

# Pain, Fatigue, and School Functioning in Children with Cerebral Palsy: A Path-Analytic Model

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**Objective** This study tests a model of how pain and fatigue, independently or in combination, relate to school functioning in pediatric cerebral palsy (CP). **Methods** One hundred eighty-nine parents of children with CP completed the Pediatric Quality of Life Inventory™ (PedsQL™) 4.0 Generic Core Scales and the PedsQL™ 3.0 Cerebral Palsy Module. Seventy-three children with CP completed the PedsQL™. Path-analytic and mediational techniques were utilized to test the a priori model. **Results** Data from both parent proxy-report and child self-report were found to have acceptable model fit. Results supported the existence of an indirect relationship between diagnostic subtypes and school functioning that was partially mediated by both pain and fatigue. **Conclusions** Pain and fatigue represent potentially modifiable targets for interventions designed to improve school functioning in children with CP.

**Key words** cerebral palsy; children; fatigue; pain; PedsQL™; school functioning.

Cerebral palsy (CP) is a nonprogressive developmental disorder that affects movement, muscle function, and cognitive functioning as a result of damage to specific areas of the brain before, at, or shortly after birth. Neurological effects can range from mild to severe. There are approximately two to three children born with CP per 1,000 live births (Panteliadis & Strassburg, 2004). Children with the spastic type of CP, which makes up about 85% of cases (Panteliadis & Strassburg), have reported significantly lower health-related quality of life (HRQOL) than healthy controls (Varni et al., 2005), reflecting the myriad of associated health complications. Spastic CP has three diagnostic subtypes that reflect which parts of the body are most affected. Hemiplegia refers to the right or left hemisphere, diplegia to lower limbs that are more affected than upper limbs, and quadriplegia to all four limbs. Children with quadriplegia often have generalized systemic problems such as swallowing

and eating disorders, seizures, and cognitive problems. This is generally assumed to be the most severe form and often results in more developmental limitations and in lower HRQOL (Varni et al., 2005; Varni, Burwinkle, Berrin, et al. 2006).

HRQOL is a subcategory of overall quality of life (QOL) that is directly related to individual health and well-being and, when measured, must at minimum incorporate ratings of physical, psychological, and social functioning as delineated by the World Health Organization (1948). Within the multidimensional HRQOL construct, some dimensions may be more negatively affected depending on the particular health condition. School functioning is of particular concern, given the negative impact of academic failure on a child's future. Previous findings with the Pediatric Quality of Life Inventory™ (PedsQL™; available at <http://www.pedsq.org>) School Functioning Scale have demonstrated significant positive

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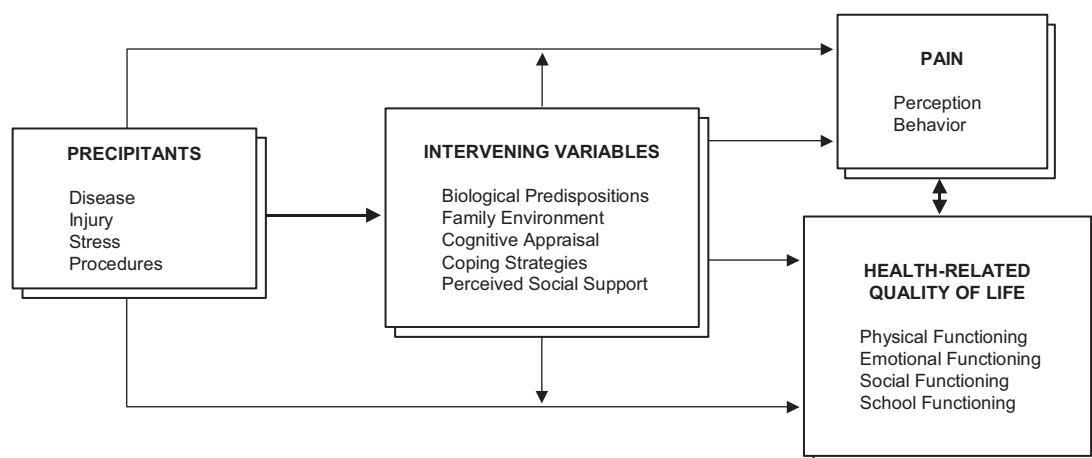
correlations (child self-report:  $r = .26$ ,  $p < .001$ ; parent proxy-report:  $r = .25$ ,  $p < .001$ ) with standardized achievement scores based on the Stanford 9 (Varni, Burwinkle, & Seid, 2006) and small positive correlations (child self-report:  $r = .17$ ,  $p < .05$ ; parent proxy-report:  $r = .11$ , NS) with standardized intelligence testing scores based on the Weschler Intelligence Scale for Children—Revised (WISC-R) (Bastiaansen, Koot, Bongers, Varni, & Verhulst, 2004), suggesting that lower school functioning as measured by the PedsQL™ is associated with lower academic achievement and, to a lesser extent, intelligence testing scores. Unfortunately, children with CP are at a significant academic disadvantage when compared with healthy controls as measured by the PedsQL™ School Functioning Scale (Varni et al., 2005; Varni, Burwinkle, Berrin, et al., 2006).

