Dysphagia limit in children with cerebral palsy aged 4 to 12 years

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AIM To assess the dysphagia limit in children with cerebral palsy (CP) according to Eating and Drinking Ability Classification System (EDACS) level, sex, and age compared to typically developing children.

METHOD Seventy-seven children with CP (54 males, 23 females; mean age 7y 6mo, SD 2y 2mo, age range 4–12y) were assessed with the Maximum Volume Water Swallow Test. Median dysphagia limit in the CP group was compared with data of typically developing children.

RESULTS The dysphagia limit of children with CP differed significantly (p<0.001) from typically developing children. The latter showed a threefold higher median dysphagia limit (22mL) compared to children with CP in EDACS level I (7mL). The higher the EDACS level, the lower the dysphagia limit in children with CP. EDACS level explained 55% of the variance in the dysphagia limit of the CP group.

INTERPRETATION Where children with CP in EDACS levels IV and V showed that their capacity met the level of their performance, children in EDACS level I had the ability to perform a maximum capacity task, but still had a threefold lower median dysphagia limit than typically developing children. Establishment of the dysphagia limit should be part of general swallowing assessment in children with CP.

Cerebral palsy (CP) is a disorder of movement and/or posture caused by a non-progressive brain lesion that persists through the lifespan and has a prevalence of more than 2 per 1000 live births.1 CP comprises a heterogeneous group of impairments that may also affect the oral motor functions needed for eating, drinking, swallowing, and articulation.2 Feeding and swallowing problems (dysphagia) can influence the safety and efficiency of feeding. This may lead to inadequate nutritional status due to prolonged feeding times,3 stress during mealtimes for the caregiver and/or the child, and impaired growth.5 Estimates of the prevalence of dysphagia in children with CP vary widely due to the different definitions and tools used, small groups of children, variability in severity of CP, and motor type. The prevalence ranges from 21% in a group of 1357 children with CP6 to 99% in a group of 166 severely affected children with intellectual impairment.7 One of the oral skills for safe swallowing is the ability to divide a liquid bolus into two or more segments in the oral cavity when it becomes too large to swallow at once; this is called piecemeal deglutition.8 When piecemeal deglutition exists in adults at or below 20mL of water, it is considered pathological and may be a sign of dysphagia.9 Even though piecemeal deglutition may be safer for swallowing liquids (people’s usual activity),9,10 portions that are too small may alter the speed of drinking and, as such, influence the efficiency of drinking.3 The upper limit of the amount of liquid that can be swallowed at once is called the dysphagia limit.11 The dysphagia limit is seen as the ‘capacity in swallowing’ and is defined as what a person can do in a standardized, controlled environment.10 Unlike in adults, little is known about the dysphagia limit in children.9 Typical values of the dysphagia limit were collected in 352 typically developing children aged 4 to 19 years, in the Netherlands, with the newly established Maximum Volume Water Swallow Test (personal communication, LEH, KvH, SG, ML, 2017). The authors showed a significant difference between two age groups (4–12y and 12–18y) and between males and females. Ozdemirkiran et al.12 showed in a small study that children with CP aged 6 to 17 years (n=12) had a significantly lower dysphagia limit than typically developing children (n=28).
The daily feeding and swallowing skills in children with CP can be classified by the validated Eating and Drinking Ability Classification System (EDACS). EDACS identifies five levels in children with CP aged 3 to 21 years. Levels range from level I: ‘eats and drinks safely and efficiently’ to level V: ‘unable to eat and drink safely’. As the EDACS describes performance rather than capacity, we were interested in how the dysphagia limit was related to EDACS level. Typically, clinicians expect that when a child has a high capacity in a certain domain of functioning it will be related to a high ease of activity performance in that domain. However, the relationship between capacity and performance has not been explored systematically in eating, drinking, and swallowing abilities in CP. One of the purposes of this study, therefore, was to explore the relationship between capacity scores of the dysphagia limit in children with CP and the level of eating and drinking performance described by the EDACS. It was hypothesized that (1) a child in EDACS level I, who is classified as eating and drinking like a typically developing peer, should have a comparable capacity of swallowing, and (2) a child in a higher EDACS level would have less ability in oral control, and thus a lower dysphagia limit. In addition, it was assumed that the EDACS level is an important determinant in explaining the variance in the dysphagia limit in children with CP.

