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CASE REPORT

A young man with herpes simplex encephalitis: Andrew and Nathan type urodynamic abnormality

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ABSTRACT

We described a case of a young man who, after recovery from herpes simplex encephalitis with lesions in the frontal and insular cortices, developed urinary incontinence. An urodynamics revealed decreased sensation, inability to initiate a volitional detrusor contraction and terminal detrusor overactivity (DO) without residual. His symptoms were initially treated with clean, intermittent self-catheterization (CIC) and over time his bladder disorder improved. This might be one form of classical 'Andrew and Nathan type' frontal urodynamic abnormality.

Keywords: herpes simplex encephalitis, frontal lobe, urinary retention, bladder sensation, detrusor overactivity

INTRODUCTION

In 1964-1966 Andrew and Nathan in London [1-3], pioneers of neurogenic bladder dysfunction of the frontal lobe origin, presented typical case records: "The patients described here were not demented, indifferent or lacking in social awareness; they were much upset and embarrassed by these symptoms… The acts of micturition and defaecation occur in a normal manner… The sensation of gradual awareness of increasing fullness of the bladder and the sensation that micturition is imminent, are impaired. When the syndrome is less pronounced, the sensation underlying the desire to micturate is absent, whereas the sensation that micturition is imminent still occurs. Then the patient is waylaid by a sudden awareness that he is about to pass urine; when neither sensation is experienced, the patient is amazed to find that he has passed urine." The cases clearly indicated abnormality of bladder sensation. However, no urodynamic confirmation has been made so far in patients with frontal lobe lesions [4,5]. We recently had a case of a 29-year-old man who, after suffering from herpes simplex encephalitis (HSE), developed severe bladder dysfunction due to frontal-insular lesions.

CASE REPORT

A 29-year-old, previously healthy man began to have high fever and coma. He was admitted to a local hospital, where he was diagnosed with HSE on the basis of increased cell count and total protein with positive herpes simplex virus type 1(HSV1) antigen by a polymerase chain reaction (PCR) test in the cerebrospinal fluid. Antiviral therapy successfully recovered his consciousness, with mild residual cognitive deterioration, disuse atrophy of the limbs, and urinary incontinence. Four months later, he was transferred to a rehabilitation hospital. His cognitive deterioration and disuse atrophy of the limbs disappeared almost completely, while he continued to have severe urinary incontinence that embarrassed this young man most, which two months later referred him to our urodynamic laboratory.

On referral to our hospital, he was alert and cooperative. Cognitive tests revealed normal general cognitive function (the Mini-Mental State Examination score of 28/30, normal > 24) but low frontal executive function (the Frontal Assessment Battery score of 13/18, normal > 16). He had normal muscle strength and deep tendon reflexes. Sensation including the perineal area was normal. He had bladder dysfunction: an International Prostate Symptom Score of 11/35 and an Overactive Bladder (OAB) Symptom Score of 12/15, both mainly due to urinary incontinence. He had urinary incontinence without warning; and before incontinence, he had no urinary sensation at all including urinary urgency.

The results of his blood test were normal. Brain magnetic resonance images (MRI) of the patient taken at 6 month after disease onset showed high signal, left-side-dominant bilateral lesions at the frontal cortex involving the middle cingulate cortex and the frontal operculum, the insular cortex, and the temporal cortex involving the temporal pole and the parahippocampal gyrus (**Fig. 1**). Brain single-photon emission computed tomography showed decreased blood flow at left medial surface of the frontal cortex etc. as seen in the MRI scan.

Urodynamic study results (6 months after disease onset)

The methods and definitions used for the urodynamic study and sphincter EMG conformed to the standards proposed by the International Continence Society [6,7].

Although he did not report any difficult urination, a free flow showed that he could not void voluntarily at all, and 315 ml of post-void residual

(PVR) was catheterized. Before the infused water volume reaches 600 ml (that provokes DO) we asked him to void; however, he was unable to voluntarily contact his bladder at all (data not shown).

