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Quantitative Analysis of Armswing in Patients with Primary Cervical Dystonia

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ABSTRACT:

Objective: Accompanying neurological signs are not expected in primary cervical dystonia (PCD), characterized by involuntary muscle contractions resulting in abnormal posture of the neck. We aimed to assess the associated arm movements during walking in patients with PCD in compared to age-, sex-, weight- and height-matched controls.

Methods: We examined 16 consecutive de novo patients with PCD and controls in a quantitative trial, and calculated the mean gait speed, mean number of steps, mean frequencies of right and left arms, mean frequencies of right and left legs, and mean arm/leg frequencies for right and left sides.

Results: Eleven patients (68,8%) had right-sided cervical dystonia, 5 patients (31,2%) had left-sided cervical dystonia. The mean speed of gait and the mean number of steps were similar between patients and controls. The mean frequencies of bilateral upper extremities and arm/leg frequencies were significantly slower in patients with PCD (p<0,05). In patient group, the mean armswing frequency was significantly lower in symptomatic side versus asymptomatic side (p=0,038). The disease duration was positively associated with the mean arm/leg frequency (p=0,042).

Discussion: Our results imply that the reduced armswing in cervical dystonia might be a feature of primary cervical dystonia.

Key words: armswing frequency, primary cervical dystonia, quantitative analysis

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ÖZET:

Primer servikal distonisi olan olgularda kol sallanmasının kantitatif analizi

Amaç: İstemsiz kas kasılması sonucu boynun anormal pozisyonu ile şekillenen primer servikal distonide (PSD) ek nörolojik muayene bulgusu beklenmemektedir. Çalışmamızda, servikal distonisi olan hastalarda yürüme sırasında kol salınımında azalma olup olmadığının video kayıtlama yapılarak araştırılması ve yaş, cinsiyet, kilo ve boy açısından eşleştirilmiş kontrol grubu ile karşılaştırılması amaçlanmıştır.

Yöntem: Çalışmamızda, yeni PSD tanısı almış ardışık 16 hasta ve 16 sağlıklı birey kantitatif bir düzenekte incelenmiş; ortalama yürüme hızları, ortalama adım sayıları, sağ ve sol kolun ortalama salınım frekansları, sağ ve sol bacağın ortalama salınım frekansları ve hem sağ hem de sol taraf için kol/bacak frekans oranı hesaplanmıştır.

Bulgular: Toplam 11 hasta (%68,8) sağ taraflı, 5 hasta ise (%31,2) sol taraflı PSD tanısı aldı. Ortalama yürüme hızları ve adım sayıları hasta ve control grubunda benzerdi. Her iki üst ekstremite ortalama salınım frekans değerleri ile kol/bacak frekans oranları PSD grubunda anlamlı olarak daha düşük bulundu (p<0,05). Hasta grubunda, ortalama kol salınım frekansı semptomatik tarafta asemptomatik tarafa kıyasla belirgin düşüktü (p=0,038). Hastalık süresi ile ortalama kol/bacak frekans oranları arasında pozitif korelasyon saptandı (p=0,042).

Tartışma: Elde ettiğimiz veriler, kol salınımında azalmanın primer servikal distoninin bir bulgusu olabileceğini göstermiştir.

Anahtar sözcükler: primer servikal distoni, kol salınım frekansı, kantitatif analiz

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INTRODUCTION

Primary cervical dystonia (PCD) is the most common form of adult-onset focal dystonia characterized by sustained, involuntary muscle contractions resulting in abnormal posture of the neck.[1,2] Other than abnormal posture and/or movements, local pain is very frequently reported among PCD patients. Any accompanying neurological signs are not expected in primary cases, but in secondary cervical dystonia. However, a recent paper by Kagi et al.[3] reported that armswing was observed to decrease in 55% of patients with adult-onset PCD versus 6% of control subjects. Asymmetry of armswing has increasingly been reported in 'benign' conditions, such as

in focal arm dystonia[4] or essential tremor.[5] The recognition of the presence of reduced armswing is important for preventing misdiagnosis, but also for the better delineation of clinical picture. On the basis of these data, we aimed to examine the patients with primary cervical dystonia in a quantitative trial to assess the associated arm movements during walking in comparison to age-, sex-, weight- and height-matched controls.

