DROOLING IN CHILDREN WITH CEREBRAL PALSY: IMPACT AND BEHAVIOURAL TREATMENT

Jan van der Burg

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DROOLING IN CHILDREN WITH CEREBRAL PALSY: IMPACT AND BEHAVIOURAL TREATMENT

ERNSTIG SPEEKSELVERLIES BIJ KINDEREN MET CEREBRALE PARESE: IMPACT EN GEDRAGSTHERAPEUTISCHE BEHANDELING

Een wetenschappelijke proeve op het gebied van de Medische Wetenschappen

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aan de Radboud Universiteit Nijmegen
op gezag van de rector magnificus
prof. mr. S.C.J.J. Kortmann,
volgens besluit van het College van Decanen
in het openbaar te verdedigen op vrijdag 30 mei 2008
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te Oss

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Contents

Voorwoord

Het schrijven van een proefschrift is een kwestie van je goed concentreren en een lange adem hebben. Een andere voorwaarde is een groep van mensen om je heen verzamelen die de bereidheid hebben je te ondersteunen met goede adviezen, tekstuele correcties, statistische analyses, boeiende discussies, ontspannende activiteiten en koppen koffie. Voordat u mijn proefschrift gaat lezen, moet u weten dat ik een fantastische groep van collega's, studenten, familieleden en vrienden om mij heen heb die ieder een onmisbare bijdrage leverden. Hen allen wil ik hiervoor hartelijk bedanken, en enkelen in het bijzonder noemen.

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Chapter 1

General introduction

Preamble

After the introduction of Botulinum Toxin type A (BTX-A) as an option for the treatment of a variety of functional problems and cosmetic purposes during the 90's, it has also been applied for the reduction of drooling in patients with Amyotrophic Lateral Sclerosis (ALS), Parkinson's Disease, and Cerebral Palsy (CP). As in cosmetic applications of BTX-A, this treatment aimed at ameliorating the physical appearance of its users, and as such also contributing to their quality of life.

Changes in the quality of life of participating children and their parents were documented during the first controlled clinical trial that evaluated the effect of BTX-A treatment on drooling in children with CP (Jongerius, 2004). The question arose as to whether behavioural science could contribute to the treatment of drooling. A systematic review of the literature on behavioural treatment eventually resulted in the development and evaluation of a self-management program, representing a non-invasive option to treat drooling in children with CP.

Drooling: definition, causes, and consequences

Blasco and Allaire (1992) define drooling as "the unintentional loss of saliva and other contents from the mouth" (p. 849). Drooling severity varies from incidental occurrence of saliva beneath the lower lip line to strings of dribble continuously falling from the mouth or chin. Drooling is a normal phenomenon up to the age of 18 months, but it persists in children with poor neuromuscular coordination, learning disability, and/or children with no structural integrity of jaws, lips or oral cavity.

Three pairs of major salivary glands (Figure 1.1) are responsible for most of the saliva production. The submandibular glands produce about 60 to 70 percent of the saliva 'in rest'. The large parotid glands are capable of producing large amounts of (watery) saliva that is secreted promptly as a reaction to tactile or gustatory stimulation of the oral mucosa, for example during eating or drinking. The sublingual glands contribute the least amount of saliva. There is general agreement that the total amount of saliva production is normal in children with CP (Senner, Logemann, Zecker & Gaebler-Spira, 2004; Tahmassebi & Curzon, 2003). Drooling results from an impaired saliva control. Senner et al. (2004) summarized multiple causes for drooling in CP. These are: increased difficulty forming a bolus, reduced lip closure, slightly less intra-oral suction, more oral residue after swallowing, poor head control, reduced ability to voluntarily control the lips, tongue and jaws, reduced intra-oral sensitivity, reduced frequency of spontaneous swallowing, oesophageal stage abnormalities, dental malocclusion, and, finally, poor coordination of orbicularis oris muscle and masseter activity.

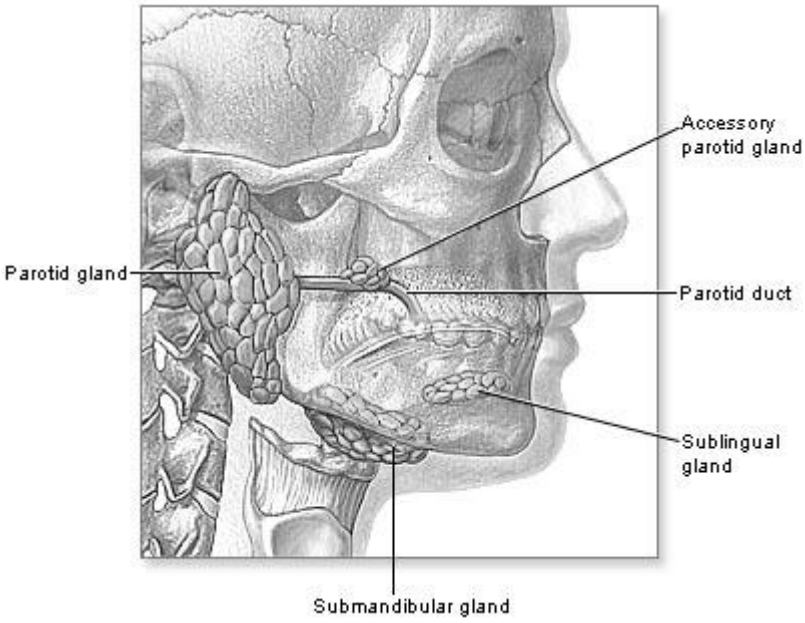

Figure 1.1 Salivary glands

The International Classification of Functioning, Disability and Health (ICF; World Health Organization, 2001) provides a model for the classification of the consequences of diseases on three levels: body functions and structures, activities, and participation. In this model, the impairment of saliva control, i.e. drooling, is situated in chapter 5 'functions of the digestive system' as part of 'Body functions and structures' (Figure 1.2). Due to their multiple impairments in body functions and structures, the development of children with CP may be at risk, with drooling being only one of their problems. For children with CP, participation in age-appropriate activities and social relations might be limited due to restricted motor functioning, learning disabilities, and communication problems (Beckung & Hagberg, 2002; Morris, Kurinczuk, Fitzpatrick, & Rosenbaum, 2006; Schenker, Coster, & Parush, 2005a, 2005b). In addition to disease-related factors, environmental factors (such as parental judgement of the impact of drooling and their reaction to their child's drooling, overt social reactions from peers and others on drooling, and the child being included in a regular or in a special school or residential facility for persons with developmental and intellectual disability) and personal factors (such as age, self-concept, perseverance) will influence the permitted activities and level of participation of drooling children with CP.

Rosenbaum and Stewart (2004) state that the 'bio-psychosocial' ICF model offers opportunities for both counselling and intervention in children and youth with CP, as well as "many possibilities to explore research questions with a fresh approach" (p. 5). They plea for multidimensional measurement of outcomes "to encompass the impact of what

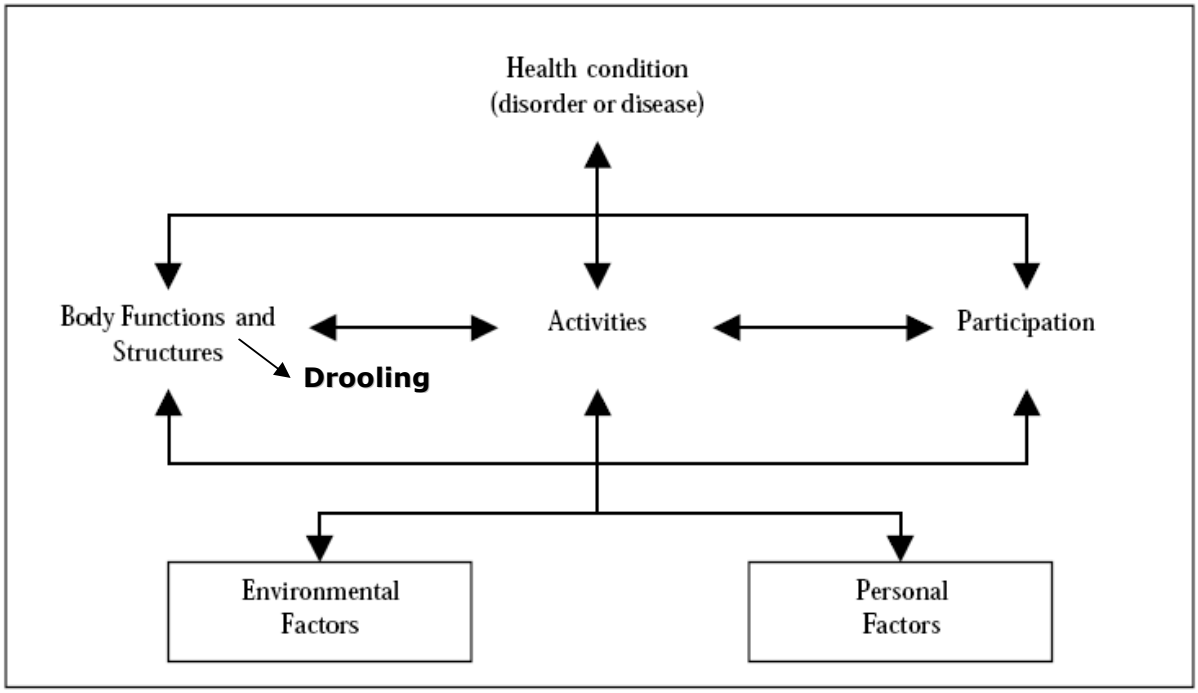

Figure 1.2 Drooling and ICF

we offer in treatment at different levels of body function and structure, activity, and participation" (p. 8). In addition, environmental and personal factors should be addressed "to capture the complex interactional nature of the life experiences of these children and their families" (p. 9). Like other isolated phenomena, very little research has been done on the impact of drooling on activities and participation of children with CP and their families.

Drooling and Quality of Life

Quality of Life (QOL) is an area of research that has received much interest in the last decade. QOL has been defined by the World Health Organisation (WHO) as 'the individual's perception of their position in life, in the context of culture and value systems in which they live and in relation to their goals, expectations, standards and concerns' (WHO, 1993). Because the research in this area is mainly focused on quality of life related to health issues, the term Health-related Quality of Life (HRQOL) was introduced. Health, as defined by the WHO (1948), consists of physical well-being, mental well-being, and social well-being, and is not merely the absence of disease or infirmity. HRQOL is considered to be a subset of QOL and refers to an individual's perception of his or her health, consisting of physical, mental, and social well-being (Waters, Mahler, Salmon, Reddihough, & Boyd, 2005). This subjective and highly individualized measure is the counterpart of the more objective taxonomy of ICF's activities and participation.

Since QOL research has been introduced, medical treatment has not only been evaluated on its effectiveness, but also on its contribution to the quality of life of the patients and their relatives. At first, questionnaires were developed to evaluate QOL issues in adults. More recently, QOL questionnaires for children were constructed, as language and content of children's QOL measures need to be appropriate to their experience and development. Two types of questionnaires are now available for children: generic and condition-specific. Generic questionnaires (e.g., Child Health Questionnaire-CHQ; Landgraf, Abetz, & Ware, 1996) are applicable for all population subgroups and are useful for comparing outcomes between subgroups. Following a longitudinal study with the CHQ, Vargus-Adams (2006) concludes that children with CP (a) have lower CHQ scores than other children, (b) on average these scores were stable over the course of one year, and (c) CHQ scores were not measurably affected by common medical interventions. Condition-specific questionnaires are applicable to one specific population subgroup only, focus on relevant aspects of QOL of this subgroup, and are useful to detect small changes (Bjornson & McLaughlin, 2001).

Meningaud, Pitak-Arnnop, Chikhani, and Bertrand (2006) state that drooling can produce significant negative effects on QOL, especially in patients with chronic neurological disabilities. Recently, several condition-specific questionnaires for children with CP have been published. Examples are the PedsQL 3.0 CP Module (Varni et al., 2006), the CP QOL-Child (Waters et al., 2007), and the CPCHILD (Narayanan et al., 2006). However, the PedsQL 3.0 CP Module and the CP QOL-Child do not even mention drooling as a symptom, nor do they assess the impact of drooling on children or their parents. Only the CPCHILD contains one item addressing the child's difficulty to 'maintain oral hygiene (keep mouth and teeth clean)'. It should be noted, however, that this item does not exclusively refer to drooling. At the time the present study was carried out, there was no validated questionnaire on HRQOL available dealing with the impact of drooling on daily life and care, social interaction, and self-esteem of children with CP.

Treatment options for drooling

Treatment options for drooling can be divided in five categories. Surgery (1) is aimed at either presenting the saliva in the back of the mouth by rerouting salivary ducts, or at reducing salivary flow by excision of salivary glands, or ligation of salivary ducts. A reduction of salivary flow can also be obtained by BTX-A injections into the salivary glands (2) or by the systemic application of anti-cholinergic drugs (3). In addition to medical approaches, several conservative (paramedical and behavioural) treatment options can be distinguished. In oral motor therapy (4), the oral phase of swallowing is trained and sensibility in the mouth area is supposed to be stimulated. In addition, mouth closure and head and posture control may be optimized. Oral appliances, used in

orofacial regulation therapy, prevent saliva from dripping out of the mouth and stimulate an optimal swallowing act. Finally, behaviour therapy (5) focuses on the optimal performance of, for instance, swallowing and wiping behaviour during daily activities.

Recently, the Dutch Institute for Healthcare Improvement CBO has published evidence-based guidelines for diagnosis and treatment of drooling in children with CP which were formulated by a group of medical and paramedical professionals (CBO, 2007). Their conclusions describe the present state of the art on the treatment of drooling. In the next section, the five treatment options for drooling are described in more detail and the evidence for their effectiveness is discussed.

Surgery

Due to motor impairment, the first phase of swallowing, that is the oral phase, is very difficult for most drooling children. Surgical rerouting of the ducts of the submandibular glands is performed to prevent saliva from entering the mouth. By presenting the saliva in the back of the mouth, the oral phase of swallowing is skipped and a strong pharyngeal stimulus to swallow is presented. In addition or alternatively, salivary flow rate is reduced by excision of salivary glands or ligation of salivary ducts.

In the CBO-guidelines, it is concluded from case-series and cohort studies (Crysdale & White, 1989; Crysdale, Raveh, McCann, Roske, & Kotler, 2001; O'Dwyer & Conlon, 1997) that, presently, the combination of rerouting the ducts of the submandibular glands and excision of sublingual glands is a preferred surgical treatment procedure. Yet, Greensmith et al. (2005) report moderate drooling after this intervention. Before considering a rerouting procedure, it is very important to rule out the possibility of the presence of posterior drooling, which refers to saliva that is spilled over the tongue through the faucial isthmus. Whenever the trigger to swallow is impaired or missing, aspiration of pooled saliva into the trachea may occur. Evidently, the surgical rerouting procedure could easily aggravate this problem. If rerouting is not an option, a two or four duct ligation or excision of, for instance, the submandibular glands are the only surgical options. However, Martin and Conley (2007) evaluated 31 patients who underwent a 2, 3 or 4 duct ligation procedure three to six years after surgery and concluded that these intra-oral surgery procedures provided minimal long-term reduction of drooling. Additional medical and surgical treatment was needed in a significant number of their patients.

Botulinum Toxin type A injections

Bilateral injection of Botulinum Toxin type A (BTX-A) in the submandibular glands reduces (and in some cases completely eliminates) drooling in children with CP for a period up to 6 months (Benson & Daugherty, 2007; Bothwell et al., 2002; Jongerius, Rotteveel, et al., 2004; Jongerius, Van den Hoogen, et al., 2004; Savarese, Diamond, Elovic, & Millis, 2004; Suskind & Tilton, 2002). It is assumed that due to neural

sprouting, the submandibular glands become reinnervated after this period and drooling returns to baseline level. There is some evidence indicating that a combination of bilateral injections of BTX-A in both submandibular and parotid glands may be more effective than injection in the submandibular glands only (Suskind & Tilton, 2002).

To inject the BTX-A at the right spot in the glands, ultrasound guidance is recommended. General anaesthesia is given to prevent pain as well as to prevent the child from moving during the injection. No major side-effects have been reported, except for incidental temporary swallowing problems due to swelling immediately after the injection. Because of the temporary reductive effect of BTX-A, this procedure has to be repeated each time severe drooling reoccurs.

Anti-cholinergic drugs

By influencing the parasympatic nervous system as a whole, anticholinergic drugs such as Glycopyrrolate, Scopolamine, and Benztropine also affect the production of saliva in the salivary glands (Camp-Bruno, Winsberg, Green-Parsons, & Abrams, 1989; Jongerius, Van Tiel, Van Limbeek, Gabreëls, & Rotteveel, 2003; Lewis, Fontana, Mehallick, & Everett, 1994; Owen & Stern, 1992; Reddihough, Johnson, Staples, Hudson, & Exarchos, 1990). But because of its systemic effect, various side-effects have been reported, such as blurred vision (because of pupil dilatation), dizziness, irritability, restlessness, disorientation, constipation, etc.. Although there are methodological flaws in studies on anticholinergic drugs, there is some evidence that they reduce drooling, but it remains unclear to what extent. However, because of their side-effects, these drugs should not be used chronically.

Many children with CP suffer from gastro-oesophageal reflux (Reyes, Cash, Green, & Booth, 1993). Because episodes of gastro-oesophageal reflux increase salivary flow rate, Heine, Catto-Smith, and Reddihough (1996) hypothesized that antireflux medication could decrease drooling severity. Unfortunately, results from a double blinded, placebo controlled cross-over trial in 9 children with CP and severe drooling showed that this approach was ineffective.

Oral motor training and orofacial therapy

Oral motor therapy is aimed at improving lip closure, fostering appropriate head and posture control, efficiency training of the oral phase of swallowing, and increasing sensibility (e.g., by using the technique of icing and brushing). Although oral motor training is advocated in each review on the treatment of drooling, systematic research into this approach is lacking and the evidence for this type of treatment is anecdotal. The Dutch CBO group of medical and paramedical professionals conclude that severe drooling cannot be treated by oral motor training alone, although there are some reports suggesting that mild drooling can be treated effectively using oral motor training alone.

Several oral appliances have been developed in an attempt to modify and improve oral motor functioning (Limbrock, Hoyer, & Scheying, 1990; Gisel, Schwartz, & Haberfellner, 1999). Other than a few case reports, there has been little research done on this type of treatment. Johnson, Reid, Hazard, Desai, and Reddihough (2004) evaluated an oral appliance, the Innsbruck Sensori-Motor Activator and Regulator, on saliva control in children with CP. Only 6 of the 18 children completed the study, the others were withdrawn. A variety of reasons for low compliance with the program and withdrawal from the study are given. Although statistically significant changes in drooling severity scores were reported for children that completed the study, conclusions about the effectiveness of this treatment cannot be drawn because of low compliance.

Behavioural therapy

Behavioural principles underlie most educational approaches used to teach adaptive behaviours to individuals with developmental disabilities and to reduce their problem behaviours (Duker, Didden, & Sigafoos, 2004). The phenomenon of drooling can also be conceived of as a behavioural problem: behaviours like swallowing and wiping are not performed at a sufficient frequency and as a result, drooling may occur. Behavioural therapy is aimed at increasing or decreasing the frequency of a target behaviour and/or collateral behaviours. In the case of drooling, the treatment procedure may be aimed at, for instance, increasing the frequency of swallowing, wiping, and/or improving head control. After individual functional analysis of these behaviours and observation of their frequency of occurrence, the target behaviour and intervention goal are determined. In behavioural therapy antecedent stimuli (that is, behaviours or events that precede the target behaviour) and/or consequent stimuli (behaviours or events that follow the target behaviour) are systematically analyzed and manipulated to alter the probability of occurrence of the target behaviour. Prior to the present study, no systematic reviews had been published on behavioural treatment for drooling. Also, the CBO (2007) failed to include behavioural therapy in their analysis of treatment options for drooling.

Outline of the study and research questions

In 2000, a controlled clinical trial was started during which children with CP who suffered from severe drooling were subsequently treated with scopolamine and BTX-A. A reduction of salivary flow was reported as an effect of both anticholinergic agents (Jongerius, Rotteveel, et al., 2004) and this reduction in salivary flow also resulted in a significant reduction in drooling in a standardized situation, as measured by the drooling quotient (Jongerius, Van den Hoogen, et al., 2004). Visual analogue scales to register parental opinion about the severity of drooling at home also showed diminished drooling (Jongerius, Van den Hoogen, et al., 2004). The objective of this thesis is to evaluate changes in drooling severity in daily life situations and its consequences for provision of

care as well as to evaluate changes in the child's social interaction, and self-esteem as a result of this treatment.

Although medical and paramedical treatment options for drooling have been reviewed repeatedly, including an analysis of their scientific evidence, options for behavioural treatment for drooling were not included in these reviews. In this thesis, we examined the evidence of behavioural treatment for drooling. In addition, we summarized components of behavioural procedures that are reported to be effective in the treatment of drooling. Finally, we conducted a pilot study on the effectiveness of a self-management program for drooling.

Research questions are:

1. Can we measure the differential impact of drooling on a subgroup of children with cerebral palsy with severe drooling?
2. Does a reduction in drooling (after consecutive treatment with scopolamine and bilateral injection of BTX-A in the submandibular glands), change the impact of drooling in daily life situations, provision of care, social interaction, and self-esteem?
3. Is there scientific evidence for the effectiveness of behavioural treatment for drooling?
4. What kind of behavioural procedures are reported to be effective in the treatment of drooling?
5. Is a self-management program effective in reducing severe drooling?

In chapter 2 of this thesis, two parent questionnaires on the impact of drooling are presented and the impact of drooling on daily life, social interaction, and self-esteem of children with cerebral palsy is illustrated. In chapters 3 and 4 the longitudinal effects of reduced salivary flow after bilateral injection of BTX-A in the submandibular glands on respectively daily life and care, social interaction, and self-esteem are described. Chapters 5 and 6 are reviews on behavioural treatment for drooling. Chapter 5 is a descriptive analysis of studies on behavioural treatment of drooling that appeared between 1970 and 2005. It focuses on available scientific evidence for the effectiveness of behavioural treatment for drooling. Chapter 6 provides a methodological critique of the literature on behavioural treatment for drooling and presents clinical guidelines and suggestions for future research. Finally, chapter 7 describes a case series study in which a self-management procedure to treat drooling is evaluated. In the general discussion conclusions and implications of the results are discussed. A summary completes this thesis.

