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# CLINICAL ASSESSMENT OF DYSPHAGIA IN CHILDREN WITH CEREBRAL PALSY USING DDS AND DMSS

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## ABSTRACT

**AIM:** This study explored whether the Dysphagia Disorder Screening and Dysphagia Management Staging Scale (DDS and DMSS) are useful in children with cerebral palsy and which factors are related to the severity of their dysphagia.

**METHODS:** Swallowing competency was measured by two raters in 60 patients (47 male, 13 female; mean age 9y8mo, SD 2y9mo) with the DDS and DMSS. Patients were represented in all levels of Gross Motor Function Classification System and in all types of CP. Mean IQ was 67.98 (SD 21.52).

**RESULTS:** Good internal consistency in subparts and total DDS was found ( $\alpha \geq .88$ ). Good interrater reliability was found for all items of DDS, except for oral pharyngeal swallowing which was moderate. Strength of relationship between individual test items of DDS and severity level on DDS was high, evidenced in discriminant function analysis. Averages of scores of participants known with dysphagia were significantly higher than subjects not known with dysphagia. Part 1 and part 2 of the DDS were positively related (Spearman's  $R = .65$ ). The severity of dysphagia was related to GMFCS level and IQ, but there was no effect of age. Significant differences were detected in the scores in patients with dyskinesia versus patients with spasticity and patients with unclassified CP. However, no difference was detected between the latter two.

**INTERPRETATION** Our results support a positive internal consistency, construct validity, and interrater reliability, as well as the relation between DDS and DMSS for the use in children with CP. Moderate interrater reliability was found for oral pharyngeal swallow and underlines the use of additional necessity of objective assessment with videomanometry in the diagnosis dysphagia in this population. DDS and DMSS are the only validated clinical assessment tools and are useful in this population as they describe the essential aspects to be considered during a mealtime observation screening. Dysphagia is complex and its severity as well as its nature is clearly linked to the type of cerebral palsy the child presents with.

## INTRODUCTION

Cerebral palsy (CP) is a common problem, the worldwide incidence being 1.5 to 3 per 1000 live births.<sup>1</sup> It is defined as *a group of disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, cognition, communication, perception, and/or behavior, and/or by a seizure disorder.*<sup>2</sup> There are three main groups of CP: spasticity (79,2%), dyskinesia (14,4%) and ataxia (3,9%). The other 2.6% consists of other types of CP.<sup>3</sup> Reilly et al.<sup>4</sup> noted that in the long list of factors reported to be associated with CP, dysphagia was not mentioned in any of the accompanying articles or subsequent correspondence about the strengths and weaknesses of the revised definition and classification. However, the prevalence of dysphagia in children with CP was explored and confirmed by several studies.<sup>5-10</sup> Dysphagia is mostly described for the total population of CP, and not separately for the three main groups. As spasticity is the most common, the spastic group is overrepresented, and dyskinetic and atactic CP underrepresented. Yet, there are not negligible differences between these groups with regard to muscle tone. In spasticity, there is a persisting increased muscle tone in one or more limbs. When the tone is varying, the CP will be classified as dyskinetic CP. In atactic CP, there is a generalized hypotonia with signs of ataxia.<sup>11</sup> These differences may suggest that the dysphagia varies by type of CP. Indeed, the swallowing mechanism relies on good coordination and cooperation progress of various muscles. Based on a parent's questionnaire, a comparison of feeding disorders in different types of neurological impairment was made by Sullivan et al.<sup>8</sup>, and differences between the three groups of CP were present. The first group 'with feeding problems' contained 10 % dyskinetic CP, 56 % spastic quadriplegia, 22% hemiplegia, 7% ataxic, 5% unclassifiable. In the second group 'without feeding problems' there were no dyskinetic responders, 45 % spastic quadriplegia, 48% hemiplegia, 4% ataxic, 3% unclassifiable. Although the use of parents questionnaires in the Sullivan study has methodological limitations,<sup>10</sup> the impression remains that the distribution of spasticity, dyskinesia, ataxia differs between patients with and without feeding problems.

Apart from the type of CP, other factors such as the severity of drooling, the severity of speech disorder, positive history of seizures, episodes of pneumonia,<sup>5,12</sup> developmental retardation<sup>5,13</sup> and severity of the functional impairment<sup>5,7</sup> can impact on the type of dysphagia. In general, the more severe the functional motor impairment, the more severe the

oral motor dysfunction is.<sup>7</sup> Waterman et al.<sup>5</sup> found that the poorer the trunk control, the higher the risk of dysphagia gets.

The link between body mass index (BMI) and dysphagia remains unclear. Calis et al.<sup>7</sup> reported in a study of 166 children with severe generalized CP with Gross Motor Function Classification System Level (GMFCS) IV or V and intellectual disability that 15% of the study population presented with a weight for height between the 25th and the 10th percentile, and that 28% had a weight for height lower than the 10th percentile. Remarkably for this group, the severity of dysphagia was positively related to the GMFCS level, but negatively related to the body mass index.

