Author/s:
McInerney, MS;Reddihough, DS;Carding, PN;Swanton, R;Walton, CM;Imms, C

Title:
Behavioural interventions to treat drooling in children with neurodisability: a systematic review

Date:
2019-01-01

Citation:
McInerney, M. S., Reddihough, D. S., Carding, P. N., Swanton, R., Walton, C. M. & Imms, C. (2019). Behavioural interventions to treat drooling in children with neurodisability: a systematic review. Developmental Medicine and Child Neurology, 61 (1), pp.39-48. https://doi.org/10.1111/dmcn.14048.

Persistent Link:
https://hdl.handle.net/11343/284599

MS MICHELLE SARAH MCINERNEY (Orcid ID : 0000-0002-9713-6566)
PROFESSOR CHRISTINE IMMS (Orcid ID : 0000-0001-9055-3554)

Article type : Systematic Review

[Systematic Review: 4 tables; 2 figures]

Behavioural interventions to treat drooling in children with neurodisability: a systematic review

MICHELLE S MCINERNEY[1]
DINAH S REDDIHOUGH[2,3,4]
PAUL N CARDING[5]
RUTH SWANTON[6]
CHLOE M WALTON[7]
CHRISTINE IMMS[8]

1 National School of Allied Health, Australian Catholic University, Sydney; **2** Neurodevelopment and Disability, Royal Children's Hospital, Melbourne; **3** NHMRC Centre of Research Excellence in Cerebral Palsy, Murdoch Children's Research Institute, Melbourne; **4** Department of Paediatrics; University of Melbourne, Melbourne; **5** National School of Allied Health, Australian Catholic University, Brisbane; **6** National School of Allied Health and Centre for Disability and Development Research at the Australian Catholic University, Sydney; **7** National School of Allied Health and Centre for Disability and Development Research at the Australian Catholic University, Brisbane; **8** National School of Allied Health and Centre for Disability and Development Research at the Australian Catholic University, Melbourne, Australia.

Correspondence to Michelle McInerney, National School of Allied Health, Australian Catholic University, Level 3, TWH, 8-20 Napier St, North Sydney, NSW 2060, Australia. E-mail: Michelle McInerney@acu.edu.au

PUBLICATION DATA

Accepted for publication 00th Month 2018.

Published online 00th Month 2018.

ABBREVIATIONS

RCT Randomized controlled trial

RoBiNT Risk of Bias assessment in N-of-1 Trials

SCED Single case experimental design

AIM To review the evidence for behavioural interventions to reduce drooling in children with neurodisability.

METHOD A detailed search in eight databases sought studies that (1) included participants aged 0 to 18 years with neurodisability and drooling; (2) provided behavioural interventions targeting drooling or a drooling-related behaviour; and (3) used experimental designs. Two reviewers extracted data from full text papers independently. Results were tabulated for comparison. The Risk of Bias assessment in N-of-1 Trials scale for single case experimental designs (SCEDs) and the Cochrane risk of bias assessment tool for randomized controlled trials (RCTs) were applied.

RESULTS Of an initial yield of 763, seven SCEDs and one RCT were included. Behavioural interventions included the use of reinforcement, prompting, cueing, self-management, overcorrection, and fading. Each assessed body functions or structures' outcomes (drooling frequency and severity); three included activity outcomes (mouth drying, head control, eye contact, and vocalizations) and none assessed participation or quality of life. While each study reported positive effects of intervention, risk of bias was high.

INTERPRETATION: Low-level evidence suggests behavioural interventions may be useful for treatment of drooling in children with neurodisability. Well-designed intervention studies are urgently needed to determine effectiveness.

What this paper adds:

- Behavioural interventions used to treat drooling included reinforcement, prompting, overcorrection, instruction, and fading.
- Interventions targeted body structures and function-level outcomes and activity-level outcomes.
- Low-level evidence supports the use of behavioural intervention to treat drooling.

First page footer]
(c) Mac Keith Press
DOI: 10.1111/dmcn.xxxxx

[Left page footer]
Developmental Medicine & Child Neurology 2018: XX: 000–000

[Right page footer]
Review

[main text]
Anterior drooling is defined as spillage of saliva from the mouth that is clearly visible, while posterior drooling is defined as saliva that spills through the oropharynx into the hypopharynx.[1] Anterior drooling, the focus of this systematic review, is common and disabling for many children with neurodisability.[2] It is estimated that 40 per cent of children between the ages of 7 years and 14 years with cerebral palsy (CP) will experience drooling.[3] In children with neurodisability the reasons for drooling include reduced awareness of drooling,[4] less frequent and inefficient swallowing,[5] and oral sensory issues,[6] which can be further compounded by postural problems or reflux.[7] The presence of contributing factors, such as the use of medications that exacerbate drooling, which is often a necessity in the life of a child with neurodisability, adds to this complexity. The health consequences of drooling can be serious and potentially life-threatening and include skin irritation and maceration,[8] and social rejection from peers.[9] Importantly, poor saliva control can also lead to reduced self-esteem and can affect the individual's successful integration into the community.[10] Additionally, there are secondary impacts in caring for the child who drools, such as greater daily care demands and increased stress levels.[11] The presence of problematic drooling therefore

has the potential to reduce the quality of life of both the child and the wider family unit.