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Chapter 2

Drooling in children with cerebral palsy: A qualitative method to evaluate parental perceptions of its impact on daily life, social interaction, and self-esteem

International Journal of Rehabilitation Research, 2006, 29, 179-182

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Abstract

Research on the treatment of drooling applies measures such as salivary flow rate, qualitative observations of drooling severity in standardized situations, and anecdotal or one-dimensional parental and teacher reports. To assess drooling severity in a range of everyday conditions, and its impact on the daily life of children and their families, two parent questionnaires were constructed. Results of baseline measurements of 43 children with Cerebral Palsy showed that the questionnaires measured the variation in drooling severity across daily life conditions, and enabled evaluation of the impact of drooling on the ability to eat, drink, and speak, on daily care, economic consequences, and social interactions. The section on the impact of drooling on self-esteem appeared not to be fully applicable for non-speaking children with a low developmental status. The questionnaires offer a qualitative method to evaluate parental perceptions of the impact of drooling and evaluate the effectiveness of interventions to reduce drooling.

Introduction

Drooling is reported to be a significant problem in 10 to 37.5% of patients with Cerebral Palsy (CP) (Van de Heyning, Marquet, & Creten, 1980). Several treatment strategies to reduce drooling have been reported such as oral motor training, behavioural therapy, the application of intra-oral devices, medication, and surgery. Because these interventions differ in intensity, duration, and invasiveness, and because they focus on different aspects of the complex problem of drooling, it is obvious that the effects and side-effects on the child's functioning vary as well.

To evaluate drooling severity and the effect of interventions, different strategies have been applied. Measuring salivary flow from the parotid and submandibular glands by the swab method (Ahlner & Lind, 1983) is highly accurate in evaluating changes in saliva production. However, unobtrusive measurement of drooling without the child being aware of it often cannot easily be obtained at a hospital or research department and a statistically significant change in salivary flow does not always necessarily imply a change in drooling severity or an amelioration of related problems in the child's daily life. To measure drooling severity in a real-life situation, Rapp (1980) observed drooling children during two daily activities, each lasting 10 minutes. Using a time-sampling method, drooling, defined as a 'string of dribble', was evaluated at 40 randomly chosen moments. The 'drooling quotient' was defined as the percentage of drooling from 80 cued observations. While this measure seems more relevant in the functional evaluation of drooling, the child's level of activity during observation threatens the validity because most children drool more when concentrating on tasks (Blasco & Allaire, 1992). The degree of drooling in daily life can also be estimated by parents and teachers on ordinal

scales (e.g. the Teacher Drool Scale; Camp-Bruno, Winsberg, Green-Parsons, & Abrams, 1989) or visual analogue scales (VAS).

Blasco (2002) questions the validity of quantitative measures of drooling and proposed the number of bandanas or shirts soaked-through in the course of a 'typical day', a 'bad day', and a 'best day' as the best semi-quantitative measure for judging treatment efficacy. He states: "The ultimate test of whether treatment will be continued or not is whether it makes the caregiver's lives easier and their child's life is improved" (p. 779). Therefore, we compiled two parent questionnaires to investigate drooling severity and its consequences for the child and the family. Results of baseline measurements in a group of severely drooling children with CP are presented.

Method

Questionnaires on drooling

Two questionnaires were constructed (see Appendix A), in cooperation with parents of drooling children who did not participate in this study. The first questionnaire covers drooling severity in specific daily life situations, the impact on oral motor activities and daily care, and economic consequences. The other questionnaire investigates the impact of drooling on social interaction and self-esteem. The first questionnaire is likely to show changes immediately after interventions, while changes in social interaction and self-esteem occur gradually with time. Thus, with two questionnaires, data can be collected at the optimal time interval for each instrument.

- *The severity of drooling in specific daily life situations*

To obtain an initial general impression, parents are asked to score drooling severity over the previous 2 weeks on a VAS scale (measuring 10 cm). Because drooling severity varies under different circumstances, additional questions focus on drooling severity at different times throughout the day and during a range of specific daily life situations (five-point ordinal scale).

- *The impact of drooling on the ability to speak, eat, and drink*

Oral motor dysfunction is a central problem in drooling children with CP, presenting inability to speak, and difficulties in eating and drinking. Interventions proposed for drooling can have both positive and negative effects on these abilities. For instance, wearing an oral appliance can reduce the intelligibility of speech, whereas reduction of salivary flow may impair the ability to swallow.

- *The impact of drooling on daily care and economic consequences*

This section investigates specific kinds of caring activities and its frequencies (supply of bibs and shawls, encouraging to swallow, wiping the mouth and chin, change of clothes, loads of wash per week), and also evaluates the damage caused to clothes, toys, books, furniture, and other equipment.

- *The impact of drooling on social interaction*

Drooling negatively influences the physical appearance of the child. As a consequence, social interaction at school and at home with peers and adults, and perhaps even within the family, is affected.

- *The impact of drooling on self-esteem*

Both parental judgment and overt reactions of the child relating to self-esteem are evaluated. Parents answer questions based on the section 'self-esteem' of the Child Health Questionnaire (CHQ-PF50; Landgraf, Abetz, & Ware, 1996): 'How satisfied do you think your child has felt about his or her (1) school ability; (2) athletic ability; (3) friendships (social contact with other children); (4) physical appearance; (5) family relationships; and (6) life overall'. In addition, parents indicate to what extent ('not at all' versus 'very considerably') drooling contributes to the level of satisfaction of the child in these areas. Furthermore, parents report their child's overt positive and negative feelings about their physical appearance, competence, and social acceptance by peers and adults. In the case of negative feelings, parents indicate if drooling is referred to as a contributing factor.

Subjects

Forty-five children were included in this study. Inclusion criteria were: pre-school and school-aged children, diagnosed with CP, for whom spasticity was the main movement problem, and who had a score of 3 or higher on the Teacher Drool Scale (Camp-Bruno et al., 1989). Written informed consent was obtained.

Results

Parents of two children did not return the questionnaires. Consequently the data of 43 children could be evaluated. The children ranged in age from 3.3 to 17.0 years (median 9.1 years). Twenty-seven were boys and 16 were girls. Eight children were ambulant, nine children had electric wheelchairs, and the other 26 used self-propelled wheelchairs. Twenty-two children could not talk. None of the children attended mainstream schools: 29 attended a special education school, while 14 attended a daycare center for children with developmental disabilities. The developmental level of 32 children was below 6 years (of which 23 scored below 4 years) as determined by either chronological age or mental capacities.

- *The severity of drooling in specific daily life conditions*

In 36 children drooling was present during the whole day and less severe at night. Three children were constantly wet, day and night. Two children drooled the most in the afternoon, one mainly in the evening and while asleep. Table 2.1 presents the specific conditions for which severe drooling was reported. Each cluster starts with the condition most frequently scored. Some parents indicated that specific conditions were not relevant

Table 2.1 Severity of drooling across conditions[a]

Condition	Children with drooling (*n*)	
	Severe[b]	**All drooling[c]**
Level of activity		
Concentrated activity	34	38
Strenuous activity	33	37
Intensive movement (sports)	29	30
Relaxed, watching TV	23	38
Walking	11	17
Sleeping	4	32
Sensorimotor factors		
Drinking	22	31
Eating	21	38
Certain tastes or foodstuffs	17	25
After brushing teeth	15	33
Talking	10	20
Smell of food	10	23
Body position		
Unsupported sit	31	38
Supported sit	29	40
Prone position	28	37
Supine position	6	34
Mood and physical condition		
Enthusiastic	32	38
Tired	30	37
Sick	22	32
Cries	22	32
Medical factors		
Allergic respiratory reactions	16	18
Swallowing medication	10	21
After swallowing medication	9	20

[a] Severity of drooling expressed in a five-point scale ranging from no drooling (= 1) to very severe drooling (= 5)
[b] Number of children with severe drooling (score 4 and 5)
[c] Number of children with any drooling in the condition (score 2 - 5)

for their child. Severe drooling occurred in conditions with either high or low activity level. In these cases the child did not pay attention to the need to swallow. During oral motor activity many children also drool severely. Severe drooling is also reported when the child is enthusiastic, tired, sick, or crying.

- *The impact of drooling on the ability to speak, eat, and drink*

The quality of speech was evaluated as poor (VAS < 33: *n* = 7), intermediate (VAS ≥ 33 and ≤ 66: *n* = 19), and good (VAS > 66: *n* = 17). The intelligibility of speech was estimated to be better for parents than for others. Forty-one children were on oral feeding, one had tube feeding, and one partial tube feeding. Most parents were satisfied with their child's eating and drinking capacity, six couples were dissatisfied about eating and 11 about drinking.

- *The impact of drooling on daily care and economic consequences*

Drooling influences the care given to the child. Twenty-three children were told to swallow on average 28 times per day. For 31 children the mouth and chin were wiped on average 74 times per day. Most children wore bibs (*n* = 36) or shawls (*n* = 15) that had to be replaced on average 6 or 7 times a day. Four children wore terry cloth wristbands. Thirty-five children received a new change of clothes during the day because of their drooling and 20 were changed more than once. On average, parents ran nine (range 2 - 25) loads of laundry per week.

As a result of drooling, parents reported damage to clothes (*n* = 33), toys (*n* = 20), books (*n* = 21), and furniture (*n* = 19). In addition, communication aids (*n* = 12), electronic communication devices (*n* = 9), computers (*n* = 12), and audio equipment (*n* = 8) were also damaged. Because of drooling, 18 of the parents reported curtailing the child's play with objects that might be damaged. Only 11 provided their child with plasticized toys or books.

- *The impact of drooling on social interaction*

Drooling is an important negative factor in the social interaction of these children (Table 2.2). Play and social interaction with children and adults is reported to be limited; for eight children even their own parents kept their distance. The parents of eight children

Table 2.2 Social consequences of drooling

	Handicap related (*n*)	**Drooling related (*n*)**
Social interaction with children		
Does not play with other children at school	10	0
Does not play with other children at home	14	5
Is avoided by other children	19	16
Social interaction with adults		
Is avoided by familiar or unfamiliar adults	16	14
Adults keep their distance	19	19
Parents keep their distance		8
Underestimation of mental ability	25	8

also reported that the unsightly look of drooling causes outsiders to assess the child as being less mentally capable than they actually are.

- *The impact of drooling on self-esteem*

The parents of eight children reported that their child was unsatisfied in one or more domains (Table 2.3). While four of the children were not happy about their social contact with peers, drooling was only cited as the main reason for two. According to the parents, none of the children was dissatisfied with his or her physical appearance. Twenty-three children were unable to express their feelings about the emotional aspects of drooling because of low developmental age (i.e. < 4 years). Thus, only 20 children could be evaluated (Table 2.4). Based on parental judgment, only a few children were reported to show overt emotional reactions to the impact of drooling on their physical appearance, their competence, and their social acceptance by adults and peers.

Table 2.3 Parental evaluation of the emotional consequences of drooling

	Not satisfied (*n*)	**Not satisfied because of drooling (*n*)**
School abilities	2	0
Participation in sports and play	3	0
Social contact with other children	4	2
Physical appearance	0	
Relations within the extended family	2	1
Total quality of life	3	0

Table 2.4 Emotional reactions of the child (*n* = 20)

	Positive (*n*)	**Negative or mixed (*n*)**	**Negative because of drooling (*n*)**
Physical appearance	3	3	3
Competence	10	4	2
Social acceptance by adults	7	1	0
Peer acceptance	3	3	2

Discussion

The presented questionnaires measure parental perceptions of variation in drooling severity across conditions in daily life and evaluate its consequences. Results from our sample show that drooling is increased where attention is diverted from control of the salivary flow, and/or when oral, or general, motor activity demands attention. Saliva control, through regular and effective swallowing during activity, is a dual task, which is difficult for children with CP. The consequences of drooling for the children and their families are considerable. Most authors recite the effects numerically to justify interventions. Anecdotic parental remarks concerning the results of drooling treatment are reported, but not as part of a systematic data collection programme. Our questionnaires enable the systematic evaluation of parental perceptions of the impact of drooling on the ability to eat, drink, and speak, on daily care and economic consequences, and on social interaction and self-esteem.

Less than half of the children from our sample are reported to experience negative consequences in social interaction at school or at home because of their drooling condition. This might mean that the impact of drooling on social interaction is either overestimated or neglected. Parents in our study often note that their drooling child is 'just like the other children' in their special school or daycare center. In these sheltered social environments, drooling seems to be accepted or ignored. Parental judgment on this issue, however, might be not sufficient to fully evaluate the impact of drooling. Possibly an additional judgment by the child's teacher would give a more realistic impression of social interaction.

From our group, only seven children expressed emotional reactions in relation to their drooling. The low developmental age of our subjects (23 below 4 years, 32 below 6 years) could explain this finding; a child needs to have sufficient social perspective to evaluate the impact of drooling and to make their feelings known. Children learn to label their characteristics in increasing differentiating categories between 2 and 7 years of age, but at age 7 the categories often lack stability and children cannot systematically test hypotheses about those characteristics that define the self. Logical thinking starts to emerge at 6 years of age, leading to self-knowledge in the form of attributes and categories that can be applied to the self. From this age on, children also gradually develop perspective skills that enable them to imagine what other people think, and in particular what others think of them (Harter, 1983). Teplin, Howard, and O'Connor (1981) found that mainstreamed children with cerebral palsy of normal intelligence begin to regard themselves as different as early as four years of age. These self-views, and their potentially negative effects on self-esteem, do not appear to crystallize until the children are in the primary grades at school. These developmental features lag behind in retarded children. In addition, for non-speaking children, it is even more difficult to

express feelings about themselves. For these reasons, the questions on self-esteem are applicable to only some of the children in the present study. Nevertheless, for those children who are able to express their feelings, this part of the questionnaire is very relevant.

Reliability of the presented data was not assessed. Theoretically, the subjective nature of parents' impressions could lead to over- and/or underestimation of the impact of drooling. However, Rosenbaum and Saigal (1996) report that parents serve as an excellent proxy for their children when assessing Health Related Quality of Life issues. Because the questionnaires are extensive, we did not build in extra questions to control for inconsistencies. It is recommended that during longitudinal effect studies the same parent (whether the father or the mother) fills in the questionnaires to control for intersubject variations. To prevent deliberate overestimation of the impact of drooling, the questionnaires should be distributed after inclusion for treatment.

In addition to quantitative measures from laboratory settings, these questionnaires offer a qualitative method to evaluate parental perceptions of the impact of drooling on daily life, social interaction, and self-esteem. Repeated administration of the questionnaires, before and after interventions, can augment the evaluation of their efficacy. Eventually, comparison of the effect of treatment strategies on these variables can support decision making regarding the preferred treatment strategy for an individual child or subgroups of children.

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Chapter 3

Drooling in children with cerebral palsy: Effect of salivary flow reduction on daily life and care

Developmental Medicine and Child Neurology, 2006, 48, 103-107

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Abstract

The purpose of this study was to investigate the effect of salivary flow reduction on daily life and provision of care in children with cerebral palsy (CP). Parents of children with CP were asked to fill in a questionnaire on the impact of drooling on the daily life of their children and their families and the data were then analyzed. Forty-five children with severe drooling (28 males, 17 females; mean age 9 years 5 months [*SD* 3 years 7 months]; range 3 to 16 years) were monitored before and after receiving medication (scopolamine and Botulinum Toxin type A) to reduce salivary flow. Type of CP included hypotonia (*n* = 1), spastic paresis (*n* = 27), and mixed motor disorders with spastic and dyskinetic paresis (*n* = 17). Eight children were independently ambulant, 37 children were wheelchair users. Thirty-four children had learning disability with a developmental age of below 6 years. Six participants dropped out of the study; data on 39 children were analyzed. Results showed that anticholinergic agents effectively reduced salivary flow. Drooling diminished substantially and this was accompanied by a significant reduction in care needs, making daily care less demanding. The amount of reported damages to communication devices and computers decreased. In addition to the evaluation of primary variables, such as the salivary flow rate, investigation of impact on daily life provides useful information about the outcome of treatment for drooling.

Introduction

Drooling in children with cerebral palsy (CP) is primarily due to oral motor dysfunction (Harris & Purdy, 1987; Hussein, Kershaw, Tahmassebi, & Fayle, 1998). It occurs in 10 to 37.5% of patients with CP (Ekedahl & Hallen, 1973; Van de Heyning, Marquet, & Creten, 1980). According to Tahmassebi and Curzon (2003) this percentage is even higher among CP children attending special schools: 58% (93 of 160) children drooled; 33% (53 of 160) drooled severely. In another population of 1437 children with CP, 34% of the parents reported that drooling was 'sometimes a problem, requiring a bib', and another 16% responded that it was 'often a problem, requiring daily clothing changes' (Bachrach, Walter, & Trzcinski, 1998).

The evaluation of interventions is mostly directed towards salivary flow. However, in some studies parents also answered open-end questions concerning the severity of drooling to evaluate treatment (Bothwell et al., 2002; Puckett, Concannan, McNaul, & Barone, 1993; Savarese, Diamond, Elovic, & Millis, 2004). It is reasonable to assume that drooling affects the daily life and care for these children. So far, no validated questionnaire has been available that addresses specific problems such as drooling in children with CP.

A controlled clinical trial was conducted recently during which children with CP were treated with scopolamine and Botulinum Toxin type A (BTX-A). A reduction of

salivary flow was seen for both anticholinergic agents (Jongerius, Rotteveel, et al., 2004); reduced salivary flow resulted in a significant reduction in drooling (Jongerius, Van den Hoogen, et al., 2004).

The objective of this study was to evaluate changes in daily life and provision of care as a result of drooling treatment. The impact of drooling on daily life was investigated before and after treatment. This was performed using a questionnaire, developed especially for this study, on items of daily living and daily care.

Method

Patients

From the outpatient clinic of the University Medical Centre, Nijmegen, the Netherlands, 45 patients with severe drooling and a diagnosis of CP were included in a controlled clinical trial as consecutive participants recruited mainly by indirect advertisement. There was no specific selection before the first outpatient visit. The effect on drooling of intraglandular BTX-A injections into the submandibular glands versus the application of scopolamine patches was investigated. The results of this part of the study was described elsewhere (Jongerius, Rotteveel, et al., 2004; Jongerius, Van den Hoogen, et al., 2004). Inclusion criteria were: males and females of pre-school and school age, diagnosis of CP, and a score of 3 or higher on the Teacher Drool Scale (TDS; Camp-Bruno, Winsberg, Green-Parsons, & Abrams, 1989). The TDS comprises a 5-point scale to define the degree of drooling: (1) 'No drooling', (3) 'Occasional drooling, on and off all day', (5) 'Constant drooling, always wet'. The TDS was only scored at entrance of the study so that conclusions could be drawn about drooling severity.

All drugs used were carefully evaluated to assess their influence on saliva secretion. Continuous use of anticholinergic drugs or benzodiazepines was not permitted in the study and no change in medication was allowed. Medication to treat drooling had to be stopped at least three months before participation. No child was excluded because of use of medication. Of all patients, 21 did not use medication before the study, four patients used benzodiazepines to treat epileptic seizures, and two patients ceased taking anti-drooling medication before the study commenced. None of the subjects had been administered BTX-A before.

Participants comprised 28 males, 17 females and ranged in age from 3 to 16 years (mean 9 years and 5 months, *SD* 3 years and 7 months). Types of CP included: hypotonia (n = 1), spastic paresis (n = 27), and mixed motor disorders with spastic and dyskinetic paresis (n = 17). Eight children were independently ambulant and 37 children were wheelchair users; 22 children did not use speech to communicate. None of the children attended mainstream schools: 29 attended a special school, and 16 attended a daycare centre for children with learning disability. Thirty-four children had learning disability and a

developmental age below 6 years as determined by psychological investigation using Dutch instruments. Written informed consent was obtained from parents after all possible adverse effects and risks related to the study had been explained to them. The Hospital's Human Research Committee approved the study.

Data collection

An extended questionnaire was developed to evaluate the impact of drooling on items of daily living (Appendix A, Questionnaire 1). The prerequisite for the items was that a team, consisting of a speech therapist, clinical pediatric psychologist, and physician in rehabilitation medicine, reached consensus on selected items regarding whether they reflected relevant aspects of the impact of drooling on daily life. Parents of three children who participated in a pilot study were invited to comment on the preliminary version of the questionnaire and this resulted in some adjustments (Jongerius et al., 2001). The questionnaire contained open-ended as well as multiple-choice questions. In addition, parents used several visual analogue scales to give their opinion on some statements about drooling and its consequences.

Data from two sections of the questionnaire are presented. The first section contains questions about drooling severity in specific daily situations, the second section contains questions about practical consequences for daily care that may be influenced by drooling itself, or, possibly, could improve after treatment of drooling (see Appendix A, Questionnaire 1).

Detailed instructions were included in the introduction of the questionnaires. Parents were asked to score average severity of each item as registered in the two weeks before assessment. Assessments were made at baseline, during the use of scopolamine, and 4, 8, 16, and 24 weeks following BTX-A injections.

Data handling

There were some missing values because of incomplete responses, non-interpretable answers, or unanswered questions. Before data analysis, missing values were imputed by using the method of 'carrying the last observation forward', meaning that missing data after scopolamine and BTX-A injections were replaced by the last available score on the same item. Missing values at baseline were estimated by imputing the mean (in case of a numerical score) or the median (in case of an ordinal score) of all participating children.

Statistical analysis

From all investigated items, we identified those situations for which the parents of at least 50% of the children scored 'severe drooling' at baseline (Table 3.1). To evaluate the severity of drooling in these situations over time we used an analysis of variance (ANOVA) for repeated measurements with degree of drooling score as the dependent variable.

To evaluate the effect size for these items, Cohen's *d* was calculated by comparing the mean baseline score of all patients with the mean at follow-up. The effect size was defined as 'large' in cases where Cohen's *d* exceeded 0.8 (Cohen, 1988).

To evaluate changes in daily parental care, ANOVA for repeated measurements was used for each individual item and Cohen's *d* was estimated. Non-parametric tests were conducted on appropriate items, such as damage to clothing, toys, books, furniture, communication aids, electric devices, or computers.

A *p*-value equal to or less than 0.05 was required for all tests. Because of multiple testing in tests of within-participant effects, Bonferroni correction was used (i.e. 0.05/number of tests).

Results

During the trial six children dropped out: four children could not complete scopolamine treatment, one child became ill (not related to trial), and one had a change of medication outside the study criteria. Data of 39 children were analyzed. A response rate of 85% was achieved.

