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PAPER

Axial rotation in Parkinson's disease

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Aims: To investigate the ability of patients with Parkinson's disease to perform a rotation around the longitudinal axis of the body. Three questions were raised. Is body rotation impaired in Parkinson's disease? Is there a level of the kinematic chain from the head to the foot at which the impairment is more severe? Is the deficit related to the general slowness of movement in Parkinson's disease?

Methods: Kinematic data were recorded. The temporal organisation of body rotation during gait initiation was analysed in 10 patients with Parkinson's disease, who were all at an advanced stage of the disease and had all experienced falls and freezing during their daily life, and in five controls. The latency of the onset of the rotation of each segment was measured by taking the onset of the postural phase of step initiation as reference value. Locomotor variables were also analysed.

Results: Body rotation was found to be impaired in patients with Parkinson's disease, as the delay in the onset of the rotation of each segment is greater than that in controls. Moreover, a specific uncoupling in the onset of shoulder and pelvis segment rotation was seen in patients. This impairment of rotation is not related only to the general slowness of movements.

Conclusion: Patients with Parkinson's disease were found to have an impairment of posturo-kinetic coordination and impaired capacity to exert appropriate ground reaction forces to orient the pelvis in space.

Many aspects of motor performance are impaired in people with Parkinson's disease. One aspect that has rarely been investigated is the ability to perform whole-body rotations around the longitudinal axis of the body. Turning in bed is markedly impaired in these people.[1] [2] A reduction in longitudinal spinal rotation in seated subjects during a reaching task has also been observed in Parkinson's disease.[3] Steiger *et al*[4] have indicated difficulty in coordinating the orientation of the superimposed axial segments along the body's spinal axis in patients with Parkinson's disease. The patients also show a marked deficit in performing a whole-body rotation on the spot while standing. Moreover, the ''impairment of turning in bed'' is usually assessed as an item of daily life activities in Unified Parkinson's Disease Rating Scale (UPDRS).[5] This axial disorder has also been described during walking (directional changes, half turns) and is associated with the ''freezing'' phenomenon. When asked to turn on the spot, patients perform the action very slowly and execute the rotation by taking little steps (Vaugoyeau *et al*, unpublished). It is not clear, however, whether the marked impairment of axial rotation is related to a specific deficit in the coordination of the superimposed segments or to a deficit caused by the change in body orientation in space. We can emphasise the role of specific constraints related to postural organisation, which requires coordination between sensory inputs and multijoint outputs. Schieppati *et al*[6] have shown that the stability limits are reduced in Parkinson's disease, which is in agreement with the postural instability described in the late stages of the disease. The static and dynamic postural deficits in patients with Parkinson's disease have been related to an impairment of postural reactions, mainly in the extensor muscles.[7] [8] Another sensory deficit concerns the body's graviceptors, more specifically those that monitor the force exerted by the extensor muscles.[9] [10] As a result, patients rely more on vision and on flexor muscles to regulate balance[11] or locomotion.[12] The deficit in producing dynamic ground reaction forces is also characteristic in Parkinson's disease,[13] especially during initiation of gait and locomotion.[14–17] These deficits, however, are related to posture and

gait in general and do not explain why rotation along the longitudinal axis is more impaired in whole-body rotation than during the initiation of gait.

This study explores the possibility that a deficit in axial body rotation while standing does exist in patients with Parkinson's disease, when performing a task that fulfils two requirements: the task has to be similar to a natural task that is easy to perform and has to involve the whole-body axis from the head to the feet, to see whether the impairment of body axis rotation may more specifically involve a given segmental level. The selected task fulfilling the two criteria was a single step at an angle of 45°, which involved a whole-body reorientation in the same direction (fig 1). Three questions were raised. Is the body rotation that takes place in this task impaired? Is there a level of the kinematical chain at which the impairment is more severe? Is the deficit related to the velocity of the movement?

METHODS
Participants
Ten patients with Parkinson's disease aged 55–70 years (mean 62.2 (SD 5.5) years) and five age-matched controls aged 56–69 years (mean 61.8 (SD 5.4) years) participated in this study after giving their informed consent. The local ethics committee (Cancer Control using Population-based Registries and Biobanks) approved the project.

Table 1 shows the clinical status of the patients. The patient group was functionally homogeneous as assessed by the UPDRS (normality test: K = 0.16, p>0.2). All patients were taking their usual antiparkinsonian drugs and were tested during the pharmacologically ''on'' status. All patients were at an advanced stage of the disease and they had all experienced falls while walking, as well as the freezing phenomenon. The controls did not have any neurological or other diseases, which may affect their postural stability or ability to perform the experimental tasks.

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Abbreviation: UPDRS, Unified Parkinson's Disease Rating Scale

Table 1 Characteristics of the disease in the 10 patients with Parkinson's disease

Characteristic	Patient									
	1	2	3	4	5	6	7	8	9	10
Duration of disease (years)	18	10	10	29	13	19	6	12	8	12
Hoehn–Yahr stage	III	III	III	IV	III	III	III	IV	III	IV
UPDRS part III in the on state	30	19	28	33	22	24	31	35	25	31
UPDRS part III in the off state	45	57	40	65	38	NA	40	48	44	44
Levodopa (mg/day); duration (years)	1750; 14	1000; 8	1000; 8	2200; 25	850; 12	800; 18	400; 5	1350; 10	1200; 8	600; 10
Other drugs, mg/day	Bromocriptin 15	Pergolide 6; Amantadine 300	Bromocriptin 40; Amantadine 300	Pergolide 3; Amantadine 300	Piribedil 100; Segiline 10	Bromocriptin 50; Segiline 10	None			Trihexipphenidyle 4
	Lisuride 0.8; Segiline 10	Bromocriptin 15; Trihexipphenidyle 10; Tolcapone 600								

UPDRS, Unified Parkinson's Disease Rating Scale.

