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Diagnostics of disturbed upright body orientation in pusher behaviour

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List of abbreviations

SCP Scale for Contraversive Pushing

BLS Burke Lateropulsion Scale

SPV Subjective postural vertical

Publication list

Journals with peer-review process

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Introduction

Pusher behaviour is characterized by an active shift of the centre of gravity towards the paretic body side; patients are thought to orient their body towards a disturbed inner reference of verticality. Pusher behaviour is very relevant in stroke rehabilitation since it hampers and prolongs the rehabilitation process [1, 2].

Patients with pusher behaviour typically push themselves away from their non-paretic body side and resist any attempt to transfer weight over the non-paretic side [3]. To increase the lateral body tilt or resistance against correction, patients show abduction or extension of their non-paretic arm and/or leg. Originally, this behaviour was referred to as pusher *syndrome* since it was observed in combination with neuropsychological symptoms, such as anosognosia, neglect or aphasia [3]. Subsequent studies found, however, no support for a syndrome [1, 4, 5]. Nonetheless there is a high prevalence of neglect or aphasia in patients with pusher behaviour [6, 7], which might be due to the close anatomical proximity of the brain structures representing the control of upright body orientation to those typically affected in patients with aphasia or spatial neglect [8, 9]. Stroke is the most frequent aetiology of pusher behaviour [2], but also few non-stroke patients showing pusher behaviour have been described [10]. In stroke patients, the behaviour is typically associated with lesions of the posterior thalamus, but also with lesions in extra-thalamic areas, such as the insular cortex, the postcentral gyrus, the middle temporal gyrus, and the inferior parietal lobule [7-9, 11-13].

There is large variation in the data reported on the frequency of pusher behaviour in stroke patients, ranging from 4.3% to 65% [1, 2, 6, 7, 10, 14-16]. The variability is caused by mainly two reasons: on the one hand, the reported study populations differed in their patient characteristics (e.g. time post stroke, severity of motor impairment, age), on the other hand, diagnostic criteria used for the classification of pusher behaviour varied considerably. While some studies used non-validated clinical diagnosis based on criteria reported by Davies [3] [1, 6, 7], other studies applied clinical scales with variable cut-off scores [2, 14-16]. Inconsistent diagnostic criteria are indeed a major issue in the research on pusher behaviour, resulting in a large uncertainty not only about the epidemiology, but also about prognostic factors, involved brain areas, and the time course of recovery from pusher behaviour. This thesis focuses on clinical examination tools relevant for the diagnostics of pusher behaviour. Study 1 compares the classification of pusher behaviour based on the two most frequently used clinical scales. Study 2 and 3 address the assessment of perceived upright body orientation during standing.

Classification of pusher behaviour using clinical scales

The Scale for Contraversive Pushing (SCP) and the Burke Lateropulsion Scale (BLS) are the two most widely used clinical scales for the diagnosis of pusher behaviour. Both scales reflect criteria set out by Davies [3], but show great variations in the selection of items and the scoring. The SCP rates the degree of postural symmetry, the presence of abduction or extension of the non-paretic extremities, and the presence of resistance to passive correction. Each of these components is tested in sitting and standing position, yielding a score between 0 and 2 per component. Originally, a cut-off score ≥1 for each component (sitting plus standing) was recommended for the diagnosis of pusher behaviour [5]. Few years later, a modified cut-off score (>0 per component) was evaluated [15]. The modified cut-off showed better diagnostic accuracy and was recommended as a less conservative alternative to the original cut-off score [15].

The BLS is less commonly used than the SCP. The scale assesses the patient's resistance to passive supine rolling, to passive postural correction when sitting and standing, and to assistance during transferring and walking [17]. The BLS is the only scale that incorporates pusher behaviour during walking. The severity of resistance is rated on a scale from 0 to 3 (0 to 4 for standing) for each item. The cut-off score which is usually used for the diagnosis of pusher behaviour is ≥2 points [18].

Both the SCP and the BLS were assumed to be reliable and valid measures for pusher behaviour with good clinical and research practicability [18]. However, the scales are differently constructed, evaluate different postures, and use different scoring. These differences may result in inconsistent classification of pusher behaviour. Yet, consistent measures to identify and follow up pusher behaviour are the prerequisite for studying the epidemiology, the underlying mechanism, prognosis, and effectiveness of therapies. Due to the need for homogenous classification, the objective of study 1 was to directly compare the classification of pusher behaviour based on the SCP and the BLS in a cohort of stroke patients with and without pusher behaviour. The clinical scales were assessed before and after three different therapeutic interventions by the same examiner. In addition to the clinical scales, standardized frontal photographs were taken to analyse postural responses and compare them to the items of the clinical scales. Diagnosis of pusher behaviour based on the SCP and the BLS showed moderate agreement. In all cases with inconsistent classification, the BLS diagnosed pusher behaviour, but the SCP did not. Patients with inconsistent classification showed mild or resolving pusher behaviour, which was primarily present during standing and/or walking. Thus, the BLS was found to be more sensitive in detecting pusher behaviour and especially useful to do so for mild or resolving pusher behaviour. In addition, the BLS was more responsive to small changes in the behaviour. Summing up, the BLS allows a more differentiated and graduated evaluation of pusher behaviour due to the wider range in its scoring. The scoring reflects the progress most patients make during rehabilitation. Though, the BLS cut-off ≥2 lacks validation. A cut-off >2 instead of ≥2 resulted in an improved agreement between the two scales in our study sample. Up to date, there is no gold standard for the diagnosis of pusher behaviour. Consequently, other criteria typically disturbed in patients with pusher behaviour, such as postural abnormalities or perceived upright body orientation should be used for the validation of cut-off values. Postural abnormalities characteristic of patients with pusher behaviour are: a lateral turn and shift of the head toward the ipsilesional side, a markedly shortened distance between the ipsilesional shoulder and the neck, and a shortening of the ipsilesional trunk with an elongation of the contralesional side [19]. Additionally, patients with pusher behaviour typically show a constant ipsilesional tilt of the non-paretic leg with respect to the trunk during slow passive body tilt in the frontal plane [20]. This abnormal postural response and the abnormal spontaneous posture might be driven by a disturbed inner representation of upright body orientation in relation to gravity. The following two sections focus on the assessment of the perceived upright body orientation during standing and its investigation in patients with pusher behaviour.

Assessment of the perceived upright body orientation during standing

The assessment of the perceived upright body orientation in relation to gravity is referred to as the subjective postural vertical (SPV). For SPV assessment, the subject is passively tilted in space and has to identify the position that he or she felt his body adjusted to the gravitational vertical. The SPV is typically measured blindfolded. Two previous studies assessed the SPV during sitting in the frontal plane in patients with pusher behaviour [5, 21]. Both studies found a considerable deviation of the SPV; however, results were contradictory with regard to the side of the deviation. One study found the SPV to be tilted about 18° to the ipsilesional side [5]. In contrast to that, the other study reported a tilt of similar magnitude to the contralesional side [21]. Both studies assessed the SPV during sitting, but they used slightly different experimental setups. The latter study used a non-motorized wheel device with the patient's head and legs restrained and the feet in contact with the ground [21]. The other study used a motor driven chair without any fixation of the head and legs [5]. The legs were hanging freely. Differences in the vestibular and somatosensory input, and restriction of spontaneous postural responses might thus explain the contradictory results of the two studies. Both studies additionally assessed visual verticality perception. Based on their respective results, the authors proposed different models to explain how the disturbed postural verticality perception leads to pusher behaviour. Karnath et al. [5] found a mismatch between an undisturbed visual verticality perception and an ipsilesionally tilted SPV, and suggested that patients actively try to compensate for this mismatch by pushing their longitudinal body axis toward the contralesional side. Additionally, they discussed that pusher behaviour might be a secondary response to the patients' unexpected experience that they lose lateral balance when trying to get up and orient the body subjectively upright. In contrast, Pérennou et al. [21] found a transmodal tilt of the visual vertical and the postural vertical to the contralesional side. They suggested that patients with pusher behaviour try to align their body with the contralesionally tilted reference of verticality.

Depending on its severity, pusher behaviour can be present in different postures, such as lying, sitting and standing, during posture transitions and/or during walking. In its severe form, pusher behaviour is present during sitting and standing (and possibly also during lying). In a less severe form or during recovery, pusher behaviour persists during standing and/or walking, but is absent during sitting. The assumption that patients with pusher behaviour orient their body towards an erroneous SPV (alignment or compensation) suggests that the internal reference of verticality is represented differently during sitting and standing. Thus, for patients who show deficient body orientation primarily during standing, the SPV during sitting might be unsuitable to detect their deficit. Therefore, it would seem especially relevant to assess the SPV of patients with mild pusher behaviour during standing, since this posture is primarily affected. The assessment of the SPV during standing in patients with pusher behaviour was subject of study 3 and is described in the next section of this thesis. Due to the lack of assessment methods, we first needed to set up an entirely new paradigm allowing the assessment of the SPV during standing. This was implemented and evaluated in study 2. In the study we determined the reliability of the SPV measurements during standing and provided normative data for healthy subjects. The test-retest reliability and the interrater reliability were evaluated for SPV measurements in the frontal and sagittal planes. Subsequently, normative values from healthy subjects aged 20 to 79 years were collected. Normative data are needed to detect and define abnormal or pathological SPV estimation in patients. In addition to the SPV error (tilt), which was calculated by averaging the six trials which were performed for SPV measurement, the SPV range was of interest. The SPV range represents the uncertainty in verticality estimation and was calculated as the difference between the maximum and the minimum values of the six trials performed. Based on the SPV error, ranges of normality for the SPV during standing were defined for the frontal plane and the sagittal plane respectively. A secondary objective of study 2 was to investigate age-related differences of the SPV during standing, since age-related changes have been reported for the SPV during sitting [22]. Similar to sitting, a backward shift of the SPV with increasing age and an increasing uncertainty in SPV estimation was found. The latter might be the result of an age-related decline of vestibular and somatosensory functions.

Perceived upright body orientation during standing in patients with pusher behaviour

Since study 2 showed that perceived upright body orientation during standing can be reliably and precisely assessed in healthy subjects using the new paradigm, we applied the same paradigm in patients with pusher behaviour. In study 3 we compared the SPV of patients with various degrees of pusher behaviour to the SPV of stroke patients without pusher behaviour, and the SPV of age-matched healthy controls. Knowledge about the SPV in patients with different levels of pusher behaviour seems very relevant for a better understanding of the mechanism leading to pusher behaviour and the time course of recovery. Finally, it might help to design specific and effective treatment approaches for patients with pusher behaviour. Pusher behaviour is considered to be a disorder that primarily affects the frontal plane. Consequently, the SPV of patients with pusher behaviour has only been investigated in the frontal plane so far. However, patients often also exhibit a posterior element to their pusher behaviour [23]. Thus, the SPV in study 3 was assessed in both the frontal plane and the sagittal plane. We found an ipsilesional SPV tilt during standing in patients with pusher behaviour, which decreased with decreasing severity of pusher behaviour. Although there was no abnormal SPV tilt in the sagittal plane, patients with pusher behaviour showed a considerably large uncertainty in verticality estimation in both planes. This indicates a general loss of sensitivity for verticality perception in space. In the study we used the BLS for classification of pusher behaviour since we have found it to be most sensitive (study 1). Accordingly, study 3 also allowed validating the BLS cut-off ≥2, which lacked validation so far. Generally, the finding that misrepresentation of body orientation is still present, even though signs of pusher behaviour are mild and primarily present in standing or walking, confirms that the BLS can be used as a valid tool to detect pusher behaviour. Interestingly, all patients with a BLS score of 2 points showed SPV tilts in the frontal plane within the ranges of normality (defined in study 2). This suggests changing the BLS cut-off to >2 instead of ≥2 for the classification of pusher behaviour, as discussed in study 1.