Identifying potential predictors of school functioning in children with CP may aid in developing intervention programs to improve academic performance. Pain (Roscigno, 2002) and fatigue (Heller, Alberto, & Meagher, 1996) are two variables that are hypothesized to be associated with academic performance for a child with CP.

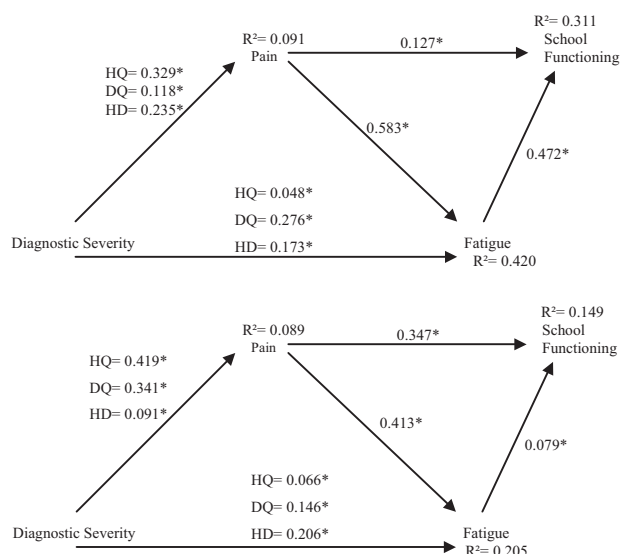
The experience of pain is common in children with a diagnosis of CP (Houlihan, O'Donnell, Conaway, & Stevenson, 2004). Varni, Burwinkle, Berrin, et al. (2006) found that children with spastic quadriplegia self-reported greater pain severity than children with either hemiplegia or diplegia; the same pattern was found for parent proxy-reports of children's pain. A significant and inverse relationship between pain severity and school functioning was also reported. Additionally, Houlihan and colleagues found a significant and positive relationship between pain and the number of school days missed.

Fatigue is another reality in the lives of children with CP and their families. For those who are ambulatory, fatigue has been documented objectively through measurement of energy expenditure, where children with CP were found to require more energy to maintain ambulatory capacity compared with normal controls (Bottos, 2003). However, parent reports of children's muscle fatigue severity have also been found to increase with the severity of diagnostic subtype regardless of ambulatory status (Varni, Burwinkle, Berrin, et al., 2006). Varni and colleagues also found that fatigue was inversely correlated with school functioning.

In sum, the preliminary research to date suggests that pain and fatigue are common to children with CP and may negatively relate to school functioning. The Biobehavioral Model of Pediatric Pain (Varni, Burwinkle, & Katz, 2004; see Fig. 1) was developed to facilitate the identification of potential predictors of pain in children and to model the relationship between pain and HRQOL (Varni et al., 1996). Varni and colleagues have argued that more effective pediatric pain interventions will be implemented if guided by a conceptual framework. This framework can also be applied to pediatric fatigue and can guide research attempting to identify predictors of fatigue as well as fatigue-related outcomes. The present study applies the conceptual framework of Varni et al.'s Biobehavioral Model to examine how pain and fatigue, independently or in combination, relate to school functioning for children with different spastic CP subtypes. Although previous work has shown simple bivariate relationships between pain, fatigue, and school functioning, no published study to date has examined these variables within the context of a systematic path-analytic model.



