

Neck Rigidity as a Physical Manifestation of Higher Brain Hypo-function/Dysfunction

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Abstract. A total of 308 patients, who received inpatient rehabilitation in department of Neurology in Sapporo City General Hospital, were studied. The patients were divided into four subgroups according to the presence or absence of Parkinsonism and the location of organic neurological lesions: group A consisted of 47 patients with brain lesions who presented with Parkinsonism; group B consisted of 135 patients with manifestations deriving from brain disorders other than Parkinsonism; group C consisted of 68 patients with manifestations deriving from spinal cord, peripheral nerve, and muscle disorders; and group D consisted of 58 patients with non-organic functional disorders and patients with organic lesions not responsible for clinical manifestations. The patients were administered a battery of 5 tests for evaluating frontal lobe and/or higher brain function. In result, our study confirmed higher brain/frontal lobe dysfunction to affect the grade of neck rigidity unless there is an interruption, in the control of peripheral organs, by the brain. Consequently the association between higher brain/frontal lobe dysfunction and the grade of neck rigidity were confirmed. Although neck rigidity may be a minor clinical manifestation, it can provide important clues to brain function.

It is occasionally noticed that stimulation evokes physical responses when various parts of the body in patients with brain dysfunction are manipulated. Some of these responses, such as sucking, snouting, grasping, and palmomental

reflexes, are termed primitive reflexes, and are considered to reflect intellectual or frontal lobe dysfunction (1). In daily clinical practice, it is often noticed that other physical responses are evoked on the same basis as primitive reflexes. We previously reported elbow flexion response (EFR) (2) and impairment in smooth pursuit eye movement (iSPEM) (3) as being two such response. We have also noticed that neck rigidity has the same nature as these responses. In order to test whether this is in fact the case, we performed a prospective clinical study.

Patients and Methods

The study subjects were 308 consecutive patients admitted to the Department of Neurology, Sapporo City General Hospital, and who received inpatient rehabilitation between April 2005 and January 2008. This study started 18 months after our previous study (3) and some patients were common to both studies. These patients were divided into 4 subgroups according to the presence or absence of Parkinsonism and the locations of organic neurological lesions: group A consisted of 47 patients with brain lesions who presented with Parkinsonism; group B consisted of 135 patients with manifestations deriving from brain disorders other than Parkinsonism; group C consisted of 68 patients with manifestations deriving from spinal cord, peripheral nerve, and muscle disorders; and group D consisted of 58 patients with non-organic functional disorders or organic lesions not responsible for clinical manifestations, including paroxysmal attacks and physical pain. The details (Table I) and profile (Table II) of each subgroup are summarized in the tables.

The patients were administered a battery of five tests for evaluating frontal lobe and/or higher brain function: three tests for evaluating primitive reflexes (sucking, snouting, and hand grasp) and two additional tests evaluating frontal lobe and higher brain function (EFR (2) and iSPEM (3)). The results of these five tests were classified as 'positive (+)' or 'negative (-)' on the basis of a consistent standard used throughout the study by the authors, all of whom are registered neurologists. The decision as to whether neck rigidity (both horizontal and vertical movements) was 'positive' or 'negative' was made solely by KS using the consistent standard

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Table I. Details of each group (308 cases).

Group A	47 Cases
Parkinson's disease (syndrome)	26
Cerebrovascular disease with Parkinsonism	12
Degenerative dementia with Parkinsonism	9
Group B	135 Cases
Cerebrovascular disease	80
Spinocerebellar degeneration	20
Multiple sclerosis	10
Brain tumor	6
Encephalitis	6
Leukoencephalopathy	5
Hypoxic encephalopathy	3
Degenerative dementia without Parkinsonism	3
Brain trauma	2
Group C	68 Cases
Peripheral neuropathy	32
Vascular disease (outside brain)	8
Myasthenia gravis	6
Multiple sclerosis (outside brain)	6
Myositis	6
HTLV-1 associated myelopathy	4
Spinal cord tumor	2
Syringomyelia	2
Spinal spondylosis	1
Periodic paralysis	1
Group D	58 Cases
Dizziness or vertigo	22
Epilepsy	14
Orthostatic dysregulation	12
Hysteria	8
Reflex sympathetic dystrophy	6
Muscular back pain	6

manual neurological maneuver independently of other clinical findings. At the same time, a registered occupational therapist examined and judged the patient's intellectual ability and the presence or absence of dementia according to the revised Hasegawa dementia scale (HDS-R; dementia 'positive': score \leq 20; dementia 'negative': score \geq 21) (4). We then tested the relationships between neck rigidity and the six items reflecting higher brain function (namely three primitive reflexes, EFR, iSPEM and dementia) using Fisher's exact probability test (5).