Rehabilitation and future directions

The reduced orientation sensitivity and the ipsilesional bias of the perceived upright body orientation in patients with pusher behaviour emphasise the need for specific rehabilitation approaches to recalibrate the impaired inner representation of verticality. So far, rehabilitation approaches for pusher behaviour for the most part focused on different forms of feedback training, that means training of postural control strategies by using visual, auditory or somatosensory cues [24-26]. A requirement of feedback training is an unimpaired orientation perception of the modality in which the feedback is provided, for example unaffected perception of visual input for visual feedback training. Though, patients with pusher behaviour typically show a large variability in the perception of the visual vertical, indicating a decreased sensitivity for visual verticality perception. Overall, evidence on the effectiveness of feedback training in patients with pusher behaviour is so far insufficient. Another approach which was used in the treatment of pusher behaviour is galvanic vestibular stimulation. Galvanic vestibular stimulation directly affects verticality perception: verticality perception shifts towards the anode during stimulation [27]. However, studies investigating the effect of galvanic vestibular stimulation in patients with pusher behaviour found only a small and unsatisfactory effect [2, 28]. In a recent study, we investigated the influence of galvanic vestibular stimulation on different methods to assess verticality perception (the subjective visual vertical, the subjective haptic vertical, and the SPV) both during and after its application [29][1]. We found that galvanic stimulation has a reversed effect on verticality perception after its application, i.e. a shift toward the cathode. So far, studies that applied galvanic vestibular stimulation in patients with pusher behaviour placed the anode over the

[1] Jeannine Bergmann is first author of this article. Together with the joint first author, she designed the study, recruited the patients, collected data, performed data analyses and interpretation of the data and wrote the article. The study is subject of another dissertation and consequently not included in the present dissertation.

ipsilesional mastoid and the cathode over the contralesional mastoid, focusing on the anodal shift of verticality perception during stimulation. The aftereffect of galvanic vestibular stimulation needs further investigation, especially its time course; however, our finding of a reversed effect after the stimulation suggests reconsidering the placement of the electrodes in future studies. Additionally, we found the SPV to be only little affected by galvanic vestibular stimulation. Assuming that pusher behaviour is correlated with a disturbed SPV, galvanic vestibular stimulation might be inappropriate to effectively affect pusher behaviour. While vestibular input seems relatively unimportant for postural verticality perception, the somatosensory input plays a major role [30, 31]. Therefore, appropriate somatosensory stimulation might be more promising than vestibular stimulation to treat pusher behaviour. In a pilot study, we compared the immediate effects of a single session of robot assisted gait training, galvanic vestibular stimulation, and conventional physiotherapy using visual feedback on pusher behaviour [32][2]. After a session of robot assisted gait training, patients showed a significant reduction in pusher behaviour compared to conventional physiotherapy. Currently, we are investigating the effectiveness of repeated robot assisted gait training on pusher behaviour in a randomised controlled trial. An interim analysis showed a larger reduction of pusher behaviour in the intervention group (two weeks of daily robot assisted gait training) compared to the control group (two weeks of conventional physiotherapy) [33]. Robot assisted gait training forces the control of upright body orientation for an extended period of time and simultaneously enhances somatosensory input during locomotion. This seems to be effective in permanently reducing pusher behaviour, possibly by recalibrating the disturbed postural verticality perception.

Future work is needed to investigate the correlation between disturbed perceived upright body orientation and pusher behaviour in more detail, especially during the rehabilitation process. The BLS and the SPV during standing seem to be useful diagnostic measures to do so.

[2] Jeannine Bergmann is co-author of this article. She organized the study, collected data, contributed to data interpretation and revised the article. The study is subject of another dissertation and consequently not included in the present dissertation.

Summary/ Zusammenfassung

Summary

Pusher behaviour reflects a severe disturbance of body orientation in space. Patients are thought to orient their body towards an erroneous internal reference of verticality. Although pusher behaviour has been increasingly studied over the last years, there is still a large amount of uncertainty about its epidemiology, underlying mechanism, prognostic factors, and effective treatment. One reason for the variable findings in previous studies might be the inhomogeneous diagnostic criteria. This thesis focuses on clinical examination tools relevant for the diagnostics of pusher behaviour following stroke: clinical scales (study 1) and the perceived upright body orientation during standing (study 2 and 3).

Study 1 directly compared the classification of pusher behaviour based on the two most frequently used clinical scales: the Scale for Contraversive Pushing (SCP) and the Burke Lateropulsion Scale (BLS). Results showed inconsistency in the classification between the two scales. The BLS was more sensitive in the classification of pusher behaviour and more responsive to small changes than the SCP. Thus, the BLS is especially useful to detect mild or resolving pusher behaviour in standing or walking.

Another diagnostic measure which is relevant in pusher behaviour is the perceived upright body orientation, which can be assessed by the subjective postural vertical (SPV). So far, SPV assessment in patients with pusher behaviour showed contradictory results and were only performed during sitting. Pusher behaviour can, however, be present in different postures, such as sitting and standing. In its severe form, it affects both sitting and standing posture, in a less severe form or during recovery, the behaviour persists during standing, but is absent during sitting. The assumption that patients with pusher behaviour orient their body towards an erroneous SPV suggests that the internal reference of verticality is represented differently during sitting and standing. Consequently, it would seem especially relevant to assess the SPV of patients with mild pusher behaviour during standing, since this posture is primarily affected. Therefore, we set up a paradigm to measure the SPV during standing in

the frontal and the sagittal planes. This new paradigm was evaluated in study 2. The test-retest and the interrater reliabilities were determined and normative data for healthy subjects provided. The study showed that SPV assessment in standing can be performed with reliable and precise results. Ranges of normality were defined. In study 3, the SPV during standing was assessed in stroke patients with and without pusher behaviour and in a healthy control group in the frontal and the sagittal planes. We included patients with different degrees of severity of pusher behaviour. The BLS was used for the classification, due to its greater sensitivity in detecting mild pusher behaviour (study 1). Study 3 revealed that patients with pusher behaviour had an ipsilesional SPV tilt during standing, which decreased with decreasing severity of pusher behaviour. Moreover, patients with pusher behaviour showed a large uncertainty in verticality estimation in both the sagittal and the frontal planes, indicating a generally disturbed sensitivity for verticality perception in space. The finding that misrepresentation of body orientation is still present, even though signs of pusher behaviour are mild, confirms that the BLS can be used as a valid tool to detect pusher behaviour. Study 3 also revealed that all patients with a BLS score of 2 points showed SPV errors in the frontal plane within the ranges of normality. This supports changing the BLS cut-off to >2 instead of using ≥2 to classify pusher behaviour.

Zusammenfassung

Patienten mit Pushersymptomatik haben eine schwere Körperorientierungsstörung. Es wird vermutet, dass sie ihren Körper an einer verkippten inneren Vertikalenreferenz ausrichten. Obschon die Pushersymptomatik in den letzten Jahren zunehmend untersucht wurde, gibt es immer noch große Unsicherheit hinsichtlich der Epidemiologie, der zugrundeliegenden Mechanismen, der prognostischen Faktoren und einer effektiven Behandlung der Störung. Ein Grund für die variablen Ergebnisse der Studien ist die Verwendung von inhomogenen diagnostischen Kriterien. Die vorliegende Arbeit befasst sich mit wichtigen diagnostischen Methoden in der Diagnostik der Pushersymptomatik nach Schlaganfall. Studie 1 untersucht die Klassifizierung der Pushersymptomatik basierend auf den zwei am häufigsten verwendeten klinischen Skalen, Studie 2 und 3 die Erhebung der subjektiven posturalen Vertikale (SPV) im Stand.

In Studie 1 wurden gleichzeitig die Skala für kontraversive Pushersymptomatik (SCP) und die Burke Lateropulsions Skala (BLS) in einer Gruppe von Schlaganfallpatienten mit und ohne Pushersymptomatik erhoben. Der Vergleich zeigte Unstimmigkeiten zwischen den zwei Skalen. Dabei war die BLS sensitiver in der Klassifizierung und zudem responsiver für kleine Veränderungen.

Neben den klinischen Skalen ist die Bestimmung der posturalen Vertikalenwahrnehmung mittels SPV eine wichtige diagnostische Methode bei Patienten mit Pushersymptomatik. Bisherige Untersuchungen der SPV bei Patienten mit Pushersymptomatik ergaben widersprüchliche Resultate bezüglich der Richtung der Verkippung und wurden ausschließlich im Sitzen durchgeführt. Die Pushersymptomatik kann verschiede Körperpositionen, wie beispielsweise den Sitz oder den Stand, beeinträchtigen. Bei einer schweren Ausprägung sind sowohl der Sitz wie auch der Stand betroffen, bei einer milden Ausprägung oder bei einer Verbesserung der Sympomatik, halten die Symptome im Stehen an, zeigen sich aber nicht mehr im Sitz. Unter der Annahme, dass Patienten mit Pushersymptomatik ihren Körper an einer verschobenen Vertikalenreferenz orientieren, scheint es besonders bei Patienten mit einer milden Form der Pushersymptomatik wichtig,

die SPV im Stand zu messen, da diese Position primär betroffen ist. Daher haben wir ein diagnostisches Verfahren entwickelt, um die SPV im Stand in der frontalen und sagittalen Ebene zu messen. Das neue Verfahren wurde in Studie 2 evaluiert. Es wurden die Test-Retest-Reliabilität und die Interrater-Reliabilität sowie Normdaten für gesunde Erwachsene erhoben. Nachdem Studie 2 gezeigt hatte, dass die SPV im Stehen reliabel und präzise gemessen werden kann, wurde sie in Studie 3 bei Schlaganfallpatienten mit und ohne Pushersymptomatik und einer gesunden Kontrollgruppe jeweils in der frontalen und der sagittalen Ebene erhoben. Es wurden Patienten mit unterschiedlichem Schweregrad der Pushersymptomatik in die Studie eingeschlossen. Zur Klassifikation wurde die BLS verwendet, da diese geeignet ist, um auch eine milde Pushersymptomatik zu erkennen (Studie 1). In Studie 3 wurde eine ipsiläsionale Verkippung der SPV in der Frontalebene gefunden, die mit abnehmendem Schweregrad der Pushersymptomatik kleiner wurde. Die Patienten mit Pushersymptomatik zeigten zudem eine auffallend große Unsicherheit der Vertikalenwahrnehmung in beiden Ebenen, was auf eine generell beeinträchtigte Sensitivität der Vertikalenwahrnehmung im Raum hindeutet. Das Ergebnis, dass auch Patienten mit milder Pushersymptomatik eine Störung der posturalen Vertikalenwahrnehmung haben, bekräftigt die Validität der BLS. Zudem zeigte Studie 3, dass bei allen Patienten mit einer Punktzahl von 2 auf der BLS die SPV im Normbereich lag. Dies spricht dafür, den BLS Cut-off Wert bei >2 und nicht bei ≥2 zu setzen, um Patienten mit Pushersymptomatik zuverlässig zu klassifizieren.

Original articles

Bergmann J, Krewer C, Rieß K, Müller F, Koenig E, Jahn K. Inconsistent classification of pusher behaviour in stroke patients: a direct comparison of the Scale for Contraversive Pushing and the Burke Lateropulsion Scale. Clinical Rehabilitation. 2014; 28:696-703. (Study 1)

Article

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Inconsistent classification of pusher behaviour in stroke patients: a direct comparison of the Scale for Contraversive Pushing and the Burke Lateropulsion Scale

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Abstract

Objective: To compare the classification of two clinical scales for assessing pusher behaviour in a cohort of stroke patients.
Design: Observational case-control study.
Setting: Inpatient stroke rehabilitation unit.
Subjects: A sample of 23 patients with hemiparesis due to a unilateral stroke (1.6 ± 0.7 months post stroke).
Methods: Immediately before and after three different interventions, the Scale for Contraversive Pushing and the Burke Lateropulsion Scale were applied in a standardized procedure.
Results: The diagnosis of pusher behaviour on the basis of the Scale for Contraversive Pushing and the Burke Lateropulsion Scale differed significantly ($\chi 2 = 54.260$, $p < 0.001$) resulting in inconsistent classifications in 31 of 138 cases. Changes immediately after the interventions were more often detected by the Burke Lateropulsion Scales than by the Scale for Contraversive Pushing ($\chi 2 = 19.148$, $p < 0.001$). All cases with inconsistent classifications showed no pusher behaviour on the Scale for Contraversive Pushing, but pusher behaviour on the Burke Lateropulsion Scale. 64.5% (20 of 31) of them scored on the Burke Lateropulsion Scale on the standing and walking items only.
Conclusions: The Burke Lateropulsion Scale is an appropriate alternative to the widely used Scale for Contraversive Pushing to follow-up patients with pusher behaviour (PB); it might be more sensitive to detect mild pusher behaviour in standing and walking.

