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Impact of Tube Feeding Route on BMI, Quality of Life Comparing PROMIS[®] and Neuro-QoL in Pediatric Patients with Cerebral Palsy

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Abstract

Background: Tube feeding is common in patients with CP, but there is no data on its impact on quality of life (QoL).

Aims: We assessed correlation between similarly categorized QoL domains measured by PROMIS[®] and Neuro-QoL in a population of pediatric patients with cerebral palsy (CP). We investigated the relationship between feeding method and body mass index (BMI), and assessed the association between feeding method and QoL.

Methods: Data were collected through retrospective medical chart review. Pearson correlation was run to determine correlation between similarly categorized Neuro-QoL and PROMIS[®] domains. Association between feeding method and BMI, as well as feeding method and QoL score, were measured with bivariable and multivariable linear regression. Mean age of participants was 9 years, 10 months (4 years, 4 months); 35 participants were male and 23 females (n=58).

Results: Pearson correlation analysis between similarly categorized QoL domains were statistically significant ($p \leq 0.05$). There was no association between feeding method and BMI, nor feeding tube method and QoL scores as measured by the following QoL domains: anxiety, depression, fatigue ($p \leq 0.05$).

Conclusion: This study found no association between feeding tube method and QoL scores, using PROMIS[®] and Neuro-QoL, in pediatric patients with CP. However, there were statistically significant correlations between similarly categorized outcome domains. Future research should focus on building accurate and robust datasets to better power studies that will assess QoL for this population and their caregivers.

Keywords: Cerebral palsy; Pediatrics; Tube feeding route; Neuro-QoL

Background

Children with cerebral palsy (CP) may be at high risk for inadequate growth and development, in part due to nutritional insufficiencies from dysphagia and dysmotility [1]. Mealtimes often provide the opportunity for social interaction and bonding, so it is preferable to feed these children in a traditional manner; however, oral feeding is often not a feasible option for ensuring optimal calorie intake in this population. In addition to difficulty swallowing, studies have reported food spillage and lengthy feeding times as additional barriers to adequate nourishment [2]. Therefore, artificial nutrition may be an appropriate option for some patients. There are two common methods of artificial feeding via the intestine. These include tubes implanted through the abdominal wall [e.g. gastrostomy tube (G-tube)], most commonly percutaneous endoscopic gastrostomy (PEG), and nasogastric tube (NGT) [3].

PEG placement is an endoscopic surgical procedure cited as a safe and reliable feeding method in children with severe neurological impairment [4]. During the procedure, a physician

places an endoscope into the stomach, via the mouth and esophagus. The endoscope is used to ensure correct positioning of the feeding tube through the wall of the abdomen and stomach [5]. Complication rates as low as 4% have been reported for PEG placement, and it is considered a less invasive procedure compared to surgical gastrostomy [6]. However, once inserted, PEGs are also associated with risks and complications, including infection, gastro-esophageal reflux, and vomiting, obstruction, and tube dislodgement [3].

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NGT placement is another option [7,8]. NGTs can be placed in patients at bedside; the tube is inserted through one of the nostrils, advanced down the esophagus and into the stomach. Patrick et al found, in a randomized control trial, that using NGT in underweight cerebral palsy pediatric patients resulted in both statistically and clinically significant weight gain compared to those patients who had standard feeding [8]. Typically, NGTs are used as a short-term option in this population to help patients achieve optimal weight. If long-term feeding assistance is needed, PEGs may be considered, but are not required [9].

While it has been demonstrated that objective nutritional outcomes, such as weight-for-height and height-for-weight, can be improved in this population using artificial feeding methods [8,10], fewer studies have assessed quality of life (QoL) outcomes for both patients and caregivers [6,11,12]. Artificial feeding methods can help these patients increase their caloric intake and attain optimal weight ranges. This may help with some QoL outcomes, but it is important to consider risks and tradeoffs to QoL, as well. For example, meals provide a time in the home for family members to gather around the table and talk to one another. While artificial feeding may eliminate a parent or caregiver's stress of providing adequate calories to her child, this may introduce new stress for the child and/or family.

