



■ CHILDREN'S ORTHOPAEDICS

Validation of a modified Care and Comfort Score and responsiveness to hip surgery in children with cerebral palsy in Gross Motor Function Classification System levels IV and V

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Aims

The purpose of this study was to assess the reliability and responsiveness to hip surgery of a four-point modified Care and Comfort Hypertonicity Questionnaire (mCCHQ) scoring tool in children with cerebral palsy (CP) in Gross Motor Function Classification System (GMFCS) levels IV and V.

Methods

This was a population-based cohort study in children with CP from a national surveillance programme. Reliability was assessed from 20 caregivers who completed the mCCHQ questionnaire on two occasions three weeks apart. Test-retest reliability of the mCCHQ was calculated, and responsiveness before and after surgery for a displaced hip was evaluated in a cohort of children.

Results

Test-retest reliability for the overall mCCHQ score was good (intraclass correlation coefficient 0.78), and no dimension demonstrated poor reliability. The surgical intervention cohort comprised ten children who had preoperative and postoperative mCCHQ scores at a minimum of six months postoperatively. The mCCHQ tool demonstrated a significant improvement in overall score from preoperative assessment to six-month postoperative follow-up assessment ($p < 0.001$).

Conclusion

The mCCHQ demonstrated responsiveness to intervention and good test-retest reliability. The mCCHQ is proposed as an outcome tool for use within a national surveillance programme for children with CP.

Cite this article: *Bone Jt Open* 2023;4-8:580–583.

Keywords: Hip, CareComfort, Cerebral palsy, Validity, Caregiver

Introduction

Hip displacement is one of the major disability-related health issues in nonambulatory children with cerebral palsy (CP) and is more likely to be seen in children in higher Gross Motor Function Classification System (GMFCS) levels.¹⁻⁴ National surveillance programmes for children with CP have been shown to reduce the incidence of hip displacement by enabling timely

management of hips at risk.^{5,6} It is important, as part of continued surveillance of children with hip displacement, to attempt to identify the extent of the clinical consequences of hip displacement such as hip pain, seating difficulties, and perineal nursing care before and after an intervention to relocate a displaced or dislocated hip. This is a challenge, as the children most commonly affected are in GMFCS levels IV and V and often unable to

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doi: 10.1302/2633-1462.48.BJO-2023-0051.R1

Bone Jt Open 2023;4-8:580–583.

Table 1. Mean and median scores per question for each dimension of the modified Care and Comfort Hypertonicity Questionnaire tools for the caregiver cohort (n = 20).

Dimension	mCCHQ score per question	
	Mean (SD)	Median (IQR)
1: Personal care	2.44 (0.41)	2.47 (2.06 to 2.79)
2: Positioning/transferring	2.17 (0.58)	2.08 (1.83 to 2.40)
3: Comfort	2.04 (0.73)	1.90 (1.40 to 2.48)
4: Interaction/communication	2.28 (0.48)	2.29 (2.00 to 2.71)
Total	2.23 (0.38)	2.17 (1.96 to 2.54)

IQR, interquartile range; mCCHQ, modified Care and Comfort Hypertonicity Questionnaire; SD, standard deviation.

express their difficulties themselves. Therefore, clinicians rely on proxy reporting by the child's carer(s).

The Cerebral Palsy Integrated Pathway Scotland (CPIPS) surveillance programme currently employs a four-point modified version of the traditional Care and Comfort Hypertonicity Questionnaire (CCHQ), introduced in 2006 by McCoy et al.⁷ The CCHQ in itself is a development of the original Caregiver Questionnaire,⁸ which was developed in 1990 to measure the perceived effort of caregivers in providing care for children with moderate to severe CP.⁹ The aim of the CCHQ was to evaluate functional needs and, to a lesser extent, quality of life in children with CP.⁷ The CCHQ is a self-reported 27-item questionnaire and the four domains covered are personal care, positioning/transferring, comfort, and interaction/communication. Although the CCHQ is rated through a seven-point Likert scale, scored from 1 (very easy) to 7 (impossible), our modified CCHQ (mCCHQ) is rated on a four-point scale. The modified Likert scale of the mCCHQ ranges from 1 (very easy) to 4 (impossible), giving a maximum possible score of 108 (Supplementary Material).

Content validity of the CCHQ has been established, as well as its ability to detect change when intrathecal baclofen was administered or the dose levels changed,¹⁰ but this has not been evaluated before and after a hip reconstruction. Furthermore, inter- and intrarater reliability and concurrent validity of the CCHQ are yet to be determined.⁷ The CCHQ seemed to offer potential advantages over more lengthy questionnaires and those that assumed that the respondent would be the child themselves.

