Interaction, communication, and stress in adults with congenital deafblindness and an intellectual disability

Kitty Bloeming-Wolbrink

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Chapter 1

General introduction

1.1 Background of the research project

The term deafblindness refers to persons with dual sensory loss. The degree of the visual and hearing disability varies within the group of people with deafblindness. The combination of a visual and a hearing disability results in a condition that is more disabling than the sum of the separate disabilities. In case of congenital deafblindness (CDB), the visual and hearing disabilities have an early onset, before the development of language. Persons with CDB have to develop communication and language abilities without their vision and hearing being intact (Dammeyer, 2014).

In this thesis, the focus is on a specific group of persons with deafblindness, namely adults with CDB and an intellectual disability (ID). Deafblindness is not uncommon in persons with an ID. Studies in the Netherlands and Austria report a prevalence ranging from 5% till even 21.4% (Evenhuis, Theunissen, Denkers, Verschuure, & Kemme, 2001; Fellinger, Holzinger, Dirmhirn, & Goldberg, 2009; Meuwese-Jongejeugd et al., 2008). Both persons with congenital and acquired deafblindness are included in these prevalence numbers. Therefore, the exact prevalence of CDB in persons with an ID is unknown.

Adults with CDB and an ID are a group of persons at high risk for not developing to their fullest potential. They often did not receive (continuous) deafblind education, for instance because of an incomplete diagnosis. Many of them live in a residential setting for persons with an ID. There, the main focus is on the ID. In general, communication partners do not have specific knowledge on deafblindness, and thus do not adjust their approach to it. Behavior that could be explained as resulting from deafblindness is often mistakenly seen as a result of the intellectual disability or autism. Living in this situation for years, one can imagine what devastating effects this will have on development. As Hart (2006) stated, the mismatch between the communicative modalities of two partners has a more profound effect on development than the sensory impairment itself. Several authors even mention the risk of an incorrect ID diagnosis in the case of deafblindness (McInnes, 1999; Narayan & Bruce, 2006).

The necessity of a specific approach for persons with CDB and an ID is recognized more and more in the practical field. This has resulted in specific group homes with trained staff in several residential settings in the Netherlands nowadays. The assumption is that offering a specific approach, adjusted to the deafblindness, will have a positive effect on development, even in the case of adults. However, scientific substantiation for this assumption is lacking.

The participants in the research project described in this thesis, are adults with CDB and an ID who live in a residential setting for persons with an ID and a visual disability. The participants have lived in residential settings for persons with an ID for many years, without (continuous) attention for their deafblindness. The main focus has been on their ID. In 2005, Project CHANGE started, including two subsequent interventions to improve the living conditions and social experiences of the participants with the overall aim to improve interaction and communication. With the start of Project CHANGE, the main focus in living conditions and social experiences shifted from the ID to the CDB. Attuning the living conditions and social experiences more and more to the CDB of the participants, was expected to improve interaction and communication.

The participants were followed for 6;6 years. During this period it was examined if the two interventions the participants subsequently experienced in the course of Project CHANGE indeed had the expected positive effects on interaction and communication. In addition, the participants' level of physiological stress, as evident from their diurnal cortisol curves, was examined directly prior to the start of Project CHANGE. This was examined because the participants were assumed to be at high risk for exposure to early and chronic stress during their lives, which could be visible in their diurnal cortisol curves.

In paragraph 1.2 of this introductory chapter, the problems in interaction and communication in case of CDB and an ID are discussed. In paragraph 1.3, the background and value of examining the participants' diurnal cortisol curves is explained. In paragraph 1.4 the outline of this thesis is presented.

1.2 Interaction and communication in persons with CDB and an ID

In this thesis, interaction is defined as the process through which two individuals mutually influence each other's behavior (Janssen, Riksen-Walraven & Van Dijk, 2003b); communication is defined as the process in which information is conveyed to and received and interpreted from others (Rowland, 2009).