Experimental procedure

The task was analysed by using a multiparametric method combining kinetic and kinematic variables. It consisted of taking a single diagonal step of an angle of 45° and ended when the other leg (trailing leg) was placed parallel to the first moving leg. Participants had to take the diagonal step accompanied by body reorientation in the direction of the step, and then end the task with their body oriented in the 45°-angle direction. A triggering signal was given by a sound emitted by a loudspeaker situated in the required step direction (45°). No instruction was given about the speed of execution. Ten trials were recorded for each participant. Before each trial, they stood erect, unsupported and barefoot on a force platform (AMTI; Advanced Mechanical Technology, Watertown, Massachussetts, USA). Participants were instructed to adopt the same initial posture with the head upright and looking straight ahead, and to take the step with the same leg—the one that they used to initiate walking. All participants but one initiated the step with the right leg.
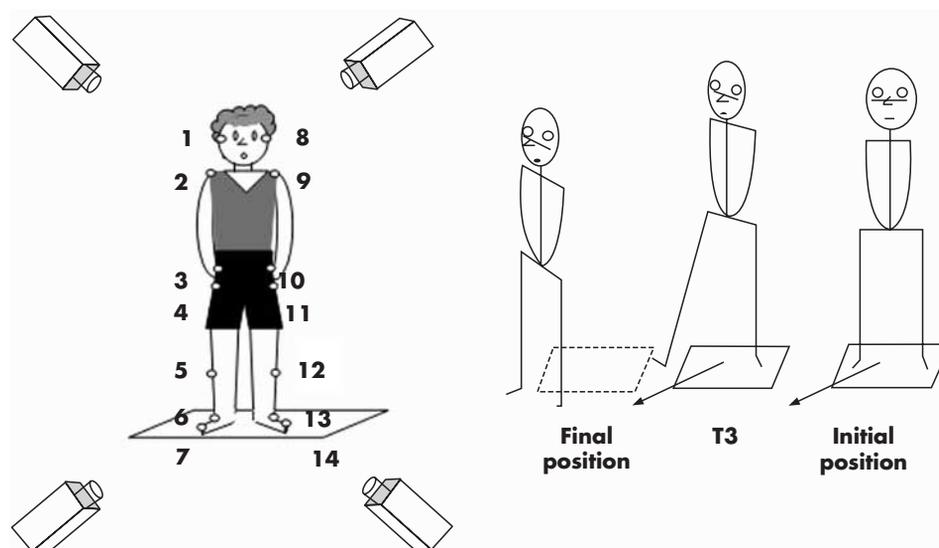
Experimental recordings

The recording duration was 3 s for each trial and horizontal and vertical components of the ground reaction forces were recorded by using a force platform with a sampling frequency of 500 Hz (AMTI). Kinematic analyses were carried out with the ELITE television image-processing system.[18] For this purpose, 14 reflective markers (1 cm in diameter) were placed symmetrically in pairs: at the level of the external angle of the eye orbit, the shoulder acromion, anterosuperior iliac spine, great trochanter, knee external malleolus and fifth metatarsus. Recordings were taken with four infrared cameras and kinematic profiles were monitored at a sampling rate of 100 Hz.

Step variables

For all variables, we analysed the first step—that is, the one performed during body rotation.

On the basis of kinetic and kinematic variables, two successive phases were defined for controls and patients with Parkinson's disease: the postural phase and the movement phase.[16] The postural phase started at T1, with the first variation of the horizontal force in the sagittal plane (Fy), and ended at T2, which corresponded to the onset of deflection of the velocity curve of the malleolus marker of the leading leg (fig 2). The movement phase corresponded to step execution, beginning at T2 and ending at T3, when the first moving leg struck the ground. T3 corresponded to the zero crossing of the velocity curve of the moving malleolus.

