Physiotherapy in cervical dystonia: Six experimental single-case studies

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Physiotherapy in cervical dystonia: Six experimental single-case studies

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The aim of the study was to explore the outcome of a physiotherapy program targeted to improve the quality of life of people with cervical dystonia (CD) by reducing pain, improving awareness of postural orientation, increasing muscle strength, and reducing the effort of moving the head and neck. In six single case studies, the primary outcome measure for each case was the Cervical Dystonia Questionnaire (CDQ) to measure the impact of the program on the individuals’ quality of life. Secondary outcome measures were identified for the different components of the physiotherapy program: Visual Analogue Scale (pain); Postural Orientation Index (postural orientation awareness); and Movement Energy Index (effort of moving head and neck). Each of the cases had the severity of their problems scored on the Toronto Western Spasmodic Torticollis Scale. The study period was 26 weeks: 2 weeks’ baseline period, 4 weeks’ treatment period, and 20 weeks’ follow-up. All measures except the Movement Energy Index (MEI) and CDQ-24 were taken three times per week for the first 6 weeks of the study and then once at 3 and 6 months. The MEI was taken once a week during the pretreatment and the treatment periods and during the first 2 weeks of follow-up and also after 3 and 6 months of follow-up. The CDQ-24 was taken once in the pretreatment period, once after completion of treatment, once 2 weeks after treatment, and once at 3 and 6 months of follow-up. Five of the six case studies reported an increase in quality of life at 6-month follow-up, as measured on the CDQ-24. Three of the six cases reported a reduction in pain and severity of the dystonia and had improved scores on the postural orientation measure at 6-month follow-up. All six patients had a reduction in the movement energy scores, but this was not significant. The outcomes of the six case studies would suggest that further investigation is required to show the effectiveness of physiotherapy programs in the management of CD.
Introduction

Cervical dystonia (CD) has been defined as involuntary twisting and turning of the neck caused by abnormal involuntary muscle contractions (Fahn, Marsden, and Calne, 1987). Deviations may occur in any single plane or in combinations of directions. The base of the neck may show sagittal or lateral deviations from the midline. Frequently, the shoulder is elevated and displaced anteriorly on the side toward which the chin is pointing (Consky and Lang, 1994). The abnormal involuntary twisting movements are characterized by co-contraction of agonist and antagonist muscles. Voluntary movement exacerbates the co-contraction (Berardelli et al, 1998). Tremor and pain are pronounced in CD (Kutvonen, Dastidar, and Nurmikko, 1997; Pal et al, 2000). Previous studies have shown impairment of health-related quality of life compared with general population data (Ben-Shlomo et al, 2002; Camfield et al, 2002; Müller et al, 2002).

The current treatment of choice consists of botulinum toxin (BTX) injections (Jankovic, 2004), with physiotherapy providing an important adjunct. In a survey study in Sweden (Silfors and Solders, 2002), the patients ranked physiotherapy as the second most effective treatment after BTX. There is limited literature on the effectiveness of physiotherapy in the management of CD. Electromyographic biofeedback in relaxation training did not prove effective in one study (Duddy and McLellan, 1995). Physiotherapy comprising passive elongation of myofascial cervical structures and exercises for postural reeducation was compared with electromyographic biofeedback in four patients, with comparable results for the two treatment programs (Smania et al, 2003). Ramdharry (2006) presented a case report outlining the rehabilitation of a patient who was able to reduce the effective dose of BTX when physiotherapy was added to a pharmacological regime. A randomized, crossover study showed that combination of a physiotherapy program with BTX treatment increased the beneficial effect of BTX, decreased perceived pain, and improved functioning of activities of daily living in 20 patients compared with treatment with BTX alone (Tassorelli et al, 2006). In the light of the sparsity of previous investigations, there seemed to be a need to develop a physiotherapy program that would be useful for patients with CD. This series of six single case studies on subjects with CD was conducted to explore the outcome of a physiotherapy program targeted to improve the quality of life of people with CD by 1) reducing neck pain, 2) improving awareness of postural orientation, and 3) increasing muscle strength and reducing the effort of moving the head and neck.

