PROTOCOL

Study Title: Evaluation of an intensive voice treatment to reduce anterior drooling in children with cerebral palsy: Protocol for a concurrent multiple-baseline, single case experimental design study.

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Abstract

Anterior drooling is common in children with cerebral palsy (CP) and poses significant risks to the child's health. Causes of drooling include oro-motor dysfunction, inefficient swallowing and reduced sensation in the orofacial musculature. Behavioural interventions are frequently recommended to reduce drooling; however, this is in the absence of high-quality research evidence. This paper describes a protocol for evaluating the effectiveness of the Lee Silverman Voice Treatment LOUD® (LSVT LOUD®) in reducing drooling; and optimising speech and swallowing in a group of children with CP. A structured and systematic visual analysis supplemented with statistical analysis will be used to analyse the data. The risk of bias in n-of-1 trials (RoBiNT) Scale [1] guided the design and implementation of the study.

Keywords: drooling, child, cerebral palsy, behavioral intervention.

Abbreviations used in text

CP	Cerebral Palsy
SCED	Single case experimental design
RoBiNT	Risk of bias in n-of-1 trials (RoBiNT)
NSW	New South Wales
ACT	Australian Capital Territory

1. Introduction

Many children with cerebral palsy (CP) experience difficulties controlling saliva [2, 3]. Potential reasons for this clinical feature of CP include: a reduction in swallowing ability [4]; oral motor dysfunction [5]; and reduced sensation in oro-facial musculature [6] resulting in the anterior loss of saliva from the mouth [7]. Drooling can be a disabling condition for children with CP adversely affecting physical and emotional health[8], social interactions and self-esteem [9, 10]. Health impacts include skin maceration and breakdown, skin infection [11], and social rejection from peers [9]. Drooling has also been shown to increase the care needs of the child, putting families under increased stress [12]. Thus, drooling has the potential to reduce the quality of life of both children with CP, and their families.

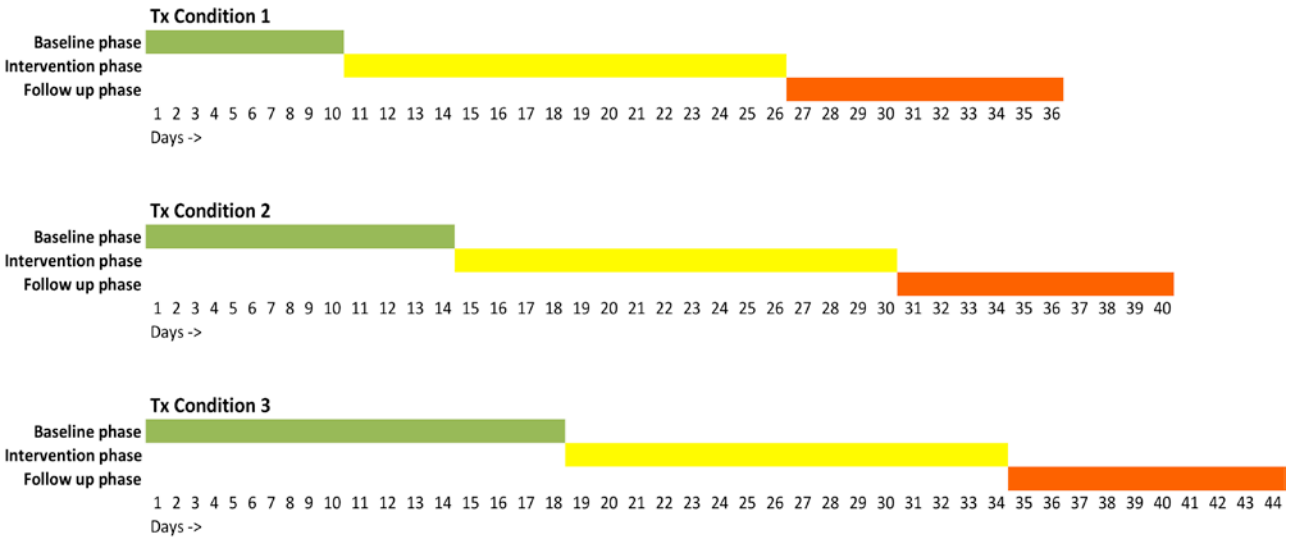
A treatment approach that holds promise in improving swallowing and reducing drooling in children with CP is the Lee Silverman Voice Treatment LOUD (LSVT LOUD®). This intensive voice treatment has already been developed, piloted, reported and implemented, and has demonstrated level I evidence in facilitating positive long-term effects on loudness of speech in adult patients with Parkinson's disease [13-16]. LSVT LOUD® is founded on principles of behavioural theories of learning, motor learning and neuroplasticity [17, 18]. Early clinical outcome research has highlighted similar speech outcomes in children with CP [19-22]. Importantly, cross-generalised effects of LSVT LOUD® to non-targeted skills such as oral motor movements and swallowing have also been reported in adults with Parkinson's disease [23-25]. As reduced swallowing function is a key contributing factor to anterior drooling in children with CP, behavioural interventions such as LSVT LOUD® that optimise swallowing, may also facilitate better management of saliva within the oral cavity, and thereby potentially reduce the anterior spillage of salivary contents, or drooling, in children with CP.