Artificial feeding methods can be seen as a technological intrusion into what should be a natural and positive emotional experience [13]. It is also important to consider that it may not be beneficial for the family to have mealtime feel more like an appointment than enjoy its social value.

Assessing QoL can be challenging in this population, since patients are often subject to "floor effect" when completing QoL surveys or having caregiver's complete questionnaires on their behalf [14]. "Floor effect" occurs when patients' severity of illness is so extensive that they almost always score the minimum value on an assessment. The lack of sensitivity in QoL questionnaires for the general population of children makes it difficult to use to assess effectiveness of interventions in pediatric patients with neuromuscular diseases [14]. However, the Patient Reported Outcomes Measurement Information System (PROMIS®) tool, developed at the National Institute of Health (NIH), and Neuro-QoL are two tools that may have enough sensitivity to capture true QoL status in these patients. While Neuro-QoL has been validated in patients with neuromuscular diseases, including CP, work is in progress for validating PROMIS® domains in this population [15]. Assessing the correlation between similarly categorized domains in these QoL instruments may aid in these efforts. PROMIS® is one of the largest QoL databases; therefore, validating it in this population would allow researchers the opportunity to have access to enough data to better support research in this area. Accurately capturing these QoL measures can aid in clinicians' decisions on how to allocate more attention and resources to this population.

We utilized data as collected for the original aims of a larger parent study to explore these objectives: correlation between similarly categorized parent-proxy and self-report domains of the QoL assessments PROMIS® and Neuro-QoL, association between method of feeding (oral vs. ever tube-fed) and BMI, and association between feeding method and QoL as measured by the following domains of both PROMIS® and Neuro-QoL, anxiety, depression, and fatigue. All objectives were explored in a pediatric population with CP.

Methods

Parent study

We analyzed data that was originally collected for identifying inflammatory markers that would effectively risk stratify pre-operative patients with neuromuscular scoliosis. This data had also been collected to assess the validity of PROMIS® and Neuro-QoL pediatric domains in patients with CP or muscular dystrophy between 5 to 17 years, as well as the validity of PROMIS® proxy-assessment in the same population.

Participants

The study population consisted of 58 patients recruited through Columbia University Medical Center, Division of Pediatric Orthopedic Surgery at Morgan Stanley Children's Hospital in New York, NY. Inclusion criteria included all pediatric patients 0 to 18 years of age with a diagnosis of CP who underwent surgery to correct spinal deformity. Patients over the age of 18 or with incomplete outcomes data (e.g. no T score for PROMIS® or Neuro-QoL) were excluded. Both patients and caregivers completed PROMIS® and Neuro-QoL questionnaires. The participants were selected into the study based off the inclusion and exclusion criteria listed above, as well as completeness of the data for each individual participant. All participants completed consent forms before filling out the questionnaires.

Variable Classification

Outcome variables

PROMIS® is a reliable measurement tool that assesses health status for physical, mental, and social well-being, through both self-reported and parent-proxy measures [15]. It was designed for use in the general population, as well as across medical conditions, though it has not been validated specifically in a pediatric population with CP. Significant work is underway to validate PROMIS® in populations with disabilities, including the work reported here, at Columbia University Medical Center.

Neuro-QoL is a validated set of self-report and proxy-report measures that specifically assesses the health-related quality of life of children and adults with neuromuscular diseases, including CP. It is comprised of item domains and scales that evaluate symptoms, concerns, and issues that are relevant across disorders, along with measures that assess areas most relevant for specific patient populations [16].

Both short-forms and computerized adaptive testing (CAT) are available for PROMIS® and Neuro-QoL. When patients and caregivers were recruited for the study, they would typically consent to the study registry during a clinical visit. During the subsequent visit, patients and caregivers were given unique login information to fill out the both the short-forms and CATs for all domains of PROMIS® and Neuro-QoL. If participants were unable or unwilling to consent to the registry or fill out the questionnaires in clinic, they were contacted via phone and e-mail and given the opportunity to complete questionnaires at home. All PROMIS® and Neuro-QoL data was available through assessmentcenter.net. If a questionnaire domain was filled out more than once for a patient for any domain, an average of the scores was calculated. Of note, for CATs, certain questions are skipped based on the respondent's answer for a previous question; conversely, short-forms require the respondent to answer all questions.