Therefore, the aims of this study were threefold: (1) to give an overview of the dysphagia limit in children with CP aged 4 to 12 years in different EDACS levels compared to typically developing children, (2) to determine whether the dysphagia limit in children with CP in EDACS level I (eats and drinks safely) differs from typically developing children, and (3) to examine the proportion of variance in dysphagia limit of children with CP, explained by EDACS level, sex, and age.

**METHOD**

**Participants**

Included in this study were children with CP aged 4 to 12 years with CP subtypes as classified by the Surveillance of CP in Europe guidelines. Children were recruited from two tertiary hospitals (UMC Groningen and Radboud UMC Nijmegen, the Netherlands), and several special needs schools and day care centres in the northern region of the Netherlands. Gross Motor Function Classification System (GMFCS) and EDACS levels were already established by trained physiotherapists and speech and language therapists at the day care centre or the child’s school. Children were excluded if (1) they were using psychiatric medication and/or sedatives, and (2) had an additional illness (i.e. tonsillitis) and/or anatomical malformations, as these might influence swallowing. Values of the dysphagia limit in typically developing children aged 4 to 12 years were retrieved from an earlier study (personal communication, LEH, KvH, SG, ML, 2017). The Research Ethics Committee of the Radboud University Nijmegen Medical Centre approved the study protocol (2017-3995). Written informed consent was obtained from the children’s parents. All data were transformed and stored anonymously.

**Assessment of the dysphagia limit**

To determine the dysphagia limit each child with CP performed the Maximum Volume Water Swallow Test. This test was used to establish the dysphagia limit in typically developing children (personal communication, LEH, KvH, SG, ML, 2017). In the test protocol, children drank tap water at room temperature by cup or by syringe while seated in a quiet room in their own (wheel)chair. The dysphagia limit in typically developing children did not seem to differ between swallowing an amount of water by cup or syringe. In the group of children with CP, assessed by the same test, the water was offered by cup or syringe, depending on the ability of the child. A verbal instruction to swallow all the water at once was given by the assessor. To determine the number of swallows needed, cervical auscultation with a neonatal stethoscope (3M™ Littmann® Classic II Infant; 3M, USA) was used. Cervical auscultation is a clinical method used to evaluate the pharyngeal phase of swallowing by listening to the sounds of swallowing and swallowing-related respiration. A stethoscope is placed on the lateral side of the neck in the region of the larynx. Starting with a very small amount of water (0.5mL), close to the amount of saliva generally swallowed, was considered to be safe. From that amount, the volume was increased to 1mL, 3mL, 5mL, 10mL, 15mL, and 20mL or more. When it became audible that the child swallowed twice on the amount of water offered, a 1mL smaller amount was given. If the child swallowed twice again, a stepwise reduction of 1mL was repeated until the child swallowed once. This final amount was given once more to check the maximum amount swallowed at once before it was established as the dysphagia limit. The authors (FVS, KvH, LEH), who were working as speech and language therapists in the tertiary hospitals, performed the assessments. The first author assessed the children at the special needs schools and the day care centres.

**Statistics**

Descriptive statistics were used (median, interquartile range, and minimum–maximum [mL]). Assumptions were checked with inferential statistics. The differences in dysphagia limit between CP versus typically developing children and between children with CP in EDACS level I versus typically developing children were tested with the Mann–Whitney U test. With multivariable linear regression the proportion of the variance in dysphagia limit in children with CP explained by the three variables, EDACS...
level, age, and sex ($R^2$), was established. When assumptions were violated, transformations were used. To attain a robust model, variables were backward selected and excluded from the model when the level of significance was $p>0.05$. Univariable linear regressions were used to assess how the individual variables, EDACS level, age, and sex, explained the variance ($R^2$) in dysphagia limit in children with CP. There were no missing data. All data analyses were performed using SPSS statistics, version 25.0 (IBM Corp., Armonk, NY, USA).