**Figure 1.** Varni's Biobehavioral Model of Pediatric Pain. Adapted from Varni et al. (1996).



**Figure 2.** (a) Parent proxy-report path model with standardized coefficients and  $R^2$  values. HD, hemiplegia/diplegia comparisons; HQ, hemiplegia/quadruplegia comparisons; DQ, diplegia/quadruplegia comparisons. \* $p < .05$ . (b) Child self-report path model with standardized coefficients and  $R^2$  values.

We proposed both direct and indirect relationships among diagnostic subtypes, pain and fatigue, and the outcome of school functioning (Fig. 2). Overall, the model proposed that diagnostic subtypes are related to school functioning via pathways mediated by pain and fatigue. Mediation refers to the presence of a third variable, which fully or partially accounts for the relationship between the variables of interest, whereas moderation occurs when a third variable's presence alters the relationship between the original variables (Baron & Kenny, 1986). Specifically, we hypothesized the following: (a) pain will mediate the relationship between diagnostic severity and school functioning; (b) fatigue will mediate the relationship between diagnostic severity and school functioning; and (c) fatigue will mediate the relationship between pain and school functioning. These hypotheses were tested using both parent proxy-report and child self-report.

## Method

### Participants

One hundred eighty-nine parents of children with spastic CP aged 5–18 (45 hemiplegia, 66 diplegia, and 78 quadriplegia) completed the PedsQL™ 4.0 Generic Core Scales and the PedsQL™ 3.0 Cerebral Palsy Module. The average age of the children whose parents completed the parent proxy-report was 8.4 years ( $SD = 4.4$ ). The sample was heterogeneous with respect to ethnicity, with 66 (35%) Hispanic, 96 (51%) White/non-Hispanic, 9 (5%) Asian or

Pacific Islander, 10 (5%) African American, and 8 (4%) “other” (8, 4% missing).

Seventy-three children with a spastic CP diagnosis aged 5–18 (27 hemiplegia, 34 diplegia, 12 quadriplegia) completed the PedsQL™ 4.0 Generic Core Scales and the PedsQL™ 3.0 Cerebral Palsy Module. The average age of children who self-reported was 10.3 years ( $SD = 3.9$ ). Not all children provided self-report because of time constraints, refusal to participate, or physical impairment. This sample is the same as that used in the study by Varni, Burwinkle, Berrin, et al. (2006), and the children are those whose parents completed the parent proxy-report.

## Measures

The PedsQL™ is a health-related quality-of-life instrument that consists of a well-validated generic core measure and some condition- and disease-specific modules. The PedsQL™ 4.0 Generic Core Scales ask questions that are not necessarily specific to CP; therefore, comparisons between CP and other chronically ill as well as healthy populations can be made. The Generic Core Scales consist of 23 items divided into four subscales: Physical Functioning (8 items), Emotional Functioning (5 items), Social Functioning (5 items), and School Functioning (5 items). Additionally, there are two summary scores: a Psychosocial Health Summary Score (derived from the Social, School, and Emotional Functioning subscales) and a Total Scale Score (from all subscales). Responses are given on a 5-point Likert scale, with 0 (*never a problem*), 1 (*almost never a problem*), 2 (*sometimes a problem*), 3 (*often a problem*), and 4 (*almost always a problem*). The self-report response scale for children aged 5–7 years was previously modified during the development of the PedsQL™ to reflect developmental ability, with a 3-point response scale instead of the 5-point scale: 0 (*not at all*), 2 (*sometimes*), and 4 (*a lot*). Each scale is then linearly transformed and reverse-scored from 0 to 100, with higher scores indicating better HRQOL (Varni, Seid, & Kurtin, 2001). The PedsQL™ 4.0 Generic Core Scales include both a parent proxy-report (items asked in third person) and a child self-report. Research on HRQOL reporting supports the value of both parent proxy-reports of their child's HRQOL and the child self-report (Varni et al., 2001, 2005).