Drooling is a multi-dimensional behaviour with multiple potential causes or contributing factors that necessitates the expertise of a multidisciplinary team.[12] Many interventions are available to a multidisciplinary team to treat drooling in children with neurodisability; however, there is no consensus about their effectiveness.[13] Botulinum neurotoxin A, surgery, and the use of medications are considered invasive treatments, whilst less invasive-treatments include behavioural, oral motor therapies, and the use of appliances.[14]

Behavioural interventions are widely recommended for drooling and it is frequently suggested that they are used before invasive treatments are considered.[15] These interventions are based on behavioural modification principles and are underpinned by the premise that all behaviour is influenced by learning principles of classical and operant conditioning.[16] The aim of these interventions is to increase target behaviours,[17] such as swallowing saliva, wiping lips and chin, improving head control, and teaching ways to manage drooling independently. Treatment techniques can include: instruction (e.g. please wipe your chin), prompting (e.g. giving a verbal prompt to swallow), positive reinforcement (e.g. praise), and/or self-management (e.g. instructions to swallow and wipe the chin)[18] (Table SI, online supporting information).

There is limited evidence to support the use of behavioural interventions. Although a previous descriptive review of 19 studies reported on the effectiveness of behavioural interventions for drooling,[18] no conclusion could be drawn regarding efficacy because of a number of methodological issues including non-experimental design, drooling measurement issues, and poor interobserver reliability.

Why should a behavioural intervention work to reduce drooling?

Behavioural interventions are concerned with analysing and modifying human behaviour, in the context in which the behaviours take place. These interventions are developed to change socially significant behaviours with the goal of improving some aspect of the person's life.[19] Behavioural interventions have their foundations in learning theory and build on the premise that human behaviour is learned through an interaction between an individual and their environment. Contemporary behavioural therapy also assumes that the individual is capable of self-directed change and that the person is the agent of that change. The focus of behavioural intervention is on

observable behaviour, current determinants of behaviour, learning experiences that promote change, tailoring treatment strategies to individual clients, and rigorous assessment and evaluation.[20]

Drooling behaviour is a suitable target for behavioural intervention because the principles of behavioural intervention can be readily applied to treat drooling. First, drooling is a socially significant human behaviour that has the potential to seriously affect the child's physical and psychosocial health,[21] and has an impact on the physical and social context.[22] Second, drooling is an overt observable behaviour, which has dimensions of frequency and intensity that can be described, recorded, and measured. In delivering a behavioural intervention, a stimulus is provided either before, during, or after the unwanted drooling behaviour occurs, to facilitate a change in the individual's responses or behaviour that will reduce or eliminate the drooling,[23] for example praising a person for not drooling for a specified period. Third, the occurrence of drooling can be influenced by contextual events, for example the person is in a poor sitting position, which results in increased head flexion, which can exacerbate drooling. By describing the functional relationship between drooling and context, hypotheses can be formulated about the reasons for the behaviour and corresponding procedures can be put in place, for example prompting carers to always place the person in the correct position. Finally, many treatments for drooling aim to increase the occurrence of desirable motor behaviours (e.g. swallowing) and decrease the occurrence of undesirable behaviours (i.e. drooling) and therefore involve opportunities to support motor learning. Behavioural intervention can facilitate motor learning through practice, stimulus selection, rehearsal, and feedback.[24]

The International Classification of Functioning, Disability and Health[25] was used as the conceptual framework for this study. The International Classification of Functioning, Disability and Health conceptualizes a person's level of 'functioning' as a dynamic interaction between his or her health conditions, environmental factors, and personal factors. Functioning includes two parts related to (1) body functions and structures which describe the anatomy and physiology/psychology of the human body and (2) activity and participation, that describe the individuals' current functional level related to skills like mobility, self-care communication, and learning.

The purpose of this review was to identify and synthesize the evidence for behavioural interventions to reduce drooling in children with neurodisability and to examine intervention effectiveness at the level of body functions and structures. The secondary purpose was to investigate whether these treatments had an effect on

activity, participation, and quality of life. The findings of this review may provide evidence to guide practice and future research in the treatment of drooling in children with neurodisability.

METHOD

This systematic review was designed in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses protocols (Figure S1, online supporting information).[26] The review protocol was registered online with the International Prospective Register of Systematic Reviews Protocols. [27] The American Academy of Cerebral Palsy and Developmental Medicine Methodology to Develop Systematic Reviews of Treatment Interventions[28] was used to determine the level of evidence of studies found.

Search strategy

A comprehensive search strategy was created using medical subject terms: 'child', 'neurodisability', 'drooling', 'behavioural', 'intervention', and associated relevant subject headings. The search was modified for the following eight electronic databases: MEDLINE, CINAHL, AMED, CENTRAL, Embase, PsycINFO, Scopus, and Web of Science (see Table SII, online supporting information for MEDLINE search strategy). The number and type of databases were chosen to minimize selection bias and increase the generalizability of this systematic review.[29] Preliminary searching began in February 2017 and the final search was completed on June 2nd, 2017. All of the retrieved articles were imported into the bibliographic database, EndNote X8 (Thomas Reuters [Scientific] LLC, Philadelphia, PA, USA) and then exported into the Covidence database for screening and management. One reviewer (MSM) also completed a manual hand search of reference lists of full text articles in English.

A limited search of unpublished sources in the grey literature was also conducted. The search terms were adapted and the following sources were searched: Trove, The Agency for Healthcare Research and Quality, Australian Institute of Health and Welfare, National Health Medical Research Council, and Registry of Open Access Repositories.