Having harmonious interactions is important for the well-being and functioning of persons of all ages (Diamond & Aspinwall, 2003; Sroufe, 1995; Trevarthen & Aitken, 2001) and is essential for a healthy development (Dammeyer, 2010). In the case of CDB, it is challenging to establish harmonious interactions and subsequent communication (Janssen, et al., 2003b; Bruce, 2005; Dalby et al., 2009; Damen, Janssen, Huisman, Ruijssenaars, & Schuengel, 2014; Dammeyer, 2010; Hart, 2010; Martens, Janssen, Ruijssenaars, & Riksen-Walraven, 2014), for several reasons outlined below.

In case of CDB, vision and hearing cannot be used in the same way as in the interaction between two sighted and hearing people. Eye gaze, nodding, facial expression, positive and negative sounds are some examples of the important roles that vision and hearing normally play in social interaction from childhood on (Pérez-Pereira & Conti-Ramsden, 1999). Interactive and communicative signals given by persons with deafblindness are often atypical, hard to interpret and unfold at a slow pace (Daelman, 2003; Janssen, Riksen-Walraven, & Van Dijk, 2003a). Communication partners are global and selective in their observation of interactive and communicative signals; they tend to look at combinations of signals and for signals they expect to occur (Daelman, 2003).

Persons with CDB experience the world mainly through their body, resulting in tactile experiences, which are different from the experiences of sighted-hearing people. Consequently, they will reflect and talk about it in a different way, for instance with bodily expressions like touching a location on the body, making a movement, or replicating a sensation (Daelman, 2003). Expressions made by people who have an ID in addition to CDB are often even more idiosyncratic and subtle, and are not always used consistently. Having an open mind for observing idiosyncratic and bodily-tactile expressions of persons with CDB and an ID is of major importance for their communication partners. Recognizing these expressions and trying to add meaning to them is a valuable contribution to improve interaction and communication.

In this thesis we look at interaction and communication. The effects on these aspects give a good indication about the effectiveness and usefulness of the interventions included in Project CHANGE, since establishing more harmonious interactions and subsequent qualitative communication is believed to have a positive effect on well-being and quality of life.

Several intervention studies have shown that interaction and/or communication with people with CDB can indeed be improved by coaching communication partners (e.g. Damen, Janssen, Ruijssenaars, & Schuengel, 2015; Janssen et al., 2003a; Martens, Janssen, Ruijssenaars, Huisman, & Riksen-Walraven, 2014a; Martens, Janssen, Ruijssenaars, Huisman, & Riksen-Walraven, 2014b). Although some of these studies included participants who have an ID alongside of the CDB, little research has been done so far on this specific group of persons.

1.3 Surplus value of knowledge about the diurnal cortisol curve

The 'stress-hormone' cortisol is produced by the adrenal glands as the end product of the hypothalamic-pituitary-adrenal axis (HPA axis) (Kirschbaum & Hellhammer, 1989). It plays a role in the person's reaction on stressors and it is required for normal physiological regulation. In healthy individuals, cortisol levels follow a circadian rhythm, reflected in the diurnal cortisol curve, with the highest levels in the early morning and the lowest levels around midnight (Lovallo, 2005). In persons with early and chronic exposure to high levels of stress, however, dysregulation of the HPA system can occur, reflected in deviancies in the diurnal cortisol curve, such as elevated cortisol levels, low morning cortisol levels, or a flattened diurnal cortisol curve with low morning levels (Bruce, Fisher, Pears, & Levine, 2009; Carlson & Earls, 1997; Carrion et al., 2002; Fries, Hesse, Hellhammer, & Hellhammer, 2005; Gunnar & Vazquez, 2001; Heim, Ehlert, & Hellhammer, 2000; Loman & Gunnar, 2010; Sánchez et al., 2005). Salivary cortisol can thus be used as a measure of physiological stress (Kirschbaum & Hellhammer, 1989), providing interesting information about the diurnal cortisol curve, and indirectly, about the possible experienced exposure to stress.

