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Generalization and maintenance of a self-management program for drooling in children with neurodevelopmental disabilities: A second case series

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ABSTRACT

In this case series (n = 10) with a non-concurrent multiple baseline design, a self-management program was shown to be effective during inpatient training in eight participants with oral-motor problems and normal intelligence or mild intellectual disabilities. They were taught to perform a self-management routine and to remain dry for increasing time intervals. In addition, the program provided differential (self-) reinforcement of swallowing, controlling and wiping behavior, explicit formulation of motivational factors, instruction and feedback for parents and teachers, and continued practice after discharge. Generalization of the initial effect to the school setting was demonstrated in seven participants. Follow-up assessments demonstrated maintenance of positive results in four children up to 24 weeks, and for two other participants until 6 weeks after discharge. These results appeared to be relevant to parents and teachers. Teaching parents and teachers to implement the procedure enables them to support the child without professional supervision in the case of relapse.

Introduction

Many children with developmental disabilities do not develop adequate saliva control. Drooling (or sialorrhea) occurs in 37–58% of children and youth with cerebral palsy (CP)\textsuperscript{1} and is generally associated with limited oromotor control as a result of muscle incoordination and sensory perception difficulties rather than excessive salivation (except for children with dyskinetic CP due to hyperkinetic oral movements\textsuperscript{2}). Chronic drooling can have significant medical and psychosocial impact on children and their families,\textsuperscript{3} such as skin irritation and breakdown, damage to toys, books and equipment, frequent clothing change, and social embarrassment and isolation. A number of treatment strategies are available, such as oromotor and orosensory strategies, behavioral treatment, oral appliances, medication, and surgery, which have different levels of evidence.\textsuperscript{1} Behavioral treatment for drooling may be considered before other treatments like medication and surgery (which may have negative side effects or are invasive) are implemented. However, empirical studies on behavioral procedures are scarce. After reviewing 19 studies including behavioral management programs published since 1970, Van der Burg and colleagues\textsuperscript{4–5} concluded that treatment procedures applied for drooling are: (a) instruction, prompting and positive reinforcement; (b) negative social reinforcement and decelerative procedures; (c) cueing techniques; and (d) self-management procedures. Although these procedures yield positive results, critical examination of experimental design of the studies revealed several methodological shortcomings. Since then, new microswitch-based techniques were developed for persons with severe/profound intellectual and multiple disabilities by Lancioni and colleagues.\textsuperscript{6–12} In their case studies, a consequent behavioral technique is used in which the wiping response of the participant activates a microswitch that automatically triggers visual, vibrotactile, and/or auditory (music) stimuli. These reinforcing stimuli promote an increase of the wiping response and lead to a reduction of the effects of drooling.

For a small group of children with oral-motor problems and normal intelligence or mild intellectual disabilities, self-management procedures for chronic drooling may be applicable. The literature on self-management procedures for drooling contains two case studies with positive results.\textsuperscript{13–14} Because rehabilitation services in our country did not offer such a specialist intervention and we were not able to send trainers to the children’s homes and schools, we developed a center based self-management program for saliva control. This program was evaluated in a case series study with 10 participants (aged between 7;0 and 19;9 years; developmental age above 6 years) with developmental disabilities and severe drooling.\textsuperscript{15} Participants were taught to perform self-management skills (i.e., self-instruction [“swallow frequently, and wipe your mouth/chin dry if necessary”], self-monitoring, self-evaluation, and self-reinforcement) and to remain dry for increasing time intervals, starting from ½, 2, 5, 10, etc. minutes to a maximum of 60 minutes. Following treatment, all participants remained dry for intervals of 30 to 60 minutes while being engaged in daily activities. For three participants, maintenance of treatment effect was established at 6 and 24 weeks. Seven participants did not maintain self-management skills at follow-up. Although the self-management procedure...
showed promising results, it was concluded that further adaptations were required to improve the efficacy, generalization, and maintenance.

If no specific actions are planned to promote generalization or maintenance, the intervention is characterized as “train and hope.” However, generalization and maintenance of behavioral change can be intentionally fostered by incorporating specific strategies into the treatment procedure. These strategies to enhance generalization may be grouped into those affecting (1) settings (i.e., training in natural settings, sequential modification, modification of the environment), (2) consequences (i.e., introducing naturally maintaining consequences, recognizing indiscriminable contingencies, training to generalize, reinforcing unprompted generalization), (3) antecedents (i.e., program common stimuli, training sufficient exemplars, training multiple exemplars, general case instruction) and (4) other strategies (i.e., train loosely, mediating generalization, functional equivalence training). These single strategies can also be combined into a comprehensive program. Taking this in consideration, we elaborated our original self-management program and included: (1) differential (self-)reinforcement of, respectively, swallowing, controlling and wiping behavior, (2) explicit formulation of personal motivational factors by the child at the start and during intervention to support intrinsic motivation; (3) extensive instruction and feedback for parents and teachers, and (4) formal practice sessions in natural settings after discharge from the center, depending on the degree in which the child succeeds in spontaneous generalization and maintenance of the self-management routine. In this study, treatment effects of the modified self-management program for drooling with a special focus on generalization across persons, settings, and time (maintenance) were evaluated.

