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Self-management treatment of drooling: A case series

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ABSTRACT

Behavioral treatment of drooling is advocated widely, but evidence of its effectiveness is lacking. In a center-based case-series study, 10 participants with severe drooling were taught self-management skills to reduce drooling. Following treatment, all participants remained dry for intervals of 30–60 min, while being engaged in daily activities. Generalization to the classroom occurred in each participant. For three participants, maintenance of treatment effect was established at 6 and 24 weeks. Seven participants failed to maintain self-management skills at follow-up. Although the self-management procedure showed promising results, further adaptations are required to improve efficacy, generalization, and maintenance.

1. Introduction

Drooling is a disabling condition for many handicapped children, adversely affecting their physical health, daily life and care, social interactions, and self-esteem. Although behavioral treatment is frequently advocated as a treatment option (e.g., Blasco, 2002; Brei, 2003), its evidence-base is limited (see Van der Burg, Didden, Jongerius, & Rotteveel, 2007a). In contrast to medical intervention studies on the treatment of drooling, the total number of participants in behavioral studies is small (i.e., $N < 60$) and participants vary considerably in age, and intellectual and motor disabilities. In addition, behavioral

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procedures to reduce drooling vary. Four types of procedures can be distinguished: (a) instruction, prompting, and positive reinforcement, (b) negative social reinforcement and decelarative procedures, (c) electronic cueing techniques, and (d) self-management procedures (Van der Burg, Didden, Jongerius, & Rotteveel, 2007b). Based on outcomes of case reports and case series it appears that all procedures (either alone or in combination) are effective, albeit with different long-term outcomes.

Self-management aims at controlling drooling without social and/or technical support and leads to independency of the individual. As opposed to external cueing, self-management techniques aim at teaching the individual to self-monitor and self-evaluate his/her physical appearance, to self-initiate an appropriate response and to self-reinforce both appropriate responses and appropriate physical appearance. However, data on self-management procedures for drooling are available from only two case reports. Thorbecke and Jackson (1982) taught self-monitoring and self-instruction following overcorrection (i.e., if the trainer observed the chin was wet, the girl had to swallow once and wipe her chin 10 times) to a 19-year-old girl with mild cerebral palsy and moderate intellectual disability. The procedure was administered mainly in the classroom, but was also executed by her teacher during trips and recesses in the playground. During self-management treatment, she had to check her chin every 5 min and repeat the teacher's instructions aloud. In case of a wet chin, she had to swallow once and wipe her chin 10 times, as in the overcorrection procedure. In subsequent treatment sessions she had to repeat these phrases first aloud, then in a whispering mode, and finally, silently. While overcorrection failed to produce a lasting response, the reduction of drooling remained stable after the addition of self-management training and following the fading out of the teacher's prompts. Dunn, Cunningham, and Backman (1987) used self-management for swallowing and positive reinforcement to eliminate drooling in a 16-year-old boy with severe spastic quadriplegia. His language comprehension and non-verbal cognitive abilities were estimated at 13 years. Two years prior to behavioral treatment, he had surgical rerouting of one salivary gland duct to redirect salivary flow, which failed to result in reduced drooling. The self-management procedure encompassed four steps: (a) monitoring mouth closure, (b) determining the need to swallow, (c) evaluating the effect of the procedure in terms of drooling prevention, and (d) verbally rewarding himself if successful. Each 60 s, he was verbally prompted to start the self-management procedure and was allowed to take a token each time he succeeded. Treatment was conducted in a hospital outpatient clinic one full day a week for a period of 10 weeks. After reduction of drooling, reinforcement was eliminated and prompts were gradually faded out. At school he was prompted to use the self-management routine in the classroom after the clinic treatment was completed. While the initial treatment effect remained stable at the 3 month follow-up, a booster training was necessary at 6 months after an increase in drooling. After the booster training, effectiveness was maintained for another 6 months. Although the authors only presented data for one individual, they claimed to have achieved a similar effect with a number of children ranging in age from 8 to 13 years with developmental ages ranging from 4 to 10 years, but without providing data.

Although these case reports seem promising, studies on self-management for drooling remain scarce. As a consequence, no general conclusions can be drawn about the effectiveness of self-management procedures for drooling in children with motor and learning disabilities. In the present study, the effectiveness of a new self-management procedure for drooling was evaluated in 10 children. Also, generalization of treatment effects to the classroom and maintenance were assessed. Finally, treatment effects on daily life and care, social interaction, and self-esteem of the participant were evaluated.

2. Method

2.1. Participants

Ten children participated in this study. Inclusion criteria were (a) severe drooling, defined as a score of 3 or higher on the Teacher Drool Scale (TDS; Camp-Bruno, Winsberg, Green-Parsons, & Abrams, 1989: see subscript Table 4), 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 or to ask or prompt the trainer to wipe their face dry, (f) the ability to maintain an upright seated 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. Table 1 presents demographic data of participants. Table 2 presents descriptions of five levels of gross motor function for children with cerebral palsy between 6 and 12 years, according to the Gross Motor Function Classification System (GMFCS; Palisano et al., 1997). Participants older than 12 years of age were also classified according to this system, as the observed GMFCS-level in 12-year-old children is highly predictive of adult motor function (McCormick et al., 2007). All participants had oral motor problems. They met the inclusion criterion in that they were able to close their mouth and swallow on demand, albeit with difficulty in the oral phase of swallowing. Two participants showed mild tongue protrusion during swallowing, three showed oral dyskinesia, and seven had mild or severe malocclusion, which did not interfere with treatment. The Human Research Committee had approved the study. Written informed consent was received from the parents of all participants.

