Objective assessment of cervical dystonia: a pilot study


Objectives – The aims were to characterize the movements in cervical dystonia (CD) by using an estimate of the mechanical power and work involved in the movements and to describe this through a movement energy index (MEI). Materials and methods – The subjects (patients \( n = 6 \), controls \( n = 6 \)) were seated in front of a screen with a laser pointer attached to a headband while they performed standardized movements. A three-dimensional motion capture system was used and a test–retest was performed. Results – The mean value of MEI was significantly higher for the patients than for the controls. There was no significant difference between MEI from test to retest for the patients but there was a significant difference between MEI from test to retest for the controls. Conclusion – This study suggests that MEI could be a useful measure for the quantification of movement dysfunction in CD and thus an objective outcome measure in comparison of different therapies.

Cervical dystonia (CD) manifests itself as involuntary muscle contractions that distort the movements of the head. The resulting abnormal head and neck postures are heterogeneous in their presentation (1). The clinical spectrum of involuntary movements and abnormal postures of the head and neck in CD varies in the rhythm, speed, amplitude, duration and direction of the dystonic movements. Deviations may occur in any single plane or in combination of directions in which the head may voluntarily move (2). The pathogenesis of CD remains unclear. However, genetic factors, trauma, impaired sensory system and impaired basal ganglia function may all play a role in the development of these symptoms (1). The current and most effective treatment, intended for the amelioration of symptoms, is botulinum toxin injections (3). For many patients with CD, physiotherapy provides an important addition to the injections.

Different clinical rating scales have been proposed to quantify the movement disorder in CD. The Tsui Scale is a four-item scale that evaluates amplitude and duration of sustained involuntary neck movements, shoulder elevation and head tremor (4). The Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS) is a composite scale with subscales for clinical severity, disability and pain (5). The TWSTRS severity scale is a 10-item objective rater assessment of torticollis severity that includes five items, which are also contained in the Tsui Scale, but it does not include tremor (6). In the CD severity scale a protractor and a wall chart are used to rate the severity of the head’s deviation from neutral in each of three planes of motion, which is then scored in 5° intervals (7). Current clinical rating scales for CD have not been developed according to the guidelines by the Scientific Advisory Group of Medical Outcomes Trust, which recommends criteria for health measurement rating scales (8). According to Cano et al. (8) this fact jeopardizes the validity of findings from studies using the clinical rating scales as an outcome measure. Cano et al. (9) have recently created a reliable and valid patient-based rating scale measuring the health impact of CD.

Electromyography has been presented as a quantitative and qualitative method for identification of dystonic muscle involvement in CD (10) and for evaluating the effect of botulinum toxin injections (11). Other instrumental methods for assessing CD that are described in the literature are the use of a piezoelectric transducer to quantify measurements (12), computer graphics (13), an
To reduce variance, a procedure of matching age control group of six healthy subjects (Table 1). An investigation group of six patients with a positive design was adopted for the study comparing used when including the subjects, and a comparison in the neck muscles. A convenience selection was of 3 months following the last injection of the toxin but they were tested after a minimum interval patients were under treatment with botulinum toxin at the University Hospital in Uppsala, Sweden. All been admitted to the neurological department at Gothenburg, Sweden, located in the gait laboratory at the University Hospital in Uppsala. This is a motion capture system (Qualysys Medical AB, Gothenburg, Sweden), located in the gait laboratory at the University Hospital in Uppsala. This is a high-speed, high-resolution digital motion capture system. Four cameras were used to measure the position of the reflective markers. The subjects were seated in front of a table with their hands on the table and feet on the floor (Fig. 1). A headband was firmly fastened around the head. A laser pointer was attached to the top headband pointing forwards, and a cluster of four markers was attached to the back of the headband. The subjects pointed at a cross-shaped target on a white paper screen (140 cm × 80 cm) situated approximately 80–90 cm in front of the head (Fig. 1) with the overhead television camera (14) and a three-dimensional cervical range of motion system (15). However, these methods are rarely used in effectiveness studies in the treatment of CD. Carpaneto et al. (16) have developed an objective method to measure posture and ranges of motion in CD using a motion capture system (Fastrack). However, the most widely used motion capture systems for movement analyses are based on reflective markers and cameras that selectively track these markers (17), a method which is used in this study. The three-dimensional trajectories of the markers can be measured by using more than one camera.

