CASE REPORT

Abnormal postural reflexes in a patient with pontine ischaemia

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SUMMARY

The control of body posture is a complex activity that needs a very close relationship between different structures, such as the vestibular system, and the muscle and joint receptors of the neck. Damage of even one of these structures can lead to abnormal postural reflexes. We describe a case of a woman with a left pontine ischaemia who developed a 'dystonic' extensor posture of the left limbs while turned on the right side. This clinical picture differs from previous reports on the subject, and may relate to ischaemic damage of a pontine structure involved in posture control, or of adjacent neural connections to be yet identified. To the best of our knowledge, this is the first case reported in the literature. Clinical examples of an altered interplay between vestibular and neck receptors are rare.

BACKGROUND

Vestibular input, joint and soft tissue mechanoreceptors, and the visual system, cooperate in the regulation of static upright posture. Each component detects and responds to specific postural perturbations, and has specific pathways by which it interacts with other postural reflexes and with higher central nervous system structures.¹

The vestibular system is involved in the control of body posture, and coordination of eyes and head movement. Its primary function is to provide sensory input regarding sustained postural stimulation.² While the semicircular canals relay afferent input on angular acceleration, such as head rotation, the utricle and saccule detect linear accelerations and the position of the head in space. Afferent information, relaying in the vestibular nuclei, projects to the thalamus, superior colliculus, reticular formation, cerebellar flocculus, cerebellar nuclei and spinal cord. Also, the cervical spine is a virtual warehouse of postural afferent input and integration. Several anatomic structures in this region are collectively responsible for maintaining an orthogonal head-on-neck position and for postural control; these include the joint facets and capsules,³ spinal ligaments⁴ and cervical muscles.⁵ The cervical facet mechanoreceptors provide afferent postural input to the trochlear, abducens, spinal trigeminal, central and lateral cervical, and vestibular nuclei,^{6–8} as well as to the cerebellar flocculus, cerebellar nuclei and vermis.

CASE PRESENTATION

An 83-year-old woman was admitted to the emergency department because of severe drowsiness following a reported 'convulsive' episode. She had

suffered from two similar events a few months before. She was on warfarin owing to atrial fibrillation and a related right hemisphere cerebral ischaemia that had occurred in 2007. A cerebral CT scan showed no recent lesion. Thus, the patient was given levetiracetam 250 mg two times a day. The next day, her clinical picture had not improved. A second CT scan was performed, showing a left pontine ischaemia with a hyperdense basilar artery (figure 1). The patient was then admitted to the Neurology ward. There, her neurological examination revealed a Glasgow Coma Scale score of 8 (E3M4V1) with drowsiness and eye opening after oral calling, global aphasia, right hemiplegia and left hemiparesis (the latter being an outcome of the 2007 ischaemic stroke). She also presented with horizontal gaze paralysis and bilateral miosis, which reacted to light poorly. Oculocephalic reflexes were absent. On the subsequent days, we noticed a 'dystonic' posture of the left limbs while the patient was being turned on her right side for nursing manoeuvres. In particular, we observed a consistent extension-abduction of the upper limb, combined with thigh and leg flexion, and plantar flexion-extrarotation of the foot, with exaggerated toe dorsiflexion. This occurred especially when the

Figure 1 CT scan showing a left pontine ischaemia (arrow). The ischaemic lesion was adjacent to the lateral and medial vestibular nuclei, pyramidal tract, nucleus of the seventh cranial nerve and, in part, the medium and superior cerebellar pedunculi. The other snapshots show a chronic cerebral vasculopathy without evidence of any new supratentorial or cerebellar ischaemia.

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patient's trunk was rotated to the right by raising her left shoulder and hip. In this instance, the head and neck followed the rest of the body, since apparently the patient was unable to react differently to her body perturbation. A somewhat similar posture occurred when she was turned on the left, by the same type of manoeuvre (figure 2). In order to investigate the possible role of brainstem structures in the development of this posture, brainstem auditory-evoked potentials were performed but were normal.