As dysphagia is common in patients with CP, thorough assessment is important. Videofluoroscopic swallow Study (VFSS) and Fiberoptic Endoscopic Evaluation of Swallow (FEES) are instrumental methods for the assessment of dysphagia. However, these methods have limitations and cannot be routinely performed on every child with CP. They remain subjective<sup>14</sup> and there is exposure to X-ray radiation during VFSS.<sup>9</sup> There are no clinical assessment tools to detect dysphagia in children with CP that are validated and standardized. At present, speech and language therapists in Flanders evaluate the swallowing process with protocols that are not specified for the group of cerebral palsy or with a selection of components from different protocols (A Meerschaert, personal communication 2009). The Dysphagia Disorder Screening and Dysphagia Management Staging Scale (DDS and DMSS)<sup>15</sup> are developed for adults with mental retardation. It is evidenced in earlier surveys that these tests can also be used for other populations,<sup>15</sup> but this potential has not yet been tested on children with CP. It is an accurate tool to localize the phase of swallowing in which the feeding problem occurs, and to provide an impression of the functional eating skills. The patients may then be referred for further assessment in order to design a treatment strategy. Depending on the nature and severity of the disorder found, treatment can sometimes be based solely on the results of the DDS. The DMSS is a 5-level scale which classifies the severity of the eating problem.

## AIM OF THE STUDY

Dysphagia is a major in children with cerebral palsy. Yet, there is a lack of clinical evaluation instruments that are validated and standardized for this population. It remains unclear which factors of the cerebral palsy (e.g. type of CP and level of functionality) are indicators for dysphagia.

The aim of the study is twofold. First, we aim to explore whether the Dysphagia Disorder Screening (DDS) and Dysphagia Management Staging Scale (DMSS) are appropriate assessment tools for the evaluation of dysphagia in children with CP. Second, we aim to assess the relationship between the severity of dysphagia and type of CP (spasticity, dyskinesia and ataxia), level of functionality (GMFCS level), cognitive function (IQ) and age.

## METHODS

### PARTICIPANTS

There were two groups of participants between 3 and 15 years of age: 21 children with CP with swallowing disorders and 40 children with CP without swallowing disorders. All children were associated with the school for special education BuBaO Dominiek Savio Institute (DSI), part of the Dienstencentrum Gid(t)s. DSI is a Flemish school for children with physical disability. The children were also associated with the medical pedagogical institute (MPI), which is specialized in children with physical disability. Only children whose swallowing disorders were described in their personal medical record of the MPI, were included in the first test group. The initial diagnosis of dysphagia, necessary for inclusion, was made by a medical specialist. Approval of the local medical research committee was obtained.

First, a list of all pupils with CP, divided in a group with and without known swallowing disorders, was generated. All CP children known with dysphagia were included in the first test group. An overview of the inclusion criteria is given in Table 1.

Table 1: inclusion criteria

| Inclusion criteria   |  |
|--|--|
| <b>Group WITH known swallowing disorders</b><br><br><b>N = 20</b>    | <ul style="list-style-type: none"> <li>- Diagnose cerebral palsy</li> <li>- Between 3 and 15 years</li> <li>- Known with swallowing disorders</li> <li>- Associated with BuBaO Dominiek Savio in Gits</li> <li>- Approval of local medical research committee</li> </ul>   |
| <b>Group WITHOUT known swallowing disorders</b><br><br><b>N = 40</b> | <ul style="list-style-type: none"> <li>- Diagnose cerebral palsy</li> <li>- Between 3 and 15 years</li> <li>- Unknown with swallowing disorders</li> <li>- Associated with BuBaO Dominiek Savio in Gits</li> <li>- Approval of local medical research committee</li> </ul> |

## INSTRUMENTS

The Dysphagia Disorder Survey (DDS) and Dysphagia Management Staging Scale (DMSS)<sup>15</sup> were used. These instruments were originally developed to screen institutionalized adults with mental disabilities for dysphagia and related eating disorders. Development of the DDS began in 1986, and the first version was revised in 1993, which resulted in the current version. The instrument could be used under the protection of a research agreement between the author J.J. Sheppard and the KULeuven. The manual for the DDS was revised and translated into Dutch in 2002. Clinical experience revealed that the DDS is also suitable for screening children and adolescents. A certification course is required for permission to use the DDS.<sup>15</sup> We used the Dutch scoring form, edition children's research.

The DDS is divided into 2 parts: (1) related factors (7 items) and (2) feeding & swallowing competency (8 items). Each item is assigned a mark. Depending on the item, the mark can vary between 0 and 1 to 0 and 4. The first part assesses evidenced dysphagia-related factors such as body mass index, diet and special feeding techniques. The second part, feeding and swallowing competency, assesses through mealtime observation the different components of the oral-preparatory phase, the oral phase, the pharyngeal phase and the esophageal phase of swallowing. Every food consistency (thick-liquid, solid and thin-liquid) is scored.<sup>15</sup> A detailed overview of the various items of the DDS is presented in Appendix 1.