Since persons with CDB appear to be vulnerable for experiencing early and chronic stress, we looked at the diurnal cortisol curve in this thesis. There are many factors that may lead to the experience of stress in persons with CDB and an ID. These include the dual sensory disability in itself, early separations from the primary caregivers because of hospital visits, an atypical communication pattern that is difficult to understand (Van Dijk, 1982; Van Dijk & Janssen, 1993), and daily situations with low predictability and low controllability (Dickerson & Kemeny, 2004; Kirschbaum & Hellhammer, 1994). Also, a lack of light is known to affect circadian rhythms, such as the production of melatonin (Arendt, 2000). So measuring the diurnal cortisol curve right before the start of Project CHANGE, before the participants experienced the changes in living conditions and social experiences, could give insight in the possible effects of early and chronic life stress expected to be experienced by adults with CDB and an ID.

Another reason to assess the participants’ diurnal cortisol curve before the start of Project CHANGE was that there is evidence that a deviant diurnal cortisol curve may be normalized by improvements in living conditions, enrichments of environment, and corrective emotional experiences (Gunnar & Quevedo, 2007) such as included in Project CHANGE. Including such an initial cortisol measure opens the possibility for future research to use subsequent cortisol measures to examine if the changes in living conditions and social experiences experienced by the participants in Project CHANGE are also visible in their diurnal cortisol curves.

1.4 Outline of the thesis

This thesis includes three empirical studies, which are described in Chapter 2, 3 and 4. Figure 1 offers an overview of Project CHANGE on a timeline, including the different periods of data collection during the Project, and an overview of the participants included in the different studies.

As shown in Figure 1, Project CHANGE consisted of two main Parts. Part 1 started with the transition (at month 0 on the timeline) of the participants to group homes specifically for persons with deafblindness. This transition implied pervasive changes in living conditions and interactions with specialized caregivers. The 13-month period prior to the transition is included in Part 1 of Project CHANGE as presented in Figure 1, because data collection started at that point in time. Part 2 of the project consisted of a two-phase intervention program aimed at fostering harmonious interactions and the use and recognition of expressions based on a bodily emotional trace (BET). Part 2 of Project CHANGE started at month 56 at the timeline; 4 years and 8 months after the start of the interventions of Part 1.

Chapter 2 describes the first study, which relates to Part 1 of Project Change. In this study, the diurnal cortisol curve of six participants was assessed, directly prior to their transition to the new group homes. Since adults with CDB and an ID seem to be at high risk for exposure to early and chronic stress - e.g. sensory deprivation, communication difficulties, and separations from primary caregivers - information about their diurnal cortisol curve is interesting.