Decisions about the specific domains to analyze from PROMIS® and Neuro-QoL were made based on completion frequency as well as comparability across the two QoL tools. All PROMIS® domains analyzed were parent-proxy, while the questions in the Neuro-QoL domains chosen were both self-report and proxy. For both assessments, CATs were filled out more completely than short-forms; therefore, they were chosen for analysis. The domains, with example questions, that fit these criteria are presented in **Table 1**.

Both Neuro-QoL and PROMIS® instruments use a T-score scoring system, which means that the mean is 50 and the standard deviation is ten 15. A T score allows for taking individual scores and transforming them into a standardized form to compare scores across QoL domains. For the Neuro-QoL and PROMIS® domains used, the mean of the US general population was 50 [15,16].

Exposure

The sole exposure investigated was feeding method, defined as either orally fed or ever having had a feeding tube prior to filling out QoL questionnaires. Feeding tubes included.

Determination of feeding method was decided through retrospective medical chart review. The first author reviewed patients’ physician notes within their medical charts. If indication of ever having an artificial feeding method was noted, the patient was categorized as ever tube-fed. If no such indication was present, the patient was categorized as orally fed.

Covariates

The following covariates were considered: body mass index (BMI), age (in years), Gross Motor Function Classification System (GMFCS), gender, and QoL domains for the same scale. We chose these covariates because of their availability in the existing database and their use in other studies, particularly GMFCS level. GMFCS was dichotomized into “higher function” (GMFCS I-III) and “limited function” (GMFCS IV-V). Patient charts were retrospectively reviewed to obtain height and weight, and BMI was calculated for all patients with both variables. Heights and weights were taken from the year that the patient’s QoL questionnaires

were completed. Not all height and weight values were available due to difficulty in obtaining these measures in wheelchair bound patients. Specific CP diagnosis and age at which patient questionnaires were completed was gathered from a tracking spreadsheet designed for the original studies’ research questions.

Statistical analysis

Univariable descriptive analysis was reported for the 58 included study participants. Frequencies were reported for binary, nominal, and ordinal variables (gender, feeding method, CP diagnosis, and GMFCS level). Mean and standard deviation were reported for continuous variables (age and BMI). All Neuro-QoL and PROMIS® data were checked for normality by looking at their distribution and probability plots and the Shapiro-Wilk test statistic. All outcome variables followed a normal or approximately normal distribution.

Pearson correlation analysis was performed to assess the correlation between similarly categorized PROMIS® and Neuro-QoL domains, specifically “Anxiety,” “Depression,” and “Fatigue”. An example model is provided **[Equation 1]**.

$$BMI = \beta_0 + \beta_1 \times \text{feeding_method} + \beta_2 \times \text{GMFCS} \quad \text{[Equation 1]}$$

For this example, model reference group was “oral” for feeding method and “GMFCS level I- III” for GMFCS level. We also assessed the association between feeding method and each QoL domain using multivariable regression models adjusting for age, gender, and other QoL domains for the same scale.

An analysis for this paper was generated using SAS software, Version 9.4. Copyright © 2013 SAS Institute Inc. Analyses were also generated using R Development Core Team, 2008. R: A language and environment for statistical computing. R Foundation for Statistical Computing, Vienna, Austria. ISBN 3-900051-07-0. URL <http://www.R-project.org>. This study was approved by Columbia University Medical Center’s Institutional Review Board under the following protocols: IRB-AAAM7459 and IRB-AAAK7607. This study was carried out in accordance with the Helsinki Declaration

Table 1: Sample questions for represented Neuro-QoL and PROMIS® domains.