Table 1. Participants (n = 10).

<table>
<thead>
<tr>
<th>Participant</th>
<th>Sex</th>
<th>Age (years; months)</th>
<th>Diagnosis</th>
<th>GMFCS</th>
<th>Setting</th>
<th>CFCS; Communication mode</th>
<th>Pre-baseline TDS</th>
</tr>
</thead>
<tbody>
<tr>
<td>A</td>
<td>male</td>
<td>9;7</td>
<td>CP extra pyramidal syndrome, dyskinetic movement impairments</td>
<td>III</td>
<td>Special school for physical and multiple disabled children</td>
<td>II; speech</td>
<td>3-4</td>
</tr>
<tr>
<td>B</td>
<td>male</td>
<td>14;2</td>
<td>CP unilateral spastic movement impairments right, strabismus convergens, psychomotor retardation</td>
<td>II</td>
<td>Special school for intellectual disabled children</td>
<td>III; speech</td>
<td>≥3</td>
</tr>
<tr>
<td>C</td>
<td>male</td>
<td>8;2</td>
<td>CP bilateral ataxia</td>
<td>III</td>
<td>Special school for physical and multiple disabled children</td>
<td>III; speech</td>
<td>4</td>
</tr>
<tr>
<td>D</td>
<td>female</td>
<td>9;7</td>
<td>CP mild unilateral spastic movement impairments right</td>
<td>II</td>
<td>Regular/Mainstreamed school</td>
<td>II; speech</td>
<td>3</td>
</tr>
<tr>
<td>E</td>
<td>female</td>
<td>6;7</td>
<td>CP bilateral spastic movement impairments, right &gt; left</td>
<td>II</td>
<td>Regular/Mainstreamed school</td>
<td>II; speech</td>
<td>4-5</td>
</tr>
<tr>
<td>F</td>
<td>male</td>
<td>8;10</td>
<td>CP bilateral spastic movement impairments</td>
<td>V</td>
<td>Special school for physical and multiple disabled children</td>
<td>III; AAC (electronic device)</td>
<td>3-4</td>
</tr>
<tr>
<td>G</td>
<td>male</td>
<td>8;11</td>
<td>Bilateral Perisylvian syndrome (Worster Drought syndrome)</td>
<td>I</td>
<td>Special school for speech and language disabled children</td>
<td>III; AAC (signs)</td>
<td>3-4</td>
</tr>
<tr>
<td>H</td>
<td>male</td>
<td>13;4</td>
<td>Psychomotor retardation, symptomatic epilepsy</td>
<td>I</td>
<td>Special school for intellectual disabled children</td>
<td>II; speech</td>
<td>3</td>
</tr>
<tr>
<td>I</td>
<td>female</td>
<td>5;11</td>
<td>CP quadriplegic mild dyskinetic movement impairments</td>
<td>I</td>
<td>Regular/Mainstreamed school</td>
<td>I; speech</td>
<td>3</td>
</tr>
<tr>
<td>J</td>
<td>male</td>
<td>10;6</td>
<td>Psychomotor retardation</td>
<td>I</td>
<td>Special school for physical and multiple disabled children</td>
<td>II; speech</td>
<td>4</td>
</tr>
</tbody>
</table>

CP: Cerebral Palsy.
GMFCS: Gross Motor Function Classification System
CFCS: Communication Function Classification System
AAC: Augmentative and Alternative Communication.
TDS: Teacher Drool Scale

**Methods**

**Participants**

Ten children participated in this study between 2008 and 2012. Inclusion criteria were: (a) severe drooling defined as a score of 3 or higher on the Teacher Drool Scale (TDS; see subscript Table 1), indicating at least “occasional drooling, intermittent all day,” (b) a developmental age of 6 years or higher, (c) some overt awareness (i.e., comments of the participant) of practical and social adverse consequences of drooling, (d) the ability to close their mouth and swallow on demand, (e) the ability to check and clean their mouth/chin, (f) the ability to sit in an upright position in a (adapted) chair, (g) no uncontrolled (epileptic) seizures, (h) no severe aggressive or hyperactive behavior, and, finally, (j) no medical treatment for drooling in the 6 months preceding participation in the present study. In addition, parents and teachers had to be willing and able to practice with the child at home or at school after inpatient intervention. All participants had a history of oromotor and orosensory training to improve saliva control without satisfying results. Prior to inclusion, all participants were evaluated for the absence of excessive saliva production (as a result of medication or gastroesophageal reflex (GERD)) and oropharyngeal or esophageal lesions. In addition, functional and pharyngeal phases of swallowing were checked and it was confirmed that all participants were able to swallow their saliva and had safe swallowing (no risk of aspiration). Participants did not receive other treatments during the intervention phase. As chronic drooling was a problematic and stigmatizing condition to both parents and participants, they actively sought help and agreed upon participation in our inpatient program. The Human Research Committee (CMO Regio Arnhem-Nijmegen) had approved the study and...
informed consent was obtained from parents. For demographic data of participants, see Table 1.

