《研究ノート》

Autonomic dysreflexia triggered by hip joint position in patients with cervical cord injury: report of two cases

Motoyuki ABE[1)], Yuka NONOMURA[2)]

[1)]Faculty of Rehabilitation and Care, Seijoh University
[2)]Department of Rehabilitation, Gifu Chuo Hospital

Abstract

Two unusual cases of autonomic dysreflexia (AD) considered to have been triggered by hip joint position in males with complete tetraplegia due to C6 and C8 cervical cord injury are reported. In both cases, frequent repeated episodes of abnormal hypertension, bradycardia, and severe headache began to occur several months after injury, clearly constituting clinical AD. The cause was entirely unknown, since none of the common trigger factors, such as urinary distension or fecal impaction, was present. Subsequent clinical observations led to the assumption that hip joint position might be the cause, and blood pressure and heart rate in different positions were measured actually. In both cases, the trigger factor was the position of the hip joint. In the first case, the trigger was the neutral and extended positions; in the second, the trigger was the abducted position. As a result of this finding, it was possible to prevent AD by adjusting the hip joint position. The mechanism of this rare AD can be inferred to be stretching of the proximal leg muscles resulting from the hip position as a noxious stimulus via the peripheral nerves. Such hip positions are common during hospitalization and rehabilitation. When AD occurs frequently without any other conceivable reason, the possibility that it may be triggered by hip joint position should be considered.

Introduction

Autonomic dysreflexia (AD) is a clinical emergency occurring in patients with high-level spinal cord injury (SCI) above the sixth thoracic level (T6) [1,2)]. AD occurs when there is no central regulation of excessive excitation of the sympathetic nervous system caused by stimulation of the pelvic organs in the paralyzed region, such as urinary distension or constipation. The afferent stimulus reaches the major splanchnic sympathetic outflow (T5-T6) and stimulates a sympathetic response. The sympathetic response causes vasoconstriction, resulting in hypertension, pounding headache, pallor, facial flushing, and goosebumps below the level of injury. This hypertension evokes parasympathetic nerve activity via the vagus nerve, causing the bradycardia reaction. This situation continues until the trigger stimulus is eliminated, and, in severe cases, it can be extremely dangerous because it carries the risk of intracerebral

hemorrhage or seizure[3,4,5].

Treatment for AD has generally comprised simple common sense, involving an attempt to resolve the AD by withdrawing the trigger stimulus. For example, AD is most commonly caused by urinary distension; in such cases, urethral catheterization to drain the urine from the bladder rapidly lowers blood pressure [1, 2]. Other reported triggers for AD include constipation, bedsores, ingrown nails, contracture, muscle spasms, pregnancy and childbirth, ejaculation, urinary tract stones, urinary tract infection, gallstones and cholecystitis, gastric ulcer, anal fissure, and bone fracture [1]. In extremely rare cases, Charcot's spine (neuropathic spinal arthropathy), breastfeeding, pulmonary edema, pulmonary embolism, and chronic pheochromocytoma [6-11] have also been reported as triggering AD.

There have been other reports of AD being triggered by passive exercise of the hip joint during rehabilitation. McGarry et al. reported that AD occurred during passive stretching of the hip joint during rehabilitation in three patients with cervical cord injury [12]. Colachis et al. reported one case of AD triggered during hip joint range of motion (ROM) exercise [13]. The fact that passive exercise of the hip joint has been reported as a trigger for AD is not well known. There have been no reports from Japan of AD occurring due to hip joint position or ROM exercise. However, we recently treated two patients with cervical cord injury in whom AD occurred as a result of a particular hip position at rest. Unlike ordinary causes of AD or the passive exercise of the hip joint described above, this is regarded as extremely rare. Thus, these two cases of AD due to a rare cause are reported, with a discussion of the hypothesized mechanism of AD and possible preventive measures. The patients and their families were informed that data from the case would be submitted for publication and gave their consent.

Case report

Case 1

The patient was a 38-year-old man. He had fallen from a height of 4 meters while jumping on a trampoline and suffered C6/7 fracture dislocation, resulting in complete C6 tetraplegia that American Spinal Injury Association (AISA) Impairment Scale was A. Anterior and posterior fusion of the cervical vertebrae and bone grafting were performed to treat the fracture dislocation. Rehabilitation therapy was initiated from the early phase, including respiratory training, ROM exercise, and muscle strengthening exercise. No subsequent neurological improvement was evident, but comprehensive rehabilitation enabled him to transfer to and drive a wheelchair independently. His finger skills were insufficient to perform urinary tract management for neurogenic bladder, which was managed by an indwelling catheter. No limitations in the ROM of the hip joint were evident on physical examination.

Course of AD

The patient's normal blood pressure was around 100/70 mmHg in the supine position. From 5 months after injury, systolic blood pressure frequently increased to over 200 mmHg during nighttime sleep and rehabilitation, with accompanying severe headache, bradycardia, and facial flushing. Although this was clearly AD, there was never any catheter blockage, constipation, bedsores, or noxious skin stimulation when it occurred; since the cause remained entirely unclear, it was treated with antihypertensive drugs. In terms of imaging findings, hip joint X-rays and abdominal CT showed no abnormalities such as renal stones, cholecystitis, or gallstones, nor was there any ectopic ossification around the hip joint.