Postural orientation was, in this study, defined as “the ability to maintain an appropriate relationship between the body segments and between the body and the environment for a task” (Shumway-Cook and Woollacott, 2001).

Method

Subjects

A sample of six patients with documented CD of varying severity, scored with the Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS) (Consky et al, 1990; Consky and Lang, 1994) (M = 49 points, range = 34–62), age (M = 48.5 years, range = 30–59), and gender (4 men, 2 women) was recruited from the waiting list for admission to the physiotherapy department of the University Hospital in Uppsala, Sweden for a 4-week inpatient program (Table 1). All the subjects had been referred to physiotherapy as in-patients by a neurologist at the mentioned hospital. Current clinical practice in this unit with physiotherapy treatment for CD as in-patients was followed. The subjects were under no other medical treatment and had not been treated with BTX during the last 3 months, at least, prior to the study.

Design

This study had an experimental multiple case study series using an ABA design (i.e., baseline no treatment; treatment; and follow-up no treatment with continuous assessment before and during treatment and during follow-up). The subjects were their own controls. The length of the pretreatment baseline period was 2 weeks, and the treatment period lasted for 4 weeks. Follow-up was performed at three points in time during a 2-week period after the end of the treatment, once 3 months after the treatment and once 6 months after the treatment. In
summary, three assessments were made during each of the above weeks, and one assessment on each of the last two follow-up occasions, which meant 26 assessments. The key characteristics of the single-case design (i.e., continuous assessment, baseline assessment, stability of performance, and the use of different assessment periods) provide rigorous control and enhance internal validity, thus permitting causal inferences to be drawn in each individual case (Kazdin, 2003). The local ethics committee at the Faculty of Medicine in Uppsala, Sweden, approved the project.

Overview of the physiotherapy program

The overarching goal of the physiotherapy program (Appendix 1) was to improve quality of life by reducing neck pain, improving awareness of postural orientation, increasing muscle strength, and reducing movement energy. The program included progressive muscle relaxation (PMR) (Gessel, 1989), exercises for isometric muscle endurance and dynamic strength (ACSM, 2000), exercises for improving coordination, balance and body perception, and muscle-stretching exercises. The treatment was scheduled to take place over a 4-week period to comply with the routines at the hospital and consisted of four phases, each lasting 1 week. The phases were the same for all patients, but the contents of each phase differed somewhat on the basis of each individual’s needs. PMR was introduced during the first phase. Given that many patients with CD experience pain, it was a major concern that the pain should be reduced initially by PMR. PMR could also be helpful in increasing subjective body perception. The rationale of the PMR procedure is that there is a relationship between muscle tension and pain. A cycle may develop whereby the muscle tension and pain act to intensify each other. Techniques such as PMR, which may result in a reduction of skeletal muscle tension, may have the potential to break this cycle and give pain relief. By promoting an awareness of the body and an understanding of the signals from the body, the PMR technique might influence the effect on pain. It may, however, have the effect of enhancing coping rather than having a specific effect on pain, which may have an impact on the patient’s overall well-being and quality of life (Carroll, 1998). In our program, once the pain had decreased and the subjective body perception increased, exercises for isometric muscle endurance and dynamic strength in the neck were added in a second phase. The exercises were mainly designed to strengthen the muscles that opposed the posture resulting from the dystonic muscle activity. The strength training resulted in more coordination of the neck muscles.

In the third phase, a variety of exercises were introduced with the aim of improving coordination, balance, and body perception, and hence overall postural orientation.

