Pelvic Autonomic Dysfunction without Paraplegia: A Sequel of Spinal Cord Stroke

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Dear Sir,

Spinal cord stroke is rare; its incidence has been reported as 1–2% of all admissions related to vascular pathology of the nervous system in several neurology departments [1, 2]. The typical clinical manifestation of spinal cord stroke is sudden onset of paraplegia, superficial but not deep modality of sensory loss below the level of the lesion, and pelvic dysfunction, e.g. urinary retention, constipation, and erectile dysfunction [3–5]. Spinal cord stroke is located at the thoracic or thoracolumbar level in the majority of cases [1–3]. Magnetic resonance imaging (MRI) scan and pathology studies have demonstrated lesions involving the bilateral central cord but relative sparing of the posterior cord, which affect pyramidal and spinothalamic pathways [1–3]. However, pelvic dysfunction without paraplegia as a sequel of spinal cord stroke is quite uncommon. We recently had such a patient, who had suffered from pelvic dysfunction for 5 years after a spinal cord stroke.

Case Report

A 57-year-old, previously healthy man suddenly developed paraplegia, urinary incontinence, and numbness in the legs, particularly in the ankles and saddle area

when he stood up while at work. He had no history of previous illnesses such as hypertension or coagulopathies. Upon admission to a local hospital, the patient had complete paraplegia and decreased pain sensation below the inguinal ligament and an indwelling urinary catheter was inserted. Laboratory investigation including coagulopathy was normal. Echography of the abdomen and the aorta was normal. Arteriography and cerebral spinal fluid analysis were not performed. Although MRI of the thoracic and lumbar spine was normal, he was diagnosed with spinal cord stroke based on the clinical findings. Intravenous steroid therapy gradually ameliorated his paraplegia. Three weeks later, he regained the ability to walk independently. However, he was found to have severe constipation that required daily laxatives and occasional fecal incontinence. After the indwelling urinary catheter was removed, he suffered from urinary retention. Because he experienced a strong urgency sensation, he was taught to perform clean intermittent self-catheterization (CISC) 8 times during the day and twice during the night. He also had occasional urinary incontinence. Two years after the onset of the disease, the patient still had urinary retention and was referred to our laboratory in order to perform pelvic

autonomic function tests. Upon referral, he had normal muscle power in the four extremities, and his deep tendon reflexes in the lower extremities were brisk bilaterally without extensor plantar reflexes. Superficial sensation determined by pin prick was decreased below the level of L₂. Position sensation in the extremities was preserved. Anal reflexes were absent. He also suffered from erectile and ejaculatory dysfunction. Again, laboratory investigation including coagulopathy was normal, and MRI scans of the cervical, thoracic and lumbar spine were normal. Tibial somatosensory evoked potentials were normal as well.

Lower Urinary Tract Function (fig. 1)

A urodynamic study [6] (Urovision, Lifetec, Inc., Houston, Tex., USA; Neuro-pack Sigma, Nihon Kohden, Inc., Tokyo, Japan) was performed to measure postvoid residual volume and medium-fill (50 ml/min) water cystometry. Simultaneously, sphincter electromyography (EMG) was carried out using a concentric needle electrode in the external anal sphincter muscles. The patient could not void, and had a postvoid residual volume of up to 200 ml (normal <30 ml). A normal volume was observed at the first sensation of the bladder filling, i.e., a volume of 124 ml (100 <

normal < 280) [7]. However, a decreased volume was observed at bladder capacity, i.e., a volume of 184 ml ($245 < \text{normal} < 600$) [7] during the storage phase. He had detrusor overactivity (normal, none). When the patient tried to start voiding, he showed an acontractile detrusor and the sphincter EMG sound was unchanged, indicating a nonrelaxing sphincter; this was followed by considerable straining. During the voiding phase, he could not void irrespective of detrusor overactivity (detrusor hyperactivity with impaired contractile function, DHIC).

Lower Gastrointestinal Tract Function (fig. 2)

