

The association of drooling and health-related quality of life in children with cerebral palsy

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Objective: To investigate the association between drooling in children with cerebral palsy (CP) and their health-related quality of life (HRQOL), as well as the possible variables that predict their HRQOL.

Method: A cross-sectional design was used for this study. Children with CP, without other identified disease, aged 2 to 6 years, who drool ($n = 33$) or did not drool ($n = 14$), were included. The dependent variables were the physical health summary scores and the psychosocial health summary scores of the Pediatric Quality of Life Inventory version 4.0. The t test, Pearson product-moment correlation, Mann-Whitney U test and stepwise regression analysis were used for statistical analysis.

Results: The physical health and psychosocial health summary scores of the children that drooled (16.29 ± 15.97 and 42.92 ± 17.57 , respectively) were lower than for the children that did not drool (31.97 ± 22.22 and 57.09 ± 12.21 , respectively; $P < 0.01$). The drooling ranking score was negatively correlated with the physical health summary score ($r = -0.355$; $P < 0.05$) and the psychosocial health summary score ($r = -0.381$; $P < 0.01$). The stepwise regression showed that gross motor development and the drooling ranking score predicted 56.6% of the variability of the physical health summary score ($R^2 = 0.566$; $P < 0.01$). The language development score predicted 25.6% of the variability of the psychosocial health summary score ($R^2 = 0.256$; $P < 0.01$).

Conclusion: Drooling was associated with a lower HRQOL. Prediction of the physical health summary score was more closely associated with gross motor development and the drooling ranking scores. Prediction of the psychosocial health summary score was more closely associated with the language development of children with CP aged 2 to 6 years.

Keywords: cerebral palsy, drooling, health-related quality of life

Introduction

Drooling is common among children with cerebral palsy (CP). The estimated prevalence has been reported from 16.8% to 58%.¹⁻⁵ Frequent drooling may cause skin maceration and infection, body fluid loss, and recurrent pneumonia.^{6,7} At school and at home, children with salivary secretions may cause damage to books, teaching materials and furniture, and it even interferes with social relationships.^{6,8,9} van der Burg et al^{8,10} reported that children with CP that drool are often avoided by other children, and familiar and unfamiliar adults (including their parents). Hockstein et al⁹ reported that drooling in children with CP could interfere with their education and increase their dependent level of care. Such studies suggest that drooling might be associated with a reduced quality of life among children with CP.

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Previous studies have shown that there are many factors that interfere with the health-related quality of life (HRQOL) in children with CP including: motor, cognitive, language, and social impairment.^{11–14} Although the prevalence of drooling is high among children with CP, there are few articles regarding its relationship to the HRQOL in these children. Most prior studies^{8,10,15} have used modified questionnaires or qualitative methods to evaluate the relationship between drooling and HRQOL. In this study, standardized measurement of the HRQOL in children with CP, with and without drooling, was investigated. In addition, the relationship between drooling and HRQOL was evaluated, as well as the factors that predict the variability of HRQOL in these children.

Materials and methods

Participants

Children with CP that attended one medical center hospital and two early intervention institutions at daytime for an early intervention and habilitation program in Taiwan were enrolled. Children with CP, aged 2 to 6 years, without drooling (with a drooling ranking score of 2 according to the Drooling Rating Scale developed by Thomas-Stonell and Greenberg¹⁶), were the control group. Children with CP, aged 2 to 6 years, with drooling (a drooling ranking score > 2), were the study group. The exclusion criteria were: (1) children with CP combined with other problems such as congenital malformation or metabolic disorder, (2) children taking anticholinergic drugs over the past 2 months, and (3) children with an acute infection or other systemic disease. Children with CP were enrolled consecutively from February 2011 to July 2011. The study was conducted according to the criteria of the Declaration of Helsinki and the review board of the university hospital approved this study. Informed consents were obtained by parents.