The DMSS is a 5-level scale which classifies the severity of the eating problem: (1) no disorder, (2) mild disorder, (3) moderate disorder, (4) severe disorder and (5) profound disorder. The criteria used to define these levels are: the extent to which the eating process has been adapted, the medical consequences and the impact on the way food is offered. The DMSS is typically used in conjunction with the DDS.<sup>15</sup>

## PROCEDURE

All children were rated in a child friendly and familiar environment, at school. The observation was done simultaneously by 2 experienced speech language therapists, who are used to working with children with CP. The scoring was done independently. No aspect of the natural feeding situation was implemented or interrupted by the test leader for the benefit of this research project. The test always started with the first part of the DDS focusing on the related factors. Measuring and weighing was done by the medical service in DSI. The child's caregiver was asked questions about the child's medical history. The second part of the DDS focuses on the actual feeding and swallowing competency. Only the subject, the test leader(s)

and the food assistant (if necessary) were present during the observation. The child was seated in its usual feeding position. This was using an ordinary chair, a wheelchair or another adapted chair. As this assessment only concerned an observation of the natural feeding position, there were no risks involved. If a child was only allowed to take thickened liquids, the items related to thin-liquid diet were not assessed, but scored as a problem. The boluses were offered in the followed order: thick-liquid, thin-liquid and solid. Informed written approval for each participant was given by the local ethical research committee.

## STATISTICAL ANALYSIS

Statistical analysis was performed with SPSS 17.0 (SPSS Inc., Chicago IL, USA). The internal consistency for the subparts 1 and 2 and for the total DDS was determined by Cronbach's alpha. Values above .80 were considered as good.<sup>16</sup> Interrater reliability was determined by Cohen's Kappa. Values between .40 and .75 were considered as reasonable, values higher than .75 as strong.<sup>17</sup> The strength of the relationship between the individual test items of the DDS and the severity of dysphagia on the DMSS was measured by means of an discriminant analysis.<sup>18</sup> Discriminant evidence of construct validity was assessed by comparing DDS averages of children known with feeding disorders and children not known with feeding disorders, using a *t*-test. Statistical significance was indicated by  $p \leq .05$ .<sup>19</sup> The correlation between the scores of part 1 and part 2 of the DDS was assessed by a Spearman correlation coefficient.<sup>19</sup>

Associations between the total score on the DDS versus IQ and age were measured by a Pearson correlation coefficient. Associations between the total score on the DDS versus type of CP and GMFCS level were assessed by a Mann-Whitney *U*-test.<sup>17</sup> The functionality levels were divided into 2 groups: level 1-2-3 and level 4-5 of the GMFCS. This division is chosen, because, from level 4 onwards, the children are wheelchair-bound. Statistical significance was indicated by  $p \leq .05$ .<sup>17</sup>

## RESULTS

### DESCRIPTION OF THE POPULATION

The population comprised of 61 children with CP, of which 60 children were included according to the criteria of this research protocol. One child was totally tube-fed and was therefore excluded. Characteristics of the participants are listed in Table 2. The current study included the entire CP-population of the primary school (N=55), and the 5 youngest children

with CP of the secondary school (N=5). The participants were evaluated within a period of 2 months. Mean scores of the total DDS, DDS Part 1 and DDS part 2 were 8.83 (range 0-34), 3.95 (range 0-13) and 4.88 (range 0-22) respectively. On the DMSS, 63.30% of all 60 children was classified as having no disorder (level 1), 18.33% mild disorder (level 2), 5.00% moderate disorder (level 3), 11.67% severe disorder (level 4) and 1.67% profound disorder (level 5).

Table 2: Characteristics participants

| Characteristics participants                       |                    |
|--|--------------------|
| <b>Age (N = 60)</b>                                |                    |
| Mean   | 9y 8mo (SD 2y 9mo) |
| Range  | 3y 1mo – 15y 8mo   |
| <b>Sex (N = 60)</b>                                |                    |
| Male   | N = 47 (78.3 %)    |
| Female   | N = 13 (21.7 %)    |
| <b>Mental abilities (N = 55)<sup>a</sup></b>       |                    |
| Mean IQ  | 68.70 (SD 21.07)   |
| Normal mental abilities (IQ ≥ 85)                  | N = 12 (21.8 %)    |
| Borderline intellectual functioning (85 > IQ ≥ 70) | N = 11 (20.0 %)    |
| Mild mental retardation (70 > IQ ≥ 55)             | N = 21 (38.2 %)    |
| Moderate mental retardation (55 > IQ ≥ 40)         | N = 7 (12.7 %)     |
| Severe mental retardation (40 > IQ ≥ 25)           | N = 4 (7.3 %)      |
| <b>Type of CP (N = 60)</b>                         |                    |
| Spastic CP   | N = 45 (75.0 %)    |
| Dyskinetic CP                                      | N = 8 (13.3 %)     |
| Atactic CP   | N = 1 (1.7 %)      |
| unclassified                                       | N = 6 (10.0 %)     |
| <b>GMFCS level (N = 60)</b>                        |                    |
| Level 1  | N = 4 (6.7 %)      |
| Level 2  | N = 27 (45.0 %)    |
| Level 3  | N = 9 (15.0 %)     |
| Level 4  | N = 7 (11.7 %)     |
| Level 5  | N = 13 (21.7 %)    |
| <b>Feeding disorder (N = 60)</b>                   |                    |
| Known with feeding disorder                        | N = 20 (33.3 %)    |
| Not known with feeding disorder                    | N = 40 (66.6 %)    |