2.2. Setting and materials

Participants resided at the children's ward of a rehabilitation clinic for about 3 weeks, with rooming-in for parents if necessary. Data collection and treatment were conducted in a separate room with a table and two chairs and enough space for a (electric) wheelchair. A mirror, a box with tissues, a so-called 'swallow-report' and stickers to document progress, a flowchart for the therapist, and an activity plan were present on the table. In addition, school and play materials as well as a laptop and a television with DVD-player were present. Parents and teachers were asked to provide school and play materials that enabled the trainer to present adequate training activities to the child similar to daily activities. For data collection a digital video camera, a stopwatch, and a notepad were at hand.

2.3. Dependent variable

Drooling was defined as saliva (either a drop or a string) present beneath the lower lip line or a string falling from the mouth for a period longer than 2 s without the individual cleaning his/her face and/or clothes. The dependent variable was latency in minutes of being dry (non-drooling) while performing daily activities. Latency recordings were scheduled throughout all phases of the study. The participant was not informed about the purpose of the recording. At the start of a latency recording session, the participant was told that he/she would be videotaped while performing activity X. Then, the participant was asked to clean his mouth and chin and the stopwatch was started. Video registration was terminated without any comment, 5 min after the trainer had observed that drooling occurred.

2.4. Design and data collection

Participants were consecutively admitted to the rehabilitation clinic. Data were collected in a non-concurrent multiple baseline design across individuals (i.e., [baseline] data collection did not overlap across participants and participants were randomly allocated to different baseline lengths; Watson & Workman, 1981) with six experimental phases: (a) baseline, (b) self-management treatment, (c) post-intervention, (e) generalization, and follow-up at (f) 6 weeks and (g) 24 weeks.

Baseline. Baseline recordings were initiated at different points in time. The number of baseline recording sessions varied from 10 to 14. Latency recordings were scheduled on 2 consecutive days and lasted either 10 or 15 min. After visual analysis showed a stable trend in baseline data, self-management treatment was administered.

Self-management treatment. In this phase, three one-to-one sessions of 90 min were scheduled daily. Each session contained one or more training trials, depending on the length of the time interval that was set, and one latency recording. Maximal duration of these latency recordings was equal to the interval length trained at that time. Initially, latency recordings were scheduled at the end of each session and later during this phase, when dry periods of 30 min were reached, these recordings

Table 1
Demographic characteristics

Participant	Sex (M/F)	Age (years; months)	Diagnosis	Motor Impairment (GMFCS)	Oral-Motor Status	Degree of Intellectual Disability (ID)	Communication mode	Setting	Previous treatment	Drooling Severity pre-baseline	
										TDS	VAS
K	F	11;10	CP, controlled epilepsy, coloboma right iris	IV	OD, S&TD	Mild; IQ 64 at age 11;9	Non-verbal, voice computer	Special school for physical and multiple disabled children	OMT, BoNT	—	—
D	M	17;0	CP quadriplegia with spastic movement impairments	V	S&TD, Mild M	Severe; MA between 4–8y	Non-verbal	Special school for physical and multiple disabled children	OMT, medication	4–5	9.6
A	M	7;10	CP quadriplegia with spastic and dyskinetic movement impairments	III	OD, S&TD, TP, Mild M	No ID; IQ 110 at age 6;0	Speaking, hardly intelligible	Special school for physical and multiple disabled children	OMT	5	9.5
S	F	9;10	CP bipyramidal syndrome with spastic movement impairments, controlled epilepsy	IV	OD, S&TD	Mild; IQ 50 at age 10;11	Speaking	Special school for physical and multiple disabled children	OMT	3–4	7.1
Mi	F	7;3	CP tetraplegia, epilepsy, speech delay	III	S&TD, Mild M	Mild; IQ 60 at age 7;8	Speaking, hardly intelligible	Special school for speech and language disabled children	OMT	5	9.7
Mo	M	10;8	CP quadriplegia spastic movement impairments	III	S&TD, Severe M	Mild; IQ 75 at age 7;9	Speaking	Special school for physical and multiple disabled children	OMT	4	9.7
L	M	9;2	Psychomotor retardation	III	Mild M, hypotonic tongue and lips	Mild; IQ 70 at age 6;5	Speaking	Special school for physical and multiple disabled children	OMT	4	6.9
Je	M	19;9	CP quadriplegia	II–III	S&TD, Severe M	Severe; MA 4;8 at age 14;1	Non-verbal, booklet with pictures, icons and alphabet	Special school for physical and multiple disabled children	OMT, BoNT, surgery (rerouting)	4	9.0
Jo	F	8;2	Worster Drought Syndrome	I	S&TD	No ID; IQ 118 at age 5;5	Speaking, hardly intelligible	Special school for speech and language disabled children	OMT	5	8.5
R	F	7;0	CP quadriplegia with spastic and atactic movement impairments	III	S&TD, TP, Mild M	No ID	Speaking	Regular/mainstreamed school	OMT	5	9.7

GMFCS = Gross Motor Function Classification System; ID = Intellectual Disability; TDS = Teacher Drool Scale; VAS = Visual Analogue Scale; CP = Cerebral Palsy; OD = oral dyskinesia; S&TD = suction and transportation difficulties in the oral phase of swallowing; TP = tongue protrusion during swallowing; M = malocclusion; MA = Mental Age; OMT = Oral Motor Therapy; BoNT = Botulinum Neuro Toxin type A injections into the salivary glands.