Forty-seven children were included in the study: 14 did not drool (mean age: 43.2 ± 13.8 months, diplegia 17%, quadriplegia 12.8%) and 33 did drool (mean age: 48.9 ± 14.4 months, diplegia 10.6%, quadriplegia 59.6%). In the group without drooling, there were eight boys and six girls, and in the group with drooling, there were 22 boys and 11 girls (Table 1). Previous studies^{4,17} have reported that drooling in infancy usually resolves by 18 months of age. The children that drool beyond 4 years of age are considered abnormal and require further treatment. Therefore, the participants were divided into groups; those less than 4 years old (>2 years and <4 years, as the age group where drooling was supposed to stop and conservative management of drooling was acceptable) and those older than 4 years of age

(≥4 years, as the age group where drooling was abnormal and required further treatment). Table 1 compares the rate drooling and HRQOL of different age groups.

Outcome measures

The ranking of drooling was evaluated using the Drooling Rating Scale as described by Thomas-Stonell and Greenberg.¹⁶ The Drooling Rating Scale was rated using two subscales: (1) drooling severity with a scale from 1 to 5, where 1 = never drools, 2 = mild drooling causing wet lips only, 3 = moderate drooling causing wet lips and chin, 4 = severe drooling where clothing becomes damp, and 5 = profuse drooling causing clothing, hands, and the subjects in general to get wet; (2) drooling frequency with a scale from 1 to 4, where 1 = never drools, 2 = occasionally drools, 3 = frequently drools, and 4 = constantly drools. The drooling ranking score was determined by adding the two subscales together (drooling severity + drooling frequency). The drooling ranking score was rated from 2 to 9, with a ranking score of 2 representing no drooling and a ranking score of 9 as the most severe level of drooling. The parents or the children's primary caregivers rated the drooling, at least 1 hour before or after meals.

The HRQOL of these children was rated using the Pediatric Quality of Life Inventory Version 4.0 (PedsQL 4.0) for toddlers (ages 2–4) and young children (ages 5–7) using the generic core scales, which combined four subscales that included: (1) Physical Functioning, (2) Emotional Functioning, (3) Social Functioning, and (4) School Functioning. The scoring dimensions included: (1) a physical health summary score that represented

Table 1 Demographic characteristics of children with cerebral palsy (CP)

	Drooling (+) no	Drooling (-) no	Statistic
Sex			$\phi = -0.091$
Boy	22	8	
Girl	11	6	
Age			$\phi = -0.042$
<4 years	15 (26–46 months)	7 (24–41 months)	
≥4 years	18 (48–71 months)	7 (49–66 months)	
Type of CP (no)			$\phi = -0.429^a$
Diplegia	5	8	
Quadriplegia	28	6	
Developmental status (month)			
Language	19.64 ± 17.90	42.36 ± 13.45	Z = -3.73 ^a
Gross motor	12.88 ± 12.54	16.36 ± 13.16	Z = -1.18
Cognition	19.76 ± 17.88	43.36 ± 17.76	Z = -3.58 ^a

Notes: Drooling (+): children with drooling; Drooling (-): children without drooling; ϕ : Chi square ϕ ; Z, Z score of Mann-Whitney U test; ^aP < 0.01.

a physical functioning scale score, and (2) a psychosocial health summary score that was the sum of the items over the number of items answered in the Emotional, Social, and School Functioning Scales. Parents or the primary caregivers rated the PedsQL 4.0 scores (internal consistency reliability: total scale score Cronbach's $\alpha = 0.9$ for parents/proxy; physical health summary score Cronbach's $\alpha = 0.88$ for parents/proxy; psychosocial health summary score Cronbach's $\alpha = 0.86$ for parents/proxy).¹⁸

The developmental status of the children was evaluated using the Developmental Screening Test for 0–6-Year-Old Children (Chinese version, content validity 0.89–0.93),¹⁹ which was also rated by the parents or primary caregivers. This test evaluated five developmental domains (language, social-personality, gross motor, fine motor, and cognition); the highest scores achieved for each domain were considered the developmental levels. The screening test was correlated with the Bayley Scales of Infant Development I (criterion-related validity $r = 0.229$ – 0.566 ; $P < 0.05$).¹⁹

Data analysis

The mean differences in HRQOL scores among the two groups were analyzed with the Mann–Whitney U test. The correlation of the drooling ranking score, the developmental status, and the HRQOL was analyzed with the Pearson product–moment correlation coefficient. Stepwise regression analysis was used for the prediction of variability of the HRQOL. Statistical analyses were performed using SPSS software (v 12.0; IBM, Armonk, NY). The significance level was set at $P < 0.05$.