<sup>a</sup> no TIQ available for 5 participants (recently included in the institute, excessive motor problems, or excessive disparity VIQ and PIQ)

## DDS & DMSS: USEFUL IN CHILDREN WITH CP?

### RELIABILITY

For the reliability tests, the results of the 60 evaluated participants were used (N = 60). Inter-item reliability of the DDS was high. The Cronbach's alpha's were higher than .80 on the individual parts 'related items' ( $\alpha = .88$ ) and 'eating and swallowing competency' ( $\alpha = .92$ ) as well as on the Total DDS ( $\alpha = .95$ ).



For almost all items of the DDS part 2, there was strong agreement ( $K > .75$ ). Only for item 13 “oral-pharyngeal swallow”, there was a reasonable degree of agreement ( $K = .57$ ). There was one pair of raters. For practical reasons, only 20 of the 60 participants were evaluated by a second rater. Cohen’s Kappa was measured for each item of the DDS part 2 and presented in Table 3. Taken together, the interrater reliability is high.

Table 3: interrater reliability

| Item of DDS part 2                  | N  | Cohen’s Kappa | Agreement            |
|-------------------------------------|----|---------------|----------------------|
| Item 8: orienting                   | 20 | K = 1.00      | Strong agreement     |
| Item 9: reception                   | 19 | K = 1.00      | Strong agreement     |
| Item 10: containment                | 20 | K = 1.00      | Strong agreement     |
| Item 11: oral transport             | 20 | K = .78       | Strong agreement     |
| Item 12: chewing                    | 20 | K = .80       | Strong agreement     |
| Item 13: oral-pharyngeal swallow    | 20 | K = .57       | Reasonable agreement |
| Item 14: post swallow               | 20 | K = 1.00      | Strong agreement     |
| Item 15: gastro-esophageal function | 20 | K = 1.00      | Strong agreement     |

#### RELATIONSHIP BETWEEN INDIVIDUAL TEST ITEMS DDS AND DMSS LEVEL

The strength of the relationship between the individual test items of the DDS and the severity of dysphagia on the DMSS was measured by means of a discriminant analysis ( $N=60$ ). A discriminant analysis identifies which items discriminate the best between the 5 levels of the DMSS and how well they predict them.<sup>18</sup>

First, the test of equality of group means was calculated. The smaller the Wilk’s lambda ( $\Lambda$ ), the more important the independent variable is for the discriminant function.<sup>18</sup> All items of the DDS appeared to be significant indicators of the DMSS level ( $p \leq .05$ ). Item 14 “post swallow” was revealed as best indicator ( $\Lambda = .189$ ,  $F = 58.902$ ,  $p < .001$ ). Also, item 3 “independence” ( $\Lambda = .248$ ,  $F = 41.745$ ,  $p < .001$ ), item 15 “gastro-esophageal function” ( $\Lambda = .312$ ,  $F = 30.365$ ,  $p < .001$ ), item 4 “adaptive utensils used” ( $\Lambda = .410$ ,  $F = 19.753$ ,  $p < .001$ ), item 7 “feeding techniques” ( $\Lambda = .271$ ,  $F = 36.979$ ,  $p < .001$ ), item 2 “diet” ( $\Lambda = .251$ ,  $F = 40.963$ ,  $p < .001$ ) and item 6 “postural control” ( $\Lambda = .653$ ,  $F = 7.316$ ,  $p < .001$ ) were revealed as good indicators. These items were respectively added to the 4 discriminant functions (stepwise statistics:  $p_{in} = .05$ ;  $p_{out} = .10$ ). The first discriminant function explained 75,7% of the variance, canonical  $R^2 = .92$ , the second 15,5%, canonical  $R^2 = .71$ , the third 6,7%, canonical  $R^2 = .44$ , and the fourth 2,2%, canonical  $R^2 = .04$ . Both in combination and when removing them one by one, these discriminant functions differentiated the DMSS level (statistics in Table 4).

