Prevalence of drooling, swallowing, and feeding problems in cerebral palsy across the lifespan: a systematic review and meta-analyses

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PUBLICATION DATA
Accepted for publication 31st May 2019.
Published online 22nd July 2019.

AIM To determine the prevalence of drooling, swallowing, and feeding problems in persons with cerebral palsy (CP) across the lifespan.
METHOD A systematic review was conducted using five different databases (AMED, CINAHL, Embase, MEDLINE, and PubMed). The selection process was completed by two independent researchers and the methodological quality of included studies was assessed using the STROBE and AXIS guidelines. Meta-analyses were conducted to determine pooled prevalence estimates of drooling, swallowing, and feeding problems with stratified group analyses by type of assessment and Gross Motor Function Classification System level.
RESULTS A total of 42 studies were included. Substantial variations in selected outcome measures and variables were observed, and data on adults were limited. Pooled prevalence estimates determined by meta-analyses were as high as 44.0% (95% confidence interval [CI] 35.6–52.7) for drooling, 50.4% (95% CI 36.0–64.8) for swallowing problems, and 53.5% (95% CI 40.7–65.9) for feeding problems. Group analyses for type of assessments were non-significant; however, more severely impaired functioning in CP was associated with concomitant problems of increased drooling, swallowing, and feeding.
INTERPRETATION Drooling, swallowing, and feeding problems are very common in people with CP. Consequently, they experience increased risks of malnutrition and dehydration, aspiration pneumonia, and poor quality of life.

Cerebral palsy (CP) is a group of permanent, but not unchanging, disorders of movement, posture, and motor function.[1] CP is a clinical diagnosis based on neurological and motor symptoms, causing functional and activity limitations. The onset of this non-progressive neurodevelopmental condition occurs in early childhood and persists throughout the lifespan.[2] People with CP experience concomitant disturbances of sensation, perception, cognition, communication, and behaviour, and are also known to experience swallowing and feeding problems, particularly during childhood.[3]

The process of swallowing is highly complex and involves many muscles in the oral cavity, larynx, and oesophagus; more than 30 nerves and muscles are involved in volitional and reflexive activities during eating and swallowing.[4] During the process of eating, food must be masticated, formed into a bolus, and transported into the pharynx, primarily driven by the tongue.[4] Fluids require initial containment and positioning of the ingested fluid in the oral cavity before its subsequent aboral propulsion into the pharynx.[5] During this initial phase of swallowing, lip closure ensures bolus containment in the oral cavity, while cyclic tongue movements, coordinated with jaw movements, process solid foods. This oral component of swallowing is mostly voluntary and involves the lips, teeth, masticatory muscles, and the tongue. Next, the pharyngeal component of swallowing will be initiated by stimulation of the superior laryngeal nerve, a branch of the cranial vagus nerve. This involuntary stage of swallowing is more reflexive.[5,6]

Whereas swallowing refers to the transport of a bolus (food, liquid, saliva) from the oral cavity to the stomach, feeding mainly describes the process of breastfeeding or bottle feeding, transition to solid foods, and/or the process of setting up, arranging, and bringing food or liquid from a plate or cup to the mouth.[7] Feeding is not limited to the

 DOI: 10.1111/dmcn.14316

actual swallowing act, but also incorporates child–caregiver interaction (e.g. responsive complementary feeding, verbal encouragement, pressure to eat, and restrictive feeding practices by caregiver) and child behaviours (e.g. self-regulatory eating practices and self-feeding skills).[8–10] Swallowing problems (dysphagia) in CP may be characterized by poor tongue function having an impact on bolus transport, delayed swallow initiation with increased risk of unsafe swallowing or aspiration, reduced pharyngeal motility, and drooling due to reduced lip closure (sialorrhoea). Feeding problems present with prolonged feeding times or delayed progression of oral feeding skills and may lead to inadequate growth.[3] Both swallowing and feeding problems are associated with dehydration, malnutrition, aspiration pneumonia, and even death.[3,11] Persons with CP experience many restrictions in eating and drinking throughout adulthood, leading to lower self-esteem, and poor quality of mealtime experiences. Such restrictions have a negative impact on social interaction, and may lead to social isolation, depression, and poor quality of life.[12–14]

A recent review by Oskoui et al.[15] estimated the pooled overall prevalence of CP to be 2.11 per 1000 live births. With the exception of those with profound intellectual deficits, most people with CP survive into adulthood.[16] As the impact of swallowing and feeding problems can be far-reaching, particularly in paediatric populations with associated developmental challenges, early diagnosis is critical to put evidenced-based interventions in place.[7] Conversely, some adults with CP may experience gradual regressive adverse changes in their eating, drinking, and swallowing as early as 30 years of age.[12] Their eating capabilities may deteriorate, which are often associated with increased coughing and choking, weight loss, or more frequent periods of respiratory health problems. As such, regular assessment of swallowing and feeding are also important in older persons with CP to monitor compliance with nutritional recommendations, ongoing safety, optimal well-being, and to ensure swallowing and feeding strategies continue to be appropriate for changing oropharyngeal function and skills.[12]

Yet, despite all the challenges associated with drooling, swallowing, and feeding in people with CP, no prevalence review for the CP population has been published so far. However, determining the prevalence of a condition is essential to guide health policy and to ensure appropriate resource allocation. Several reviews have been published on the prevalence of drooling, swallowing, and feeding problems in specific populations: for example, the prevalence of drooling in Parkinson disease;[17] swallowing problems in stroke, Alzheimer disease, head injury, Parkinson disease, and multiple sclerosis;[18,19] and feeding problems in infants born very preterm and patients in intensive care units.[20,21] For the CP population, data on the prevalence of drooling, swallowing, and feeding problems can be retrieved from individual studies. Several factors need to be taken into consideration when calculating the prevalence of drooling, swallowing, and feeding problems in CP. Most studies include rather small samples and, as such, sample sizes should be weighted. The studies have relatively heterogeneous patient characteristics and therefore confounding variables such as age, motor functioning (i.e. varying Gross Motor Function Classification System [GMFCS] levels) and level of intellectual functioning should be considered. In addition, the measures and variables to determine prevalence rates may vary and should be considered when comparing prevalence data between studies. The purpose of this systematic review was to retrieve all published data on drooling, swallowing, and feeding problems in persons with CP across the lifespan, and to determine the pooled prevalence estimates of drooling, swallowing, and feeding problems using meta-analyses.