This paper presents a protocol to evaluate the effects of LSVT LOUD® on the target behaviours; drooling severity and impact; swallowing and feeding competency; and speech intelligibility at single word, sentence and conversational level. It is anticipated that the current project will contribute to evidence for managing and treating drooling, and also build on existing evidence in the treatment of motor speech impairments in children with CP. The International Classification of Functioning, Disability and Health- Child & Youth Version (ICF-CY) (World Health Organization [26] provided the conceptual framework for the study.

2. Methods

2.1. Study Design

A SCED involves the repeated measurement of an individual's behaviour in the presence and absence of the intervention, thereby enabling the individual to serve as their own control [27]. A concurrent multiple baseline SCED will be used to investigate the effects of LSVT LOUD® on the primary outcome of drooling, and the secondary outcomes of swallowing and speech. The application of randomization will be made to phase onset with the starting point of intervention differing for each participant (see Fig. 1 below). The baseline phase will be followed immediately by a 16-session treatment period over four weeks. The follow up period will take place 12-weeks after the last treatment session has taken place.



2.2. Participants

Six children between the ages of 7 and 18 years with a diagnosis of CP who have problematic drooling and who live in Sydney will be sought.

Inclusion criteria: (i) confirmed diagnosis of CP (ii) Intelligence Quotient (IQ) falling within the average to moderate range of disability (iii) evidence of frequent drooling as determined by parent or child and affecting the child's physical or social health (iv) severity of drooling has remained stable over the past 3 months (v) aged 7 to 18 years (vi) Level 1 and 2 communicators on the Communication Function Classification System (CFCS) [28](vii) verbal communicators who are able to produce an 'ah' vocalisation (viii) language: have the ability to produce at least 3-4 worded utterances (ix) speech: overall speech intelligibility must be greater than 30% and/or have a diagnosis of not greater than 'moderate to severe' dysarthria (x) demonstrated high compliance with previous speech pathology interventions (as pre-determined by parent and/ or speech pathologist) (xi) ability to maintain independent head control (xii) hearing within normal limits/ no dual diagnosis of hearing impairment.

Exclusion criteria: (i) intellectual ability is greater than mild impairment (ii) presence of additional vocal pathology e.g. vocal nodules (iii) history of hoarse voice (iv) inconsistent drooling (periods when drooling is not present) (v) currently on medications that cause drooling e.g. clonazepam (vii) currently receiving other treatment for drooling e.g. block of oral sensory motor therapy.

2.3. Procedure

Ethical approval for this study was received from the Australian Catholic University (ACU) (ref: 2018-142H) and the Cerebral Palsy Alliance (CPA) (ref: 2018-08-03). The six participants will be recruited in two samples of three participants (n=6). Concealed randomisation will be used to allocate each participant to one of three treatment baseline conditions: a baseline length of 10 days, 14 days or 18 days, which means the starting point of intervention will be staggered, and different for each participant. All participants will begin the baseline phase on the same day. A concurrent design enhances internal validity by controlling for maturation effects and minimizing environmental influences [1, 29].