Selection criteria

Studies were eligible if they (1) included participants between the ages of 0 and 18 years with a diagnosis of neurodisability and drooling; (2) provided behavioural interventions that targeted drooling or a drooling-related behaviour; and (3) used experimental study designs including randomized controlled trials (RCTs), clinical controlled trials, and single case experimental design (SCED) studies.

Study selection

Through initial screening of articles by the primary reviewer (MSM), duplicates, review articles, conference proceedings, editorials, poster presentations, and articles in languages other than English were removed. After this, two reviewers (MSM and CMW) independently applied selection criteria to title, abstract, and full text articles. Most differences were resolved through consensus. A third party (CI) was consulted if differences were unresolved until a final decision was reached.

Data extraction

A data extraction form was first created, customized, and trialled. Two reviewers (MSM and CMW) then independently used this form to extract data on the following variables: study design, participant number and diagnostic information, behavioural intervention, target behaviour definition, outcomes of interest, outcome measure used, main findings, and level of evidence.

Data synthesis

It was not possible to complete a meta-analysis as heterogeneity was found in: (1) population diagnoses; (2) data collection methods and measures used in the included studies; and (3) study outcomes reported. A narrative approach was therefore used to synthesize the data on key study variables, study quality, and risk of bias.

Risk of bias assessment

Risk of bias assessment was performed at a study level. The Risk of Bias assessment in N-of-1 Trials (RoBiNT) Scale[30] was used to assess the risk of bias in studies where single case experimental methodology was used. In RCTs, we used the Cochrane Collaboration Tool[31] to assess risk of bias. Two independent reviewers (MSM and CMW) completed risk of bias assessment on RCTs. SCED studies were examined using the RoBiNT Scale by two reviewers (MSM and RS). Cases where there were different ratings were resolved by consensus.

RESULTS

Study design

The search identified 763 papers. There were 93 studies that were assessed for full text eligibility against selection criteria and a final total of eight papers were included. Seven studies used a SCED approach and one was an RCT (Table I). Four of the eight articles were published between 1977 and 1989, with the remaining four published between 2006 and 2011. All included studies used an experimental design with control. Four of the SCED studies used a withdrawal/reversal (ABAB) design and three used a concurrent multiple baseline design.

Participants

The eight included studies involved a total of 35 participants who received behavioural interventions (Table I). Of these, 25 were included in the single RCT. The age range of the participants was 5 years to 17 years.

Behavioural interventions

In all studies, a combination of behavioural intervention methods was used, for example prompting and reinforcement, rather than only a single method. Reinforcement was used in all eight studies,[32–39] with positive reinforcement in seven studies,[32-35, 37–39] and token economy reinforcement in two studies.[32,37] Prompting was used in six studies.[33–37,39] Three of the studies used the term 'cueing' instead of 'prompt', in which a visual, auditory, or vibratory stimulus prompt was provided by an assistive technology device to make the correct response more likely, for example swallowing, wiping the mouth[33-35] (see Table SI for behavioural intervention definitions). The other strategies included self-management,[37] overcorrection (positive punishment),[38] instruction,[39] and fading[34] (Table I).

Outcomes assessed

Each study aimed to reduce a target behaviour related to either drooling frequency and/or severity. Frequency-related outcomes in two studies included measures of drooling frequency[32] and drooling drops per minute.[37] Four studies assessed drooling-severity related outcomes defined as percentage of chin wetness,[34] percentage of wet time samples,[33] pools of saliva,[39] and drooling status.[38] Activity-level outcomes were measured in three studies and included mouth drying as a response,[34,35] improved

head control, eye contact, and imitative vocalization.[36] None of the studies assessed outcomes related to participation or quality of life.

Effectiveness of interventions

Each study reported a positive result related to drooling and drooling-related behaviours (Table I). No adverse effects were reported. Two studies described reduced drooling frequency. In the RCT,[32] drooling frequency decreased significantly in the experimental group after token economy and positive reinforcement compared to the non-experimental group ($p<0.01$). In this study, the participants received a token (e.g. star sticker) and verbal reinforcement after cessation of drooling for a specified length of time. Opportunity was provided to collect more star stickers (by not drooling) and once four were collected, these could be exchanged for a reward. In a second study, drooling was reportedly eliminated in one individual in all task situations, after a combination of behavioural intervention methods including prompting, positive and token reinforcement, and self-management.[37]

In the remaining studies, a drooling-severity related behaviour was the outcome of interest, with visual inspection of the data suggesting that drooling severity dropped below baseline levels in three out of six studies.[33-35] Statistical testing was used in only one SCED study[35] with a significant difference in drooling severity demonstrated ($p<0.01$) after a period of positive reinforcement and cueing using assistive technology. None of the SCED studies completed a structured visual analysis whereby the data within and between phase patterns were systematically evaluated in terms of level, slope or trend, variability, immediacy of effect, and consistency of data patterns across similar phases.[30] No robust conclusions could therefore be drawn about the size of effect or the clinical impact that behavioural intervention could have on drooling.