OUTCOME AND FOLLOW-UP

During hospitalisation, the patient's condition worsened, since she developed pneumonia and subsequent respiratory distress. Therefore a cerebral MRI or further neurophysiological investigations could not be performed. She was then transferred to another hospital for long-term care.

DISCUSSION

The maintenance and control of posture and movement depends on inputs not only from the labyrinths but also from the neck. In particular, damage to neck muscles in humans may produce gait disturbances, dizziness and nystagmus.^{9 10} Neck muscle perturbation in humans modulates the excitability of motor neurons, innervating postural muscles.^{11 12} The vestibular and neck afferent inputs are involved in the perception of the position and motion of the trunk-in-space.¹³ Hence, the trunk-in-space may be computed by the combination of the head-in-space signal (vestibular) and the head-on-trunk signal (neck proprioceptive). In physiological models, neck rotation holding the head (which eliminates the role of vestibular receptors),¹⁴ implies extensor responses of contralateral and flexor responses of ipsilateral limbs, due to activation of neck mechanoreceptors. The opposite response can be observed during head

rotation holding the neck (figure 3). However, little is known about the neuronal basis of vestibulospinal reflex activity in response to changes of head-on-trunk position.^{15–17} In general, there is strong evidence that neck-related information is processed by the vestibular nuclei.^{14 18} Furthermore, information from the vestibular system and neck mechanoreceptors can converge on the reticulospinal neurons in the medullary reticular formation¹⁹ and on cerebellar structures.^{20 21} Admittedly, signals on trunk-on-head position, derived from neck proprioceptors, can reach the cerebellum along private (direct or indirect) pathways, bypassing the vestibular nuclei. Furthermore, some neurons in both cerebellum and brainstem areas show a weak vestibular sensitivity,²² while in others, the gain of the vestibular response is generally higher than that caused by neck rotation.²³ These different neuronal responses to vestibular and neck proprioceptive stimuli could be involved in the modulation of the complex interplay at basis of vestibulospinal reflexes. Thus, the anatomic-physiological mechanisms of both the physiological and pathological reflexes observed in limbs as a consequence of head rotation holding the neck or neck rotation holding the head are somewhat obscure.

Owing to the progressive worsening of the clinical condition of the patient, this report lacks refined neuroimaging and an exhaustive neurophysiological study, which could have allowed for a more detailed anatomic-physiological explanation of the clinical picture. However, the CT scan performed a few days after the beginning of the symptoms, showed the new left pontine ischaemia while the supratentorial images demonstrated a chronic cerebral vasculopathy without clear evidence of acute ischaemic lesions in the basal ganglia, frontal lobe other structures, that could be involved in the genesis of these abnormal body postures. For these reasons, we suppose that the pontine ischaemia may have caused an alteration in the vestibular/neck

Figure 2 The 'dystonic' extensor posture while turned on the right and left sides.

Combined cervical and vestibular reflexes

Figure 3 Postural reflexes resulting from a combination of cervical and vestibular reflexes.

mechanoreceptors extensor/flexor control, determining a physiologically extensor response of the left limbs while also turning the patient on the right by holding her by the trunk followed by the head and the neck, and also using the right as a unique 'block'. Thus, the patient postural reflexes behaved in a manner somewhat opposite to that observed in the normal animal model.

Brainstem ischaemias are quite frequent clinical events but, very rarely, impairment of postural reflexes are observed in these cases. To the best of our knowledge, this is the first case report approaching the complex interactions between the vestibular and neck inputs in terms of pathological limb reflexes in humans—pointing out a very particular clinical feature in well-defined neurological pictures. Bedside observations are best suited to challenge animal models in this particular neurophysiological field.

Learning points

- The vestibular system as well as muscle and joint receptors of the neck are involved in the control of body posture.
- Brainstem ischaemia can interfere with the connections between neck receptors and vestibular system causing abnormal body postures.
- More animal and human models are needed to fully understand these complex relations.

Competing interests None declared.

Patient consent Obtained.