Previous validation research with the PedsQL™ 4.0 Generic Core Scales has shown internal consistency reliabilities that generally exceed the .70 standard for group comparisons among children with CP (Varni et al., 2005). Construct validity in CP has been demonstrated using the known-groups method (Varni, Burwinkle, Berrin, et al., 2006).

The PedsQL™ 3.0 Cerebral Palsy Module was developed as a condition-specific HRQOL instrument (Varni, Burwinkle, Berrin, et al., 2006). The PedsQL™ 3.0 Cerebral Palsy Module includes 35 items that are divided into seven subscales: Daily Activities (9 items), School Activities (4 items), Movement and Balance (5 items), Pain and Hurt (4 items), Fatigue (4 items), Eating Activities (5 items), and Speech and Hearing (4 items). Like the Generic Core Scales, responses are given on the same 5-point response scale, with the self-report scale for children aged 5–7 years previously modified to reflect developmental ability. Previous validation research with the PedsQL™ 3.0 Cerebral Palsy Module Scales has shown internal consistency reliabilities that generally exceed the .70 standard for group comparisons (Varni, Burwinkle, Berrin, et al., 2006). This validation research was conducted on the same child sample as this study.

### Procedure

This investigation was conducted as part of a larger validation study assessing the PedsQL™ in a CP population. Participants were recruited through clinic appointments at Children's Hospital and Health Center, San Diego, and medical therapy units (MTUs) throughout Southern California. Children were screened by using medical records and excluded if other comorbid conditions such as mental retardation or Down's syndrome were present. The consent forms and measures took approximately 30 min to complete, and each parent/child dyad received \$50 as a token of appreciation for participation. The Institutional Review Boards at Children's Hospital and Health Center, San Diego, San Diego State University, and the State of California approved this study.

### Data Analyses

The overall model was tested for goodness-of-fit via path analysis. Owing to limitations with the chi-square likelihood ratio test statistics, many researchers (e.g., Tanaka, 2000) have suggested using multiple measures of model fit. In this study, the following measures were employed: (a) the Satorra–Bentler-Scaled  $\chi^2$  ( $S-B\chi^2$ ; Satorra & Bentler, 2001), a statistical test of model fit that corrects for non-normality if it exists, assumes the correct model and assesses the absolute fit of the model to the data; (b) the Comparative Fit Index (CFI; Bentler, 1990), a fit index that tests the proportional improvement in fit by comparing the model with a baseline, with values  $>.90$  indicating reasonable model fit; and (c) the Root Mean Square Error of Approximation (RMSEA; Steiger, 1990), a closeness in fit test with values  $<.08$  indicating reasonable model fit. In evaluating the statistical significance of

individual model parameters (i.e., path coefficients), a significance level of  $p < .05$  was used.

To investigate Hypotheses 1 through 3, Baron and Kenny's (1986) test of mediation was used, in which there are four steps: (a) the mediator is regressed on the independent variable; (b) the dependent variable is regressed on the independent variable; (c) the dependent variable is regressed on the mediator; and (d) the dependent variable is regressed on both the independent variable and the mediator. If significant regressions are found for Steps 1–3, in Step 4 mediation is shown if the independent variable becomes nonsignificant or is reduced in significance.

### Results

Descriptive statistics for the sample are summarized in Table I. For the pain and fatigue scales, children reported less pain and fatigue (reflected in higher scores on the scales) than did their parents via proxy-report. School functioning scores showed a similar pattern, with parents reporting poorer school functioning than did their children (lower scores indicating poorer school functioning). For the parent proxy-report,  $t$  tests revealed no significant differences in PedsQL™ scores with regard to gender or ethnicity. Additionally, there were no significant differences in proxy ratings between parents of children who completed a self-report and parents of children who did not with regard to reports of children's pain, fatigue, or school functioning. Child self-report  $t$  tests revealed no significant differences in PedsQL™ scores for gender or ethnicity.