Keywords

Stroke, pusher syndrome, Scale for Contraversive Pushing, Burke Lateropulsion Scale

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Introduction

Pusher behaviour is characterized by an active lateral tilt of the body and resistance to passive correction of the tilted posture.[1] Patients with pusher behaviour show an erroneous internal reference of verticality.[2,3] This leads to a shift of the centre of gravity toward the paretic side and can result in loss of balance and falls.[4,5]

Pusher behaviour is very relevant in stroke rehabilitation, because it prolongs inpatient treatment.[6] However, there is much uncertainty about its prevalence, what may be due to heterogeneous diagnostic criteria.[7-9]

Clinical scales have been proposed for the diagnosis of pusher behaviour, e.g. the Scale for Contraversive Pushing and the Burke Lateropulsion Scale. Both scales reflect criteria set out by Davies[1] to distinguish patients with pusher behaviour; however, classification based on these scales might be inconsistent, for they show great variations in the selection of items and the scoring. The Scale for Contraversive Pushing rates the degree of postural symmetry, the presence of abduction or extension of the non-paretic extremities, and the presence of resistance to passive correction. The Burke Lateropulsion Scale assesses the degree of action or reaction of the patients to keep or change a position. It is the only scale that incorporates pusher behaviour in supine rolling and in walking. The clinimetric properties and the clinical applicability of the two scales were recently reviewed by Babyar et al.[10] While the Scale for Contraversive Pushing is more extensively evaluated, the Burke Lateropulsion Scale also shows evidence of clinical and research practicability.[7,11,12]

Consistent measures are urgently needed to identify and follow-up pusher behaviour. They are a prerequisite for studying the epidemiology, the underlying mechanisms, prognostic factors, and the effectiveness of therapies.

The aim of our study was to compare the classifications of pusher behaviour based on the Scale for Contraversive Pushing and the Burke Lateropulsion Scale in the same sample of stroke patients.

Methods

The present study is a secondary analysis of a cross-over study on the effects of different therapeutic interventions on pusher behaviour. The methods and primary results of the study were reported in detail elsewhere.[13]

Patients

Patients with hemiparesis due to a unilateral hemispheric stroke were enrolled in the study. Additional inclusion criteria were age ≥18 years, inability to stand unassisted, but previous ability to walk independently before stroke. Exclusion criteria due to therapeutic interventions were body weight above 150 kg, body height below 1.60 meters and above 1.90 meters, unstable cardiac disease, metal implants, brain tumour, meningitis, epilepsy, vestibular disorders, eye muscle paralysis, neurodegenerative movement disorder, unstable fracture, severe osteoporosis, contractures or spasticity of the lower extremities.

The Ethics Committee of the Ludwig-Maximilians University Munich approved the study in accordance with the Declaration of Helsinki. Written informed consent was given by all patients or their legal representatives.

Assessments and procedure

The Scale for Contraversive Pushing includes three components: (1) the symmetry of spontaneous body posture (rated with 0, 0.25, 0.75, or 1 point), (2) the use of non-paretic extremities (0, 0.5, or 1 point), and (3) the resistance to passive correction of the tilted posture (0 or 1 point).[3,14] Each component is tested in sitting and standing position, yielding a maximum score of 2 per component. For a diagnosis of pusher behaviour all three components must be present. Karnath et al.[3] originally recommended a cut-off score equal to or greater than one (cut-off ≥1) for each component (sitting plus standing). A less conservative cut-off score greater than zero (cut-off >0) for each component was evaluated by Baccini et al.[7,12], who found improved diagnostic accuracy.

The Burke Lateropulsion Scale assesses the patient's resistance to passive supine rolling, to passive postural correction when sitting and standing, and to assistance during transferring and walking.[11] The score for each item is rated on a scale from 0 to 3 (0 to 4 for standing) and is based on the severity of resistance or the tilt angle when the patients starts to resist the passive movement. The cut-off for the diagnosis of pusher behaviour is ≥2 points.[10]

Standardized frontal photographs were made to study postural responses and compare them to the items of the clinical scales. Head, trunk, and leg orientation were measured in three positions: spontaneous sitting on the physiotherapist's bench with feet having ground contact, spontaneous sitting with legs hanging freely, and standing. Detailed instruction can be found in the supplementary material appendix.

Patients in the study underwent three different therapeutic interventions in a pseudo-random order over 1 week. Immediately before and after each therapy, the Scale for Contraversive Pushing and the Burke Lateropulsion Scale were assessed by the same blinded and trained examiner, and standardized photographs were taken. The following assessment sequence was defined to apply the measures in a single procedure: assisted transfer from the wheelchair to the therapy bench toward the non-paretic side (relevant for the Scale for Contraversive Pushing, B-sitting and the Burke Lateropulsion Scale, transfer), supine rolling (Burke Lateropulsion Scale, supine), sitting on the bench with the feet having ground contact and the knees at a 90° angle (Scale for Contraversive Pushing, A-sitting and photograph), passive correction of the body position (Scale for Contraversive Pushing, C-sitting), sitting on the bench without feet having ground contact and hands in the lap (photograph), passive tilting to the paretic and non-paretic side (Burke Lateropulsion Scale, sitting), assisted standing (Scale for Contraversive Pushing, A-standing and photograph), assisted standing with passive tilting and correction (Scale for Contraversive Pushing, C-standing and Burke Lateropulsion Scale, standing), assisted walking (Burke Lateropulsion Scale, walking) and transfer via stance and toward the paretic side back into the wheelchair (Scale for Contraversive Pushing, B-walking).

Statistics

The chi-square test and Cohen's kappa coefficient (k) were calculated for the classification of pusher behaviour and the detection of changes to estimate the agreement between the Scale for Contraversive Pushing and the Burke Lateropulsion Scale.

For comparisons of the leg, trunk, and head position between groups, ANOVAs were performed and posthoc Bonferroni tests were applied.

Data were analyzed with the statistical package IBM SPSS Statistics 19. The statistical α-level was set at 0.05.

Results

Twenty-three patients with unilateral hemispheric stroke were enrolled in the study (mean age 68 ± 10 years; 6 females; 19 right brain hemisphere damaged; 1.6 ± 0.7 months post stroke). Ten of them were classified as pushers by the Scale for Contraversive Pushing at first study visit (Scale for Contraversive Pushing score 3.25 ± 2.00 (median ± interquartile range), Burke Lateropulsion Scale score 7.5 ± 4.0). Immediately before and after each of the three therapeutic interventions the data was assessed, resulting in a total of 138 data sets.

The diagnoses of pusher behaviour based on the Scale for Contraversive Pushing (cut-off >0) and the Burke Lateropulsion Scale are shown in Table 1 ($\chi 2(1) = 54.260$, $p < 0.001$; $k = 0.564$, $SE = 0.062$). They resulted in an inconsistent classification for 31 data sets, which originated from nine patients.

A comparison of the original and the modified Scale for Contraversive Pushing cut-off scores revealed that the original cut-off (≥1) missed pusher behaviour in two cases compared to the modified cut-off (>0). For further analysis, the cut-off >0 was used.

The Scale for Contraversive Pushing was taken as reference standard to calculate the sensitivity and specificity of the Burke Lateropulsion Scale, which resulted in 100% and 67%, respectively.

Changes were estimated as difference between the scores immediately before and after a therapeutic intervention. The number of detected changes is shown in Table 2 and significantly differed between the Scale for Contraversive Pushing and the Burke Lateropulsion Scale ($\chi 2(1) = 19.148$, $p < 0.001$) and showed moderate agreement ($k = 0.500$, $SE = 0.103$).

Table 1. Classification of pusher behavior based on the Scale for Contraversive Pushing and the Burke Lateropulsion Scale.

Burke Lateropulsion Scale	Scale for Contraversive Pushing (cut-off >0)		
	Pusher behaviour	No pusher behaviour	Total
Pusher behaviour	44	31	75
No pusher behaviour	0	63	63
Total	44	94	138

Table 2. Changes of pusher behaviour detected on the Scale for Contraversive Pushing and the Burke Lateropulsion Scale.

Burke Lateropulsion Scale	Scale for Contraversive Pushing		
	Change	No change	Total
Change	16	13	29
No change	3	37	40
Total	19	50	69

According to the classifications based on the Scale for Contraversive Pushing and the Burke Lateropulsion Scale, data sets were divided into a group with consistently positive diagnosis of pusher behaviour ($PB^{+/+}$), a group with inconsistent diagnosis of pusher behaviour ($PB^{-/+}$), and a group with a consistently negative diagnosis ($PB^{-/-}$).

All 31 cases of $PB^{-/+}$ were classified as pushers on the Burke Lateropulsion Scale, but not on the Scale for Contraversive Pushing. For these cases the item scores were examined. $PB^{-/+}$ showed signs of pusher behaviour mostly in the standing items: 27 of 31 cases showed no points on the Scale for Contraversive Pushing components in sitting and 25 cases no points on the Burke Lateropulsion Scale sitting items. In standing 23 of 31 cases scored on the Scale for Contraversive Pushing component A (symmetry of body posture), 13 cases on the component C (resistance to correction), but only three cases on the component B (use of non-paretic extremities).

In seven of 31 cases no points were scored on the Scale for Contraversive Pushing, neither in sitting nor in standing, however all of them, except one case, scored only on the Burke Lateropulsion Scale standing and walking items. Regarding the entire $PB^{-/+}$ group, even 20 cases scored on the Burke Lateropulsion Scale on the standing and walking items only.

The values of the head, trunk, and non-paretic leg positions determined by photographs, and the results of the ANOVAs and the posthoc comparisons are shown in Table 3.

Discussion

The Scale for Contraversive Pushing and the Burke Lateropulsion Scale showed moderate agreement in the diagnosis of pusher behaviour with higher sensitivity but lower specificity for the Burke Lateropulsion Scale in comparison to the Scale for Contraversive Pushing. The scales resulted in inconsistent classifications in patients with mild or resolving pusher behaviour. In these patients the Burke Lateropulsion Scale might be especially useful to detect pusher behaviour in standing and walking.

The Scale for Contraversive Pushing and the Burke Lateropulsion Scale resulted in 22.5% of cases in inconsistent classifications. In all these cases, the Burke Lateropulsion Scale diagnosed pusher behaviour but the Scale for Contraversive Pushing did not.

Table 3. Mean values (±SD) of head, trunk and leg orientation in spontaneous sitting and standing position.

Group	Sitting with feet having ground contact			Sitting without feet having ground contact			Standing		
	Head	Trunk	Leg	Head	Trunk	Leg	Head	Trunk	Leg
$PB^{+/+}$ (°)	−0.3 ± 4.5	−4.2 ± 8.9	−7.6 ± 8.0	−1.9 ± 4.5	−5.7 ± 9.6	−4.8 ± 6.5	−1.7 ± 6.0	−2.4 ± 6.8	4.6 ± 4.7
$PB^{-/-}$ (°)	−0.5 ± 5.2	−1.2 ± 5.3	−8.9 ± 7.1	−0.4 ± 5.2	−1.6 ± 4.7^{**a}	−3.1 ± 5.6	0.2 ± 5.6	1.8 ± 5.6^{**a}	−1.2 ± 5.8^{**a}
$PB^{-/+}$ (°)	1.2 ± 4.7	1.7 ± 5.9^{**a}	−13.9 ± 6.5^{**a}, ^{**b}	1.4 ± 4.2^{*a}	1.3 ± 4.8^{**a}	−6.1 ± 5.2	1.1 ± 4.6	3.5 ± 5.8^{**a}	2.2 ± 5.2^{*b}
F	1.367	7.032	7.285	4.395	10.469	2.986	2.674	9.959	15.874
p	0.258	0.001	0.001	0.014	<0.001	0.054	0.073	<0.001	<0.001

$PB^{+/+}$, pusher behaviour on the Scale for Contraversive Pushing and pusher behaviour on the Burke Lateropulsion Scale; $PB^{-/-}$, no pusher behaviour on the Scale for Contraversive Pushing and no pusher behaviour on the Burke Lateroupulsion Scale; $PB^{-/+}$, no pusher behaviour on the Scale for Contraversive Pushing but pusher behaviour on the Burke Lateropulsion Scale.

Post-hoc comparisons: * $p < 0.05$, ** $p < 0.01$, [a] compared to $PB^{+/+}$, [b] compared to $PB^{-/-}$.