Advertisements in the monthly newsletter, together with the participant information letter and consent forms, will be sent to the Cerebral Palsy Alliance (CPA) and to the New South Wales (NSW)/Australian Capital Territory (ACT) CP Register for distribution. Interested parties will be invited to contact the lead researcher. If needed, the advertisement will also be sent to schools for children with developmental disability in the Sydney metropolitan area following relevant approvals. Once interest is solicited, the lead researcher will email or mail the participant information letters and consent forms for participants and their parent(s)/ caregiver(s) to the interested families. For those participants not able to provide written informed consent, verbal assent will be obtained from the participant and documented; and the parent/ guardian will be asked to complete an informed consent form. Following brief telephone screening, individuals who are not eligible will also be informed by the lead researcher over the phone.

Each data collection and treatment session for each participant will be audio- and videorecorded**.** Quantitative data on drooling, swallowing and speech will be collected for each participant in three phases; (1) baseline (2) intervention and (3) follow up. Five repeated measures of drooling will be taken; and three repeated measures of each of the swallowing and speech variables will be taken in each phase. An assessor (research assistant) who is independent of the practitioner will collect all data on primary and secondary outcomes (See Fig. 2 below). The interventionist will be a speech pathologist who is trained in both LSVT LOUD® and the online treatment version (LSVT e- LOUD®). A daily treatment protocol sheet from the LSVT LOUD® treatment manual will be used as a guide in each treatment session, whereby each ingredient is checked off against the protocol to capture data on what was implemented.

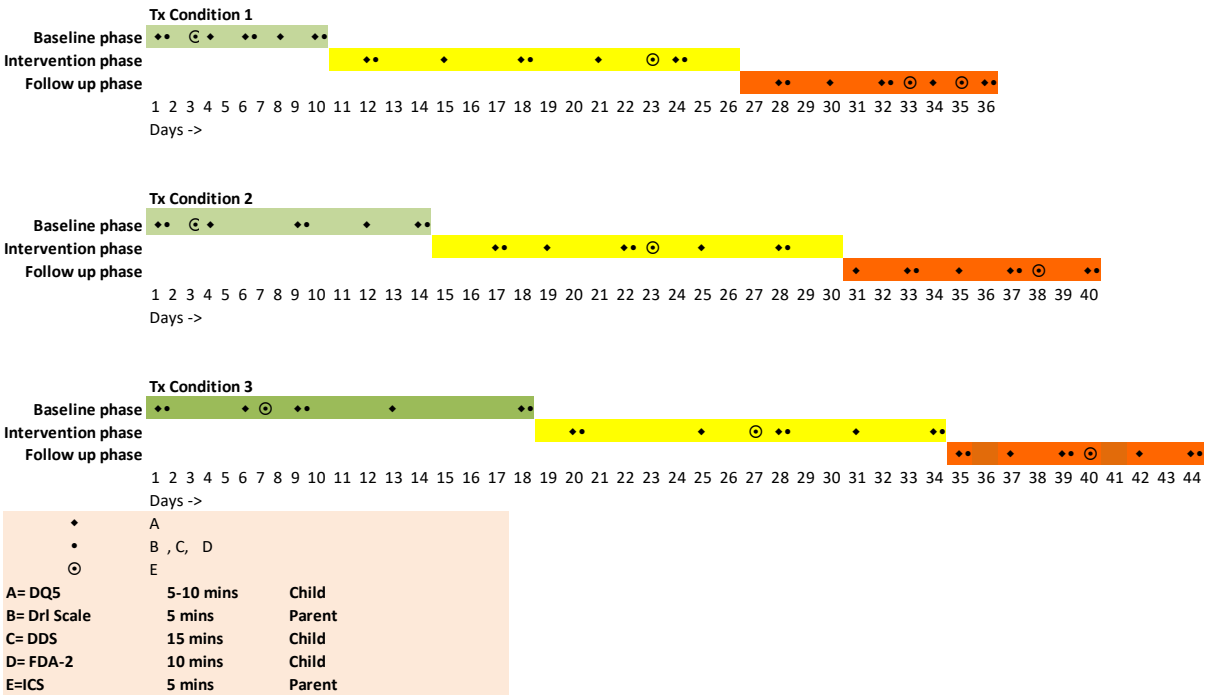


Fig. 2. Example of data collection plan for one participant

3. Dependent variables and outcome measures

Detailed baseline data related to drooling, speech and swallowing will be collected after enrolment and prior to commencement of the baseline data collection. After participants are enrolled in the