Figure 1 Experimental set-up and task. On the left: representation of the subject in the initial position with the location of the 14 markers; on the right: experimental task; single diagonal step with reorientation.
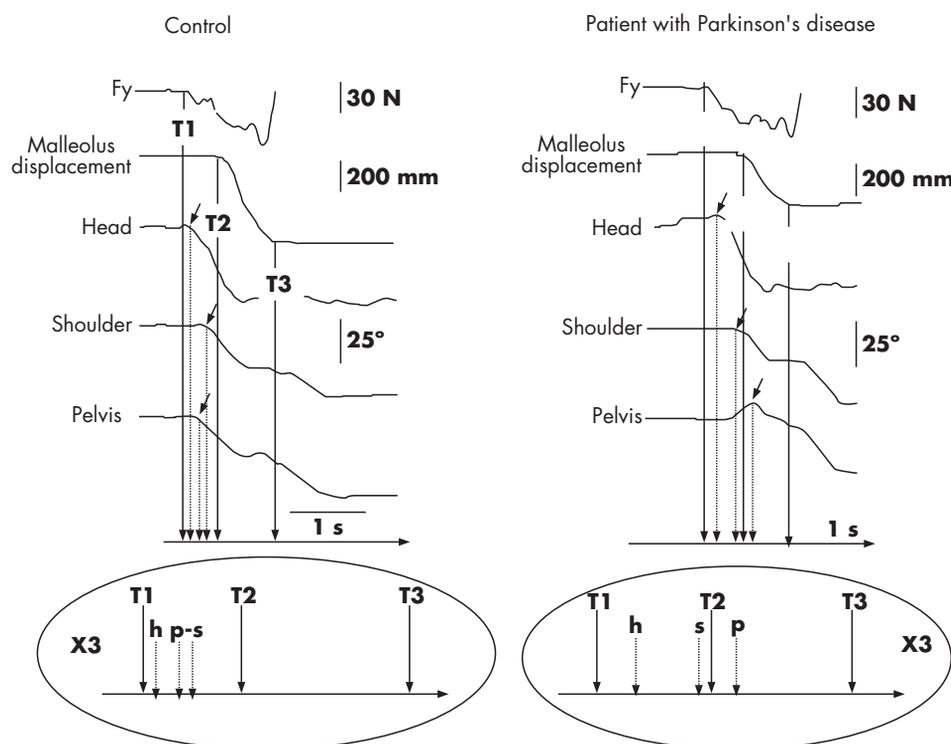
Figure 2 Single trial recording of body rotation: left, control; right, a patient with Parkinson's disease. Top: kinetic (Fy: horizontal forces in the sagittal plane), kinematic (malleolus displacement) and angular data. The horizontal rotation around the vertical axis (yaw), angular variation of the head, the shoulders and the pelvis are shown. The small arrows indicate the onset of rotation of each segment. Bottom: representation of the temporal organisation of the rotation. Controls begin shoulder and pelvis rotation simultaneously with their axial body rotation, whereas patients with Parkinson's disease start shoulder (s) rotation before pelvis (p) rotation. Trunk rotation is uncoupled. T1, start of the postural phase (first variation in the horizontal forces in the sagittal plane); T2, end of the postural phase corresponding to the beginning of the movement phase (first variation in the velocity of the malleolus marker); T3, end of the movement phase (first contact of the foot with the floor); X3, temporal organization enlargement by 3.

Step length was measured in the step direction from the trajectory of the malleolus marker of the first moving leg. To verify this (45°), the angular direction of the step was measured, from the curve of the malleolus marker displacement of the moving leg, as the angle change between the initial anteroposterior direction and the final direction defined by the path of the step.

Body rotation

Whole-body reorientation in the step direction was investigated. The horizontal rotation around the vertical axis (yaw) of three segments was considered—those of the head, the shoulder and the pelvis—then corresponding to the lines between markers 1 and 8 (head), 2 and 9 (shoulders) and 3 and 10 (pelvis).

Angular variables

Two angles for each segment were defined as follows:

1. the angle between the initial position and the position at T3 (that corresponds to the end of the first diagonal step; $\alpha1$ for the head, $\beta1$ for the shoulders and $\gamma1$ for the pelvis);
2. the angle between the initial position and the end of the body rotation ($\alpha2$ for the head, $\beta2$ for the shoulders and $\gamma2$ for the pelvis).

Temporal variables

For each segment, the onset time of the rotation was measured, which corresponded to the onset of angular variation from the initial position in the horizontal plane. The latencies of the onset of rotation of each segment were calculated by taking the first variation in the horizontal forces

in the sagittal plane (T1) initiating the postural phase as a reference. These time intervals were called head (Th), shoulder (Ts) and pelvis (Tp) onset latencies.

Statistical analysis

Data were analysed with the Statistica program Statsoft. Descriptive statistics are reported as mean (SD). The mean of each group corresponds to the average of the mean values of the 10 trials performed for each participant.

Differences between the groups were tested with the Mann–Whitney U test. The comparison between latency of segments in each group was made with the Wilcoxon's signed rank test (for within-subjects comparisons).

One-sample analysis (t test) was used to compare the step trajectory to the 45° required. To determine whether the segmental rotation was simultaneous with the onset of the postural phase, we compared the onset of the latencies of each segmental rotation to 0, by using one-sample analysis (t test). A correlation analysis between the body's kinematic variables and step velocity was carried out in both groups. A coefficient of correlation not significantly different from zero would show that the impairment of rotation would not be related only to the general slowness of movements. We checked that all the data analysed with a t test were normally distributed (Kolmogorov–Smirnoff test).

Differences with $p < 0.05$ were considered to be significant.

RESULTS

To assess how accurately the instructions of step direction in both control and patient groups were followed, original and final orientations of the head were compared with 45° (fig 3). Step direction was successfully carried out. The deviation of the direction of step trajectory from the required 45° was not
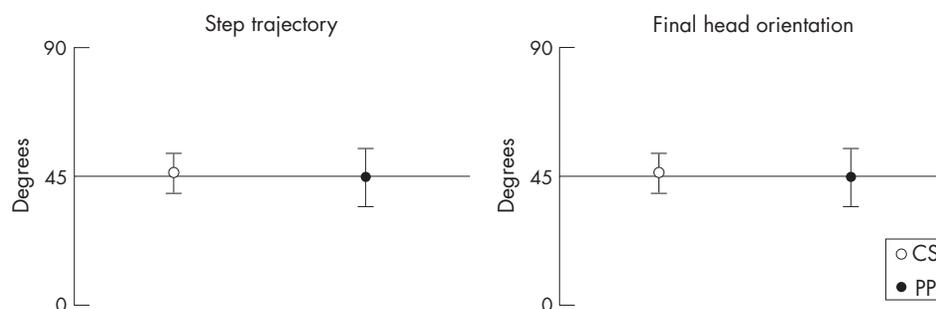
Figure 3 Mean (SD) step trajectory (left) and final head orientation (right) of controls (CS, white) and patients with Parkinson's disease (PP, black).

significantly different for controls (t = 1.8; p>0.05) and patients with Parkinson's disease (t = 0.2; p>0.05), and the final orientation of the head was not significantly different from 45° for controls (t = −0.29; p>0.05) and patients with Parkinson's disease (t = 0.92; p>0.05).