What this paper adds

- Drooling, swallowing, and feeding problems are very common in persons with cerebral palsy (CP).
- The prevalence of drooling, swallowing, and feeding problems is 44.0%, 50.4%, and 53.5% respectively.
- There are limited data on the prevalence of drooling, swallowing, and feeding problems in adults.
- Higher Gross Motor Function Classification System levels are associated with higher prevalence of drooling, swallowing, and feeding problems.
- There is increased risk for malnutrition, dehydration, aspiration pneumonia, and poor quality of life in CP.

METHOD

The methodology and reporting of this systematic review were based on the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement and checklist.[22] The PRISMA statement and checklist are designed to enhance the essential and transparent reporting of systematic reviews.[23]

Eligibility criteria

To be eligible for inclusion in this systematic review, articles were required to report on prevalence data of the proportion of people with CP in the sample with either drooling and/or swallowing and/or feeding problems, or data could be retrieved from the article by calculation. Drooling was defined as the involuntary loss of saliva, liquid, or food from the oral cavity as a result of incomplete lip closure. Swallowing and feeding problems were interpreted broadly; for example, problems could refer to dysphagia, feeding difficulties, problems with self-feeding, or masticatory problems. Specifically, swallowing problems refer to problems in the transport of a bolus from the oral cavity to the stomach and could be characterized by poor tongue function, delayed swallow initiation, or reduced pharyngeal motility. Feeding problems refer to problems with breastfeeding or bottle feeding, transition to solid foods, and/or the process of setting up, arranging, and bringing food or liquid from a plate or cup to the mouth. Feeding problems incorporated both child–caregiver interaction and child behaviours, and could present with prolonged feeding times or delayed progression of oral feeding skills. Studies solely reporting on malocclusion, dental caries, dysarthria, dyspraxia, or weight loss were

excluded. Studies on oesophageal problems, such as regurgitation and vomiting, as well as studies on eating disorders, such as anorexia or bulimia, and behavioural eating aversions or picky eaters, were beyond the scope of this review.

Studies where people with CP were included based on the preselection criteria that they had confirmed drooling, swallowing, or feeding problems were excluded. To make the systematic review viable, sample sizes of the included studies had to exceed 30 patients. Study inclusion was not limited by study design (e.g. cross-sectional, cohort, case–control study design). Only original peer-reviewed articles were included; therefore, conference abstracts, reviews, case reports, student dissertations, and editorials were excluded. All studies had to be published in English. To be included in the systematic review, articles had to meet all eligibility criteria.

Data sources and search strategies

A literature search was performed in five different databases: AMED (1995–2017), CINAHL (1937–2017), Embase (1902–2017), MEDLINE (1946–2017), and PubMed (1966–2017), with the following dates of coverage. All publication dates up to 4th November 2018 were included. To identify the most recent publications, searches with subject headings were supplemented by free-text words using a publication limit of at least 1 year before November 2018. All search strategies are presented in Table SI (online supporting information). Additionally, reference lists of included studies were searched by hand.

Study selection

Two independent abstract reviewers performed a stepwise eligibility assessment. First, titles and abstracts were screened for eligibility. At least one reviewer screened all records. A random sample (50%) of the records was reviewed by both reviewers to determine the interrater reliability between reviewers as calculated by a weighted kappa.[24] After excluding records that did not meet the inclusion criteria for this review, the full-text articles of the remaining records were retrieved for further assessment. Both reviewers assessed all full-text articles for eligibility. Differences of opinion about the eligibility of articles were settled by consensus.

Methodological quality assessment

The methodological quality of included studies was assessed using the STROBE checklist for cohort, case–control, and cross-sectional studies (combined).[19,25] The following four domains were assessed for quality: study design and setting; study participants; outcomes; and eligibility criteria. Studies were assigned a score of 1 for each domain assessed when they contained the information listed in the checklist and could be replicated using the information provided, giving a maximum total quality assessment score of 4. If minor quality concerns were identified within a domain, a 0.5 score was allocated, whereas in case of a study clearly not meeting quality criteria, a score of zero was allocated. For example, for studies that did not provide GMFCS level or similar data on functional limitations of the included study participants, a 0.5 score was given for the criterion of study participants. Studies that were published before the introduction of the GMFCS were not penalized for not reporting on GMFCS.[26] However, not providing an adequate definition of swallowing and feeding problems was considered a more serious limitation and therefore a score of 0 was given for the outcomes criterion. All STROBE assessments were consensus-based ratings by two authors.

In addition, the methodological quality of all included articles was determined using AXIS, a critical appraisal tool used to assess the quality of cross-sectional studies.[27] The AXIS tool consists of 20 items or criteria with 'Yes' or 'No' response options. The authors added a numerical scale by assigning a value of 1 for 'Yes' ratings or 0 for 'No' ratings for each item. All but two items were scored such that 'Yes' was indicative of better methodological quality. To create a uniform scoring method, the scoring of these two items was reversed. Thus, high scores ('Yes' or 1) came to indicate higher methodological quality and low scores ('No' or 0) lower quality. Next, to achieve overall quality scores per study, sum scores were calculated as follows: the minimum total score is 0 and the maximum score is 20. AXIS scores were based on consensus ratings between two authors.