Domains	Neuro-QoL Example	PROMIS® Example
Anxiety n=19 n*=13	In the past 7 days I become anxious when I go back to the hospital or clinic	In the past 7 days my child worried about what could happen to him/her
	Not at all	Answer Choices
	A little bit	Never
	Somewhat	Almost Never
	Quite a bit	Sometimes
	Very Much	Often
		Almost Always
Depression n=16 n*=14	In the past 7 days it was hard for me to have fun	In the past 7 days it was hard for me child to have fun
	Never	
	Almost Never	
	Sometimes	
	Often	
	Almost Always	
Fatigue n=11 n*=23	In the past 7 days I had trouble starting things because I was too tired	In the past 7 days my child was so tired it was hard for him/her to pay attention
	None of the time	
	A little bit of the time	
	Some of the time	
	Most of the time	
	All of the time	

Note: Neuro-QoL n and PROMIS® n* are for number of questions in each domain, respectively

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of 1975 and 2000 revisions, including that participants were not worse off at the end of the study period.

Results

Description of the study sample is presented in **Table 2**. 72 patients were eligible for this study. 8 were excluded due to lack of Neuro-QoL and PROMIS® data and 6 were excluded for having a neuromuscular disease diagnosis that was not CP specific. 58 patients were included in the study for analysis. Most patients in this sample were male, and the average age for all subjects was 9.86 (4.30) years. 9 distinct CP diagnoses were represented based on ICD-9 code records with diplegic and quadriplegic CP most highly represented.

GMFCS levels were approximately proportionate, with GMFCS

II being most highly represented. Most patients were classified as “oral” for feeding method, and the average BMI for the group was 16.78 (3.46).

Three sets of similarly categorized PROMIS® and Neuro-QoL domains were assessed for correlation (**Table 3**). Pearson correlations between similarly categorized Neuro-QoL and PROMIS® domains were all statistically significant at alpha level 0.05, with the fatigue domains being most highly correlated. Correlations between QoL measures are presented in **Table 4**. BMI was not significantly greater among patients who were categorized as “ever tube-fed,” controlling for GMFCS, age, and gender. Multivariable models were adjusted for age, gender, and other QoL domains for the same scale. There were no significant associations between feeding method and QoL domains at an alpha level of 0.05 (**Table 5**).

Table 2: Characteristics of CP patients with Neuro-QoL and PROMIS® scores.

n=58	n=35
Gender (Male)	
Age in years	9 years, 10 months (4 years, 4 months)
CP Diagnosis (%)	
Agenesis of the corpus callosum	2.08
Congenital hemiplegic CP	2.08
CP	8.33
CP NOS	22.92
Diplegic CP	29.17
Hemiplegic CP	22.92
Quadriplegic CP	29.17
Right hemiplegic CP	2.08
Spastic quadriplegic CP	2.08
GMFCS level (%)	
I	25
II	39.58
III	16.67
IV	14.58
V	25
GMFCS dichotomized (%)	
Higher Function (I-III)	67.24%
Limited Function (IV-V)	32.76%
Feeding method	
Ever Tube-Fed (%)	20.83
BMI	16.78 (3.46) (n missing=21)

Table 3: Mean anxiety, depression, and fatigue scores as measured by PROMIS and Neuro-QoL.

QoL Domain	Neuro-QoL Mean (SD)	PROMIS® Mean (SD)
Anxiety	53.14 (6.85)	53.22 (10.64) (n missing=9)
Depression	49.46 (7.75)	49.91 (10.26) (n missing=10)
Fatigue	50.36 (8.38) (n missing=2)	51.85 (10.84) (n missing=12)

Table 4: Pearson Correlations between similarly categorized Neuro-QoL and PROMIS® domains.

Neuro-QoL		Anxiety	Depression	Fatigue
PROMIS®	Anxiety	0.4491*	--	--
	Depression	--	0.4604*	--
	Fatigue	--	--	0.6253*

*Pearson Correlation is statistically significant at the $p \leq 0.05$ level

Table 5: Linear regression coefficients comparing ever tube-fed vs. oral feedings for selected Neuro-QoL and PROMIS® domains.