In the fourth phase, muscle-stretching exercises were applied. This order of sequence of the exercise phases was based on the concept that

<table>
<thead>
<tr>
<th>Patient</th>
<th>Sex</th>
<th>Age/years</th>
<th>Illness duration/years</th>
<th>Severity of CD/TWSTRS</th>
<th>Neckpain/VAS</th>
<th>Time from BTX/months</th>
<th>Physical trauma</th>
<th>Tremor</th>
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<tr>
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</tr>
<tr>
<td>2</td>
<td>F</td>
<td>30</td>
<td>5</td>
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<td>No</td>
</tr>
<tr>
<td>3</td>
<td>M</td>
<td>55</td>
<td>16</td>
<td>61.75</td>
<td>9</td>
<td>Never BTX</td>
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<td>Yes</td>
</tr>
<tr>
<td>4</td>
<td>M</td>
<td>59</td>
<td>6</td>
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<td>56</td>
<td>9</td>
<td>46</td>
<td>4</td>
<td>&gt; 6</td>
<td>No</td>
<td>Yes</td>
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</tbody>
</table>

M, male; F, female; TWSTRS: Toronto Western Spasmodic Torticollis Rating Scale (Consky et al., 1990) maximum points = 85; VAS: Visual Analogue Scale (Jensen and Karoly, 2001); BTX, botulinum toxin.
reduced pain, reduced severity of dystonia, decreased disability, and increased body perception, and thus improved postural orientation, would give the best conditions for muscle-stretching exercises, thus lowering the energy consumption when the patient performed voluntary movements of the head (Appendix 1).

Two physiotherapists, experienced in the field of CD, provided the intervention. The patients met the same physiotherapist at 45-minute sessions twice a day, except on Fridays, when there was only one session. Exercises managed by the patients themselves were introduced according to each individual’s need and were supervised by the physiotherapist. The program had a cumulative character, and the conditions in the individual patient guided the therapist in the number of repetitions, the body positions, and the time needed for each exercise during the therapy sessions.

Measures

Cranio cervical Dystonia Questionnaire (CDQ-24)

CDQ-24 is a disease-specific questionnaire designed to evaluate quality of life in patients with CD or blepharospasm (Müller et al, 2004). It contains 24 items with five subscales: stigma, emotional well-being, pain, activities of daily living, and social/family life. According to the authors, high scores on this measure (maximum 96 points, non-transformed) indicate negative outcome and reflect low quality of life. Internal consistency reliability is satisfactory for all subscales, and test-retest reliability is generally high.

Visual Analogue Scale (VAS)

The VAS was used for rating current pain in the neck (Jensen and Karoly, 2001). VAS scores range from 0 = no pain to 10 = worst imaginable pain and thus a negative outcome. VAS has been widely used in pain research and shows good construct validity and good sensitivity to treatment.

Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS)

The TWSTRS is a clinical rating scale that assesses severity, disability, and pain in CD (Consky et al, 1990; Consky and Lang, 1994). TWSTRS combines a physician-based severity scale (0–35), with patient-based scales for disability (0–30) and pain (0–20) with a total score of 85 points. High scores on this scale indicate negative outcome and reflect higher levels of severity, disability, and pain. In a study by Salvia et al (2006), the interobserver reliability for the total score of TWSTRS was excellent ($r_s = 0.99$), and for the disability and the pain scales it was good ($r_s > 0.88$); for the global severity scale, however, the reliability was defined as moderate ($r_s = 0.63$).

Cervical Dystonia Postural Orientation Index (POI)

The POI represents a self-rated measure of the ability for postural orientation in nine standardized positions, including standing and sitting, developed by one of the authors (LZ). Each position is rated on a horizontal 11-grade numerical scale where 0 = no difficulty in holding the position and 10 = impossibility of holding the position. High scores on this scale (maximum 90 points) reflect negative outcome with decreased ability for postural orientation. No formal testing of the reliability and validity of the scale has yet been performed. Face validity has been ensured through peer-review and patient feedback sessions.