Data extraction

A data extraction form was developed to retrieve data from the included studies. Data were extracted on study setting and country; sample characteristics; description of participants' motor and cognitive functioning; description of key terminology related to drooling, swallowing, and feeding problems; assessment methods; and prevalence data. Two authors were involved in the data extraction process. Data retrieved from all articles were reviewed by both authors and discrepancies were resolved through consensus.

Data items, risk of bias, and synthesis of results

Data were retrieved from the included studies using comprehensive extraction forms. The risk of bias was assessed at individual study level using the STROBE checklist for cohort, case–control, and cross-sectional studies (combined) and the AXIS tool. Interrater reliability for independent abstract selection between two reviewers was established based on weighted kappa calculations. Final study selection and quality assessments were the result of consensus-based ratings of two authors. Likewise, all extracted data were reviewed by both authors and discrepancies resolved through consensus. As none of the reviewers has formal or informal affiliations with any of the authors of the included studies, no evident bias in quality assessment or data extraction was expected.

Meta-analysis

For the purpose of the meta-analyses, data on swallowing problems were included if they related to any problem

during the swallowing process; however, data on visceral swallowing or masticatory difficulties due to incomplete teeth were excluded. Feeding problems referred to both eating and drinking problems, including contextual and behavioural factors such as parental stress during mealtimes, problems with breast or bottle feeding, or problems with self-feeding due to developmental delays or motor impairments. Only studies where prevalence data were comprehensively reported were included in the meta-analyses; studies that only reported data on specific aspects of either swallowing or feeding problems were excluded. For example, studies were excluded from the meta-analyses if prevalence data were available for only single items such as 'exhaustion while feeding', 'difficulties biting', or 'feeding is less enjoyable', in the absence of an overarching construct that accounted for the multidimensionality of feeding problems. Similarly, studies were excluded from the meta-analyses when swallowing problems were defined using single items, such as 'difficulties swallowing solids' and therefore not accounting for other aspects of the swallowing process.

Data from medical registers were not included in the meta-analyses given that the quality criteria for completeness, reliability, and validity of data may not have been met sufficiently.[28] For those studies that used a longitudinal design, only the most recent prevalence estimates were considered, disregarding previous, repeated measurements over time. In case of intervention studies, only pretreatment data were included. Finally, if the same study populations were used for prevalence estimates in different publications, selected data were included in the meta-analyses avoiding the use of data on the same participants more than once in determining pooled prevalence estimates; only the most comprehensive and relevant data for drooling, swallowing, and feeding were selected based on clinical consensus by two reviewers.

Descriptive statistics were used to summarize study characteristics and data extraction. Estimates of pooled prevalence weighted by sample size using random-effects models for summary statistics were separately calculated for drooling, swallowing, and feeding problems, as it was unlikely that the included studies have the same true effect due to variations in sampling, outcome measurement, and participant characteristics.

Subsampling was chosen as the predominant analytic technique for this review, as the small number of studies with both available and suitable data limited the viability of conducting meta-regression using multiple covariates. Data were extracted from the included studies to measure the overall within- and between-group prevalence of drooling, swallowing, and feeding. Overall within-group prevalence accounted for all studies with data for drooling, swallowing, and feeding. An overall between-group prevalence was conducted to determine potentially confounding variables as a function of type of assessment (parent vs clinical assessment) and CP severity (GMFCS levels). As such, for overall between-group prevalence, data were grouped and pooled by type of assessment for drooling and swallowing, as data containing these categories were not available for feeding. Data were also grouped and pooled by GMFCS levels (levels I–V individually and grouped I–III and IV–V) for drooling, swallowing, and feeding. Publication bias was assessed using the classic fail-safe N test. The test calculates the number of additional studies that would nullify the measured effect (N), if added to the analysis. A large N is indicative that it is unlikely the meta-analysis is compromised by publication bias. All statistical analyses were performed using software package Comprehensive Meta-Analysis Version 3.3.070 (Biostat; Englewood, NJ, USA).

RESULTS

Study selection

A total of 339 citations were identified across the five databases (AMED, CINAHL, Embase, MEDLINE, and PubMed), representing 258 independent studies after deleting duplicates. Two independent reviewers screened all records. The agreement between reviewers, as determined by weighted kappa, was 0.91 (95% confidence interval [CI] 0.84–0.98), indicating excellent interrater reliability.[24] Eighty-two full-text articles were assessed for eligibility, of which 38 articles met the inclusion criteria. In addition, four articles were identified after checking the reference lists of the included articles, resulting in a total of 42 included articles. A flowchart of the selection process according to PRISMA is shown in Figure S1 (online supporting information).[22]

Description of studies

All included studies are summarized in detail in Table SII (online supporting information). Data were grouped under the following subheadings: reference; study quality as reported by STROBE score; study setting and country; sample characteristics (number, sex, and age); description of participants' motor and cognitive functioning (e.g. motor type, GMFCS level, intellectual disabilities); description of drooling, swallowing, and feeding problems; assessment methods used for prevalence calculations; and prevalence data.

Methodological quality

STROBE scores can vary between 0 and 4. A score of 4 indicates that all items for all four domains (study design and setting, study participants, outcomes, and eligibility criteria) were adequately met. STROBE scores for the 42 included studies ranged from 2 to 4, with an average STROBE score of 3.2: score of 2 (n=1 study),[29] score of 2.5 (n=8 studies),[30–37] score of 3 (n=11 studies),[38–48] score of 3.5 (n=18 studies),[49–66] and score of 4 (n=4 studies).[67–70] The most common methodological issues included absent or incomplete definitions of outcome variables, no reporting of GMFCS levels or other information on CP severity, or minor inconsistencies in data analyses. Further details on STROBE scores can be found in Table SIII (online supporting information).