Baseline investigations

Table 3.1 presents conditions in which severe drooling occurred. Each cluster starts with the condition most frequently scored. Because some items were not applicable for each of the children, the actual number of involved individuals is given. Severe drooling occurred particularly during strenuous activities with a high concentration level, such as 'walking' and 'sport'. Severe drooling was also observed when insufficient attention was given to swallowing (e.g. when watching television).

Severe drooling appeared to influence daily care and attention given to the children. For example, 18 children were told to swallow a mean of 25 times per day. The chins of 26 children were wiped a mean 73 times per day. Thirty-one children wore bibs or shawls which had to be replaced a mean of seven times a day. Thirty-one children were redressed every day, 16 more than once a day. Five children wore terry cloth wristbands because of drooling. Parents had a mean of nine (range 2-25) loads of wash per week. Damage to clothes (n = 30), toys (n = 17), books (n = 17), and furniture (n = 18) was also reported. In addition, communication aids (n = 9), electronic communication devices (n = 8), computers (n = 10), and audio equipment (n = 6) were damaged by drooling. Sixteen parents reported curtailing the child's play with objects that could be damaged.

Table 3.1 Severity of drooling across conditions[a]

Condition	Children with drooling (*n*)	
	Severe[b]	All drooling[c]
Level of activity		
Concentrated activity	29	33
Strenuous activity	28	32
Intensive movement (sports)	24	25
Relaxed, watching TV	21	33
Walking[d]	11	17
Sleeping	4	28
Sensorimotor factors		
Eating	20	34
Drinking	19	26
Certain tastes or foodstuffs	16	21
After brushing teeth	13	29
Teething	12	15
Talking	9	17
Smell of food	9	19
Body position		
Unsupported sitting	26	33
Prone position	25	33
Supported sitting	24	35
Supine position	6	30
Mood and physical condition		
Enthusiastic	27	33
Tired	25	32
Unwell	19	29
Cries	18	27
Medical factors		
Broncho-pulmonary reactions	14	16
Using medication	9	18
After taking medication	8	17

[a] Severity of drooling expressed in a five-point scale ranging from no drooling (= 1) to very severe drooling (= 5)
[b] Number of children with severe drooling (score 4 and 5)
[c] Number of children with any drooling in the condition (score 2-5)
[d] Including assisted walking

Table 3.2 Multivariate test, tests of within-participant effects, and effect size of drooling in daily living following intervention

Situation	**Multivariate test**	**Tests of within-participant effects**		**Effect-size: Cohen's *d***				
	HT^2 (*p*)	***F* (df_1, df_2)**	***p*[a]**	**Sc**	**B4**	**B8**	**B16**	**B24**
Concentrated activity	1.65 (0.002)	5.4 (5, 110)	<0.001	5.530	4.009	4.880	4.331	3.254
Strenuous activity	1.48 (0.002)	7.2 (5, 115)	<0.001	6.580	4.491	5.532	4.848	3.235
Intensive movement (sports)	1.40 (0.056)	4,5 (5, 75)	0.001	4.530	3.785	4.535	4.755	3.437
Relaxed, watching TV	1,23 (0.008)	6,4 (5, 110)	<0.001	4.350	3.846	3.939	4.623	2.287
Eating	2.21 (<0.000)	6.1 (5, 120)	<0.001	4.647	3.675	3.728	4.290	2.441
Drinking	1.57 (0.009)	3.4 (5, 95)	0.007	2.956	2.460	3.099	2.404	1.475
Unsupported sitting	1.56 (0.001)	6.7 (5, 120)	<0.001	5.053	5.106	5.690	5.246	3.613
Prone position	2.82 (<0.001)	6.9 (5, 110)	<0.001	4.648	5.103	4.548	4.330	2.004
Supported sitting	1.86 (<0.001)	8.2 (5, 145)	<0.001	6.472	5.555	6.087	5.613	4.489
Enthusiastic	1.13 (0.009)	6.2 (5, 115)	<0.001	6.029	4.397	4.763	4.058	2.684
Tired	0.91 (0.046)	6.2 (5, 100)	<0.001	4.350	4.191	3.994	4.191	2.218
Unwell	1.91 (0.248)	0.8 (5, 45)	0.561	- 0.339	0.434	0.761	1.449	-0.443

HT^2, Hotelling's Trace; *df*, degrees of freedom; Sc, scopolamine treatment; B4, B8, B16, B24, 4, 8, 16, and 24 weeks after BTX-A injections.
[a] Significance set at $p < 0.004$.

Daily living: *repeated measures analyses*

A multivariate ANOVA (MANOVA) with a repeated measurements design was conducted for items on which at least 50% of the population demonstrated severe drooling at baseline. Significant changes after intervention were recorded for nearly all situations (Table 3.2). Significance was not reached for 'intensive movement' (Hotelling's trace [HT^2] = 1.40; $p = 0.056$) and 'is unwell' ($HT^2 = 1.91$; $p = 0.248$). In all other daily situations, drooling severity decreased significantly during scopolamine application and 4, 8, 16, and 24 weeks after BTX-A injection (Figure 3.1). Except for 'is unwell', Cohen's *d* exceeded 0.8 in all comparisons, indicating a large effect size (Table 3.2).

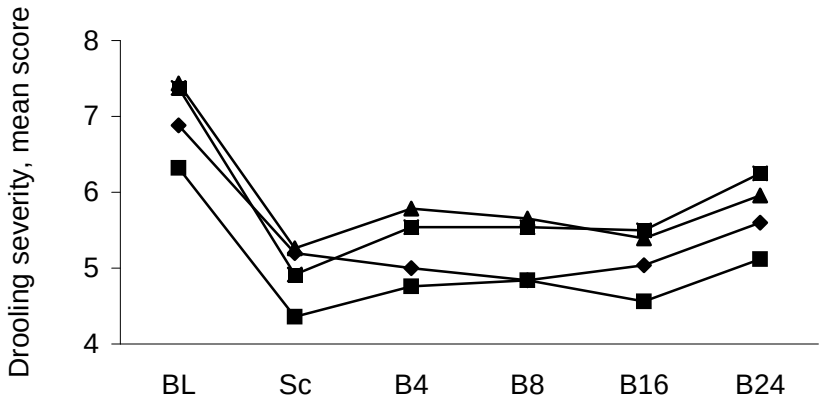

Figure 3.1 Drooling severity scores at baseline (BL), scopolamine treatment (Sc), and 4, 8, 16, and 24 (B4, B8, B16, and B24) weeks after BTX-A injection. ♦, unsupported sitting; ■, eating; ▲, concentrated activity; ■, enthusiastic.

Practical consequences: repeated measures analyses

Measures taken to minimize the practical consequences of drooling (provision of bibs, shawls, terry cloth wristbands, plasticized toys, or books) were analysed. A MANOVA with a repeated measurement design did not show significant change over time in the *number* of separate measures. In addition, no significant change was found in the *frequency* of urging the child to swallow (HT^2 = 0.23; p = 0.289), changing shirts or sweaters (HT^2 = 0.34; p = 0.094), changing pants (HT^2 = 0.12; p = 0.620), and the number of wash loads per week (HT^2 = 0.06; p = 0.881).

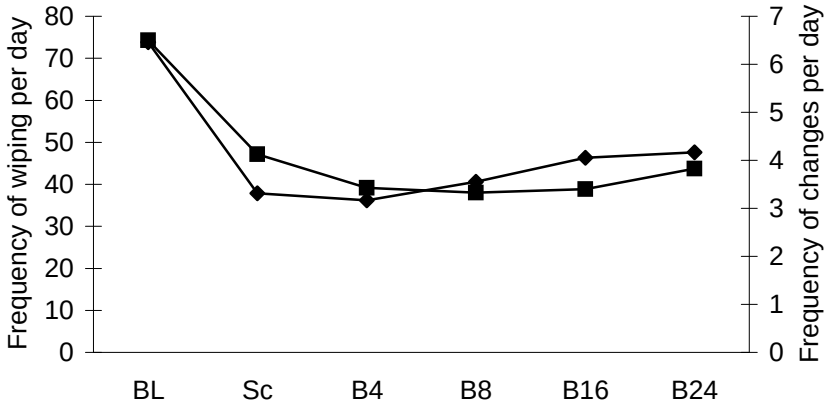

Figure 3.2 Frequency of specific actions at baseline (BL), scopolamine treatment (Sc), and 4, 8, 16, and 24 (B4, B8, B16, and B24) weeks after BTX-A injection. ■, frequency of wiping children's mouths and chins; ♦, mean frequency of changing bibs or shawls

However, a significant decrease was demonstrated in the *frequency* of wiping the child's mouth or chin (Fig. 3.2). For this item, multivariate tests (HT^2 = 0.53; p = 0.033) and the tests of within-participants effects (ANOVA F = 6.7; df = 5,155; p < 0.001) were significant and Cohen's d ranged from 2.350 to 3.463 comparing baseline scores with the scores during scopolamine treatment or 4, 8, 16, and 24 weeks after BTX-A injection.

The frequency of changing bibs or shawls decreased significantly (Fig. 3.2). Multivariate tests (HT^2 = 0.79; p = 0.002) and tests of within-participant effects (ANOVA F = 7.6; df = 5,175; p < 0.001) were significant, Cohen's d ranged from 2.459 to 3.683.

No significant change in the damage to cloths, toys, books, or furniture was reported as a result of therapy. However, a continuous decrease existed, from baseline up to 24 weeks after BTX-A injection, in the damage to communication aids, electric devices, and computers (Chi-square = 13,11; df = 5; p = 0.022).

Discussion

Daily care of children with drooling appears to be demanding and has practical consequences. A detailed questionnaire was developed to document the impact of drooling on daily life and provision of care. A significant reduction in salivary flow rate and drooling was demonstrated as a result of the administration of anticholinergic agents (Jongerius, Rotteveel, et al., 2004; Jongerius, Van den Hoogen, et al., 2004). This was accompanied by a decrease in the frequency of wiping the children's mouths and chins and the frequency of changes of bibs or shawls, making daily care less demanding. Additionally, a decrease in damage to communication aids, electric devices, and computers was demonstrated. No significant change occurred for the items: 'the frequency of changing clothes', or 'the number of wash loads per week'. Frequencies of changing clothes probably did not change because there might have been reasons other than drooling for changing clothes, for which we did not score. This could also explain why there was no change found in the number of washes. Despite a reduction of drooling, parents may have continued to use bibs or shawls for reasons other than drooling, leaving the amount of clothes to be washed at the same level.

This study demonstrated that a reduction in salivary flow has significant positive effects on daily life and provision of care. The reduction of salivary flow should not be the only target in itself in the treatment of drooling. It is recommended that drooling treatments (surgery, medication, behaviour therapy, etc.) are also evaluated in relation to impact on daily life as described in the current questionnaire.

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Chapter 3

Supplement

Drooling in children with cerebral palsy: effect of salivary flow reduction on daily life and care
Additional results

Effects of salivary flow reduction on eating, drinking, and speech

In the published articles (chapter 3 to 4) we did not report on the effect of Botulinum Toxin type A (BTX-A) treatment on eating, drinking, and speech. These data are presented in this supplement.

Eating, drinking, and speech: repeated measures analyses

No significant changes were demonstrated in a MANOVA with repeated measures design for the ability to drink and eat. However, from tests of within-participant contrasts it appeared that parents were significantly less satisfied about their child's eating after scopolamine treatment compared to baseline, but not after BTX-A treatment.

For changes in the ability to speak, only 12 children could be analysed (the others were non-speaking). Therefore, we cannot draw firm conclusions. Nevertheless, *t*-tests suggested that the intelligibility of speech for parents significantly improved (t = -1,870; one sided p = 0.044) only at 4 weeks after BTX-A treatment. The intelligibility of speech for outsiders (those who do not interact with the child on a daily base) significantly improved after scopolamine (t = -2,376; one sided p = 0.018) and 4 weeks (t = -2,517; one sided p = 0.015) and 16 weeks (t = -2,426; one sided p = 0.016) after BTX-A treatment. Satisfaction about speech also significantly increased after scopolamine (t = -2,421; one sided p = 0.017) and 4 weeks (t = -3,654; one sided p = 0.002) and 16 weeks (t = -2,838; one sided p = 0.008) after BTX-A treatment.

Chapter 4

Social interaction and self-esteem of children with cerebral palsy after treatment for severe drooling

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Abstract

The impact of salivary flow reduction following medication (scopolamine and Botulinum Toxin type A [BTX-A]) on social interaction and emotional development (self-esteem) was evaluated in a group of 45 children with cerebral palsy who suffered from severe drooling. The children ranged in age from 3 to 16 years (median 9.1 years); 28 were male, 17 female. A questionnaire to document the impact of drooling on social interaction and self-esteem for both the children and their parents was developed and administered during the use of scopolamine and up to 24 weeks after intraglandular BTX-A injections in the submandibular glands. The reduction of drooling was related to increased social contacts with peers. In addition, parents perceived that the impact of drooling on the level of the child's satisfaction on physical appearance, relations within the extended family, and life in general increased. Although medication led to (temporary) positive changes, many social and emotional consequences remained unchanged. Interventions to treat drooling should not only be evaluated using measurements of drooling, but the consequences on social interaction and self-esteem should also be assessed.

Introduction

Several studies have reported the multiple adverse social and psychological effects, including poor self-esteem, of handicaps experienced by older children, adolescents, and young adults with cerebral palsy (CP; Cruickshank, 1952; Minde, 1978). Mainstreamed children of normal intelligence with CP regard themselves as being different, starting as early as four years of age (Teplin, Howard, & O'Connor, 1981). However, these self-views and their potentially negative effects on self-esteem do not seem to have crystallized in the cognitive and social mindset of the children until after they have started primary school.

Drooling is one of the disabling conditions. It is reported to be a significant problem in 10-37.5% of patients with CP (Ekedahl & Hallen, 1973; Van de Heyning, Marquet, & Creten, 1980). Parents of 1437 children with CP reported that 34% sometimes found drooling to be a problem, while 16% responded that it was often a problem (Bachrach, Walter, & Trzcinski, 1998). This percentage was even higher among CP children attending special schools: 58% (93 of 160) children, aged 4-18 years, drooled; 33% (53/160), drooled severely (Tahmassebi & Curzon, 2003).

Drooling radically influences daily care given to the child, for example, frequent instruction to swallow. Parents develop the habit of wiping the child's mouth and chin repeatedly. To protect their clothes, children wear bibs, shawls or terry cloth wristbands that have to be continually changed, not to mention damp clothes that are often changed at least once a day, resulting in extra loads of laundry per week. The drooling child

damages clothes, toys, books, and furniture, not to mention communication aids, computers, and audio equipment.

The psychological consequences of drooling should not be underestimated as drooling negatively influences the physical appearance of a child. The unsightly nature of drooling and the salivary spray occurring when the child talks, sneezes or coughs can result in some degree of alienation (Finkelstein & Crysdale, 1992). As a consequence, social interaction with peers or adults even including family members, is at risk. The mental capacity of the drooling child is frequently assumed to be lower than it is. In short, the development of self-esteem, largely determined by reactions from the child's social environment, is negatively affected.

The impact of drooling on social interaction and self-esteem justifies medical, surgical, and behavioural interventions to reduce drooling. However, to date no systematic investigation to assess the effect of such treatment on the child's development of self-esteem has been conducted.

In a recent controlled clinical trial, the anticholinergic treatment whether with Botulinum Toxin type A (BTX-A) or scopolamine was shown to significantly reduce salivary flow from the submandibular glands (Jongerius, Rotteveel, et al., 2004). Salivary flow reduction diminishes drooling in a standardized situation, as measured by the drooling quotient (Rapp, 1980). Visual analogue scales to register parental opinion about the severity of drooling at home have also shown diminished drooling (Jongerius, Van den Hoogen, et al., 2004).

The objective of this study was to evaluate the change that the above-mentioned interventions have produced in the child's social interaction and self-esteem.

Method

Subjects

A total of 45 children were included in a controlled clinical trial to compare the effect on drooling between intraglandular BTX-A injections into the submandibular glands and treatment with transdermal applications of scopolamine. Inclusion criteria were: pre-school and school age children with a diagnosis of CP, who had a score of 3 or higher on the Teacher Drool Scale (Camp-Bruno, Winsberg, Green-Parsons, & Abrams, 1989). This scale is a 5-point level to measure the degree of drooling: (1) indicating 'No drooling', (3) 'occasional drooling, on and off all day', (5) 'constant drooling, always wet'. Written informed consent and ethical approval for this study were obtained.

Data collection

In this study, a list of questions was developed to evaluate the impact of drooling on social interaction and self-esteem, partly based on the section self-esteem of the Child Health Questionnaire (CHQ-PF50; Landgraf, Abetz, & Ware, 1996). The research team,

consisting of a speech therapist, a clinical paediatric psychologist, and a paediatric rehabilitation physician, agreed that the items of the questionnaire covered relevant aspects of social interaction and self-esteem for drooling children. The parents of three children participating in a pilot study (Jongerius et al., 2001) commented on the preliminary version of the questionnaire, resulting in minor adjustments. The questionnaire, containing multiple-choice questions and Visual Analogue Scales (VAS), evaluated parental experiences as well as personal reactions of the child. Assessments were scheduled at baseline, during scopolamine treatment, and 8 and 24 weeks following BTX-A injections.

Data handling

To describe the data, the VAS scores were divided into three equal parts: 0-32, 33-66, and 67-100 mm. Missing values occurred because 23 of the 156 lists were not returned, furthermore questions were left unanswered or had non-interpretable answers. Before data analysis, list-wise missing values were imputed by using the method of 'carrying the last observation forward'. This meant that if one or two of the scores for an item after scopolamine or BTX-A treatment were missing, the last available score on that question was filled in (carried forward). If both assessments on a specific question at 8 and 24 weeks following BTX-A injections were missing, these subjects were not included in the statistical analysis for that question. Missing baseline values were estimated by imputing the mean (numerical score) or median (ordinal score) of the positively assessed participants.

Data analysis

Items concerning social interaction were analyzed using non-parametric tests. To evaluate the change in the parent's impression concerning the emotional development of their child, we used analysis of variance techniques for repeated measures, with alpha set at 0.05.

To assess possible bias by exclusion or drop out, we compared the distribution of the entire sample with the distribution of these cases on the factors 'developmental level' and 'treatment success'. Developmental level was scored in five categories: (1) developmental age (DA) below 4 years; (2) DA between 4 and 6 years and IQ $\leq$ 70; (3) DA between 4 and 6 years and IQ > 70; (4) DA above 6 years and IQ $\leq$ 70; (5) DA above 6 years and IQ > 70. Treatment success was defined as follows: a responder to therapy had a minimal flow rate reduction of 30% in combination with a reduction of the drooling quotient of at least 50%.

Statistical analyses for emotional reactions reported by the children themselves could not be performed because of the small number of children with meaningful answers.

Results

A total of 45 children (28 male and 17 female) were included: median age 9.1 years (range 3 to 16 years). Of these children, 22 could not talk. Eight children walked without an aid; 37 were wheelchair-bound. None of the children attended mainstream schools; 29 attended a special education school, while 14 attended a daycare center for children with developmental disabilities. The DA of 23 children was below 4 years as ascertained by psychological investigation. Six children did not complete the trail: for four children the scopolamine treatment was terminated because of adverse effects before the prescribed period ended, one changed anti-epileptic medication (a reason for exclusion from the study), and one did not attend the required follow-up sessions because of illness not related to the trial. The questionnaires of 39 children could be analyzed. As a result, the response rate was 85% (133 out of the initial 156 lists were returned).

Baseline investigations

Social interaction. Drooling appeared to have a remarkably negative influence on the social interaction (Table 4.1). The child's play with peers and his/her social interaction with both children and adults were limited. A striking finding was that, for six of these drooling children even their own parents kept their distance. In addition, parents of seven children reported that the unsightly appearance of their child caused outsiders to underestimate the child's mental ability.

Table 4.1 Social consequences of drooling at baseline (n = 39)

	Handicap related (*n*)	**Drooling related (*n*)**
Social interaction with children		
Does not play with other children at school	10	0
Does not play with other children at home	12	3
Is avoided by other children	16	13
Social interaction with adults		
Is avoided by familiar or unfamiliar adults	14	12
Adults keep their distance	16	16
Parents keep their distance		6
Underestimation of mental ability	21	7

Emotional development. Table 4.2 presents the parental impression of the child's self-esteem along with the overt emotional reactions of the child. The parents of seven children reported their child to be dissatisfied (VAS score 0-32) in one or more domains.

Three children were not happy about their social contact with peers, with one indicating drooling to be the main reason (VAS score 67-100). None of the children were dissatisfied with their physical appearance. Teplin, Howard, and O'Connor (1981) stated that between 4 and 8 years of age, children with CP become increasingly aware of their handicap but maintain positive self-esteem until early elementary school, when some become more distressed as they compare themselves with their non-handicapped peers. Out of the 39 children who completed our study, 20 were unable to express their feelings due to low DA (i.e., < 4 years). Consequently, only 19 children could be evaluated on the occurrence of overt emotional reactions. According to the parents, only a few of the children manifested emotional reactions pertaining to their physical appearance, competence or social acceptance by adults and peers.

Table 4.2 Emotional consequences of drooling at baseline (*n* = 39)

Parental evaluation (*n* = 39)		
	Not satisfied (*n*)	Not satisfied because of drooling (*n*)
School abilities	2	0
Participation in sports and play	2	0
Social contact with other children	3	1
Physical appearance	0	-
Relations within the extended family	1	0
Total quality of life	3	0

Emotional reactions of the child (*n* = 19)			
	Positive (*n*)	Negative or mixed (*n*)	Negative because of drooling (*n*)
Physical appearance	3	3	3
Competence	10	3	2
Social acceptance by adults	7	1	0
Peer acceptance	3	3	2

Changes across time

Social interaction. No significant change across time was found in social interaction as a result of treatment, with the exception of 'is avoided by other children' (chi-squared = 8.33; *df* = 3; *p* = 0.043). The valid percentage of this item showed a significant decrease, indicating that gradually participants were less frequently avoided by other children after treatment: 35,1% at baseline; 33,3% at scopolamine; 25,0% at 8 weeks after BTX-A, and 21,4% after 24 weeks.