Results

The proportions of patients in each subgroup who tested positive for neck rigidity (both horizontal and vertical), three

Table II. Profile of each group.

	Group A	Group B	Group C	Group D
Number	47	135	68	58
Age (years)	72.2	63.8	45.4	62.8
SD	8.7	15.2	22.6	16.1
Gender F/M (number)	25/22	59/76	26/42	25/33
Duration (years)	4.4	3.6	5.4	7.6
SD	2.1	1.8	1.7	2.6
HDS-R	19.9	20.6	26.3	20.5
SD	5.6	6.8	3.6	6.7

primitive reflexes (sucking, snouting and hand grasping), EFR (2), iSPEM (3), and dementia are shown in Table III. In this table, significant relationships between neck rigidity and all six items are denoted by asterisks. In every group, the frequency of vertical neck rigidity was higher than that of horizontal neck rigidity.

Statistical analysis showed neck rigidity (both horizontal and vertical) to have a significant relationship with dementia but not with the other five items for group A. There was a significant relationship between horizontal neck rigidity and all six items in group B and two out of the six items in group D. There was significant relationship between vertical neck rigidity and three out of the six items in group B and two of the six items in group D. There was no significant relationship between neck rigidity (horizontal or vertical) and any of the six items in group C. Significant relationships between horizontal and vertical neck rigidity were seen in all groups.

Discussion

Primitive reflexes (sucking, snouting and hand grasping) are traditionally considered to be indicators of frontal lobe dysfunction (1, 6, 7), and we have already demonstrated that EFR (2) and iSPEM (3) can indicate higher brain/frontal lobe dysfunction. Neck rigidity sometimes appears by itself and is sometimes accompanied by various degrees of rigidity in other parts of the body (1). Although it appears as a manifestation of meningeal irritation (8), cervical spine lesions (9) or muscle diseases (10, 11), in nearly all cases it is related to extra -pyramidal function (12). Some subgroups of patients with Parkinsonism, such as these with progressive supranuclear palsy and corticobasal degeneration, are characterized by neck rigidity and dementia (6, 13, 14), while some patients with Parkinsonism show neither marked neck rigidity nor dementia. Although neck rigidity is known to be a manifestation of extra -pyramidal dysfunction (15, 16), a relationship between neck rigidity and higher brain/frontal lobe dysfunction has not yet been reported.

In group A, with Parkinsonism features, neck rigidity had a significant relationship with dementia, while showing no

Table III. Proportion (%) of positive results for each test in each group.

	Group A		Group B		Group C		Group D	
	H	V	H	V	H	V	H	V
Sucking	28.3		18.2	***	0		7.4	
Snouting	50.0		47.0	***	17.2		22.0	**
Hand grasping	21.7		18.9	***	1.7		7.4	***
EFR	56.5		47.0	**	22.4		30.9	
iSPEM	56.5	*	34.8	***	5.2		13.2	***
Dementia	52.2	*	40.2	***	6.9		11.8	*
Horizontal neck rigidity	50.0	/	31.1	/	8.6	/	11.8	/
Vertical neck rigidity	84.8	*	51.5	***	31.0	*	33.8	*

Significant difference of each test against horizontal(H)/vertical(V) neck rigidity is shown by asterisks; *:p<0.5, **:p<0.1, ***:p<0.01, /:between the same item. EFR:Elbow flexion response, iSPEM:impairment in smooth pursuit eye movement.

significant relationship with the other five items including primitive reflexes. In contrast, Huber and Paulson reported a significant relationship between primitive reflexes and the severity of Parkinson's disease as indicated by the Hoehn-Yahr scale (7). Although we did not evaluate the severity of Parkinsonism, Huber evaluated neither higher brain function nor muscle rigidity. Our observations show brain dysfunction to be related to neck rigidity in all subgroups, even in the presence of Parkinsonism (group A). We speculate that the discrepancy between their study and ours may have arisen due to this difference in background factors. In group B, without Parkinsonism, the current results confirm significant relationships between neck rigidity (both horizontal and vertical) and all or most of the six items studied, probably because higher brain function tends to manifest in bodily form when there is no interference by Parkinsonian movement disorders. In group C, damage to descending pathways mediating brain commands prevents higher brain function from having an influence on terminal organs. In addition, aging of the brain would have been mild because the patients in this group were relatively young as compared to those in the other groups. These factors might explain why there was no significant relation between neck rigidity (horizontal or vertical) and the six items including dementia in this group. As there were no organic lesions in group D, higher brain/frontal lobe dysfunction might have manifested more clearly in the peripheral organs of these patients than in those of groups A and C.