AXIS scores ranged between 13 and 20 (maximum score). Two studies scored less than 15,[33,37] whereas five studies received the maximum score.[45,54,60,63,68] Mean score was 17.2 (SD 1.7).

Participants

The 42 studies included an estimated total of 23 169 participants. The number of participants per study ranged from 30 to 14 806, with a median participant number of 120 (interquartile range [IQR] 55–186): 30≤*n*<50 (*n*=8 studies);[29,33–38,49] *n*<100 (*n*=10 studies);[39,42–44,47,48,50,63,64,69] *n*<200 (*n*=14 studies);[30–32,40,41,46,51–53,61,62,65,67] *n*<500 (*n*=5 studies);[45,58,60,66,68] *n*≥500 (*n*=5 studies).[54,55,57,59,70] Age ranged from birth to 79 years (mean 10y 1mo [SD 11y]); however, only six of the 42 studies included adults (>18y), of which two studies included adults only.[31,59] Data were retrieved from studies conducted across 20 countries, mainly from disability registers, special needs schools, adult group homes, and clinical centres.

Assessment methods

Prevalence data of drooling, swallowing, and feeding problems were determined using different types of assessments. Studies used clinical assessments (*n*=13 studies);[31,33,35,39,41,44,48–50,52,61,62,69] parent or carer questionnaires or interviews (*n*=7 studies);[29,42,45,46,63,64,68] or a combination of both (*n*=12).[30,34,36,37,40,43,51,53,56,65–67] Nine studies used data from medical registers or charts,[34,45,54,55,57–60,70] of which three added data from either clinical assessments,[47] or parent or carer questionnaires.[38,60] Many measures were designed by the authors only for the purpose of their study, whereas some studies used standardized measures from the literature (e.g. Schedule for Oral Motor Assessment, Pre-Speech Assessment Scale, Pediatric Evaluation of Disability Inventory, Dysphagia Disorder Survey, Thomas-Stonell Greenberg Saliva Severity Scale).

Meta-analyses

Studies included participants with a great variety of drooling, swallowing, and feeding problems. The concept of drooling or sialorrhoea was generally clearly defined and referred to involuntarily spillage of saliva from the mouth. However, feeding and swallowing problems were not always well defined and showed great heterogeneity between studies. Swallowing problems referred to, for example, dysphagia, signs of pharyngeal impairment (e.g. choking, gurgly voice), or impaired oro-motor skills.

Nine studies included data from medical registers and were excluded from meta-analyses.[34,45,54,55,57–60,70] To further reduce heterogeneity in the data when conducting meta-analyses, the two studies that included adult participants only were also excluded.[31,59] Four studies used a longitudinal design;[29,34,50,65] thus, only the most recent prevalence estimates were considered. Finally, to avoid using data on the same participants more than once in determining pooled prevalence estimates, only selected data from seven studies were included in the meta-analyses.[50–53,65–67] Table SII provides an overview of prevalence estimates as retrieved from the literature; data used for meta-analyses have been marked.

Drooling

Drooling prevalence data were available from 13 studies,[29,37,40,41,44,45,47,52,56,60–62,69] with a pooled prevalence estimate of 44.0% (95% CI 35.6–52.7; Table I; Fig. S2, online supporting information). The between-group differences were not significant when comparing clinical assessments (*n*=8 studies)[37,44,47,52,56,61,62,69] with parents and carers reports (*n*=5 studies),[29,40,41,45,60] with pooled prevalence estimates of 50.8% (95% CI 41.7–59.9) and 34.2% (95% CI 24.2–45.8) respectively. Two studies provided prevalence data for each of the five GMFCS levels individually.[52,56] Pooled prevalence estimates stratified by GMFCS level were: 22.0% for level I (95% CI 6.9–51.9), 36.0% for level II (95% CI 23–51.4), 36.0% for level III (95% CI 10.8–72.3), 64.4% for level IV (95% CI 44.7–80.3), and 85.7% for level V (95% CI 45.5–97.7). Data from three studies provided pooled prevalence estimates for GMFCS levels I to III combined and GMFCS levels IV to V combined.[40,52,56] The pooled prevalence estimates were 24.3% (95% CI 12.2–42.6) and 68.7% (95% CI 54.1–80.4) respectively. The overall between-group differences were not significant.

This meta-analysis of drooling incorporated data from 13 studies, which yielded a z-score of –1.359 and corresponding two-tailed *p*-value of 0.174. As the combined result is not statistically significant, the fail-safe N (which addresses the concern that the observed significance may be spurious) is not relevant.

Swallowing

Prevalence data on swallowing problems across 10 studies resulted in an estimated pooled prevalence of 50.4% (95% CI 36.0–64.8) (Table II; Fig. S3, online supporting information).[29,30,34,35,39,40,47,56,65,68] Six studies used clinical assessment,[30,34,35,39,47,65] and four studies used parent or carer report to determine swallowing prevalence,[29,40,56,68] resulting in pooled prevalence estimates of 68.4% (95% CI 46.2–84.4) and 29.9% (95% CI 16.5–47.9) respectively. The overall between-group differences were not significant. Pooled prevalence estimates stratified by GMFCS level retrieved from three studies were: 16.3% for level I (95% CI 10.2–24.9), 51.7% for level II (95% CI 32.8–70.1), 60.4% for level III (95% CI 43.0–75.5), 84.2% for level IV (95% CI 71.3–91.9), and 97.9% for level V (95% CI 90.7–99.6).[30,56,65] The overall between-group differences were not significant. Pooled prevalence estimates for GMFCS levels I to III combined (*n*=3 studies)[40,56,65] were 23.9% (95% CI 10.8–44.9) and 88.3% (95% CI 45.7–98.5) for GMFCS levels IV to V combined (*n*=4 studies).[30,40,56,65] The overall between-group differences were not significant.