We decided to evaluate intra- and inter-rater reliability, and responsiveness to surgical intervention, of our four-point mCCHQ questionnaire to assess hip pain, overall function and the outcome of hip surgery for children with CP in the CPIPS surveillance programme.

Methods

This study was reviewed by the South East Scotland Research Ethics Service and was deemed not to require ethical approval under the terms of the Governance

Arrangements for Research Ethics Committees. This was a population-based cohort study in children with CP from the CPIPS surveillance programme, initiated in 2013.¹¹ Over 95% of all children with CP are registered in the programme, and one of its main aims is to monitor hip displacement and dislocation.¹¹ Informed consent for use of data is gained for each patient as they are enrolled on to the CPIPS programme.

Assessment of reliability of mCCHQ score. To evaluate the test-retest reliability of the mCCHQ, 20 caregivers participating in the CPIPS programme were invited to complete the mCCHQ questionnaire on two occasions three weeks apart.

Assessment of responsiveness to change. To assess responsiveness of the mCCHQ to hip intervention, the CPIPS database of 2,403 children with CP in Scotland was interrogated to identify patients in GMFCS levels IV and V who had undergone surgery for hip displacement or dislocation between 2018 and 2021, and who had a preoperative score and postoperative score collected at a minimum of six months following their hip surgery. Data on the classification of CP¹² and GMFCS levels of the children were retrieved from the CPIPS database.

Statistical analysis. Test-retest reliability examined the relation between repeated evaluations of the mCCHQ score. The standard deviations (SDs) of the differences between the initial and three-week mCCHQ scores for individual domains and the total score were calculated to summarize the absolute differences obtained on repeat testing. Intraclass correlation coefficients (ICC) (two-way random effects model with absolute agreement) were applied to determine the test-retest reliability of the day 0 mCCHQ scores compared with the three-week mCCHQ scores. Scores for ICC coefficient range from 0 to 1, where the former shows no reliability, and the latter exhibits perfect reliability. Cicchetti and Sparrow¹³ and Fleiss et al¹⁴ have suggested that a score of < 0.40 is poor, 0.40 to 0.59 is fair, 0.60 to 0.74 is good, and > 0.74 is excellent.

Significance of responsiveness to change between preoperative score and minimum six-month follow-up was assessed using the paired *t*-test. A significance level < 0.05 was deemed to be statistically significant. In this part of the study, we deemed it important clinically to evaluate the average total score per dimension, rather than mean score of individual questions. This was to determine which dimensions of the mCCHQ were more or less affected by the intervention, and where the statistical significance lay within the dimensions, as well as whether there was a significant change in the overall score. "Not applicable" (N/A) responses are available in the database.

Results

All questionnaires were completed by carers of the children without supervision from clinical staff, suggesting that the questionnaire was easy to complete. A total of

Table II. Overall preoperative and six-month postoperative modified Care and Comfort Hypertonicity Questionnaire scores for study cohort (n = 10).

Dimension	Preoperative mean score (SD)	Minimum six-month mean score (SD)	Mean difference (SEM)	p-value*
1: Personal care	18.4 (6.22)	15.5 (5.44)	2.90 (1.48)	0.041
2: Positioning/transferring	16.0 (3.92)	11.3 (3.86)	4.7 (1.37)	0.007
3: Comfort	10.2 (5.63)	9.30 (4.81)	0.90 (1.32)	0.513
4: Interaction/communication	16.2 (2.39)	14.6 (3.06)	1.60 (0.79)	0.074
Overall Score	60.8 (14.2)	50.7 (12.5)	10.1 (1.72)	< 0.001

*Paired t-test.

mCCHQ, modified Care and Comfort Hypertonicity Questionnaire; SD, standard deviation; SEM, standard error of the mean.

20 carers completed the mCCHQ questionnaire on the first occasion, and 19 completed this three weeks later. Of the patients, 16 were GMFCS level V and four were level IV. Mean and median scores per question for each dimension of the mCCHQ for the first timepoint are given in Table I.

Test-retest reliability of the mCCHQ score. Test-retest reliability was calculated for the mCCHQ score between day 0 and three-week test occasions. During this time period, there was no interval change in health status as ascertained by clinical assessment with a complete review of systems. ICCs demonstrated the highest interobserver reliability for personal care (ICC 0.76), followed by comfort (ICC 0.71), positioning/transferring (ICC 0.67), and was lowest for interaction/communication (ICC 0.54) of the mCCHQ. Test-retest reliability for the overall mCCHQ score was good (ICC 0.78). The SD of differences for the overall mean score per question between initial mCCHQ and three-week mCCHQ was 0.24, with no difference in any dimension being greater than 0.48. Therefore, on re-testing we can be 95% confident that the difference in overall score per question will be less than 0.5. No score was deemed to have poor reliability, with all scores being at least moderately reliable or better. Any questions that were “N/A” preoperatively were also “N/A” postoperatively in the same patient.