Comparison between groups
Analysis of the step variables
Phase duration, step length, step velocity
The duration of the postural phase was significantly longer in the patients than in the healthy controls (330.30 (78) *v* 239.30 (70.38) s; U = 3.62; p<0.001). The duration of the movement phase was significantly shorter in patients than in controls (787.20 (149.2) *v* 861.7 (110.9) s; U = −2.54; p<0.05) (fig 4).

The step length in the patients with Parkinson's disease group was significantly decreased compared with that in the control group (302.40 (77) *v* 422.30 (8.32) mm; U = −6.67; p<0.001). Step velocity was reduced in patients compared with that in controls (389.5 (94.94) *v* 495 (10.9) mm/s; U = −5.12; p<0.001).

Analysis of body rotation
Figure 5 shows the respective angular rotation amplitudes of the head, the shoulders and the pelvis at T3 (α1, β1, γ 1) and at the end of the task (α2, β2, γ2). Figure 6 shows the latencies from T1 to the onset of rotation for the three segments studied (head, shoulders and pelvis).

Amplitude of rotation of segments
When compared with healthy controls, patients with Parkinson's disease presented with a significant decrease in the amplitude of pelvis rotation at T3 (35° (11.2°) and 38.3° (10.7°); U = 5.15; p<0.05; fig 5). Rotations of the head and

shoulder, however, were similar in both groups at T3 and at the end of the task.

Onset of rotation of segments
For the three segments studied (head, shoulders and pelvis), the latencies of the onset of the rotation from T1 (Th, Ts and Tp, respectively) were significantly longer in patients with Parkinson's disease than in controls: head, U = −3.51; p<0.001; shoulders, U = −2.34; p<0.05; and pelvis, U = −3.65; p<0.001. This indicates a global delay in the onset of body rotation in patients with Parkinson's disease (fig 6).

Comparison within groups
Temporal organisation of body rotation
The comparison between groups showed a significant delay in the onset of rotation of each segment in patients with Parkinson's disease (fig 6). To determine possible changes in the organisation of rotation, the latencies of head, shoulders and pelvis were statistically analysed for both groups.

In controls, the latency of head rotation (Th) was not significantly different from 0, indicating that the onset of the postural phase (T1) and the onset of head rotation (Th) were almost simultaneous. The latencies of both shoulder and pelvis rotation were significantly different from 0 (t = 5.74; p<0.001 and t = 4.08; p<0.01, respectively), indicating a delay with respect to T1. Latency of onset of head rotation (Th) was significantly shorter than those of shoulder (Ts; t = −3.82; p<0.001) and pelvis rotation (Tp; t = 3.49; p<0.001), indicating that the onset of head rotation preceded shoulder and pelvis rotation. We found no significant difference, however, between the latencies of shoulder and pelvis rotation. Therefore, controls initiated body rotation
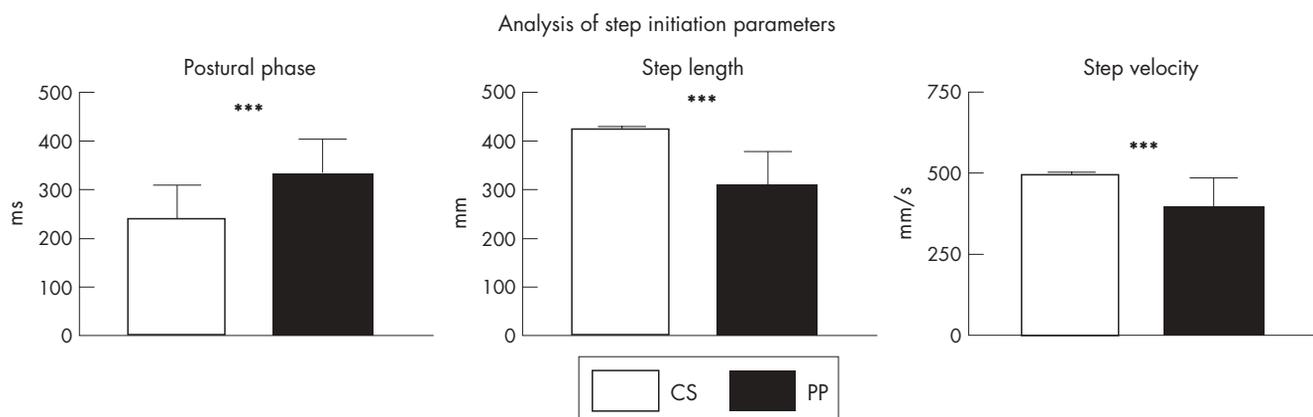
Figure 4 Bar graphs of step initiation variables, comparison between controls (CS, white) and patients with Parkinson's disease (PP, black). Mean (SD) values are given. ***p<0.001.
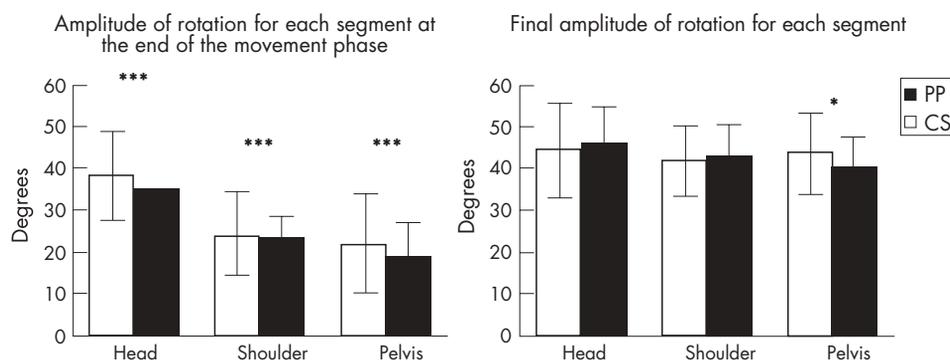
Figure 5 Bar graphs of amplitude of rotation for each segment. Comparison between controls (CS, white) and patients with Parkinson's disease (PP, black). The mean (SD) values are given. *p<0.05 and ***p<0.001.