As a result of careful observation, the possibility was raised that AD might not be triggered by one of its typical causes but because of the position of the hip joint. To verify this hypothesis, blood pressure and heart rate were measured by using noninvasive blood pressure monitor continuously in a range of leg positions. Figure 1 shows the results of the mean blood pressure and heart rate measurements taken while the patient was sitting in a wheelchair or in the supine position with both hip joints flexed at angles of 0º and 20º and extended at an angle of 20º. The data shown in Figure 1a were taken 2-3 min after each position was adopted. During this process, the patient was immediately returned to a safe position if blood pressure increased to a dangerous high level. Hypertension and bradycardia were observed with the hip joints flexed at 0º and extended at 20º, meaning that AD had been triggered. AD was not evident when the hip joints were slightly flexed, indicating that this was an effective position. As a preventive procedure, while the patient was lying down, the hip joints were placed in a slightly flexed position; thereafter, the incidence of AD during nighttime sleep and rehabilitation decreased markedly.

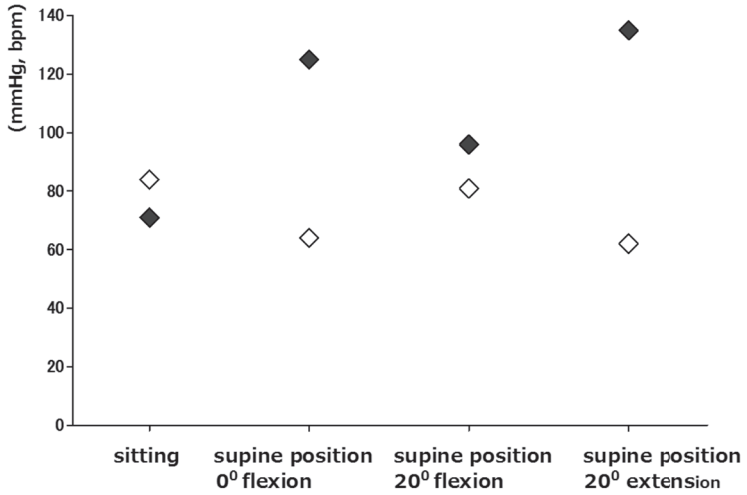

Fig.1 Response to alteration of hip joint angles of Case 1

Filled dots represent the mean blood pressure and empty dots represent bpm（beats per minute）.

Case 2

The patient was a 41-year-old man. He had fallen from the roof of his house and suffered a C6/7 fracture dislocation, resulting in complete C8 tetraplegia that AISA Impairment Scale was A. He was admitted to our hospital for rehabilitation 1 month after having undergone anterior and posterior fusion of the cervical vertebrae. No subsequent neurological improvement was evident, but comprehensive rehabilitation enabled him to transfer to and drive a wheelchair independently as Case 1. Intermittent self-catheterization was introduced for neurogenic bladder, and he could manage by himself independently. Physical findings revealed no limitations in the ROM of the hip joint.

Course of AD

The patient's normal blood pressure in the supine position was around 100/60 mmHg. From 4 months after injury, systolic blood pressure would increase to around 200 mmHg at nighttime with accompanying headaches, and this was particularly frequent during intermittent catheterization. There was no decrease in blood pressure even when urine was drained to prevent accumulation in the bladder. In this case as well, AD was clearly occurring, but there was never any bladder distension, constipation, or noxious skin stimulation when it occurred; the cause remained entirely unclear. As in Case 1, in terms of imaging findings, hip joint X-rays and abdominal CT showed no abnormalities.

The possibility was raised that AD might not be triggered by one of its normal causes but because of the abducted position of the hip joint, as not in Case 1. To verify this hypothesis, the patient was placed in a supine position with the hip joints flexed at 0º, and unilateral abducted at 20º and bilateral abducted at 20º in the supine position. Blood pressure and heart rate were measured in the same way as in Case 1. The patient had no constipation at this time, and urine had been drained from the bladder by intermittent catheterization. Figure 2 shows the mean blood pressure and heart rate measurements in each position. The data shown in the figure were taken 2-3 min after each position was adopted. The patient was immediately returned to a safe position if blood pressure increased to a dangerous high level. Obvious AD was not observed when the hip joints were flexed at 0°or slightly flexed. AD was observed when the hip joints were abducted. That was present when bilateral hip joints were abducted at 20º, and also with unilateral hip joint abduction to 20º. In this case, the trigger factor for AD was confirmed to be hip joint abduction, and when the patient avoided positions requiring hip joint abduction during intermittent catheterization, the incidence of AD was found to decrease markedly, as in Case 1.