**Setting and materials**

Participants were admitted to the child rehabilitation center for children with physical and multiple disabilities at the Child Rehabilitation Department of Sint Maartenskliniek (Nijmegen, the Netherlands) for three weeks. Treatment and data collection were conducted in a separate, child friendly decorated room with a table and two chairs and enough space for an (electric) wheelchair. Present on the table were a mirror, a box of tissues, a so-called “swallowing-report” and stickers to document the progress of the child during the treatment, a flowchart of the procedure, and school and play materials. In addition, a computer with internet was available. Parents and teachers were asked to bring school and play materials that enabled the trainer to present adequate training activities to the child, activities that were similar to his/her daily activities at home and at school. For data collection, a digital video camera, a stopwatch and a note-book were at hand. In an advanced phase of the intervention, treatment was occasionally conducted during activities close to our center.

**Dependent variable**

Drooling was defined as saliva (either a drop or a string) present beneath the lower lip line or falling from the mouth for a period longer than 2 seconds without the participant cleaning his/her face and/or clothes. The dependent variable was latency in minutes of being dry (non-drooling) while performing daily activities. Specific activities were chosen from the child’s current school and leisure activities and at random selected for recording sessions. Latency recordings were scheduled throughout all phases of the study. During the treatment phase, latency recordings were not scheduled within half an hour after a meal. Prior to the latency recording, the participant was asked to wipe his/her chin or mouth dry. After the participant cleaned his/her chin or mouth, the stopwatch and video recording was started by the trainer. As soon as drooling was observed, the stopwatch was stopped and the latency score was written down in the note-book. Video recording was terminated without any comment to the participant who continued the activity for several minutes until there was a natural break.

**Design and data collection**

Participants were consecutively admitted to the rehabilitation center. Data were collected in a non-concurrent multiple baseline design across individuals (i.e., data collection across participants did not overlap and participants were randomly allocated to different baseline lengths). The present study design consists of 4 experimental phases: (a) baseline, (b) self-management treatment, (c) generalization, and (d) follow up with data collection at 6 and 24 weeks and intermediate phone contact with the trainer after 3 and 12 weeks. Baseline recordings (a) were initiated at different points in time during the first two days of admission to the center. The number of baseline recordings varied from 8 to 14 and lasted maximum 15 minutes to be able to gather latency scores while the child performed different kind of activities in the course of one or two days. After visual analysis showed a stable or decreasing trend in baseline latency scores, self-management treatment (b) was started on the second or third day of admittance to the center. Three one-to-one sessions of 90 minutes were scheduled daily. Each session contained one or more training trials and one latency recording. The number of training trials in each session depended on the length of the time interval that was set by the trainer. The maximum duration of the latency recording was equal to the interval length trained at that time. At first, a latency recording was scheduled at the end of each session. When the participant could remain dry for 30 minutes or more, latency recordings were only scheduled at the end of the last training session of the day to ensure that sufficient time for training remained. Generalization (c) of treatment effect to the participants’ natural context (i.e., school) was assessed in the week following discharge from the center, during 4 trials scheduled on one day. This generalization assessment was repeated at 6 and 24 weeks after discharge (d: follow up). The maximum duration of the latency recording during generalization and follow up equaled the maximum time interval the participant remained dry during initial treatment. At 3 and 12 weeks after discharge the trainer contacted the parents and the teacher by phone and discussed the current status of drooling and the child’s performance of the self-management routine, without formal data collection.

**Reliability**

Reliability of latency recordings was determined for each participant by a second rater, who independently but not simultaneously, scored 18% of all videotaped sessions in a random order, equally distributed across the baseline, intervention and post intervention phase. This independent observer was not familiar with the participants, nor did she know in which phase of the study the videotape was taken. Intra-class correlation (SPSS 21: ICC = 2,1) (two way random model, absolute agreement, single measures) was computed for all reliability checks (overall), and in addition for each participant and across each experimental phase. Overall ICC was .795. The ICC for participants were: A .999; B .969; C .824; D 1.000; E .508; F .122; G .999; H .991; I .104; J .924. The ICC’s for experimental phases were respectively: .755 (baseline), .678 (intervention), and .818 (post intervention: .518 (generalization), 1.000 (6-weeks follow up) and .987 (24-weeks follow up)).