Positive values indicated an ipsiversive tilt relative to the earth-vertical, i.e. a tilt to the side of the brain lesion; negative values, a contraversive tilt, i.e., a tilt to the unaffected side of the brain.

When the original cut-off criterion of the Scale for Contraversive Pushing was used, which has been suggested by Karnath et al.[3], two more cases were inconsistently classified. Baccini et al.[12] found an excellent agreement between the cut-off >0 and the clinical diagnosis of pusher behaviour, whereas the original cut-off failed to detect pusher behaviour in patients with slight symptoms. Even with the cut-off >0, all signs described by Davies[1] must be present for the diagnosis of pusher behaviour. Consequently, we recommend the use of the cut-off >0 and refer to it in the following discussion.

Since there is no gold standard for the diagnosis of pusher behaviour and proof of validity was not carried out by an expert rating, we calculated the sensitivity and the specificity of the Burke Lateropulsion Scale compared to the Scale for Contraversive Pushing. The Burke Lateropulsion Scale has a higher sensitivity but a lower specificity than the Scale for Contraversive Pushing for detecting pusher behaviour and might produce more false-negative diagnoses.

We also found the Burke Lateropulsion Scale to be more responsive to small changes than the Scale for Contraversive Pushing. This supports the suggestion of Babyar et al.[10] that the Burke Lateropulsion Scale might be more useful for monitoring patients with pusher behaviour as well as for assessing small changes in their status. The clinical relevance of the detected changes is not yet clear; however, small improvements are important for the rehabilitation process and might facilitate the mobilisation and therapy of the patients. In a recent study, Clark et al.[15] showed that the Burke Lateropulsion Scale can be used to monitor progress and recovery during rehabilitation. The wider range of the Burke Lateropulsion Scale allows a more differentiated and graduated evaluation of pusher behaviour; the scale can be used to grade the severity of pusher behaviour across the full continuum of scores and reflects the progress most patients make during rehabilitation.[10]

The cases inconsistently classified by the Burke Lateropulsion Scale and the Scale for Contraversive Pushing ($PB^{-/+}$), showed signs of pusher behaviour mainly in the standing but not in the sitting items.

64.5% of them scored on the Burke Lateropulsion Scale on the standing and walking items only. These two items seem to be crucial for the inconsistent classification between the scales. While walking is not included in the Scale for Contraversive Pushing, both scales address resistance in standing. However, the Scale for Contraversive Pushing rates resistance to an upright position, whereas the Burke Lateropulsion Scale additionally determines resistance to moving the patient 10 degrees past midline. Resistance past midline is only measured in standing and scored with one point. Thus standing is the only item rated on a scale from 0 to 4. The authors established this weighting to emphasise features thought to be most characteristic of pusher behaviour.[11]

Another important difference between the two scales, is that the Burke Lateropulsion Scale rates exclusively resistance to passive correction through a larger variety of postures (lying, sitting, standing, transferring, and walking), while the Scale for Contraversive Pushing addresses resistance in only one component in sitting and standing, respectively. On the Burke Lateropulsion Scale resistance is scored on a scale from 0 to 3 (0 to 4 for standing), on the Scale for Contraversive Pushing, however, according to the 'all-or-nothing' principle, with either 1 point (resistance is shown) or 0 points (resistance is not shown).

As mentioned, the walking item of the Burke Lateropulsion Scale is very relevant for the inconsistent classification of the two scales. However, we observed some difficulties in the assessment of this item. All patients included in this study were not able to stand unassisted and most of them needed either a lot of assistance to walk or were not able to walk at all. Consequently, the walking item was very difficult for severely impaired patients to do and for the examiner to rate. The authors of the scale recommended that, if it is not possible to assess patients in standing or walking due to marked lateropulsion they should be scored as having maximum deficit for those tasks that could not be tested.[11] However, it was not always evident during the assessment of the scales in our study, if standing and walking were impossible due to the severity of lateropulsion or due to other impairments.

Like Clark et al.[15] we also had problems in detecting small body tilts or determining the degree of tilt in the sitting and standing items of the Burke Lateropulsion Scale. In our study, the examiner was on the paretic side and assisted the patient while assessing the scales. It might be useful to have the examiner in front of the patient to judge deviation from verticality and responses of the trunk or the limbs. However, at the same time, the examiner has to move the patient and feel the potential resistance against the movement. Standardized photographs of the patient in a frontal view might help identifying body tilts that the examiner has difficulties detecting while sitting or standing on the patient's side.

When we compared the item scores and the body positions determined by photographs the following was evident: patients of $PB^{-/+}$ scored only three times on the Scale for Contraversive Pushing component B in standing, but more than 74% scored on the component A, i.e., abduction of the non-paretic leg was rarely observed, but in many cases a contraversive body tilt. In contrast, data of the photographs revealed abduction of the non-paretic leg and an average slightly ipsiversively tilted trunk position. This indicates that, although these patients were able to bring their upper body to an upright position or even past midline, they were unable to place their centre of gravity over the base of support in standing. Also in sitting, photographs revealed a noticeable tilt of the non-paretic leg in the $PB^{-/+}$ group. Despite the clinical scales detected no pusher behaviour, the postural responses of these patients seem not completely recovered. Further research is needed to improve the understanding of the mechanism behind pusher behaviour and its recovery process.

Summing up, the Burke Lateropulsion Scale seems to be an appropriate alternative to the widely used Scale for Contraversive Pushing and especially useful to detect patients with very mild pusher behaviour and to track small changes in the behaviour. However, until now, there are no data available on sensitivity, specificity, and internal consistency of the scale. Concurrent validity was estimated by correlating the lateropulsion score with the Fugl-Meyer Balance score and the FIM motor score.[11] Patients are thought to show pusher behaviour when scoring two or greater on the scale, but this cut-off value has not been validated to our knowledge and is inconsistently used in the literature. Babyar et al.[10,16] applied the cut-off ≥2, while Clark et al.[15] in a recent study used the cut-off >2. In our sample, a cut-off value >2 instead of ≥2 improves the agreement between the scales from 77.5% (107 of 138) to 85.5% (118 of 138). As there is no gold standard for the diagnosis of pusher behaviour, a validation of the cut-off score against postural abnormalities or the subjective postural vertical might be meaningful.

There are some limitations to this study, including the small number of patients ($n = 23$). However, this number is comparable to other studies investigating patients with pusher behaviour and the total number of analyzed data sets is quite high as six measurements per patient were included.[3,12] At the same time, the repeated measurement design could be a limitation of the study, since each measurement was analyzed as independent measure for comparison of classifications, what might have biased the result. We performed another chi-square test for comparison of classification including only the data at first study visit and the test was highly significant ($p < 0.001$). Thus, the repeated measurements do not seem to significantly distort our results.

A limitation with regard to the photographs is the dependency on the angulation of the focal point of the camera. We tried to minimise this bias by using a standardised protocol. Furthermore, body orientation was only determined in the frontal plane and deviations in the horizontal or sagittal plane were not taken into account.

Clinical messages

- The Burke Lateropulsion Scale is more responsive to small changes and more sensitive in the classification of pusher behaviour than the Scale for Contraversive Pushing.
- The Burke Lateropulsion Scale is especially useful to detect mild or resolving pusher behaviour in standing and walking.

Contributors

JB recruited the patients, collected the data, performed data analysis and interpretation of data and wrote the paper. CK designing the study, performed acquisition and interpretation of data and revised the paper. KR designed the study, performed acquisition of data and revised the paper. FM, EK and KJ contributed to the conception of the study and the interpretation of data analysis and revised the paper.

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Conflict of interest

The authors declare that there is no conflict of interest.

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The subjective postural vertical in standing: Reliability and normative data for healthy subjects

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Abstract Impaired verticality perception can cause falls, or even the inability to stand, due to lateropulsion or retropulsion. The internal estimate of verticality can be assessed through the subjective visual, haptic, or postural vertical (SPV). The SPV reflects impaired upright body orientation, but has primarily been assessed in sitting position. The internal representations of body orientation might be different between sitting and standing, mainly because of differences in somatosensory input for the estimation of SPV. To test the SPV during standing, we set up a paradigm using a device that allows movement in three dimensions (the Spacecurl). This study focused on the test–retest and interrater reliabilities of SPV measurements (n = 25) and provides normative values for the age range 20–79 years (n = 60; 10 healthy subjects per decade). The test–retest and interrater reliabilities for SPV measurements in standing subjects were good. The normality values ranged from –1.7° to 2.3° in the sagittal plane, and from –1.6° to 1.2° in the frontal plane. Minor alterations occurred with aging: SPV shifted backward with increasing age, and the variability of verticality estimates increased. Assessment of SPV in standing can be done with reliable results. SPV should next be used to test patients with an impaired sense of verticality, to determine its diagnostic value in comparison to established tools.

Keywords Spatial cognition · Motor control · Aging

The human sense of verticality is constructed and updated by integrating vestibular, somatosensory, and visual inputs (Barra et al., 2010). Verticality perception is impaired in different neurological disorders. Its disturbance—for example, in stroke—causes latero- or retropulsion and falls, both of which are major challenges for patient neurorehabilitation (Karnath & Broetz, 2003; Manckoundia, Mourey, Pérennou, & Pfitzenmeyer, 2008; Pérennou et al., 2008).

Different methods have been used to assess verticality perception: the subjective visual vertical (SVV; i.e., adjusting a bar that is visually compared with the gravitational vertical), the subjective haptic vertical (SHV; adjusting a bar to the gravitational vertical without visual control), and the subjective postural vertical (SPV; adjusting the body to the gravitational vertical). Most likely, SVV, SHV, and SPV test different but overlapping aspects of verticality control and yield complementary information (Pérennou et al., 2014). The SVV is the measure investigated most often. It is frequently used in the diagnosis of vestibular disorders, but is poorly correlated with postural impairment (Bonan et al., 2007; Karnath, Ferber, & Dichgans, 2000; Pérennou et al., 2008). The SPV is altered in subjects with deficits of upright body orientation, both in the frontal and sagittal planes—for example, in subjects with pusher behavior or retropulsion after hemispheric lesions (Karnath et al., 2000; Manckoundia, Mourey, Pfitzenmeyer, Van Hoecke, & Pérennou, 2007; Pérennou et al., 2008). Furthermore, the SPV is influenced by the aging process: With increasing age, the SPV shifts backward and body alignments are less accurate (Barbieri, Gissot, & Pérennou, 2010). These age-related changes might be the consequences of a decline in sensory function (Manckoundia et al., 2008).

So far, SPV measurements have been mainly made with subjects in a sitting position, by using various motor-driven

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machines (e.g., Bisdorff, Wolsley, Anastasopoulos, Bronstein, & Gresty, 1996; Karnath et al., 2000) or a nonmotorized paradigm, the so-called wheel paradigm (Pérennou, 2006). We hypothesized that the internal representations of body orientation might be different between sitting and standing, mainly because of differences in somatosensory inputs. Somatosensory inputs—that is, contact, proprioceptive, and visceral cues—play a major role in verticality perception (Bronstein, 1999). In sitting, several contact cues from the chair are available—for example, pressure cues on the back, under the buttocks, and on the back sides of the legs. These cues are not present during standing. Instead, upright stance involves pressure cues from the soles under the feet and somatosensory feedback from the ankle joints. Although sensory input from the lower extremities seems relatively unimportant for SPV estimation in sitting (Mazibrada et al., 2008), both contact and proprioceptive input might significantly contribute to the SPV in standing.

SPV assessment during standing might be especially relevant for postural disorders primarily affecting the standing posture. Several authors have reported that pusher behavior in its severe form is expressed in both sitting and standing positions. In a less severe form, or when the patient has progressed during rehabilitation, pusher behavior is no longer present in sitting, but continues in standing position (Babyar, Peterson, Bohannon, Pérennou, & Reding, 2009; Bergmann et al., 2014; Premoselli, Cesana, & Cerri, 2001). Considering that patients with pusher behavior attempt to align their body with an erroneous SPV (Pérennou et al., 2008), this suggests that the internal reference of verticality is represented differently during sitting and standing. Thus, for patients with deficient body orientation in standing, SPV assessment in sitting might not be able to detect the deficit. That is why we set up a paradigm to measure the SPV during standing using the Spacecurl. The Spacecurl is a cardanic suspension apparatus that so far has been used as a therapeutic approach for patients with neuropathy (Lauenroth, Knipping, & Schwesig, 2012) or back pain (Müller, Schwesig, Leuchte, & Riede, 2001). The purpose of this study was to investigate the reliability and normative values of SPV during standing using this paradigm. Healthy subjects were examined, and values in the sagittal and frontal planes were collected. A secondary objective was to investigate age-related differences in SPV during standing.