Table 4: Wilks' Lambda of the Canonical Discriminant Function

| Test of Function(s) | Wilks' Lambda | Chi-square | df | Sig.   |
|---------------------|---------------|------------|----|--------|
| 1 through 4         | ,008          | 258,497    | 28 | < .001 |
| 2 through 4         | ,101          | 121,505    | 18 | < .001 |
| 3 through 4         | ,354          | 55,017     | 10 | < .001 |
| 4                   | ,739          | 16,013     | 4  | .003   |

The classification results revealed that 97.4% of the participants in DMSS level 1 were correctly classified, 90.9% of the participants in level 2, 100% of the participants in level 3, 85.7% of the participants in level 4 and 100% of the participants in level 5.

#### DISCRIMINANT EVIDENCE

On average, participants with known feeding disorders received significantly higher grades ( $M = 21.55$ ,  $SE = 10.22$ ) on the total DDS than participants without feeding disorders ( $M = 2.75$ ,  $SE = 3.18$ ),  $t(22.637) = -7.855$ ,  $p < .01$ ,  $r = .86$ . Each individual item of the second part 'feeding and swallowing competency' of the DDS distinguishes participants known and not known with dysphagia. Statistics are listed in Table 5, with parallel the results of previous studies.

Table 5: t-test of each item of DDS part 2 'eating and swallowing competency'

|                  | Task component (items on the DDS part 2)  | Study population                     |   | Literature: potential problems during swallowing in children with CP  |
|------------------|---|--------------------------------------|---|---|
|                  |   | known with feeding disorders (N =20) | Not known with feeding disorders (N=20) |   |
| Oral preparation | <u>Orienting:</u><br>alerting to food/nipple, moving toward food, mouth opening             | M = .85<br>SE = .221                 | M = .15<br>SE = .150                    | oral motor dysfunction:<br><ul style="list-style-type: none"> <li>- Reduced lip closure and drooling<sup>5,7,12</sup></li> <li>- Reduced <i>tongue mobility and coordination</i><sup>7,20</sup></li> <li>- Excessive tongue thrusting<sup>7,20</sup></li> <li>- Abnormal oral reflexes<sup>5,7</sup></li> <li>- Insufficient oral intake<sup>6,7</sup></li> </ul> |
|                  | <u>Reception:</u><br>Timing, strips spoon, bites off, sips from cup, appropriate bolus size | M = 2.05<br>SE = .276                | M = .25<br>SE = .099                    |   |
|                  | <u>Containment:</u><br>No dribbling or ejecting food or liquid                              | M = 1.45<br>SE = .294                | M = .15<br>SE = .109                    |   |
|                  |   | $t(38) = -2.621$<br>$p < .05$        |   |   |
|                  |   | $t(38) = -6.136$<br>$p < .05$        |   |   |
|                  |   | $t(38) = -4.138$<br>$p < .05$        |   |   |

|                         |   |                                    |                      |   |
|-------------------------|---|------------------------------------|----------------------|---|
|                         | <u>Chewing:</u><br>chew adequate for bolus, no special placement required             | M = 1.00<br>SE = .000              | M = .15<br>SE = .082 |   |
|                         |   | $t(38) = -10.376$<br>$p < .05$     |                      |   |
|                         | <u>Oral transport:</u><br>No residual in mouth after swallow, efficient bolus transit | M = 2.65<br>SE = .150              | M = .30<br>SE = .128 | <ul style="list-style-type: none"> <li>- Prolongation of oral transit time<sup>6,20</sup></li> <li>- Fragmentation of the bolus over the dorsum of the tongue<sup>6,20</sup></li> <li>- Residue in the sulci<sup>6,20</sup></li> <li>- Pharyngeal pooling prior to swallow in vallecula and/or pyriform sinuses<sup>20</sup></li> <li>- Abnormal bolus propulsion:               <ul style="list-style-type: none"> <li>- Excessive tongue thrusting<sup>7,20</sup></li> <li>- Reduced tongue mobility and coordination<sup>7,20</sup></li> </ul> </li> </ul> |
| <b>Oral initiation</b>  |   | $t(37.059) = -11.928$<br>$p < .05$ |                      |   |
|                         | <u>Oral-pharyngeal swallow:</u>   | M = 2.55<br>SE = .185              | M = .35<br>SE = .182 | Delayed initiation of the swallowing movement <sup>20</sup>   |
|                         | Prompt, sequential liquid swallow, no gagging or multiple swallows                    | $t(38) = -8.493$<br>$p < .05$      |                      | <ul style="list-style-type: none"> <li>- Pharyngeal pooling prior to swallow in vallecula and/or pyriform sinuses<sup>20</sup></li> <li>- Reduced pharyngeal peristalsis<sup>20</sup></li> <li>- Residue after swallowing<sup>20</sup></li> <li>- (silent) aspiration before, during or after swallowing<sup>20, 21</sup></li> <li>- Respiratory problems<sup>20,21</sup></li> </ul>  |
|                         | <u>Post swallow:</u><br>Absent coughing, wet breath sounds, wet voice                 | M = 1.55<br>SE = .336              | M = .00<br>SE = .000 |   |
| <b>Pharyngeal phase</b> |   | $t(38) = -4.610$<br>$p < .05$      |                      |   |
|                         | <u>Gastro-esophageal function:</u><br>Absent vomiting or rumination                   | M = .75<br>SE = .298               | M = .00<br>SE = .000 | Beyond the scope of this project  |
| <b>Esophageal phase</b> |   | $t(38) = -2.517$<br>$p < .05$      |                      |   |

## CONVERGENT EVIDENCE

To investigate the correlation between the sub-scores from part 1 and part 2 (N=60), Spearman's correlation coefficient was calculated. The results ( $R = .65$  with  $p < .01$ ) indicated that the correlation between part 1 and part 2 of the DDS is high.