study, a case history form will be sent to each parent to complete and they will be asked to return it to the lead researcher. The case history questionnaire was created by the lead researcher and included questions about the child's relevant medical history and demographic details. The questionnaire content is also based on the following relevant measures: The Saliva Control Assessment form [30]; The Drooling Frequency and Severity Scale [31]; Communication Function Classification System [28]; and The Australian Therapy Outcome Measures (AusTOMs) [32]. A follow up phone call will be conducted with each parent to verify responses on the case history form and facilitate accuracy in the recording of baseline data.

The primary dependent variables in this study are *anterior drooling*, or the anterior spillage of saliva beyond the lip margin; and *drooling impact*, or the influence of drooling on the individual's life as perceived by a parent. Secondary dependent variables are related to speech intelligibility and swallowing. Single-word speech intelligibility is defined as the perceived clarity of speech at a single word level; sentence intelligibility, clarity of speech at sentence level, and conversational speech intelligibility referred to clarity of speech at a conversational level. Swallowing and feeding competency is defined as the task components of oral preparatory, oral and pharyngeal phases of swallowing.

Drooling Quotient 5 ($DQ5^A$)[33]: Measure of Drooling Severity (Body Functions & Structures (BFS) Level): The $DQ5^A$ is an objective valid measure of drooling frequency and severity for children with CP and other developmental disabilities. It is an observational tool whereby a trained observer records the number of times the individual drools whilst they are undertaking an activity e.g. building a block tower, reading out loud. It takes 5-10 minutes to complete. The $DQ5^A$ has been demonstrated to be a sensitive and specific measure of drooling severity (0.61 and 0.75 respectively using Youden Index[1]*). A trained observer records the number of times the individual drools in a five-minute period whilst they undertake a preferred activity in a sitting position (e.g. building a block tower, reading out loud). Drooling episodes within the five-minute period are counted as present when saliva is visualised beyond the lip margin. A score of '1' is given for episodes where drooling occurs and '0' for no drooling. A drooling 'quotient' score is expressed as a percentage of observed drooling episodes (intervals with new saliva) and the total number of intervals (0 = no new saliva, 100 = 100% of the intervals new saliva) [33]. The Drooling Quotient has good test-retest reliability in children who have stable drooling with the intra-rater intraclass correlation coefficient (ICC) reported to be >.86 [33]..

The Drooling Impact Scale (Drl Scale)[34]: The Drl Scale[34] is a subjective measure of the impact of drooling in children with neurological conditions. It is a parent-completed questionnaire consisting of 10 items, to rate the degree to which drooling has affected their life over the previous week on a 10-point scale (1= not at all, 10= constantly). Examples of questions include '*How much skin irritation has your child had due to drooling?*'. It takes 5 minutes to complete. The Drl Scale is valid, reliable and responsive to change in children with developmental disabilities who have undergone saliva control interventions. It has demonstrated good face, content and construct validity, with significant correlations between scores and carer's global ratings of change (0.69, $p<0.001$). It has good test-retest reliability in children who have stable drooling [34] with the ICC reported to be 0.95 [35].