Table 2
Description of the five level Gross Motor Function Classification System[a] between the 6th and 12th birthday

I	Children walk indoors and outdoors, and climb stairs without limitations. Children perform gross motor skills including running and jumping but speed, balance, and coordination are reduced
II	Children walk indoors and outdoors, and climb stairs holding onto a railing but experience limitations walking on uneven surfaces and inclines, and walking in crowds or confined spaces. Children have at best only minimal ability to perform gross motor skills such as running and jumping
III	Children walk indoors and outdoors on a level surface with assisted mobility device. Children may climb stairs holding onto a railing. Depending on upper limb function, children propel a wheelchair manually or are transported when travelling for long distances or outdoors on uneven terrain
IV	Children may maintain levels of function achieved before age 6 or rely more on wheeled mobility at home, school, and in the community. Children may achieve self-mobility using a power wheelchair
V	Physical impairments restrict voluntary control of movement and the ability to maintain antigravity head and trunk postures. All areas of motor function are limited. Functional limitations in sitting and standing are not fully compensated for through the use of adaptive equipment and assistive technology. At level V, children have no means of independent mobility and are transported. Some children achieve self-mobility using a power wheelchair with extensive adaptations

[a] For more details, see http://www.canchild.ca/Portals/0/outcomes/pdf/GMFCS.pdf.

were scheduled at the end of each training day. To assess whether the treatment effect was carried over to the next day, the first session of each day started with a latency recording (i.e., pretest latency).

Post-intervention. Because drooling severity may vary across different types of activities, latency scores during treatment may be spuriously influenced by the kind of activities that are performed during assessment. To evaluate if drooling severity decreased during *all* activities, post-intervention tests were scheduled during which the participant performed the same activities as during baseline recordings. After self-management training was stopped, again 10–14 latency recording sessions of either 10 or 15 min were scheduled on 2 consecutive days.

Generalization and follow-up. After discharge from the rehabilitation clinic, generalization of treatment effect to the individual's natural context (i.e., school) was assessed during four sessions, scheduled on 2 consecutive days. These generalization assessments at school were repeated at 6 and 24 weeks after discharge (follow-up). Maximal duration of latency recordings during generalization and follow-up was equal to the longest time interval attained during treatment. No formal training was in effect after discharge.

2.5. Reliability

Reliability of latency recording was determined by a second rater, who independently (but not simultaneously) scored 10% of all videotaped sessions, equally distributed across experimental phases and participants. Raters were considered to be in agreement if latency scores were exactly the same or differed plus or minus 2 s. Inter-rater reliability was 91.2%.

2.6. Procedure

During latency recordings and training sessions, participants performed daily activities derived from their personal educational school plans and according to their educational level, as well as leisure activities. During baseline sessions neither instruction nor comment on drooling was in effect.

During treatment, participants were taught to perform self-management skills and to remain dry for increasing time intervals. Pre-set time intervals were $^{1}/_{2}$, 1, 2, 5, 10, 15, 20, 25, 30, 35, 40, and 45 min. The initial trial of the first treatment session began with a time interval just below the mean latency of being dry during baseline. If the child succeeded to remain dry during three consecutive trials, the next larger interval was set. If the child failed to remain dry during three consecutive trials or during five trials in sum, the previous interval was in effect again until the child succeeded in three consecutive trials. Once the participant had attained the 45 min interval, time intervals were extended either by 5, 10, or 15 min, depending on the participant's estimated potential (determined from learning curve and latency scores during treatment) to a maximum of 60 min.

At the beginning of the first training session, the trainer made the following verbal statements: *You often have wet clothes and a wet chin and this is not good. To avoid getting wet you must swallow, check your chin and if it's wet, wipe it and swallow again. But this is rather difficult! That's why we are going to practice this here in the clinic and in the future also at school and at home.* 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 is 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.* If 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 thinking to his or herself. After instruction, the trainer announced the length of the next time interval and set the stopwatch.

The child was verbally praised if he/she remained dry during an entire interval and received a sticker in his/her swallow-report. 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 s, 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, 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 next trial started with the instruction: *If you are dry, we can start.*

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 weekends, the training was interrupted as the child went home. To prevent relapse and to support generalization, the child was told to practice at home during the weekend. Two or three times a day parents asked their child to stay dry for a time interval somewhat shorter than the interval that was currently trained at the clinic, so that the child could easily be successful. The parents gave the child a sticker in his/her swallow-report if he/she succeeded. If not, parents were instructed to mark a cross in the swallow-report, without negative verbal feedback. After discharge from the rehabilitation clinic, parents and teachers received verbal and written instructions on the continuing administration of occasional positive feedback for the self-management for drooling. Continuation of formal training by parents, teachers, or therapists was discouraged, except for infrequent short trials such as parents performed during weekends.