MATERIAL AND METHODS

We examined and video-recorded 16 consecutive de novo patients with PCD during one year. All patients were diagnosed by a neurology specialist on movement disorders, and none exhibited any concomitant abnormal neurological findings, or postural/kinetic tremor. Past histories of the patients were unremarkable; none of them had any history of neuroleptic use or use of antiemetic agents that might have parkinsonian side effects. Patients with dystonia on other parts of the body accompanying to the cervical dystonia were excluded. None of the patients had previous botulinum toxin injections or any antidystonic medication.

We positioned a camera at a fixed location under constant conditions, where patients with PCD were asked to walk back and forth a standardized distance of 5 m five times for each side of the body, and arm movements were visualized and recorded using the time function of the camera. The subjects were asked to 'walk at normal speed'. To compare our findings in patients, we had an age- and sex-matched control group of 26 volunteers (spouses/family members or hospital staff), who were video-recorded within the same protocol. None of the patients or controls had any pain disorder or deformities of the skeletal system that could have had any impact on walking. The study was approved by the local ethics committee, and informed written consent was obtained from all participants.

We later analyzed the duration of a walk along a pre-set distance in seconds, the number of steps taken to walk a pre-set distance, and the number of bilateral arm swings while walking. A step was defined as from the beginning of the lift of the heel of one foot off the floor until the beginning of the next lift of the same foot. Similarly, an armswing was defined as an entire circuit of an arm back and forth beginning from one point and returning to it. Examinations were recorded separately for the right and left sides, and the mean values of walking for five times were calculated for all measures. The mean gait speed, mean number of steps, mean frequencies of right and left arms (number of arm swings/time to walk the pre-set distance), mean frequencies of right and left legs (number of steps/time to walk the pre-set distance), and mean arm/leg frequencies for right and left sides (number of arm swings/number of steps) were then calculated.

The statistical analysis of the results was performed using the chi-square test and Spearman's correlation ratio, as appropriate. The parametric variables were compared using the paired-samples t test, independent samples t test and one-way ANOVA, accordingly. The statistical significance was set as a p value equal to or less than 0,05.

RESULTS

A total of 16 patients with adult-onset primary cervical dystonia and 26 age-, sex-, weight- and height-matched healthy control subjects were enrolled into the study. The mean age of patients at examination was 44,0±11,7 years versus 50,2±9,4 years in control group (p=0,196). Five patients were men (31,3%) and 11 were women (68,7%); while there was 10 men (38,5%) and 16 women (61,5%) in the control group (p=0,269). The mean age at disease onset was 34,0±10,2 years in patients with PCD (varying between 18 and 49 years), and the mean disease duration was 10,1±6,9 years (between 1 and 21 years). Eleven patients (68,8%) had right-sided cervical dystonia, and 5 patients (31,2%) had left-sided cervical dystonia. Four patients (25,0%) displayed some degree of retrocollis, and 3 patients (18,7%) had anterocollis, in addition to laterocollis.

The mean speed of gait and the mean number of steps were not different between patients with PCD and control subjects (Table 1). The mean frequencies of both upper extremities, however, were significantly slower in patients with PCD in compared to controls, while there was no

Table 1: The comparison of gait and armswing parameters in patients with PCD and control subjects

Variables	Patients with PCD (n=16)	Control subjects (n=26)	p value
Mean speed of gait (seconds)	3,22 ± 0,78	3,10 ± 0,72	0,814
Mean number of steps	3,11 ± 0,67	2,88 ± 0,68	0,766
Mean frequency of right arm	0,66 ± 0,28	0,70 ± 0,15	0,042
Mean frequency of left arm	0,76 ± 0,27	0,81 ± 0,17	0,038
Mean frequency of right leg	0,82 ± 0,22	0,77 ± 0,11	0,753
Mean frequency of left leg	0,80 ± 0,22	0,80 ± 0,14	0,833
Mean arm/leg frequency for right side	0,77 ± 0,34	0,91 ± 0,13	0,011
Mean arm/leg frequency for left side	0,93 ± 0,13	1,00 ± 0,10	0,026

Table 2: The comparison of the symptomatic and asymptomatic sides in 16 patients with PCD

Variables	Symptomatic side	Asymptomatic side	p value
Mean arm frequency	0,68 ± 0,33	0,72 ± 0,32	0,038
Mean leg frequency	0,80 ± 0,22	0,80 ± 0,22	0,850
Mean arm/leg frequency	0,84 ± 0,28	0,86 ± 0,27	0,184

significant difference in the mean frequencies of lower extremities (Table 1). The mean arm/leg frequencies were also smaller in patients with PCD than those in controls. The comparison of symptomatic and asymptomatic sides in patients with PCD is given in Table 2. The mean armswing frequency was significantly lower on symptomatic side (0,68±0,33) versus asymptomatic side (0,72±0,32, p=0,038). The mean leg or arm/leg frequencies were similar, though (Table 2).