Results

There were no significant correlations with sex and age of those that drooled compared to those that did not drool (Table 1). However, the diagnosis of diplegia or quadriplegia was associated with a significant difference; there were more children with quadriplegia that drooled and fewer with diplegia that drooled ($P < 0.01$, Table 1). Table 1 also shows a comparison of the developmental status between those that drooled and those that did not drool. The language developmental status (in months) of the children that drooled was significantly lower compared to the children that did not drool (19.64 ± 17.90 vs 42.36 ± 13.45 ; $P < 0.01$). The cognitive developmental status of the children that drooled was lower than in the children that did not drool (19.76 ± 17.88 vs 43.36 ± 17.76 ; $P < 0.01$). There was no significant difference in the gross motor developmental status between the children that drooled and those that did not drool (12.88 ± 12.54 vs 16.36 ± 13.16 ; $P > 0.05$).

There were no significant differences in the HRQOL (physical health summary score and psychosocial health summary score) by sex (18.96 ± 17.43 vs 24.49 ± 22.16 , $t = -0.95$, $P > 0.05$ and 49.20 ± 16.68 vs 43.51 ± 18.31 , $t = 1.09$, $P > 0.05$, respectively; Table 2) and by age group (25.57 ± 19.66 vs 16.90 ± 28.25 , $t = 1.57$, $P > 0.05$ and 52.13 ± 16.43 vs 42.75 ± 17.19 , $t = 1.91$, $P > 0.05$, respectively). The physical health summary scores of the quadriplegic group were significantly lower than those of the diplegic group (34.19 ± 18.69 vs 15.90 ± 17.11 , $t = 3.20$, $P < 0.01$); however, there were no significant differences observed on the psychosocial health summary scores (53.48 ± 10.70 vs 44.72 ± 18.82 , $t = 1.58$, $P > 0.05$). For the children that drooled, both the physical health summary scores and the psychosocial health summary scores were significantly lower than the scores for the children that did not drool (31.97 ± 22.22 vs 16.29 ± 15.97 , $t = 2.73$, $P < 0.01$ and 57.09 ± 12.21 vs 42.92 ± 17.57 , $t = 2.74$, $P < 0.01$, respectively).

In order to determine which variables (developmental status and drooling ranking scales) could be enrolled in stepwise regression to predict the variability of HRQOL, correlations between the variables and the HRQOL were studied first (Table 3). Those predictor variables which were significant correlated with HRQOL were enrolled in the stepwise regression study. The drooling ranking score was negatively correlated with the physical health summary score ($r = -0.355$; $P < 0.05$) and psychosocial health summary score ($r = -0.381$; $P < 0.01$). The developmental status (language, gross motor, and cognition) of the children was

Table 2 Comparison of sex, age, type of CP, and drooling with the HRQOL

	Physical summary score		Psychosocial summary score	
	Mean \pm SD	t	Mean \pm SD	t
Sex				
Boy	18.96 \pm 17.43	-0.95	49.20 \pm 16.68	1.09
Girl	24.49 \pm 22.16		43.51 \pm 18.31	
Age				
<4 years	25.57 \pm 19.66	1.57	52.13 \pm 16.43	1.91
\geq 4 years	16.90 \pm 28.25		42.75 \pm 17.19	
Type of CP				
Diplegia	34.19 \pm 18.69	3.20 ^a	53.48 \pm 10.70	1.58
Quadriplegia	15.90 \pm 17.11		44.72 \pm 18.82	
Drooling				
Drooling (–)	31.97 \pm 22.22	2.73 ^a	57.09 \pm 12.21	2.74 ^a
Drooling (+)	16.29 \pm 15.97		42.92 \pm 17.57	

Notes: Statistics with the t test; ^a $P < 0.01$.

Abbreviations: CP, cerebral palsy; HRQOL, health-related quality of life; SD, standard deviation.