Treatment with botulinum toxin to improve functional and psychological outcome and relieve pain is effective but expensive (18). The physiotherapy treatment is expensive as well, caused by recurrent periods of treatment as inpatients or outpatients at a neurology clinic, and therefore the effectiveness of the interventions needs to be evaluated. Current clinical rating scales have not been developed following recommended guidelines (8). This highlights the need for an objective way to access the impact of the disorder and the outcome of treatment.

The spasmodic and jerky movement of CD will often manifest itself in high amplitude and rapidly changing accelerations of the head. The energy needed to accelerate the head, as well as the energy needed to decelerate the head, is provided through the force generation of the muscles. The aim of our study was to quantify the irregular movements of the CD patient’s head by using an estimate of the mechanical power and work involved in the movements, described through a movement energy index (MEI), which is an objective measure for assessment of CD severity.

Objective measurement may be useful as a supplement to the clinical assessments of the individual patient.

Materials and methods

The CD subjects were adult patients with documented movement disorder who had previously been admitted to the neurological department at the University Hospital in Uppsala, Sweden. All patients were under treatment with botulinum toxin but they were tested after a minimum interval of 3 months following the last injection of the toxin in the neck muscles. A convenience selection was used when including the subjects, and a comparative design was adopted for the study comparing an investigation group of six patients with a control group of six healthy subjects (Table 1). To reduce variance, a procedure of matching age and gender was performed. To study the reliability of the motion capture system and the analysis, a test–retest was performed after 1 day (19). As the number of subjects was limited, this investigation was considered a pilot study.

The severity of dystonia was quantified (Table 1) using the individual TWSTRS severity subscale (6) and the Tsui scale (4). Scores for the TWSTRS severity subscale range from 0 to 35 where 35 represents severe excursion. Scores for the Tsui scale range from 0 to 25 where 25 represents severe dystonia. The TWSTRS severity subscale and the Tsui scale are clinician-based outcome measures (4, 5). The TWSTRS severity subscale contains the judgement of the maximal excursion besides a duration factor, effect of sensory tricks, shoulder displacement, range of motion and time for which the patient is able to maintain the head within 10° of neutral position (5). The Tsui scale contains, besides amplitude of sustained movements, the judgement of duration of sustained movement, shoulder elevation and tremor (4). The TWSTRS has been tested for reliability and validity with acceptable results (2). No formal test for validity has been performed for the Tsui scale but the reliability is considered to be good (4).

All the subjects performed a number of pre-defined standardized movements with the head. The movements were measured using a ProReflex motion capture system (Qualysys Medical AB, Gothenburg, Sweden), located in the gait laboratory at the University Hospital in Uppsala. This is a high-speed, high-resolution digital motion capture system. Four cameras were used to measure the position of the reflective markers. The subjects were seated in front of a table with their hands on the table and feet on the floor (Fig. 1). A headband was firmly fastened around the head. A laser pointer was attached to the top headband pointing forwards, and a cluster of four markers was attached to the back of the headband. The subjects pointed at a cross-shaped target on a white paper screen (140 cm × 80 cm) situated approximately 80–90 cm in front of the head (Fig. 1) with the...
The procedure started with the subjects maintaining the beam pointed at the centre of the cross for 4 s. This was followed by 12 trials where the subjects were asked to trace horizontal and vertical lines as fast as possible, with sufficient inter-trial rest. The subjects attempted to track a horizontal black line three times from left to right and three times from right to left. This was followed by movements in a vertical direction. Three movements were made from the top to the bottom of the screen (flexion) and three movements were made from the bottom to the top (extension) over a range of approximate 90°.

Calculation of MEI

The energy index used in this article is a measure that is proportional to the mechanical work required to obtain the measured movement of the head. To obtain a measure that can be used for comparison between subjects, mechanical work needs to be modified to account both for the differences in body mass and for the difference in the speed with which the subjects performed the task.

Translations of the head are small in the experiments and are assumed to be negligible. In the following discussion we consider only rotations of the head about its centre of mass. We did not measure the centre of mass. As is clear from the rest of the derivation, it is not needed. Consider the resultant torque (sum of torques produced by agonist against antagonist muscles) that generates the rotation of the head. The mechanical power produced by this torque is given as the scalar product of the torque, T and the angular velocity of the head, ω:

\[ P = T \cdot \omega. \]

The angular velocity vector is estimated directly from the marker data, whereas the torque vector is estimated from the marker data using inverse dynamics. This method estimates the internal forces and torques (the torque T, in our case) by applying Newton’s second law of motion.