2.7. Statistical analyses

To evaluate effects of treatment for all activities that were presented at baseline observations, mean latency scores and range for baseline and post-intervention tests were determined and Cohen's *d* effect sizes were calculated.

2.8. Social validity

In addition to objective evaluation of the self-management treatment of drooling by observational data (i.e., latency), we investigated whether parents and teachers noted changes in drooling severity at home and at school and/or its consequences in daily life. To assess drooling severity throughout the day, parents and teachers completed the TDS and a Visual Analogue Scale (i.e., VAS) during 10 consecutive days preceding baseline recordings, during 10 days immediately following discharge from the clinic, and during 10 days at 6 and 24 weeks follow-up. Scores on the 10 cm VAS could vary-from 0 to 100 (0 = 'very severe drooling', 100 = 'no drooling'). In addition, a shortened version of the

questionnaires by Van der Burg, Jongerius, Van Limbeek, Van Hulst, and Rotteveel (2006) was administered 1 week preceding baseline recordings, and at 6 and 24 weeks following discharge to evaluate parental perception of the impact of drooling on daily life and care, social interactions, and self-esteem. A teacher version of this questionnaire was developed and administered at the same points in time.

3. Results

Fig. 1 depicts the latency (i.e., number of minutes dry) during baseline, treatment, generalization, and follow-up at 6 and 24 weeks for each participant (post-intervention tests are not depicted in Fig. 1). It shows that after a 3-week self-management treatment all participants were able to remain dry for intervals of 30–60 min while engaged in daily activities.

The learning curves of K, D, S, and Je contained no relapse to a shorter interval. L and Mo had one relapse, for Jo and R there were two relapses, and for Mi there were five relapses to a shorter interval.

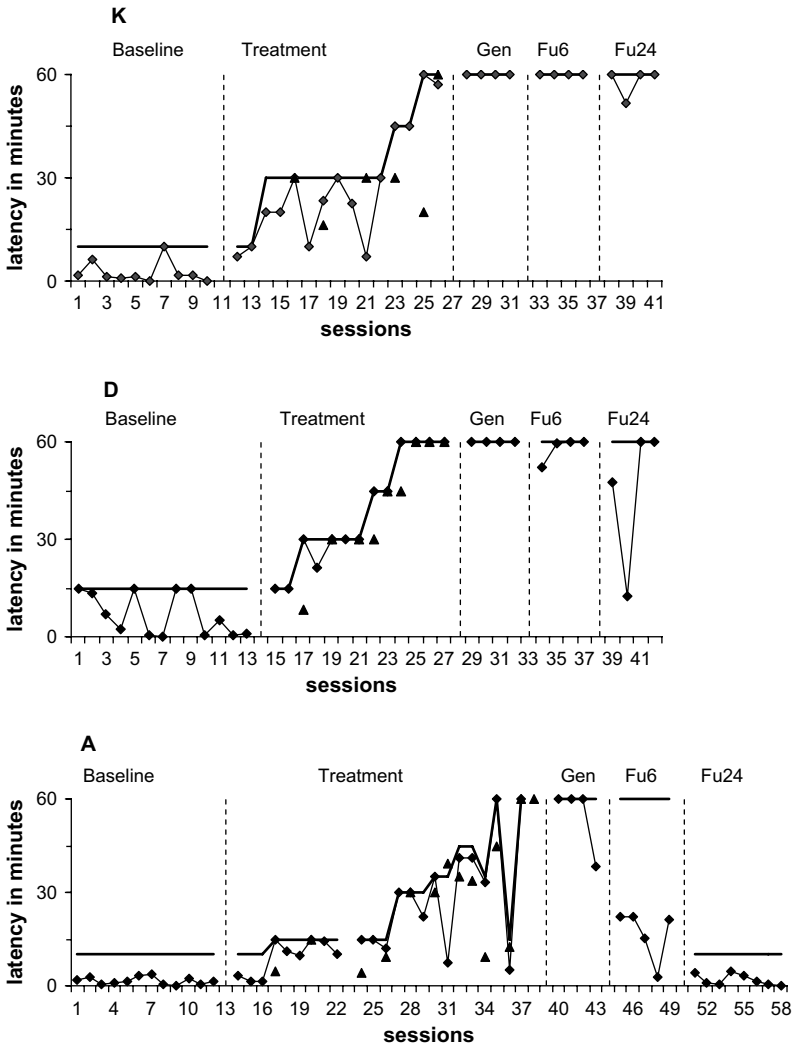

Fig. 1. Latency (in min) during conditions of baseline, treatment, generalization (Gen), and follow-up at 6 (Fu6) and 24 (Fu24) weeks.
—◆— latency —— maximum ▲ pretest latency