## FACTORS RELATED TO THE SEVERITY OF DYSPHAGIA IN CHILDREN WITH CP

Associations between child characteristics and the score on the DDS are described in Table 6. IQ was significantly associated with the score on the DDS. However, the age of the child was not. Participants with a better functionality level, scored significantly better on the DDS. For the factor ‘type of CP’, no comparison was made with atactic CP, as only 1 subject presented with this type of CP. Significant differences were detected in the scores in patients with dyskinesia versus patients with spasticity and patients with unclassified CP. However, no difference was detected between the latter two.

Table 6: Associations between child characteristics and result on the DDS

| Child variable             |                         | Statistics influence of child variable on DDS score <sup>a</sup> |
|----------------------------|-------------------------|--|
| 1. Age in years, mean (SD) | 9.68 years (2.71 years) | Pearson's $R = .032, p \geq .05$                                 |
| 2. IQ, mean (SD)           | 67.98 (21.52)           | Pearson's $R = -.271, p = .049$                                  |
| 3. GMFCS, <i>n</i>         |                         |  |
| Level 1-2-3                | 40                      | $Z = -4.355, p = .000$   |
| vs Level 4-5               | 20                      |  |
| 4. Type of CP, <i>n</i>    |                         |  |
| Spasticity                 | 46                      | $Z = -4.049, p = .000$   |
| vs Dyskinesia              | 8                       |  |
| Spasticity                 | 46                      | $Z = -1.634, p \geq .05$   |
| vs other                   | 7                       |  |
| Dyskinesia                 | 8                       | $Z = -2.137, p = .033$   |
| vs other                   | 7                       |  |

<sup>a</sup> Pearson's  $R$  for interval/ratio variables, Z-score based on Mann-Whitney  $U$ -test categorical variables

## DISCUSSION

Our study population consisted of children from 3 to 15 years of age, diagnosed with CP in the Dominiek Savio Institute, and is comparable to the CP population described in the literature. Bax et al.<sup>3</sup> presented a European Cerebral Palsy Study including 61.9% male and 38.1 % female persons. In the current study, the number of males is slightly higher (78.3%) and the number of females lower (21.7%). In addition, the distribution of CP type (75.0% spasticity, 13.3% dyskinesia and 1.7% ataxia) corresponds with numbers from the European Cerebral Palsy Study (spasticity 79.2%, dyskinesia 14.4% and ataxia 3.9%). In previous studies about dysphagia and CP, the proportion of participants with severe hypertonic or hypotonic generalized CP were mostly included.<sup>7,10</sup> The present study opted for a heterogeneous group, consisting of children with different types of CP and different degrees

of severity. As a result, the percentage of children, classified with a feeding disorder, is much lower (33.33%) in the present study, and does not correspond with findings by Calis et al.<sup>10</sup> and Reilly et al.,<sup>7</sup> respectively 99% and 91%. However, Waterman et al.<sup>5</sup> diagnosed 27% of the children with CP in special school with swallowing problems, using radiographic or clinical evidence of dysphagia, which was similar to our population.

The first aim of the study was whether the DDS and DMSS<sup>15</sup> are useful for the assessment of swallowing disorders in children with CP. It is evidenced in earlier surveys that the DDS can also be used for other populations than the original group of people with mental disabilities. This finding was never assessed in children CP, a population with distinct physical disabilities.<sup>15</sup> Our results support a positive internal consistency, construct validity, and the interrater reliability of the DDS as well as the relation between DDS and DMSS. Since there are no other validated dysphagia screening tools specifically targeting children with CP, the current results cannot be compared with previous validity and reliability test results used in this population.

The internal consistency reliability for both part one and part two and for the total DDS was high, as in the original group of institutionalized adults with mental disabilities.<sup>15</sup> The dysphagia-related items in part 1 and the mealtime-observation items in part 2 measure the same skill, namely the presence of dysphagia in children with CP. Combined (total DDS), they have a higher Cronbach's alpha than each separate part. Both parts are important and using them both leads to the most complete screening.