Risk of bias: SCED studies

Total internal validity scores of the RoBiNT Scale[30] across the studies ranged from 2 to 5 (out of a possible score of 14). All seven studies met the criterion for experimental design (Table II) but none used randomization. No blinding of either assessors or people involved in the intervention took place; however, it is acknowledged that blinding of participants and interventionists is not possible when implementing behavioural interventions. External validity scores ranged from 3 to 8 out of a possible score of 16 (Table II). The lowest scoring categories pertained to

those of baseline characteristics, generalization, data analysis, and replication. Apart from the 'design with control' category, there were fundamental issues across all categories related to internal and external validity (Table II). Total RoBiNT scale scores ranged between 6 and 12 (out of a score of 30), with four of the seven SCEDs obtaining a score below 10 (Table II).

Risk of bias: the RCT

One single blind, two-group RCT was included in this review (Table I). The process of allocation concealment lacked sufficient detail. Blinding of participants and interventionists was not possible, and any measures to blind outcome assessors were not described. Outcome data on premeasures and postmeasures were provided but there was no information on dropout rates, indicating an unclear risk of attrition bias. Plausible bias associated with items related to performance, detection, and attrition weakens confidence in results presented.

DISCUSSION

Positive outcomes were reported in all eight studies in this systematic review that included a total of 35 participants with different clinical presentations. Despite the prevalence of drooling in children with neurodisability and the likely common use of behavioural interventions, very few experimental studies have been undertaken to determine whether these interventions work. All eight studies targeted outcomes at a body functions and structures level with only three studies targeting activity outcomes. A reduction in drooling behaviour related to frequency or severity in neurodisability populations was reported in each study. An increase in an activity related to drooling was also reported, for example mouth drying. No study used outcome measures related to participation or quality of life and no validated measures were used to assess outcomes of interest in included studies. Significant quality issues related to recruitment, risk of bias, and external validity were present in all eight studies, greatly limiting the interpretation as to how effective behavioural interventions are, in reducing drooling in children with neurodisability.

All the included SCED studies met the criteria for experimental study design, however none used randomization of phase order or onset. The application of randomization is a critical component of SCEDs where threats to internal validity can be experimentally controlled and opportunities for statistical testing can be

facilitated.[40] The use of randomization further enables the researcher to determine whether there is a causal or functional relationship between the manipulated independent variable (behavioural intervention) and dependent variable (drooling behaviour).[41]

Visual analysis is the predominant method used to analyse data in SCED research. Data were displayed graphically and a visual inspection of the data occurred in the included studies, but a comprehensive visual analysis was not completed in any study. Though visual analysis remains the SCED standard, experts strongly advocate the use of both visual and statistical analysis methods.[40,41] This includes the requirement to provide information on changes to the level of the score from the measure of the dependent variable (target behaviour) after manipulation of the independent variable (treatment); emerging trends or slopes in the data; and commenting on variability in the data.[30,40] Only three of the seven SCED studies discussed trends in the data and only one study used statistical analysis methods. The premise of SCED research is to investigate if there is a change in the person's targeted behaviour of interest after a period of intervention. We cannot know what the effect of any clinical intervention is without sufficient analysis of the data.

What are the important elements of a behavioural intervention?

This review highlighted many fundamental issues about implementing behavioural interventions to treat drooling in children with neurodisability. First, a functional analysis of the baseline condition, that is drooling, that includes identifying factors that are maintaining the drooling behaviour is needed,[17] for example head control or the child's positioning. As part of this analysis, adequate information must be provided about the baseline characteristics of the individual, including diagnosis and severity of condition. This will enable the development of effective function-based treatment.[42] Second, a clear definition of the target behaviour (drooling) is needed to know what the person does that signifies a drooling event and to facilitate accurate recording of that event. Third, treatment procedures need to be specified, outlining what needs to be said or done in treatment to signify that active ingredients are being delivered.[43] In specifying the content and procedures involved in delivering a treatment, the intervention 'fidelity' is partly ensured. Fidelity is an important part of best practice,[44] as higher levels of treatment fidelity are associated with higher gains attributable to intervention.[45] Finally, a multifaceted intervention approach is

warranted as drooling is a complex behaviour to treat and a single intervention alone is unlikely to be effective in eliminating or reducing drooling significantly.

How should drooling be measured?

One of the hallmarks of behavioural intervention is its emphasis on measuring the behaviour before, during, and after the intervention so that any change resulting from that intervention is captured.[19] To provide an accurate measure of drooling, an operational definition of the target behaviour, that is easily observable and measurable, needs to be provided. This infers that the drooling behaviour of interest is identified and defined in precise terms using action verbs; for example, any saliva that spills from the mouth that is clearly visible. In addition, the method of measurement must be described in terms of how data are recorded, what instruments were used, and what constitutes a correct or incorrect response.[30] Ongoing assessment of the behaviour should also be done well beyond the point of intervention to truly measure change.[19]

Since the publication of four of the included studies, two validated measures of drooling, the Drooling Quotient[46] and Drooling Impact Scale,[47] have become available. The Drooling Impact Scale is a subjective scale completed by parents, which has good test–retest reliability and has been shown to be responsive to change in children who have undergone saliva control interventions. The Drooling Quotient is a reliable and objective measure of drooling severity. It is recommended that the available validated measures be used to reduce the potential error in measuring drooling.