with the head at the beginning of the postural phase and recruited the shoulders coupled with the pelvis.

In patients with Parkinson's disease, the three latencies (head, shoulders and pelvis) were significantly different from 0 (t = 7.35; p<0.001; t = 16.24; p<0.001 and t = 10.24; p<0.001, respectively). The latency of head rotation was significantly shorter than those of shoulder (Ts; t = 3.57; p<0.001) and pelvis rotation (Tp; t = 4.71; p<0.001). In contrast with controls, the latency of shoulder rotation was significantly shorter than that of pelvis rotation (Tp; t = 3.18; p<0.01). Obviously, patients with Parkinson's disease started their rotation with the head long after the onset of the postural phase, then continued with the rotation of the shoulders and finally with that of the pelvis. The shoulders were not recruited together with the pelvis but in a top–down sequential organisation.

Influence of step velocity on the sequential onset of shoulder and pelvis rotation

To test the influence of step velocity on the pattern of rotation onset, the coefficient of correlation of the delay between the onset of shoulder and pelvis rotation and step velocity was calculated.

In controls, the coefficient of correlation was not significantly different from 0 ($r^2 = 0.07$), indicating that there is no relationship between the step velocity and the timing of rotation onset. In other words, this indicates that step velocity does not influence the timing of the onset of rotation.
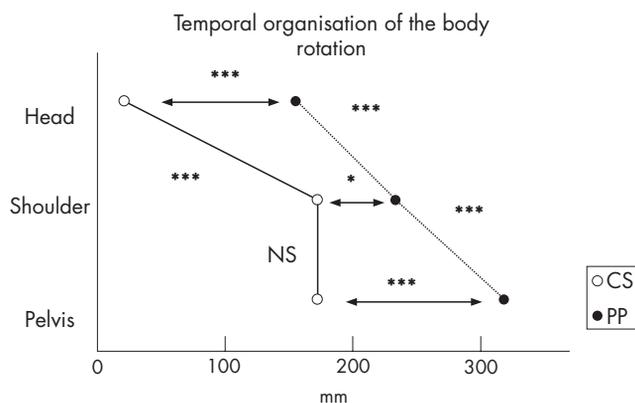
Figure 6 Representation of the temporal organisation of the rotation for patients with Parkinson's disease and controls: comparison between groups and between levels (head, shoulders and pelvis). Horizontal arrows: comparison between groups for a given level. Comparisons between the latencies of each anatomical segment. NS, non-significant; *p<0.05 and ***p<0.001.

In patients with Parkinson's disease, the same analysis indicates that the latency between shoulders and pelvis rotation does not depend on step velocity ($r^2 = 0.1$). Therefore, the uncoupling between onset of shoulder and pelvis rotation is not because of the general slowing of the movement. A set of values with a high delay between shoulder and pelvis, however, is seen only for high-velocity values.

DISCUSSION

The analysis of body rotation in the population with Parkinson's disease shows a specific impairment of the process of axial rotation. The principal results were (1) a decrease in the gait initiation performance, (2) a coupled shoulder and pelvis rotation and (3) a delayed onset of pelvis rotation with respect to the shoulder, suggesting a specific deficit in coordinating shoulder and pelvis rotation, not due to the slowness in the performance of movement in Parkinson's disease.

Deficits of temporal organisation of body rotation in patients with Parkinson's disease

As shown in fig 2, in controls, head rotation was the first event accompanying the onset of the postural phase (T1), which corresponded to the generation of propulsive forces in the sagittal plane. During the onset of the postural phase, the trunk segment (shoulder and pelvis) started its rotation almost simultaneously. Thus, at the onset of the movement phase (T2), all segments had largely initiated their rotations and the rotation process continued progressively during the execution of the diagonal step. At the end of the movement phase (T3), more than half the trunk rotation had been completed and the rotation of the head segment was almost fully achieved. In patients with Parkinson's disease, the onset of rotation of each segment was clearly delayed in comparison with that of controls. The onset of head rotation intervened only at the middle of the postural phase, whereas that of shoulder rotation intervened at the end of this phase, and that of pelvis rotation intervened even later, with the onset of the movement phase.

Slower movements were not significantly different from faster ones, with a coupled rotation of the shoulder and the pelvis. By contrast, the slowest axial rotations observed in patients with Parkinson's disease were performed in a manner similar to those of controls. In patients, the delay between shoulder and pelvis rotation increased with the velocity of rotation, indicating that the delay in pelvis rotation with respect to the shoulder is not due to the slowness of performance of movement in Parkinson's disease but due to an impairment that increases with the velocity of the task.