**Procedure**

During latency recordings and training sessions the participants performed daily activities such as schoolwork, as well as leisure activities such as playing outside (football, running, swinging), watching television or playing computer games. After baseline recordings and before the start of intervention, the participants’ personal reasons for participation in the intervention (motivating factors such as “If I manage to stay
dry, my schoolwork will not get wet” or “... I will be allowed to participate in cooking class”) were formulated on a so-called “motivation list” in the swallow report. Occasionally, additional comments on this list were made during intervention. In the case of reduced motivation during the course of the training, the trainer reminded the participants of their personal reasons for participation by showing and talking about this list. The intervention consists of three phases, each phase covering about one week.

**Phase 1.** During the first phase the participants were taught to perform self-management skills and to remain dry for increasing time intervals. Pre-set time intervals were 1/5, 1/2, 5, 10, 15, 20, 25, and 30 minutes. The first training interval started with a time interval just below the mean latency score during baseline, with a minimum of 30 seconds and a maximum of 5 minutes. If the participant succeeded to remain dry during 3 consecutive trials, the next larger interval was set. If the participant failed to remain dry during 3 consecutive trials or during 5 trials in sum, the previous interval was in effect again until the participant remained dry in 3 consecutive trials.

The instruction procedure for the use of self-management skills for saliva control was adopted from Van der Burg et al. At the beginning of the first training session, the trainer told the child that drooling is not an acceptable condition and that he/she needs to practice to swallow, check the chin and wipe if it is wet, to remain dry. Then, the trainer gave the instruction: If you are dry, we can start. In subsequent trials, this was the first instruction. If the child exhibited self-control (i.e., checked if there was any saliva on the mouth or chin) and -if necessary- self-care (i.e., wiping), positive verbal feedback was given. If not, the trainer gave the instruction: Wipe your chin, so we can start. After the child wiped his/her chin with a tissue, a positive remark was given. If not, the trainer wiped the child’s mouth and chin dry with a tissue, while giving negative verbal feedback: A wet chin is not nice, and you know that. It’s not good if you do not clean your chin. Following, a procedure of positive practice was administered: Your chin is dry now. Look in the mirror. This is great! To prevent your chin from getting wet, you must swallow. Let’s practice this three times together... Good! Check your chin, it feels dry, and it looks good! Remember to swallow to keep your chin dry. When you feel you are wet, wipe your mouth and chin immediately and swallow. To enhance self-instruction, the trainer asked: So, what do you have to do to stay dry? Then, the trainer initially guided the child to say: Swallow, check my chin, and wipe. Eventually, the child was taught to use this phrase during the training sessions: first aloud, then whispering, and finally in inner speech. After instruction, the trainer announced the length of the next time interval and set the stopwatch.

The child was verbally praised and received a sticker in his/her swallow-report if he/she remained dry during an entire trial. This report was shown daily to significant others, like parents, ward staff, and visitors. If drooling occurred during the interval without the child wiping the chin within 2 seconds, the trainer immediately interrupted the activity and made the child look in the mirror saying: Look, your chin is wet, that’s not nice. What a pity. If the child was non-cooperative and/or underachieving compared to former performance, a negative remark was added: You did not try hard enough. A sticker was then denied, and a cross was marked in the swallow-report. Immediately hereafter, the positive and supportive tone of the interaction was restored and the next trial started with the instruction: If you are dry, we can start.

**Phase 2.** The second phase of the intervention started once the child was familiarized with the self-management routine and remained dry for at least 10 minute intervals. During the morning sessions in phase 2, the procedure of phase 1 was continued. When the child succeeded to stay dry for 30 minutes, the time interval was extended stepwise with 5–15 minutes up to a maximum of 60 minutes, depending on the child characteristics and personal goals. In addition to verbal instruction, a script of pictographs depicting the self-management routine was introduced for E, F and G in this phase. Initially, they were guided by the trainer through self-instruction according to this script, and as they learned to instruct themselves, the instruction by the trainer was gradually faded.

In the afternoon sessions in phase 2, differential evaluation and reinforcement on the performance of the self-management routine (i.e., swallowing, controlling the chin and wiping) was added to the procedure to be sure that the participant executed all elements of the self-management routine adequately and frequently enough. Performance of the self-care skills was judged by criteria. The criterion for swallowing was 1 to 2 times per minute, based upon the results of Senner et al. who reported an average frequency of swallowing in children with CP who did not drool of about 21 (with a maximum of 32) times per 20 minutes. The criterion for wiping was wiping the chin, mouth or clothing within 2 seconds after saliva was below the lower lip or directly falls from the mouth. The criterion for checking the chin was 2 times per 15 minutes. After each trial, the child evaluated the performance of each self-care skill. If the performance met the criterion, the child registered a green dot; if it did not, he/she registered a red dot and if the performance was “in between” an orange dot was marked in the swallow-report. In case there was no need to wipe because the child stayed dry, a green stripe was marked. Verbal feedback was given accordingly. First, the trainer guided the child through this evaluation and self-registration procedure. Then, the child was asked to evaluate his/her own performance with feedback from the trainer, and eventually the child performed this procedure without help. To prevent over- or underestimation by the child, the opinion of the trainer about the implementation of the self-care skills was decisive.