This meta-analysis for swallowing incorporated data from 10 studies yielding a z-score of 0.055 and corresponding two-tailed *p*-value of 0.956. As the combined

Table I: Meta-analyses for drooling

Study	(Sub)group	Prevalence (%)	95% CI	*p*
Motion et al.,[29] Franklin et al.,[37] Erkin et al.,[40] Hegde and Pani,[41] Morales-Chávez et al.,[44] Reid et al.,[45] Waterman et al.,[47] Benfer et al.,[52] Edvinsson and Lundqvist,[56] Sullivan et al.,[60] Tahmassebi and Curzon,[61] Santos et al.,[62] Sedky[69]	Overall within-group prevalence	44.0	35.6–52.7	0.174
Franklin et al.,[37] Morales-Chávez et al.,[44] Waterman et al.,[47] Benfer et al.,[52] Edvinsson and Lundqvist,[56] Tahmassebi and Curzon,[61] Santos et al.,[62] Sedky[69]	Clinical assessment	50.8	41.7–59.9	0.860
Motion et al.,[29] Erkin et al.,[40] Hegde and Pani,[41] Reid et al.,[45] Sullivan et al.[60]	Parent or carer report	34.2	24.2–45.8	0.008[a]
Motion et al.,[29] Franklin et al.,[37] Erkin et al.,[40] Hegde and Pani,[41] Morales-Chávez et al.,[44] Reid et al.,[45] Waterman et al.,[47] Benfer et al.,[52] Edvinsson and Lundqvist,[56] Sullivan et al.,[60] Tahmassebi and Curzon,[61] Santos et al.,[62] Sedky[69]	Overall between group	44.6	37.5–51.9	0.146
Benfer et al.,[52] Edvinsson and Lundqvist[56]	GMFCS level I	22.0	6.9–51.9	0.065
Benfer et al.,[52] Edvinsson and Lundqvist[56]	GMFCS level II	36.0	23.0–51.4	0.073
Benfer et al.,[52] Edvinsson and Lundqvist[56]	GMFCS level III	36.0	10.8–72.3	0.461
Benfer et al.,[52] Edvinsson and Lundqvist[56]	GMFCS level IV	64.4	44.7–80.3	0.150
Benfer et al.,[52] Edvinsson and Lundqvist[56]	GMFCS level V	85.7	45.5–97.7	0.075
Benfer et al.,[52] Edvinsson and Lundqvist[56]	Overall between group	45.2	34.8–56.0	0.381
Erkin et al.,[40] Benfer et al.,[52] Edvinsson and Lundqvist[56]	GMFCS levels I–III	24.3	12.2–42.6	0.008[a]
Erkin et al.,[40] Benfer et al.,[52] Edvinsson and Lundqvist[56]	GMFCS levels IV–V	68.7	54.1–80.4	0.013[b]
Erkin et al.,[40] Benfer et al.,[52] Edvinsson and Lundqvist[56]	Overall between group	52.6	40.2–64.7	0.684

Criteria for meta-analyses: comprehensive measure, most recent prevalence estimates (longitudinal design), pretreatment data (intervention studies), selected data when same study populations used in different publications (clinical consensus), no data from medical registers. [a]$p<0.01$; [b]$p<0.05$. CI, confidence interval; GMFCS, Gross Motor Function Classification System.

result is not statistically significant, the fail-safe N (which addresses the concern that the observed significance may be spurious) is not relevant.

Feeding

Data from eight studies were included in the meta-analysis on feeding problems,[30,33,38,40,43,45,46,68] resulting in an estimated pooled prevalence of 53.5% (95% CI 40.7–65.9; Table III; Fig. S4, online supporting information). Prevalence data were based on parent or carer report (n=6 studies),[30,38,40,45,46,68] clinical assessment (n=1 study),[33] or combined data on parent or carer report and clinical assessment (n=1 study).[43] Owing to the limited numbers per type of assessment, no group differences were analysed. When determining pooled prevalence estimates by GMFCS level (n=2 studies),[43,68] the results were as follows: 25.0% for level I (95% CI 6.3–62.3), 50.0% for level II (95% CI 16.8–83.2), 51.8% for level III (95% CI 7.6–93.4), 58.6% for level IV (95% CI 20.2–88.8), and 89.4% for level V (95% CI 64.6–97.5). GMFCS levels I to III combined (n=2 studies)[40,43] and GMFCS levels IV to V combined (n=3 studies)[40,43,68] yielded pooled prevalence estimates of 19.8% (95% CI 1.5–79.6) and 70.3% (95% CI 45.3–87.2) respectively. No overall between-group differences were significant.

This meta-analysis of feeding problems incorporated data from eight studies, which yielded a z-score of 0.55 and corresponding two-tailed p-value of 0.579. As the combined result is not statistically significant, the fail-safe N (which addresses the concern that the observed significance may be spurious) is not relevant.