Table 3 Correlations between drooling ranking, developmental status, and HRQOL

	Physical health summary (r)	Psychosocial health summary (r)	Drooling ranking (r)	Language (r)	Gross motor (r)
Drooling ranking	-0.355 ^a	-0.381 ^b			
Language	0.477 ^b	0.522 ^b	-0.589 ^b		
Gross motor	0.716 ^b	0.383 ^b	-0.123	0.458 ^b	
Cognition	0.503 ^b	0.516 ^b	-0.537 ^b	0.900 ^b	0.501 ^b

Notes: r, Pearson correlation coefficient; ^a $P < 0.05$; ^b $P < 0.01$.

Abbreviation: HRQOL, health-related quality of life.

positively correlated with both the physical health summary score ($r = 0.477$, 0.716 , and 0.503 , respectively; $P < 0.01$) and the psychosocial health summary score ($r = 0.522$, 0.383 , and 0.516 , respectively, $P < 0.01$). Thus the drooling ranking score and the developmental status of language, gross motor, and cognition factors were included in the stepwise regression analysis because they were significantly correlated with HRQOL. The results of the stepwise regression are shown in Table 4. For variability of the physical health summary score, the developmental level of language and cognition was excluded after the stepwise regression (due to $P > 0.1$). The drooling ranking score and gross motor development predicted 56.6% of the variation of the physical health summary score ($R^2 = 0.566$; $P < 0.01$). For the prediction of variability of the psychosocial health summary score, the drooling ranking score, gross motor development, and cognitive development scores were excluded after stepwise regression (due to $P > 0.1$). The language development level predicted 25.6% of the variation of the psychosocial health summary score ($R^2 = 0.256$; $P < 0.01$).

Discussion

The frequency of drooling in the quadriplegic children was greater than in the diplegic children. This result is compatible with the findings of Hegde and Pani³ that showed that patients with quadriplegia had the most severe drooling, followed

by children with diplegia and the least affected children had athetoid CP. But other reports showed opposite results and they found the drooling is more prevalent and intense in children with dyskinetic CP than in children with spastic CP.^{5,20} The main etiologies of drooling include: impaired postural control as well as oral-motor and swallowing abnormalities.^{3,20-22} The quadriplegic children with CP are more likely to develop drooling due to their more extensive brain dysfunction and poor oral motor and sensory function compared to the diplegic children with CP. In this paper, there was no dyskinetic-type CP enrolled for study so we cannot compare the drooling problem with other CP subtypes.

The results of this study showed that the language and cognitive development of the children that drooled was lower than in the children that did not drool; however, the gross motor development showed no significant difference between these two groups (Table 1). These findings are compatible with the report of Senner et al²³ that showed that children who drooled had more severe dysarthria and impaired nonverbal intelligence, but their gross motor status was not more impaired; although impaired motor control was considered one of the contributing factors of drooling.^{21,22}

Children at different developmental stages may have different findings and explanations for their disease.^{24,25} Younger children were less likely to be perceived as performing less well on the HRQOL questions than older children.²⁶ It is possible that finding no significant HRQOL differences between age groups, in this study, was due to the younger ages of our study groups. Previous studies^{12,14} also revealed that the HRQOL (both physical and psychosocial) of quadriplegic children was lower than that of diplegic children. In this study, the physical health summary scores of quadriplegic children were lower than diplegic children; however, the psychosocial health summary score was not. The possible explanations for this result include: (1) most of the subjects attended early intervention institutions or a hospital program, where they possibly had a more supported and structured environment and received less negative feedback from social interaction;^{10,14} (2) most of the subjects were young and

Table 4 Stepwise regression of drooling ranking and developmental status

	Physical health summary score				Psychosocial health summary score			
	β	t	P	Tolerance	β	t	P	Tolerance
Drooling ranking	-0.271	-2.772	<0.01		(-)			0.653
Language	(-)			0.501	0.522	4.103	<0.01	
Gross motor	0.683	6.977	<0.01		(-)			0.790
Cognition	(-)			0.519	(-)			0.191

Note: (-): excluded in stepwise regression.

cognitively impaired; the impairment was known since birth and they received support from their caregivers to accomplish daily activities, and therefore had fewer experiences of negative psychosocial well-being;^{11,27} (3) the number of subjects was too small to show a statistically significant difference in psychosocial health scores.