**Phase 3.** During the third phase, the child performed the self-management routine and self-care skills adequately and independently. The self-management routine was established by continuous repetition. The trainer monitored the correct implementation of the routine, and supported the child by giving positive feedback in the case of success and negative feedback and additional instruction if the child failed. During the afternoon session the time interval was gradually extended. Eventually, the child was taught to whisper self-instruction or use internal speech; this way the self-management routine will not be disturbing in the child’s natural environment after discharge.
Parent and teacher training

After baseline, parents and ward staff were instructed to give occasional positive feedback during the day, contingent upon the child swallowing, being dry, or performing self-care related to drooling. During the (two) weekends, the training was interrupted as the child stayed at home. To prevent relapse and to foster generalization, the child was instructed to practice at home during the weekend, under supervision of his/her parents. Therefore, parents were instructed by the trainer during the last training session of the week. After phase 1, they received verbal and written instructions on the behavioral techniques (such as instruction, feedback, positive reinforcement) that were used during the treatment for drooling. After several trials of modeling of the procedure by the trainer, parents practiced the instructions, feedback and reinforcement during one or more trials with their child. Afterwards, the trainer gave parents verbal feedback on their performance. During the weekends, parents instructed their child to stay dry two or three times a day for a time interval somewhat shorter than was currently trained at the center, so that the child could easily be successful. The parents gave a sticker in the swallow-report if the child succeeded. If not, parents were instructed to mark a cross in the swallow-report, but to refrain from negative verbal feedback. Parents were asked to videotape one trial at home. After each weekend, the trainer and the parents evaluated this videotaped trial together and feedback on the child’s performance and on the parent’s adherence to the training procedure was given.

At the end of phase 2, parents also learned to implement the differential evaluation and reinforcement of the self-care skills. During the next weekend, parents practice the differential evaluation and reinforcement 2 times a day and one additional trial in which they ask their child to stay dry for a time interval that equals the time interval currently trained at the center. Parents were asked to videotape one trial in which they practiced the differential evaluation and reinforcement of the self-care skills. After the weekend, the trainer and the parents evaluated this videotaped trial together and parents received feedback on their performance.

At the end of phase 3, the self-management training was evaluated with the parents. Parents received tips and tricks for maintaining the results of the training at home. Parents were instructed to continue daily practicing at home during the first 6 weeks after discharge, implementing differential evaluation and reinforcement of the self-care skills twice a day, and one additional trial in which the child was asked to stay dry for the maximum time interval achieved during the training.

After generalization assessments at school (in the first week after discharge), teachers were instructed at school in much the same way as parents were during intervention: they received written instructions on the behavioral techniques and practiced the instruction, feedback and reinforcement procedures that were first modeled by the trainer. Teachers were instructed to continue daily practicing (including the differential evaluation and reinforcement of the self-care skills) at school for the first 6 weeks after discharge from the center. The swallow-report remained to be used for registration of the execution of trials as well as for both the evaluation of drooling and the execution of the self-management routine by the child. In addition, parents and teachers were instructed to give occasional positive feedback during the day, contingent upon the child swallowing, being dry, or performing self-care related to drooling.

During follow-up, the trainer had contact with the parents and teacher twice by telephone (at 3 and 12 weeks after discharge) and visited the child’s school twice (at 6 and 24 weeks after discharge). At each contact, the trainer discussed the current status of drooling and the child’s performance of the self-management routine with the parents and teacher. If the results from the initial training at the center were maintained, the frequency of practicing at home and school was gradually reduced. If relapse occurred, the trainer advised to increase the frequency of practicing. During the school visits, the trainer observed the teacher practicing with the child and gave feedback and additional instruction if necessary.

Data analyses

After visual analysis of graphed line data, we performed statistical analyses for single subject data. To evaluate effects of treatment for each participant, the change in latency scores during subsequent phases compared to baseline was determined by calculating either Tau or Tau U23 (see http://www.singlecaseresearch.org/). These effect sizes for data from single subject designs are based on all pairwise data comparisons and examine data non-overlap between phases (Tau), while controlling for undesirable positive baseline trend (Tau U).