DISCUSSION

Systematic review findings

This is the first systematic review and meta-analyses on prevalence estimates of drooling, swallowing, and feeding problems in CP. A total of 42 studies were included from five different literature databases. Most studies included wide age ranges of participants, from infancy to late teenage years, without differentiating the age groups in the prevalence data. Furthermore, 10 studies provided limited background information on the degree of intellectual disabilities for included participants, but no stratified prevalence data were reported based on categories of intellectual functioning.[30,31,34,35,39,46,47,57,63,70] Nineteen studies described GMFCS levels of their participants,[30,40,43,45,50–58,63,65–69] of which all but four presented prevalence data by GMFCS level.[45,57,63,69] Remarkably, even though CP is a lifelong condition, only two of 36 studies focused exclusively on adult populations,[31,59] exposing a substantial knowledge gap in the lifespan experiences of drooling, swallowing, or feeding in people with CP.

Based on this review, the need for consensus on best evidence-based practice in daily clinics and research on drooling, feeding, and swallowing problems in persons with CP is evident. Without guidelines on best evidence-based practice, differences in definitions on concepts like feeding and swallowing problems, the use of measures with unknown or poor psychometric characteristics, and great variability in the selected assessment and outcome variables will remain a challenge and have a negative impact on the healthcare planning and management of persons with CP. The Eating and Drinking Ability Classification System is a newly developed

Table II: Meta-analyses for swallowing problems

Study	(Sub)group	Prevalence (%)	95% CI	*p*
Motion et al.,[29] Calis et al.,[30] Reilly Skuse et al.,[34] Thommessen et al.,[35] Del Giudice et al.,[39] Erkin et al.,[40] Waterman et al.,[47] Edvinsson and Lundqvist,[56] Benfer et al.,[65] Fung et al.[68]	Overall within group prevalence	50.4	36.0–64.8	0.956
Calis et al.,[30] Reilly Skuse et al.,[34] Thommessen et al.,[35] Del Giudice et al.,[39] Waterman et al.,[47] Benfer et al.[65]	Clinical assessment	68.4	46.2–84.4	0.102
Motion et al.,[29] Erkin et al.,[40] Edvinsson and Lundqvist,[56] Fung et al.[68]	Parent or carer report	29.9	16.5–47.9	0.030[a]
Motion et al.,[29] Calis et al.,[30] Reilly Skuse et al.,[34] Thommessen et al.,[35] Del Giudice et al.,[39] Erkin et al.,[40] Waterman et al.,[47] Edvinsson and Lundqvist,[56] Benfer et al.,[65] Fung et al.[68]	Overall between group	45.4	31.5–60.0	0.537
Edvinsson and Lundqvist,[56] Benfer et al.[65]	GMFCS level I	16.3	10.2–24.9	<0.001[b]
Edvinsson and Lundqvist,[56] Benfer et al.[65]	GMFCS level II	51.7	32.8–70.1	0.866
Edvinsson and Lundqvist,[56] Benfer et al.[65]	GMFCS level III	60.4	43.0–75.5	0.239
Calis et al.,[30] Edvinsson and Lundqvist,[56] Benfer et al.[65]	GMFCS level IV	84.2	71.3–91.9	<0.001[b]
Calis et al.,[30] Edvinsson and Lundqvist,[56] Benfer et al.[65]	GMFCS level V	97.9	90.7–99.6	<0.001[b]
Calis et al.,[30] Edvinsson and Lundqvist,[56] Benfer et al.[65]	Overall between group	49.2	41.1–57.4	0.857
Calis et al.,[30] Edvinsson and Lundqvist,[56] Benfer et al.[65]	GMFCS levels I–III	23.9	10.8–44.9	0.017[a]
Calis et al.,[30] Erkin et al.,[40] Edvinsson and Lundqvist,[56] Benfer et al.[65]	GMFCS levels IV–V	88.3	45.7–98.5	0.071
Calis et al.,[30] Erkin et al.,[40] Edvinsson and Lundqvist,[56] Benfer et al.[65]	Overall between group	34.2	17.8–55.5	0.143

Criteria for meta-analyses: comprehensive measure, most recent prevalence estimates (longitudinal design), pretreatment data (intervention studies), selected data when same study populations used in different publications (clinical consensus), no data from medical registers. [a]$p<0.05$; [b]$p<0.01$. CI, confidence interval; GMFCS, Gross Motor Function Classification System.

Table III: Meta-analyses for feeding problems

Study	(Sub)group	Prevalence (%)	95% CI	*p*
Calis et al.,[30] Reilly et al.,[33] Dahl et al.,[38] Erkin et al.,[40] Martinez-Biarge et al.,[43] Reid et al.,[45] Stallings et al.,[46] Fung et al.[68]	Overall within group prevalence	53.5	40.7–65.9	0.592
Martinez-Biarge et al.[43]	GMFCS level I	25.0	6.3–62.3	0.178
Martinez-Biarge et al.[43]	GMFCS level II	50.0	16.8–83.2	1.000
Martinez-Biarge et al.,[43] Fung et al.[68]	GMFCS level III	51.8	7.6–93.4	0.956
Martinez-Biarge et al.,[43] Fung et al.[68]	GMFCS level IV	58.6	20.2–88.8	0.692
Martinez-Biarge et al.,[43] Fung et al.[68]	GMFCS level V	89.4	64.6–97.5	0.006[a]
Martinez-Biarge et al.,[43] Fung et al.[68]	Overall between group	58.9	40.0–75.6	0.356
Erkin et al.,[40] Martinez-Biarge et al.[43]	GMFCS levels I–III	19.8	1.5–79.6	0.321
Erkin et al.,[40] Martinez-Biarge et al.,[43] Fung et al.[68]	GMFCS levels IV–V	70.3	45.3–87.2	0.108
Erkin et al.,[40] Martinez-Biarge et al.,[43] Fung et al.[68]	Overall between group	64.0	39.9–82.6	0.251

Criteria for meta-analyses: comprehensive measure, most recent prevalence estimates (longitudinal design), pretreatment data (intervention studies), selected data when same study populations used in different publications (clinical consensus), no data from medical registers. [a]$p<0.01$. CI, confidence interval; GMFCS, Gross Motor Function Classification System.

classification framework aimed at improving the classification of eating and drinking abilities in persons with CP.[71] However, the Eating and Drinking Ability Classification System requires additional psychometric evaluation and wider consultation before its implementation in clinics and research can be justified. Furthermore, the Eating and Drinking Ability Classification System uses 'safe' eating and drinking as the premise of the classification, without explicit reference to how silent aspiration should be measured.