Mechanisms

What could be specifically impaired in the capacity of patients with Parkinson's disease to exert simultaneous rotation of the upper and lower trunk? A general deficit may be the reduced reliance on proprioceptive inputs to control the motor output[19] and the biased functioning of the segmental circuits related to Ia and Ib afferents.[20–23] The muscles participating in axial rotation are also antigravity muscles, and the impaired functioning of these circuits may be increased compared with that of the flexor muscles.[10]

The main impairment may concern the control of pelvis rotation. The pelvis position in space depends on two control systems. The first is based on a top–down organisation by using the head position in space as a reference frame and calculating the various segment positions in space from the head position. The second is the bottom–up control, which, when a person is standing, regulates the position of the centre of gravity in space as well as to that of the pelvis, where the centre of gravity is located, by using the feet–support interface as a reference frame.[24 25] The bottom–up control would be mainly related to equilibrium maintenance and would be more severely impaired in patients with Parkinson's disease, owing to the reduced capacity to generate ground reaction forces,[14–17] in connection with a biased assessment of the effort exerted by the extensor muscles against gravity.[26] By contrast, the control of head and trunk rotation, which depends on the descending chain, would be less impaired in those with Parkinson's disease, and the onset of shoulder rotation would occur earlier than that of pelvis rotation.

An impairment of coordination between the top–down and bottom–up control can also be put forward to explain the parkinsonian impairment in whole-body rotation. In Parkinson's disease, the difficulty of performing two motor tasks simultaneously has been described for a long time:[27] this phenomenon is well known in clinical practice, with the classic description of a patient being unable to walk and simultaneously to put his wallet into his inside pocket.[28] This underlines the role of the basal ganglia and supplementary motor area output in providing the person with an internal timing cue for movement and postural sequences.[29 30]

Study limitations

These data on a small group of selected patients with advanced Parkinson's disease (Hoehn–Yahr stages III and IV), however, are clearly not applicable to patients at earlier stages of disability without any axial motor impairment during daily living.

Conclusions

The specific impairment of temporal organisation of the axial rotation in patients with Parkinson's disease may reflect two aspects. The first would be related to a general role of the basal ganglia in body orientation in space. The delayed onset of the head rotation in patients with Parkinson's disease as compared with that in controls may be related to the impaired orientation in patients with Parkinson's disease. The second aspect would be a major deficit in coordinating the descending control of body segment orientation starting from the head and acting on the shoulder with the ascending control of the pelvis orientation in space starting from the feet. The difficulty in Parkinson's disease to produce ground reaction forces responsible for positioning of the centre of gravity in space would explain the uncoupling between shoulder and pelvis rotation and the delayed onset of pelvis rotation. This impaired generation of ground reaction forces may be due to a deficit relating to the extensors Ib input, monitoring the muscle effort against gravity.

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PostScript ....

Rostral cingulate motor area and paroxysmal alien hand syndrome

Alien hand syndrome (AHS) is characterised by abnormal motor behaviour of the contralateral upper limb, which is subjectively experienced as involuntary or alien induced.[1] The affected hand often shows a grasp reflex and an instinctive grasp reaction, as well as elements of "magnetic apraxia" associated with frontal lobe damage. The most common frontal type of AHS is repeatedly observed in patients with lesions in the supplementary motor area, anterior cingulate gyrus, medial prefrontal cortex, and anterior corpus callosum. Involuntary uncontrolled movements in AHS usually remain unchanged or improve gradually over periods of varying length. A paroxysmal form of alien hand syndrome has been described very rarely. In these exceptional cases, focal epileptic seizures were suspected to be the pathophysiological substrate for paroxysmal alien limb phenomena.[2 3]

Case report

We report a 61 year old right handed man with a paroxysmal form of alien hand syndrome resulting from an ischaemic lesion within the rostral part of the right cingulate motor area (CMA) (fig 1A). Short episodes with typical spontaneous involuntary movements of his left hand (groping, scratching, grasping) developed suddenly four days before patient's admission to our ward. At that time, normal glycaemia and a transient increase in blood pressure (200/100 mm Hg) were observed. A neurological examination revealed normal findings with the exception of positive grasp reflex on the affected upper limb. Magnetic resonance imaging (MRI) on the same day disclosed circumscribed hyperintense lesion with a 12 mm diameter in the right anterior cingulate cortex. Cerebrospinal fluid examination and an interictal EEG was normal. All "alien hand" episodes started and terminated suddenly; they occurred every 15 minutes and their duration was one to three minutes. The patient was fully conscious during the seizures. He experienced the abnormal motor behaviour of his left hand as involuntary. He was unable to control these hand movements despite great effort. The abnormal movements were repeatedly directed towards external stimuli. A gradual increase in seizure frequency was observed in subsequent days, and discrete myoclonic jerks and slight tonic posturing of the left upper extremity were added to the typical hand automatisms. An FDG-PET investigation showed focal hypermetabolism of the right CMA (corresponding precisely to the structural MRI lesion) and repeated video-EEG monitoring clearly revealed the ictal epileptic mechanism of the "alien hand" seizures (fig 1, panels B and C). A neuropsychological examination revealed no speech problems or problems with bimanual coordination. The paroxysmal complex motor activities of the left upper limb completely disappeared immediately after antiepileptic drug treatment (levetiracetam) was begun.

Control MRI (done after three days, two weeks, and three months) confirmed an ischaemic aetiology of the structural lesion.