Social validity

In addition to objective evaluation of the self-management treatment of drooling by observational data (i.e., latency), we also investigated what parents and teachers thought about the drooling severity at home and school and if they noticed any changes in this severity. Therefore parents and teachers completed a Visual Analogue Scale (i.e., VAS) for drooling severity at baseline, and blind to their previous ratings during the generalization phase immediately following discharge, and at 6 and 24 weeks follow-up. Scores on the VAS could vary from 0 to 100 (0 = very severe drooling and 100 = no drooling).

Results

Figure 1 depicts the latency scores (i.e., number of minutes dry) during baseline, treatment, generalization, and follow-up at 6 and 24 weeks for each participant. The immediacy effect was lacking for most of the participants as the first time interval during intervention was deliberately set just below the mean baseline latency score to increase the chance of initial success which was expected to be motivating for the child. Figure 1 shows that after a three-week self-management treatment all but one (i.e., F) participants were able to remain dry for intervals of 30 to 60 minutes while engaged in daily activities. Right after discharge from the center, eight participants succeeded to stay dry during one or more generalization trials at school, resulting in equivalent latency scores
compared to the end of the intervention phase. For one participant (I), latency scores during generalization and follow-up trials relapsed, but maintained well above baseline level. At 6 weeks follow-up, seven participants (A, B, C, D, E, H, J) remained dry for their personal maximum time interval during one or more trials at school, and five of them (A, B, C, D, J) also succeeded in doing so at 24 weeks follow-up. For four participants (i.e., G, H, I, J), due to practical circumstances in the classroom and/or school program, one or more generalization and follow-up trials had to be terminated before the maximum latency could be determined (see Figure 1, these data points are marked as open diamonds). As a consequence, these latency scores underestimate the child’s ability to stay dry while engaged in daily activities.

Figure 1. Latency (in minutes) during conditions of baseline, intervention, generalization (Gen), and follow-up after 6 (FU6) and 24 weeks (FU24). Assessment during generalization and follow-up sessions in the school were sometimes disrupted because the school program changed unexpectedly.
In Table 2, individual values of Tau and Tau-U (percentage non-overlapping data corrected for positive baseline trend) for contrasts between baseline and, respectively, intervention, generalization, follow-up at 6 (FU6) and 24 (FU24) weeks are presented, including a weighted average. The contrast between baseline and intervention was significant for eight participants (A, B, C, D, E, G, I and J), indicating an increase in latency scores. For all of them, the individual weighted average scores of Tau/Tau U for all contrasts between experimental phases ranged between 0.66 and 0.99 and were significant. For four of them (A, B, C, D), the intervention effect maintained until the follow-up at 24 weeks. For two (E and J) the effect maintained until 6 weeks follow up, but Tau at 24 weeks was not significant. Although visual inspection of the learning curve of F suggests some initial effect during intervention, Tau U for the contrast between baseline and intervention (controlling for a positive baseline trend) appeared to be not significant. For G, there was an inconsistent pattern across phases after intervention. For I, the difference between the baseline and generalization phase was significant, but the effect was not maintained at follow-up assessments.

Table 3 shows the VAS scores from parents and teachers at baseline, generalization and follow-up. Unfortunately, there were many missing values. Although there are clear differences between participants, VAS scores seem to be congruent with the reported changes in latency scores. The group mean VAS scores increased between baseline and generalization with 50 points according to the parents and 60 points according to the children’s teachers, indicating a substantial reduction in drooling severity. At 6 and 24 weeks follow-up there was a decrease (teachers rated drooling more severe than parents did), but after 24 weeks there was still a mean difference of around 35 points above baseline level for both parents (34.6 points) and teachers (35.6 points), indicating that drooling severity was still below the baseline level.

**Discussion**

A self-management program for children who experience chronic and severe drooling was shown to be effective in improving latency scores. For all participants, VAS scores seem to be congruent with the reported changes in latency scores. The group mean VAS scores increased between baseline and generalization with 50 points according to the parents and 60 points according to the children’s teachers, indicating a substantial reduction in drooling severity. At 6 and 24 weeks follow-up there was a decrease (teachers rated drooling more severe than parents did), but after 24 weeks there was still a mean difference of around 35 points above baseline level for both parents (34.6 points) and teachers (35.6 points), indicating that drooling severity was still below the baseline level.

### Table 2. Individual values of Tau and Tau-U for contrasts between baseline and respectively intervention, generalization, follow-up at 6 (FU6) and 24 (FU24) weeks.