In the current review considerable variability was observed in selected outcome measures. Most studies used clinical assessments or combined these with parent or carer questionnaires, some used parent or carer questionnaires only or used data from medical registers. An obvious advantage of using existing medical registers is the direct access to patient data that may have been collected over a longer period of time in relatively large patient cohorts.[45,54,55,57–60,70] However, criteria for data quality, including data completeness and reliability and validity of data, may not have been adequately achieved in medical registers.[28] As such, data may lack comprehensiveness (i.e. not including sufficiently detailed data for research purposes) or inclusiveness (i.e. raising doubts around population selections).[72]

Six studies[30,51,52,65–67] used the Dysphagia Disorder Survey,[73] the most psychometrically robust measure in non-instrumental swallowing and feeding assessment in paediatrics according to a recent review by Speyer et al.[7] However, only limited information was available on their reliability and validity for most outcome measures, and many studies used measures that were developed for single use only.

There was also considerable variation in the variables used to capture prevalence of drooling, swallowing, and feeding problems. Some studies only reported on specific aspects of the problems, without presenting data on the overarching constructs; for example, seven studies presented data on self-feeding or assistance during feeding.[35,36,47,55,59,64,65] The ability to self-feed is an important aspect of feeding but does

not represent feeding problems as a comprehensive construct. Other studies only reported, for example, on lip competence,[37] oral skills,[48] modified diet,[47] or cough during oral intake.[36] These variables do contribute to feeding or swallowing problems; however, individually, they do not capture the complete, overarching constructs, and were therefore excluded from meta-analyses. Also, occasionally, some doubts about underlying causes of identified problems arose. For example, chewing problems can be associated with the presence of CP, but also with general weakness and incomplete teeth. Further, authors used different variables to measure the same construct. When considering drooling, for example, selected variables to assess drooling differed considerably, such as presence or absence of: 'drooling saliva', 'excessive drooling' or of 'drooling most of the time', ordinal severity scales of drooling, or ordinal frequency scales of drooling. This is despite having consensus about what constitutes drooling in most studies. Similar issues arose for variables on swallowing and feeding problems.

Studies only presenting group data on separate aspects of feeding and swallowing were excluded from meta-analyses, if group data could not be linked to individual outcomes; data could not be combined into more comprehensive reports on swallowing or feeding problems. However, exclusion of articles from meta-analyses does not imply that these studies lack sufficient methodological quality. The main reason for excluding studies from the meta-analyses related to the suitability of the data for conducting the meta-analysis.

Meta-analyses findings

Twenty-three of 42 studies were included in the meta-analyses and resulted in pooled prevalence estimates of 44.0% (95% CI 35.6–52.7) for drooling, 50.4% (95% CI 36.0–64.8) for swallowing problems, and 53.5% (95% CI 40.7–65.9) for feeding problems. These prevalence rates are very high. As feeding problems referred to eating and drinking, including contextual and behavioural factors, the prevalence of feeding problems was expected to be higher than that of swallowing problems. Feeding is a more general term comprising a larger variety of contributing factors and variables, compared with swallowing.

Even though there was a clear under-reporting by parents, the prevalence estimates for drooling did not show significant between-group differences when comparing parent or carer reports with clinical assessment. Possibly, carers' lack of awareness of drooling may result from having to deal with many other problems associated with CP on a regular basis, and drooling may be considered a nuisance without serious consequences. Owing to large confidence intervals, prevalence estimates for both swallowing and feeding showed no significant between-group differences, even though data indicated a similar trend for lower prevalence estimates by parents or carers. If there was higher homogeneity in the data between studies, it would most likely have resulted in significant differences with higher prevalence when assessed by clinicians. The construct of swallowing problems is more complex to measure, compared with the construct of drooling, which likely increased data heterogeneity. Further, no significant between-group differences were found for GMFCS levels, but, as expected, a clear trend emerged in which more severely impaired functioning in CP was associated with increased problems of drooling, swallowing, and feeding. Interestingly, the general prevalence trends showed marked increases from one GMFCS level to the next, except for the increase from GMFCS level II to III, with increases only ranging from 0.0% to 1.8% (see Tables I–III).

No meta-analyses were conducted based on age, as age groups per study differed greatly. Further, only very limited data were available for adult populations; as such, conducting a subgroup analysis was not viable. In terms of the adult population, Strauss et al. reported on self-feeding prevalence (aspect of feeding) based on a database of developmental disabilities,[59] whereas Henderson et al. identified adults with dysphagia based on a health status survey without providing further details on defining or diagnosing dysphagia.[31] In addition, group comparison between participants with and without an intellectual disability was not possible, as only a few studies reported on participants' cognitive functioning using diverse outcome measures.

Limitations

Despite achieving high interrater reliability between independent reviewers during the PRISMA article selection process, this systematic review and meta-analysis is subject to limitations. Most of the included studies showed minor methodological shortcomings, according to the STROBE or AXIS scores. Only two of 42 studies were awarded the maximum STROBE score of 4,[67,68] meeting all criteria for study quality, whereas five studies achieved the maximum AXIS score of 20.[45,54,60,63,68] All other studies presented with minor methodological issues. In addition, meta-analyses conducted are subject to heterogeneity in study designs, patient populations, and outcome measures and variables. Studies differed in defining constructs of swallowing, feeding, and drooling, and showed great variability in selected outcome measures and variables. Therefore, even though the studies included in meta-analyses were selected with great caution to include only studies showing sufficient similarity based on clinical consensus judgement, the results should still be interpreted with caution.