RESULTS

Seventy-seven children with CP (54 males, 23 females; mean age [SD] 7y 6mo [2y 2mo]) participated in this study. Children attended regular schools (8%), special needs schools (73%), and day care centres (19%). Table 1 shows the characteristics of both the typically developing children from a previous study (personal communication, LEH, KvH, SG, ML, 2017) and the children with CP from the current study. In both groups, the data of the dysphagia limit was non-normally distributed. The dysphagia limit was significantly higher for typically developing children (median 22.0mL) than for children with CP (median 7.0mL); ($U$ 1279.5, $p<0.001$, $r=0.65$) and differed significantly between different EDACS level groups; ($\chi^2[4]$=33.256, $p<0.001$). Data of dysphagia limit in the total group with CP, in children with CP in EDACS level I, and typically developing children are presented in Table 2. The dysphagia limit of typically developing children (median 22.0mL) differed significantly from the subgroup of children with CP in EDACS level I (median 7.0mL); ($U$ 855.0, $p<0.001$, $r=0.37$), see Table 2. Data of the dysphagia limit of children with CP classified by EDACS level are shown in Table 3.

Variance in dysphagia limits explained by EDACS level, age, and sex

A natural log transformation on the dysphagia limit was performed because assumptions of multivariable linear regression were not met. A significant regression equation was found ($F[3,73]=33.216$, $p<0.001$), with 58% of the variance in dysphagia limit ($R^2=0.58$) explained by EDACS level, age, and sex together, regression equation was found ($F[3,73]=33.216$, $p<0.001$). The model with only significant variables (EDACS level and age) explained 57% of the variance in dysphagia limit. Sex turned out to be a non-significant variable in this model. No difference in dysphagia limit was found between males and females in this group. In the univariable models, the EDACS level was significant and explained 55% of the variance in dysphagia limit in children with CP.

DISCUSSION

Our study supported the relationship between swallowing capacity and eating and drinking performance in children with CP using the dysphagia limit and EDACS levels. It showed that children with CP had a significantly lower dysphagia limit than typically developing children in the same age range. As the dysphagia limit was highly related to EDACS level, we found when EDACS level increased, the severity in eating and drinking problems also increased, and a smaller median and spreading of the dysphagia limit was established. A better functional performance in eating and drinking (EDACS level I or II) showed a better capacity for controlling liquids (higher dysphagia limit). Although CP is more prevalent in males,\(^{1,19}\) no difference was found in the capacity task of swallowing thin liquids

| Characteristics          | Children with CP, $n=77$ | Typically developing children, $n=218$ |
|--------------------------|--------------------------|---------------------------------------|
| Sex                      |                          |                                       |
| Male                     | 54 (70)                  | 104 (48)                              |
| Female                   | 23 (30)                  | 114 (52)                              |
| Age                      |                          |                                       |
| Mean age (SD), y:mo      |                          |                                       |
| 4y                       | 7.6 (2.2)                | 8.2 (2.2)                             |
| 5y                       | 8 (10)                   | 29 (13)                               |
| 6y                       | 13 (17)                  | 29 (13)                               |
| 7y                       | 9 (12)                   | 28 (13)                               |
| 8y                       | 8 (10)                   | 31 (14)                               |
| 9y                       | 15 (20)                  | 36 (17)                               |
| 10y                      | 11 (14)                  | 33 (15)                               |
| 11y                      | 2 (3)                    | 29 (13)                               |
| School                   |                          |                                       |
| Regular school           | 6 (8)                    | 218 (100)                             |
| Special needs school     | 56 (73)                  |                                       |
| Day care centre          | 15 (19)                  |                                       |
| Type of CP\(^{a}\)       |                          |                                       |
| Spastic unilateral       | 17 (22)                  | NA                                    |
| Spastic bilateral LE\(\rightarrow\) UE | 12 (16) |                                       |
| Dyskinetic               | 10 (13)                  |                                       |
| Ataxia                   | 5 (6)                    |                                       |
| Worster-Drought          | 1 (1)                    |                                       |
| GMFCS level              |                          |                                       |
| I                        | 20 (26)                  | NA                                    |
| II                       | 10 (13)                  |                                       |
| III                      | 11 (14)                  |                                       |
| IV                       | 21 (27)                  |                                       |
| V                        | 15 (20)                  |                                       |
| EDACS level              |                          |                                       |
| I                        | 26 (34)                  | NA                                    |
| II                       | 20 (26)                  |                                       |
| III                      | 12 (16)                  |                                       |
| IV                       | 11 (14)                  |                                       |
| V                        | 8 (10)                   |                                       |
| EDACS levels of assistance|                        |                                       |
| Independent              | 41 (53)                  | 218 (100)                             |
| Requires assistance      | 15 (20)                  |                                       |
| Totally dependent        | 21 (27)                  |                                       |
| Maximum volume water swallow test |      |                                       |
| Offered by cup           | 43 (56)                  | 218 (100)                             |
| Offered by syringe       | 33 (43)                  |                                       |
| Offered by straw         | 1 (1)                    |                                       |