<table>
<thead>
<tr>
<th>Participant</th>
<th>Baseline – Intervention</th>
<th>Baseline – Generalization</th>
<th>Baseline – FU6</th>
<th>Baseline – FU24</th>
<th>Weighted Average&lt;sup&gt;a&lt;/sup&gt;</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Tau(-U) SD p</td>
<td>Tau(-U) SD p</td>
<td>Tau(-U) SD p</td>
<td>Tau(-U) SD p</td>
<td>Tau(-U) p</td>
</tr>
<tr>
<td>A</td>
<td>0.98 0.23 &lt;0.01*</td>
<td>1.00 0.34 &lt;0.01*</td>
<td>1.00 0.34</td>
<td>1.00 0.34 &lt;0.01*</td>
<td>0.99 &lt;0.01*</td>
</tr>
<tr>
<td>B</td>
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<td>1.00 0.34</td>
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<td>0.98 &lt;0.01*</td>
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<tr>
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<td>0.84 &lt;0.01*</td>
</tr>
<tr>
<td>D</td>
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<td>1.00 0.34 &lt;0.01*</td>
<td>1.00 0.34</td>
<td>1.00 0.34 &lt;0.01*</td>
<td>0.94 &lt;0.01*</td>
</tr>
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<td>1.00 0.37</td>
<td>0.30 0.34 0.38</td>
<td>0.80 &lt;0.01*</td>
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<tr>
<td>F&lt;sup&gt;b&lt;/sup&gt;</td>
<td>0.18 0.27 0.49</td>
<td>-0.67 0.39 0.09</td>
<td>-0.97 0.39</td>
<td>-0.36 0.39 0.35</td>
<td>-0.39 0.03*</td>
</tr>
<tr>
<td>G</td>
<td>0.68 0.25 0.01*</td>
<td>0.61 0.48 0.11</td>
<td>0.70 0.32</td>
<td>0.63 0.34 0.07</td>
<td>0.66 &lt;0.01*</td>
</tr>
<tr>
<td>H</td>
<td>0.19 0.26 0.48</td>
<td>0.23 0.32 0.46</td>
<td>-0.25 0.34 0.47</td>
<td>-0.22 0.38 0.56</td>
<td>0.01 0.94</td>
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<tr>
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<td>0.71 0.27 0.01*</td>
<td>0.67 0.32 0.04*</td>
<td>0.63 0.34 0.07</td>
<td>0.67 &lt;0.01*</td>
</tr>
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</table>

<sup>a</sup>Weighted average of four contrasts: Baseline – Intervention, Baseline – Generalization, Baseline – FU6 and Baseline – FU24.

<sup>b</sup>Corrected for positive trend in baseline.

<sup>*</sup>p < 0.05.
eight out of ten participants during initial training. In addition to the original program in which participants were taught to perform a self-management routine and to remain dry for increasing time intervals, the current program provided differential (self-)reinforcement of, respectively, swallowing, controlling and wiping behavior, explicit formulation of motivational factors, extensive instruction and feedback for parents and teacher(s), and continued practice at home and at school after discharge. Nine participants were able to remain dry for at least 30 minutes while engaged in daily activities after intervention. Immediately after discharge from the child rehabilitation center, generalization of the initial effect to the school setting was demonstrated in seven participants. In addition to amendments in the in-patient program, formal practice sessions after discharge were encouraged, tailored to the needs of the child. At follow-up assessments 6 and 24 weeks after discharge, maintenance of positive results was demonstrated in seven and four participants, respectively. Three of them preserved full effect, the other four children showed (somewhat) lower latency scores compared to the end of the intervention phase. Still, the VAS-scores were well above baseline level and the results appeared to be relevant to the parents and teachers. Two children returned to latency scores at the baseline level.

There were some limitations in the current study. Reliability checks for latency scores were taken from video recorded sessions and unfortunately not all of them appeared good enough to make accurate judgments on the appearance of saliva below the lower lip. The main reasons for this were low technical quality (i.e. backlight, instable focus), movement artifact, and hands of the child in front of their mouth and chin. This was particularly the case during generalization and follow-up latency recordings in the classroom. Nevertheless, the overall ICC was quite good and ICC’s across experimental phases and for most individual participants were good. However, for the generalization phase and participant E the ICC was moderate and for participants F and I it was low. Thus, conclusions from their latency scores and learning curves must be interpreted cautiously. Although the ICC of their latency recordings was low, participants F and I were not excluded from analysis because we did not want to ameliorate our results by leaving F out, nor did we want to exclude I because of the positive changes in VAS-scores from their parents and teacher, which may indicate some kind of change in drooling severity in daily life.

While nine participants show a stable or decreasing trend in baseline scores, E’s first latency scores are below 1 minute, but increase during her last baseline trials. One could argue that a specific intervention was not needed. However, latency scores right after the start of intervention return below 15 minutes, indicating that her drooling problem was not solved by maturation. During baseline, the child’s ability to stay dry may have been underestimated because latency scores were maximized at 15 minutes. Six participants sometimes scored on this maximum (median 2 times (range 0–8) during 8–14 baseline assessments). This might have inflated the effect sizes found in children that repeatedly reached a maximum score during baseline.