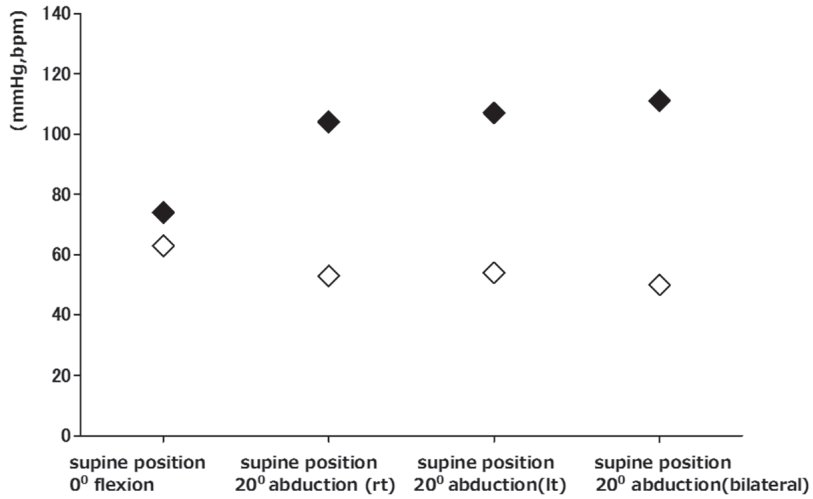

Fig.2　Response to alteration of hip joint angles of Case 2

Filled dots represent the mean blood pressure and empty dots represent bpm（beats per minute）.

Discussion

Two cases of AD with an unusual cause were described. In both patients, AD was triggered during daily life activities or rehabilitation by the commonplace factor of hip position. This is an extremely rare trigger factor that has not been previously reported. Hip joint position of extension or abduction triggered severe AD in each patient, though the joint position differed between the cases. Surprisingly, AD occurred easily during nighttime sleep in the supine position with the hip joint in the neutral and extended position in Case 1. We found also that AD even occurred when the unilateral and bilateral hip joint was in the abducted position in Case 2, a position similar to that applied for the hip joint position stretching during rehabilitation or during intermittent catheterization. In both cases, a particular hip joint position was understood to constitute the factor triggering AD.

The reason why that was triggered by hip joint position remains speculative, but we propose and discuss one possible mechanism. The possibility that abdominal organs such as the bladder or intestines received some type of stimulus from hip joint neutral or abduction position cannot be ruled out. However, we consider this to be unlikely, as a full bladder and constipation were absent in both cases, and blood pressure and heart rate were measured in a variety of hip positions, with AD being triggered as a result. In addition, in both cases, abdominal CT and hip joint X-rays revealed no abnormalities. In the present two cases, we inferred that positions of hip joint extension or abduction resulted in stimulation of the femoral nerve, which innervates the iliopsoas muscle, and the obturator nerve, which innervates the adductor muscles, and this stimulus arrived at the lumbar spinal cord as an afferent impulse, causing AD. In both cases, when that was triggered it was expressed as a mass reflex, increasing

muscle spasticity in the legs and trunk.

There have been no previous reports of cases completely similar to the present cases, but the differences between these cases and two other reports relating to the hip joint are worthy of note. McGarry et al. reported AD triggered by passive stretching (straight leg raising) of the hip joint in three patients with cervical cord injury [12]. With respect to the AD trigger, they claimed that stretching of the hip joint or proximal leg muscle, innervated by the fourth and fifth lumbar and first sacral segments, was the stimulus for the AD. Unlike the present cases, in their report, AD occurred when the hip joint was flexed on passive stretching. Colachis et al. reported a single case of AD occurring during hip joint ROM exercise and causing transient aphasia [13]. They suggested that the stimulus causing the AD was increased bladder pressure during ROM exercise, but did not present direct evidence. In the present cases, the actual occurrence of that was measured in both patients when the bladder was empty. The hip joint angle when that occurred in each position was also under 20º, a position that is assumed to have little effect on increasing internal bladder pressure.

AD is a high-risk complication that can be life-threatening in patients with cervical or high thoracic cord injury, and not only rehabilitation staff but all medical professionals concerned with SCI should be aware of its occurrence. In severe cases, it may cause central nervous disorders, such as cerebral hemorrhage or seizure [3, 4, 5]. If the cause of AD is unknown, a variety of different antihypertensive drugs may be used [14], and once the cause of AD can be identified, it is generally easy to deal with. In Case 1, the method for improvement was to place the hip joint in a slightly flexed position; in Case 2, the strategy was to avoid hip joint abduction during intermittent catheterization. These simple procedures had an excellent clinical effect, with a marked decrease in the frequency of that. The fact that there was no occurrence of severe hypertension after these preventive procedures were implemented also demonstrates their effectiveness.

In conclusion, AD in the present two cases was triggered by simple hip joint positions. AD in patients with SCI is often caused by bladder distension or constipation and can generally be dealt with easily. However, if the trigger factor is not well known or the cause seems to be unusual, the fact that hip joint position also constitutes a trigger factor for that should be considered.

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