The interrater reliability was high in all items, except in item 13 'oral pharyngeal swallow.' The score on the DDS is not dependent on the rater. This means that any certified person could assess a child, and that this leads to reliable results. An explanation for the strong agreement between the two raters in item 8 to 12 can be found in the clear description of how to rate the items and to the objective observation of movements in and around the mouth. Item 13, the oral pharyngeal swallow, had only a reasonable agreement between raters. The rating is based on 'indications' of dysphagia during the pharyngeal phase, such as multiple swallows, than on objective observation of movements, which could explain this discrepancy. There is an absolute agreement in item 14 and 15, because the information is derived from the same anamnestic interview with a caregiver. However, these items are not based on objective observation of movements either. The above result suggests that problems during the oral (preparatory) phase could be objectively detected using the DDS. In order to diagnose problems in the pharyngeal phase, more objective methods such as high resolution

manometry and impedance measurements<sup>14</sup> are strongly recommended, as problems during this phase should have serious health effects<sup>21</sup> and the scoring of this item on the DDS is rather subjective and less reliable.

The strength of the relationship between individual test items (DDS) and severity (DMSS) was tested by discriminant analysis and was found to be adequate. Item 14, post-swallow, assesses the effectiveness of the pharyngeal phase during the observation, using visible signs such as coughing and wet breath sounds. This item was the best predictor of DMSS level. As aspiration is the most direct life-threatening consequence of dysphagia, this finding seems logical. The criteria of level 4 and 5 are persisting secondary problems in the airways, which may be associated with aspiration.<sup>15</sup> Explicit attention to these signs during and after the meal is important. Also the items 'diet' and 'feeding techniques' are added to the discriminant functions, which is not unusual, as these are also criteria to determine the DMSS level. The nutritional status is a criterion as well, but it is not added to the discriminant functions. An explanation could be found in Calis et al.<sup>10</sup> who stated that in case the nutritional status becomes critical, tube feeding (supplementary or total) is often chosen, resulting in a good nutritional status for patients in level 5. Although dependence on intake of food does not directly lead to a disorder on the DMSS, this item was added to the discriminant function. Similar as to the use of adapted cutlery and postural control, dependence on intake of food is related to a higher GMFCS level and also to a higher score on the DMSS. The task description, used in the DDS and linked to the DMSS, combines both body function and activity level of the ICF-model.<sup>22</sup>

This study indicates that the DDS is able to differentiate between children with and without swallowing disorders. On average, participants with known feeding disorders presented with significantly higher scores on the DDS. Moreover, the effect size (.86) is large, which means that the magnitude of the effect is strong.<sup>17</sup> The standard error of the mean (SEM) of the total DDS score is low in participants without feeding problems, which suggests that the variation is minimal. However, the SEM is much higher in participants with feeding problems, which suggest that the scatter is wider and the variation is bigger. This finding is logical, because levels of severity are related to this score. In clinical use, this means that a score difference of 10.22 is necessary to be sure that a 'true' improvement took place, rather than the difference being due to measurement errors.<sup>17</sup> If the SEM were calculated for groups, based on DMSS level, we expect that the SEM would be lower, as there would be more consistency within

groups. A wider scatter in children with feeding problems, means that there are opportunities for differentiation according to severity, which benefits the treatment.

Additionally, to item-averages, there are significant differences between children with and without feeding problems. Items 8 to 12 evaluate the various components of the oral phase of swallowing. Earlier findings of problems in this phase could be linked to the different items of the DDS. For instance, reduced lip closure<sup>5,7,12</sup> could lead to problems with reception (item 9), containment (item 10) and chewing (item 12). Problems with chewing could be caused by a reduced tongue mobility and coordination<sup>7,20</sup>. Since the DDS evaluates each task component, the severity of restriction is indirectly taken into account. A slight reduced lip closure could lead to only a deviant score on the item 'reception', while a more prominent reduced lip closure could also lead to problems with containment (item 10). Unfortunately, little attention is given in the DDS to the aspects of oral sensitivity and oral reflexes. Nevertheless, abnormal oral reflexes and abnormal oral sensibility are present in the population of children with CP.<sup>5,7</sup>

The sub-scores from part 1 and part 2 correlate ( $R = .646$ ). Part 1 discusses the *related* items, and part 2 the *actual* feeding and swallowing competency so both parts support each other, but are not exactly the same.

In conclusion to the first aim of the study, the DDS and DMSS are useful screening instruments. Though, mealtime observation screening cannot guarantee the absence of silent aspiration. As reported in Calis et al.<sup>10</sup>, the DDS supports referral for objective clinical and instrumental evaluation and treatment in children at risk for aspiration. Indeed, detailed information on the exact mechanisms during the pharyngeal phase as well as specific time measurements cannot be established by mealtime observation, which is reflected in the low interrater reliability of the item 'oral pharyngeal swallow' in comparison with other items. Nevertheless, the DDS and DMSS are useful and unique, because they describe the essential aspects to be considered during a mealtime observation screening.

The second aim of the study focused on the influencing factors on dysphagia. A sample representative of the population of children with CP led to uneven distribution among the factors. Most of the children had spasticity, and a minority dyskinetic, atactic or unclassified CP.

