Review

A descriptive analysis of studies on behavioural treatment of drooling (1970–2005)

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A descriptive analysis was conducted on studies on the behavioural treatment of drooling (published between 1970 and 2005). The 17 articles that met the inclusion criteria described 53 participants (mean age 14y 7mo, [SD 4y 9mo]; range 6–28y). Sex of 87% of the participants was reported: 28 male, 18 female. For 60% of the participants the degree of learning disability was reported, varying from severe/profound (*n*=24, 75%), moderate (*n*=4, 13%), to mild (*n*=2, 6%), while two participants (6%) had no learning disabilities. Forty-two participants (79%) were diagnosed with cerebral palsy. Behavioural procedures included instruction, positive and negative reinforcement, overcorrection and restitution, verbal and automatic cueing, and/or self-management. Effective behavioural procedures are reported in children with and without learning disability and/or motor impairment. Even participants with profound learning disability may benefit from behavioural intervention. However, the evidence base in terms of number of studies in this area is limited. Fifteen studies used a single participant design; two studies implemented an experimental-comparison group design. Some of these studies were poorly designed and methodological flaws were identified. Therefore, conclusions about efficacy of behaviour therapy for drooling and/or best practice cannot be drawn, although our analysis suggests that this approach is promising. However, future research on this topic is needed. After years of research focused on medical treatment, the option of behavioural treatment to reduce drooling should be reconsidered in relation to the medical management of this problem.

Various treatment strategies to reduce drooling have been reported, including oral-motor training, proper body positioning, behavioural therapy, the use of intra-oral devices, medication, and surgery. Their effectiveness and relative contribution to the management of drooling has been discussed in a number of narrative reviews.[1–8] There is general agreement that behavioural treatment is one of the viable options that should be implemented before other, more intrusive, strategies are considered. For example, Blasco[2] states: 'In some cases, usually those more mildly involved, behavioural techniques (…) are easy to institute and will work.' Brei[3] concludes that behavioural interventions are relatively time-consuming and more likely to be effective for children with better cognitive functioning and milder drooling. Further, he concludes that behavioural interventions usually do not totally eliminate drooling, that spontaneous generalization does not occur, and that its long-term benefits are uncertain. A critical note is made on the small sample size described in effect studies on behaviour modification. However, rigorous single-participant design methodology is considered more convincing than studies on oral-motor treatment that most often lack experimental rigour.[1]

Some important shortcomings of the above reviews should be mentioned. First, none of these reviews cover all published studies on behaviour therapy for drooling. Second, no systematic analysis of methodological issues or procedural aspects of behavioural intervention has been carried out. In this paper, we present a descriptive analysis of all behavioural treatment studies on drooling published between 1970 and 2005.

Method

LITERATURE SEARCH

Searches in Medline, PubMed, PsychInfo, and Web of Science using the keywords 'drooling' and 'sialorrhoea' in combination with 'behaviour therapy', 'behaviour treatment', 'behav-

iour training', and 'behaviour modification' yielded a database of articles on behavioural treatment of drooling. To include studies from the USA, searches were also performed with the keyword 'behavior'. In addition, the references from the above retrieved articles and general reviews on drooling were scrutinized for additional relevant titles. Inclusion criteria were: (1) empirical data were presented on the use of a behavioural intervention as treatment for drooling; (2) participants were diagnosed with developmental disabilities (e.g. cerebral palsy [CP], learning disability*); and (3) published between 1970 and 2005. Exclusion criteria were: (1) participants were treated exclusively by any of the following approaches: oral-motor training, proper body positioning, the application of intra-oral devices, medication, or surgery; and (2) behavioural interventions focused at other problem behaviour neglecting to report systematic data presentation on drooling severity before and after intervention.

STUDY VARIABLES

Data pertaining to the participants' sex, age, degree of learning disability, motor impairment, former (medical) treatment for drooling, and drooling severity were collected. Information about the setting in which the behavioural treatment was conducted was also recorded. We investigated how changes in drooling severity were evaluated. Also, data on interobserver agreement were registered. Since conclusions from research in applied settings are dependent on the type of experimental design used, we also assessed if, and how, experimental control was obtained. Although the main purpose of all studies was to reduce drooling, the behaviour targeted for intervention varied (e.g. swallowing, wiping the mouth and chin, or exercising self-control). Behavioural treatment focuses on changing stimuli either preceding the target behaviour (i.e. antecedent procedures) or following the target behaviour (i.e. consequent procedures). We assessed target behaviour and treatment type and determined whether a specific apparatus was used. To estimate the cost of the behavioural treatment, we calculated frequency (i.e. training sessions per day or week) and duration (in weeks or sessions) of instruction by the trainer.