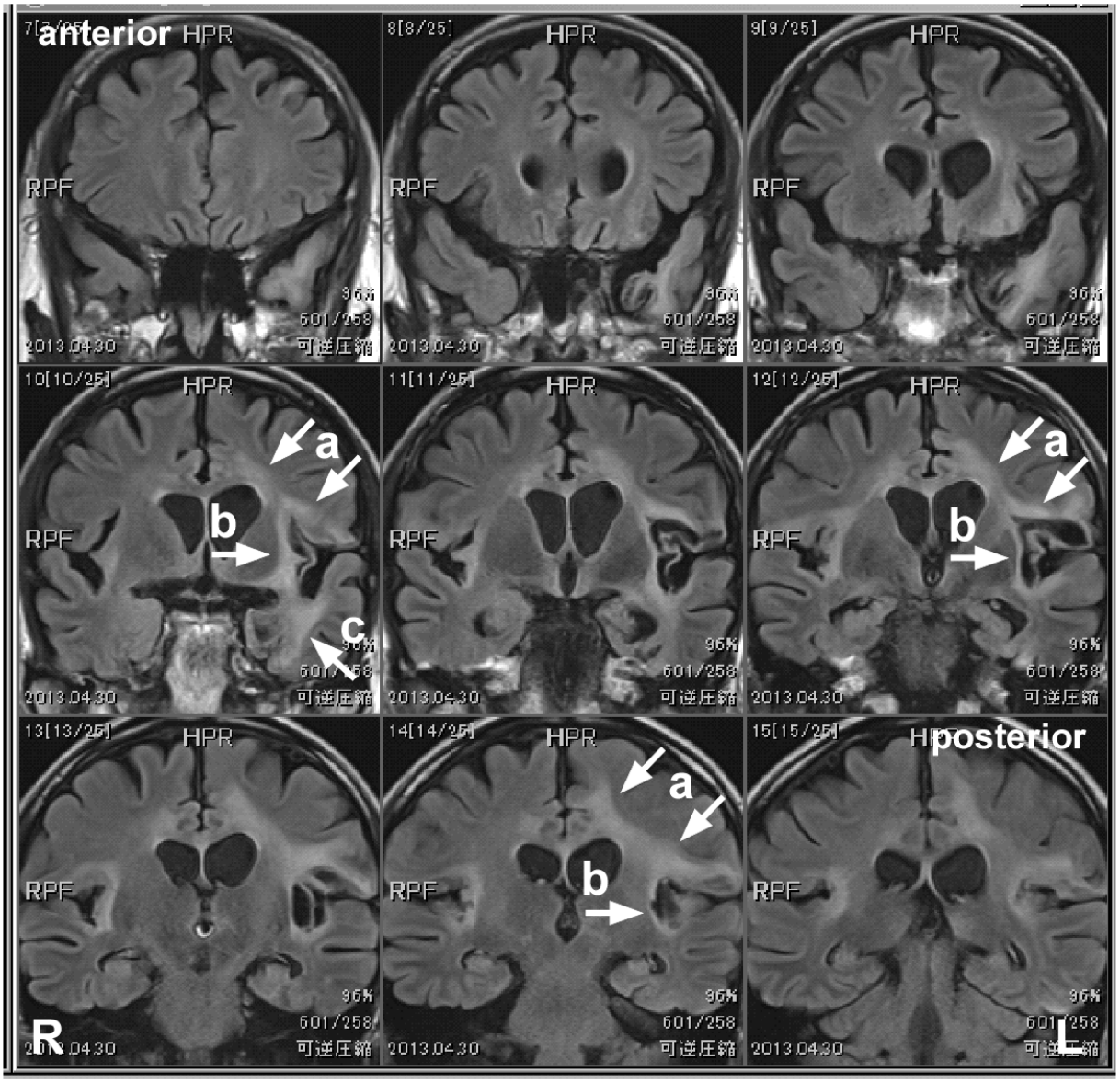

Figure 1. MRI of the patient taken at 6 months after disease onset. Fluid attenuated inversion recovery (FLAIR) coronal MRI images of the patient showed high signal, left side dominant lesions bilaterally at (arrow a) the frontal cortex involving the middle cingulated cortex and the frontal operculum (with cystic formation), (arrow b) the insular cortex (with cystic formation), and (arrow c) the temporal cortex involving the temporal pole and the parahippocampal gyrus (with cystic formation).