Movement Energy Index (MEI)

The MEI is a measure of the energy expenditure in active movements of the head as measured with a three-dimensional motion capture system (Qualisys AB, Gothenburg, Sweden). A high score indicates negative outcome and a movement that is energetic in excess of what is needed for the task at hand (voluntary movements of the head in the sagittal and the horizontal planes). In a methodological pilot study (Zetterberg et al, 2005), a mean value of 0.167 was found for the controls (SD = 0.188). A comparison revealed no significant difference between test and retest in patients with CD. However, the control group, while scoring significantly lower than the CD group, showed a small decrease from test to retest, suggesting a learning effect. Even if we cannot rule out such...
a learning effect in the CD patients, it is plausible that the effect will diminish quickly once the patients become familiar with the test situation and the personnel.

Time frame for taking measurements

Current pain intensity in the neck scored on VAS, severity of dystonia, disability and pain rated on TWSTRS, and ability for postural orientation scored on POI were measured three times a week during all phases and once at the follow-up assessment after 3 and 6 months. The head movement evaluations with MEI were carried out once a week during the pretreatment and the treatment periods and during the first 2 weeks of follow-up, and also after 3 and 6 months of follow-up. Quality of life was scored on CDQ-24 once in the pretreatment period, once after completion of treatment, once 2 weeks after treatment, and once at 3 and 6 months of follow-up. The measures were taken in the mornings always before the physiotherapy treatment started for the day. One of the authors (LZ), who was not treating the patients, performed the testing.

Data analyses

Data related to dependent variables in each patient were plotted in graphs and visually interpreted with regard to trend. Linear regression analysis was used to test the significance of trend changes from baseline to treatment and from treatment to follow-up. The regression lines for MEI had to be interpreted with caution because of the few measurement occasions. Wilcoxon signed rank test was used to test any differences in CDQ-24 results between periods.

Results

The results for VAS, TWSTRS, POI, and MEI are presented in graphs, two for each patient, in Figures 1a–f. The first graph gives scores and regression lines from the pretreatment baseline, treatment, and short-term follow-up periods. The second graph also shows the 3- and 6-month follow-up assessments. The four phases of the treatment (T1–T4) are shaded. It should be noted that the regression lines for MEI are based on fewer data points than those for the other measurements and are therefore more sensitive to variability in the measure. The results for CDQ-24 are presented in Table 2.

In the following, the results are given for each individual patient regarding trends and regression lines for the different measures, as observed in the graphs, and changes in trends with p values, as derived from statistical analyses.

Patient 1

The trend for VAS was slightly negative (unfavorable) during treatment and vaguely positive (favorable) during follow-up. The trends for TWSTRS and POI were slightly positive during treatment and continued to be positive during follow-up. MEI did not change during treatment but displayed a slightly negative trend at follow-up.

The statistical analyses showed a significant positive change in trend between the treatment period and the follow-up period concerning both VAS ($p = 0.016$) and TWSTRS ($p = 0.039$). Scores for POI displayed positive significant changes in trends both between the pretreatment period and the treatment periods ($p = 0.008$) and between the treatment period and follow-up ($p = 0.037$).

The results for CDQ-24 scores are presented in Table 2, where it is seen that the scores after treatment and after 2 weeks, 3 months, and 6 months of follow-up were all decreased compared with those in the pretreatment period.

Patient 2

VAS showed a positive trend during the pretreatment period and a stable positive trend during treatment, which persisted at follow-up. For TWSTRS, there was a positive trend during the pretreatment period and this continued during treatment. A minor negative trend was discernible during follow-up. POI and MEI exhibited slightly more positive trends during treatment compared with the pretreatment period. The trend for POI continued in a positive direction at follow-up, whereas the follow-up trend for MEI was slightly negative.
Statistical analysis showed a significant positive change in trend for VAS during the treatment compared with the pretreatment period ($p = 0.049$). The scores for TWSTRS showed a significant negative change in trend, with increasing scores in the follow-up period compared with the treatment period ($p = 0.030$). A significant positive change in trend between the pretreatment and follow-up periods was observed for POI ($p = 0.030$) and MEI ($p = 0.019$).}