Individual studies were analyzed with regard to treatment effectiveness by visual inspection of graphic or numerical data and authors' evaluation.

INTERRATER RELIABILITY

A second rater independently reviewed four of the experiments[12,13,16,19] that were randomly chosen from the total

*North American usage: mental retardation.

Table I: Demographic characteristics, setting, and time investment for intervention

Study	*Sample size, n*	*Sex M/F*	*Age, y*	*Degree of learning disability*	*Motor impairment (MI)*	*Setting*	*Instruction frequency (mins); duration treatment*
Barton et al.[9]	2	2M	24, 28	Moderate (IQ 45); Severe (IQ 30)	CP	LD residential setting	– (10–15); >500&900s
Barton & Madsen[10]	1	M	11	Severe	–	Special education class	5/wk; 24s
Connis & Rusch[11]	1	M	22	Moderate	–	Vocational training in a food service programme	5/wk; 36d
Drabman et al.[12] exp 1	3	2M/1F	7, 12, 15	Profound	1 CP, 2 no MI	Nonresidential LD centre	5/wk; 12–15wk
Drabman et al.[12] exp 2	2	1M/1F	14, 15	Mild (IQ 63); Severe (IQ 39)	no MI	Nonresidential LD centre	5/wk; 19&15d
Dunn et al.[13]	1	M	16	No LD	CP	Special class in a regular public school	1d/wk; 10wk
Garber[14]	1	M	14	–	CP	Speech and hearing clinic	3s/wk (25); 6wk
Jones[15]	8[a]	–	12–16	–	CP	Residential school for children with SLD CP	5/wk; 3wk
Koheil et al.[16]	12	9M/3F	6–18	No severe LD	CP	Children's hospital and school/regular environment	–
Lancioni et al.[17,18]	2	1M/1F	27, 20	Moderate (IQ 40); Severe (IQ 35)	1 no MI; 1 CP	LD residential setting, activity centre and farm	1–2s/d (30); 35s
Lancioni et al.[19]	2	2M	16, 11	Mild (IQ<70); No LD (IQ>70)	1 CP, 1 no MI	Residential setting for the deaf	–;32&26s
Poling et al.[20]	1	M	17	Moderate	–	School for LD	5d/wk; 25d
Rapp[21]	8[b]	3M/5F	13–15	Profound & severe (MA: 1½–6y)	CP	Residential school for children with SLD CP	5/wk (1s); 1wk
Rapp & Bowers[22]	7	3M/4F	15–16	Severe (MA: 4–8y)	CP	Residential school for children with SLD CP	5/wk (1s); 1wk
Richman & Kozlowski[23]	1	F	9	Severe	CP	LD residential setting	4s/wk (30); 20wk
Thorbecke & Jackson[24]	1	F	19	Moderate (IQ 47)	CP	Special developmental school and at home (generalization)	5/wk; 25d
Trott & Maechtlen[25]	1	F	11	Severe	no MI	Self-contained special class in a public elementary school	2s/d(15,25–30); 3wk

MA, mental age; LD, learning disability; CP, cerebral palsy; SLD, severe learning disabilities; s, sessions; d, days; /d, per day; wk, weeks; /wk, per week; –, not stated. [a]One patient was treated previously in study by Rapp.[21]; [b]Rapp[21] also reports data on the follow-up of the Rapp and Bowers[22] group and on a comparison group.

sample (34% of the participants). If both reviewers recorded the same descriptive category for the study variables, they were considered to be in agreement. Interobserver agreement was calculated for each study variable by dividing the number of agreements by the number of agreements plus disagreements and multiplying by 100%. Mean interobserver agreement for study variables was 81.5% (range 68–95%).

Results

Seventeen articles, published in a total of 11 journals between 1970 and 2005, were identified.[9–25] Most articles (76%) were published between 1977 and 1987. After 1994, no study on behaviour treatment for drooling appeared. The number of participants in the experiments varied from one (eight studies), two (four studies), three (one study), seven (one study), eight (two studies), to 12 (one study). Most articles described one experiment only. The selection included two series of articles in which (some of the) participants were treated repeatedly.[15,17,18,21,22] That is, the follow-up for the experiment by Rapp and Bowers[22] was reported in the study by Rapp,[21] in which the procedure was also repeated with a second intervention group. Jones[15] conducted another study at the same school. The most recent study by Lancioni et al.[18] is a follow-up of their 1992 study, reporting data for the same participants. The study by Drabman et al.[12] described two experiments. In the present review each experiment was analyzed separately, including follow-up data published in subsequent articles.