CONCLUSION

The current review retrieved 42 articles reporting on prevalence of drooling, swallowing, and feeding problems in persons with CP. Pooled prevalence estimates determined by meta-analyses were as high as 44.0% (95% CI 35.6–52.7) for drooling, 50.4% (95% CI 36.0–64.8) for swallowing problems, and 53.5% (95% CI 40.7–65.9) for feeding problems, indicating that persons with CP are at high-risk for malnutrition and dehydration, aspiration pneumonia, and, subsequently, poor quality of life. As too few studies reported on adult populations, all pooled data were based on younger populations (0–18y).

Future studies should include adult populations with CP to address existing knowledge gaps to account for individual characteristics like CP severity, age categories, and the presence of intellectual disabilities. Furthermore, studies should use outcome measures with robust psychometric properties when reporting prevalence data on drooling, swallowing, or feeding.

ACKNOWLEDGEMENTS

The authors have stated that they had no interests that might be perceived as posing a conflict or bias.

SUPPORTING INFORMATION

The following additional material may be found online:

Table SI: Search strategies per literature database

Table SII: Characteristics of included studies

Table SIII: STROBE scores based on the checklist for cohort, case–control, and cross-sectional studies

Figure S1: Flow diagram of the reviewing process according to PRISMA.

Figure S2: Forest plot of drooling prevalence.

Figure S3: Forest plot of swallowing problems prevalence.

Figure S4: Forest plot of feeding problems prevalence.

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RESUMEN

PREVALENCIA DE PROBLEMAS DE SIALORREA, DEGLUCIÓN Y ALIMENTACIÓN EN PARÁLISIS CEREBRAL A LO LARGO DE LA VIDA: UNA REVISIÓN SISTEMÁTICA Y METAANÁLISIS

OBJETIVO Determinar la prevalencia de problemas de sialorrea/babeo, deglución y alimentación en personas con parálisis cerebral (PC) a lo largo de la vida

MÉTODO Se llevó a cabo una revisión sistemática utilizando cinco bases de datos diferentes (AMED, CINAHL, Embase, MEDLINE y PubMed). El proceso de selección fue completado por dos investigadores independientes y la calidad metodológica de los estudios incluidos se evaluó utilizando las directrices STROBE y AXIS. Se realizó un metaanálisis para determinar las estimaciones de prevalencia agrupadas en problemas de babeo, deglución y alimentación con análisis de grupos estratificados por tipo de evaluación y nivel del Sistema de Clasificación de la Función Motora Gruesa.

RESULTADOS Se incluyeron un total de 42 estudios. Se observaron variaciones sustanciales en las medidas y variables de resultados seleccionadas, y los datos sobre adultos fueron limitados. Las estimaciones de prevalencia agrupadas determinadas por metaanálisis fueron tan altas como 44,0% (intervalo de confianza [IC] del 95% 35,6–52,7) para babeo, 50,4% (IC 95% 36,0–64,8) para problemas de deglución y 53,5 % (IC 95% 40,7–65,9) para problemas de alimentación. Los análisis de grupo para el tipo de evaluaciones no fueron significativos; sin embargo, el funcionamiento más severo en PC se asoció con problemas concomitantes de aumento de sialorrea, deglución y de la alimentación.

INTERPRETACIÓN Problemas relacionados con sialorrea, tragar, y de alimentación son muy comunes en personas con PC. En consecuencia, ellos experimentan mayores riesgos de desnutrición y deshidratación, neumonía por aspiración y mala calidad de vida.

RESUMO

PREVALÊNCIA DE PROBLEMAS COM SIALORRÉIA, DEGLUTIÇÃO E ALIMENTAÇÃO EM PARALISIA CEREBRAL AO LONGO DA VIDA: UMA REVISÃO SISTEMÁTICA E METANÁLISE

OBJETIVO Determinar a prevalência de problemas com sialorréia, deglutição e alimentação em pessoas com paralisia cerebral (PC) ao longo da vida.

MÉTODO Uma revisão sistemática foi realizada utilizando cinco bases de dados diferentes (AMED, CINAHL, Embase, MEDLINE, e PubMed). O processo de seleção foi realizado por dois pesquisadores independentes e a qualidade metodológica dos estudos incluídos foi avaliada usando as diretrizes STROBE e AXIS. Metanálises foram realizadas para determinar as estimativas de prevalência agrupada de problemas de sialorréia, deglutição e alimentação, com análises estratificadas por tipo de avaliação e nível do Sistema de Classificação da Função Motora Grossa.

RESULTADOS Um total de 42 estudos foram incluídos. Variações substanciais nas medidas de resultado e variáveis selecionadas foram observadas, e dados em adultos são limitados. As estimativas de prevalência agrupada determiadas pela metanálise chegaram a 44,0% (intervalo de confiança [IC] 95% 35,6–52,7) para sialorréia, 50,4% (IC 95% 36,0–64,8) para problemas com deglutição, e 53,5% (IC 95% 40,7–65,9) para problemas de alimentação. Análises agrupadas por tipo de avaliação não foram significativas; no entanto, comprometimento funcional mais severo em PC foi associado com mais problemas concomitantes de salivação deglutição e alimentação.

INTERPRETAÇÃO Problemas de sialorréia, deglutição e alimentação são muito comuns em pessoas com PC. Consequentemente, elas apresentam risco aumentado de malnutrição e desidratação, pneumonia por aspiração e pior qualidade de vida.