The path-analytic model fit well to the data (Fig. 2) for the parent proxy-report, with  $S-B\chi^2(2, n = 182) = 3.23, p = .199$ ; CFI = .993; RMSEA = .058. The child self-report model fit was found to be acceptable, with  $S-B\chi^2(2, n = 66) = 2.25, p = .325$ ; CFI = .996; RMSEA = .044. General guidelines suggest that a model is considered a good fit for the data if the  $S-B\chi^2$  values are *not* significant,

**Table I.** Scale Descriptives for PedsQL™ 4.0 Generic Core and Cerebral Palsy Module

	Number of items	<i>n</i>	Mean	<i>SD</i>
Child self-report				
School functioning	5	77	65.61	22.18
Pain	5	71	73.59	23.13
Fatigue	4	73	68.09	21.45
Parent proxy-report				
School functioning	5	196	52.47	21.39
Pain	5	219	65.29	25.98
Fatigue	4	226	59.68	24.42

if the CFI value is between .9 and 1 (Bentler, 1990), and if the RMSEA value is  $<.8$  (Steiger, 1990).

Standardized path coefficients ( $\beta$ ) and  $R^2$  values are reported in Fig. 2. The parent proxy path-analytic model variables explained 9% of the variance for pain, 42% for fatigue, and 31% for school functioning. These results suggest that diagnostic severity and pain explain large amounts of reported fatigue in CP, as do pain and fatigue for school functioning. Furthermore, there appear to be other important explanations for pain variability beyond diagnostic severity, which did not account for much variance. The child self-report model explained 9% of the variance for pain, 20% for fatigue, and 14% for school functioning, which suggests similar conclusions for pain variability. However, children's self-reported diagnostic severity and pain comprised a smaller role in fatigue presence than did the reports from their parents; additionally, less variance in children's reports of school functioning was explained by pain and fatigue.

For Hypothesis 1, we tested whether pain was a mediator between diagnostic severity and school functioning. For the parent proxy-report, Step 1 of the analysis showed that diagnostic severity and pain were significantly associated ( $\beta = -.243, p = .001; R^2 = .059, p = .001$ ). In Step 2, diagnostic severity was significantly associated with school functioning ( $\beta = -.232, p = .001; R^2 = .054, p = .001$ ). Step 3 showed that pain and school functioning were significantly associated ( $\beta = .403, p < .001; R^2 = .163, p < .001$ ). In Step 4, both diagnostic severity ( $\beta = -.154, p < .05$ ) and pain ( $\beta = .366, p < .001$ ) were significantly associated with school functioning ( $R^2 = .185, p < .001$ ). The relationship between diagnostic severity and school functioning was still significant but was lower than Step 2, suggesting partial mediation of pain. The Sobel test, however, found that this mediated effect was not statistically significant,  $z = .87, p > .05$ . A more severe diagnosis is associated with increased pain, which in turn is associated with lower school functioning, yet a more severe diagnosis (independent of pain) is also associated with lowered school functioning.

For the child self-report, pain was not found to be a mediator between diagnostic severity and school functioning. Step 1 showed that diagnostic severity and pain were significantly associated ( $\beta = -.273, p < .05; R^2 = .074, p < .05$ ). However, in Step 2 of the mediational analysis, the relationship between diagnostic severity and fatigue was not found to be significant. This was surprising, as this direct relationship was shown to be significant in the path-analytic testing of the model.

For Hypothesis 2 (testing fatigue as a mediator between diagnostic severity and school functioning),

Step 1 showed that diagnostic severity and fatigue were significantly associated ( $\beta = -.255, p < .001; R^2 = .065, p < .001$ ) for the parent proxy-report. In Step 2, diagnostic severity was significantly associated with school functioning ( $\beta = -.232, p = .001; R^2 = .054, p = .001$ ). Step 3 showed that fatigue and school functioning were significantly associated ( $\beta = .420, p < .001; R^2 = .163, p < .001$ ). In Step 4, both diagnostic severity ( $\beta = -.137, p < .05$ ) and pain ( $\beta = .385, p < .001$ ) were significantly associated with school functioning ( $R^2 = .185, p < .001$ ). The relationship between diagnostic severity and school functioning was still significant but was lower than Step 2, suggesting partial mediation of fatigue. The Sobel test confirmed that this mediated effect was statistically significant,  $z = -3.09, p = .001$ . Thus, children with a more severe diagnosis tended to experience more fatigue, which in turn was associated with lower school functioning as reported by parents.