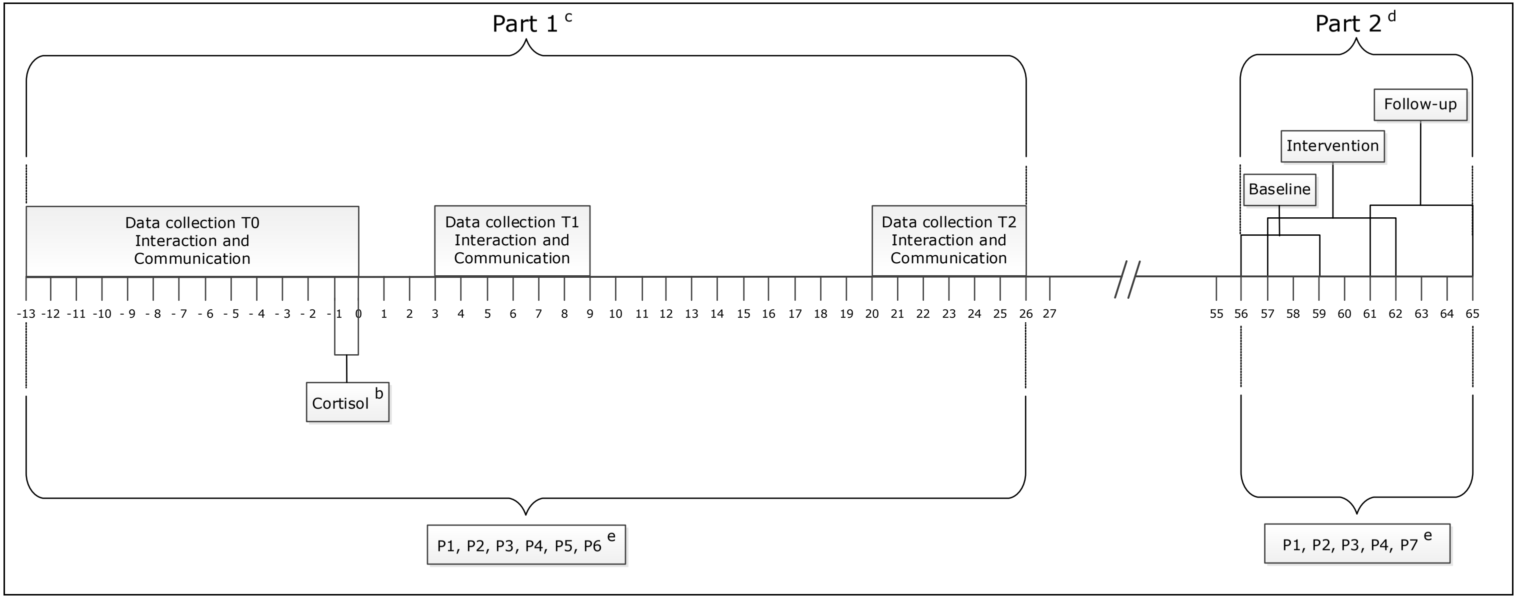

Figure 1. Overview of Project CHANGE on a timeline [a].

[a] The numbers (-13 – 65) below the timeline refer to the months before and after the participants' transition to the new group homes (indicated by 0).

[b] The results are reported in Chapter 2.

[c] The results are reported in Chapter 3.

[d] The results are reported in Chapter 4.

[e] P1-P7 refer to the participants in this study. The numbers do not correspond with the participant numbers in Chapter 4.

Chapter 3 describes the second study, which relates to Part 1 of Project CHANGE. Six participants were included in this study. They experienced a transition to group homes specifically for persons with deafblindness, and interactions with specialized caregivers. In this study, the effects on interaction between the participants and their caregivers, and the effects on the participants' expressive communication skills were examined, using observational data and file information collected at T0, T1, and T2.

Chapter 4 describes the third study, which relates to Part 2 of Project CHANGE. In this study, the effects of a two-phase intervention program, specifically aimed at fostering harmonious interactions (Phase I of the intervention program) and the use and recognition of participant expressions based on a BET (Phase II of the intervention program), were examined. This was done using observational data. Five participants were included in the study. Four of them also took part in the earlier studies (Chapter 2 and 3).

Chapter 5, the final chapter of this thesis, comprises a summary, main conclusions, and a general discussion of the main findings of the three empirical studies. The strengths and limitations of the research project are discussed, as well as recommendations for researchers and the practical field that emerged from the main findings.

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Chapter 2

Stress in adults with congenital deafblindness and an intellectual disability: Information from their cortisol curves

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Abstract

Adults with congenital deafblindness and an intellectual disability are at high risk for exposure to chronic and severe stress. Sensory deprivation, separations from primary caregivers, and difficulties in communication, from birth on, make daily life stressful for them. Chronic and high stress can lead to deviancies in the cortisol curve. This, in turn, can have negative effects on mental and physical health. Nothing is known yet about the cortisol curve of adults with congenital deafblindness and an intellectual disability, but our hypothesis is that it will be deviant.

In this study, the cortisol curve of six participants with congenital deafblindness was compared to the curve of 40 typical controls. Remarkably, in spite of all past and present stress factors, the cortisol curve of the participants was rather normal. Possible explanations for this finding are discussed, amongst others individual differences in experiencing stress. Implications for further research are discussed.