Method

Subjects

The reliability of SPV measurements was determined in 25 healthy subjects (age 34.4 ± 9.7 years [mean ± standard deviation], 19 to 56 years [range]; 15 females, 10 males). In addition, the normative values were collected from 60 healthy subjects aged 20 to 79 years (ten subjects per decade). Exclusion criteria were acute cardiac disease, arterial aneurism, thrombosis, unstable spinal column, neuroses/psychoses, advanced pregnancy; body height <145 cm and >195 cm, and body weight >150 kg. Subjects had to be free of any vestibular or balance deficit. Subjects >50 years of age underwent a neurological examination, including test of pallesthesia and a head-impulse test for vestibular function. The study was approved by the Ethics Committee of Ludwig-Maximilians University (LMU) Munich in accordance with the Declaration of Helsinki. All participants gave their written informed consent.

Apparatus and experimental procedure

The Spacecurl (Physio Boerse, Wittlich, Germany) is a cardanic suspension apparatus consisting of three concentric rings that allows rotation in three-dimensional space. The rings can be fixed so as to permit the rotation of the subject around each axis separately. The subject stands in the center of the apparatus (with the subject's hip approximately at the center of rotation) on a platform attached to the innermost ring, and is secured by padded holders on the hip (Fig. 1).

The settings of the platform and the holders were adjusted for each subject before making the first measurement and were retained for the following measurements. The level of the platform was chosen according to the body height of the subject. For a body height of 160 cm, the platform was adjusted to 14 cm. The platform level was lowered for taller persons and raised for smaller persons (1 cm for 2 cm of body

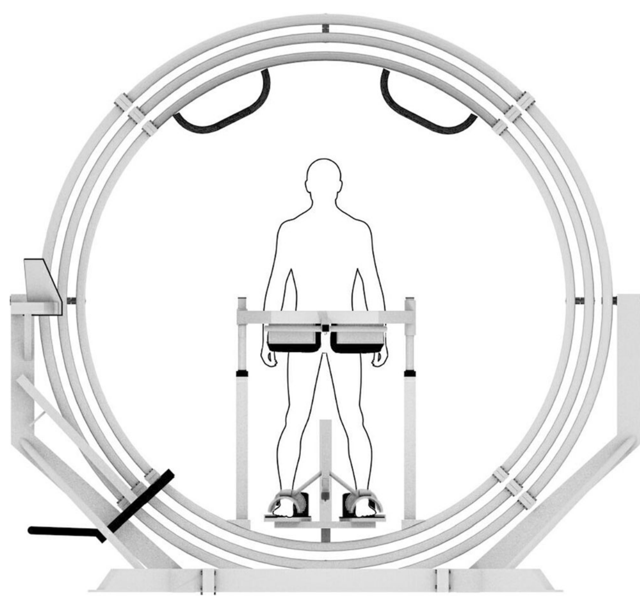

Fig. 1 Schematic illustration of a subject standing in the Spacecurl. The subject stands on a platform and is secured by padded holders at the hips and feet. The Spacecurl model is published with the kind permission of Klaus-Hendrik Wolf of the Peter L. Reichertz Institute for Medical Informatics, University of Braunschweig–Institute of Technology, and Hannover Medical School, Germany

height). The padded holders were adjusted to the level of the iliac crests and of the lumbar lordosis of the back. These holders were tightly fixed in such a way that the subject stood upright. The feet were secured by padded brackets. These brackets were only loosely attached without affecting the load under the subject's feet.

An SPV measurement was made by an examiner and an assistant. The examiner gave standardized instructions and moved the rings of the Spacecurl. The assistant handled the computer. Before starting the measurement, the subject was instructed to stand in an upright body position, while placing his/her hands on the support frame right in front of the trunk. To rule out any visual input, the subject wore a pair of opaque goggles.

The SPV was first assessed in the sagittal plane and afterward in the frontal plane, using the method of magnitude production. Six trials per plane were conducted, with the start positions in random order (12°, 15°, and 18°). From the start position, the Spacecurl was rotated back in the direction of the earth vertical or across until the subject had verbally identified the position that he or she felt to be upright. The subject was allowed to make small adjustments until he or she was satisfied that a vertical position had been reached. Subsequently the subject was tilted to the next start position. The Spacecurl was rotated manually as steadily and smoothly as possible by the examiner at a velocity of 1.0°–1.5° per second (feedback was provided on the computer screen).

Deviations from the earth vertical were measured with the Wireless Inertial Measurement Unit (IMU BT02-0300F05, Memsense, Rapid City, USA) placed on the support frame of the Spacecurl right in front of the subject, approximately on the level of the subject's body center. Data were transmitted wirelessly between the sensor and computer via the Bluetooth protocol and were recorded using a EyeSeeCam software module. The data were analyzed using a MATLAB-based program.

Experimental designs

To determine the test–retest and interrater reliability of the SPV measurements, the SPV was measured two times a day on two consecutive days. The study design is shown in Fig. 2. Measurement 1, Measurement 2, and Measurement 3 were made by the same examiner, whereas Measurement 4 was done by another examiner. The data from Measurements 1 and 2 were used to estimate test–retest reliability, and the data from Measurements 3 and 4 to estimate interrater reliability. Between both Measurements 1 and 2 and Measurements 3 and 4, the subject had a standardized rest period of 20 min to relax on a chair. No feedback about his/her performance was given to the subject before the four SPV measurements were completed.

For the normative SPV values, only one session was necessary to measure the roll and pitch planes (as described above). The normative data were all assessed by the same two examiners.

Data and statistical analysis

The SPV was described in terms of the difference between the subject's perceived vertical and the gravitational vertical. In the sagittal plane, forward deviations of the SPV were given a positive sign, backward deviations a negative sign. In the frontal plane, rightward deviations were indicated by a positive sign and leftward deviations by a negative sign. The SPV error was obtained by averaging the six trials per measurement, and the SPV range was calculated as the difference between the maximum and minimum values of the six trials (Baccini, Paci, Del Colletto, Ravenni, & Baldassi, 2014).

Test–retest reliability and interrater reliability were computed for SPV measurements in the sagittal and the frontal planes separately. To determine the consistency between measurements, the intraclass correlation coefficient (ICC) with the 95% confidence interval (95% CI) was used. The ICC(2,6) model was applied to test the test–retest reliability, and the ICC(3,6) model was used to estimate the interrater reliability (Shrout & Fleiss, 1979). The standard error of measurement (*SEM*) was defined as the square root of the mean squared error. To calculate the minimal detectable change (MDC), the

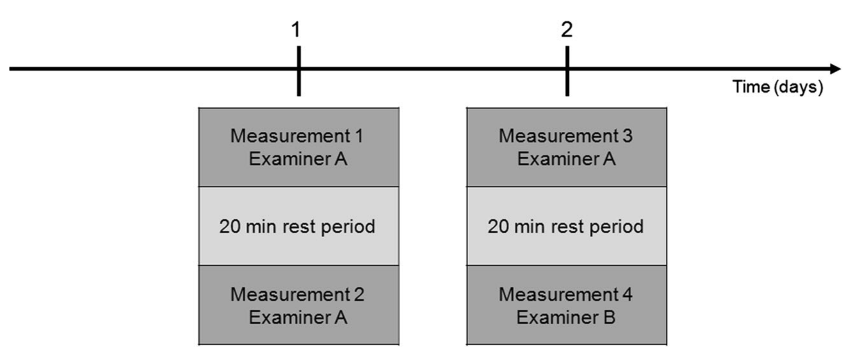

Fig. 2 Study design to determine the test–retest and interrater reliabilities of measurements of the subjective postural vertical using the Spacecurl. Four tests were performed on two consecutive days. Measurement 4 was carried out by a different experimenter than Measurements 1, 2, and 3. To estimate test–retest reliability, Measurements 1 and 2 were compared; to estimate interrater reliability, Measurements 3 and 4

SEM was multiplied by 1.96 and by the square root of 2 (Weir, 2005).

The degree of agreement between measurements was determined by calculating the mean difference between measures (d) and the 95% limits of agreement (LOA: d ± 1.96 *SD*), displayed by Bland–Altman plots (Bland & Altman, 1986). For comparison of the SPV errors and the SPV ranges between the age decades, univariate analyses of variance (ANOVAs) were performed for both planes, and post-hoc Tukey tests were applied. Correlation analyses between age and either the SPV error or the SPV range were carried out with the Pearson test. All calculations were considered significant at the 5% alpha level. Statistical analysis was performed using the statistical package SPSS Statistics 17.0.

Results

Reliability

All 25 subjects included in the reliability experiment completed the four SPV measurements. Table 1 presents ICCs with the 95% CIs, *SEM*s, and MDCs for estimation of the test–retest and interrater reliabilities. Bland–Altman plots (Fig. 3) show mean differences between the measures and 95% LOAs for the test–retest and interrater reliabilities in the sagittal and frontal planes.

Normative data and age dependency

The mean SPV error and mean SPV range per age decade, and the results of the ANOVAs, are listed in Table 2. The largest difference between the age groups for the range of SPV in roll was found between subjects 40–49 years and subjects 60–69 years of age, but this difference did not reach significance in the post-hoc test (p = .064).

Table 1 Reliability parameters of subjective postural vertical measurements in the sagittal and frontal planes

	ICC (95% CI)	*SEM* (°)	MDC (°)
Test–Retest			
Sagittal plane	.70 (.31–.87)	0.7	1.9
Frontal plane	.73 (.40–.88)	0.5	1.3
Interrater			
Sagittal plane	.66 (.23–.85)	0.8	2.3
Frontal plane	.73 (.39–.88)	0.5	1.3

ICC, intraclass correlation coefficient; 95% CI, 95% confidence interval; *SEM*, standard error of measurement; MDC, minimal detectable change

Because the SPV errors did not differ between the various age decades, the ranges of normality were calculated for the whole group of subjects from 20 to 79 years of age. The average SPV (mean ± *SD*) for all subjects was 0.3° ± 1.0° in the sagittal and –0.2° ± 0.7° in the frontal plane. Thus, the values of normality (mean ± 2 *SD*s) in the sagittal plane ranged from –1.7° to 2.3°, and in the frontal plane, from –1.6° to 1.2°.

Moderate, statistically significant correlations between age and SPV error and between age and SPV range were found for the sagittal plane (error: r = –.262, p = .043; range: r = .385, p = .002), but not for the frontal plane (p > .110). Scatterplots for the correlations in the sagittal plane are shown in Fig. 4.

Discussion

This was the first time that SPV measurements made with standing subjects have been evaluated for their test–retest and interrater reliabilities and that respective normative values have been given for the sagittal and frontal planes. We found overall good reliability in healthy subjects and minor changes with aging—that is, increased variance of the estimations. In the past, various devices have been used to measure the SPV, but mainly in sitting subjects. SPV assessment in a standing position might be relevant for postural impairments that primarily affect the standing posture.

Reliability

The present study revealed good reliability parameters for the SPV measurements with the Spacecurl in healthy subjects. The ICCs were .73 for both the test–retest and interrater reliabilities in the frontal plane, with a standard error of measurements of 0.5°. The reliability in the sagittal plane was slightly worse than in the frontal plane, in particular the interrater reliability. However, the reproducibility was still reasonable, with the standard error of measurements being smaller than 1°.

On the basis of our results, changes of the SPV are assumed to be clinically relevant if they are ≥1.3° in the frontal plane and ≥1.9° in the sagittal plane. These MDCs are similar to the LOAs illustrated in the Bland–Altman plots. Generally, the plots show good agreement, with very small differences between the measurements.

Nevertheless, the reliability assessed in healthy subjects is not necessarily applicable to those measured in very old people or patients with impaired balance. Assessments of test–retest and interrater reliability should therefore be performed in the respective sample of interest to confirm the potential clinical utility of these measures.