During bladder filling, he reported the first sensation at 400 ml (100 ml <normal< 300 ml), but after that, his bladder sensation was only minimum, and his bladder capacity exceeded 600 ml (200 ml <normal< 600 ml); we then stopped infusing saline into the bladder in order to avoid over-distension bladder injury. After provocation by coughing, he showed detrusor overactivity (DO) twice (**Fig. 2**, downward arrowheads 1,2). The sphincter EMG activity disappeared on DO that normally increases in order to prevent incontinence, therefore we regarded this as uninhibited sphincter relaxation (upward arrowheads). Together with the second DO he leaked involuntarily. We asked him to continue urinating voluntarily. However, he could only strain voluntarily with an intermittent urinary flow. Pressure-flow analysis of this flow (mostly involuntary leaking together with DO) showed normal maximum urinary flow rate (42 ml/s), low average urinary flow rate (7 ml/s, normal >10); no obstruction [Abrams unobstructed; Schafer 1 (normal<2)]; strong detrusor (Schafer strong, Watts factor 42.6 watts/m^2); no detrusor-sphincter dyssynergia; and minimum [PVR volume (20 ml)].

In summary, he could not voluntarily void at all with large PVR before his bladder volume reaches 600 ml. During bladder filling, his bladder sensation was markedly decreased. When his bladder volume reaches 600 ml, urinary leakage was noted at the time of DO without PVR.

In order to ameliorate his bladder condition, we first taught him to perform clean, intermittent self-catheterization (CISC) three times a day in order to avoid over-distension bladder injury. Along with CISC, urinary incontinence was resolved. Two months later (8 months after disease onset), he became able to sense his bladder before an amount of 400 ml as well as to void voluntarily without medication. The number of CISC could be tapered to once a day, but urinary incontinence did not re-appear. After his voluntary voiding, his PVR volume was 80 ml and CISC was stopped.

DISCUSSION

The present patient had the following: a 29-year-old man had a history of HSE 6 months ago. Brain MRI scans revealed left-side-dominant bilateral lesions at the middle cingulate (medial frontal) and insular cortices. After recovery from high fever and coma by antiviral therapy, he had no significant cognitive decline or gait difficulty. However, he had severe urinary incontinence that embarrassed this young man most. His prostate volume was normal. An urodynamic test revealed decreased sensation, inability to initiate a volitional detrusor contraction, and urinary leakage was noted at the time of DO without PVR. After starting CISC three times a day, bladder dysfunction ameliorated spontaneously (8 months after disease onset). While brainstem HSE can lead to bladder dysfunction [8], to the best of our knowledge, this is the first cortical HSE case to show neurogenic bladder dysfunction.