STUDY VARIABLES

Participants and setting

The selected studies described 53 participants receiving behavioural treatment for drooling (see Table I; patients participating in more than one study were only counted once). The sex of 87% of the participants was reported: 28 males and 18 females. The mean age was 14 years 7 months (SD 4y 9mo) with age ranging from 6 to 28 years: 12 participants were between 6 and 12 years, 35 participants were between 13 and 18 years, and six participants were over 18 years. For 60% of the participants the degree of learning disability was reported, varying from severe/profound (n=24: 75%), moderate (n=4: 13%), to mild (n=2: 6%), while two participants (6%) had no learning disability. Forty-two participants (79%) were diagnosed with CP: hemiplegia (n=5), diplegia (n=3), quadriplegia (n=11), and type of CP not specified (n=23). Functional level of CP could not reliably be determined from the short and global descriptions of participants. Six participants had no motor impairment. Previous medical treatment was seldom mentioned. Dunn et al.[13] reported that their patient was treated by surgery to reroute the duct of one salivary gland 2 years before behavioural treatment. Drooling severity was indicated by scores on different dependent measures, and by global descriptions like 'sufficient to require the programme', 'profuse', 'excessive', 'continuously', and 'chronic'. No standardized measures for drooling severity were used in the studies.

Dependent variables and reliability

Across the selected studies the definitions varied for drooling and additional dependent variables, for assessment procedures and their frequency, and for activities performed during assessments. In four studies[14–16,22] no interobserver reliability was reported. In the other studies, interobserver reliability appeared to be consistently high: above 70%.

Design

Rapp[21] used an experimental comparison group design without randomization and presented data for a comparison group for the patient group in both her first[22] and second[21] studies. Although the comparison group was comparable with both intervention groups on chronological and mental age, the percentage of drooling during baseline differed substantially: 62% and 75% for the intervention groups versus 31% for the comparison group. The remaining 15 studies used a single participant design: six experiments with AB (baseline and intervention) design,[11,12,14,15,25] six used a reversal design,[9,13,16,19,20,24] in which each participant served as his/her own control. Five studies used a multiple baseline design, either across behaviours[23] (one), across activities[13] (one), across settings[10] (one), or across participants[17–19] (two). However, sufficient experimental control was not always achieved in these studies (i.e. AB designs) and trends in baseline data invalidate conclusions on the effect of intervention. Garber[14] started intervention after only two baseline sessions in which a decreasing trend of drooling was already apparent, and in two other studies[11,23] drooling rate also showed a decreasing trend during baseline.

Thirteen studies presented follow-up data at one or more intervals for a maximum of 1 year after intervention. Lancioni et al.[18] reported the most extensive follow-up data: at a 1- to 2-monthly period during the first 18 months after the intervention.[17]

Target behaviour and type of treatment

Target behaviours for treatment were swallowing,[9,12,14–16,21,22,24] wiping,[9,10,12,17–19,22] non-drooling,[10,12,14,20,22,25] mouth closure,[11] head control and imitative vocalization,[23] and self-management.[13,24] In antecedent procedures participants had received verbal, auditory (beeps or electromyography feedback), visual, or vibratory cues at different time schedules to elicit the target behaviour. In eight studies[9,15–19,21,22] (involving 40 participants), cueing devices were used. Initially, participants received instructions to swallow or wipe following the cue. In some procedures[9] cues were gradually faded, while in others[17,18] a specific cueing frequency was maintained, adjusted to the child's needs.

In consequent procedures, participants were positively reinforced after performing the target behaviour. In five studies, negative social reinforcement (e.g. a negative reprimand like 'Your chin is wet, it looks horrible') was given whenever drooling occurred,[11,12,20,24,25] while in three of these experiments wiping was used as overcorrection and restitution procedure.[12,24,25] Only two studies focused on the development of self-management skills to reduce drooling. Thorbecke and Jackson[24] taught the participant in their second treatment phase to self-evaluate her chin (wet or dry) and to instruct herself by internalizing the self-statements used in the overcorrection procedure. Dunn et al.[13] taught the participant to monitor mouth closure, determine the need to swallow, evaluate the success of drooling prevention, and to verbally reward himself.