![Figure 1.](image-url) Figure 1. (a–f) Graphs for each patient with scores and regression lines from the Visual Analogue Scale (VAS) (Jensen and Karoly, 2001); Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS) (Consky et al, 1990); Postural Orientation Index (POI); and Movement Energy Index (MEI) (Zetterberg et al, 2005). The first graph illustrates scores for pretreatment (baseline; days 1–14), treatment (T1–T4) and follow-up (days 43–56). The second graph also illustrates scores for pretreatment, treatment, and follow-up but with scores in addition at 3- and 6-month follow-up. The different phases of the treatment (T1–T4) are shaded.
and the treatment periods was noted for POI ($p = 0.030$).

The CDQ-24 scores, measuring quality of life, were reduced during the treatment and follow-up periods compared with the pretreatment scores (Table 2).

**Patient 3**

VAS showed a minor positive trend during the pretreatment period and a distinct positive trend during treatment. A negative trend was observed throughout the follow-up. In the
graphs for TWSTRS scores, there was a positive trend during the pretreatment period, and this continued during the weeks of treatment. During follow-up, a distinct negative trend was evident. For POI, a positive trend was observed during treatment, in contrast to a lack of change in the pretreatment period, and the positive trend remained stable during follow-up. The opposite was seen for MEI, with a negative trend during the 4 weeks of treatment.

The statistical analyses showed significant negative changes in trends of scores for VAS ($p < 0.0001$), TWSTRS ($p = 0.0001$), and MEI ($p = 0.0003$) at follow-up compared with the treatment period. Scores for POI showed slightly positive significant changes in trends at follow-up.
compared with treatment ($p = 0.0002$). For MEI, there was significant positive change in trend at follow-up compared with the treatment period ($p = 0.0003$).

This patient had reduced scores for quality of life, as measured with CDQ-24, during all assessments periods (Table 2).

**Patient 4**

The VAS scores showed a straight negative trend before treatment and a clear positive trend during treatment. In the follow-up period, the trend had a negative direction. For TWSTRS, there was a minor negative trend during the
pretreatment period, a positive trend during treatment, and a negative trend at follow-up. POI displayed a slightly positive trend during treatment, but the trend became slightly negative during follow-up. The graphs for MEI illustrate a clearly positive trend before treatment but a distinct change in level and trend during the treatment period. The trend during follow-up was positively stable.

On statistical analysis, VAS showed a significant positive change between pretreatment and treatment ($p=0.003$) and a significant negative change between treatment and follow-up ($p=0.010$). Scores from TWSTRS changed...
significantly in the negative direction between treatment and follow-up \((p = 0.047)\), and this was also seen for POI \((p = 0.040)\).

This patient experienced a favorable marginal effect in quality of life as measured with CDQ-24, with similar scores during all assessment periods. An increased score was noted, however, at the 6-month follow-up (Table 2).

**Patient 5**

The trends for VAS as illustrated on the graphs were very similar during the different periods. The trend for TWSTRS was negative during the pretreatment period but showed an obvious positive trend during treatment. POI and MEI followed the same changes in trends,
with negative trends, before treatment and a positive trend during treatment. During follow-up, the trends for POI and TWSTRS were again negative. MEI showed a stable trend during follow-up, with no changes compared with the end of treatment.

The statistical analysis revealed a significant positive change in trend for TWSTRS between the pretreatment and treatment periods \( (p = 0.001) \). When the follow-up scores for TWSTRS were compared with the treatment scores, a significant negative change in trend \( (p = 0.002) \) was observed. The POI score showed a significant positive change in trend in the treatment period compared with the pretreatment scores \( (p = 0.007) \), and a negative change in trend was noted at follow-up compared with the treatment scores \( (p = 0.023) \).

Compared with the pretreatment scores, the CDQ-24 scores after treatment and at 2 weeks’ and 3 months’ follow-up were all reduced, and the same was found at the 6-month follow-up (Table 2).