Fatigue was not found to be a mediator between diagnostic severity and school functioning for the child self-report, as Step 1 for this mediational analysis, the relationship between diagnostic severity and fatigue, was not found to be significant.

Hypothesis 3 tested fatigue as a mediating variable between pain and school functioning. In Step 1 for the parent proxy-report, pain was significantly associated with fatigue ( $\beta = .622, p < .001; R^2 = .387, p < .001$ ). In Step 2, pain was significantly associated with school functioning ( $\beta = .403, p < .001; R^2 = .163, p < .001$ ). In Step 3, fatigue was significantly associated with school functioning ( $\beta = .420, p < .001; R^2 = .176, p < .001$ ). In Step 4, both pain ( $\beta = .233, p < .001$ ) and fatigue ( $\beta = .280, p < .001$ ) were significantly associated with school functioning ( $R^2 = .212, p < .001$ ). The relationship between pain and school functioning was still significant but was lower than Step 2, suggesting partial mediation of fatigue. The Sobel test, however, found that this mediated effect was not statistically significant,  $z = .97, p > .05$ . Thus, children with more pain tended to experience more fatigue, which in turn was associated with lower school functioning. However, children with greater pain also experienced lower school functioning independent of the fatigue they experienced.

Fatigue was not found to be a mediator between pain and school functioning for the child self-report. In Step 1, pain was significantly associated with fatigue ( $\beta = .407, p = .001; R^2 = .166, p = .001$ ). In Step 2, pain was significantly associated with school functioning ( $\beta = .379, p < .01; R^2 = .144, p < .01$ ). Step 3 in this analysis, the relationship between fatigue and school functioning, was not found to be significant, although there was a trend toward significance ( $\beta = .230, p = .061; R^2 = .053, p = .061$ ).

## Discussion

This study sought to further define the interrelationships among diagnostic subtype, pain, fatigue, and school functioning within the spastic subtype of pediatric CP using a path-analytic model. Although previous studies (Varni, Burwinkle, Berrin, et al., 2006) have reported bivariate relationships among these constructs, this is the first study to our knowledge to examine the direct and the indirect paths among these variables in spastic CP within the context of an overall model.

This study found a good overall fit for the model utilizing path-analytic techniques, suggesting that pain and fatigue are both important variables in understanding how diagnostic subtype translates into problems in everyday living, such as school functioning. Specific mediational analyses for the indirect effects of pain and fatigue on the relationship between diagnostic severity and school functioning found partial support. Findings suggest that not only are more severe diagnoses associated with lowered school functioning but also associated pain and fatigue, and in some cases, pain and fatigue partially explain the association between CP diagnosis and performance at school. Additionally, increased pain was associated with reduced school functioning indirectly through fatigue, suggesting that fatigue may be a key variable in this process; however, the direct relationship between pain and school functioning also appeared to be an important factor. Thus, these data provide some support for the key roles of both pain and fatigue as potential points of intervention to improve school functioning and ultimately increase HRQOL in children with spastic CP.

However, all child self-report mediational analyses were nonsignificant. This finding is particularly interesting when the good overall fit of the child self-report model is considered. Two of the nonsignificant regression findings are for the relationship between diagnostic severity and fatigue. We suspect this may be due to decreased power, as child self-report sample sizes were smaller than those of parent proxy-reports. Another potential explanation for this finding is that, when pain is introduced into the overall model, the error variance associated with CP severity is reduced, thus resulting in statistical significance of the pathway. It is also possible that, during modeling, the standard errors are smaller, which makes the effect significant.