To derive the torque it is necessary to know the acceleration of the head as well as the external forces acting on it. The only external force acting on the head is gravity and as it acts through the centre of mass, it does not contribute to the torque. The net joint torque is therefore equal to the moment of inertia of the head times the angular acceleration:

\[ T = I\ddot{\theta} = I\ddot{\alpha}, \]

where \( \alpha \) denotes the angular acceleration, and \( I \) is with respect to the centre of mass. To reduce the variability due to difference in body mass of different subjects, a scaling of the torque with respect to the inertial property of the head is motivated. The reason being that two different subjects should obtain the same value when performing the task identically. This simplifies the expression of the mechanical power to:

\[ P_s = I^{-1}T \cdot \omega = \alpha \cdot \omega. \]

The expression may be denoted scaled power. It is positive if the acceleration and velocity vectors have the same direction, and negative if the directions are opposite.

The (scaled) work performed in an interval of time is computed by integrating the previous expression:

\[ W_s = \int_{t_1}^{t_2} P_s dt = \int_{t_1}^{t_2} \alpha \cdot \omega dt. \]

Assuming that the storing and releasing of elastic energy in ligaments and tendons can be neglected for the movement under study, then both positive and negative power (and resulting positive and negative work) are the result of actions by the muscles. As it is the complete work performed by the muscles we want to approximate, whether it produces positive or negative power, the absolute value of the power is considered:

\[ P_a = |\alpha \cdot \omega|. \]
With the assumptions made, the $W_a$ measure, when calculated over a trial, is related to the mechanical energy required by the muscles when producing the movement.

The subjects were asked to perform the task (tracking horizontal and vertical lines) as fast as possible. To account for differences in the time used to complete the task, $W_a$ was divided by the duration of the trial, thus in effect computing the average scaled power $P_a$. This gives the proposed movement energy index:

$$\text{MEI} = \frac{\int_{t_1}^{t_2} |\alpha \cdot \omega| \, dt}{t_2 - t_1}$$

The calculation of the MEI was implemented in software using the Java programming language.

Data analysis was based on the mean values of the repeated movements in each direction. For comparison between the groups, mean values were calculated from the movements of both test and retest, whereas for the test–retest comparison, mean values were calculated separately for each of the two occasions. Comparison of the MEI mean values for each subject and each direction of the two groups was made with the Mann–Whitney $U$-test. The test–retest comparison was made using the Wilcoxon matched-pairs signed ranks test.

The procedure in the present study was in accordance with current ethical standards. Subjects were given information saying that participation was voluntary and that they could drop out of the study at any time without having to give a reason.

**Results**

The MEI varied between 0.004 and 0.65 for the CD patients in the centre position and between 0.06 and 3.99 for the active movements. For the controls the index varied between 0.001 and 0.008 in the centre position and between 0.04 and 0.55 for the active movements (Table 2).

There was a significant difference between the groups concerning the MEI values in all movement directions ($P < 0.01$). The mean value of the MEI was significantly higher for the CD patients (Fig. 2). It is also clear from Fig. 2 that, for almost all the subjects in this study, performing active movements of the head tended to demand more energy than keeping the head in the centre. There was no significant difference between the mean values of MEI for all directions, from test to retest for the CD patients. However, there was a significant difference between the mean values of MEI from test and retest for the controls ($P < 0.05$), MEI mean values being consequently

![Figure 2. Mean values of movement energy index (MEI) for each subject and each direction. The controls are represented by dots and patients by crosses. The $x$-axis describes the standardized movements. The $y$-axis contains MEI on a logarithmic scale.](image-url)

<table>
<thead>
<tr>
<th>CD ($n = 6$)</th>
<th>Controls ($n = 6$)</th>
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<tbody>
<tr>
<td>Test Mean</td>
<td>Range</td>
</tr>
<tr>
<td>C</td>
<td>0.16</td>
</tr>
<tr>
<td>L → R</td>
<td>0.80</td>
</tr>
<tr>
<td>R → L</td>
<td>0.52</td>
</tr>
<tr>
<td>U → D</td>
<td>0.31</td>
</tr>
<tr>
<td>D → U</td>
<td>0.20</td>
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</tbody>
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The left column indicates the directions of the movements: C, centre; L, left; R, right; U, up; D, down.
higher in the test than in the re-test. There was only some overlap between CD patients and controls, as illustrated in Fig. 2. One CD patient performed the left to right movement within the bounds of the healthy subjects and another CD patient performed the down–up movement within the bounds of the healthy subjects.