### Patient 6

The scores for VAS, TWSTRS, and POI showed positive changes in trends during treatment compared with the pretreatment period, but negative trends were noted during follow-up. For MEI, there was a positive trend before treatment and also to some extent during treatment and during follow-up.

The statistical analyses disclosed significant positive changes in trends for VAS \( (p = 0.005) \) and POI \( (p = 0.0003) \) during treatment compared with the pretreatment period. During follow-up, changes in trends were observed for VAS \( (p = 0.0005) \) and TWSTRS \( (p = 0.0004) \) in comparison with the trends during treatment.

This patient exhibited reduced scores for CDQ-24 during treatment and at follow-up, but the score at the 6-month follow-up was almost the same as that before treatment (Table 2).

All patients, except one, were found to have reduced scores for CDQ-24 (Table 2), indicating increased quality of life, after treatment compared with the pretreatment scores. Statistical analysis confirmed this finding and showed significant changes in the whole group when pretreatment was compared with treatment \( (p = 0.027) \), follow-up after 2 weeks \( (p = 0.046) \), and follow-up after 3 months \( (p = 0.028) \). No

### Table 2. Total non-transformed scores from the cranio-cervical dystonia questionnaire (CDQ-24) (Müller et al, 2004), maximum non-transformed points = 96, for each patient at pretreatment (Pre), post treatment (Post), and at follow-up at 2 weeks (F1), 3 months (F2), and 6 months (F3).

<table>
<thead>
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<th>Patient</th>
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<th>Post</th>
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<tr>
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<td>48</td>
<td>29</td>
<td>23</td>
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<td>46</td>
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</tbody>
</table>

### Table 3. Raw data from pretreatment (Pre), post treatment (Post), and follow-up at 6 months of Visual Analogue Scale (VAS) (Jensen and Karoly, 2001); Toronto Western Spasmodic Torticollis Scale (TWSTRS) (Consky et al, 1990); Postural Orientation Index (POI); and Movement Energy Index (MEI) (Zetterberg et al, 2005) for each of the six patients.

<table>
<thead>
<tr>
<th>Patient</th>
<th>VAS Pre</th>
<th>VAS Post</th>
<th>TWSTRS Pre</th>
<th>TWSTRS Post</th>
<th>POI Pre</th>
<th>POI Post</th>
<th>MEI Pre</th>
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<td>Post</td>
<td>6 months</td>
<td>Pre</td>
<td>Post</td>
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significant change was established between the pretreatment scores and the scores at follow-up after 6 months ($p = 0.074$).

Raw data of VAS, TWSTRS, POI, and MEI from one occasion before treatment, after treatment, and at 6 months’ follow-up are presented in Table 3.

**Discussion**

This series of six single-case studies on subjects with CD was conducted to explore the impact on a 4-week inpatient physiotherapy program on the quality of life by reducing pain, improving awareness of postural orientation, increasing muscle strength, and reducing the effort of moving the head and neck. All patients who participated in this study displayed generally positive effects after a 4-week period of physiotherapy in terms of neck pain, severity of dystonia, disability, postural orientation, movement energy expenditure, and quality of life.

The main effect of the physiotherapy program in our study was that the quality of life, as measured with CDQ-24, increased in all the participants, except one, during the study period and that the improvement was maintained at the 6-month follow-up.

Higher TWSTRS total scores are explaining reduced health-related quality of life (Skogseid, Malt, Røislien, and Kerty, 2007). The participant in our study who did not experience improved quality of life scored 62 points out of 85 on TWSTRS at pretreatment and demonstrated only marginal reduction in these scores during treatment and follow-up.