Outcome	β -value for "ever tube-fed"	β -value Unadjusted	SE	SE Unadjusted	<i>p</i>	<i>p</i> Unadjusted
Neuro-QoL						
Anxiety	-1.0877	1.7538	2.1889	2.3904	0.621	0.465
Depression	3.0832	3.095	2.1843	2.687	0.1644	0.254
Fatigue	-2.8698	2.171	2.8002	3.061	0.3105	0.481
PROMIS®						
Anxiety	0.7663	0.4947	2.8789	3.966	0.7915	0.901
Depression	0.0359	0.1872	2.5433	3.8346	0.9888	0.961
Fatigue	-2.9011	-2.16	3.0649	4.487	0.3499	0.633

Discussion

Our findings support the hypothesis that there is a significant correlation between PROMIS® and Neuro-QoL for the domains anxiety, depression, and fatigue. To our knowledge, whether these two QoL measures correlate has not been studied and could help to validate PROMIS® in this population. Future research should examine all other comparable domains between the two QoL instruments and examine if correlation is present for a larger group of patients.

Feeding method was not associated with BMI, nor was it associated with anxiety, depression, and fatigue domains of the QoL assessments, Neuro-QoL and PROMIS®. Based on prior studies [8], tube feeding can be very effective in reversing wasting in CP. Our study suggests that no tube feeding route (NGT, PEG or G-tube, and GJ-tube) is superior for weight gain. This could be due to our small sample size and the fact that we were not comparing weight pre- and post- enteral nutrition for individual patients. However, our data could also reflect no true association between feeding method and BMI, suggesting that methods for choosing and delivering optimal feeding methods in this population have improved.

Additionally, the hypothesis that ever having a feeding tube is associated with higher QoL scores is not supported in this study. This result is consistent with the finding that QoL, as rated by parents, did not increase following insertion of G and GJ tube in neurologically impaired children [10]. Due to small sample size, and a lack of randomization, it was also difficult to say the true direction of the associations. There could be no difference in QoL for several reasons. Our data could have been accurate in reflecting no association between feeding method and QoL, implying that enteral nutrition may have more positive impact than negative when it comes to patient and caregiver well-being.

Mahant et al. prospectively assessed QoL of neurologically impaired children (n=45) before and after gastrostomy and gastrojejunostomy tube insertion and found QoL as rated by parents did not increase. However, Sullivan et al found that children with cerebral palsy (n=57) experienced significant improvements in social functioning, mental health, energy/vitality and general health perception as reported by caregivers 12 months after gastrostomy placement. Again, our results, coupled with these studies, suggest non- superiority of gastric vs. nasal tubes.

There were limitations in this study. Small sample size (n=58) reduced power to identify significant relationships. No association was found between feeding method and BMI, nor feeding method and specific QoL domains. However, it is impossible to decipher

whether this is because there is truly no association, or the results are due to lack of statistical power. A related limitation was missing BMI data. Not having these measures for all patients made it difficult to present complete descriptive statistics, as well as adjust for these variables in linear regression analyses. It can be challenging to obtain these measures in this population because patients may be non-ambulatory. Another limitation was choosing which forms to analyze, CATs or short-forms. The decision to only analyze CATs was made after study participants completed the questionnaires. This decision was based on availability of data, though it is a potential source of bias. The reference population for the T-score distribution scoring system is another limitation. A US general population is used as the reference population for most PROMIS® and Neuro-QoL domains [14,15]. This is not surprising considering neuromuscular diseases are uncommon and there is need for substantially more research in this population, including accurate BMI and QoL data. With the lack of data currently available, it is not possible to have a reference population that reflects the patients represented in this study.

Researching fewer common diseases comes with both challenges and opportunities. Arguably the most difficult component for these investigations is recruiting an adequate sample size. Either large, multi-center data or collaboration is needed, or researchers must be able to adopt specialized study techniques and statistical methods to maximize a small dataset [17]. Limiting consideration to only a pediatric subgroup of this population makes it even more difficult to study these disorders, but that does not detract from the importance and need for meaningful research for these disorders. Future research should continue to focus on efforts for more robust methodology to better capture the data necessary to answer QoL questions specific to this population.

Conclusion

Comparative effectiveness studies of feeding tube route are lacking. Perhaps these could be accomplished by communication across hospital networks that specialize in this patient population to attain optimal sample sizes necessary for future work. With this information, we may be able to make more informed decisions when it comes to deciding feeding methods for these patients, and how it will affect their and their caregivers' quality of life.

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