Implications for research

First, it is recommended that a distinction be made between anterior and posterior drooling and used consistently in conducting research to facilitate transparency, collaboration, and direction in future research. For example, the anterior drooling definition provided by the American Academy of Cerebral Palsy and Developmental Medicine[1] refers to the anterior spillage of saliva from the mouth that is clearly visible beyond the lip margin. A distinction between anterior and posterior drooling was not made in any of the included studies (Table II). Furthermore, providing an operational definition of the drooling behaviour targeted in the study describes the dependent variable, which is an important external validity construct.

Second, experimental, as opposed to non-experimental or quasi-experimental studies, are needed to better demonstrate a causal relationship between the intervention and the effects on drooling.[41] When proof of concept is present, robust RCTs are warranted. SCED studies are well suited to investigating preliminary effects of interventions,[40,48] but better-quality studies that address important internal and external validity constructs are needed. Randomizing the interventions phase order or onset helps to reduce the threats to internal validity and this is recommended so researchers can improve the credibility of study findings.[41] Future SCED studies should include a randomization component to minimize the risk of bias and to better detect any intervention effects. The RoBiNT Scale[30] was designed as an appraisal tool; it does, however, also provide a very useful guide to researchers designing future SCEDs. Completion of high quality SCEDs will improve the evidence base for the role of behavioural interventions in the treatment of drooling.

Third, it is recommended that researchers use the available validated measures of drooling severity and impact. None of the included studies used a validated measure of drooling, instead using terms such 'problematic drooling' and 'chronic drooling', and it was therefore difficult to gauge the baseline drooling status before commencement of intervention. In addition, research that focuses on drooling outcomes at an activity, participation, and quality of life level, in addition to a body functions and structures level, is needed. Through investigating the effects of behavioural interventions on these outcomes relevant to the child's life stage, the potential of these treatments to improve health and development can be evaluated. To fully investigate the potential effect of behavioural interventions, an objective, reliable, and valid means of assessing and describing all relevant clinical characteristics of drooling and its contributing factors is needed, which currently does not exist. The development of a validated tool that assesses all aspects of the drooling behaviour, beyond severity and its impact, is warranted.

Finally, a descriptive analysis of drooling is an important part of the functional assessment, which is a necessary component before implementing a behavioural intervention. The International Classification of Functioning Disability and Health – Children and Youth Version[49] is an extension of the original International Classification of Functioning, Disability and Health[25] framework, and can be used to describe and organize information about drooling using a coding classification system. Through using this, the complexity of drooling behaviour can be highlighted (Table SIII, online supporting information). This descriptive outcome is the functional

profile of the individual, not a diagnosis, and may facilitate the planning of comprehensive assessment and intervention. The identification of all components of this complex behaviour is critical for best practice to improve health outcomes for children with neurodisability who drool.

Strengths and limitations of the review

This systematic review aimed to answer a specific and focused clinical intervention question. The finding of only eight small experimental studies, despite a search of published and grey literature to locate relevant research, suggests that this area is under-researched. Four of the included studies were published over 30 years ago (1977–1989). This left four contemporary studies in this area from which to draw evidence. Significant advances in areas of practice closely related to drooling, for example swallowing, have occurred since the completion of the older studies. It is possible that studies published in languages other than English may have yielded additional information.

Conclusion

The few experimental studies identified that were designed to reduce drooling in children with neurodisability demonstrated positive outcomes, although findings are tempered with high risk of bias. By designing studies that use experimental methods and addressing fundamental quality issues related to internal and external validity, researchers would be in a better position to test the efficacy of such interventions. This will inform us as to how effective and potentially valuable behavioural intervention could be. Activity and participation level outcomes need to be targeted in this clinical intervention research, in addition to quality of life. Given the reported prevalence of drooling in this population, priority research is warranted to advance the evidence base related to behavioural interventions for drooling in current practice.

Acknowledgements

The authors acknowledge the support of the NHMRC-funded Centre of Research Excellence in Cerebral Palsy (#1057997). The authors have stated that they had no interests that could be perceived as posing a conflict or bias.

SUPPORTING INFORMATION

The following additional material may be found online:

Table SI: Behavioural intervention definitions

Table SII: Search strategy in MEDLINE

Table SIII: Description of drooling using International Classification of Functioning Disability and Health – Children and Youth coding

Figure S1: Preferred Reporting Items for Systematic Reviews and Meta-Analyses flow diagram

REFERENCES

1. Delsing C, Glader LJ, Hughes A, et al. Sialorrhea in cerebral palsy [Internet]. Milwaukee, WI: American Academy of Cerebral Palsy and Developmental Medicine, 2018. Available at: https://www.aacpdm.org/publications/care-pathways/sialorrhea (accessed 12 February 2018).

2. Blasco PA, Allaire JH. Drooling in the developmentally disabled: management practices and recommendations. *Dev Med Child Neurol* 1992; **34**: 849–62.

3. Reid SM, McCutcheon J, Reddihough DS, Johnson H. Prevalence and predictors of drooling in 7- to 14-year-old children with cerebral palsy: a population study. *Dev Med Child Neurol* 2012; **54**: 1032–6.

4. Little SA, Kubba H, Hussain SS. An evidence-based approach to the child who drools saliva. *Clin Otolaryngol* 2009; **34**: 236–9.

5. Senner JE, Logemann J, Zecker S, Gaebler-Spira D. Drooling, saliva production, and swallowing in cerebral palsy. *Dev Med Child Neurol* 2004; **46**: 801–6.

6. Reilly S. The limitations in interpreting the evidence for behavioral interventions for drooling. *Evid Based Commun Assess Interv* 2008; **2**: 3–5.