Comment

Studies on primates, and increasing evidence in humans, support the notion that the anterior cingulate cortex is strongly involved in the preparation and execution of movements. A specific role may be played by an area that encompasses the ventral bank of the cingulate sulcus and is located just behind and in front of the vertical plane that passes through the anterior commissure (VCA). This region corresponds to the simian rostral cingulate motor area (rCMA); in humans it contains gigantopyramidal neurones which resemble the Betz pyramidal cells of the primary motor cortex. The projections of this area 24c (rCMA) target mainly the rostral portion of the SMA (medial area 6aβ).[4] Interestingly, electrical stimulation of this area, undertaken recently in one epileptic patient, provoked "an irresistible urge to grasp something, …, accompanied by a wandering arm movement contralateral to the stimulation side".[5]

Based on this finding, an engagement of CMA in compulsive goal directed motor behaviour in humans was suggested. Our present observation clearly demonstrated that even limited impairment of the rostral part of the CMA may produce the clinical alien hand syndrome. It is noteworthy that the structural lesion in AHS patients often encroaches on the supplementary motor area and the middle section of the corpus callosum, and it is therefore difficult to define the precise involvement of the anterior cingulate cortex.

Unequivocal proof of the epileptic origin of paroxysmal AHS represents another important aspect of the present case report. To date, only three patients with an epileptogenic lesion within the frontomedial cortex manifesting with paroxysmal alien limb phenomena have been analysed in the literature. In the light of the seriously limited published sources, our findings provide evidence of an underlying epileptic mechanism in paroxysmal AHS. The ictal EEG pattern in our subject was obtained on the third day after his admission, when a slight tonic posturing of the left upper extremity had already been
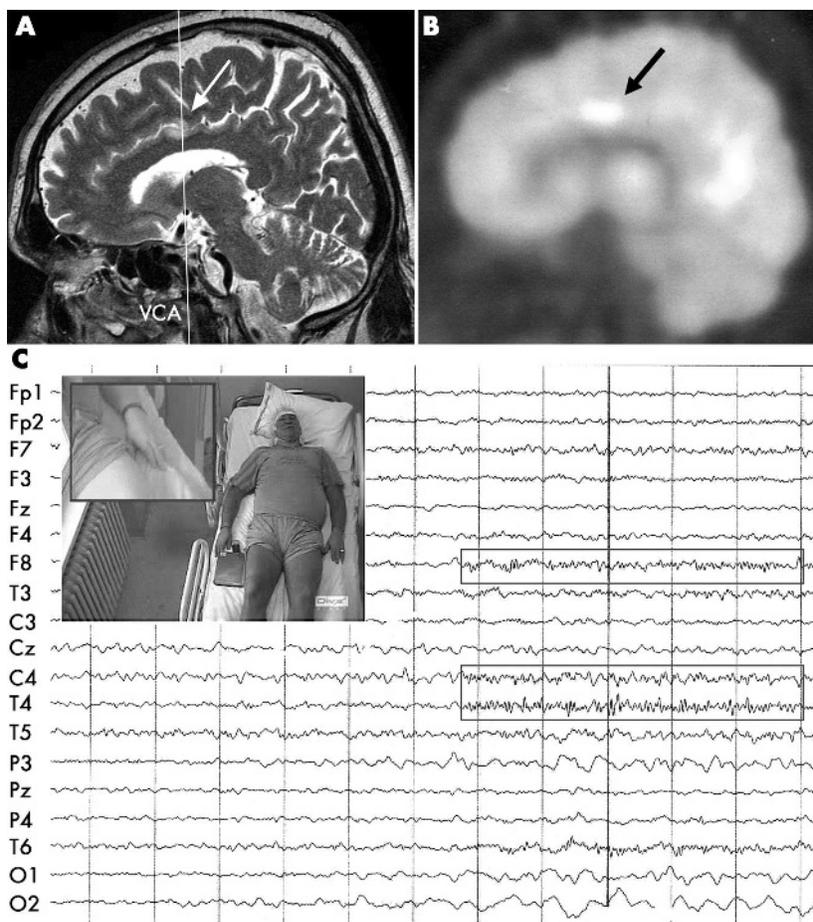
Figure 1 (A) Sagittal T2 weighted magnetic resonance imaging showing the hyperintense lesion in the rostral cingulate motor area (CMA) (day 4); VCA, vertical plane passing through the anterior commissure. (B) FDG-PET scan. (C) Video-EEG trace (ictal pattern (spiking) in the right F-C-T region time locked with an alien hand episode).

added to the typical "alien hand" automatisms (fig 1C). The spreading of the ictal activity from rCMA to premotor and motor cortical areas resulting in the additional tonic phenomena is highly probable. The ictal EEG recordings from the date of the patient's admission with pure ictal "alien hand" signs revealed only very discrete non-specific changes in the central and right paracentral region (Cz and C4). In this case, an underlying ictal activity in the "hidden" lesional/perilesional region is strongly anticipated. But absolute certainty that the rCMA is solely responsible for the paroxysmal "alien hand" syndrome could only be drawn from invasive EEG investigation or electrical cortical stimulation. This treatment was not necessary for clinical reasons and thus was not undertaken in our patient.

In conclusion, rostral CMA very probably plays a crucial role in the production of ictal automatic motor behaviour of the contralateral hand. This finding may further implicate the participation of this part of the caudal anterior cingulate cortex in the genesis of ictal limb automatisms in epileptic patients.