Data are $n$ (%) unless otherwise stated. *Cerebral palsy (CP) subtypes classified according to the Surveillance of Cerebral Palsy in Europe.\(^{15}\) Gross Motor Function Classification System (GMFCS): level I, walks without limitations to level V, transported in a manual wheelchair. Eating and Drinking Ability Classification System (EDACS): level I, eats and drinks safely and efficiently to level V, unable to eat or drink safely – tube feeding may be considered to provide nutrition. NA, not applicable; LE, lower extremities; UE, upper extremities, where lower extremities are more spastic than the upper extremities.
Dysphagia limits in the total group with CP, in children with CP in EDACS level I, and typically developing children

| Table 2 | Dysphagia limits in the total group with CP, in children with CP in EDACS level I, and typically developing children |
|-----------------|-------------------------------------------------|-----------------|-----------------|
|                | Children with CP, n=77 | Children with CP in EDACS level I, n=26 | Typically developing children, n=218 |
| Dysphagia limit median (IQR), mL | 3.0 (1.8–7.0) | 7.0 (4.0–14.0) | 22.0 (16.5–30.0) |
| Dysphagia limit min-max, mL | 0.3–41.0 | 2.0–41.0 | 3.0–70.0 |

*Mann–Whitney U test. CP, cerebral palsy; EDACS, Eating and Drinking Ability Classification System; IQR, interquartile range; min-max, minimum–maximum.

Dysphagia limits per EDACS level in the total group of children with cerebral palsy

| Table 3 | Dysphagia limits per EDACS level in the total group of children with cerebral palsy |
|-----------------|-------------------------------------------------|-----------------|-----------------|
|                | EDACS level | n | Median, mL | IQR, mL | Min-max, mL |
|                | I           | 26 | 7.0 | 4.0–14.0 | 2.0–41.0 |
|                | II          | 20 | 3.0 | 2.0–6.8 | 1.0–38.0 |
|                | III         | 12 | 2.5 | 1.3–3.6 | 0.5–15.0 |
|                | IV          | 11 | 2.0 | 1.0–2.0 | 0.5–2.0 |
|                | V           | 8  | 0.5 | 0.5–0.5 | 0.3–1.0 |

*Kruskal–Wallis H test p=0.001 for all Eating and Drinking Ability Classification System (EDACS) levels. EDACS: level I, eats and drinks safely and efficiently to level V, unable to eat or drink safely – tube feeding may be considered to provide nutrition. IQR, interquartile range.

between males and females in this study. The results are in accordance with the outcome reported by Ozdemirkiran et al. and show the importance of knowledge of sip volume. Children with CP have problems with swallowing liquids related to a timing deficit with delayed pharyngeal swallow initiation. In addition, children who are completely dependent on the caregiver while being fed are at greater risk of aspiration because of oversized boluses that may be given. Moreover, some children with CP have trouble coughing, making them prone to aspirations and respiratory tract infections. It is reported that children with aspiration pneumonia have higher rates of mortality, are more likely to require intensive care unit level of care, and have higher 30-day readmission rates. Given the above-mentioned problems, knowledge of the child’s dysphagia limit is essential to support safe swallowing.