The main predictors of quality of life in dystonia have been identified as poor body concept, sense of disfigurement, functional disability, and depression. When improving health care for patients with dystonia, efforts should not only focus on reducing the severity of dystonia but also consider factors critical for good quality of life (Page, Butler, and Jahanshahi, 2007). The result from the physiotherapy program given in this study support the statement by Page, Butler, and Jahanshahi (2007), saying that interventions aimed at modifying the negative body concept and sense of disfigurement may have a large impact on improving quality of life in dystonia. One case report (Faircloth and Reid, 2006) suggests that a cognitive-behavioral approach could be helpful and effective in the management of CD where there is increased health anxiety. The study recognized the organic origin of the dystonia but emphasised the role of psychological factors maintaining and exacerbating symptoms and disability. Jahanshahi and Marsden (1990) also stated that because patients with CD may have a negative body concept and exhibit high levels of functional disability, a cognitive-behavioral intervention would be a suitable approach for the management of depression, negative body concept, and disability for these patients.

To identify factors predicting disability, possibly to influence, is a task for future studies in CD. A challenge is to perform a self-management program to perceive the positive effect of increased quality of life experienced during a physiotherapy program as in this study.

Because of the sparsity of studies in this area, the clinical physiotherapist is left with little guidance on which symptoms in CD respond to what treatment approach. The current PT program was divided into four phases in all patients but with individual setups. The basic rationale underlying the order of sequence of the treatment phases was that PMR was meant to reduce neck pain but also to enhance body perception and increase awareness of underused muscles. There is no documented evidence of effectiveness of PMR in reducing neck pain in patients with CD. However, PMR is one component of pain relief provided for patients with pain from cancer (Sloman, 1995) and osteoarthritis (Baird and Sands, 2004). Our study showed that three patients experienced a reduction of neck pain as scored on VAS during treatment. Two patients scored 2 on VAS for neck pain at pretreatment and that must be considered as a limited possibility for improvement. Their main problem was not neck pain.

Once pain was alleviated and body perception of the underused muscles had begun, the program could move on into more complex exercises. In our experience, strengthening of the underused muscles in the neck help to resist dystonia and facilitate voluntary movements in the direction toward the dystonic side of the head. The benefit of this training achieved during treatment for the patients, however, was not maintained during follow-up, when TWSTRS
demonstrated increased severity, disability, and pain compared with the findings during the treatment period. However, three patients did not reach as high TWSTRS scores at 6 months’ follow-up as during pretreatment.

It has been claimed by Anastopoulos, Nasios, Mergner, and Maurer (2003) that patients with CD have a somewhat inaccurate knowledge of their head posture but can effectively use neck proprioceptive input and vestibular cues when estimating head and trunk displacement. To improve postural orientation in patients with CD, stimulation of the vestibular, somatosensory, and visual systems should therefore include a variation of voluntary movement exercises as was done during phase three. Four patients showed improvement of postural orientation during treatment, indicating that these exercises had a positive effect on the ability to maintain a more appropriate relationship between the body segments and the environment for a task.

It is important to consider not only the degree to which a patient can carry out a movement but how and with what effort the patient performs the movement. All six patients had a reduction in the movement energy scores recorded at post-treatment, but this was not significant. Maybe the use of an energy concept, applied as MEI, captures a subjective feeling of less demanding active movements with the head after 4 weeks of physiotherapy as applied in this study.

The outcome measures were chosen with the expectation of capturing different aspects of CD. CDQ-24 addressed the special problems of patients with craniocervical dystonia regarding quality of life (Müller et al, 2004). On the basis of recent literature, Cano et al. (2004) recommended TWSTRS as the current clinician-based outcome measure of choice for CD. POI was developed prior to the start of the study to assess the concept of postural orientation. The present lack of reliability and validity of the POI instrument could affect the interpretation of the result. MEI is a measure of active head movements and is related to the energy expended during the movements. This index must be regarded as a snapshot of the patient’s ability to perform active head movements at the time of assessment and is therefore affected by day-to-day variation in this ability.

In clinical physiotherapy the focus is on the individual patient. The key characteristics of experimental single-case designs are baseline assessments, continuous assessment, stability of performance, and use of different phases (Kazdin, 2003). It would have been preferable to have longer phases with additional assessments to more clearly show any causal relationships and to strengthen the validity of the results. Specifically, an extended pretreatment period would have given a better picture of the status of each patient prior to the intervention. However, for practical reasons, the preferred guidelines of the design could not be completely achieved.