7. Leung AK, Kao CP. Drooling in children. *Paediatr Child Health* 1999; **4**: 406–11.

8. Blasco PA. Management of drooling: 10 years after the Consortium on Drooling, 1990. *Dev Med Child Neurol* 2002; **44**: 778–81.

9. Van der Burg J, Jongerius P, van Limbeek J, van Hulst K, Rotteveel JJ. Social interaction and self-esteem of children with cerebral palsy after treatment for severe drooling. *Eur J Pediatr* 2006; **165**: 37–41.

10. Reddihough D, Johnson H. Assessment and management of saliva control problems in children and adults with neurological impairment. *J Dev Phys Disabil* 1999; **11**: 17–24.

11. Bella GP, Garcia MC, Spadari-Bratfisch RC. Salivary cortisol, stress, and health in primary caregivers (mothers) of children with cerebral palsy. *Psychoneuroendocrinology* 2011; **36**: 834–42.

12. Daniel SJ. Multidisciplinary management of sialorrhea in children. *Laryngoscope* 2012; 122 (Suppl. 4): s67–8.

13. Walshe M, Smith M, Pennington L. Interventions for drooling in children with cerebral palsy. *Cochrane Database Syst Rev* 2012; **11**: CD008624.

14. Bavikatte G, Sit PL, Hassoon A. Management of drooling of saliva. *Br J Med Pract* 2012; **5**: a507.

15. Meningaud JP, Pitak-Arnnop P, Chikhani L, Bertrand JC. Drooling of saliva: a review of the etiology and management options. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2006; **101**: 48–57.

16. Burton LJ, Westen D, Kowalski RM, editors. Psychology, Fourth Australian and New Zealand edition. Milton, Qld: Wiley, 2015.

17. Corey G. Theory and practice of group counseling, Ninth Edition. Boston, MA: Cengage Learning, 2015.

18. Van Der Burg JJ, Didden R, Jongerius PH, Rotteveel JJ. A descriptive analysis of studies on behavioural treatment of drooling (1970–2005). *Dev Med Child Neurol* 2007; **49**: 390–4.

19. Miltenberger R. Behavior modification: principles and procedures, Fifth edition. Belmont, CA: Wadsworth Cengage Learning, 2012.

20. Corey G. Theory and practice of counseling and psychotherapy, Eighth Edition. Pacific Grove, CA: Thomson, Brooks/Cole, 2009.

21. Chang SC, Lin CK, Tung LC, Chang NY. The association of drooling and health-related quality of life in children with cerebral palsy. *Neuropsychiatr Dis Treat* 2012; **8**: 599–604.

22. Tahmassebi JF, Curzon ME. Prevalence of drooling in children with cerebral palsy attending special schools. *Dev Med Child Neurol* 2003; **45**: 613–7.

23. Kazdin AE. Behavior modification in applied settings, Sixth Edition. Belmont, CA: Wadsworth/Thomas Learning, 2001.

24. Valvano J. Activity-focused motor interventions for children with neurological conditions. *Phys Occup Ther Pediatr* 2004; **24**: 79–107.

25. World Health Organization. The International Classification of Functioning, Disability & Health (ICF): an overview [Internet]. Geneva, Switzerland: WHO, 2001. Available at: https://www.cdc.gov/nchs/data/icd/icfoverview_finalforwho10sept.pdf (accessed 18 April 2017).

26. Hutton B, Moher D, Cameron C. The PRISMA extension statement. *Ann Intern Med* 2015; **163**: 566–7.

27. National Institute for Health Research. PRSOPERO International Prospective Register of Systematic Reviews Protocols [Internet]. York, UK: NHS, 2017. Available at: http://www.crd.york.ac.uk/PROSPERO/display_record.php?ID=CRD42017060963 (accessed 19 February 2017).

28. Darrah J, Hickman, R, O'Donnell, M, Vogtle, L, Wiart, L. The American Academy of Cerebral Palsy and Developmental Medicine methodology to develop systematic reviews of treatment interventions. Milwaukee, WI: American Academy of Cerebral Palsy and Developmental Medicine, 2008. Available at: https://www.aacpdm.org/UserFiles/file/systematic-review-methodology.pdf (accessed 5 May 2018).

29. Vassar M, Yerokhin V, Sinnett PM, et al. Database selection in systematic reviews: an insight through clinical neurology. *Health Info Libr J* 2017; **34**: 156–64.

30. Tate RL, Rosenkoetter U, Wakim D, et al. The risk-of-bias in N-of-1 trials (RoBiNT) scale: an expanded manual for the critical appraisal of single-case reports. St Leonards, NSW: John Walsh Centre for Rehabilitation Research, 2015.

31. Higgins JPT. Cochrane Handbook for Systematic Reviews of Interventions. Cochrane Collaboration. Chicester, UK: Wiley, 2008.

32. Sethy D, Mokashi S, Chia Swee H. Effect of a token economy behaviour therapy on drooling in children with cerebral palsy. *Int J Ther Rehabil* 2011; **18**: 494–9.

33. Lancioni GE, Coninx F, Manders N, Driessen M. Use of automatic cueing to reduce drooling in two multihandicapped students. *Journal of the Multihandicapped Person* 1989; **2**: 201–10.