Emotional development. Parental report on the child's satisfaction with school abilities, sports and play participation, social contact with peers, physical appearance, relations within the extended family, and life in general did not change significantly as a result of treatment. Nevertheless, parents reported a significant increase of *perceived impact of drooling* on the level of child's satisfaction on physical appearance (Hotelling's trace [HT^2] = 3,160; *df* = 3,23; *p* = 0.044), relations within the extended family (HT^2 = 3,363; *df* = 3,24; *p* = 0.035), and life in general (HT^2 = 3,243; *df* = 3,25; *p* = 0.039). To control for the possibility that these significant results are caused by selective analysis of children (whether those with low developmental level or those who did not respond to the treatment), we compared the factors 'developmental level' and 'treatment success' for those subjects excluded from statistical analysis on these questions (because of missing post treatment data) to those of the entire sample. They were equally distributed and statistical analysis did not indicate a significant difference (Chi-square test for independence [StatXact]: chi-squared = 9.767, asymptotic *p* = 0.6364; exact *p* = 0.6757). As mentioned, we cannot report the overt reactions of the participating children to the emotional factors investigated in this study. The parents of only seven of these children reported that their child expressed negative feelings about drooling in relation to one or more emotional factors during this study. There was no change for this outcome variable before and after treatment.

Discussion

Significant changes in social interaction and self-esteem were found after treatment of drooling in a sample of 39 children with CP. Following diminished drooling, social contacts with peers increased. In addition, parents indicated that the perceived impact of drooling on the level of child's satisfaction concerning physical appearance, relations within the extended family, and life in general increased as a result of anticholinergic medication or intraglandular injections of BTX-A.

Scopolamine as well as intraglandular BTX-A injections are effective interventions to reduce salivary flow rate and the spilling of saliva from the mouth, but do not totally eliminate drooling in all instances (Jongerius, Van den Hoogen, et al., 2004). If drooling is not totally inhibited, many social and emotional consequences may remain unchanged. This would explain why the investigated factors did not change.

Only a few parents reported an overt emotional reaction by the child concerning drooling. Several reasons can account for this. Many children in our sample could not express their feelings because of limited developmental age or other cognitive and/or communication disabilities. In addition, to improve the reliability of the responses, we asked parents at each assessment to consider only 'the past 4 weeks'. This may have resulted in an underestimation of the child's reaction during the entire 24 weeks post

BTX-A intervention period. Finally, the children in this study attend special schools or daycare centers for children with developmental disabilities. These social environments are likely to be tolerant of drooling as it is seen as one of the inevitable consequences of the handicap. The staff are positively engaged with these children, trying to create a stimulating social interaction, and to emphasize the relative strengths of each child to foster the child's self-esteem. This may also explain why drooling children express positive feelings about their physical appearance, their competence, and their social acceptance by adults and peers and why they express few negative emotions.

The increased perception of the importance of drooling on some aspects of emotional development may be explained by the specific effect of the mode of treatment. While both treatments effectively reduce drooling, the treatment effect gradually decreased during the 24 weeks following BTX-A injection, leading to a gradual increase in drooling. Before treatment, most children had a history of severe drooling for many years, during which the parents and the children resigned themselves to the situation. After the successful treatment and a period of reduced drooling, they might be disappointed because of the recurrence of drooling. As a result the importance of the problem could have been re-evaluated as being less acceptable than before the treatment.

It is recommended that any intervention to treat drooling such as surgery, medication or behavioural therapy, be evaluated not only with primary outcome variables such as the salivary flow but also for the effect on social interaction and self-esteem. While standardized and validated instruments for social interaction and self-esteem of drooling CP children with a wide range of motor, mental, and communication disorders are not available, our questionnaire can be used to obtain an impression. Although our results do not substantiate an improvement for all social and emotional aspects assessed, the inhibition of drooling remains important for practical reasons. If drooling can be reduced for longer periods or even permanently, positive social interaction over the years could lead to increased positive self-esteem and acceptance by peers and adults, both within the family as well as in a wider social sphere.

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Chapter 5

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

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Abstract

A descriptive analysis was conducted on studies on the behavioural treatment of drooling (published between 1970 and 2005). The 17 articles, which met the inclusion criteria, described 53 participants (mean age 14 years 7 months, [*SD* 4 years 9 months]; range 6-28 years). 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.

Introduction

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 (Blasco, 2002; Blasco & Allaire, 1992; Brei, 2003; Crysdale, Raveh, McCann, Roske, & Kotler, 2001; Hussein, Kershaw, Tahmassebi, & Fayle, 1998; Lloyd Faulconbridge, Tranter, Moffat, & Green, 2001; Meningaud, Pitak-Arnnop, Chikhani, & Bertrand, 2006; Nunn, 2000). 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 (2002) states: "In some cases, usually those more mildly involved, behavioural techniques (…) are easy to institute and will work" (p. 780). Brei (2003) 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 generalisation 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 (Blasco & Allaire, 1992).

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', 'behaviour 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); (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 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 (Drabman, Cordua y Cruz, Ross, & Lynd, 1979; Dunn, Cunningham, & Backman, 1987; Koheil, Sochaniwskyj, Bablich, Kenny, & Milner, 1987; Lancioni, Coninx, Manders, & Driessen, 1989) that were randomly chosen from the total 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 (Barton, Leigh, & Myrvang, 1978; Barton & Madsen, 1980; Connis & Rusch, 1980; Drabman et al., 1979; Dunn et al., 1987; Garber, 1971; Jones, 1982; Koheil et al., 1987; Lancioni, Brouwer, & Coninx, 1992, 1994; Lancioni et al., 1989; Poling, Miller, Nelson, & Ryan, 1978; Rapp, 1980; Rapp & Bowers, 1979; Richman & Kozlowski, 1977; Thorbecke & Jackson, 1982; Trott & Maechtlen, 1986). 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 twelve (one study). Most articles described one experiment only. The selection included two series of articles in which (some of the) participants were treated repeatedly (Jones, 1982; Lancioni et al., 1992, 1994; Rapp, 1980; Rapp & Bowers, 1979). That is, the follow-up for the experiment by Rapp and Bowers (1979) was reported in the study by Rapp (1980), in which the procedure was also repeated with a second intervention group. Jones (1982) conducted another study at the same school. The most recent study by Lancioni et al. (1994) is a follow-up of their 1992 study, reporting data for the same participants. The study by Drabman et al. (1979) described two experiments. In the present review each experiment was analysed 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 5.1; 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* 4 years 9 months) with age ranging from 6 to 28 years: 12 participants were between 6 and 12 years, 35

Table 5.1 Demographic characteristics, setting, and time investment for intervention

Study	**Sample size, *n***	**Sex M/F**	**Age (years)**	**Degree of learning disability**	**Motor impairment**
Barton et al. (1978)	2	2M	24, 28	Moderate (IQ 45); Severe (IQ 30)	CP
Barton & Madsen (1980)	1	M	11	Severe	-
Connis & Rusch (1980)	1	M	22	Moderate	-
Drabman et al. (1979) experiment 1	3	2M,1F	7, 12, 15	Profoundly	1 CP, 2 no MI
Drabman et al. (1979) experiment 2	2	1M,1F	14, 15	Mild (IQ 63); Severe (IQ 39)	no MI
Dunn et al. (1987)	1	M	16	No LD	CP
Garber (1971)	1	M	14	-	CP
Jones (1982)	8[a]	-	12-16	-	CP
Koheil et al. (1987)	12	9M,3F	6-18	No severe LD	CP
Lancioni et al. (1992, 1994)	2	1M,1F	27, 20	Moderate (IQ 40); Severe (IQ 35)	1 no MI, 1 CP
Lancioni et al. (1989)	2	2M	16, 11	Mild (IQ < 70); No LD (IQ > 70)	1 CP, 1 no MI
Poling et al. (1978)	1	M	17	Moderate	-
Rapp (1980)	8[b]	3M,5F	13-15	Profound & Severe (MA: 1½-6 years)	CP
Rapp & Bowers (1979)	7	3M,4F	15-16	Severe (MA: 4-8 years)	CP
Richman & Kozlowski (1977)	1	F	9	Severe	CP
Thorbecke & Jackson (1982)	1	F	19	Moderate (IQ 47)	CP
Trott & Maechtlen (1986)	1	F	11	Severe	no MI

Note: MA, mental age; LD, learning disability; CP, cerebral palsy; MI, motor impairment; -, not stated.
[a]: One patient was treated previously in study by Rapp (1980)
[b]: Rapp (1980) also reports data on the follow-up of the Rapp & Bowers (1979) group and on a comparison group

participants were between 13 and 18 years, and 6 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

Table 5.1 Continued

Study	**Setting**	**Instruction frequency (minutes); duration treatment**
Barton et al. (1978)	LD residential setting	- (10-15); >500&900s
Barton & Madsen (1980)	Special education class	5/wk; 24s
Connis & Rusch (1980)	Vocational training in a food service programme	5/wk; 36d
Drabman et al. (1979) experiment 1	Nonresidential LD centre	5/wk; 12-15wk
Drabman et al. (1979) experiment 2	Nonresidential LD centre	5/wk; 19&15d
Dunn et al. (1987)	Special class in a regular public school	1d/wk; 10wk
Garber (1971)	Speech and hearing clinic	3s/wk (25); 6wk
Jones (1982)	Residential school for children with SLD CP	5/wk; 3wk
Koheil et al. (1987)	Children's hospital & school/regular environment	-
Lancioni et al. (1992, 1994)	LD residential setting, activity centre and farm	1-2s/d (30); 35s
Lancioni et al. (1989)	Residential setting for the deaf	-;32&26s
Poling et al. (1978)	School for LD	5d/wk; 25d
Rapp (1980)	Residential school for children with SLD CP	5/wk (1s); 1wk
Rapp & Bowers (1979)	residential school for children with SLD CP	5/wk (1s); 1wk
Richman & Kozlowski (1977)	LD residential setting	4s/wk (30); 20wk
Thorbecke & Jackson (1982)	Special developmental school and at home (generalization)	5/wk; 25d
Trott & Maechtlen (1986)	Self-contained special class in a public elementary school	2s/d(15,25-30); 3wk

Note: LD, learning disability; CP, cerebral palsy; SLD, severe learning disabilities; s, sessions; d, days; /d, per day; wk, weeks; /wk, per week; -, not stated.

the short and global descriptions of participants. Six participants had no motor impairment. Previous medical treatment was seldom mentioned. Dunn et al. (1987) report that their subject 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 (Garber, 1971; Jones, 1982; Koheil et al., 1987; Rapp & Bowers, 1979) no interobserver reliability was reported. In the other studies, interobserver reliability appeared to be consistently high: above 70%.

Design. Rapp (1980) used an experimental comparison group design without randomization and presented data for a comparison group for the patient group in both her first (Rapp & Bowers, 1979) and second (Rapp, 1980) 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 (Connis & Rusch, 1980; Drabman et al., 1979; Garber, 1971; Jones, 1982; Trott & Maechtlen, 1986), six used a reversal design (Barton et al., 1978; Dunn et al., 1987; Koheil et al., 1987; Lancioni et al., 1989; Poling et al., 1978; Thorbecke & Jackson, 1982), in which each participant served as his/her own control. Five studies used a multiple baseline design, either across behaviours (one: Richman & Kozlowski, 1977), across activities (one: Dunn et al., 1987), across settings (one: Barton & Madsen, 1980), or across participants (two: Lancioni et al, 1992, 1994; Lancioni et al., 1989). 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 (1971) started intervention after only two baseline sessions in which a decreasing trend of drooling was already apparent, and in two other studies (Connis & Rusch, 1980; Richman & Kozlowski, 1977) 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. (1994) reported the most extensive follow-up data: at a 1- to 2-monthly period during the first 18 months after the intervention (Lancioni et al., 1992).

Target behaviour and type of treatment. Target behaviours for treatment were swallowing (Barton et al., 1978; Drabman et al., 1979; Garber, 1971; Jones, 1982; Koheil et al., 1987; Rapp, 1980; Rapp & Bowers, 1979; Thorbecke & Jackson, 1982), wiping (Barton et al., 1978; Barton & Madsen, 1980; Lancioni et al., 1992, 1994; Lancioni et al., 1989; Rapp & Bowers, 1979), nondrooling (Barton & Madsen, 1980; Drabman et al., 1979; Garber, 1971; Poling et al., 1978; Rapp & Bowers, 1979; Trott & Maechtlen, 1986), mouth closure (Connis & Rusch, 1980), head control and imitative vocalization (Richman & Kozlowski, 1977), and self-management (Dunn et al., 1987; Thorbecke & Jackson, 1982). 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 (involving 40 participants: Barton et al., 1978; Jones, 1982; Koheil et al., 1987; Lancioni et al., 1992, 1994; Lancioni et al., 1989; Rapp, 1980; Rapp & Bowers, 1979), cueing devices were used. Initially, participants received instructions to swallow or wipe following the cue. In some procedures (Barton et al., 1978) cues were gradually faded, while in others (Lancioni et al., 1992, 1994) 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 (Connis & Rusch, 1980; Drabman et al., 1979; Poling et al., 1978; Thorbecke & Jackson, 1982; Trott & Maechtlen, 1986), while in three of these experiments wiping was used as overcorrection and restitution procedure (Drabman et al., 1979; Thorbecke & Jackson, 1982; Trott & Maechtlen, 1986). Only two studies focused on the development of self-management skills to reduce drooling. Thorbecke and Jackson (1982) taught the participant in their second treatment phase to self-evaluate her chin (wet or dry) and to instruct herself by internalising the self-statements used in the overcorrection procedure. Dunn et al. (1987) 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 5.1), it appears that some interventions using cueing devices (Jones, 1982; Rapp, 1980; Rapp & Bowers, 1979) are less time-consuming than other procedures. Although the studies by Barton et al. (1978) and Richman & Kozlowski (1977) are extensive (20 or more weeks), the remaining intervention procedures take between three and 15 weeks.

Effectiveness. All authors, except Jones (1982) claim positive results and many of them anecdotally describe the related positive changes in quality of life of the participant(s). In 13 experiments (19 participants: Barton et al., 1978; Barton & Madsen, 1980; Connis & Rusch, 1980; Drabman et al., 1979; Dunn et al., 1987; Garber, 1971;

Lancioni et al., 1992, 1994; Lancioni et al., 1989; Poling et al., 1978; Richman & Kozlowski, 1977; Thorbecke & Jackson, 1982; Trott & Maechtlen, 1986) 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 (Drabman et al., 1979; Dunn et al., 1987; Lancioni et al., 1989). From the studies reporting numerical data (35 participants: Jones, 1982; Koheil et al., 1987; Rapp, 1980; Rapp & Bowers, 1979), five participants had drooling rate zero during intervention and 28 participants showed a reduction in drooling rate of more than 50% compared with baseline.

Generalisation and follow-up. Most authors both trained participants and evaluated effectiveness of their intervention in daily activities. Barton et al. (1978), Barton and Madsen (1980), and Dunn et al. (1987) assessed generalisation to respectively 'work', 'class', and 'school'. In four experiments (Garber, 1971; Jones, 1982; Koheil et al., 1987; Richman & Kozlowski, 1977) training and/or assessment were conducted in a specific room or during a few activities without assessing the generalisation of treatment effect.

Most studies report maintenance of treatment effect at follow-up. Four studies do not report follow-up data. Barton et al. (1978) 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 (Drabman et al., 1979; Dunn et al., 1987) drooling returned to baseline level at follow-up. Dunn et al. (1987) 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 last 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,

Richardson, Rosenberg, and Haynes (1997). Up to the present day, no randomized controlled trails (level I) have been performed. Using an experimental-comparison group design, the level of evidence for the studies of Rapp and Bowers (1979) and Rapp (1980) 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 and 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 type A (BTX-A; e.g. Meningaud et al., 2006). 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, 2003). 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. (1987) shows, behavioural therapy can also be effective following surgery. Greensmith et al. (2005) 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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Chapter 6

Behavioural treatment of drooling: A methodological critique of the literature with clinical guidelines and suggestions for future research

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Abstract

Many children with developmental and learning disabilities suffer from the consequences of chronic drooling. Behavioural treatment for drooling should be considered before other, more intrusive treatments like medication and surgery are implemented. However, empirical studies on behavioural procedures are scarce. This article reviews 19 behavioural studies published since 1970. Treatment procedures are (a) instruction, prompting, and positive reinforcement; (b) negative social reinforcement and decelarative procedures; (c) cueing techniques; and (d) self-management procedures. Although these procedures yield positive results, critical examination of experimental methodology of the studies reveals several methodological shortcomings. Guidelines for clinical use of behavioural treatment for drooling are presented, and recommendations are given for future research in this area.

Introduction

Drooling is the unintentional loss of saliva from the mouth. It is a normal phenomenon in infancy and usually subsides as oral motor function matures. Drooling while awake after four years of age is considered abnormal (Crysdale, 1989). In children with cerebral palsy (CP), persistent drooling is primarily due to oral motor dysfunction: inefficient (particularly in the oral phase) and/or infrequent swallowing and/or poor lip closure (Harris & Purdy, 1987; Hussein, Kershaw, Tahmassebi, & Fayle, 1998; Sochaniwskyj, Koheil, Bablich, Milner, & Kenny, 1986). In addition, Burgmayer and Jung (1983) state that saliva running down the lower lip or chin causes an aversive sensation in infants. In the normal development of swallowing, the young child learns to avoid this unpleasant sensation. If this aversive sensation does not occur in CP, swallowing as avoidance behaviour does not develop spontaneously. Drooling is reported in 10% to 37.5% of patients with CP (Ekedahl & Hallen, 1973; Van de Heyning, Marquet, & Creten, 1980). This percentage appears to be even higher among CP children attending special schools: 58% of 4- to 18-year-old children were reported to drool, with 33% (53/160) of the total school population drooling severely (Tahmassebi & Curzon, 2003). Drooling affects physical health and well-being negatively. Children suffer from chronically irritated, chapped or macerated skin over the chin and perioral region leading to cheilitis in 67% of the cases (Wilson & Henderson, 1999). Sometimes there is a chronic loss of fluids and nutrients, and in case of posterior drooling (Jongerius, Van Hulst, Van den Hoogen, & Rotteveel, 2005) children with inadequate swallowing frequently aspirate their own saliva, which may cause recurrent pneumonia. In addition, drooling has an adverse effect on social and emotional functioning (Van der Burg, Jongerius, Van Limbeek, Van Hulst, & Rotteveel, 2006).

Given the adverse consequences of drooling, treatment is warranted. Various treatment strategies have been reported, such as oral motor training, proper positioning, behavioural therapy, the use of intraoral devices, medication, and surgery (Blasco & Allaire, 1992). Although drooling is not caused by hypersalivation (Tahmassebi & Curzon, 2003), anticholinergic drugs are implemented to reduce salivary flow. Although this medication can be effective for drooling, adverse side-effects are frequently reported (Jongerius, Van Tiel, Van Limbeek, Gabreëls, & Rotteveel, 2003). Recently, several studies appeared in which the positive effects of injections of Botulinum Toxin type A into the parotid and/or submandibular salivary glands on salivary flow rate and drooling are reported (Banerjee, Glasson, & O'Flaherty, 2006; Jongerius et al., 2004). Surgical procedures include salivary glands excision, salivary duct rerouting, and a combination of these procedures (Crysdale, Raveh, McCann, Roske, & Kotler, 2001). The first procedure reduces salivary flow rate, whereas as a result of the second procedure the saliva enters the mouth at the back of the tongue, thus avoiding the oral phase of swallowing.

Behavioural procedures to treat drooling should be considered before other, more intrusive treatments like medication and surgery are implemented (Blasco, 2002; Brei, 2003; Hussein et al., 1998; Lloyd Faulconbridge, Tranter, Moffat, & Green, 2001; Meningaud, Pitak-Arnnop, Chikhani, & Bertrand, 2006; Nunn, 2000). However, empirical studies on behavioural procedures are scarce. Since 1970, 19 studies have been published in scientific journals. Across studies, various types of behavioural treatment procedures were evaluated on a total sample of 56 participants, who varied considerably in age and in learning and motor disabilities. Therefore, conclusions about the best type of behavioural therapy for drooling cannot yet be drawn.

In recent years there has been a paucity of behavioural studies on drooling as the focus has been on medication and surgical interventions. However, medication has only a temporary effect (Jongerius et al., 2004) and surgery does not always totally eliminate drooling (Greensmith et al., 2005). Given their potential effectiveness, behavioural procedures should be reconsidered. In the present study, we present a narrative review of behavioural studies published since 1970, focusing on type of treatment procedure and the experimental methodology instead of outcome. Our aim is to provide: (a) a description of treatment procedures, (b) a critical examination of the experimental methodology, (c) guidelines for the clinical use of behavioural treatment for drooling, and (d) recommendations for future research in this area.

Method

A database of articles on behavioural treatment of drooling was compiled from searches in PsychInfo, Web of Science, Medline, and PubMed, using the keywords *drooling* and *sialorrhoea* in combination with *behaviour therapy, behaviour treatment, behaviour*

training, and behaviour modification. To include studies from the USA, searches were also performed with the keyword *behavior*. This database was supplemented with articles retrieved from the references of selected articles and general reviews on drooling. Inclusion criteria were (a) detailed descriptions of behavioural procedures as treatment for drooling were presented and (b) participants were diagnosed with developmental disabilities (e.g., CP, learning disability).

Each article was assessed for a set of study variables including participants' characteristics such as gender, age, level of learning disability, motor impairment, former (medical) treatment for drooling, and drooling severity. Treatment procedures were analyzed, and target behaviour and treatment type were determined for each experiment. We also assessed whether a specific apparatus was used. In addition, information about the setting in which the behavioural treatment was conducted was recorded. Methodological aspects were also investigated, such as the definition of the dependent variables and how they were measured. In addition, data on interobserver agreement were registered. Finally, the type of experimental design was assessed.

To assess interrater reliability, a second rater independently reviewed four of the experiments that were randomly chosen from the total sample (including 18 [32%] of the participants). If both reviewers recorded the same descriptive category for the study variables, they were considered to be in agreement. Reliability was calculated for each study variable by dividing number of agreements by number of agreements plus disagreements and multiplying by 100%. Mean interobserver agreement score for study variables was 84.75% (range 72% to 100%).

Individual studies were analyzed with regard to treatment effectiveness by calculating mean baseline reduction (MBLR; Campbell, 2004). This effect size is based on a comparison of baseline data with data from the treatment phase that immediately follows it (i.e., AB design). For each participant, MBLR was calculated by subtracting the mean of treatment observations from the mean of baseline observations, then dividing by the mean of baseline observations and multiplying by 100. If interventions consisted of consecutive phases, the mean MBLR score per phase was calculated. In the case of a reversal design (i.e., ABAB) with reversal conditions similar to baseline conditions, MBLR was calculated independently for both the first baseline-first treatment and the second baseline-second treatment data. In the latter case, both MBLR scores were summed and divided by two. In the case of a multiple baseline design, MBLR was calculated for each behaviour, setting, or participant. These MBLR scores were summed and divided by the number of behaviours, settings, or participants.

