After suffering a brainstem stroke, a 62-year-old man developed locked-in syndrome including loss of horizontal eye movement and increased anal tone. Magnetic resonance imaging (MRI) of the patient revealed a massive stroke in the pons and right cerebellum, which seemed to involve the pontine micturition/defecation center (Barrington’s nucleus) and the rostral pontine reticular formation (RPRF). As his increased anal tone was intractable to medical treatment, he required intermittent catheterization with an anal bougie tube. In light of the reported cases, our patient developed increased anal tone presumably due to pontine defecation center and RPRF lesion.

**Key words** anal tone, brainstem stroke, pontine defecation center

1. **INTRODUCTION**

A “pontine defecation center” has been demonstrated in experimental animals, which is located adjacent to the pontine micturition center (Barrington’s nucleus) and the locus ceruleus.1–4 We recently had a male patient who, after a brainstem stroke that involved the pontine defecation center, developed locked-in syndrome and together with increased anal tone.

2. **CASE REPORT**

A 62-year-old man with hypertension and coronary stent gradually developed dizziness on walking and a staggering gait. Three weeks later, he was referred to our hospital. He had no history of constipation. At referral, he had a macro-square-wave jerk at rest and leftward nystagmus on left gazing, but without apparent cerebellar or other neurologic dysfunction. Brain magnetic resonance imaging (MRI) revealed right-side dominant, small high-signal spots on diffusion-weighted images in the pons. MR angiography revealed diffuse irregular stenosis in the vertebrobasilar artery. Laboratory tests revealed mild diabetes (HbA1C 7.2%, normal <5.8). He was diagnosed with progressive pontine infarction. On admission, he was started on 160 mg/day intravenous sodium ozagrel, an anti-platelet agent, and oral drugs for hypertension and diabetes. To measure water balance and because of immobility, he was inserted with an indwelling Foley catheter. Because of sudden onset of disease in the patient, we did not know of lower urinary tract symptoms. We did not perform urodynamics because of the patient’s immobility. He had bowel movements four times a week without defecation difficulty. Abdominal X-ray was normal. However, two weeks after admission, he suddenly developed tetraplegia following vertigo. Tendon reflexes were slightly increased and plantar reflexes showed a bilateral extensor response. Eye movements were abolished horizontally but preserved vertically. An MRI scan revealed a massive stroke in the pons and right cerebellum (Fig. 1). He was started on 1.5 mg/day warfarin through a nasal tube, which prevented further exacerbation. Five days after occurrence of locked-in syndrome, he was started on tube feeding. However, he became severely constipated. Abdominal X-ray showed marked gas in the colon (Fig. 2), whereas the bowel sound was well preserved. Digital examination revealed extremely increased anal tone; through which feces did not come out. We did not know whether increased anal tone was reflexive or spontaneous as we did not perform an anal physiology test. Because of increased anal tone the patient needed an intermittent catheterization with an anal bougie tube. Although we did not perform an anal physiology test, his constipation was considered secondary to increased anal tone, which simulated anismus. A pontine lesion on MRI seemed to involve the pontine defecation center in the tegmentum (Fig. 1a–c). Mosapride citrate, a selective serotonin 4 receptor agonist, and magnesium oxide did not ameliorate his condition. Stoma was not indicated because of coronary heart disease. The patient’s increased anal tone remained unchanged for the following two months, and he was referred to a local rehabilitation hospital.

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Received 2 November 2011; revised 28 December 2011; accepted 9 January 2012.

DOI: 10.1111/j.1757-5672.2012.00143.x
3. DISCUSSION

It is remarkable that our patient developed both locked-in syndrome and increased anal tone after suffering a brainstem stroke. Locked-in syndrome (pseudocoma) describes patients who are awake and conscious but selectively de-efferented (i.e. have no means of producing speech, limb or facial movements). As described by Plum and Posner, voluntary motor paralysis prevents subjects from communicating by word or body movement. Usually, but not always, the anatomy of the responsible lesion in the brainstem is such that locked-in patients are left with the capacity to use vertical eye movements and blinking to communicate their awareness of internal and external stimuli.\(^5\)

Whereas colonic motility seemed to be preserved in our case, increased anal tone did not respond to current prokinetic medications. This condition needed intermittent catheterization with an anal bougie tube. Previously, Weber et al.\(^6\) reported a similar observation with ours, although detailed location of the lesions were not included. They suggested that brainstem stroke can interfere with coordinated relaxation of the anal sphincter. Increased anal tone (without voluntary defecation) or anismus (with an attempt of voluntary defecation) has been reported in multiple sclerosis,\(^7\) spinal cord injury,\(^8\) and Parkinson’s disease.\(^9\)

The defecation reflex is thought to be a spinal reflex. In addition, experimental studies indicated that Barrington’s nucleus is critical for supraspinal control of colonic and anorectal motility by bulbospinal pathways, and at least in part, by vagal pathways.\(^10,11\) Barrington’s nucleus is located in or adjacent to the locus ceruleus in humans.\(^11,12\)
On MRI images, both sides (right-side dominant) of the pontine micturition/defecation center were involved in our case. On the other hand, the rostral pontine reticular formation (RPRF), located in the ventromedian side of the pontine micturition center, regulates muscle tone. Stimulation of this area (RPRF) suppresses antigravity limb muscle tone (such as REM atonia or locked-in syndrome), external urethral sphincter activities as well as bladder activity. Therefore, the increased anal sphincter tone is considered to be due to destruction of the bilateral pontine reticular formation. In fact, on the MRI images, both sides of RPRF were impaired in our case.

Our patient developed increased anal tone presumably due to bilateral lesion in the pontine micturition/defecation center and the rostral pontine reticular formation.

Disclosure

None of the authors have any conflict of interest relevant to the study.

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