Dementia and various primitive reflexes are known to be manifestations of higher brain dysfunction, and neck rigidity was confirmed herein also to be associated with higher brain function. Our study confirmed higher brain/frontal lobe dysfunction as affecting the grade of neck rigidity unless there is an interruption in the control of peripheral organs by the brain. The reason for the frequency difference between horizontal and vertical neck rigidity was not elucidated

herein and requires further study. Although neck rigidity may be a minor manifestation of brain dysfunction, it can provide us important clues to brain function. Thus, careful observation of patients is essential. We urge clinicians to closely monitor all phenomena in everyday clinical settings.

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References

- Mestre T and Lang AE: The grasp reflex: a symptom in need of treatment. *Mov Disord* 25: 2479-2485, 2010.
- Sudo K, Matsuyama T, Goto Y, Matsumoto A and Tashiro K: Elbow flexion response as another primitive reflex. *Psychiatry Clin Neurosci* 56: 131-137, 2002.
- Sudo K, Mito Y, Tajima Y, Matsumoto A, Oikawa O and Tashiro K: Smooth pursuit eye movement – a convenient bedside indicator for evaluating frontal lobe and intellectual function. *In Vivo* 24: 795-797, 2010.
- Okumiya K, Matsubayashi K, Wada T, Kimura S, Doi Y and Ozawa T: Effects of exercise on neurobehavioral function in community-dwelling older people more than 75 years of age. *J Am Geriatr Soc* 44: 569-572, 1996.
- Agresti A: A survey of exact inference for contingency tables. *Statistical Science* 7: 131-153, 1992.
- Aries MJ, Debruyne H, Engelborghs S, Le Bastard N, Somers N, Gorissen D, Pickut BA and De Deyn PP: Reversal of head drop after discontinuation of olanzapine in a DLB patient. *Mov Disord* 23: 1760-1762, 2008.
- Huber SJ and Paulson GW: Relationship between primitive reflexes and severity in Parkinson's disease. *J Neurol Neurosurg Psychiatry* 49: 1298-1300, 1986.
- Herbowski L and Kawalec P: Pseudomeningeal syndrome of Dupre associated with cervical discopathy--case report. *Acta Neurochir (Wien)* 152: 329-331, 2010.

- 9 Ohnari H, Sasai K, Akagi S, Iida H, Takanori S and Kato I: Investigation of axial symptoms after cervical laminoplasty, using questionnaire survey. *Spine J* 6: 221-227, 2006.
- 10 Franzén E, Paquette C, Gurfinkel VS, Cordo PJ, Nutt JG and Horak FB: Reduced performance in balance, walking and turning tasks is associated with increased neck tone in Parkinson's disease. *Exp Neurol* 219: 430-438, 2009.
- 11 Flanigan KM, Kerr L, Bromberg MB, Leonard C, Tsuruda J, Zhang P, Gonzalez-Gomez I, Cohn R, Campbell KP and Leppert M: Congenital muscular dystrophy with rigid spine syndrome: a clinical, pathological, radiological, and genetic study. *Ann Neurol* 47: 152-161, 2000.
- 12 Kobayashi M, Ikeda K, Kinoshita M and Iwasaki Y: Amyotrophic lateral sclerosis with supranuclear ophthalmoplegia and rigidity. *Neurol Res* 21: 661-664, 1999.
- 13 Gresty M: Stability of the head in pitch (neck flexion-extension): studies in normal subjects and patients with axial rigidity. *Mov Disord* 4: 233-248, 1989.
- 14 Fukushima-Kudo J, Fukushima K and Tashiro K: Rigidity and dorsiflexion of the neck in progressive supranuclear palsy and the interstitial nucleus of Cajal. *J Neurol Neurosurg Psychiatry* 50: 1197-1203, 1987.
- 15 Vanek Z and Jankovic J: Dystonia in corticobasal degeneration. *Mov Disord* 16: 252-257, 2001.
- 16 Askmark H, Eeg-Olofsson K, Johansson A, Nilsson P, Olsson Y and Aquilonius: Parkinsonism and neck extensor myopathy: a new syndrome or coincidental findings? *Arch Neurol* 58: 232-237, 2001.

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