Description of treatment procedures

The goal of any behavioural treatment procedure is to establish, increase, or decrease specific behaviour by systematically manipulating its antecedent stimuli (events preceding the target behaviour) and/or consequent (events following the target behaviour) stimuli. In the case of drooling, behavioural treatment aims at increasing target behaviours such as swallowing, wiping, head control, mouth closure, nondrooling, and performing self-control. Studies describing behavioural interventions for drooling are summarized in Table 6.1, with MBLR scores to document effectiveness for each procedure. In this section we provide a description of four types of treatment procedures: (a) instruction, prompting, and positive reinforcement; (b) negative social reinforcement and decelarative procedures; (c) cueing techniques; and (d) self-management procedures.

Instruction, prompting, and positive social reinforcement

Instruction and prompting are both antecedent techniques-that is, preceding the target behaviour. For example, the child is instructed to wipe the face whenever the chin is wet or is prompted to swallow if the trainer spots saliva on the child's lip or before beginning to talk.

Positive social reinforcement is an example of a consequent technique. Following the performance of target behaviour, a reinforcing stimulus such as a positive remark is given, occasionally paired with a token or an edible. In several studies, positive reinforcement for nondrooling behaviour was given. This behavioural technique is called Differential Reinforcement of Other (nonproblem) behaviour (DRO).

In most studies reported, combinations of these behavioural techniques were often implemented. For instance, Garber (1971) successfully reduced drooling during articulatory therapy in a 14-year-old boy by prompting him to swallow before speaking. In addition, he was verbally praised for swallowing and rewarded with pennies for being dry during increasing time intervals. In a related study, Barton and Madsen (1980) prompted an 11-year-old boy who has severe learning disability to wipe his face contingent on drooling and praised him for both wiping and nondrooling for which he also received drinks. DRO was also implemented by Richman and Kozlowski (1977). After positive reinforcement of head control and imitative vocalization in a 9-year-old girl who has severe learning disability, they found an improvement in these target behaviours in addition to a reduction in drooling.

Negative social reinforcement and decelarative procedures

Negative social reinforcement and decelarative procedures are both consequent procedures. Contingent on the occurrence of drooling, an aversive stimulus such as a

verbal warning is given, and sometimes a more intrusive decelarative procedure is added. Connis and Rusch (1980) report a significant decrease in drooling in a 22-year-old kitchen aid who has moderate learning disability as a result of a treatment package consisting of reprimands for drooling, instructions to keep his mouth closed and praise for being dry, which were all gradually faded. Poling, Miller, Nelson, and Ryan (1978) also report a significant decrease in drooling in a 17-year-old boy with moderate learning disability, during treatment, which consisted of negative comments on drooling behaviour and verbal praise and tokens for nondrooling. Burgmayer and Jung (1983) used a brief period of withdrawal of attention (time-out) by the trainer contingent on drooling.

In some studies, mild to moderate punishment is given contingent on drooling. Kay, Harchik, and Luiselli (2006) trained a 17-year-old student with autism who attended a public high school. After preteaching sessions in which he was taught to respond to the instructions 'swallow' and 'wipe your mouth', regular checks (increasing from 5 to 15 minutes) were made by the aid in the classroom, community vocational site, and cooking class throughout the school day. If his face was dry, he was praised and an edible treat was presented to him. If his face was wet, he was mildly punished by requiring him to wipe his mouth and swallow three times successively.

Trott and Maechtlen (1986) also implemented a decelarative procedure through overcorrection involving restitution and positive practice. In this type of procedure, the participant was forced to restore the undesired state extensively and to perform desired behaviour in a somewhat excessive frequency. An 11-year-old girl who has severe learning disability was physically guided to clean any saliva that was on her clothing, the furniture, or materials with a paper towel (i.e., restitution). Positive practice consisted of helping the child press a tissue lightly under her lips for 30 seconds. Two daily sessions of 15 to 30 minutes were scheduled during the day. During the remainder of the day she was frequently complimented on her appearance and praised when her face was dry.

Drabman, Cordua y Cruz, Ross, and Lynd (1979) described two studies in which the most intrusive overcorrection procedure was used. In the first study (with 3 participants who had profound learning disabilities, aged 7, 12, and 15 years) the teacher checked if the child's chin was wet at specified time intervals, which were decreased from 30 to 7.5 minutes. If the chin was wet, a verbal reprimand was given, and the child was prompted to wipe his or her chin 50 times. In the second study, both children (aged 14 and 15; IQ 39 and 63, respectively) were asked to wipe their mouths and swallow whenever they were drooling. They were abundantly praised whenever the teacher noticed the participants' lips and chin were dry and also at 'dry' checks (every 7.5 minutes) by the teacher. If their chin appeared wet when checked, they received a verbal reprimand and were prompted to wipe their chin 50 times.

Table 6.1 Type of treatment, apparatus, sample size, and effect size (MBLR)

Study	**TARGET BEHAVIOUR treatment type**
Garber (1971)	ND penny reward system; S prompt, verbal praise
Barton et al. (1978)[a] subject M	S verbal prompt GF, tokens GF; W auditory cue contingent on drooling GF
Barton et al. (1978)[a] subject K	S verbal prompt, noncontingent visual & auditory cue, tokens GF; W auditory cue contingent on drooling GF
Richman & Kozlowski (1977)	HCIV positive social Rf
Poling et al. (1978)	ND positive social Rf, tokens GF; D negative social Rf
Drabman et al. (1979) experiment 1	ND positive social Rf; D negative social Rf, overcorrection (wiping 50 times)
Drabman et al. (1979) experiment 2	ND positive social Rf; D negative social Rf, overcorrection (wiping 50 times); S & W instruction, contingent on drooling
Rapp & Bowers (1979)	ND positive social Rf; S noncontingent auditory cue, verbal praise; W verbal & visual instruction, noncontingent auditory cue, verbal praise
Rapp (1980)	S non-contingent auditory cue, positive social Rf
Barton & Madsen (1980)	ND positive social Rf, drinks; W prompting: contingent on drooling, praise GF
Connis & Rusch (1980)	D negative social Rf; MC instruction
Jones (1982)	S noncontingent auditory cue, positive social Rf
Thorbecke & Jackson (1982)	D negative social Rf, overcorrection (wiping 10 times); S instruction, positive social Rf; SM internalized self-instruction (negative Rf, overcorrection)
Burgmayer & Jung (1983)	ND positive social Rf; D negative social Rf (a brief period no attention); S instruction, fluid in the back of the throat, auditory cue, positive social Rf
Trott & Maechtlen (1986)	ND positive social Rf; D overcorrection, positive practice (wiping 30 seconds)
Koheil et al. (1987)	S instruction, auditory cueing, EMG biofeedback
Dunn et al. (1987)	SM prompts GF, verbal Rf, token system for self-control (mouth closed and swallow)
Lancioni et al. (1989)[a] subject 1	W vibratory cues (replaced by visual cues), positive social Rf
Lancioni et al. (1989)[a] subject 2	W vibratory cues, positive social Rf
Lancioni et al. (1992, 1994)	W (brief and/or flexible) auditory cues, positive social Rf
Kay et al. (2006)	ND positive social Rf, edible treat; D wiping and swallowing 3 times

Note: MBLR, mean baseline reduction; ND, nondrooling; D, drooling; S, swallowing; W, wiping; HCIV, head control and imitative vocalization; MC, mouth closed; GF, gradually fading; Rf, reinforcement; SM, self-management; EMG, electromyograph; CD, cueing device.
[a] Because type of treatment in Barton et al. (1978) and Lancioni et al. (1989) differs slightly across participants, treatment for both participants is described separately.

Table 6.1 Continued

Study	Apparatus	Sample Size (*n*)	MBLR
Garber (1971)		1	88
Barton et al. (1978)[a] subject M	CD	1	-27
Barton et al. (1978)[a] subject K	CD	1	20
Richman & Kozlowski (1977)		1	48
Poling et al. (1978)		1	83
Drabman et al. (1979) experiment 1		3	41
Drabman et al. (1979) experiment 2		2	85
Rapp & Bowers (1979)	CD	7	83 [b]
Rapp (1980)	CD	8	87 [b]
Barton & Madsen (1980)		1	85
Connis & Rusch (1980)		1	69
Jones (1982)	CD	8	-9
Thorbecke & Jackson (1982)		1	73
Burgmayer & Jung (1983)	CD	1	- [c]
Trott & Maechtlen (1986)		1	93
Koheil et al. (1987)	EMG, CD	12	73
Dunn et al. (1987)		1	99
Lancioni et al. (1989)[a] subject 1	CD	1	86
Lancioni et al. (1989)[a] subject 2	CD	1	100
Lancioni et al. (1992, 1994)	CD	2	85
Kay et al. (2006)		1	76

[b] Data (tables) by Rapp (1980) are not entirely consistent with both text and data presented in Rapp and Bowers (1979).
[c] Burgmayer and Jung (1983) do not present numerical data.

Cueing techniques

Several studies report on a specific prompting procedure by electronic cueing. An electronic cueing device emits visual, auditory, or tactile cues (i.e., antecedent stimuli) to increase the frequency of swallowing or wiping the mouth and chin. Cueing frequency varies across studies from once in every 15 seconds (Barton, Leigh, & Myrvang, 1978) to every 5 minutes (Burgmayer & Jung, 1983). In the first studies, the device was situated on a table, whereas in later studies, portable cueing devices were used.

In their extensive study of more than 500 sessions per participant, Barton et al. (1978) trained 2 men who had severe to moderate learning disabilities (24 and 28 years; IQ 45 and 30, respectively). Swallowing was verbally prompted and rewarded with tokens. The second participant also received a visual cue every 15 seconds to pace swallowing. To elicit a wiping response, an auditory cue from a buzzer on the table was presented contingent upon the occurrence of drooling. Both electronic cues were gradually faded out.

Rapp and Bowers (1979) described a portable cueing device (a so-called dribble box) that electronically prompted 7 children with CP (age 15 to 16 years, developmental age 4 to 8 years) to swallow after a 2- to 3-second auditory cue at fixed time intervals. After the stimulus-response chain was established during an undemanding task, the children wore the device attached to their clothes throughout the day. While Rapp (1980) used a small box pinned to the child's clothes, Burgmayer and Jung (1983) used a watch, which produced an auditory cue of about 30dB at 5 minutes intervals to elicit regular swallowing in an 8-year-old male with CP and severe learning disability.

Lancioni, Brouwer, and Coninx (1992) evaluated different cueing procedures with 'brief' versus 'flexible' auditory cues in 2 participants who had learning disabilities (aged 24 and 28 years). Brief cues with a fixed duration (2.5 seconds) were given at fixed intervals (2 minutes). Flexible cues lasted until the target response occurred, after which a new interval started. Lancioni et al. (1992) concluded that flexible cues are particularly effective in producing a consistent response, whereas brief cues may be sufficient to ensure a response from some subjects, but not from all.

Koheil, Sochaniwskyj, Bablich, Kenny, and Milner (1987) also reported an intervention using an auditory cueing procedure, but their 12 participants (aged 6 to 18 years; inclusion criterion-no severe learning disability) first received EMG feedback and visual feedback (mirror) in the research department to increase swallowing. The participants were taught to swallow in response to a cueing device (a so-called Accularm) that produced an auditory signal through earphones every 30 to 40 seconds, depending on the child's needs. In the next phase the cueing device was used in the child's natural environment. If the response to the cue was poor, the auditory cue was transmitted through speakers so that staff or other children could prompt the child to swallow.

Cueing techniques for drooling have also been adapted for people who are deaf. Lancioni, Coninx, Manders, and Driessen (1989) trained 2 participants who were deaf (age 11 and 16 years; the first with motor and learning disabilities, the second learning disabled without motor disabilities) through the use of tactile and visual cues that lasted until the wiping response was performed. If the response was performed spontaneously, the cue was avoided. The portable cueing device consisted of an electronic circuitry, which included a timer (placed at the participant's belt), a vibrator (at the waist or connected with the ear mold of the hearing aid) or a light (at the inside of the frame of the participants eye-glasses), and a response sensor (a wireless transmitter on the wrist that, whenever in close proximity, activated a tiny receiver at the shoulder). The wiping response was registered and resulted in the interruption of cueing and/or resetting of the timer to a new interval.

Self-management

To develop internal control of swallowing or wiping, as opposed to external cueing, self-management techniques aim at teaching the child 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. Data on self-management procedures for drooling are available for only two children. After implementation of an overcorrection procedure (e.g., if the trainer observed the chin was wet, the girl had to swallow once and wipe her chin 10 times), Thorbecke and Jackson (1982) taught self-monitoring and self-instruction to a 19-year-old female with CP and moderate learning disability. During self-management training, she herself had to check her chin and repeat the phrases containing both feedback and instruction. In subsequent training sessions, she was required to say these sentences first aloud, then in a whisper, and finally think to herself. Although the overcorrection procedure did not produce a lasting response when baseline conditions were reinstated, the reduction in drooling after implementation of self-monitoring and self-instruction did remain stable during and after the fading out of the trainers' prompts to carry out the self-instruction procedure.

In the last study reported, Dunn, Cunningham, and Backman (1987) used a procedure of self-control for swallowing with positive reinforcement to eliminate drooling in a 16-year-old boy with severe spastic quadriplegia. His mental abilities were fairly good-language comprehension and non-verbal cognitive abilities were estimated at approximately 13 years. The self-control procedure contained four steps: (a) monitor mouth closure, (b) determine the need to swallow, (c) evaluate the success of the procedure in terms of drooling prevention, and (d) verbally reward himself if successful. Every 60 seconds the boy was verbally prompted to start the self-control procedure and

was allowed to take a token each time he succeeded. After reduction in drooling, reinforcement was eliminated and prompts were gradually faded out.

Critical examination of experimental methodology

Before clinical guidelines for behavioural treatment can be given, we need to examine the methodology used in the studies reviewed. In this section, we will discuss important methodological issues pertaining to measurement of drooling, interobserver reliability, experimental design, generalization, and maintenance (Table 6.2).

Measurement of drooling

Across studies, conclusions on the effectiveness of behavioural procedures were based on different definitions of drooling and differing measurement techniques were used. The definition of drooling varies widely, ranging from 'saliva on lower lip', 'saliva under the lower lip leading to a wet chin', to 'a string of dribble falling from the mouth or chin'. In addition, techniques to measure drooling vary. In 15 studies, drooling was measured by direct observation, either by examining the participant's face or by checking the therapist's finger after wiping the participant's chin. In 7 studies, the observation period started after cleaning the mouth and chin, whereas in others the assessment started without first cleaning the face. In 4 studies the participants themselves were involved in evaluating the drooling severity. In 2 studies the amount of drooling was measured by the weight of a bib (e.g., Burgmayer & Jung, 1983) or the quantity of saliva collected in a chin cup (e.g., Koheil et al., 1987). As indicated in Table 6.2, there was a wide range of activities performed by participants during assessment. Some children perform regular activities in daily situations, whereas in other studies, they perform only a few standardized activities, not executed in their natural environment. These differences in measurement hinder comparison of results across studies.

Interobserver reliability

Interobserver reliability levels for dependent measures were satisfactory in most studies (Table 6.2). However, 5 studies (Burgmayer & Jung, 1983; Garber, 1971; Jones, 1982; Koheil et al., 1987; Rapp & Bowers, 1979) lacked reliability data, precluding valid conclusions on the intervention effects reported.

Experimental design

Evidence for effective treatment procedures comes from carefully designed studies. In applied behavioural research, most studies use single-subject designs. With the exception of Rapp (1980), who used an experimental control group design, behavioural studies on drooling predominantly used single-subject designs, such as the AB design (n = 7) and reversal-design (n = 6). Six studies used a multiple-baseline design, either across behaviours, across activities, across settings, or across participants. Although the

internal validity of studies using reversal or multiple-baseline designs is strong, the external validity of all studies with single-subject designs is at risk because of the small number of participants (mean number = 2.9; range 1 to 12). Thus, the evidence base is relatively small because individual characteristics of participants could account for the variance in treatment success. In addition, some methodological flaws were evident. For instance, Garber (1971) started intervention after only two baseline sessions, in which a trend for decreased drooling was already apparent. For two other studies (Connis & Rusch, 1980; Richman & Kozlowski, 1977), the drooling rate also showed a decreasing trend during baseline. These methodological flaws limit the validity of conclusions.

Generalization and maintenance

If behavioural intervention takes place in a therapeutic setting, generalization to daily life situations –where attention is diverted from drooling intervention per se- has to be evaluated and, if necessary, intentionally fostered. However, Garber (1971), Jones (1982), Koheil et al. (1987), and Richman and Kozlowski (1977) failed to do so: changes in drooling were evaluated exclusively in therapeutic sessions and/or sessions at the research department.

Maintenance of treatment effect is another important issue in the choice of the intervention type. Interventions with lasting results are needed. Thirteen studies presented follow-up data at one or more intervals for a maximum of 1 year after intervention. Lancioni, Brouwer, and Coninx (1994) reported the most extensive follow-up data: at a 1- to 2-monthly period during the first 18 months after the intervention study (Lancioni et al., 1992). Unfortunately, 5 studies failed to report any follow-up data (Barton et al., 1978; Garber, 1971; Kay et al., 2006; Lancioni et al., 1989; Poling et al., 1978). Although treatment was reported to be effective, there was no evidence to support maintenance of these intervention effects.

Guidelines for clinical use of behavioural treatment for drooling

After careful consideration of the four types of behavioural intervention for drooling and the methodological issues concerning the studies of these behavioural treatments, some general guidelines can be derived for implementation of such interventions in a clinical setting. Instructions, prompts, praise, and tangibles (tokens) for performing target behaviours such as nondrooling, swallowing, wiping, keeping the mouth closed, and maintaining upright head position are all commonly applied in daily interaction with children who suffer from drooling. Unfortunately, such approaches remain without a lasting effect because they are only intermittently and not systematically applied. Only systematic implementation of (a combination of) these techniques in a behavioural treatment program is effective. Although instructions, prompts, praise, and tangibles are

Table 6.2 Definition of drooling and additional dependent variables, activity during assessment, interobserver reliability, and design

Study	Definition of drooling and additional dependent variables	Activity during assessment
Garber (1971)	Saliva outside the mouth; wiping	Articulatory therapy
Barton et al. (1978)	Chin dry or wet; swallows; wipes; coughs	Training and real life situations
Richman & Kozlowski (1977)	Saliva on the lips or mouth: wet or dry [b]; head control; imitative vocalization	Language training
Poling et al. (1978)	Visually evident presence of excessive saliva	Classroom activities
Drabman et al. (1979) experiment 1	Chin and lower lip dry, wet or very wet [a,b,c]	Classroom and playground activities
Drabman et al. (1979) Experiment 2	Chin and lower lip dry, wet or very wet [a,b,c]	Classroom and playground activities
Rapp & Bowers (1979)	String of dribble	Informal group sessions
Rapp (1980)	String of dribble	Informal group sessions
Barton & Madsen (1980)	Drooling; remnants; wiping	Classroom activities
Connis & Rusch (1980)	A drool beneath lower lip line [b,c]	Vocational training in food service program
Jones (1982)	String of dribble	Doing as little as possible; tower building [d]
Thorbecke & Jackson (1982)	Chin under lower lip dry, wet or soaking[a,b]	Classroom and playground activities, group excursions
Burgmayer & Jung (1983)	Weight of bib; swallows	Training and everyday situations
Trott & Maechtlen (1986)	Saliva on lips > 3 sec. [b]	Real-life situations (not at mealtime)
Koheil et al. (1987)	Drooling rate ml/hr; swallow frequency (EMG)	Watching video
Dunn et al. (1987)	Number of drips falling from lip or chin	Playing PC games, card playing, conversation, reading
Lancioni et al. (1989)	Saliva outside the lower lip: wet or dry [b]	Classroom, vocational room, and resource room activities
Lancioni et al. (1992, 1994)	Saliva outside the lower lip: wet or dry; wiping	Farm-related jobs; occupational activities with peers
Kay et al. (2006)	Pools of saliva diameter > 1 inch	Classroom activities; community vocational site; cooking

Note: -, no data; PI, partial interval recording; MTS, momentary time sampling; BL, baseline; FU, follow-up; R, reversal design; MB, multiple baseline across; EC, experimental-control.
[a] check finger after wiping mouth and/or chin.
[b] dry start of assessment procedure.
[c] participant involved in assessment procedure.
[d] at baseline activity not described.

Table 6.2 Continued

Study	**Interobserver reliability % (mean %)**	**Design (FU in weeks)**
Garber (1971)	-	AB
Barton et al. (1978)	80-100	R
Richman & Kozlowski (1977)	80-95 (92)	MB behaviours (4 & 52)
Poling et al. (1978)	71	R
Drabman et al. (1979) experiment 1	66-100 (98-99)	AB (26)
Drabman et al. (1979) experiment 2	98-100 (99)	AB (26)
Rapp & Bowers (1979)	-	EC-group (1 & 39)
Rapp (1980)	99	EC-group (1 & 13)
Barton & Madsen (1980)	87-100	MB settings (20)
Connis & Rusch (1980)	100	AB (2)
Jones (1982)	-	AB (3&8)
Thorbecke & Jackson (1982)	91.96-100	R (8&26)
Burgmayer & Jung (1983)	-	AB (12&52)
Trott & Maechtlen (1986)	90-100	AB (10)
Koheil et al. (1987)	-	R[e] (4)
Dunn et al. (1987)	98	MB activities; R (13, 26 & 52)
Lancioni et al. (1989)	56-100 (93-99)	MB participants; R
Lancioni et al. (1992, 1994)	0-100 (89)	MB participants[f] (13-78)
Kay et al. (2006)	89-100 (91-96)	MB settings

[e] design replication across participants.
[f] during intervention and follow-up also assessments under baseline condition.

useful elements in behavioural interventions, withdrawal of these procedures usually leads to a return to baseline-level drooling (e.g., Poling et al., 1978).

Self-management for drooling without social or technical support is the ultimate goal of intervention and leads to maximum independence of the participant. Although Dunn et al. (1987) present evidence for the effectiveness of this procedure for only one subject with no learning disability, they claimed to have achieved a similar effect in a variety of children ranging in age from 8 to 13 years with developmental levels from 4 to 10 years. Contraindications for the use of self-management procedures seem to be a low-grade mental ability (severe and profound learning disability) and a developmental age below 4 years.