From data on frequency and duration of instruction (see Table I), it appears that some interventions using cueing devices[15,21,22] are less time-consuming than other procedures. Although the studies by Barton et al.[9] and Richman and

Kozlowski[23] are extensive (20 or more weeks), the remaining intervention procedures take between three and 15 weeks.

Effectiveness

All authors, except Jones,[15] claim positive results and many of them anecdotally describe the related positive changes in quality of life of the participant(s). In 13 experiments[9–14,17–20,23–25] (19 participants) graphic data are presented. Visual inspection of these data shows that drooling severity after behaviour therapy is well below baseline level in all experiments and sometimes even reduced to (near) zero.[12,13,19] From the studies reporting numerical data[15,16,21,22] (35 participants), five participants had drooling rate zero during intervention and 28 participants showed a reduction in drooling rate of more than 50% compared with baseline.

Generalization and follow-up

Most authors both trained participants and evaluated effectiveness of their intervention in daily activities. Barton et al.,[9] Barton and Madsen,[10] and Dunn et al.[13] assessed generalization to respectively 'work', 'class', and 'school'. In four experiments[14–16,23] training and/or assessment were conducted in a specific room or during a few activities without assessing the generalization of treatment effect.

Most studies report maintenance of treatment effect at follow-up. Four studies do not report follow-up data. Barton et al.[9] report no formal follow-up data, but the length of their $n=2$ study seems extremely extensive with more than 500 and 900 sessions, respectively. For two participants[12,13] drooling returned to baseline level at follow-up. Dunn et al.[13] scheduled a training refresher after which drooling once again was eliminated for a half-year period. The majority of participants ($n=24$, 59%) who were trained with a cueing device still used these devices at follow-up.

Discussion

Behavioural therapy may be an effective treatment option for drooling. Substantial reduction and even elimination of drooling was reported in the studies reviewed. Effective behavioural procedures were implemented in participants who were 6 years of age and above, with and without learning disability and/or motor impairment. Even participants with profound learning disability may benefit from behaviour intervention.

However, it should be noted that the evidence base, in terms of number of studies in this area, is limited. This may be the result of a publication bias since only effective studies are accepted for publication. In the past 35 years, only 17 empirical studies have been published worldwide, in which various treatment procedures are evaluated on 53 participants varying considerably in age, and learning and motor disabilities. Some of these studies are poorly designed and we have identified methodological flaws. The evidence coming from these studies can be graded against the standards set by Sackett et al.[26] Up to the present day, no randomized controlled trials (level I) have been performed. Using an experimental-comparison group design, the level of evidence for the studies of Rapp and Bowers[22] and Rapp[21] could be rated at level III, but the experimental and comparison groups were not comparable on the dependent variable drooling. All other studies were case reports or case series (level IV) with experimental designs often used in behavioural intervention studies. Because behavioural interventions are complex and demanding, it is difficult to include a large number of participants when conducting such studies in daily clinical practice. Carefully designed trials ($n=1$) have internal validity if reversals or multiple baselines are implemented, but need replication to enhance external validity of their conclusions. Therefore, conclusions about efficacy of behaviour therapy for drooling and/or best practice cannot be drawn yet, although our analysis suggests that this approach is promising.

The most recent study on behavioural treatment for drooling was published more than 10 years ago. Recent research and earlier reviews on this subject focus on surgery and on treatment with botulinum toxin (BTX-A; e.g. Meningaud et al.[7]). Such interventions are time efficient and generalization over situations is assured. However, they are also invasive, irreversible (surgery) or only temporarily effective (BTX-A), and participants do not learn to control drooling by themselves. Although the costs of the behaviour treatments presented here cannot be determined, these seem to be high because of the labour intensity. However, costs of surgery, (repeated) BTX-A injections, and long-term oral motor treatment may be considerable as well.

Behavioural treatment is assumed to be more effective for children with better cognitive functioning and milder drooling (e.g. Brei[3]). Although this issue was not addressed in the literature so far, this review has not substantiated this assumption either. In earlier reviews, behavioural therapy was considered as the intervention technique to be used before medical treatment. However, as the study of Dunn et al.[13] shows, behavioural therapy can also be effective following surgery. Greensmith et al.[27] reported that 2 and 5 years after surgery (bilateral submandibular duct transposition [$n=67$] combined with bilateral sublingual gland excision [$n=41$]), participants still frequently drooled with moderate severity (i.e. wet lips and chin). Perhaps additional behavioural treatment would improve the effectiveness of the surgery. The option of behavioural treatment to reduce drooling should be reconsidered in relation to the medical management of the problem.

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