Very little work has been conducted in this area. This present report confirms the findings in the hitherto solitary studies on the effect of physiotherapy in CD (Ramdharry, 2006; Smania et al, 2003; Tassorelli et al, 2006), all of which have yielded promising results. Differences in methodology make it difficult, however, to compare the results of the studies. However, the content and the procedure of the physiotherapy intervention in the above three studies seems to be similar to those in the current study.

In summary, the present data do to some extent support the use of the type of physiotherapy given in this study, but the program needs to be replicated in studies with a controlled group study design before the generality of the outcomes can be discussed. However, the patients with CD in this study experienced a positive effect of the physiotherapy treatment given.

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Appendix 1: Physiotherapy Program in Cervical Dystonia

The program includes progressive muscle relaxation (PMR), exercises for isometric muscle endurance and dynamic strength, coordination, balance, body perception, and muscle-stretching exercises. The program consisted of four phases.

Phase 1
On the 1st day of the program, the physiotherapist explains the setup of the whole program. The patient is interviewed concerning relevant medical, physical, and psychosocial information, and an individualized physical examination is carried out. The intervention begins with a session of PMR according to the method of Jacobson, and this continues twice a day during the 1st week. The main objective of the PMR is to alleviate pain, increase the subjective body perception, and achieve muscle relaxation in general. The program for the PMR is modified for each individual patient to find a position in which the treatment can be carried out as comfortably as possible. The patient tries to strain a chosen muscle and subsequently relax that muscle. Once the relaxation exercises are being well performed, the physiotherapist encourages the patient to practice them on his/her own.

Phase 2
In phase 2, exercises for increasing isometric muscle endurance and dynamic strength in the neck muscles are introduced. Both exercises are carried out with the patients in a standing position and also in a supine, prone, and lateral position lying on a board, which is leaning against a wall-bar (height 83 cm). An example of an exercise aimed to achieve isometric muscle endurance in the neck muscles in a supine position is active pressing of the back of the head down onto the surface for approximately 5 seconds. An example of a dynamic exercise in a lateral position is active lateral flexion of the head while lying on a board, which is resting against a wall-bar. An illustration of how the training can be accomplished while standing in a walking position is when the patient has a rubber tube (Latex 4.0 × 1.0) fixed at eye level on a wall-bar and placed around the forehead while he/she performs weight transfers backward and forward. A starting point with 5 × 3 repetitions is a common introduction to these exercises.

Phase 3
Phase 3 comprises exercises aimed at improving coordination, balance, body perception, and postural orientation. The goal of the exercises is to incorporate these different aspects and may be exemplified by an exercise with a ball of medium size. While walking in different directions, the physiotherapist bounces the ball in altered directions to the patient. The patient is stimulated to automatically turn the head toward the ball (to be able to see and catch it) and grasps the ball with both hands. Exercises for promoting balance are carried out on different surfaces and on balance plates. Attempts to improve body perception in a sitting, standing, and walking position are made with manual, verbal, and visual guidance. For instance, the physiotherapist places a hand on the patient’s shoulder and instructs her/him to “lower your shoulder” while sitting in front of a mirror. The exercises also focus on equilibrium of the pelvic position in addition to symmetry of the position of the feet and head. In this phase, exercises for increasing the dynamic strength of the trunk and upper extremities with machine equipment are added.

Phase 4
In the fourth phase, active muscle-stretching exercises are introduced for the neck muscles, for the muscles involved in scapular movements, and for the muscles of the spine, trunk, and legs according to the needs of the individual patient. The physiotherapist prolongs the stretching and increases the number of repetitions as required.

In addition
The physiotherapy program has a cumulative design. The number of repetitions and the types of exercises that are added from one phase to another will vary according to each individual’s needs as decided by the physiotherapist.