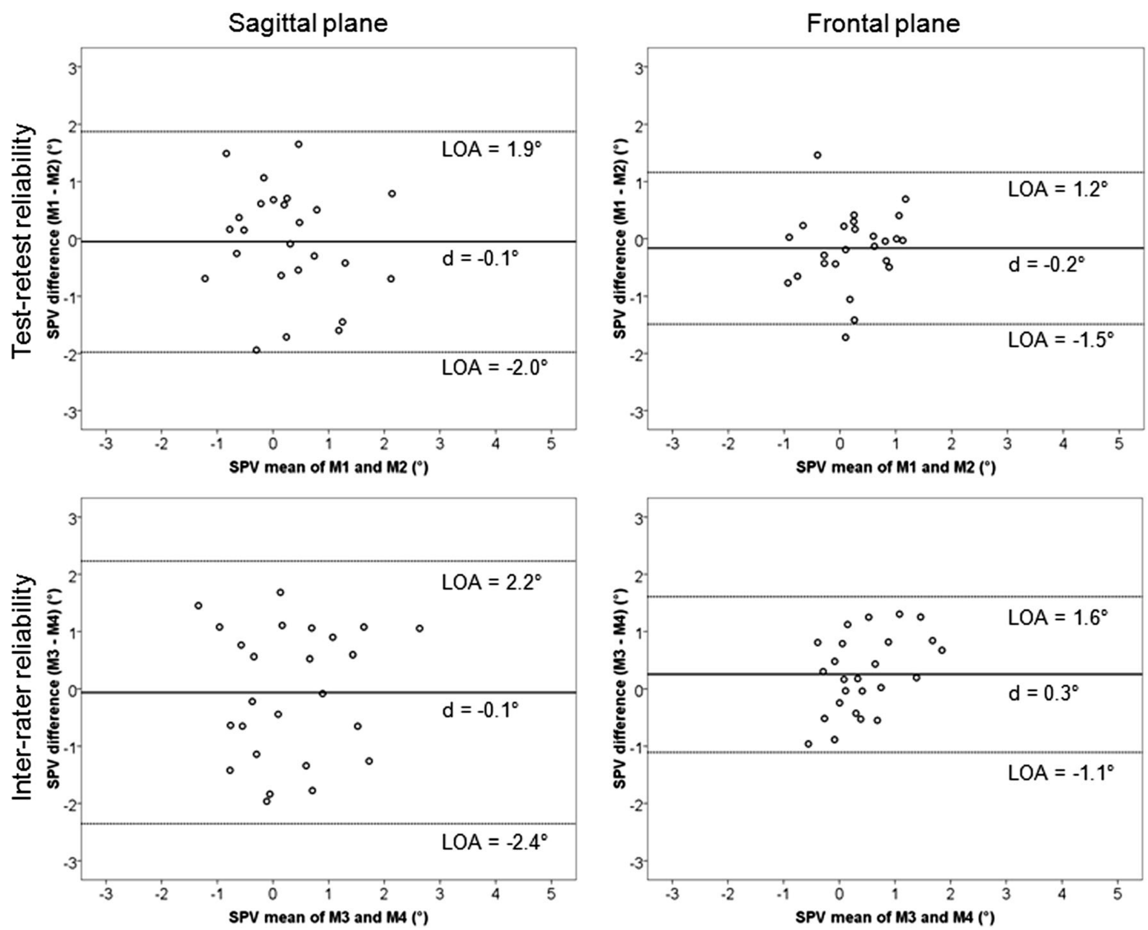

Fig. 3 Bland–Altman plots of the SPV in the sagittal and frontal planes: Differences between measurements versus the means of the measurements, with the mean difference (**d**) and 95% limits of agreement (LOA). SPV, subjective postural vertical; M, measurement

Normative data

Normative values were collected over the age range. Since there was no significant difference in the SPV errors between decades, we calculated the ranges of normality for the whole group: –1.7° to 2.3° in the sagittal plane, and –1.6° to 1.2° in the frontal plane. For the sagittal plane, the range is similar to the values Barbieri et al. (2010) found in young adults in a sitting position (<50 years; –2.4 to 1.5°). The only difference was that we found less backward tilt, most likely due to the different testing devices (see below). For older subjects (≥50 years), Barbieri et al. found a larger and more backward-shifted range of normality (–4.0° to 1.7°). In the frontal plane, normative values for sitting SPV were given by Pérennou et al. (2008), who found a larger range than in our results. Similar to Pérennou et al. (2008), we found an almost symmetrical distribution of the normative values around the gravitational vertical in the frontal plane. This is in contrast to the sagittal plane, in which SPV values were distributed asymmetrically but with differences between standing and sitting. Whereas we observed an average slight forward tilt of the SPV in standing, Barbieri et al. found a backward tilt in sitting. As they discussed, it is likely that the backward-tilted SPV in sitting might be due to a methodological bias caused by the wheel paradigm that they used. When sitting in the wheel paradigm, the main contact points giving somatosensory information are under the buttocks and on the back. Since the

Table 2 Error and range (mean ± *SD*) of subjective postural vertical for different age decades, with results of the ANOVAs

	Sagittal Plane		Frontal Plane	
Age (years)	Error (°)	Range (°)	Error (°)	Range (°)
20–29 (7f)	0.2 ± 1.0	3.2 ± 1.3	–0.5 ± 0.7	2.2 ± 1.0
30–39 (3f)	0.9 ± 0.9	4.3 ± 1.9	–0.5 ± 0.5	2.6 ± 1.2
40–49 (4f)	0.6 ± 0.8	4.4 ± 1.5	–0.1 ± 0.5	3.6 ± 1.6
50–59 (8f)	0.2 ± 1.0	5.0 ± 1.4	0.0 ± 0.6	2.6 ± 1.2
60–69 (5f)	0.1 ± 0.6	5.2 ± 2.1	0.1 ± 1.0	2.1 ± 0.8
70–79 (7f)	–0.2 ± 1.2	5.3 ± 2.1	–0.3 ± 0.7	3.3 ± 1.3
F	1.652	2.041	1.871	2.532
p	0.162	0.087	0.115	0.039

f, female

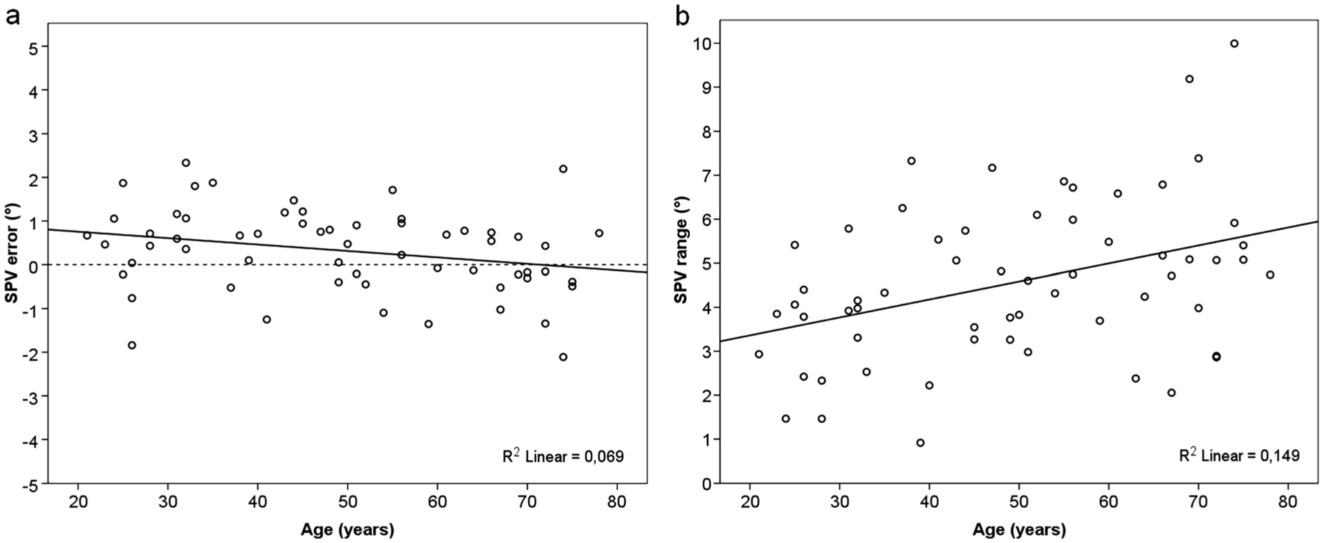

Fig. 4 Scatterplots illustrating the correlations between (**a**) subjective postural vertical (SPV) error and age and (**b**) SPV range and age, in the sagittal plane. The SPV error is slightly forward-tilted in younger subjects and approaches the earth vertical with increasing age. The SPV range increases with increasing age

internal estimate of verticality seems to be tilted to the side from which one gets more somatosensory information (Barra et al., 2010), the SPV might be shifted backward during sitting. In our paradigm, the main contact surfaces were almost symmetrical on the front and back, due to the padded holders on the hip as well as on the feet. There was thus no preponderance of one side.

Age dependency

Consistent with the findings of Barbieri et al. (2010), we found age-related changes of the SPV: a slight backward shift of the SPV error and a larger SPV range with increasing age. In the present study, the SPV error shifted from a small forward tilt in younger subjects toward the earth vertical with aging. In the work of Barbieri et al., the SPV shifted from an average slightly backward tilt in younger subjects to a more distinctly backward tilt in older subjects. The larger SPV range found in both studies in older subjects indicates increased uncertainty in verticality perception. Similarly, Bisdorff et al. (1996) observed larger sector widths of the SPV with aging—that is, a loss of sensitivity for the perception of body verticality. This reduced sensitivity may reflect an age-related decline of vestibular and somatosensory functions (Choy, Brauer, & Nitz, 2003; Nusbaum, 1999). These sensory systems are involved in creating and updating the central representation of verticality (Barra et al., 2010). In particular, the somatosensory system is supposed to be important for the SPV—for example, to improve the stability of the verticality representation (Barbieri et al., 2010; Barra et al., 2010; Bringoux, Marin, Nougier, Barraud, & Raphel, 2000).

Saeys et al. (2012) determined the influence of somatosensory loss on the perception of verticality in stroke patients during sitting. They differentiated between skin-related and joint-related somatosensory information and found a stronger relationship between the SPV and skin-related somatosensory input. In the Spacecurl, the contact area between the padded holders and the body is relatively small; however, the pattern of pressure at the hip and the pressure distribution under the feet might have affected the SPV measurements. Studies on patients with complete and partial somatosensory loss or on subjects with experimentally disturbed body sense (vibration) might help to determine the influences of somatosensory information on the SPV. When measuring SPV during sitting, Mazibrada et al. (2008) found a large error of the SPV in the frontal plane in a patient with Guillain–Barré syndrome and with severe symmetrical loss of peripheral sensation. The SPV accuracy considerably improved after recovery. In contrast, two patients with paralysis from Th 6–7 down did not show a significant error of the SPV. The authors concluded that somatosensory input from the trunk and the shoulders is especially important for the perception of verticality during sitting, whereas input from the lower limbs is less important, at least during sitting. For the SPV during standing, however, somatosensory input from the lower limbs and feet may play a major role.

A potential limitation of the Spacecurl as a measurement tool for the SPV is that the fixation on the hip not only provides the subject with somatosensory information but also forces the subject into an upright posture that might differ from the spontaneous subjective upright posture. Furthermore, the feet and hip fixations hamper postural control strategies such as the ankle or hip strategy in the sagittal plane. Nonetheless, the fixations are necessary to secure the subject. Since the Spacecurl was originally constructed for therapy, it provides no fixation of the trunk or head. For SPV measurements, its advantage is that no unnatural pressure cues are provided by holders or fixation straps on the upper body. However, the muscle activity needed to control the trunk and the head relative to gravity might increase the proprioceptive input available for SPV estimation, and might consequently lead to more precise and robust SPV measurements (as in the natural condition). This might account for the rather small variability in our data. Previous studies assessing the SPV had restrained the trunk and head and had allowed for rather less postural activity. In our study, the trunk and head were free to move, and postural control was needed to a greater extend. It remains to be determined whether complete restraint or more free standing is better suited to measure the inner representation of body orientation in space.

