adjacent to the pseudomeningocele. However, when the patient was in the upright position, CSF would be largely drained into the pseudomeningocele by the gravity force, resulting in a decrease in intracranial pressure (ICP) or intracranial hypotension, similarly found in the CSF leakage from other causes [19]. Intracranial hypotension potentially increases cerebral perfusion pressure (CPP) if mean arterial pressure (MAP) is not changed since CCP is equal to MAP minus ICP. To maintain the CCP at its homeostatic level, the body has to lower the MAP using the mechanisms that are remained after SCI such as lowering the heart rate by increasing the vagal tone, bringing the patient a significant decrease in blood pressure. Due to the loss of sympathetic tone and other previously described factors which potentially prevent appropriate reactions to the postural challenge after SCI, the patient, therefore, faced an intractable PH. Figure 3 summarizes the proposed mechanisms of pseudomeningocele-induced PH in this patient.

According to our hypothesis that pseudomeningocele might contribute to PH development, a question of whether it is necessary to investigate for pseudomeningocele, i.e., request for MRI, in all patients with SCI who develop PH, and if not, when should we investigate it. To all our knowledge, there is no standard consensus for prescribing post-spinal surgery imaging, especially MRI, in real-life clinical settings [17, 20] Routine post-operative MRI were prescribed only in research settings [21]. Corona-Cedillo, et al. (2021) comprehensively reviewed the imaging assessment of the postoperative spine issue and recommended that routine imaging should be plain x-ray or computerized topography (CT) to evaluate the alignment of the spines and instrument [17]. MRI should be prescribed only in patients with abnormal clinical features indicating complications, such as surgical site swelling or mass, deformity, or unexplainable pain [17]. From this report, we would also include unexplainable PH as one of the indications. On the other hand, patients with SCI who have intractable PH should be physically examined for excluding a mass at the surgical site, in addition to the weekly routine examination. If the examination reveals any suspected signs or the patient still has intractable and unexplainable PH, especially in patients with tetraplegia, a spinal MRI should be considered to rule out pseudomeningocele.

CONCLUSIONS

Pseudomeningocele could be one of the precipitating factors of PH in patients with SCI, especially those with tetraplegia. The mechanism of pseudomeningocele-induced PH in this patient might be related to cerebral perfusion pressure hemostasis. Healthcare providers should consider investigating pseudomeningocele in patients who have intractable and unexplainable PH. A further study aiming to evaluate the prevalence, as well as to explore the mechanism of pseudomeningocele-induced PH is needed to elucidate this uncommon cause of the common condition.

DATA AVAILABILITY

The datasets generated and/or analyzed during the current study are available from the corresponding author on reasonable request.

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AUTHOR CONTRIBUTIONS

BR and SP designed the research question, collected and analyzed the data, and wrote and revised the final version of the manuscripts.

COMPETING INTERESTS

The authors declare no competing interests.

ETHICS

We certify that in our institute, a single, deidentification case report is waived from ethical approval.

ADDITIONAL INFORMATION

Supplementary information The online version contains supplementary material available at https://doi.org/10.1038/s41394-023-00572-8.

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