[1] The Youden Index is the likelihood of a positive test result in persons with the condition versus those without the condition. It combines sensitivity and specificity into a single measure and has a value between 0 and 1 (a score of 1 being the perfect score)

***Dysphagia Disorders Survey (DDS*) [36] (BFS & Activity Level):** The Dysphagia Disorders Survey (DDS) [36] is a standardized screening tool of swallowing and feeding disorders in children with developmental disability. The individual is observed having something to eat and drink, and the certified clinician completes a 15-item survey on their swallowing and feeding skills. The DDS takes 10-15 minutes to complete. For each item, the clinician rates 0 for 'good function' or '1' for deficient function (definitions of what constitutes 'good' and 'deficient' are provided in the manual). This provides a numerical score and percentile rankings for swallowing and feeding abilities in children and adults with developmental disability. The DDS [36] has strong internal consistency, with a Cronbach alpha score of .89 for part 2, and demonstrates high-sensitivity, specificity, positive and negative predictive values [36]. The DDS meets the criteria for face, content, convergent and construct validity. Inter-rater reliability is considered to be good with an average ICC of .97 [36].

***Frenchay-Dysarthria Assessment-2nd edition (FDA-2) [37]*:** Perceptual assessment is central to the evaluation of speech outcomes [38]. Data on speech intelligibility of untrained spoken items (single word, sentence and conversational level) will be collected. The intelligibility subtest from the FDA-2 [37] was chosen to assess intelligibility at a single word, sentence-level and conversational level. This was chosen as a) no validated measure was available for both age range of participants and diagnosis b) stimulus items in subtest are phonetically balanced to provide a more reliable sample of speech intelligibility and c) previous research completed with children with CP has shown the subtest to be a feasible measure [39]. In assigning a perceptual rating, the clinician then judges the person's intelligibility at each level using a 5-point Likert scale. This scale will also be used to judge the intelligibility of trained speaking items (phrase/ sentence level intelligibility) before, during and after the intervention. The validity and reliability of the FDA has been established by several studies with diagnostic groups with differing neurological conditions, for example, with stroke [40]. The FDA-2 has good inter-rater reliability agreement with an average ICC of 91% (.91)[37].

4. Data Analysis

In a SCED, data are evaluated by comparing an individual's performance during the baseline with performance during the experimental phase [41]. A systematic visual analysis is the traditional method of interpreting the effects of the intervention studies using SCEDs [1, 42], and is considered the gold standard for assessing quantitative procedures [43, 44]. Data collected on drooling (frequency and severity), swallowing (oral preparatory, oral and oropharyngeal stages of swallows) and speech (word, sentence, conversational intelligibility) measures will be entered into SPSS and plotted on graphic displays e.g., line graphs. Five visual inspection criteria and two supplemental statistical methods will be completed to facilitate a systematic visual analysis and will include the evaluation of level, trend, variability, immediacy of effect (see Table 1 below). This systematic examination of graphed data enables the level of functional relationship between the manipulation of an independent variable and a change in the dependent variable to be assessed. It also facilitates judgment as to whether there is strong, moderate, or no, evidence for a treatment effect on drooling, swallowing and speech**.** Results on all measures will be tabulated and descriptive statistics including measures of central tendency will be presented for each of the three measures.

Table 1. Visual inspection criteria

Criterion	Definition
Stability	Baseline data for each behaviour describe the current level of performance and predict future performance, and it is important that baseline data are relatively stable. Little variability and the absence of trend indicate a stable rate of performance. The stability envelope will be calculated for each outcome in each phase. For stability, 80% data must fall within 25% of median value for that phase [45].
Changes in means across phases	Mean rate of the behaviour shows a change from phase to phase in the expected direction.
Change in trend (slope)	Direction of slope changes from phase to phase, for example, no slope (horizontal line) in baseline and an accelerating/ increasing slope during intervention phase indicates a change in behaviour with introduction of the intervention. Trend also refers to rate of change/ progress.
Shift in level	A level refers to the change in behaviour (dependent variable value) from the last day of one phase, e.g. baseline, and the first day of the next phase, e.g. intervention. An abrupt shift facilitates interpretation of an effect from treatment.
Latency of change	Period of time between the onset or end of one phase and changes in performance (e.g. from baseline to intervention). The closer in time that behaviour change occurs after the conditions have been altered, the easier it is to attribute the change to the intervention.