Although latency scores increased in most participants, we did not measure child correct use of self-management strategies. This is important to demonstrate a functional relation with latency scores and to allow for conclusions about the cause of poor response during the training.

Due to practical circumstances in the classroom and/or school program, one or more generalization and follow-up trials had to be terminated before maximum latency could be determined in four participants. It should be noted that this only influences the effect size Tau in case these latency scores are in the range of baseline scores. Inspection of the data shows that J has two ties (baseline and generalization scores of 15 minutes) and both G and H had one tie, while all other latency scores during disrupted trials were well above the baseline level. If latency scores during disrupted generalization and follow-up trials were above the maximum baseline score of the child, it did not affect the effect size.

In contrast to latency recordings in the post intervention phases, baseline recordings of latency were not taken in the home and school of the participant but in the center. As our participants were well aware of the purpose of their admission to the center, this may have prompted them to be more alert on saliva control and thus reduced their baseline drooling severity. VAS-scores of their parents and teachers were collected as a secondary measure to be able to compare between pre-baseline and post-treatment drooling severity. Unfortunately, there were many missing values as the compliance of parents and teachers to return post intervention VAS-data was low, despite repeated requests. Perhaps they did not recognize the importance of their written reports after exchange of detailed information in personal contact with the trainer during and after the intervention. As a consequence, the support of conclusions from this secondary measure is not entirely convincing.

In order to prevent relapse and foster generalization, parents and teachers were taught how to practice the self-management routine at home and at school in order to support the child whenever drooling would increase again. We did not measure how much additional training the participants received at home and at school after discharge from the center. As a consequence, we cannot draw unambiguous conclusions about the cause of decreasing latency scores for some children at follow-up assessments. Several factors may have caused this, such as low intensity of standardized training during the follow-up phase. In addition, personal factors may have contributed to decreased latency scores such as the severity of the child’s oral motor problems, the child’s incorrect use of self-management strategies, difficulty in incorporating the self-management procedure in care routine at home or at school, the child’s lack of motivation to remain dry, the child’s or caregivers’ perceived lack of result or lack of motivation to practice the self-management routine, and a lack of time or opportunities to provide extra training. In spite of being taught how to do so, some parents or teachers might not have given the child sufficient support.

In a former case series, the intervention effect at 24 weeks follow-up maintained for only 3 out of 10 children while the
remaining 7 children quickly or gradually returned to baseline levels of drooling. In the present study, 3 of 9 successful participants maintained satisfying latency scores until 24 weeks after intervention. Four other children showed some decay but the effect was still large and clinically relevant for them and their parents. In two children drooling returned to baseline level at follow-up. Of course, our cohort of participants is too small to perform effective subgroup analysis and no definite conclusions can be drawn from the comparison of two case series. In addition, some essential differences between the participants of the former (first group) and the present (second group) case series existed in: 1) gross motor functioning, 2) drooling severity at baseline and 3) age-range. The median score on the Gross Motor Function Classification System in the first group was higher (median III; range I-V), indicating more severe gross motor problems than in the second group (median II; range I-V). Pre-baseline drooling severity as measured by the TDS was a little higher in the group of participants in our first study (median 4; range 3–4 to 5) compared to the second one (median 3–4; range 3 to 4–5). Finally, the first group was somewhat older than the second group (age range first group 7;0–19;9 years; second group 5;11–14;2 years). However, in both case series, most of the participants learn to perform the self-management routine and increase their latency scores, while some of them succeed in generalization and maintenance of the effects at school and at home. A clear advantage seems to be that parents and teachers in the second case series learned to implement the procedure at home and school so they can support their child to stay dry without professional supervision. In the case of a relapse of drooling, they may reinstate the procedure themselves. Additional techniques such as the introduction of automatic cueing to prompt the child at regular time intervals to perform the self-management routine during daily activities may be helpful.

In many countries, an inpatient program for drooling is not available as these kinds of low tech, non-medical interventions are not conducted in an inpatient setting. Future research should focus on implementing the self-management program for drooling in a school- and/or home-based program for children with normal intelligence or mild intellectual disabilities. This way, generalization is less of a problem because training takes place in the child’s own environment. Of course, detailed instruction and coaching of parents and teachers seems important to ensure procedural integrity and maximal compliance, and training of participating children has to be carefully planned for several weeks in their daily program at school and at home. Also, the child’s correct use of self-management strategies must be measured to demonstrate a functional relation with latency scores and to allow for analysis of poor response during the training. To support parents and teachers in implementing the self-management program in their own situation, the use of telehealth may be considered, as this is currently being introduced in swallowing evaluation and in the implementation of behavioral procedures by parents at home and intervention by teachers at school. However, not every parent will be prepared to be enrolled in support via telehealth.

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Declaration of interest

The authors report no declarations of interest.

References