The relation between dysphagia limit (capacity) and EDACS level (performance) is comparable with gross motor studies. It was found that severity in daily life mobility (performance), classified by the GMFCS, is closely related to gross motor capacity in children with CP. However, to our knowledge, the relationship between capacity and performance has not been studied in pediatric swallowing studies in children with CP. Children in EDACS level I, classified as ‘eats and drinks safely and efficiently’, are generally regarded as comparable with their typically developing peers. Our study found that children in EDACS level I still showed a three-fold smaller median (7.0mL) dysphagia limit compared to their typically developing peers (22.0mL). Although their performance showed a safe and efficient eating and drinking ability, their capacity is limited compared to typically developing children. A similar difference between children with CP in GMFCS levels I and II and typically developing children was also found by Verschuren and Takken. Results of aerobic capacity of children with CP were significantly below the values observed in typically developing children. We hypothesized that as motor functioning is influenced by motor capacity in daily life mobility, in CP, daily eating and drinking performance is influenced by the swallowing capacity (dysphagia limit). The less capacity in swallowing the more challenging the efficiency and safety in daily life eating and drinking will be.

EDACS level explained more than half of the variance in dysphagia limit in the CP group. This is in concordance with gross motor studies, where motor capacity alone is not the sole factor contributing to daily life mobility. The actual skills of eating and drinking are not only influenced by oral motor performance, but also by personal and social factors, such as parents’ judgements about their children’s eating and drinking. Disturbances of sensation, perception, cognition, communication, and behaviour, as well as epilepsy and secondary musculoskeletal problems, are included in the definition of CP and should be taken into account. Children with more severe eating and drinking difficulties (EDACS levels IV and V), do not have a well-integrated sensorimotor system for oral pharyngeal swallowing, a prerequisite for controlling a liquid bolus. Their swallowing function is mostly disturbed at all stages (i.e. oral, pharyngeal, and oesophageal stage). Children in EDACS levels IV and V showed that their capacity for controlling and swallowing a liquid bolus at one time (oral stage) was very limited or not possible. They were not able to adapt to control thin liquids to improve their capacity of swallowing. Their maximum capacity is comparable with their usual saliva swallowing or swallowing small amounts of liquids. Therefore, their capacity met the level of their performance.

Dividing the bolus into multiple swallows may alter the speed of eating and drinking. Although the relation between dysphagia limit and mealtime duration is currently unclear, the dysphagia limit can be used to explain to parents or caregivers why drinking is so challenging. The findings may also have implications for therapy aimed at improving the capacity and performance (i.e. safe and efficient eating and drinking) in daily life situations.

Several strengths and limitations of our study should be noted. A strength is the number of participating children...
with CP, with a representative distribution of males and females and the major clinical characteristics in line with other data.27 Children with severe CP in EDACS levels IV or V, or classified in GMFCS levels IV and V are less represented in the group with CP and also in our study.15,27 This small group in our study may be a limitation, but they showed less variation in dysphagia limits, which may be interpreted as representative for their maximum capacity of swallowing thin liquids. It should be noted that the assessors were well experienced in swallowing assessment. A limitation for clinical practice might be that less experienced assessors might underestimate the dysphagia limit in children with CP.17 Another limitation is that some children (n=6) did not completely understand the instruction ‘swallow at once’ as given in the protocol. However, they were able to control an increasing amount of water and to swallow. Although it was unclear if these children presented their maximum capacity, we decided to include them in this study because it was the maximum achievable in these children.

In typically developing children older than 12 years, the dysphagia limit increases as they grow older (personal communication, LEH, KvH, SG, ML, 2017). Although we have gained some insight into the relationship between swallowing capacity and daily-life eating and drinking performance in children with CP, our study reveals nothing about the change in swallowing capacity when these children grow older. From a developmental perspective, it is therefore of interest to investigate this relationship over time.

With the carefully developed protocol of the non-invasive Maximum Volume Water Swallow Test, establishing the dysphagia limit might be realizable, although the reliability of the test still requires further research. To support safety and efficiency in eating and drinking, establishing the dysphagia limit in children with CP should be a part of the eating and drinking assessment.

ACKNOWLEDGEMENTS
We gratefully acknowledge all the children and their parents for participating in this study. We also wish to thank the participating tertiary hospitals, rehabilitation centres, special needs schools, and their affiliated speech and language therapists who helped to recruit the children. The authors have stated that they had no interests which might be perceived as posing a conflict or bias.

DATA AVAILABILITY STATEMENT
This statement will be published alongside our manuscript.
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