Another potential explanation regarding the lack of significance for the child self-report data relates to an important secondary finding of this study. Parents rated their child's functioning on all of the domains as significantly lower than their child self-reported, which replicates

the findings from past HRQOL research (Varni et al., 2001). This could be due to the tendency of an observer to infer negative conclusions about the child's experience using physical, observable behaviors because they have access to little else (Varni et al., 2001). The child, on the contrary, could have adapted to their circumstance, thereby reporting higher scores. Children might also differentiate more readily between pain and fatigue than their parents, as supported by stronger bivariate correlations between these variables found for the parent proxy-report (Varni, Burwinkle, Berrin, et al., 2006). It therefore becomes even more important for health care providers to consider both child and the parent reports because of cross-informant differences.

Taken together, these results suggest that children with more severe subtype diagnoses are experiencing more pain and fatigue according to their parents, which are affecting school performance to varying degrees. However, the children themselves might feel differently, which is a research area worthy of further exploration. These findings may suggest opportunities to improve the school performance of children with CP, by targeting the potentially modifiable dimensions of pain and fatigue. There are a variety of current treatments available for pain and fatigue in CP. Botulinum toxin A (O'Brien, 2002), a localized muscle relaxant, has been shown to reduce both pain and fatigue. Other muscle relaxants such as diazepam (Mathew & Mathew, 2005) and baclofen (Vargus-Adams, Michaud, Kinnett, McMahon, & Cook, 2004) appear to have similar effects. Intensive physical therapy (PT) can serve to increase muscle strength and flexibility (Kanda, Pidcock, Hayakawa, Yamori, & Shikata, 2004). Surgical procedures such as muscle lengthening and selective dorsal rhizotomy are also beneficial (Miller, Johann-Murphy, & Cate, 1997). No longitudinal studies were found that have measured improvements in HRQOL (or school functioning) as a result of the available treatments. If future studies find that these treatments result in improved HRQOL, they will provide further model support.

There are several potential limitations of the study. Data collection included a screening and exclusion for comorbidity; therefore, no children with the additional diagnoses of mental retardation, Down's syndrome, or other conditions were included. This study was cross-sectional in nature, and not all children provided self-report. Those who did participate were not asked to contribute data on intellectual ability, which might feasibly account for differences between groups. The previously cited findings that the PedsQL™ School Functioning Scale scores are modestly related to intelligence testing

scores suggest that only a small percentage of the variance will be accounted for by child intelligence. Nevertheless, future studies will need to consider and control for this covariate in children with spastic CP.

In sum, the results of this study identify pain and fatigue as important components affecting HRQOL of children with spastic CP and suggest the possibility that interventions to reduce pain and fatigue in children with spastic CP may improve school functioning. Given current limitations in medical approaches to modifying the disease itself, in contrast to the availability of options for treating pain and fatigue, this approach holds promise for reducing disease burden and improving children's functioning. Future intervention studies will be needed to evaluate the effectiveness of such an approach.