If a self-management procedure appears to be ineffective or is contra-indicated, intervention by cueing techniques should be considered. Forty-one participants in this review were trained with electronic cueing devices for swallowing or wiping, albeit with variable success. For instance, during the 3 studies at the Meldreth Manor School, the positive effects of auditory cueing seem to decrease over time. While Rapp and Bowers (1979) and Rapp (1980) reported most promising results, Jones (1982) reported no significant improvement although he used the same procedure to introduce and implement the dribble boxes. Jones further observed a slackening of interest in the use of dribble boxes and questions about the effectiveness of the cueing device among the staff of the institute. Some considered that only certain children benefited, whereas other pupils could not maintain the habit of swallowing or wiping on the cue. Thus, apparently only introducing a cueing device is not in and of itself sufficient to reduce drooling. Additional behavioural strategies such as instructions, prompts, praise, and tangibles seem to be necessary not only at the start of the therapy but also during the course of the treatment.

Once participants treated for drooling become accustomed to the cueing device, they often remain dependent on the device to remain dry. Lancioni et al. (1994) scheduled assessment sessions with and without the cueing device throughout their study. They concluded that their subjects remained dry with the cueing device but did not when the device was not used. They proposed that some participants may only achieve a device-assisted ('artificial') control of drooling. Without a positive attitude toward the program and substantial positive reinforcement from the social environment, these children failed to maintain the learned behaviour. Therefore, before employing a cueing device, one should assess in advance if continuous cueing is acceptable in the participant's social setting, whether at home, school, or work. Perhaps tactile cueing should not only be considered for deaf people but also in cases where auditory cueing could be socially unacceptable, just as one sometimes turns off the ring tone of the mobile phone and only uses the vibrating call alert. Because electronic devices are susceptible to breakage, an extra device needs to be at hand.

Cueing devices are not readily available and must be constructed by the clinicians themselves. The authors using these devices provided some advice. Hart and McCrady (1977) also described an inexpensive electronic device that could be easily built. None of the studies described how optimal cueing intervals were established. The frequency of cueing varied across studies from once every 5 minutes to once every 15 seconds. Optimal cueing frequency was adjusted to the child's needs. It seems reasonable to assume that the child's drooling severity is a contributing factor. Those with severe drooling should be cued more often than those with milder drooling. In addition, the minimal percentage of effective cues (e.g., performance of target behaviour after a cue) is another determining factor. Children who drool are often unable to perform dual tasks (e.g., swallowing or wiping while performing another activity). If they are allowed to miss some cues while concentrating on another task or activity, cueing frequency should be increased. The available literature on drooling does not contain a systematic evaluation of which factors justify a certain cueing frequency. For clinical use, a cueing frequency of every 15 seconds seems appropriate at introduction to establish the stimulus-response chain. If the child is successful in maintaining dry, the trainer can systematically increase the interval as long as the child continues to be dry.

Although overcorrection was also reported to be an effective treatment option for drooling, its aversive nature makes its use socially acceptable only if the nonaversive procedures described above have been shown to be ineffective. However, a careful evaluation of the undesired side effects resulting from this kind of procedure should be conducted.

Treatment should be focused on reducing drooling while the child performs a variety of his or her daily activities (e.g., Dunn et al., 1987). However, generalisation of learned behaviours to a variety of different daily situations and activities is often a problem in the learning disabled population and, therefore, has to be assessed and intentionally fostered. For some participants in the reviewed studies, drooling returned to baseline level after intervention. If long-term follow-up data are collected, the need for supplemental training ('booster sessions') to reinstate the initial treatment effect can be assessed. For instance, in the study by Dunn et al. (1987), the initial treatment effect remained stable at 3-month follow-up, but a booster training was necessary at 6 months to reinstate the effect, which was then maintained for another 6 months.

Recommendations for future research

The following recommendations concerning methodology for future empirical research can be made. The conceptualisation of what constitutes drooling requires specification. In several studies drooling was defined as 'a string of dribble' falling from the mouth. Although this is certainly the most severe level of drooling, the negative social impact of

drooling is just as apparent when saliva perpetually accumulates on the chin beneath the lower lip line. Hence, eliminating strings of dribble does not necessarily imply that the child's face is dry. Therefore, a consistent definition such as 'saliva on the chin, beneath the lower lip line', is strongly recommended to define the dependent variable when evaluating interventions to treat drooling.

The conditions during assessment must be carefully chosen and defined. Because children vary in their intellectual levels and daily activities, it is not possible to select one standard activity or situation to validly determine treatment effect. In addition, drooling severity is variable between activities and conditions both between as well as within participants. Jones (1982) stated that reliable (i.e., test-retest) data could be obtained from ten 10-minute sessions. However, the data validity can only be ascertained by choosing representative situations and activities comparable to the child's daily activities. To evaluate the effect of intervention strategies, drooling has to be assessed in both standardized situations during training as well as during regular daily life situations. In addition, unobtrusive measurement of drooling by direct observation may be impossible because the child is aware of being observed. To reduce the threat to validity, a standardized measure of drooling severity during the day based either on the parents' or the teachers' judgments, such as the Teacher Drool Scale (TDS: Camp-Bruno, Winsberg, Green-Parsons, & Abrams, 1989) or a Visual Analogue Scale (e.g., Jongerius et al., 2004) should supplement data on drooling from direct observation. The TDS is a five point scale, measuring (a) no drooling; (b) infrequent drooling, small amount; (c) occasional drooling, on and off all day; (d) frequent drooling, but not profuse; and (e) constant drooling, always wet. In addition, the use of the TDS allows for comparisons between children across studies. This may provide us with information on treatment effectiveness for specific subgroups in the drooling population.

Evidence-based practice demands scientific evaluation of treatment procedures with internally valid designs and interobserver reliability checks. Detailed description of participants for dependent variables and mental and motor abilities enables the clinician to determine whether the procedure would be successful in a specific case. In addition, if behavioural treatment procedures prove effective in studies with a single-subject design, the external validity is enhanced by replication of the same procedure in a larger group of drooling children.

Because behavioural interventions are usually time-consuming, the positive effects from reducing or eliminating drooling in everyday life can motivate both children and caregivers to adhere to the program. Therefore, systematic data collection on practical, social, and emotional consequences of (reduced) drooling is necessary (Van der Burg et al., 2006). Future research should also focus on minimizing instruction time by

choosing efficient behavioural procedures, adjusted to the learning potential of the child that drools.

Conclusion

Clearly, behavioural treatment has been shown to be effective to increase swallowing and wiping and to decrease drooling, but procedures need both elaboration and evaluation in future studies. Guidelines for best practice in behavioural treatment, differentiated for age groups and level of learning and motor disability, must be developed. In this way, behavioural therapy can contribute toward improving the quality of life of children who drool and that of their parents and others who are involved and can add to the multidisciplinary management practice of drooling.

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Chapter 7

Self-management treatment of drooling: A case series

Submitted for publication

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Abstract

Behavioural 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 to 60 minutes, 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.

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 behavioural 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 behavioural studies is small (i.e., $N < 60$) and participants vary considerably in age, and intellectual and motor disabilities. In addition, behavioural 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 ten 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 minutes and repeat the teacher's instructions aloud. In case of a wet chin, she had to swallow once and wipe her

chin ten 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 behavioural 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 seconds, 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 3 months 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.

Method

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 7.4), 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 behaviour, and, finally, (j) no medical treatment for drooling in the 6 months preceding participation in the present study. For demographic data of participants, see Table 7.1. Table 7.2 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.

Setting and materials

Participants resided at the children's ward of a rehabilitation clinic for about three 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 an (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.

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 seconds without the individual cleaning his/her face and/or clothes. The dependent variable was latency in minutes of being dry (nondrooling) 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.

Table 7.1 Demographic characteristics

Participant	Sex (M/F)	Age (years; months)	Diagnosis	Motor Impairment (GMFCS)	Oral motor Status	Degree of Intellectual Disability (ID)
K	F	11;10	CP, controlled epilepsy, coloboma right iris	IV	OD, S&TD	Mild IQ 64 at age 11;9
D	M	17;0	CP quadriplegia with spastic MI	V	S&TD, Mild M	Severe MA between 4-8y
A	M	7;10	CP quadriplegia with spastic & dyskinetic MI	III	OD, S&TD, TP, Mild M	No ID IQ 110 at age 6;0
S	F	9;10	CP bipyramidal syndrome with spastic MI, controlled epilepsy	IV	OD, S&TD	Mild IQ 50 at age 10;11
Mi	F	7;3	CP tetraplegia, epilepsy, speech delay	III	S&TD, Mild M	Mild IQ 60 at age 7;8
Mo	M	10;8	CP quadriplegia spastic MI	III	S&TD, Severe M	Mild IQ 75 at age 7;9
L	M	9;2	Psychomotor retardation	III	Mild M, hypotonic tongue and lips	Mild IQ 70 at age 6;5
Je	M	19;9	CP quadriplegia	II-III	S&TD, Severe M	Severe MA 4;8 at age 14;1
Jo	F	8;2	Worster Drought Syndrome	I	S&TD	No ID IQ 118 at age 5;5
R	F	7;0	CP quadriplegia with spastic and atactic MI	III	S&TD, TP, Mild M	No ID

GMFCS, Gross Motor Function Classification System; ID, Learning Disability; CP, Cerebral Palsy; MI, movement impairments; 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

Then, the participant was asked to clean his mouth and chin and the stopwatch was started. Five minutes after the trainer had observed that drooling occurred, video registration was terminated without any comment.

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 6 experimental phases: (a) baseline, (b) self-management treatment, (c) post-intervention, (e) generalization, and

Table 7.1 Continued

Participant	Communication mode	Setting	Previous treatment(s)	Drooling Severity pre-baseline	
				TDS	VAS
K	Non-verbal, voice computer	Special school for physical and multiple disabled children	OMT, BTX-A	-	-
D	Non-verbal	Special school for physical and multiple disabled children	OMT, medication	4-5	9.6
A	Speaking, hardly intelligible	Special school for physical and multiple disabled children	OMT	5	9.5
S	Speaking	Special school for physical and multiple disabled children	OMT	3-4	7.1
Mi	Speaking, hardly intelligible	Special school for speech and language disabled children	OMT	5	9.7
Mo	Speaking	Special school for physical and multiple disabled children	OMT	4	9.7
L	Speaking	Special school for physical and multiple disabled children	OMT	4	6.9
Je	Non-verbal, booklet with pictures, icons and alphabet	Special school for physical and multiple disabled children	OMT, BTX-A, surgery (rerouting)	4	9.0
Jo	Speaking, hardly intelligible	Special school for speech and language disabled children	OMT	5	8.5
R	Speaking	Regular/mainstreamed school	OMT	5	9.7

TDS, Teacher Drool Scale; VAS, Visual Analogue Scale; OMT, Oral Motor Therapy; BTX-A, Botulinum Toxin type A injections into the salivary glands

follow-up at (f) 6 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 two consecutive days and lasted either 10 or 15 minutes. 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 minutes 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

Table 7.2 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

that time. Initially, latency recordings were scheduled at the end of each session and later during this phase, when dry periods of 30 minutes were reached, these recordings 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 to 14 latency recording sessions of either 10 or 15 minutes were scheduled on two 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 two 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.

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 seconds. Inter-rater reliability was 91.2%.

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 ½, 1, 2, 5, 10, 15, 20, 25, 30, 35, 40, and 45 minutes. 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 3 consecutive trials, the next larger interval was set. If the child failed to remain dry during 3 consecutive trials or during 5 trials in sum, the previous interval was in effect again until the child succeeded in 3 consecutive trials. Once the participant had attained the 45 minutes interval, time intervals were extended either by 5, 10, or 15 minutes, depending on the participant's estimated potential (determined from learning curve and latency scores during treatment) to a maximum of 60 minutes.

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 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, 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.

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.

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 one 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 (see Appendix B). A teacher version of this questionnaire was developed and administered at the same points in time.

Results

Figure 7.1 depicts the latency (i.e., number of minutes dry) during baseline, treatment, generalization, and follow-up at 6 and 24 weeks for each participant (Note: post intervention tests are not depicted in Figure 7.1). It shows that after a three-week self-management treatment all participants were able to remain dry for intervals of 30 to 60 minutes while engaged in daily activities.

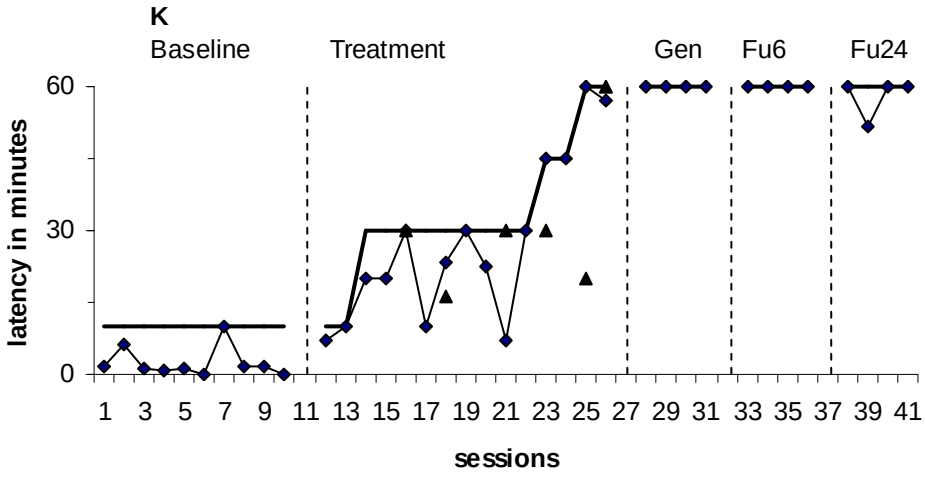
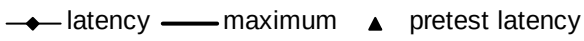


Figure 7.1 Latency (in minutes) during conditions of baseline, treatment, generalization (Gen), and follow-up at 6 (Fu6) and 24 (Fu24) weeks.



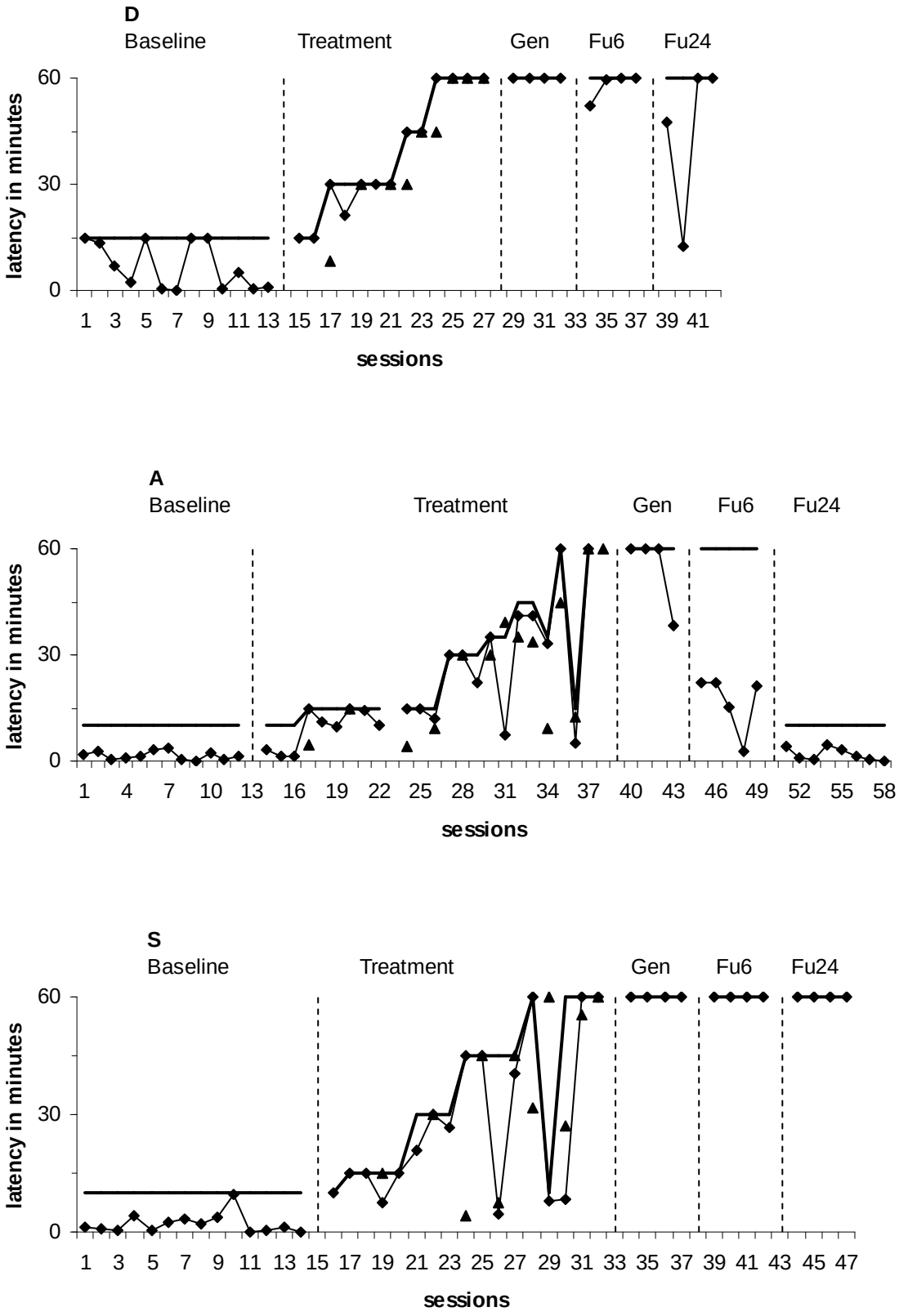


Figure 7.1 Continued

latency maximum pretest latency

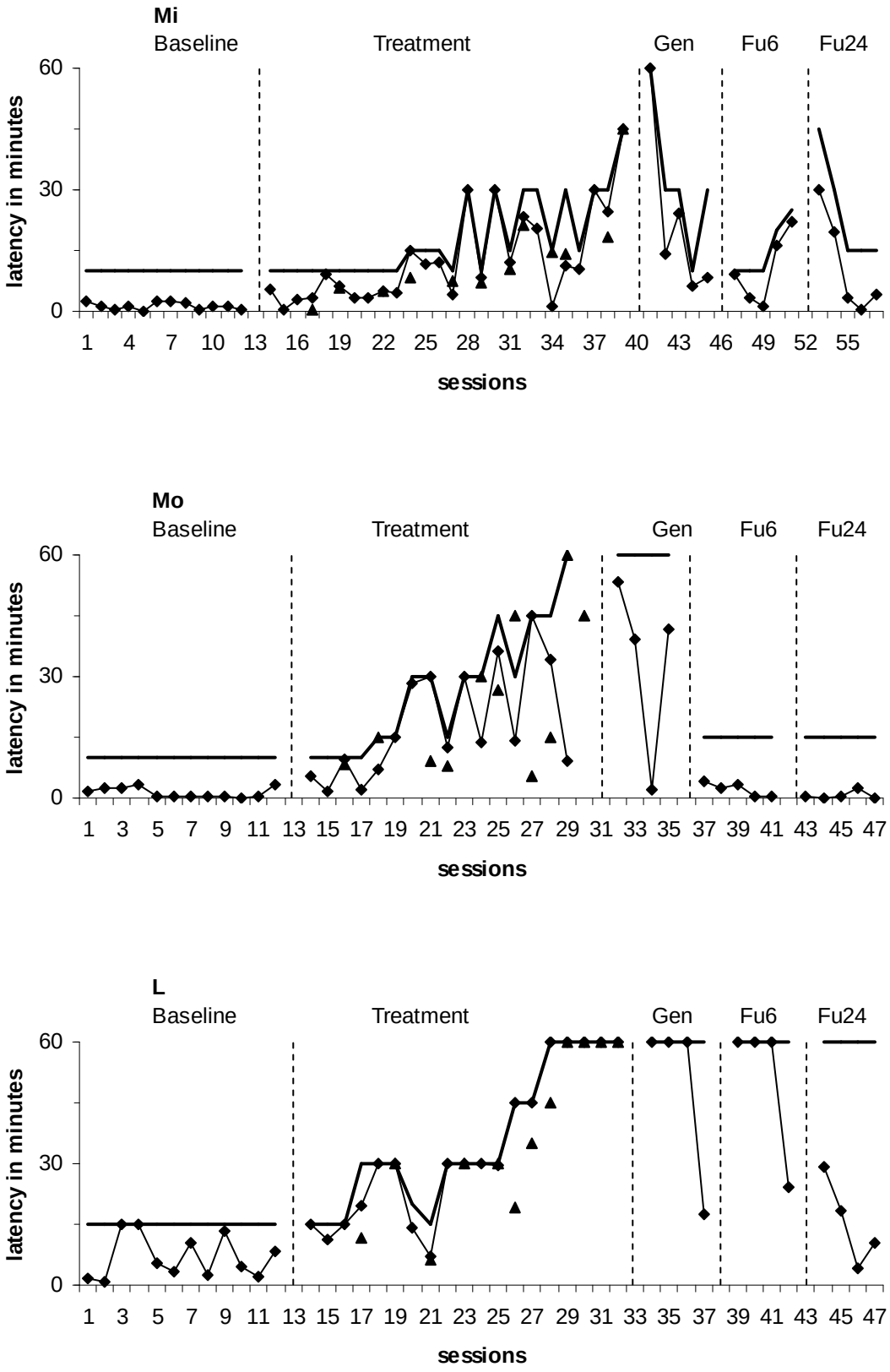


Figure 7.1 Continued

latency maximum pretest latency

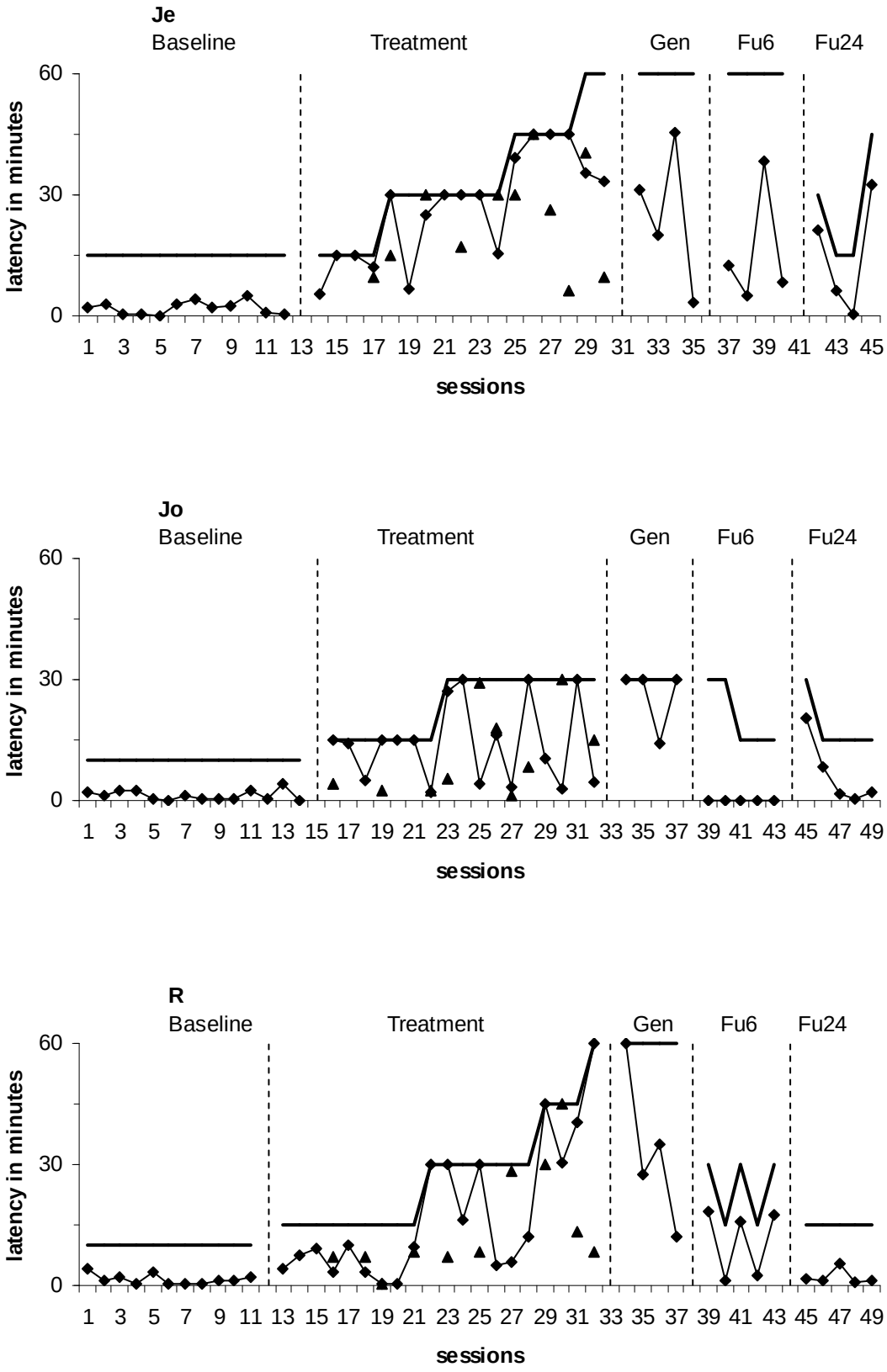


Figure 7.1 Continued

latency maximum pretest latency

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. 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 minute interval again. His learning curve did not show any further relapse.