Another limitation, especially in comparisons with studies assessing the SPV in sitting, is the comparatively small tilt of the start positions. A maximum tilt angle of 18° was chosen, because larger angles were hardly tolerable by several subjects, due to the upper body being free to move. Larger angles in these subjects caused fear and undesired postural reactions. Future measurements with patients should determine whether these start positions are suitable for detecting SPV deviations in patients with severely impaired verticality perception.

In conclusion, we found precise and reliable SPV estimations for healthy subjects in standing using the Spacecurl. We found on average a slightly forward-tilted SPV, which approached the earth vertical with increasing age. The clinimetric properties of the SPV in standing have to be further investigated in patients with deficits of postural control and/or upright body orientation. SPV measurements in patients with somatosensory, vestibular, or central disorders will lead to a better understanding of the pathways forming the inner model of verticality perception. The Spacecurl is a promising tool for these kinds of experiments.

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The Subjective Postural Vertical Determined in Patients with Pusher Behavior During Standing

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Background: The subjective postural vertical (SPV), i.e., the perceived upright orientation of the body in relation to gravity, is disturbed in patients with pusher behavior. So far, the SPV has been measured only when these patients were sitting, and the results were contradictory as regards the side of the SPV deviation.
Objective: The objective was to investigate the SPV in patients with different degrees of severity of pusher behavior while standing.
Methods: Eight stroke patients with pusher behavior, ten age-matched stroke patients without pusher behavior, and ten age-matched healthy control subjects were included. The SPV (SPV error, SPV range) was assessed in the pitch and the roll planes. Pusher behavior was classified with the Burke Lateropulsion Scale (BLS).
Results: In the pitch plane, the SPV range was significantly larger in pusher patients than in patients without pusher behavior or healthy controls. The SPV error was similar for groups. In the roll plane, the SPV error and the SPV range were significantly larger and more ipsilesionally tilted in the pusher group than in the other two groups. There was a significant correlation between the SPV error in the roll plane and the BLS score.
Conclusions: The study revealed that patients with pusher behavior had an ipsilesional SPV tilt that decreased with decreasing severity of the behavior. The large uncertainty in verticality estimation in both planes indicates that their sensitivity for the perception of verticality in space is generally disturbed. These findings emphasize the importance of specific rehabilitation approaches to recalibrate the impaired inner model of verticality.

Keywords: subjective postural vertical, verticality perception, stroke, pusher behavior

Introduction

Stroke patients who actively shift their body weight across the midline toward the side of the hemiparesis and resist passive correction of the tilted posture have what is called pusher behavior or lateropulsion.[1] Depending on its severity, this behavior compromises sitting, standing, transferring, or walking and can lead to a loss of balance and falls.

Pusher behavior is a major issue in neurorehabilitation, since it hampers and prolongs the rehabilitation process. Patients with pusher behavior need about 4 weeks longer to reach the same functional outcome level as stroke patients without pusher behavior[2,3] or are only half as efficient and effective in their rehabilitation outcome.[4]

Patients with pusher behavior are thought to orient their body toward a disturbed internal reference of verticality. The subjective postural vertical (SPV) reflects the perceived upright orientation of the body in relation to gravity; it is altered in patients with pusher behavior.[5,6] However, previous investigations reported contradictory results as regards the side of the SPV deviation: one study found the SPV to be tilted about 18° to the ipsilesional side,[5] while another reported a tilt of similar magnitude to the contralesional side.[6] Both studies assessed the SPV with the patient in a sitting position. Different models were discussed to explain how the SPV leads to pusher behavior. Karnath et al[5] found a mismatch between the visual vertical and the orientation of body verticality. They suggested that patients with pusher behavior try to compensate for this mismatch by pushing their longitudinal body axis toward the contralesional side. By contrast, a transmodal tilt of the visual and the postural vertical to the contralesional side was found by Perennou et al.[6] The authors suggested that patients with pusher behavior try to align their body with the contralesionally tilted reference of verticality.

As stated above, pusher behavior can compromise different postures. In its severe form, pusher behavior

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is present during sitting and standing (and possibly also during lying). In a less severe form or when the patient has progressed during the rehabilitation process, pusher behavior persists during standing or walking, but is absent during sitting.[7,8] If patients with pusher behavior orient their body toward an altered internal reference of verticality, it would seem especially relevant to assess the SPV of patients with mild pusher behavior during standing, since this posture is primarily affected. SPV assessment in patients with various degrees of severity of pusher behavior might also help to understand the mechanisms leading to pusher behavior and to design specific rehabilitation programs tailored to these patients.

The objective of this study was to assess the SPV in patients with different levels of pusher behavior while standing. This behavior ranged from severe pusher behavior, which compromised sitting and standing posture, to mild pusher behavior, which affected only standing or walking. The SPV was assessed in the roll (frontal) and pitch (sagittal) planes and compared to the SPV of stroke patients without signs of pusher behavior and the SPV of healthy subjects.

Methods

Participants

Eight stroke patients with pusher behavior (BLS score ≥2) and ten age-matched stroke patients without signs of pusher behavior participated in this study. All stroke patients had had a hemiparesis due to a unilateral hemispheric stroke less than 6 months before inclusion in the study. Additionally, ten age-matched healthy control subjects were enrolled. Exclusion criteria for all participants were acute cardiac disease, known arterial aneurism or thrombosis, unstable spinal column, neuroses/psychoses, pregnancy, body height <145 cm or >195 cm, body weight >150 kg, and age <18 or >90 years. An inclusion criterion for patients was the ability to tolerate 30 min of passive standing. Independent standing, however, was not required, since the measurement device allows support of the patient during standing.

The study was approved by the Ethics Committee of XXX in accordance with the Declaration of Helsinki. All participants or their legal representatives gave their written informed consent.

Assessments and procedure

The Burke Lateropulsion Scale (BLS) was used to classify pusher behavior. The scale assesses the patient's resistance to passive supine rolling, to passive postural correction when sitting and standing, and to assistance during transferring and walking.[9] Each item is scored from 0 to 3 (0–4 for standing) and is based on the severity of resistance or the angle at which the patient starts to resist passive movement. A higher score reflects more severe pusher behavior. For classification of pusher behavior, the cutoff score ≥2 was used.[7]

In addition, the Scale for Contraversive Pushing (SCP) was applied. The SCP is another clinical scale for detecting and rating pusher behavior.[5,10] The SCP has three components: the symmetry of spontaneous body posture, the use of the non-paretic extremities, and the resistance to passive correction. Each component is rated in sitting and standing positions.

Since the aim of this study was to also examine patients with mild forms of pusher behavior, the BLS was used for classifying the level of pusher behavior. It is more sensitive than the SCP for detecting mild or resolving pusher behavior.[8]

The SPV assessment during standing was performed using the Spacecurl® (Physio Boerse, Wittlich, Germany). The apparatus and the experimental procedure are described elsewhere.[11] For SPV assessment, the blindfolded subject stood in the center of the device. All subjects were secured by padded holders on the hip. Additionally, padded holders for knee fixation were used in patients. Consequently, also patients who were not able to stand unsupported could be measured with the device. The subject was passively tilted to given start positions (12°, 15°, or 18°) in the pitch (sagittal) and roll (frontal) planes. SPV was first assessed in the pitch plane and afterward in the roll plane; six trials were performed per plane. Start positions were presented in an unpredictable order, alternating between front and back, or right and left, respectively. From the start position, the subject was rotated backward in the direction of the earth vertical or across until the subject indicated that he or she felt upright. The subject was allowed to make small adjustments until satisfied that a vertical position had been reached. Angular deviations from the earth vertical were measured with the Wireless Inertial Measurement Unit (IMU BT02-0300F05; Memsense, Rapid City, USA); values were transmitted wirelessly and recorded using a EyeSeeCam software module. A MATLAB-based program was used to analyze the data.

The Barthel Index[12] was collected to determine functional disability of patients. The Barthel Index measures independence in activities of daily living. In addition to the total Barthel Index, the item mobility was of interest to provide information about the mobility level of patients.

Data and statistical analysis

The SPV was described in terms of the difference between the subject's perceived vertical and the gravitational vertical. In the pitch plane, forward SPV deviations were indicated by a positive sign, backward deviations by a negative sign. In the roll plane, ipsilesional deviations for patients and rightward deviations for healthy controls were given a positive sign, contralesional or leftward deviations, and a negative sign. The SPV error was defined as the mean SPV of the six trials per plane. The SPV range was calculated as the difference between the maximum and the minimum SPV values of the six trials. The SPV range represents the uncertainty in verticality perception.[13]

Demographic and clinical characteristics of the participants were compared between groups using the chi-square test (for comparison of proportions), a Mann–Whitney *U*-test (for ordinal variables), a one-way ANOVA, or Student's *t*-test (for continuous variables). A one-way ANOVA was used to evaluate differences in the SPV error and the SPV range between groups. In case of significant results, subsequent *post hoc* tests were performed using the Bonferroni procedure.

Correlations of the SPV errors and the SPV ranges with the BLS score or the SCP score were analyzed with Spearman's tests.

The significance level for α was set at 0.05. Statistical analysis was performed using the statistical software package SPSS Statistics 17.0.

Results

Participant characteristics are shown in Table 1. The three groups (pusher $n = 8$, non-pusher $n = 10$, controls $n = 10$) did not differ in age ($F(2) = 0.120$, $p = 0.888$) or gender ($X^2(2) = 3.369$, $p = 0.186$). Patients of the pusher group and patients of the non-pusher group were similar in terms of the etiology of the lesion ($X^2(1) = 0.678$, $p = 0.410$), the side of the lesion ($X^2(1) = 0.055$, $p = 0.814$), and the time since lesion ($t(16) = 0.221$, $p = 0.828$). The total Barthel Index significantly differed between the two patient groups ($U = 3.500$, $Z = -3.272$, $p = 0.001$). Patients of the pusher group showed more severely impaired performance in activities of daily living than stroke patients without pusher behavior. However, the groups did not significantly differ with regard to their mobility level (Barthel Index item mobility; $X^2(4) = 4.915$, $p = 0.178$).

Mean SPV values for the three groups and results of the ANOVAs are shown in Table 2. In the pitch plane, there was a significant difference between groups for the SPV range, but not for the SPV error. *Post hoc* tests revealed a significantly greater SPV range for the pusher group than for the non-pusher group ($p < 0.001$) and the healthy control group ($p < 0.001$). In the roll plane, both the SPV error and the SPV range significantly differed between groups. *Post hoc* tests revealed significant differences of the SPV error between the pusher group and both, the non-pusher group ($p = 0.015$), and the healthy control group ($p < 0.001$). The pusher group showed larger and more ipsilesional SPV deviations than the non-pusher group and the control group (Figure 1). Regarding the SPV range in the roll plane, *post hoc* tests revealed a significantly larger SPV range for the pusher group than for the non-pusher group ($p < 0.001$) and the healthy control group ($p < 0.001$).

SPV errors and ranges for each subject of the pusher group, the non-pusher group, and the healthy control group are shown in Table 1. A recent study defined the ranges of normality for the SPV during standing: −1.7–2.3° in the pitch plane and −1.6–1.2° in the roll plane.[11] According to these ranges, the SPV errors of 50% (4/8) of patients of the pusher group and 40% (4/10) of patients of the non-pusher group were abnormal in the pitch plane. In the roll plane, the SPV errors of 50% (4/8) of the pusher patients and 20% (2/10) of the non-pusher patients were outside the ranges of normality. SPV errors of the two non-pusher patients were only slightly outside the ranges of normality (<0.5°), while all abnormal SPV errors in the pusher group were more than 1.5° outside these ranges.

There was a significant positive correlation between the SPV error in the roll plane and the BLS score ($r_s = 0.663$; $p = 0.037$), while the relationship between the SPV error and the SCP was not significant ($r_s = 0.575$, $p = 0.068$). Patients with larger deviations of the SPV showed higher scores on the pusher scales. The SPV error in the pitch plane and the SPV ranges did not correlate with the BLS or the SCP.

Discussion

Patients with pusher behavior show evidence of a misperception of their body's orientation to gravity. Two previous studies found large SPV deviations in the roll plane, but the deviations were opposite as regards their direction from the side of the lesion.[5,6] So far, SPV has been assessed in patients with pusher behavior only while in a sitting position. However, since the standing posture is primarily affected in patients with mild forms of pusher behavior, SPV assessment during standing is thought to be especially relevant. This is the first study to assess the SPV in stroke patients with pusher behavior while standing. In this study, pusher behavior ranged from very mild forms that compromised primarily standing and/or walking to more severe forms that affected both standing and sitting postures.