34. Lancioni GE, Singh NN, O'Reilly MF, et al. Technology-assisted programs to promote mouth drying and reduce the effects of drooling with two persons with developmental disabilities. *J Dev Phys Disabil* 2009; **21**: 555–64.

35. Lancioni GE, Singh NN, O'Reilly MF, et al. Two persons with multiple disabilities use a mouth-drying response to reduce the effects of their drooling. *Res Dev Disabil* 2009; **30**: 1229–36.

36. Richman JS, Kozlowski NL. Operant training of head control and beginning language for a severely developmentally disabled child. *Jour Beh Ther Exp Psych* 1977; **8**: 437–40.

37. Dunn KW, Cunningham CE, Backman JE. Self-control and reinforcement in the management of a cerebral-palsied adolescent's drooling. *Dev Med Child Neurol* 1987; **29**: 305–10.

38. Drabman RS, Cordua Y Cruz G, Ross J, Lynd S. Suppression of chronic drooling in mentally retarded children and adolescents: effectiveness of a behavioral treatment package. *Behav Ther* 1979; **10**: 46–56.

39. Kay S, Harchik A, Luiselli J. Elimination of drooling by an adolescent student with autism attending public high school. *J Posit Behav Interv* 2006; **8**: 24–8.

40. Smith J. Single-case experimental designs: a systematic review of published research and current standards. *Psychol Methods* 2012; **17**: 510–50.

41. Kratochwill T, Levin J. Single-case intervention research: methodological and statistical advances.Washington, DC: American Pyschological Assocation, 2014.

42. Roscoe E, Schlichenmeyer KJ, Dube WV. Functional analysis of problem behavior: a systematic approach for identifying idiosyncratic variables. *J Appl Behav Anal* 2015; **48**: 289–314.

43. Hart T. Treatment definition in complex rehabilitation interventions. *Neuropsychol Rehabil* 2009; **19**: 824–40.

44. Krasny-Pacini A, Evans J. Single-case experimental designs to assess intervention effectiveness in rehabilitation: a practical guide. *Ann Phys Rehabil Med* 2017; **61**: 164–79.

45. Hinckley JJ, Douglas NF. Treatment fidelity: its importance and reported frequency in aphasia treatment studies. *Am J Speech Lang Pathol* 2013; **22**: S279–84.

46. Van Hulst K, Lindeboom R, van der Burg J, Jongerius P. Accurate assessment of drooling severity with the 5-minute drooling quotient in children with developmental disabilities. *Dev Med Child Neurol* 2012; **54**: 1121–6.

47. Reid SM, Johnson HM, Reddihough DS. The Drooling Impact Scale: a measure of the impact of drooling in children with developmental disabilities. *Dev Med Child Neurol* 2010; 52: e23–8.

48. Portney LG, Watkins MP. Foundations of clinical research: applications to practice, Third Edition. Philadelphia, PA: F.A. Davis Company, 2015.

49. World Health Organization. International Classification of Functioning Disability and Health: Child and Youth version [Internet]. Geneva, Switzerland: WHO, 2007. Available at: http://apps.who.int/iris/bitstream/handle/10665/43737/9789241547321_eng.pdf;jsessionid=BE065DF8DE8C1200A339C2490C65F7F0?sequence=1 (accessed 4 June 2018).

Study and aim	Level of evidence design	Participants diagnosis and age	Total *n*	Target behaviour definition	Behavioural intervention	Outcomes of interest	Measure	Main findings
Sethy et al.[32] Effect of a token economy behaviour on drooling	Level II RCT	CP and mild ID; problematic drooling; 5–12y	25	Saliva that spilled over the lower lip and fell out of the mouth	1. Token economy reinforcement 2. Positive reinforcement	BFS: Drooling frequency A: N/A P: N/A	Continuous recording=frequency of drooling episodes for 20mins	Significant decrease in drooling frequency in experimental group (n=12) compared to non-experimental group (n=13) (p<0.01). Reduction not maintained
Lancioni et al.[34] Technology-assisted promotion of mouth drying to reduce effects of drooling	Level IV SCED: withdrawal/ reversal ABAB design	Multiple DD; persistent drooling problem; 16y	1	Chin wetness: saliva appearing below the lower lip Mouth drying response: passing mouth and chin over spongy surface to activate two touch/pressure microswitches	1. Prompting 2. Fading 3. Positive reinforcement	BFS: Drooling severity (chin wetness) A: Mouth drying response P: N/A	5s interval recording of chin wetness Electronic recording of mouth-drying response	Increase in mouth drying Decrease in chin wetness Improvements not maintained

Table I: Characteristics of included studies

Dunn et al.[37] Self-control and reinforcement in the management of drooling in one adolescent with CP	Level II SCED: MBD; three situations ('social', 'cognitive', and 'no' demand)	Spastic quadriplegia CP; ID and non-verbal; long history of drooling; 16y	1	The number of drops falling from the lip or chin	1. Self-management (self-control) 2. Prompting 3. Positive reinforcement 4. Token economy reinforcement	BFS: Drooling frequency A: N/A P: N/A	Continuous recording=frequency of drooling episodes	Self-control was the most effective treatment component. Drooling eliminated in all three situations
Lancioni et al.[35] Two persons with multiple disabilities use a mouth drying response to reduce the effects of their drooling	Level IV SCED: withdrawal/ reversal ABAB design	Down Syndrome, visual impairment and non-verbal; long history of drooling; 12y 2mo	1	Chin wetness: saliva appearing below the lower lip Mouth drying response: bringing/pushing the napkin to mouth (to activate two sensors)	1. Prompting 2. Positive reinforcement	BFS: Drooling severity (chin wetness) A: Mouth drying response P: N/A	Electronic recording of mouth-drying response Interval recording of chin wetness using 5s intervals	Significant differences in mouth-drying frequencies and chin wetness ($p<0.01$)