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Atypical micrographia associated with corticostriatal white matter lesions in systemic lupus erythematosus

Micrographia is a heterogeneous condition in which various parts of the CNS may be involved. An anatomical substrate for micrographia, however, remains to be established. Here, we report on a patient with systemic lupus erythematosus (SLE), who presented with atypical micrographia, which was associated with bilateral lesions in the corticostriatal white matter.

Case report

A 30-year-old right-handed woman was diagnosed as having SLE in 2002. She had concomitant leukocytopenia, arthritis, nephritis and high titres of antinuclear antibodies, which satisfied the American Rheumatism Association criteria for SLE. Thereafter, maintenance treatment using corticosteroids was started. She was admitted to the Niigata University Medical and Dental Hospital, Niigata, Japan, in February 2005, because of a high fever and headache with affective incontinence.

Examination showed her muscle strength and the sensory function of her extremities to be normal. She did not have involuntary movements or akinetic–rigid symptoms, as her gait was normal and no rigidity was observed in the neck, body and extremities. No impairment was seen in the rapid alternative movements of her hands. She was well oriented and cooperative. Aphasia was absent; her speech was well articulated and grammatically correct, and she had no difficulty in naming objects. She had no abnormalities in praxis, showing an excellent capacity in imitating and pantomiming, and in using tools with either hand. She could perform a fist–palm-alternating task swiftly. Orofacial apraxia, visuospatial disturbance, unilateral spatial neglect or visual agnosia was not observed. She had no memory impairment. No general intellectual deterioration was seen; her score in the revised Hasegawa Dementia Scale, which is widely used for intellectual screening in Japan, was 28 of 30 (cut-off 20/30).

During the neuropsychological evaluation, the quality of the patient's spontaneous handwriting deteriorated—that is, the characters or drawings were always small. To evaluate this symptom in detail, we asked the patient to write a Japanese character and a Roman alphabet repetitively, and draw a triangle, a circle, a square and a star in a manner identical to the samples given by the examiner. The initial characters of the patient's handwriting were smaller than those in the samples, and the small size of the characters was constant throughout the sequence of her handwriting (fig 1A). Furthermore, a marked initial reduction in size was observed in the symbols drawn compared with the corresponding samples (fig 1B). These copying tasks were carried out using her dominant (right) hand. Neither hesitation nor slowness in handwriting was observed. The patient could correctly evaluate the size of objects, as she could sort various objects on the table by size. She acknowledged the disorder, complaining that she could write only such small characters or symbols despite great efforts to write in the same size as that in the samples.

Examination of CSF showed a normal cell count, although a mild rise was observed in IgG level. The patient tested negative for serum antiphospholipid antibodies. MRI of the brain carried out on day 2 of admission showed hyperintensity signal lesions in the bilateral dorsal part of the striatum and adjacent white matter on T2-weighted and diffusion-weighted images (fig 2A). These lesions were contrast enhanced with gadolinium. The other parts of the CNS, however, were intact. Single-photon emission CT using ethyl cystine dimer labelled with technetium-99m carried out on day 12 of admission showed a decreased perfusion of the bilateral striatum and surrounding white matter.

After treatment with 1000 mg of methylprednisolone for 5 days, the initial reduction in the size of handwriting had improved (fig 1C). MRI of the brain carried out 3 months after the treatment showed that the white matter lesions had reduced in size (fig 2B).

Informed consent was obtained from the patient before carrying out the evaluations and giving treatment.
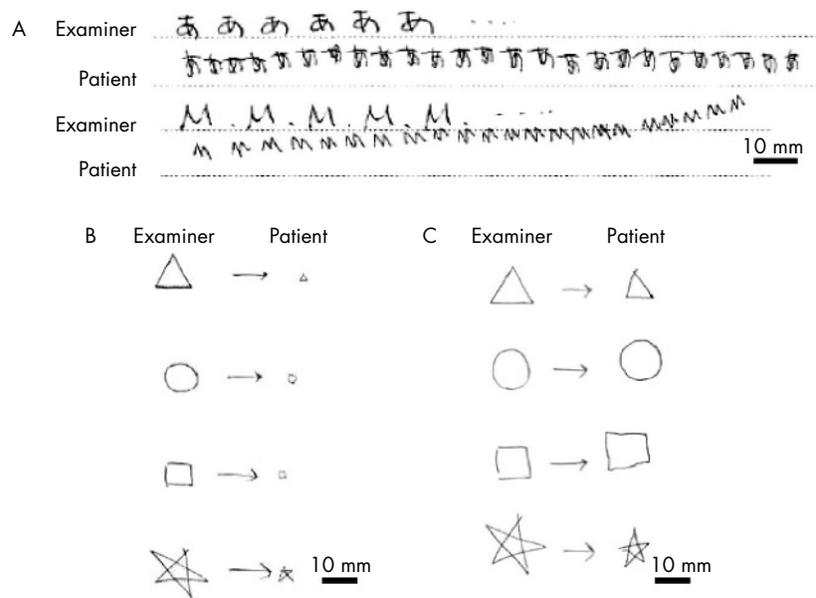
Figure 1 (A) Repetitive writing of a Japanese character and a Roman alphabet was requested. The sizes of the characters and letters in the patient's handwriting are smaller than those in the examiner's handwriting, and this small size is constant throughout the handwriting sequence. (B) Before the steroid pulse treatment, a marked initial reduction in size is observed on drawing a triangle, a circle, a square and a star. (C) The initial reduction in size is obviously improved after the steroid pulse treatment.