The age, sex, handedness, weight and height of the participants showed no effect on gait and armswing parameters. The age at disease onset similarly showed no effect on these parameters. The disease duration, on the other hand, was longer in patients with slower arm/leg frequency on the asymptomatic side (p=0,042). The presence of retrocollis or anterocollis showed no effect on gait and/or armswing parameters (p=0,862).

DISCUSSION

Our study showed that the associated armswing movements during walking are bilaterally reduced in patients with primary cervical dystonia, being more prominent on the symptomatic side. There is one observational study by Kagi et al.,[3] who investigated 100 consecutive patients with PCD and observed that 55% of patients had reduced armswing; of these, 85,5% had unilateral decrease in armswing, and only 14,5% had bilaterally reduced arm swing. The quantitative analysis of armswing in our study showed that not only on the symptomatic side but bilateral armswing frequencies were affected in patients with PCD, being significantly slower than those in healthy subjects. Furthermore, the armswing frequency on the symptomatic side was slower than asymptomatic side in patients with PCD. The leg frequencies, on the other hand, were not affected, showing no significant difference between patients and controls, as well as between symptomatic and asymptomatic sides.

Primary cervical dystonia is accepted as a disorder of basal ganglia dysfunction leading to the increased brainstem interneuronal excitability via alterations in descending projections to inhibitory neurons.[6,7] Among brainstem structures, the changes in the modulation of the vestibulospinal tract are of particular interests in the pathophysiology of PCD.[8,9] Recent evidence suggested that the neuronal control of rhythmic arm movements and interlimb coordination are regulated by brainstem structures, namely the proprioceptively modulated central pattern generator activity (CPG).[10,11] These rhythmic arm movements are managed by the alternating, with some overlapping, activation of opposing muscle

groups. Such alternating activation is produced by two mutually inhibiting groups of interneurons that are facilitated by different descending systems and which in turn facilitate, for example, flexor and extensor motoneurons.[12] It has been also shown that, other than CPG activity, sensory feedback mechanisms via vestibulospinal tracts, as well as corticospinal, reticulospinal, and rubrospinal tracts in addition with and reflex control loops are able to modulate proprioceptive feedback mechanisms effective in activating appropriate limb muscles.[10,13-15] From another point of view, Mazziotta et al.[16] reported that cortical plasticity and learning-based dedifferentiation of sensory feedback information from the hand could contribute to the genesis of repetitive strain injuries and produce focal dystonia.

In a recent study by Filip et al.,[17] the authors investigated whether patients with cervical dystonia exhibited impaired performance on a motor-timing task known to require cerebellar input using a motor-timing computer task, and showed the involvement of cerebellum – although this neuronal structure is not noted traditionally as one of the major sources of dystonia development. The authors therefore suggested that the cervical dystonia patients are impaired at integrating incoming visual information with motor responses during the prediction of upcoming actions. As another target of involvement, parieto-motor cortical connectivity was also demonstrated to be impaired in cervical dystonic patients in a study of transcranial magnetic stimulation by Porcacchia et al.,[18] which was associated with slower reaching movements. Depending on reflexive-voluntary interactions, malfunctioning proprioceptive feedback was also suggested to contribute the pathophysiology of cervical dystonia.[19] In regard to these recent studies, it is now better known that the ability to manipulate normal postural reactions to head-trunk rotations is impaired in patients with cervical dystonia.

As one the limitations of our study, the study population is rather small and larger longitudinal studies are warranted to demonstrate our findings. Another important limitation is that we could not measure the amplitudes of the arm movements but only the number of armswings during a pre-set distance of walking. Although a decrease in frequency indirectly show the absence of armswing in some of the steps, a detailed computerized analysis of armswings including amplitude measurements would better demonstrate any impairment in this context.

In summary, reduced armswing might be a phenomenological feature of primary cervical dystonia caused by the subtle motor system dysfunction involving basal ganglia-brainstem pathways. On the other hand, decreased associated arm movements could simply result from the disturbed proprioceptive sensations over the dystonic cervical musculature, which leads to altered modulation of rhythmic arm movements at brainstem level. Because the decrease in armswing has now been commonly reported in many 'benign' movement disorders, the anatomical and pathophysiological mechanisms underlying the reduced armswing movements should further be studied.

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