Responsiveness of mCCHQ to hip intervention. The surgical intervention cohort comprised ten children with a mean age of 8.6 years (4 to 13) who had both pre- and postoperative mCCHQ scores at a minimum of six months postoperatively (mean follow-up 14.6 months (SD 3.8)). There were seven patients who were GMFCS level V and three who were level IV. Eight patients had a combined Dega pelvic osteotomy and proximal femoral varus derotation osteotomy (of which six were bilateral) and two had proximal femoral varus derotation osteotomies alone (both left-sided). In all cases, any questions that were “N/A” preoperatively were also “N/A” postoperatively.

The mean preoperative and six-month postoperative mCCHQ scores for the cohort are shown in Table II. There was a significant improvement in the domains of personal care, positioning/transferring, and overall mCCHQ score from preoperative assessment to six-month postoperative follow-up assessment ($p < 0.001$).

Discussion

The most important finding of this preliminary study was that the mCCHQ score was a reliable, responsive tool that could be used to evaluate the effect of surgery in managing a displaced or dislocated hip in CP children who were GMFCS levels IV and V.

The mCCHQ score is a health-related quality of life measure that includes items of clinical relevance to hip displacement, and therefore should reflect the effects of hip interventions on quality of life accurately. This study shows that all dimensions in the mCCHQ were reduced by hip intervention, suggesting an improvement in quality of life. The overall change in score was statistically significant ($p < 0.001$). The comfort dimension did not reach statistical significance, which is of interest as pain is often cited by clinicians as the main reason for hip surgery in this patient group. The authors feel this may be due to the relatively low numbers in the intervention group, as discussed below. There was one question (occasionally two) that carers determined “N/A” within the intervention group, but the same questions remained “N/A” after intervention and were thus not relevant to the hip surgery.

There were limitations to this study. The number of respondents could have been greater, but we felt that 20 respondents would suffice for test-retest reliability of the mCCHQ. We had also anticipated identifying more children from the CPIPS database who had mCCHQ scores at least six months after reconstructive surgery. We concluded that the small number was because surgical services were disrupted by the COVID-19 pandemic and continued to be so after the relaxation of restrictions, with the recovery of clinical services from this period continuing up to the time of writing. Future studies on responsiveness of the score to intervention will contain greater numbers. These should give more detailed information on the effect of hip intervention on the domains, and perhaps even allow assessment of the individual questions.

There are a number of other recognized quality of life measures for children with CP, each with their own advantages and disadvantages. The CPCHILD¹⁵ and CP QOL-Child¹⁶ measures have been considered to have a high quality of construction, face validity, and concurrent

validity. However, their sensitivity to change has not previously been reported.¹⁷ The PEDSQL 3.0 measure has data on sensitivity, but has a disproportionate focus on function rather than quality of life which, although linked, does not always correlate.¹⁸ A balanced valid score for use in a children's CP national registry is required to be acceptable to stakeholders, responsive to intervention, have good utility, sufficient content, and concurrent validity, and show acceptable test-retest reliability.

Our conclusions are that the mCCHQ is responsive to hip interventions and has good test-retest reliability. The mCCHQ is thus proposed as an outcome tool for use in GMFCS levels IV and V within the setting of a national surveillance programme for children with CP.



Take home message

- The four-point modified Care and Comfort Hypertonicity Questionnaire (mCCHQ) score is a valid outcome assessment tool which shows responsiveness to intervention and good test-retest reliability.
- The mCCHQ is proposed as validated outcome measure for use within a national surveillance programme for children with cerebral palsy.

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Supplementary material



The modified Care and Comfort Hypertonicity questionnaire (mCCHQ) currently administered as part of the Cerebral Palsy Integrated Pathway Scotland programme.

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Funding statement:

- The authors disclose receipt of the following financial or material support for the research, authorship, and/or publication of this article: this work was supported by a small grant from CPIPS which funded the open access publication fee for this study.

Data sharing:

- The data that support the findings for this study are available to other researchers from the corresponding author upon reasonable request.

Ethical review statement:

- Following review by the South East Scotland Research Ethics Service, ethical approval for this study was not required under the terms of the Governance Arrangements for Research Ethics Committees.

Open access funding:

- The open access funding for publication of this article was provided by CPIPS.

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