Lancioni et al.[33] Use of automatic cueing to reduce drooling in two students with multiple disabilities	Level IV SCED: withdrawal/ reversal design	Student 1: Deafness; motor impairment, ID; drooled consistently; 16y 10mo Student 2: Deafness and ID; 11y 2mo	2	Wet: saliva appearing outside the lower lip Dry: No saliva appearing outside the lower lip	1. Prompting 2. Positive reinforcement 3. Extinction (of positive reinforcement)	BFS: Drooling severity (percentage of wet time samples) A: N/A P: N/A	Interval recording: judged if dry or wet at the end of 20s intervals	Percentage of time samples in which students were wet reduced during treatment
Richman and Kozlowski[36] Operant training of head control and beginning language for a child with severe developmental disability	Level IV SCED: withdrawal/ reversal design	Multiple DD; quadriplegic spastic CP; baseline drooling status not reported; 9y 8mo	1	The presence of saliva on the lips or mouth	1. Prompting 2. Contingent reinforcement of all behaviours	BFS: Drooling severity (dry or wet) A: (1) Eye (initiation and maintenance of eye contact) (2) Head (independent maintenance) (3) Vocal (imitation of vocalization) P: N/A	Interval recording: all measures taken over 10min periods (20s intervals)	Eye, head, vocal behaviours increased with treatment and mean decreases in behaviour during reversal periods. Mean levels of drooling decreased in treatment period

Drabman et al.[38] Suppression of chronic drooling in children and adolescents with intellectual disability; effectiveness of a behavioural treatment package	Level II SCED: multiple baseline design across three participants	'Profound mental retardation' and chronic drooling. Participant 1 7y; participant 2 had left hemiparesis, 15y; participant 3 had cleft palate and orofacial anomaly; 12y	3	Wet: any moisture (besides perspiration) present on the finger Very wet: presence of drool dripping from the child's chin Dry: if saliva was not wiped off by the finger nor was dripping below the chin	1. Positive reinforcement (praise if dry) 2. Over-correction (if wet)	BFS: Drooling severity (dry, wet, or very wet) A: N/A P: N/A	Interval recording every 30min	Baseline 'dry' rates of 10% improved to 50%–60% after treatment Overcorrection for wetness and positive reinforcement for dryness lead to 'durable and stable reductions in drooling'
Kay et al.[39] Elimination of drooling by an adolescent student with autism	Level II SCED: multiple baseline design across three	ASD and 'mental retardation'; 'rudimentary speech'; chronic drooling; 17y	1	'Saliva pool' – 1 inch or greater in diameter	1. Instruction – 'swallow' and 'wipe your mouth' 2. Prompting (physical prompt)	BFS: Drooling severity (pools of saliva on surfaces) A: N/A P: N/A	Interval recording: 5min initially; increased to every 15min in each setting	Steady reduction and eventual elimination of drooling in the three settings

	settings				3. Positive reinforcement (praise)			

RCT, randomized control trial; CP, cerebral palsy: ID, intellectual disability; y, years; BFS, body functions and structures-level outcome; A, activity-level outcome; N/A, not applicable; P, participation-level outcome; SCED, single case experimental design; DD, developmental disability; MBD, multiple baseline design.

Table II: Risk of bias for SCED studies using the Risk of Bias assessment in N-of-1 Trials (RoBiNT) Scale[a]

Internal validity subscale	Study and internal validity scoring								Study and external validity scoring						
	Lancioni et al.[34]	Dunn et al.[37]	Lancioni et al.[35]	Lancioni et al.[33]	Richman and Kozlowski[36]	Drabman et al[38]	Kay et al.[39]	External validity subscale	Lancioni et al.[34]	Dunn et al.[37]	Lancioni et al.[35]	Lancioni et al.[33]	Richman and Kozlowski[36]	Drabman et al.[38]	Kay et al.[39]
Design	2	2	2	2	2	2	2	Baseline characteristic	0	0	0	0	0	0	0
Randomization	0	0	0	0	0	0	0	Setting	1	1	1	1	1	1	1
Sampling of behaviour	0	0	1	1	1	0	2	Dependent variable	2	0	2	2	2	0	0
Blinding of assessor	0	0	0	0	0	0	0	Independent variable	1	1	1	1	2	0	0
Blinding of people involved in intervention	0	0	0	0	0	0	0	Raw data record	1	2	1	1	2	1	1
Interrater agreement	2	0	0	2	1	1	0	Data analysis	0	0	1	0	1	0	0
Treatment adherence	0	0	0	0	0	0	0	Replication	0	0	1	1	0	1	1
								Generalization	0	0	0	0	0	0	0
Total internal validity score	4	2	3	5	4	3	4	Total external validity score	5	4	7	6	8	3	3

[a]The RoBiNT Scale[30] uses a 3-point scoring system (2=items meet stringent criteria; 1=standards met with reservations; 0=failure to meet minimum standards).