The results of this study showed that the physical health summary scores and psychosocial health summary scores were significantly lower in the children with CP that drooled than in the children with CP that did not drool. These results were compatible with previous studies showing that drooling may lead to health-related problems such as skin maceration, recurrent pneumonia, and malnutrition.^{7,28} Although our result showed a significant level of correlation between drooling and psychosocial HRQOL, but the correlation coefficient showed only a lower level of correlation ($r = -0.381$; $P < 0.01$). Some studies have reported that drooling was associated with impaired social relationships of these children with adults and their peers; however, few showed negative emotional reactions due to drooling.^{8,10,15} van der Burg et al¹⁰ reported that children with CP who drooled showed few negative emotional reactions because they attended special education schools and the drooling problem seems to be acceptable and ignored in these places.

Although the language and cognitive developmental status was positively correlated with the physical health summary score, these two variables were excluded from the stepwise regression model because of the statistical result of $P \geq 0.1$. Thus, the most important variables left in the stepwise regression model were gross motor development and rank of drooling, and they predicted 56.6% of the variation of the physical health summary score. In predicting the variation associated with the psychosocial health summary score, the level of drooling, gross motor development and cognitive development were excluded from the stepwise regression model ($P \geq 0.1$); the remaining language development predicted 25.6% of the variation of the psychosocial health summary score. Dickinson et al¹¹ showed that gross motor development correlated most with the physical wellness of children with CP; intellectual disability and language impairment significantly interfered with psychosocial wellness. However, drooling was not investigated as a problem that interfered with the quality of life in this prior study. In our study, only the ranking of drooling/gross motor development and language development were considered as important factors associated with the physical health summary score and psychosocial health summary score. There are likely other factors involved that were not evaluated in this study.

Further investigation of these possible variables (eg, family or institutional factors, Gross Motor Function Classification System of children with CP) that might be correlated with the HRQOL of children with CP requires further research.

Although cognitive development was excluded in the stepwise regression model, its importance cannot be overlooked due to nearly the same correlation coefficient associated with the psychosocial health summary score as with the language development score. However, the excluded variables did not add much to predicting the variability of the psychosocial health summary score, when it was included.

Because the subjects in this study were too young or too cognitively impaired, a parent/proxy report was used to assess the drooling and HRQOL of the enrolled children. A bias might have been introduced by parents that were bothered by their children's drooling and inclined to rate their children's HRQOL lower. As in the study by Davis et al,²⁹ there was discordance between parent/proxy and child self-report because they responded to the HRQOL questionnaire items differently. But in this study, the definitions of drooling severity and frequency were clearly defined for parents and primary caregivers. In addition, the standard HRQOL questionnaires were used for the HRQOL evaluation, and were not likely significantly affected by bias. Although a self-reported HRQOL is standard for the perceived HRQOL, the parents are valuable proxies to assess their children's HRQOL if the children are too young or too cognitively impaired to complete a self-reported HRQOL.^{10,30} Therefore, the findings of this study based on parent/proxy reports showed that children with CP that drooled had a lower HRQOL are important.

Study limitations

This was a cross-sectional study and focused only on 2- to 6-year-old children with diplegic and quadriplegic CP from a university hospital and early intervention institutions. Because our subjects in this paper were young and recruited from localized areas in Taiwan, and contained only two CP subtypes, the results cannot be generalized to all ages and all groups of children with CP.

Conclusion

In conclusion, the standard assessment inventory was used to evaluate the correlation of drooling on the HRQOL of children with CP. The more severe the drooling was (without considering the type of CP), the lower the physical and psychosocial health quality of life was in the children with CP. The gross motor development level and ranking of drooling predicted the

physical health score better, and the language development level predicted the psychosocial health score better. With regard to providing early intervention programs for children with CP, their developmental status should be assessed as well as their drooling problem, which has a negative correlation on their HRQOL. The focus of this study was on young children with CP; further evaluation is needed to determine similar correlations among older children with CP.

Disclosure

No commercial party having a direct financial interest in the results of the research supporting this article has or will confer a benefit upon the authors or upon any organization with which the authors are associated.

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