The pusher group showed abnormal SPV deviations in the roll plane, but not in the pitch plane. Individual SPV estimations in the pusher group were on average either vertically aligned or ipsilesionally tilted. Ipsilesional SPV tilts in patients with pusher behavior are in line with the findings of Karnath et al,[5] who measured the SPV in sitting patients. Similar to the model Karnath et al[5] discussed, we suggest that pusher behavior might result from an active compensation for a conflict between reference

Table 1 Demographics and characteristics

ID	Group	Age (years)	Gender	Lesion location	Etiology	Side of lesion	Time since lesion (days)	BI total	BI mobility	SCP score	BLS score	SPV error pitch (°)	SPV range pitch (°)	SPV error roll (°)	SPV range roll (°)
1	P	79	M	T, F, P	I	R	103	40	5	1.75	3	6.8	19.7	4.0	21.2
2	P	77	F	T, F, P	I	R	32	10	0	3	4	–2.2	20.5	2.9	14.1
3	P	70	F	F, P	I	R	77	40	5	1	2	–2.2	8.1	0.0	18.6
4	P	86	M	P, O	H	R	14	15	0	6	15	–3.9	10.6	6.2	14.1
5	P	52	F	BG	H	L	180	35	5	3	6	1.1	8.7	0.5	8.8
6	P	67	F	F, P	I	L	32	30	5	0.75	2	1.5	13.6	0.5	5.3
7	P	75	F	BG	H	R	50	35	5	2.5	4	0.6	12.4	5.5	10.2
8	P	74	M	T	I	R	40	35	5	0.25	2	–0.4	13.6	0.7	15.9
9	NP	60	M	P	H	R	39	50	5			3.4	7.7	0.9	5.0
10	NP	81	F	BG	I	L	51	45	5			–1.2	3.7	0.9	1.7
11	NP	69	F	BG	I	R	60	50	5			–2.2	10.5	0.3	9.1
12	NP	52	M	T,F,P	I	R	39	45	5			–2.0	9.4	0.2	5.8
13	NP	72	M	P	I	R	42	70	5			–2.1	5.8	–0.5	4.0
14	NP	74	M	F, P	I	R	154	70	5			1.0	7.4	1.6	11.2
15	NP	76	M	T, P	I	L	34	85	10			–0.8	3.4	1.5	3.0
16	NP	80	M	T, F, P, BG	I	R	122	50	5			0.2	6.1	–0.1	11.5
17	NP	72	M	BG	H	L	39	100	15			2.0	5.6	–0.1	2.3
18	NP	75	M	T, F, P, BG	I	R	30	35	10			–0.8	9.5	–1.6	2.7
19	C	70	F									–0.5	7.4	–0.3	4.1
20	C	74	M									2.2	5.9	–1.2	3.3
21	C	72	F									0.4	2.9	–1.2	6.1
22	C	72	M									–1.3	2.9	–0.4	3.0
23	C	80	M									0.4	7.6	1.0	8.5
24	C	72	M									–0.2	5.1	0.0	3.0
25	C	75	F									–0.5	5.4	–0.3	4.1
26	C	52	M									0.3	4.6	–1.4	1.9
27	C	71	M									0.2	2.4	–1.3	2.8
28	C	67	F									–1.0	2.0	–1.5	3.2

Note: P = pusher, NP = non-pusher, C = controls, F = female, M = male, BG = basal ganglia; FL = Frontal lobe; TL = Temporal lobe; PL = Parietal lobe, I = Ischemia, H = intracerebral hemorrhage, l = left, r = right, BI = Barthel index.

Table 2 SPV error and SPV range (mean ± SD)

		P	NP	Controls	ANOVA
Pitch plane	Error (°)	0.2 ± 3.3	–0.3 ± 1.9	0.0 ± 1.0	$F = 0.080$, $p = 0.923$
	Range (°)	13.4 ± 4.6	6.9 ± 2.4	4.6 ± 2.0	$F = 18.843$, $p < 0.001$
Roll plane	Error (°)	2.5 ± 2.5	0.3 ± 1.0	–0.6 ± 0.8	$F = 10.078$, $p = 0.001$
	Range (°)	13.5 ± 5.2	5.6 ± 3.7	4.0 ± 1.9	$F = 16.041$, $p < 0.001$

Note: P = pusher, NP = non-pusher.

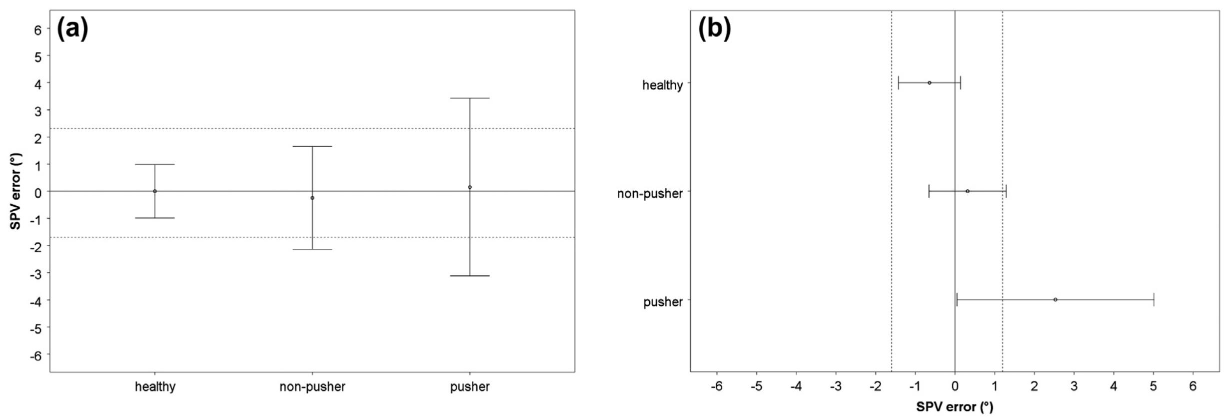

Figure 1 Mean (±SD) SPV error of the healthy control group, the non-pusher group, and the pusher group in (a) the pitch plane and (b) the roll plane. Dashed lines indicate the ranges of normality (pitch plane: –1.7–2.3°, roll plane: –1.6–1.2°).[13]

systems.[5] However, the ipsilesional SPV tilts Karnath et al[5] found were larger (about 18°) than ours. Differences in the base of support and additional degrees of freedom in the hip, knee, and foot joints during standing might increase the sensory input available for SPV estimation and consequently lead to more precise SPV measurements during standing than during sitting. Moreover, the patients in Karnath's study[5] had more severe forms of pusher behavior than those in this study. The correlation we found between SPV deviations and the severity of pusher behavior supports the assumption that patients with more severe forms of pusher behavior show larger deviations of their internal representation of verticality.[6] In a recent study, Mansfield et al[14] determined the SPV in sitting chronic stroke patients whose pusher behavior had resolved. They found no difference of the SPV between patients with a history of pusher behavior and patients without a history of pusher behavior. Consequently, one might assume that the recovery of the impaired SPV accompanies the recovery of pusher behavior. An important finding of our study is that also patients with mild forms or almost resolved pusher behavior, who would not have been diagnosed to have pusher behavior when using the SCP scale, (still) showed abnormal SPV deviations in the roll plane. Whether the SPV deviations were only abnormal during standing or possibly also during sitting was not determined in this study. Generally, the SCP is used for the classification of pusher behavior; however, the BLS seems more suitable for detecting mild signs of pusher behavior affecting standing and walking.[8] Our finding that misrepresentation of body orientation is still present, even if signs of pusher behavior are mild and primarily present in standing or walking, underlines that the BLS can be used as a valid tool to detect pusher behavior. Another aspect is that all patients with a BLS score of 2 showed an SPV error in the roll plane within the ranges of normality. This supports changing the BLS cutoff to >2 instead of using ≥2 to classify pusher behavior, which was already discussed by Bergmann et al.[8]

Another study that assessed the SPV in patients with pusher behavior during sitting[6] contradicted our results and the results of Karnath et al.[5] The former found large contralesional SPV deviations in patients with pusher behavior. While they restrained the head,[6] there was no fixation of the head in our study or in the study of Karnath et al.[5] Spontaneously, patients with pusher behavior typically turn and shift their head laterally toward the ipsilesional side.[1,15] Differences in vestibular inputs due to different head positions might explain differences in the SPV estimations. However, vestibular input seems relatively unimportant for SPV estimation.[16,17] On the other hand, the restriction of the head in a rather unnatural position might have biased somatosensory input from the neck and possibly caused compensatory postural responses. Another difference in the experimental setting of Pérennou et al[6] and Karnath et al[5] was the restriction of the legs. In the study of Pérennou et al[6], patients legs were strapped and the feet were in contact with the ground. In

Karnath et al.'s[5] study, the legs of the patients freely hung. Johannsen et al[18] studied spontaneous postural responses of patients with pusher behavior during passive body tilt. They found a constant ipsilesional tilt of the non-paretic leg corresponding to trunk orientation, which was most pronounced during body tilt in the ipsiversive direction. Fixation of the legs hampers these spontaneous postural responses and might bias afferent inputs. In our study, leg position was restricted due to the hip fixation and the platform for the feet, but the trunk was free to move. In general, it remains to be determined whether complete restraint or a more free and natural posture better reflects the inner representation of the body's orientation in relation to gravity. We decided for a more natural position with the trunk and the head free to move. The position required a certain amount of muscle activity and allowed postural reactions.

Pusher behavior is considered to be a disorder that primarily affects the patient's perception in the roll plane. Consequently, SPV has to date only been assessed in the roll plane for patients with pusher behavior. However, patients often also show a posterior element to their pusher behavior.[3] To the best of our knowledge, this study is the first to also determine the SPV in the pitch plane for patients with pusher behavior. SPV deviations in the pitch plane did not differ for the pusher group compared to the non-pusher group or the healthy control group. However, the SPV range was significantly larger for patients with pusher behavior than for patients without pusher behavior or for controls. Patients with pusher behavior showed a noticeable variability and uncertainty in verticality estimation in both the pitch and the roll planes. This indicates a general loss of sensitivity for the perception of verticality in space. For visual verticality perception, an increased noise in encoding of visual orientation in patients with pusher behavior was discussed.[19] A lower signal-to-noise ratio in coding systems might possibly also account for the large uncertainty in verticality estimation by patients with pusher behavior.

There are some limitations to this study. First, the sample sizes per group were small. Only eight patients with signs of pusher behavior were included in the study. Nevertheless, this number is comparable to other studies investigating patients with pusher behavior, but studies with a larger number of patients covering the whole range of severity of pusher behavior are needed to verify the validity of our findings. Secondly, we interpreted the large SPV ranges found in patients with pusher behavior as a decreased sensitivity for orientation perception. Nonetheless, we cannot exclude that patients were not able to perceive upright orientation of the body and were primarily guessing during SPV assessment. Experimental limitations, such as the active control of the head and trunk position or the relatively small tilt of the start positions, are discussed in Bergmann et al.[11]

This study investigated the SPV during standing in a cohort of patients with different levels of pusher behavior. Future work is needed to determine whether the SPV differs in this cohort during standing compared to sitting.

Conclusions

The study revealed that patients with pusher behavior had an ipsilesional SPV tilt while standing. The SPV tilt decreased with decreasing severity of pusher behavior. Patients with a BLS score of 2 points showed SPV tilts in the roll plane within the ranges of normality, indicating a BLS cutoff >2 to be more valid to classify pusher behavior than the hitherto used cutoff ≥2. Although there was no abnormal SPV deviation in the pitch plane, there was a considerably large uncertainty of verticality estimation in both planes. These findings emphasize the importance for specific rehabilitation approaches to recalibrate the impaired perception of verticality in patients with pusher behavior. The somatosensory system plays a crucial role in SPV estimation, especially for the stability of verticality representation. Consequently, appropriate somatosensory stimulation might improve the decreased orientation sensitivity of patients with pusher behavior and possibly recalibrate the biased SPV deviation.

Conflict of interest

There is no conflict of interest.

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Ethics

The study was approved by the Ethics Committee of Ludwig-Maximilians University (LMU) Munich in accordance with the Declaration of Helsinki. All participants or their legal representatives gave their written informed consent.

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