Scores from MEI in the centre position had a moderate correlation with scores from the Tsui scale ($r_s = 0.7$) but there was no correlation between scores from the active movements of MEI and the Tsui scale. There was also a moderate correlation between the MEI scores for active movements and scores from TWSTRS severity subscale ($r_s = 0.6$) but there was no correlation between the centre position and the TWSTRS severity subscale. There was no correlation analyses made between MEI and the TWSTRS disability or pain sections.

### Discussion

The results of this investigation showed a significant difference for MEI between a group of patients with CD and a group of healthy volunteers. In general, the movements performed by the CD patients required more energy than the movements of the healthy subjects. However, there was a slight overlap between the groups, which means that some CD patients were able to perform movements of the head within the bounds of the healthy subjects for some of the trajectories. It is interesting to note that the results showed no significant difference between MEI from test to re-test for the CD patients. However, there was a significant difference between MEI for the controls. This might be due to an effect of learning, which the CD patients were unable to obtain. The movement disorder seemed to dominate over the effect of learning. The long-term effects of botulinum toxin injection on the performance of the voluntary movements of the head cannot be completely excluded.

Berardelli et al. (20) indicated in a review article that the twisting movements of dystonia are characterized by co-contraction of agonist and antagonist muscles and that voluntary movement have to exacerbate the co-contraction of the antagonist muscle pairs. The co-contractions may then contribute considerably to the energy required in the head movements. Therefore, the MEI probably underestimates the energy required for the head movements in CD. However, the study emphasizes the significant difference in the MEI values for the CD patients and the healthy controls.

It is important to create reference data for outcome measures before large studies are performed. The MEI values for the healthy subjects showed little distribution, which might indicate the likely range of MEI values one could expect for active head movements in healthy subjects. However, more healthy subjects need to be measured to estimate norm values.

A sample size calculation was not carried out beforehand and further studies are needed for being able to consider what differences in the index as being meaningful.

We used a non-parametric hypothesis test (Mann–Whitney $U$-test), which is independent of the magnitude of the difference. For this test, the smallest sample size that can show a significance difference between two groups is six individuals in each group. A difference between the groups is significant only if all the pairs show a similar difference (all positive or all negative).

The voluntary movements of the head are, to some extent, restricted in CD according to Carpeneto et al. (16). Besides the increase in the voluntary range of motion after treatment with botulinum toxin and/or physiotherapy, patients with CD often find it less demanding to perform the voluntary movements compared with before treatment. Maybe the use of an energy concept, applied as an MEI in this study, might capture the subjective feeling of an easier and smoother way to perform active movements for CD patients after treatment if measured before and after treatment. To check this hypothesis further studies are needed.

The MEI focuses on the performance of active movements of the head in CD, more specifically quantification of these movements. The clinical rating scales used in this study focus on the severity of the disorder. This difference in contents with MEI and the clinical rating scales may explain the moderate correlations between the scores from the clinical rating scales and MEI.

Motor dysfunction in patients with CD seems to lead to excessive effort, beyond what is necessary for performing these simple movement tasks. It is important to consider not only the degree to which a patient can carry out a task but how and with what effort the patient performs the task (21). MEI therefore seems to be one useful outcome measure for detecting and quantifying the movement disorder in CD. This objective measure could have a wide area of application in the field of CD. It could be used to measure the effect of therapy and to consider a change in the therapeutic approach. It could also help in communicating the results of treatment to patients and other health professionals.

The MEI would be practical enough to be considered in a larger clinical trial ($n > 50$) with
two or more measurement occasions. The assessment situation, including the analyses in the ‘head motion program’, requires 20–30 min for one patient. This time needed for MEI is greater when compared with a conventional clinical rating scale, e.g. the Tsui scale. However, the conventional clinical rating scales seem to assess different dimensions of CD.

Consequently, the advantage of using the motion capture system used in this study was the increased possibility of a more standardized and objective measure. With appropriate software tools the method is easy to use. One limitation of the method is the requirement of a sitting position. Some patients find that the symptoms of CD are maximal during dynamic activity such as walking.

The MEI focuses on the external aspect of CD and so far it is not known if this measure can capture changes that are important for the patients themselves. Certainly, there is an association between impairment and functional well-being, but the two are not identical. From the patient’s point of view, motor disturbances are not the only important aspects of the disorder, but also the impact of the disorder on the ability to perform daily activities or to fulfil social roles (22). This also needs to be investigated.

In conclusion, MEI can be a useful alternative measure for detecting and quantifying movement disorder in CD. MEI provides an objective assessment of the movement disorder in CD and could supplement existing scoring systems based on clinical judgement. It may enable an objective outcome measure of the response to treatment and in comparing the effectiveness of different therapies.

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References