In Table 7.3, 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 weeks follow-up (Figure 7.1). 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 weeks 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 weeks follow-up. While latency scores for Mi and Je remained above baseline level at 6 and 24 weeks follow-up, 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 7.3 Mean (range) latency in seconds at baseline and post intervention, and Cohen's *d* effect size

	Baseline		**Post intervention test**		**Effect size**
Participant	**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

Table 7.4 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 Figure 7.1, 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

Table 7.4 Individual and group scores on VAS (mean) and TDS (median) at baseline, generalization, 6 weeks follow-up (Fu 6) and at 24 weeks follow-up (Fu 24)

		VAS[a] (mean)				TDS[b] (median)			
Participant		**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

generalization, but decreased slightly to 62.4 (*SD* = 34.5; range 7.8-99.6) at 6 weeks follow-up, and 53.8 (*SD* = 38.7; range 12.7-100) at 24 weeks 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 weeks follow-up, and 3 (parents range 1-4; teacher range 1-5) at 24 weeks 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 weeks follow-up compared to baseline results.

Reduction of drooling after a three-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 and chin by parents 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 weeks 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 weeks 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 weeks follow-up. Whereas 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 6 weeks 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 weeks follow-up), social acceptance by peers (none at baseline, four at 6 weeks follow-up), and social acceptance by adults (one at baseline, six at 6 weeks 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 weeks 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 weeks follow-up), and social acceptance by peers (one at baseline, three at 6 weeks follow-up).

Discussion

From results of this case series, it may be concluded that the self-management treatment was effective in increasing time of nondrooling in a group of 10 children with severe drooling. The number of minutes of being dry increased to intervals of 30 to 60 minutes, indicating that after a three-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 weeks follow-up. 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 weeks 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 both to swallow and wipe her face when wet, just as we did. The latter combination of target behaviours 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 ten 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 to 30 minutes 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 two seconds. 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 observers' finger across participants' chin beneath the lower lip. During baseline, the individuals' chin was wiped dry afterwards 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 (nondrooling) while performing daily activities. We did not use other techniques, such as for example, time-sampling techniques, because samples of 60 minutes 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 three 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 behavioural 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 three weeks 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 their parents' and teachers' attention for nondrooling (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.

Behavioural 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 behavioural 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 behavioural procedure. Further research is also needed to explore the ways in which behavioural treatment can be supported by medical interventions.

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Chapter 8

General discussion

In his annotation, Blasco (2002) made an aside on the measurement of drooling and questions if the evaluation of treatment outcome by very precise quantification of drooling is any more meaningful than rough estimates like counting the number of bandanas or shirts soaked. For clinical purposes at least, these rough quantifications seemed the most relevant to him because: "The ultimate test of whether treatment will be continued or not is whether or not it makes caregivers' lives easier and their child's life is improved." (p. 779). Thus, if drooling severity is significantly decreased by intervention but the impact of drooling remains high, the treatment is evidence-based but may be not clinically relevant. As a consequence, researchers should not be satisfied with the effectiveness of their treatment if it is not accompanied by improvement in the quality of life.

Drooling Questionnaires

The severity of drooling in daily life situations and its impact on the ability to speak, eat, and drink, daily care and economic consequences, social interaction, and self-esteem can all be successfully investigated by our questionnaires (see Appendix 1). As such, they offer both information about the impact of drooling on ICF domains of activities and participation, and Health Related Quality of Life. From data collected with these questionnaires, it appears that drooling has a differential impact on children with cerebral palsy (CP), depending on the severity of disabilities. In addition, the questionnaires are sensitive to change. Contingent on reduction of salivary flow rate after bilateral submandibular treatment with Botulinum Toxin type A (BTX-A) and a corresponding decrease in drooling severity (measured by the drooling quotient, and VAS and TDS scores), the impact of drooling changed in a positive direction. Subsequently, with the diminishing of the effect of BTX-A, flow rate and drooling severity returned to baseline levels and the impact scores returned toward baseline level as well. To evaluate the impact of drooling and treatment efficacy, these questionnaires add to existing condition-specific questionnaires for children with cerebral palsy that do not cover this topic. By utilizing parental report, the questionnaires are applicable for all drooling children, even for those with profound intellectual disabilities.

The drooling questionnaires also enable us to compare the effect of different types of treatment (surgery, medication, behavioural therapy, oral motor training) on the severity of drooling in specific daily life situations, on the ability to speak, eat, and drink, on daily care and economic consequences, as well as on social interaction and self-esteem. Finally, the differential effect of treatments across subgroups of drooling children (based on, for instance, severity of drooling or degree of learning disability) can be documented. Perhaps data from the questionnaires can support future decisions

regarding the preferred treatment strategy for an individual child or subgroups of drooling children.

Psychometrics

The questionnaires on drooling measure the construct 'impact of drooling on daily life, social interaction, and self-esteem', and all questions clearly refer to this subject. Both content and face validity of both questionnaires are adequate. After careful selection of questions, parents of children with CP who drool confirmed that the questionnaires cover all relevant aspects in which drooling influences their daily life. Predictive validity seems to have no relevance, since it is not our aim to *predict* anything from the results of our questionnaires: neither in relation to another variable nor to another point in time. The primary focus is to evaluate and document changes in the impact of drooling on daily life, social interaction, and self-esteem of CP children and their parents after treatment. Investigation on concurrent validity requires an instrument that measures the same construct. Unfortunately, there is no such instrument available yet. Data shown in this thesis suggest that results on the drooling questionnaires covariate with primary measures of drooling severity. Reliability of the questionnaires was not assessed.

To enhance homogeneity, it is recommended that throughout a study the same parent (either the mother or the father) completes the questionnaires to prevent intersubject variations. Also, questionnaires should be sent and completed after inclusion for treatment to prevent parents from deliberately exaggerating the impact of drooling to be considered for treatment. Retrospective telephone interviews on long term effect and QOL outcome of treatment for drooling are frequently reported (Martin & Conley, 2007; Syeda, Ahsan, & Nunez, 2007). The accuracy of answers from these interviews, several years after treatment, is questionable. By asking parents to report on changes in the last two to four weeks, more reliable answers on the questionnaires can be expected.

Short version

Although parents were eager to participate in the study by Jongerius (2004), reactions during follow-up assessments at the clinic revealed that the original questionnaires were too labour-intensive and were administered too often (six times in the course of 24 weeks). To reduce non-responding and superficial answering, we designed a short version containing questions from both questionnaires. This short version was applied in the study on self-management treatment of drooling and administered only three times: at baseline, and 6 and 24 weeks after intervention.

The short questionnaire (see Appendix B) can be used to assess the effect of any treatment for drooling. In future research, we intend to collect data on drooling severity and its impact on the life of children and their parents before and after different kinds of treatment.

Behavioural therapy

Based on the reviews of the literature in this thesis it appears that, although promising behavioural programs for drooling have been described, the procedures are very different from each other, the total number of participants (e.g., *N* = 55) is small, and the evidence-base is limited. This holds particularly true for self-management procedures for drooling, because data from only two successful case studies have been reported (i.e., Dunn, Cunningham, & Backman, 1987; Thorbecke & Jackson, 1982). Our case series provides additional support for the effectiveness of a self-management treatment of drooling, although the procedure needs further elaboration for cases in which generalization and maintenance is insufficient. Although Dunn and colleagues (1987) conclude that: "it is unlikely that the self-control is dependent on age and developmental factors, as a similar effect had been found in a variety of children seen at our clinic, ranging in age from eight to thirteen years with developmental levels from four to 10 years" (p. 309), our study of 10 cases shows that with follow-up evaluation, differential long-term effects of a self-management program for drooling emerge that may be related to differences in age and/or developmental factors.

During the behavioural intervention study, two children dropped out after inclusion. One of them with an incorrect neurological diagnosis at inclusion, showing severe loss of both strength and (oral) motor function during treatment. The other drop-out began to exhibit non-compliant behaviour during the study. Initially, this participant also showed some progress, but when interval length was increased, it appeared that the self-management procedure became difficult for her to adhere to, and was too demanding. Her non-compliance ultimately caused early discharge from the program. These cases show the need for careful selection of candidates for this self-management treatment and to exclude children with severe non-compliant behaviour and children with occasional loss of strength and function due to, for instance, epilepsy or neurological decay.

Social validity of treatment results

Although effectiveness of different kinds of treatment is frequently claimed on the basis of significant decrease in flow rate and drooling severity, the amount of drooling following treatment may still be considerable. For instance, Greensmith et al. (2005) defined a clinically significant improvement as an improvement by at least 1 point on the Thomas-Stonell and Greenberg classification (1988), which consists of a 5-point scale for drooling severity and a 4-point scale for drooling frequency (see Table 8.1). After bilateral submandibular duct transposition combined with bilateral sublingual gland excision, the median score for the frequency of drooling of 67 of their patients with 2 years follow-up data decreased from 4.0 to 2.9, and the median score for the severity of drooling

decreased from 4.8 to 3.0. For 41 patients followed to 5 years both the median frequency and severity scores remained at 3. The same holds true for the results of bilateral submandibular injections of BTX-A (Jongerius, 2004): mean scores on the Teacher Drool Scale decreased from 5 (constant drooling, always wet) to 3 (occasional drooling: intermittent all day) at eight weeks after injections, with a slight increase at 24 weeks (mean TDS 3.8). These mean scores from both studies indicate that after treatment, drooling is still observed several times a day. Since the social disabling effect of drooling is already apparent when any saliva is below the lower lip line on the chin for a few seconds, these medical treatments do improve the condition, but do not resolve the problem of drooling.

Table 8.1 Drooling severity and frequency ranks (Thomas-Stonell & Greenberg, 1988)

Severity		**Frequency**
1. Dry	Never drools	1. Never drools
2. Mild	Only the lips are wet	2. Occasional drooling
3. Moderate	Wet on the lips and chin	3. Frequent drooling
4. Severe	Clothing becomes damp	4. Constant drooling
5. Profuse	Clothing, hands, tray, and objects become wet	

In our intervention study, drooling was defined as saliva (either a drop or a string) present beneath the lower lip line or a string falling directly from the mouth for a period longer than 2 seconds without the individual cleaning his/her face and/or clothes. The dependent variable was latency in minutes of dry periods until drooling occurred while performing daily activities (dual task). From this point of view, our definition of the dependent variable was rather conservative compared to earlier studies: if any saliva appeared beneath the lower lip line and the child did not react within 2 seconds the latency recording was stopped immediately, even though the child may have reacted soon after this period with the appropriate responses and stayed dry for another substantial time interval. Thus, compared to preceding studies, our data may be underestimating the treatment effect.

Final remarks

Reviews on the management of drooling generally advocate the oral motor and behavioural approach as a first approach, medication and BTX-A injections as a secondary option, and surgery for those who fail to respond to the former treatment modalities. Although this seems the most plausible sequence of treatment strategies as it

moves from least to most intrusive, this sequence itself is not evidence-based. In our behavioural study, participants were often treated with BTX-A or even had surgery before inclusion. Behavioural treatment should be considered both before, but also after these medical interventions if drooling is still a problem.

As we learned from this study, the impact of drooling is dependent on personal and environmental factors. In addition, the best option for treatment of drooling is also dependent on personal and environmental factors. Which treatment option is preferred at a specific point in time depends on multiple factors, such as severity of drooling, type of drooling (posterior or anterior), (oral) motor ability, age, intellectual ability, intrinsic motivation, and the overt social reactions from peers and others at home or at school (either mainstreamed or residential). Because the decision making process on treatment for drooling requires multifactor analysis, we are convinced that a multidisciplinary approach is necessary. Multidisciplinary expert teams that evaluate and treat this problem should consist of a speech therapist, physiatrist, child neurologist, ENT (Ear, Nose and Throat) surgeon, and - based on the conclusions of this thesis - a behavioural therapist. Detailed and standardized description of demographic variables of participants in effect studies on all kinds of treatment for drooling, both medical and behavioural, is necessary to eventually formulate a clinical decision making model for differential indication of treatment options across childhood.

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Summary

Chapter 1

Severe drooling negatively affects many aspects of daily life, social interactions, and the development of self-esteem. As such, it influences the quality of life of both parents and children with severe drooling. The first part (i.e., chapters 2 to 4) of this thesis addresses the measurement of the impact of severe drooling on the lives of children with cerebral palsy (CP) and their parents. In addition, the longitudinal effects of reduced salivary flow after scopolamine and bilateral injection of Botulinum Toxin Type A (BTX-A) in the submandibular glands on the impact of drooling is evaluated. The second part (i.e., chapters 5 to 7) is focused on behavioural treatment of drooling, its evidence-base and the development and evaluation of a new self-management programme.

Chapter 2

To assess drooling severity in a variety of everyday conditions, and its impact on the daily life of children and their families, two parent questionnaires were constructed. The first questionnaire covers drooling severity in specific daily life situations, its impact on oral motor activities and daily care, and its economic consequences. The second questionnaire measures the impact of drooling on social interactions and self-esteem. Results of baseline measurements of 43 children with CP and severe drooling showed that the questionnaires measured the variation in drooling severity across daily life conditions, and enabled evaluation of the impact of drooling on the ability to eat, drink, and speak, as well as on daily care, economic consequences, and social interactions. The section of the questionnaire that addresses the impact of drooling on self-esteem appeared not to be fully applicable for non-speaking children with a low developmental status. However, this may have been due to the fact that our sample included 32 children with a developmental level below 6 years (of which 23 scored below 4 years). It was concluded that the questionnaires offer a qualitative method to evaluate parental perceptions of the impact of drooling and to evaluate the effectiveness of interventions to reduce drooling.

Chapter 3 and 4

The impact of salivary flow reduction following medication (scopolamine and intraglandular BTX-A injection in the submandibular glands) on daily life and provision of care (chapter 3), and on social interaction and self-esteem (chapter 4) was evaluated in a group of 45 children with CP (age 3 to 16 years) who suffered from severe drooling. Eight children were independently ambulant, whereas 37 children used a wheelchair. Thirty-four children had a learning disability with a developmental age below 6 years. Both questionnaires were administered during the use of scopolamine and up to 24 weeks after BTX-A injection. Six participants dropped out of the study; data on 39

children were analyzed. Results showed that anticholinergic agents effectively reduced salivary flow. Drooling diminished substantially and this was accompanied by a significant reduction in care needs, making daily care less demanding. The amount of reported damage to communication devices and computers decreased. The reduction of drooling was also related to increased social interactions with peers. However, parents perceived that the impact of drooling had increased on the level of the child's satisfaction on physical appearance, relations within the extended family, and life in general. This may reflect their disappointment as the treatment effect gradually decreased during the 24 weeks following BTX-A injection, leading to a gradual increase in drooling. Unfortunately, although medication led to (temporary) positive changes, many social and emotional consequences of drooling remained unchanged during this study.

Chapter 5 and 6

In chapter 5, a descriptive analysis is presented on studies on the behavioural treatment of drooling (published between 1970 and 2005). The 17 articles which met the inclusion criteria described 53 participants (age 6 to 28 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 disabilities. Forty-two participants (79%) were diagnosed with CP. Behavioural procedures included instruction, positive and negative reinforcement, overcorrection and restitution, verbal and automatic cueing, and/or self-management. It was concluded that 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 of behavioural intervention in terms of number of studies and their quality in this area is limited. Fifteen studies used a single subject design; two studies implemented an experimental-control group design. Some of these studies are poorly designed and methodological flaws were identified. Therefore, general conclusions about efficacy of behaviour therapy for drooling and/or best practice cannot be drawn yet, although our analysis suggests that behavioural treatment in general is promising.

In chapter 6, 19 behavioural studies published since 1970 were reviewed to formulate guidelines for clinical use of behavioural procedures for drooling. It is concluded that (a) instruction, prompting, and positive reinforcement are effective only if systematically applied and if not withdrawn after an initial effect is established, (b) negative social reinforcement and decelarative procedures are socially acceptable only after non-aversive procedures have been shown to be ineffective, (c) cueing techniques should be considered if self-management procedures appear not to be effective or contraindicated; their introduction must be carefully planned; participants often remain dependent on these

devices, which are not readily available and often susceptible to damage, and (d) until now, self-management procedures for drooling have rarely been studied (*n* = 2) and such procedures do not seem to be applicable in participants with severe and profound retardation and a developmental age below 4 years.

Chapter 7

The effectiveness of a self-management procedure for drooling was evaluated in a center-based case-series study (*n* = 10). Participants were between 7;0 and 19;9 years of age, and suffered from poor motor and oral motor function and severe drooling. During the 3-week intervention, they were taught self-monitoring skills and to frequently swallow and wipe their mouth and chin whenever drooling occurred. Data were collected in a non-concurrent multiple baseline design across individuals. Latency recordings, defined as the time interval in minutes without drooling, showed that all participants remained dry for intervals of 30 to 60 minutes while being engaged in daily activities. For all participants generalization to the classroom occurred, and for three participants maintenance of treatment effect was established at 6 and 24 weeks. However, seven participants failed to maintain self-management skills at follow-up. In five participants drooling returned to baseline level. Parental and teachers' judgment of drooling severity during the day showed positive results up to 24 weeks follow-up and, moreover, positive changes in the impact of drooling on daily care, social interactions, and self esteem were reported as well. Although the self-management procedure showed promising results, further adaptations are required to improve generalization and maintenance.

Chapter 8

The questionnaires developed in this study offer information about the impact of drooling on ICF domains of activities and participation and on Health Related Quality of Life. Based on the data collected with these questionnaires, it appears that drooling has a differential impact on children with CP, depending on the severity of disabilities. In addition, the questionnaires are sensitive to change. Because the original questionnaires appeared too labour-intensive and were perceived as having been administered too frequently (six times in the course of 24 weeks), a short version containing questions from both questionnaires was designed in order to reduce non-responding and superficial answering. In future research, this short version can be used to compare the effect of different types of treatment (e.g., surgery, medication, behavioural therapy, oral motor training) on drooling severity and its impact on the lives of children and their parents. In addition, the differential effect of treatments across subgroups of drooling children (based on, for instance, severity of drooling or level of learning disability) can be documented.

Behavioural 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, BTX-A injections, and surgery. Reviews on the management of drooling generally advocate the oral motor and behavioural approach as basic, medication and BTX-A injections as a secondary option, and surgery for those who fail to respond to the former treatment modalities. Although this seems the most plausible sequence of treatment strategies as it moves from least to most intrusive, this sequence itself is not evidence-based. Analysis of studies on medical treatment showed that, although effectiveness of this type of treatment is frequently claimed on the basis of a significant decrease in flow rate and drooling severity, the amount of drooling following treatment may still be considerable. Since the socially disabling effect of drooling is already apparent when any saliva is below the lower lip line on the chin for a few seconds, although such medical treatments do improve the condition, they cannot be said to resolve the problem of drooling. It was concluded that behavioural treatment should be considered both before, but also after these medical interventions if drooling is still a problem.

Samenvatting

Hoofdstuk 1

Ernstig kwijlen heeft een negatieve invloed op veel praktische aspecten van het dagelijks leven, de sociale interactie en de ontwikkeling van zelfwaardering. Als zodanig beïnvloedt kwijlen de kwaliteit van leven van zowel ouders als kinderen die ernstig kwijlen. Het eerste deel van dit proefschrift (hoofdstuk 2 tot en met 4) gaat over het meten van de impact van ernstig kwijlen op het leven van kinderen met cerebrale parese (CP) en hun ouders. Bovendien wordt het longitudinaal effect van gereduceerde speekselvloed na behandeling met scopolamine en bilaterale injectie van Botuline Toxine type A (BTX-A) in de submandibulaire speekselklieren, op de impact van het kwijlen geëvalueerd. Het tweede deel (hoofdstuk 5 tot en met 7) beschrijft de mogelijkheden van gedragstherapeutische behandeling van kwijlen, de empirische evidentie voor deze aanpak en de ontwikkeling en evaluatie van een nieuw zelfmanagement programma.