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## References

- Baron, R. M., & Kenny, D. A. (1986). The moderator-mediator variable distinction in social psychological research: Conceptual, strategic, and statistical considerations. *Journal of Personality and Social Psychology*, 51, 1173–1182.
- Bastiaansen, D., Koot, H. M., Bongers, I. L., Varni, J. W., & Verhulst, F. C. (2004). Measuring quality of life in children referred for psychiatric problems: Psychometric properties of the PedsQL™ 4.0 Generic Core Scales. *Quality of Life Research*, 13, 489–495.
- Bentler, P. M. (1990). Comparative fit indexes in structural models. *Psychological Bulletin*, 107, 238–246.
- Bottos, M. (2003). Ambulatory capacity in cerebral palsy: Prognostic criteria and consequences for intervention. *Developmental Medicine and Child Neurology*, 45, 786–790.
- Heller, K. W., Alberto, P. A., & Meagher, T. A. (1996). The impact of physical impairments on academic performance. *Journal of Developmental and Physical Disabilities*, 8, 233–245.
- Houlihan, C. M., O'Donnell, M., Conaway, M., & Stevenson, R. D. (2004). Bodily pain and health-related quality of life in children with cerebral palsy. *Developmental Medicine and Child Neurology*, 46, 305–310.
- Kanda, T., Pidcock, F. S., Hayakawa, K., Yamori, Y., & Shikata, Y. (2004). Motor outcome differences between two groups of children with spastic diplegia who received different intensities of early onset physiotherapy followed for 5 years. *Brain and Development*, 26, 118–126.
- Mathew, A., & Mathew, M. C. (2005). Bedtime diazepam enhances well-being in children with spastic cerebral palsy. *Pediatric Rehabilitation*, 8, 63–66.
- Miller, A. C., Johann-Murphy, M., & Cate, I. M. (1997). Pain, anxiety, and cooperativeness in children with cerebral palsy after rhizotomy: Changes throughout rehabilitation. *Journal of Pediatric Psychology*, 22, 689–705.
- O'Brien, C. F. (2002). Treatment of spasticity with botulinum toxin. *The Clinical Journal of Pain*, 18, 182–190.
- Panteliadis, C. P., & Strassburg, H. M. (Eds.) (2004). *Cerebral Palsy: Principles and Management*. Stuttgart, NY: Georg Thieme.
- Roscigno, C. I. (2002). Addressing spasticity-related pain in children with spastic cerebral palsy. *Journal of Neuroscience Nursing*, 34, 123–133.
- Satorra, A., & Bentler, P. M. (2001). A scaled difference chi-square test statistic for moment structure analysis. *Psychometrika*, 66, 507–514.
- Steiger, J. H. (1990). Structural model evaluation and modification: An interval estimation approach. *Multivariate Behavioral Research*, 25, 173–180.
- Tanaka, J. S. (2000). Statistical models for change. In J. Rappaport & E. Seidman (Eds.), *Handbook of community psychology* (pp. 697–723). Dordrecht, Netherlands: Kluwer Academic Publishers.
- Vargus-Adams, J. N., Michaud, L. J., Kinnett, D. G., McMahon, M. A., & Cook, E. F. (2004). Effects of oral baclofen in children with cerebral palsy. *Developmental Medicine and Child Neurology*, 46, 787–789.
- Varni, J. W., Burwinkle, T. M., Berrin, S. J., Sherman, S. A., Artavia, K., Malcarne, V. L., et al. (2006). The PedsQL™ in pediatric cerebral palsy: Reliability, validity, and sensitivity of the Generic Core Scales and Cerebral Palsy Module. *Developmental Medicine and Child Neurology*, 48, 442–449.
- Varni, J. W., Burwinkle, T. M., & Katz, E. R. (2004). The PedsQL™ in pediatric cancer pain. A prospective

- longitudinal analysis of pain and emotional distress. *Journal of Developmental and Behavioral Pediatrics*, 25, 239–246.
- Varni, J. W., Burwinkle, T. M., & Seid, M. (2006). The PedsQL™ 4.0 as a school population health measure: Feasibility, reliability, and validity. *Quality of Life Research*, 15, 203–215.
- Varni, J. W., Burwinkle, T. M., Sherman, S. A., Hanna, K., Berrin, S. J., Malcarne, V. L., et al. (2005). Health-related quality of life of children and adolescents with cerebral palsy: Hearing the voices of the children. *Developmental Medicine and Child Neurology*, 47, 592–597.
- Varni, J. W., Rapoff, M. A., Waldron, S. A., Gragg, R. A., Bernstein, B. H., & Lindsley, C. B. (1996). Chronic pain and emotional distress in children and adolescents. *Journal of Developmental and Behavioral Pediatrics*, 17, 154–161.
- Varni, J. W., Seid, M., & Kurtin, P. S. (2001). The PedsQL™ 4.0: Reliability and validity of the Pediatric Quality of Life Inventory™ Version 4.0 Generic Core Scales in healthy and patient populations. *Medical Care*, 39, 800–812.
- World Health Organization. (1948). *Constitution of the World Health Organization basic document*. Geneva, Switzerland: World Health Organization.