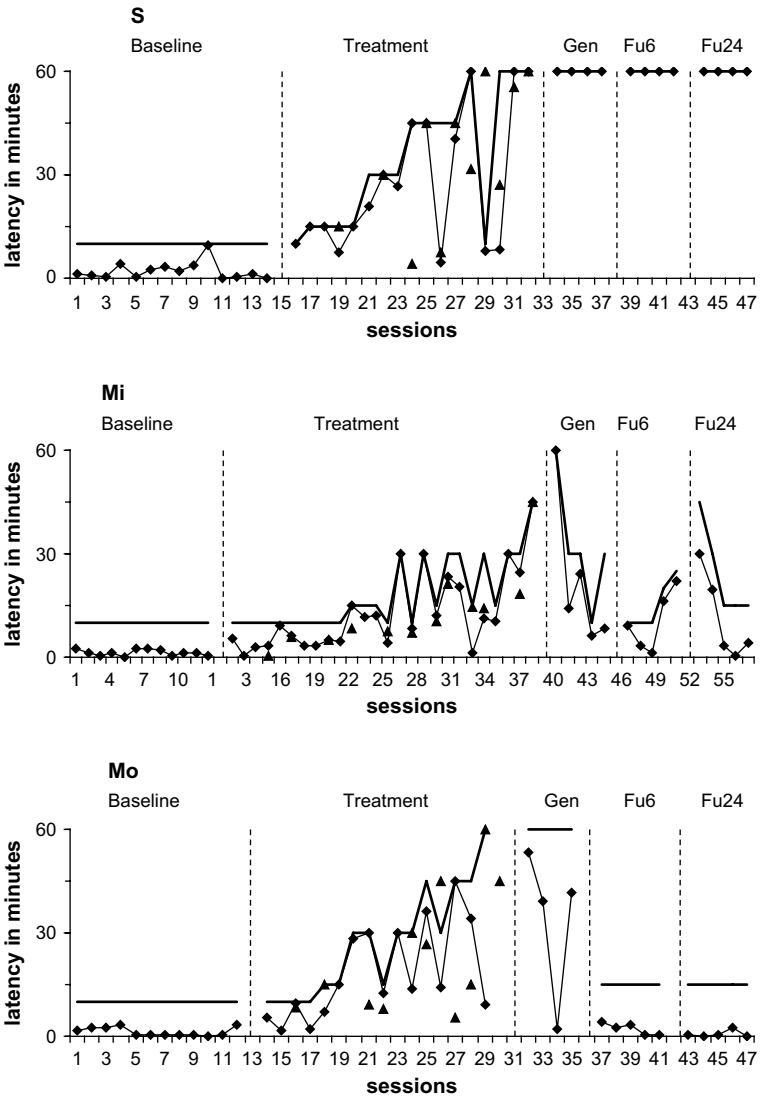

Fig. 1. (*continued*).

Participant A was released from the hospital for a week during the intervention period because he became ill (although his illness was unrelated to the treatment or stay at the clinic). After his return, a latency recording showed that the training had to start at a 5 min interval again. His learning curve did not show any further relapse.

In Table 3, mean and range in latency scores at baseline and post-intervention are presented. Cohen's *d* effect size was large for all participants, ranging from −1.13 to −11.80.

For all participants, generalization to the classroom occurred and for K, D, and S maintenance of treatment effect was established at 6 and 24 week follow-ups (Fig. 1). However, the other participants could not maintain their self-management skills and their latency scores gradually decreased. Whereas L maintained the treatment effect at 6 week follow-up, there was a reduction to baseline level after 24 weeks. For A, Mi, Je, and R, there was a partial loss of treatment effect at 6 week follow-up. While