A small test capsule (containing 20 radiopaque markers: one marker corresponding to 1.2 h) was ingested once a day for 6 days shortly after breakfast, and a week later, an abdominal X-ray was obtained [5, 8]. He had a slightly prolonged total colonic transit time (CTT) of 47.0 h (normal 16.0–48.0 h) [9], comprising a right CTT of 8.4 h (normal 3.6–7.2 h), a left CTT of 18.0 h (normal 2.4–19.2 h) and a rectosigmoid CTT of 20.4 h (normal 7.2–26.4 h). Static anal manometry showed a normal resting pressure of 74 cm H₂O (normal >40 cm H₂O) and a squeeze pressure of 114 cm H₂O (normal >50 cm H₂O). Videomanometry was performed, simultaneously measuring rectal pressure, abdominal pressure, sphincter pressure, sphincter EMG activity and fecal flow by the same urodynamic computer [8, 10]. A normal volume was observed at the first sensation of rectal filling, i.e., a volume of 250 ml ($50 < \text{normal} < 280$) and at rectal capacity, i.e., a volume of 350 ml ($140 < \text{normal} < 600$) during the storage phase. During rectal filling, he had infrequent small phasic rectal contractions of 10 cm H₂O ($10 < \text{normal} < 50$) with leaking (normal, none). In the defecation phase, he had no rectal contractions with marked strain (strain type defecation) and defecatory difficulty. The motor unit potentials of the external sphincter were examined by the same EMG computer. He had normal motor unit potentials with a mean duration of 7.71 ms (normal <10 ms) [11].

He was started on 1.5 mg/day of clonazepam for numbness, 15 mg/day of mosapride, a 5-HT₄ selective agonist [10] for constipation, and 60 mg/day of urapidil, an α -adrenergic antagonist for urinary retention. In contrast to the improvement

Fig. 1. Urodynamic recording of the patient's bladder. In the storage phase (a), the urodynamic study showed a normal volume of 124 ml at the first sensation, but a decreased volume at the bladder capacity of 184 ml, together with marked detrusor overactivity (arrowheads). In the voiding phase (b), the patient could not contract his bladder voluntarily at all, indicating an acontractile detrusor, whereas involuntary detrusor overactivity appeared repeatedly without urinary flow (arrowheads). The sphincter EMG sound was unchanged, indicating a nonrelaxing sphincter. Note that the sweeping speed in the voiding phase was 5 times faster than that in the storage phase. Flow = Urinary flow; P_{ves} = vesical (bladder) pressure; P_{abd} = abdominal (rectal) pressure; P_{diff} = differential detrusor pressure = P_{ves} – P_{abd}.

in numbness and constipation, urinary retention was unchanged for the following 3 years. He performed CISC 4 times during the daytime and used a night balloon [12] during the nighttime, which improved his sleep quality significantly. His major disability was still sacral autonomic dysfunction.

Discussion

Although MRI scans of this case were normal, a sudden appearance of neurological signs strongly suggested spinal

cord stroke in the anterior spinal artery area [1–3] at or above the L₂ segment. A remarkable feature of our patient was the dissociation between his recovery from paraplegia and pelvic autonomic dysfunction; in other words, while his paraplegia disappeared almost completely, his urinary retention remained to the extent that he needed CISC over a period of 5 years. Although we could not find similar cases, Inatomi et al. [13] previously reported 3 patients with spinal cord infarction whose symptoms started with urinary dysfunction. The first case (a 55-year-old man) noticed difficulty in

Fig. 2. Videomanometric recording of the patient's rectum. In the storage phase, videomanometry showed a normal volume at the first sensation of 250 ml and at the rectal capacity of 350 ml. However, he had smaller phasic rectal contractions only infrequently (arrowheads). In the defecation phase, he had no rectal contrac-

tion with marked strain (strain type defecation) with defecatory difficulty. Flow = Urinary flow; P_{rect} = rectal pressure; P_{abd} = abdominal (bladder) pressure; P_{diff} = differential detrusor pressure = $P_{\text{rect}} - P_{\text{abd}}$; P_{ana} = anal pressure.

urination, followed by an abrupt onset of a burning pain below his neck 2 days later. His muscle power and deep reflexes were normal. The second case (a 66-year-old woman) had sudden difficulty in urination and numbness on the thighs bilaterally. She had normal muscle power but had slight hyperreflexia in the extremities. The third case (a 66-year-old man) had a sudden onset of difficulty in urination. Three days after the onset, a transverse lesion below the T_{10} dermatome was completed.

Pelvic autonomic function tests in our case revealed DHIC in the bladder [4, 14], suggesting suprasacral spinal cord lesion. The pathophysiology of DHIC in spinal cord lesions is that a single partial lesion in the spinal autonomic pathways disrupts

the spino-bulbo-spinal micturition reflex arc, and below the lesion, a C-fiber-mediated novel sacral micturition reflex arc can emerge [8]. Pelvic autonomic function tests also revealed a prolonged CTT [5] and decreased spontaneous phasic contractions [5], but without neurogenic sphincter EMG [11], presumably suggesting a suprasacral cord lesion. Tibial somatosensory evoked potentials were normal in our case. These findings indicated that the lateral medullary reticulospinal tract [10, 13, 15, 16] just inside the pyramidal tract, e.g. the descending pathway to the bladder and the bowel, was selectively affected in our case.

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