Hoofdstuk 2

Om de ernst van het kwijlen in verschillende dagelijkse situaties en de impact ervan op het dagelijks leven van kinderen en hun ouders te meten, werden twee vragenlijsten voor ouders geconstrueerd. De eerste vragenlijst inventariseert de ernst van het kwijlen in specifieke dagelijkse situaties, en de invloed van het kwijlen op mondmotorische activiteiten en de dagelijkse verzorging, en de economische consequenties ervan. De tweede vragenlijst inventariseert de impact van kwijlen op sociale interacties en zelfwaardering. Resultaten van basislijn metingen bij 43 kinderen met CP die ernstig kwijlen, lieten zien dat de vragenlijsten variatie in ernst van kwijlen meten en geschikt zijn voor de evaluatie van de impact van kwijlen op het eten, drinken, en spreken, evenals op de dagelijkse verzorging, de economische consequenties en sociale interacties. De sectie van de vragenlijst over de impact van kwijlen op de zelfwaardering bleek niet volledig toepasbaar bij niet-sprekende kinderen met een laag ontwikkelingsniveau. Dit zou echter het gevolg kunnen zijn van het feit dat het ontwikkelingsniveau van 32 kinderen uit deze steekproef lager dan 6 jaar was (waarvan 23 kinderen onder 4 jaar scoorden). Geconcludeerd werd dat de vragenlijsten een kwalitatieve methode bieden om de indruk van ouders over de impact van kwijlen te inventariseren en het effect van interventies gericht op het verminderen van kwijlen te evalueren.

Hoofdstuk 3 en 4

Het effect van reductie van speekselvloed na medicatie (scopolamine en bilaterale BTX-A injecties in de submandibulaire speekselklieren) op de praktische gevolgen van kwijlen in het dagelijks leven en de dagelijkse verzorging (hoofdstuk 3), en op de sociale interactie en de zelfwaardering (hoofdstuk 4) werd geëvalueerd in een groep van 45 kinderen met

CP (leeftijd 3 tot en met 16 jaar) die ernstig kwijlden. Acht kinderen konden zelfstandig lopen zonder hulpmiddel, 37 kinderen gebruikten een rolstoel. Vierendertig kinderen hadden leer- en ontwikkelingsproblemen met een ontwikkelingsniveau beneden 6 jaar. Beide vragenlijsten werden afgenomen gedurende het gebruik van scopolamine en tot 24 weken na de BTX-A injecties. Zes deelnemers vielen gedurende het onderzoek uit (z.g. drop-outs); data van 39 kinderen konden worden geanalyseerd. De resultaten toonden aan dat anticholinergica de speekselvloed effectief verminderden. Het kwijlen verminderde substantieel en dit ging gepaard met een significante vermindering in verzorgingsbehoefte, waardoor de dagelijkse zorg voor ouders minder veeleisend werd. Het aantal gerapporteerde beschadigingen van communicatieapparaten en computers verminderde eveneens. De vermindering van kwijlen was ook gerelateerd aan toegenomen sociale contacten met leeftijdgenoten. Ouders rapporteerden ook dat in hun ogen de invloed van kwijlen op de tevredenheid van hun kind met het fysieke uiterlijk, de relaties in de familie en het leven in het algemeen toenam. Mogelijk komt hierin de teleurstelling tot uitdrukking dat het effect van de behandeling langzaamaan verminderde gedurende de 24 weken na de BTX-A injecties waardoor het kwijlen weer toenam. Hoewel medicatie tot (tijdelijke) positieve veranderingen leidde, bleven veel sociale en emotionele consequenties helaas onveranderd gedurende deze studie.

Hoofdstuk 5 en 6

In hoofdstuk 5 wordt een beschrijvende analyse gepresenteerd van studies naar gedragstherapeutische behandelingen van kwijlen (gepubliceerd tussen 1970 en 2005). De 17 artikelen die aan de inclusiecriteria voldeden, beschrijven de behandeling van 53 deelnemers (leeftijd 6 tot en met 28 jaar). Voor 60% van de deelnemers werd het niveau van verstandelijke beperking gerapporteerd, variërend van ernstig en zeer ernstig (*n* = 24: 75%), matig (*n* = 4: 13%), tot mild (*n* = 2: 6%), terwijl 2 participanten (6%) geen leerproblemen hadden. Twee-en-veertig deelnemers (79%) hadden de diagnose CP. Gedragstherapeutische procedures die werden toegepast waren: instructie, positieve en negatieve bekrachtiging, overcorrectie en restitutie, verbale en automatische cueing, en/of zelfmanagement technieken. Geconcludeerd werd dat effectieve gedragstherapeutische programma's voor kwijlen zijn gerapporteerd voor kinderen met en zonder leerproblemen en/of motorische problemen. Zelfs participanten met zeer ernstige leerproblemen kunnen baat hebben bij gedragstherapeutische behandeling voor kwijlen. Echter, de empirische evidentie voor gedragstherapeutische behandeling voor kwijlen in termen van aantallen studies en de kwaliteit ervan is beperkt. Vijftien studies gebruikten een single subject design; twee studies implementeerden een experimental-control group design. Sommige studies waren zwak van opzet en hadden methodologische tekortkomingen. Daarom kunnen nog geen algemene conclusies worden

getrokken over de effectiviteit van gedragstherapie voor kwijlen, hoewel de analyse suggereert dat deze methode in het algemeen een veelbelovende optie is.
In hoofdstuk 6 worden 19 studies naar gedragstherapeutische behandeling van kwijlen (gepubliceerd sinds 1970) besproken met als doel om richtlijnen te formuleren voor de klinische toepassing van gedragstherapeutische procedures in de behandeling van kwijlen. Geconcludeerd wordt dat (a) instructie, prompting, en positieve bekrachtiging alleen effectief zijn wanneer deze technieken systematisch worden toegepast en niet worden afgebouwd nadat een initieel effect tot stand is gebracht; (b) negatieve sociale bekrachtiging en straffende procedures pas sociaal aanvaardbaar zijn als niet-aversieve procedures ineffectief zijn gebleken; (c) cueing technieken overwogen kunnen worden als zelfmanagement procedures niet effectief blijken of gecontraïndiceerd zijn; de introductie ervan moet zorgvuldig gepland worden; gebruikers blijven vaak afhankelijk van het cueingapparaat en deze apparaten zijn niet makkelijk verkrijgbaar en vaak gevoelig voor storing; en (d) zelfmanagement procedures voor kwijlen zijn tot nu toe zelden bestudeerd (n = 2) en deze procedures lijken niet toepasbaar bij personen met ernstige en zeer ernstige leerproblemen en een ontwikkelingsleeftijd onder 4 jaar.

Hoofdstuk 7

De effectiviteit van een zelfmanagement programma voor kwijlen werd geëvalueerd in een centrum-gebonden case-series studie (n = 10). Participanten (leeftijd 7;0-19;9 jaar) waren bekend met motorische beperkingen en ernstig kwijlen. Tijdens een 3-weekse interventieperiode leerden zij zelf-monitoring vaardigheden, regelmatig slikken en zodra speekselverlies optrad, hun mond en kin direct af te vegen. Data werden verzameld in een non-concurrent multiple baseline design across individuals. Latentietijd metingen (gedefinieerd als tijdsinterval in minuten waarin de participant niet kwijlt) toonden aan dat alle deelnemers na de interventie langere periodes van uiteindelijk 30 tot 60 minuten ononderbroken droog bleven terwijl ze dagelijkse activiteiten uitvoerden. Generalisatie van het effect van de behandeling naar de klassensituatie op de eigen school trad op bij alle participanten. Voor drie participanten bleef het effect van de interventie na 6 en 24 weken in stand. Echter, voor zeven participanten was dit niet het geval en voor vijf van hen nam het kwijlen weer toe tot basislijn niveau. De beoordeling van de ernst van kwijlen door ouders en leerkrachten lieten positieve resultaten zien tot aan de metingen 24 weken na de interventie, en zij rapporteerden tot aan de laatste metingen ook positieve veranderingen in de impact van kwijlen op dagelijkse verzorging, sociale interactie en zelfwaardering van de deelnemers. Hoewel deze studie veelbelovende resultaten had, moet de procedure worden aangepast om generalisatie en handhaving van het effect te verbeteren.

Hoofdstuk 8

De vragenlijsten die in deze studie ontwikkeld werden, geven informatie over de impact van kwijlen op de ICF domeinen van activiteiten en participatie en over gezondheidgerelateerde kwaliteit van leven. Uit resultaten van deze vragenlijsten blijkt dat kwijlen een gedifferentieerde impact heeft op kinderen met CP, afhankelijk van de ernst van hun beperkingen. Daarnaast blijken deze vragenlijsten gevoelig voor (het meten van) veranderingen in de impact van kwijlen. Omdat de oorspronkelijke vragenlijsten te arbeidsintensief bleken en ouders aangaven dat ze te vaak in korte tijd werden aangeboden (zes keer in een periode van 24 weken), werd een korte versie met een selectie van vragen uit beide vragenlijsten gemaakt, om niet of slechts oppervlakkig antwoorden te verminderen. In toekomstig onderzoek kan deze korte versie gebruikt worden om de effecten van verschillende behandelingsmethoden (chirurgie, medicatie, gedragstherapie, mondmotorische therapie) op de ernst van het kwijlen en de impact ervan op het leven van kinderen en ouders te evalueren. Daarnaast kan het gedifferentieerde effect van de behandeling op verschillende subgroepen van kwijlende kinderen (gebaseerd op bijvoorbeeld de ernst van het kwijlen of de ernst van leerproblemen) gedocumenteerd worden.

Gedragstherapie voor kwijlen is een optie die kan worden beschouwd in relatie tot de voor- en nadelen van mondmotorische therapie, training met orthodontische hulpmiddelen, medicatie, BTX-A injecties en chirurgische ingrepen. Overzichtsartikelen over de behandeling van kwijlen pleiten in het algemeen voor mondmotorische therapie en gedragstherapie als de basale optie, medicatie en BTX-A injecties als secundaire optie, en chirurgie voor hen die niet afdoende reageren op eerder genoemde behandelingsmodaliteiten. Hoewel dit een plausibele sequentie van behandelstrategieën lijkt vanwege het feit dat op deze manier van de minst naar meest ingrijpende aanpak wordt gegaan, is er geen empirische evidentie voor deze sequentie zelf. De analyse van studies over medische behandeling van kwijlen laat zien dat, hoewel vaak effectiviteit wordt geclaimd op basis van significante vermindering van speekselvloed en van de ernst van het kwijlen, de mate van kwijlen na behandeling nog altijd aanzienlijk kan zijn. Omdat het sociaal beperkend effect van kwijlen al merkbaar is als zich slechts enige seconden enig speeksel onder de onderste liprand op de kin bevindt, verminderen deze medische ingrepen wel de ernst van het kwijlen, maar niet noodzakelijk ook de ernst van het probleem. Daarom moet gedragstherapie zowel voorafgaand aan, maar ook volgend op de medische behandeling worden overwogen als het kwijlen voor het kind en zijn ouders een probleem blijft.

Appendix A

Questionnaires on drooling

Questionnaire 1

- *The severity of drooling in specific daily life situations*

1. During the past two weeks the degree of drooling was

   [----------]

   very severe no drooling

2. Indicate the degree of drooling for each time during the day during the past two weeks.

	Time of the day	**Degree of drooling**				
	morning	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	afternoon	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	evening	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	night	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)

3. Mark all conditions in which your child is drooling in the first column. If yes, indicate the degree of drooling for each condition in the past two weeks.

	Condition	**Degree of drooling**				
	supported sit	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	unsupported sit	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	prone position	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	supine position	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	walking	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	intensive movement (sports)	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	ill	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	tired	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	eating	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	smell of food	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	certain tastes or foodstuffs	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	drinking	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	talking	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	sleeping	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	concentrated activity	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	relaxed watching TV	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	strenuous activity	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	enthusiastic	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	swallowing medication	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	after swallowing medication	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	after brushing teeth	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	crying	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	allergic respiratory reactions	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)

4. During the past two weeks the child suffered from a dry mouth

[----------]

very often never

- *The impact of drooling on the ability to speak, eat, and drink*

5. For us, parents, the child's speech is

[----------]

not intelligible always intelligible

6. For outsiders (those who do not interact with the child on a daily base) the child's speech is

[----------]

not intelligible always intelligible

7. The quality of speech during the past two weeks is

[----------]

very poor very good

8. We are contented with the child's speech

[----------]

strongly disagree strongly agree

9. How does your child eat?
   a. oral feeding
   b. tube feeding
   c. partially oral and partially tube feeding

10. During the past two weeks the child's eating was

[----------]

very poor very good

11. We are contented with the child's eating

[----------]

strongly disagree strongly agree

12. During the past two weeks the child's drinking was

[----------]

very poor very good

13. We are contented with the child's drinking

[----------]

strongly disagree strongly agree

- *The impact of drooling on daily care and economic consequences*

14. What measures do you take because of drooling?
    a. child wears bib
    b. child wears shawl
    c. child wears terry cloth wristbands
    d. child has washable toys
    e. books are plasticized
    f. other measures, ……..

15. How often is his/her mouth or chin wiped dry? …. times per hour **or** …. times per day

16. How often is he/she told to swallow? …. times per hour **or** …. times per day

17. How often is his/her bib or shawl replaced? …. times per day

18. How often are his/her clothes changed? …. times per day

19. How many loads of wash do you do? …. loads of wash per week

20. Has there been damage to
    a. clothes yes / no
    b. toys yes / no
    c. books yes / no
    d. furniture yes / no
    e. communication aids yes / no
    f. electronic communication devices yes / no / not relevant
    g. computer yes / no / not relevant
    h. audio equipment yes / no / not relevant

21. Did you curtail your child's playing with objects that might be damaged by drooling?
    yes / no

Questionnaire 2

- *The impact of drooling on social interaction*

1. Does your child play with other children at school?
   0 yes
   0 no: because of drooling? yes / no / don't know
   0 don't know

2. Does your child play with other children at home?
   0 yes
   0 no: because of drooling? yes / no / don't know
   0 don't know

3. Did you notice your child being avoided by other children?
   0 yes: because of drooling? yes / no / don't know
   0 no

4. Did you notice your child being avoided by familiar or unfamiliar adults?
   0 yes: because of drooling? yes / no / don't know
   0 no

5. Did you notice that adults keep their distance from your child?
   0 yes: because of drooling? yes / no / don't know
   0 no

6. Did you notice that familiar or unfamiliar adults underestimate the mental capacity of your child?
   0 yes: because of drooling? yes / no / don't know
   0 no

7. Did you notice that you yourself kept your distance from your child because of drooling?
   0 yes
   0 no

- *The impact of drooling on emotional development (self-esteem)*

Parent impression

During the past 4 weeks, how satisfied do you think your child has felt about:

8. his/her school ability?

[----------]

very dissatisfied very satisfied

To what extent does drooling contribute to the level of satisfaction of your child in this domain?

[----------]

not at all very important

9. his/her participation in sports and play?

[----------]

very dissatisfied very satisfied

To what extent does drooling contribute to the level of satisfaction of your child in this domain?

[----------]

not at all very important

10. his/her social contact with other children?

[----------]

very dissatisfied very satisfied

To what extent does drooling contribute to the level of satisfaction of your child in this domain?

[----------]

not at all very important

11. his/her physical appearance?

[----------]

very dissatisfied very satisfied

To what extent does drooling contribute to the level of satisfaction of your child in this domain?

[----------]

not at all very important

12. his/her relations within the family?

[----------]

very dissatisfied very satisfied

To what extent does drooling contribute to the level of satisfaction of your child in this domain?

[----------]

not at all very important

13. his/her life in general?

[----------]

very dissatisfied very satisfied

To what extent does drooling contribute to the level of satisfaction of your child in this domain?

[----------]

not at all very important

Emotional reactions of the child

Although some drooling children cannot express their feelings on the following subjects because of either their age and/or cognitive and/or communication disabilities, we ask you in this section to report only your child's reactions.

During the past 4 weeks, did you notice your child expressing any overtly positive and/or negative feelings about:

14. his/her physical appearance?
    - 0 no
    - 0 yes, positive feelings
    - 0 yes, negative feelings: because of drooling? yes / no

15. his/her competence?
    - 0 no
    - 0 yes, positive feelings
    - 0 yes, negative feelings: because of drooling? yes / no

16. his/her social acceptance by peers?
    - 0 no
    - 0 yes, positive feelings
    - 0 yes, negative feelings: because of drooling? yes / no

17. his/her social acceptance by adults?
    - 0 no
    - 0 yes, positive feelings
    - 0 yes, negative feelings: because of drooling? yes / no

Appendix B

Questionnaire on drooling, short version

Questionnaire (short version)

- *The severity of drooling in specific daily life situations*

1. During the past two weeks the degree of drooling was

   [----------]

   very severe no drooling

2. Mark all conditions in which your child is drooling in the first column. If yes, indicate the degree of drooling for each condition in the past two weeks.

	Condition	**Degree of drooling**				
	supported sit	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	unsupported sit	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	prone position	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	supine position	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	walking	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	intensive movement (sports)	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	ill	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	tired	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	eating	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	smell of food	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	certain tastes or foodstuffs	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	drinking	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	talking	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	sleeping	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	concentrated activity	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	relaxed watching TV	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	strenuous activity	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	enthusiastic	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	swallowing medication	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	after swallowing medication	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	after brushing teeth	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	crying	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)
	allergic respiratory reactions	none (1)	mild (2)	moderate (3)	severe (4)	very severe (5)

- *The impact of drooling on daily care and economic consequences*

3. How often is his/her mouth or chin wiped dry? …. times per hour **or** …. times per day

4. How often is he/she told to swallow? …. times per hour **or** …. times per day

5. How often is his/her bib or shawl replaced? …. times per day

6. Has there been damage to communication aids, electronic communication devices, computer, and/or audio equipment? yes / no

7. Has there been damage to floors and/or furniture? yes / no

- *The impact of drooling on social interaction*

8. Did you notice your child being avoided by other children?
   0 yes: because of drooling? yes / no / don't know
   0 no

9. Did you notice your child being avoided by familiar or unfamiliar adults?
   0 yes: because of drooling? yes / no / don't know
   0 no

10. Did you notice that familiar or unfamiliar adults underestimate the mental capacity of your child?
    0 yes: because of drooling? yes / no / don't know
    0 no

- *The impact of drooling on emotional development (self-esteem)*

<u>Parent impression</u>

During the past 4 weeks, how satisfied do you think your child has felt about:

11. his/her social contact with other children?

[----------]

very dissatisfied very satisfied

To what extent does drooling contribute to the level of satisfaction of your child in this domain?

[----------]

not at all very important

12. his/her physical appearance?

[----------]

very dissatisfied very satisfied

To what extent does drooling contribute to the level of satisfaction of your child in this domain?

[----------]

not at all very important

13. his/her relations within the family?

[----------]

very dissatisfied very satisfied

To what extent does drooling contribute to the level of satisfaction of your child in this domain?

[----------]

not at all very important

14. his/her life in general?

[----------]

very dissatisfied very satisfied

To what extent does drooling contribute to the level of satisfaction of your child in this domain?

[----------]

not at all very important

Emotional reactions of the child

Although some drooling children cannot express their feelings on the following subjects because of either their age and/or cognitive and/or communication disabilities, we ask you in this section to report only your child's reactions.

During the past 4 weeks, did you notice your child expressing any overtly positive and/or negative feelings about:

15. his/her physical appearance?
    - 0 no
    - 0 yes, positive feelings
    - 0 yes, negative feelings: because of drooling? yes / no

16. his/her social acceptance by adults?
    - 0 no
    - 0 yes, positive feelings
    - 0 yes, negative feelings: because of drooling? yes / no

17. his/her social acceptance by peers?
    - 0 no
    - 0 yes, positive feelings
    - 0 yes, negative feelings: because of drooling? yes / no

Curriculum vitae

Jan van der Burg wordt op 4 april 1964 geboren in Oss en groeit daar gezond en gelukkig op. Na het doorlopen van de kleuterschool St. Janneke en de basisschool Don Bosco, begint hij in 1976 aan het VWO op het Titus Brandsma Lyceum aldaar. Als hij in 1982 het Atheneum-B succesvol heeft afgerond, begint hij aan de studie Pedagogische en Andragogische Wetenschappen (PAW) aan de Katholieke Universiteit Nijmegen en verhuist naar deze stad. In 1987 studeert hij af als orthopedagoog met als specialisatie ontwikkelings- en leerproblemen bij lichamelijk gehandicapten, een keuze die logisch voortkomt uit zijn vrijwilligerswerk bij Stichting Aktie-Fix, een vrijwilligersorganisatie die weekendopvang realiseert voor gehandicapte kinderen en jongeren. Hier houdt hij niet alleen een hechte vriendengroep aan over, maar ontkiemt er ook een liefdevolle relatie met Chrétienne van Lent. Aansluitend aan het afstuderen vervult hij zijn vervangende dienstplicht in het Bio-Revalidatiecentrum voor Kinderen te Arnhem en is werkzaam in een project waarin de revalidatiebehandeling ten aanzien van ademhalingsondersteuning voor kinderen en jongeren met een spierziekte wordt opgezet, en in een wetenschappelijk onderzoeksproject naar de stimulering van de spelontwikkeling van gehandicapte peuters. Na deze leerzame periode gaat hij in 1989 als orthopedagoog bij Centrum Bartiméus te Zeist aan de slag in de ambulante thuisbegeleiding voor blinde en slechtziende baby's, peuters en kleuters. In 1992 trouwt hij met Chrétienne en verhuist terug naar Oss. In 1993 krijgt hij de unieke gelegenheid om in de St. Maartenskliniek te Nijmegen te gaan werken, waar hij sindsdien met veel plezier werkt op de peuterrevalidatie en de kleuterafdeling van de St. Maartenschool. In 1995 wordt Anne geboren, en in 1998 volgt Sjoerd! In 1999 gaat een langgekoesterde wens in vervulling om, naast het klinisch werk, ook wetenschappelijk onderzoek te gaan doen in het UMC St. Radboud (afdeling revalidatiegeneeskunde en IKNC) te Nijmegen. Parallel daaraan krijgt hij in 2002 ook de kans om op de afdeling 'Orthopedagogiek: Leren en Ontwikkeling' van de Radboud Universiteit te Nijmegen, als docent, zijn vak uit te dragen aan orthopedagogen in opleiding.