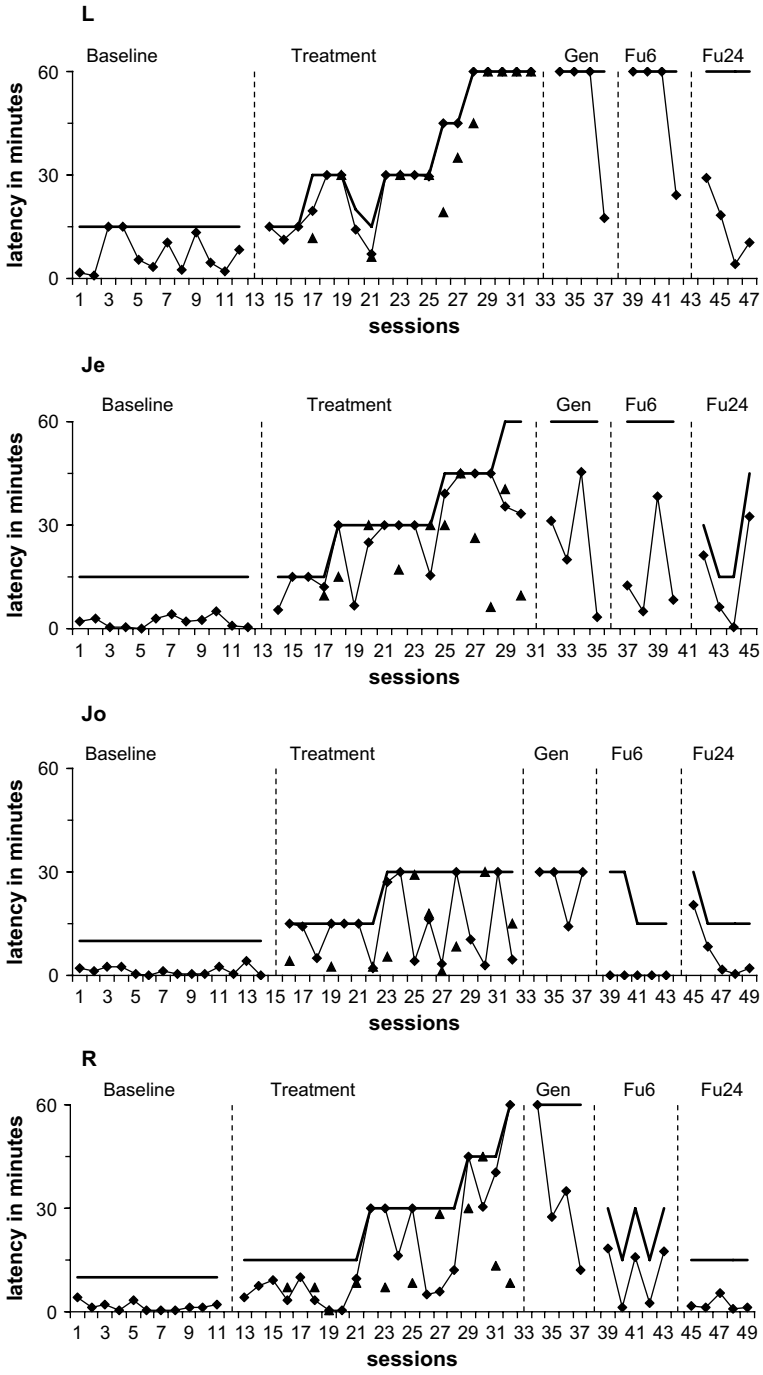

Fig. 1. (*continued*).

Table 3
Mean (range) latency in seconds at baseline and post-intervention, and Cohen's *d* effect size

Participant	Baseline		Post-intervention test		Effect size
	Mean	Range	Mean	Range	Cohen's *d*
K	97.3	7–399	600.0	600[a]	−4.41
D	452.3	17–900[a]	900.0	900[a]	−1.13
A	103.0	23–238	896.1	853–900[a]	−11.80
S	143.1	14–583	600.0	600[a]	−3.05
Mi	90.8	11–175	598.5	582–600[a]	−8.95
Mo	91.7	15–233	600.0	600[a]	−6.13
L	422.1	61–900[a]	900.0	900[a]	−1.50
Je	127.2	8–312	700.7	123–900[a]	−1.99
Jo	89.2	2–266	541.3	174–600[a]	−3.48
R	109.1	27–260	600.0	600[a]	−5.96

[a] Maximum score.

latency scores for Mi and Je remained above baseline level at 6 and 24 week follow-ups, drooling severity of A and R returned to baseline level at 24 weeks. Latency scores for Mo and Jo returned to baseline level at 6 weeks.

Table 4 shows parents' and teachers' reports of drooling severity in daily life on the VAS and TDS during baseline, generalization, and follow-up at 6 and 24 weeks. The scores for individual participants during generalization reflect the positive effect of the intervention on drooling severity in daily life as shown in Fig. 1, and similar trends during the maintenance phase compared to data from direct observation at school. Participants' mean score on the VAS (parent report) increased from 20.1 (SD = 18.7; range 6.5–54.1) during baseline to 74.0 (SD = 25.2; range 43.7–99.4) during generalization, but decreased slightly to 62.4 (SD = 34.5; range 7.8–99.6) at 6 week follow-up, and 53.8 (SD = 38.7; range

Table 4
Individual and group scores on VAS (mean) and TDS (median) at baseline, generalization, 6 week follow-up (Fu 6), and at 24 weeks follow-up (Fu 24)

Participant		VAS[a] (mean)				TDS[b] (median)			
		Baseline	Generalization	Fu 6	Fu 24	Baseline	Generalization	Fu 6	Fu 24
K	Parents	8.8			94.7	5			1
	Teacher				44.0				3.5
D	Parents	17.6	97.1	96.6	97.4	5	1	1	1
	Teacher		96.0	91.2	87.3		1	1	2
A	Parents	7.9	49.1	35.1	12.7	5	3	3	4
	Teacher		65.1	24.4			2	4	
S	Parents		99.4	99.6	100.0	3	1	1	1
	Teacher		97.5	97.8	56.7		1	1	1
Mi	Parents	14.0	77.7		17.9	5	2		4
	Teacher		38.5	55.8	53.4		2	2	3
Mo	Parents	6.5	43.7	7.8	15.3	4.5	3	3	3
	Teacher	6.6	58.5	12.0	9.8	5	3	5	5
L	Parents	54.1	95.1		70.1	4	2		3
	Teacher	49.4	94.2	57.6	58.3	2	2	3	2
Je	Parents	31.7	74.0		55.1	3.5	2	3	2
	Teacher	25.8	54.3	91.0	65.8	5	3	1	2
Jo	Parents		55.5	54.1			4	3	
	Teacher		69.3	56.7	14.1		4	2	5
R	Parents			81.1	21.0	5	1	2	4
	Teacher	32.0				5	2	2	3
Group	Parents	20.1	74.0	62.4	53.8	5	2	3	3
	Teacher	28.4	71.7	60.8	48.7	5	2	3	3

[a] VAS: score 0 = very severe drooling; score 100 = no drooling.
[b] TDS: score 1 = No drooling; score 2 = Infrequent drooling, small amount; score 3 = Occasional drooling, intermittent all day; score 4 = Frequent drooling, but not profuse; score 5 = Constant drooling, always wet.