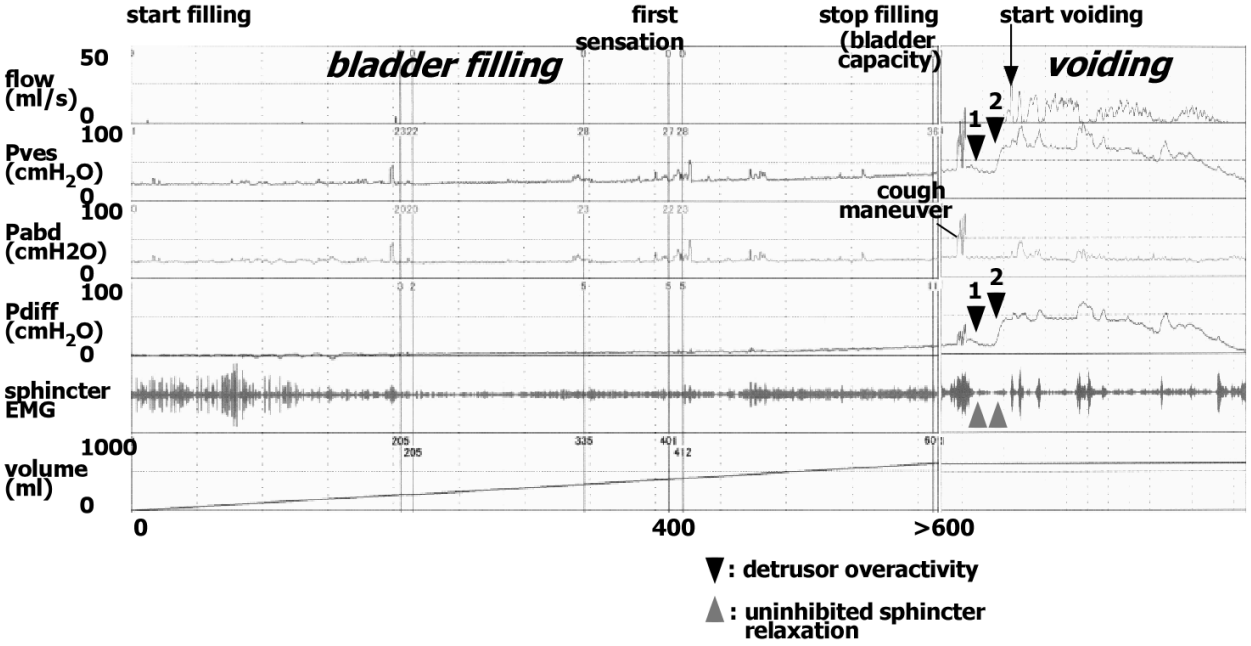

Figure 2. Urodynamic recording of the patient 6 months after disease onset. Flow, urinary flow; Pves, vesical (bladder) pressure; Pabd, abdominal (rectal) pressure; Pdiff, differential detrusor pressure = Pves - Pabd; EMG, electromyography.

In 1964-1966 Andrew and Nathan [1-3] reported a series of patients with bladder dysfunction due to frontal tumor, post-aneurysm surgery, etc. In those papers, cystometry recording clearly showed the presence of DO. However, bladder sensation during storage and bladder function during voiding were not presented urodynamically, while bed-side observation of these patients indicated decreased bladder sensation. The present case urodynamically showed decreased bladder sensation due mostly to left-side-dominant bilateral lesions at the middle cingulate and insular cortices [9]. It is well documented that bladder afferent signals reach the frontal lobe including the prefrontal, cingulate and insular cortices in experimental animals [10,11] and humans[12,13]. Therefore, it is reasonable to assume that lesions in the cingulate and insular cortices lead to decreased bladder sensation. DO is a motor phenomenon, and commonly occurs in lesions in the cingulate and insular cortices [14] that mainly inhibit the micturition reflex [11]. It is noteworthy that before the occurrence of DO, he was unable to contract his bladder, which was observed with decreased bladder sensation. This combination of bladder disorder is previously not well documented. It is reasonable to assume that triggering/initiation of bladder contraction needs appropriate sensory input from the lower urinary tract [11]. It is documented that dorsal rhizotomy interrupted the micturition reflex [15] and lessens DO in patients with intractable urinary incontinence. Sensory de-afferentiation within brain might have interfered voluntary voiding in our patient.