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REFERENCES

- 1 Morningstar MW, Pettibon BR, Schlappi H, *et al.* Reflex control of the spine and posture: a review of the literature from a chiropractic perspective. *Chiropr Osteopat* 2005;9:13–16.
- 2 Goldberg ME, Hudspeth AJ. The vestibular system. In: Kandel ER, Schwartz JH, Jessell TM, eds. *Principles of neural science*. Vol 4. McGraw-Hill Companies, 2000:514–24.
- 3 Grod JP, Diakow PR. Effect of neck pain on verticality perception: a cohort study. *Arch Phys Med Rehabil* 2002;83:412–15.
- 4 Sjölander P, Johansson H, Djupsjöbacka M. Spinal and supraspinal effects of activity in ligament afferents. *J Electromyogr Kinesiol* 2002;12:167–76.
- 5 Schieppati M, Nardone A, Schmid M. Neck muscle fatigue affects postural control in man. *Neuroscience* 2003;121:277–85.
- 6 McLain RF. Mechanoreceptor endings in human cervical facet joints. *Spine* 1994;19:495–501.
- 7 Yoganandan N, Knowles SA, Maiman DJ, *et al.* Anatomic study of the morphology of human cervical facet joint. *Spine* 2003;28:2317–23.
- 8 Wyke B. Neurology of the cervical spinal joint. *Physiotherapy* 1979;65:72–6.
- 9 Biemond A, De Jong J. On cervical nystagmus and related disorders. *Brain* 1969;92:437–58.
- 10 Cope S, Ryan GMS. Cervical and otolith vertigo. *J Laryngol Otol* 1959;73:113–20.
- 11 Aiello I, Rosati G, Sau GF, *et al.* Interaction of tonic labyrinth and neck reflexes in man. *Ital J Neurol Sci* 1992;13:195–201.
- 12 Rossi A, Mazzocchio R, Nuti D. Tonic neck influences on lower limb extensor motoneurons in man. *Electromyogr Clin Neurophysiol* 1986;26:207–16.
- 13 Mergner T, Huber W, Becker W. Vestibular-neck interaction and transformation of sensory coordinates. *J Vestib Res* 1997;7:347–67.
- 14 Gdowski GT, McCrea RA. Neck proprioceptive inputs to primate vestibular nucleus neurons. *Exp Brain Res* 2000;135:511–26.
- 15 Hlavacka F, Njiokiktjien C. Postural responses evoked by sinusoidal galvanic stimulation of the labyrinth: influence of head position. *Acta Otolaryngol (Stockh)* 1985;99:107–12.
- 16 Fransson PA, Karlberg M, Sterner T, *et al.* Direction of galvanically induced vestibulo-postural responses during active and passive neck torsion. *Acta Otolaryngol (Stockh)* 2000;120:500–3.
- 17 Kennedy PM, Inglis JT. Interaction effects of galvanic vestibular stimulation and head position on the soleus H reflex in humans. *Clin Neurophysiol* 2002;113:1709–14.
- 18 Anastasopoulos D, Mergner T. Canal-neck interaction in vestibular nuclear neurons of the cat. *Exp Brain Res* 1982;46:269–80.
- 19 Pompeiano O, Manzoni D, Srivastava UC, *et al.* Convergence and interaction of neck and macular vestibular inputs on reticulospinal neurons. *Neuroscience* 1984;12:111–28.
- 20 Kotchabhakdi N, Walberg F. Cerebellar afferent projections from the vestibular nuclei in the cat: an experimental study with the method of retrograde axonal transport of horseradish peroxidase. *Exp Brain Res* 1978;31:591–604.
- 21 Precht W, Volkind R, Blanks RH. Functional organization of the vestibular input to the anterior and posterior cerebellar vermis of cat. *Exp Brain Res* 1977;27:143–60.
- 22 Anastasopoulos D, Mergner T, Becker W, *et al.* Sensitivity of external cuneate neurons to neck rotation in three-dimensional space. *Exp Brain Res* 1991;85:565–76.
- 23 Kleine JF, Guan Y, Kipiani E, *et al.* Trunk position influences vestibular responses of fastigial nucleus neurons in the alert monkey. *J Neurophysiol* 2004;91:2090–100.

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