12.7–100) at 24 week follow-up. Mean VAS-scores from teacher reports are comparable to those reported by parents. Median score on the TDS from both parents and teachers decreased from 5 (parents range: 3–5; teacher range: 2–5) during baseline to 2 (range 1–4) during generalization, and slightly increased to 3 (parents range 1–3; teacher range 1–5) during 6 week follow-up, and 3 (parents range 1–4; teacher range 1–5) at 24 week follow-up. Thus, on both VAS and TDS reports from parents as well as teachers, drooling severity throughout the day was still lower at 24 week follow-up compared to baseline results.

Reduction of drooling after a 3-week treatment also resulted in positive changes in daily care, social interaction, and self-esteem, as reported by parents and teachers. For instance, from the parent questionnaire it appeared that the mean frequency of wiping the child's mouth by parents and chin decreased from 8.5 (SD = 7.0; range 0–20) times per hour at baseline, to 2.6 (SD = 4,7; range 0–15) times at 6 week follow-up. The mean frequency of urging the child to swallow decreased from 6.4 (SD = 4.1; range 1–10) times per hour at baseline to 1.8 (SD = 3.1; range 0–10) times at 6 week follow-up, and changes of bibs or shawls decreased from 5.6 (SD = 3.8; range 0–10) times per day at baseline to 1.3 (SD = 1.4; range 0–4) times at 6 week follow-up. Although four parents reported that their child was avoided by other children because of drooling during baseline, only one parent reported that this was the case at the 6 week follow-up. After intervention, more participants than before intervention were reported to overtly express positive feelings about their physical appearance (one participant at baseline, four participants at 6 week follow-up), social acceptance by peers (none at baseline, four at 6 week follow-up), and social acceptance by adults (one at baseline, six at 6 week follow-up), and fewer participants expressed negative feelings on these measures in relation to their drooling than before treatment. Mean scores on most items of the questionnaire returned toward baseline levels at 24 week follow-up assessment. Teachers also reported that more children express overtly positive feelings about their physical appearance (two participant at baseline, four participants at 6 week follow-up), and social acceptance by peers (one at baseline, three at 6 week follow-up).

4. Discussion

From results of this case series, it may be concluded that the self-management treatment was effective in increasing time of non-drooling in a group of 10 children with severe drooling. The number of minutes of being dry increased to intervals of 30–60 min, indicating that after a 3-week intervention period all participants were taught to perform self-management skills while engaged in their daily activities. Parental and teachers' judgment of drooling severity during the day also showed positive results following treatment. The self-management program also resulted in positive changes in the impact of drooling on daily care, social interactions, and self-esteem, as reported by parents and teachers. For all participants, generalization to the classroom occurred, but only three participants maintained treatment effects at 6 and 24 week follow-ups. Seven participants could not maintain the high level of performance of the self-management skills at follow-up. Latency scores returned to baseline level for five participants. Nevertheless, parents and teachers still rated drooling severity (VAS and TDS) below baseline level at 24 week follow-up.

This case-series, along with two case reports (Dunn et al., 1987; Thorbecke & Jackson, 1982), provides evidence of promising results of a self-management procedure. However, there were differences in procedures and methodology between our study and the previously published studies. First, while Dunn et al. (1987) focus on mouth closure and swallowing only, Thorbecke and Jackson (1982) taught the participant to both swallow and wipe her face when wet, just as we did. The latter combination of target behaviors is preferable because it enables the child not only to prevent drooling, but also, if necessary, to restore his/her physical appearance. Second, if wiping is adequately performed (i.e., after one or two wipes the lips and chin are dry), it may be not necessary to instruct the individual to wipe 10 times (Thorbecke & Jackson, 1982). Although this type of positive practice strengthens the wiping response during training, it is socially undesirable when excessive wiping becomes a habit in natural situations. Additionally, children suffering from severe drooling may have chronically irritated, chapped, or macerated skin over the chin and peri-oral region, and excessive wiping can further worsen this adverse condition. For these reasons, wiping frequency in our protocol did not exceed three times. Third, prompting and data recording by the teacher at 5–30 min intervals throughout the day

(Thorbecke & Jackson, 1982) is labor-intensive. In our experience, introduction of a self-management procedure for drooling and/or data collection by a teacher should be avoided because it increases the risk of procedural failures and discontinuation of the training program. Introducing the procedure at the hospital and generalizing the treatment program to the classroom after reduction of drooling is established (see Dunn et al., 1987), prevents overworking the teacher, and enables monitoring adherence to the procedure by the therapist. Finally, the definition of drooling and its measurement in earlier studies is debatable. Dunn et al. (1987) defined drooling as the number of drops falling from the lip or chin. However, the social disabling effect of drooling is already apparent when saliva is below the lower lip line on the chin for one or 2 s. Although Dunn et al. (1987) claimed positive effects of their treatment, it remains unclear if their participants' face was fully dry or that drops of saliva were no longer falling from his lip or chin. Therefore, to obtain socially valid conclusions from intervention studies, drooling should be defined as 'saliva below the lower lip line on the chin or a string of dribble falling straight from the mouth'. Thorbecke and Jackson (1982) evaluated drooling severity by wiping the observer's finger across participants' chin beneath the lower lip. During baseline, the individual's chin was wiped dry afterward to ensure that previous drooling did not affect the next observation. However, there is a risk that this routine becomes a discriminative stimulus for the individual to attend and react to drooling before the self-management procedure is administered.