In conclusion, we described a case of a young man who, after recovery from HSE with lesions in the frontal and insular cortices, developed urinary incontinence. An urodynamic test revealed decreased sensation, inability to initiate a volitional detrusor contraction, and urinary leakage was noted at the time of DO without PVR. His symptoms were initially treated with clean, intermittent self-catheterization (CIC) and over time his bladder disorder improved. This might be one form of classical 'Andrew and Nathan type' frontal urodynamic abnormality.

References

1. Andrew J, Nathan PW (1964) Lesions on the anterior frontal lobes and disturbances of micturition and defaecation. Brain 87: 233-262. PMID: 14188274
2. Andrew J, Nathan PW (1965) The Cerebral Control of Micturition. Proc R Soc Med 58: 553-555. PMID: 19994438
3. Andrew J, Nathan PW, Spanos NC (1966) Disturbances of micturition and defaecation due to aneurysms of anterior communicating or anterior cerebral arteries. J Neurosurg 24: 1-10. doi: 10.3171/jns.1966.24.1.0001. PMID: 5903296
4. Wyndaele JJ (1993) Is impaired perception of bladder filling during cystometry a sign of neuropathy?. Br J Urol 71: 270-273. PMID: 8477312
5. Tsunoyama K, Sakakibara R, Yamaguchi C, Uchiyama T, Yamamoto T, et al. (2011) Pathogenesis of reduced or increased bladder sensation. Neurourol Urodyn 30: 339-343. doi: 10.1002/nau.20953. PMID: 21305587
6. Abrams P, Cardozo L, Fall M, Griffiths D, Rosier P, et al. (2002) The standardisation of terminology of lower urinary tract function: report from the Standardisation Sub-committee of the International Continence Society. Neurourol Urodyn 21: 167-178. PMID: 11857671
7. Sakakibara R, Uchiyama T, Yamanishi T, Kishi M (2008) Sphincter EMG as a diagnostic tool in autonomic disorders. Clin Auton Res 19: 20-31. doi: 10.1007/s10286-008-0489-5. PMID: 18780143
8. Sakakibara R, Hattori T, Fukutake T, Mori M, Yamanishi T, et al. (1998) Micturitional disturbance in herpetic brainstem encephalitis; contribution of the pontine micturition centre. J Neurol Neurosurg Psychiatry 64: 269-272.

PMID: 9489547

9. Fowler CJ, Griffiths D, de Groat WC (2008) The neural control of micturition. Nat Rev Neurosci 9: 453-466. doi: 10.1038/nrn2401. PMID: 18490916
10. Tai C, Wang J, Jin T, Wang P, Kim S, et al. (2009) Brain switch for reflex micturition control detected by FMRI in rats. J Neurophysiol 102: 2719-2730. doi: 10.1152/jn.00700.2009. PMID: 19741099
11. de Groat WC, Wickens C (2012) Organization of the neural switching circuitry underlying reflex micturition. Acta Physiol (Oxf) 207: 66-84. doi: 10.1111/apha.12014. PMID: 23033877
12. Griffiths DJ, Fowler CJ (2012) The micturition switch and its forebrain influences. Acta Physiol (Oxf) 207: 93-109. doi: 10.1111/apha.12019. PMID: 23164237
13. Sakakibara, , Tateno F, Yano M, Takahashi O, Sugiyama M, et al. (2013) Tolterodine activates the prefrontal cortex during bladder filling in OAB patients: a real-time NIRS-urodynamics study. Neurourol Urodyn 33: 1110-1115. doi: 10.1002/nau.22471. PMID: 24038213
14. Sakakibara R, Fowler CJ, Hattori T (1999) Voiding and MRI analysis of the brain. Int Urogynecol J Pelvic Floor Dysfunct 10: 192-199. PMID: 10430014
15. Liao J, Cheng C, Lee S, Chen G, Chen K, et al. (2006) Impaired micturition reflex caused by acute selective dorsal or ventral root(s) rhizotomy in anesthetized rats. Neurourol Urodyn 25: 283-289. doi: 10.1002/nau.20220. PMID: 16496396