The type of CP influences the score on the DDS. Studies in which there is a clear distinction between the types of CP, are rare. Results usually pertain to the entire group of children with CP only.<sup>5,7,20</sup> As suspected from the parent's questionnaires by Sullivan et al.<sup>8</sup>, the current

study reports significant differences between the types of CP. Patients with dyskinetic CP are often identified with dysphagia. This finding is new and further investigation on the origin of dysphagia in this group is recommended. Children with dyskinetic CP have only limited movement control, which is also pernicious to the feeding situation. Ramritu et al.<sup>23</sup> indicated the presence of dyskinesia as a risk factor, that is associated with dysphagia, because of reduced ability to chew, to manipulate the bolus in the mouth and to swallow. Also, the use of muscle relaxants administered to children with spasticity may affect ability to swallow.<sup>23</sup>

The cognitive function, here expressed as IQ, was significantly associated with the score on the DDS. As previously suggested, the readiness to make the transition to a next milestone in the acquisition of eating skills relates more closely to developmental age than chronological age.<sup>24</sup> Our data confirm that dysphagia is related to lower mental abilities.<sup>5,13</sup> However, there is a variety of physical disorders in children with CP, the mental capacities of the CP-child have a clear impact on the severity of feeding disorders.

The GMFCS level significantly influences the score on the DDS. The clinical reality is that wheelchair-bound children with CP are at significantly higher risk of suffering from dysphagia than others. Since the presence of dysphagia is mainly examined in children with severe CP, it was suspected that these children face greater risk of suffering from dysphagia.<sup>7,10,20</sup> Calis et al.<sup>10</sup> exposed differences between GMFCS level 4 and 5 as well. Level 5 was significantly associated with a more severe level of dysphagia. This supposition is confirmed. Although it certainly should be mentioned that dysphagia was also present in children with GMFCS level 1, 2 or 3. So the functional impairment does not completely explain the origin of dysphagia.

Finally, no effect of age was detected. There were no higher or lower grades on the DDS with increasing age. As the youngest participant was 3 years old, no conclusions should be made about the development of feeding skills in children with CP. At the age of 3, a lot of feeding skills are acquired in normally developing children, but need to be refined.<sup>25</sup> Indeed, Reilly et al.<sup>7</sup> found that the majority of children with CP between 12 and 72 months have clinically significant oral motor dysfunction. A larger study with younger children with CP might give clarification about the development of feeding skills.

In conclusion, dysphagia in children with CP is complex and has shown to be influenced by several factors such as (1) GMFCS level, (2) type of CP and (3) IQ. Further research with a



larger sample and objective and detailed information on the exact swallowing mechanisms should clarify the origin of dysphagia in these children with CP.

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APPENDIX 1: ITEMS OF THE DDS (SHEPPARD, 2002)

| DDS part 1: related factors                   |  |
|---|--|
| <b>1. Body mass index</b>                     | $\text{BMI} = \frac{\text{weight (kg)}}{\text{height}^2 (\text{m}^2)}$ <p>a ratio of weight and height ( ). The calculation is useful for children who are older than 2 years. In our study, the BMI is interpreted by means of special tables for typically developing children.<sup>26</sup></p> |
| <b>2. Diet</b>                                | the feeding consistencies during daily mealtimes   |
| <b>3. Independence</b>                        | the extent of assistance needed by eating the meal   |
| <b>4. Adaptive utensils used</b>              | the use of special eating utensils such as special cups, spoons and plates   |
| <b>5. Positioning</b>                         | the special tools used to create the best possible position during the meal  |
| <b>6. Postural control</b>                    | the ability to sit upright   |
| <b>7. Feeding techniques</b>                  | eating techniques that are used by the caregiver for dependent eaters (e.g. mouth control) and eating techniques used by independent eaters to compensate for eating and swallowing problems   |
| DDS part 2: feeding and swallowing competency |  |
| <b>8. Orienting</b>                           | the will to accept the impending food bolus  |
| <b>9. Reception</b>                           | removal off the bolus of the cutlery, regulating the size of the bolus and eating rate   |
| <b>10. Containment</b>                        | to keep the food in the mouth to be ready to swallow   |
| <b>11. Oral Transport</b>                     | to put food in the correct position to swallow   |
| <b>12. Chewing</b>                            | chewing of food that is so coarse that chewing is necessary  |
| <b>13. Oral-pharyngeal swallow</b>            | the efficiency of the swallow from the beginning to the end, the ease of swallowing, the rapid consecutive swallowing and finally the presence of choking and multiple swallowing movement   |
| <b>14. Post swallow</b>                       | the effectiveness of the pharyngeal phase during the observation, one look for visible signs of pharyngeal stasis, and inadequate protection of the respiratory tract (coughing, changes in the frequency and the ability to breath and "wet" or gurgling breath sounds)                           |
| <b>15. Gastro-esophageal function</b>         | symptoms that may indicate an (increased risk of) impairment in the esophageal phase of swallowing   |