Main goal of the treatment was to train the child to remain dry for increasing time intervals. For this purpose, we used latency in minutes of being dry (non-drooling) while performing daily activities. We did not use other techniques, for example, time-sampling techniques, because samples of 60 min would have resulted in a disproportional increase of baseline recordings during the stay at the rehabilitation clinic. In addition, we aimed at the elimination of drooling and not merely a reduction of the percentage of intervals without drooling, since the stigmatizing effect of drooling is apparent whenever any saliva is present outside the mouth.

Some shortcomings of the present study need to be mentioned. In this study, procedural integrity was not assessed because the child's performance of self-management skills was not measured during treatment and following phases. In addition, the results of parents' and teachers' scores on VAS and TDS should be interpreted with caution, as parents and teachers were not blind to the experimental conditions and the purpose of the study, which may have resulted in bias.

To facilitate generalization, the self-management program could be conducted in the natural environment of the child at home and at school right from the beginning. However, we choose to treat the participants during a 3 week period in a clinical setting because in general the natural context of the child at home and at school does not allow for this kind of intensive behavioral treatment. During treatment, the focus of the child and parents was on acquisition of self-management skills and reduction of drooling, while not being distracted by other activities or events at school or at home that could interfere with the intensive training. To simulate the natural environment as much as possible, we both trained and measured the effect of treatment while the child was performing daily activities with objects (books, toys, etc.) from their homes and schools, so-called common salient physical stimuli (Stokes & Osnes, 1989). To become independent from the trainer and to normalize social interactions, instructions and prompts were gradually faded as the time intervals were increased in a stepwise manner.

Anecdotal reports of parents and teachers revealed several personal factors and events that may have negatively influenced generalization and maintenance in this study. These personal factors and events were either common for the population (e.g., stress from family or school problems, limited motivation, pain, inflamed tonsils) or more or less characteristic of individuals with developmental disabilities (such as loss of posture control because of growth or physical deterioration, impaired ability to attend to more than one task at a time, especially in a complex and demanding environment at school or at home).

In our study, demographic characteristics of participants varied considerably in terms of age, developmental age, (oral) motor functioning, and intellectual functioning. It remains to be determined to what extent these factors have influenced effectiveness, generalization, and maintenance of self-management. Although maintenance of effect in our group was best among the children with high GMFCS scores (i.e., K and S at level IV, D at level V), indicating severe gross motor problems, no final conclusions can be drawn on this small number of participants. Future research with detailed

description of demographic and medical characteristics of participants is warranted to establish inclusion criteria (e.g., minimum age, minimum developmental age, minimum intellectual functioning) for the self-management program to be effective and to identify critical factors for generalization and maintenance.

Duker, Didden, and Sigafoos (2004) state that it is tempting for trainers and others to attribute the failure to maintain and generalize newly acquired responses to some deficit in the learner. However, lack of generalization and maintenance may be the result of shortcomings in instructional procedures. For instance, the trainer in our study may become the only or most salient discriminative stimulus to perform the self-management skills, and the natural environment may not be sufficiently prepared to ensure that newly acquired responses will enable the learner to gain reinforcement, so the responses are not maintained once training ends. As Stokes and Osnes (1989) pointed out, additional procedures during and/or after treatment are needed to promote generalization and maintenance. In future studies, this may be accomplished by incorporating functional mediators such as important persons from the child's natural environment (e.g., parents, teachers, and/or peers) in the treatment procedure as common salient social stimuli, both during the 3 week intervention period and following discharge, during the generalization and maintenance phase. Another strategy to promote generalization and maintenance is to recruit natural consequences by teaching the child to ask for their parents' and teachers' attention for non-drooling (for instance with cueing questions like: 'How do I look?') to elicit positive remarks on their physical appearance when dry.

Dunn et al. (1987) noted the need for additional treatment at 6 months after intervention, to establish long-term generalization of their self-management procedure. In our study, some participants gradually lost their level of performance of self-management skills as early as a few days after discharge. If maintenance fails to occur, additional training is needed. This may be either a booster training at the hospital (if the participant completely fails to exhibit the self-management skills) or an outpatient period of short training sessions in which the self-management procedure is supported by parents and teachers. This last option brings the self-management procedure under the control of natural stimuli (e.g., the child's parents and teachers). To perform this additional training, parents and teachers need to learn the instructional procedure for the self-management skills for drooling, including appropriate motivation and feedback techniques. Instructional courses and video-feedback sessions for parents and teachers may be scheduled during admission to the hospital or following discharge. Formal training sessions with the child by parents and teachers in their natural environment should be carefully planned and monitored, since they often have many other responsibilities and priorities.

Behavioral treatment for drooling is an option that may be considered in relation to the advantages and disadvantages of oral motor therapy and related training with oral appliances, medication, Botulinum Toxin type A injections and surgery (Van der Burg et al., 2007a, 2007b). Although the present study shows promising results for a self-management procedure, it needs further adaptations to improve efficacy, generalization, and maintenance. Also, this kind of behavioral treatment is not suitable for all children with severe drooling. If self-management skills cannot be learned and maintained, (automatic) cueing strategies for swallowing and/or wiping may be a good alternative behavioral procedure. Further research is also needed to explore the ways in which behavioral treatment can be supported by medical interventions.

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