4.1 Further data analysis methods

Two non-overlap indices methods, the Two Standard Deviation Band Method and the Nonoverlap of all pairs (NAP) (for phases A + B), are additional data analysis methods chosen to support and facilitate an accurate interpretation of findings [46]. These indices are used when a change in behaviour is determined from an initial visual analysis as being potentially significant. The ***two standard deviation band method*** is used when there is baseline stability, a difference in means, and either a change in level or trend between the baseline and intervention phases. To complete a two-standard deviation band, the mean and standard deviation from the baseline phase data will be computed, the "band" representing two standard deviations (2 *SD*s) will be drawn around the baseline mean onto the graph, and the intervention phase data points that fell outside the "band" will be determined. A rule of thumb offered is that if at least two consecutive intervention phase data points fall outside the band; a statistically significant change is observed, as the probability of this happening by chance is less than the criterion of $p<.05$ [47].

NAP is a nonparametric technique for measuring nonoverlap or dominance for two phases. There are a number of reasons why the NAP was chosen as the additional data analysis method. NAP is included in robust standards for evaluating single case research [48] and is a strong indicator of performance change between phases [49, 50]. Importantly, the NAP index has also been chosen as it requires neither well-conforming data, for example, baseline stability, nor large data sets, rather it is based on the relative standing of individual data points [46]. As drooling can be highly variable in nature [35], the researcher was aware of the possibility that baseline instability may occur for some of the participants. Specifically, NAP is deemed a better indicator of performance change compared to other non-overlapping indices such as percentage of non-overlapping data, as it is a 'complete' nonoverlap index due to individually comparing all baseline with intervention data points (nA x nB points) [46]. In addition, NAP is relatively easy to calculate by hand and is known to be less prone to human error compared to other hand-calculated indices such as percentage of data points exceeding the median [50].

NAP will be calculated for all outcomes for all participants. NAP will be particularly useful if there is a potential effect in the absence of baseline stability or uncertainty about an effect, and to calculate the size of the effect detected. NAP is score of probability, whereby 'a score drawn at random from the treatment phase will exceed (overlap) that of a score drawn at random from the baseline phase' [50], with scores typically ranging from .5 to 1. If datapoints from the baseline and treatment phases cannot be easily differentiated, there is a 50% chance that scores from one phase will exceed those of the other, and it is recommended in this case, that NAP scores be re-scaled from 0-100 [51]. NAP scores range from 0 to 1 and can be interpreted as small (<.65), medium (.66 to .92) or large (>.93) [50] (see supplementary appendix for worked example of NAP calculation).

5. Discussion

Anterior drooling is common and significantly affects the health of children with CP. Behavioural intervention is a sound theoretical non-invasive approach for treating drooling but supported by low-level research evidence. High-quality experimental studies on the effectiveness of interventions to treat anterior drooling are urgently needed. This paper describes a study protocol for the evaluation of an intensive voice treatment, LSVT LOUD®, targeting drooling, speech, and swallowing in children with CP using a concurrent multiple baseline SCED. To our knowledge, there is no previous research

evaluating the effects of LSVT LOUD® on anterior drooling in children with CP. Our hypothesis is that LSVT LOUD® will reduce anterior drooling in children with CP. The assessment of methodological quality using the (RoBiNT) Scale [1] directly informed the conceptual development and planning of this intervention study. It will facilitate addressing some of the methodological pitfalls of previous research to optimise the internal and external validity of the intervention study including providing relevant baseline characteristics, blinding outcome assessors and optimising sampling of data.

SCEDs are a useful alternative experimental design choice when evaluating intervention effectiveness [42, 52, 53]. SCEDs are advantageous when compared to group-level designs for numerous reasons. This experimental research design is very suitable when there is little known about an intervention's effects and can provide a more accurate assessment of the impact of an intervention for each individual [54, 55]. SCEDs are useful when conducting research with low-incidence heterogeneous populations and can be delivered at relatively low-cost. Importantly, when implemented rigorously, SCEDS can also provide level 1 evidence regarding an intervention's effectiveness [56]. Collectively, these reasons highlight the SCED to be a worthwhile and valid method of testing the effectiveness of interventions. It is anticipated that this protocol will assist other researchers who are using a SCED method to evaluate the effects of interventions in early phases of clinical outcome research.

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Declaration of competing interest

The authors declare that there is no conflict of interest.

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