2.1 Introduction

Individuals with congenital deafblindness (CDB) (dual sensory loss from birth) have a high prevalence of mental and behavioural disorders such as mental retardation, obsessive compulsive disorders, mood disorders, behavioural disorders, anxiety, psychosis and pervasive developmental disorders (Dammeyer, 2011; Hoevenaars-van den Boom, Antonissen, Knoors & Vervloed, 2009; Luiselli, 1992; Van Dijk, 1982). In the group with acquired (post-lingual) deafblindness (ADB), individuals reported that the dual sensory loss negatively impacted their mental well-being which leads to increased anxiety, stress, depression and loss of self-esteem (Bodsworth, Clare, Simblett & Deafblind UK, 2011; Capella-McDonnall, 2005; McDonnell, 2009).

Adults with CDB and an intellectual disability (ID), the target group of this study, face many challenges that may cause severe and chronic stress. Aside from their sensory deprivation, many of these adults have a history of social deprivation resulting from severe communication difficulties and stressful life experiences. Many of these people did not receive any deafblind-specific education or communication support adapted to their needs.

Although stress is a normal part of life, toxic stress can have very serious consequences. Toxic stress is defined as strong, frequent or prolonged activation of the body’s stress management system and it is provoked by stressful events that are chronic, uncontrollable and/or experienced without the person having access to support from caring persons (National Scientific Council on the Developing Child, 2009). Early in development, toxic stress can impair emotional well-being, exploration and curiosity, school achievement and other qualities (Nelson, Greenfield, Hyte, Shaffer, & Paul, 2011). Toxic stress is also known to lead to an increased risk of many conditions, like heart disease, diabetes, depression and anxiety disorders (National Scientific Council on the Developing Child, 2009).

Salivary cortisol can be used as a biological indicator of stress (Hellhammer, Wüst, & Kudielka, 2009). Cortisol is a glucocorticoid hormone that is produced by the adrenal glands as the end product of the hypothalamic-pituitary-adrenal axis (HPA axis). Cortisol is released in response to various biochemical agents and psychosocial stimuli, such as a challenging situation (Kirschbaum & Hellhammer, 1989). It plays a role in a person's reaction to stressors and it is required for normal physiological regulation. In healthy individuals, cortisol levels show a circadian rhythm called the diurnal cortisol curve; levels peak in the early morning hours and are at their lowest around midnight (Lovallo, 2005).

The rhythmic pattern of cortisol secretion, resulting in the diurnal cortisol curve, is important for maintaining normal organ regulation and adaptive functioning. Impairment of this rhythm has been associated with poor long-term health (Kirschbaum & Hellhammer, 1989; Lovallo, 2005). In the short term, activations of the HPA axis tend to support adaptive functioning. However, there is substantial evidence to suggest that early and chronic exposure to high levels of stress leads to dysregulation of the HPA system (Fries, Hesse, Hellhammer, & Hellhammer, 2005; Gunnar & Vazquez, 2001) and increases the risk of physical and behavioural problems (Gunnar & Quevedo, 2007). Elevated cortisol levels have been found, for example, in emotionally maltreated children (Bruce, Fisher, Pears, & Levine, 2009) and in children who have a history of exposure to trauma and show symptoms of post-traumatic stress disorder (PTSD) (Carrion et al., 2002). Chronic stress and prolonged hyperactivity of the HPA system are believed to eventually result in down-regulation of the system or hypocortisolism, characterized by blunted cortisol responses to stressors and a flattened diurnal cortisol curve with low morning values (Fries et al., 2005; Heim, Ehlert, & Hellhammer, 2000; Loman & Gunnar, 2010). Low morning cortisol levels have been observed, for instance, in nonhuman primates exposed to repeated maternal separations (Sánchez et al., 2005), in children raised in neglectful institutions (Carlson & Earls, 1997) and in foster children, especially those with a history of neglect (Bruce et al., 2009).

Although adverse circumstances are likely to have a negative effect on cortisol levels, there are individual differences. Some of the variability in cortisol levels across individuals seems to be due to genetic differences (Clements, 2012; Nicolson, 2007) and coping styles (Nicolson, 2007). Nelson et al. (2011) also mention temperamental characteristics as a protective factor against toxic stress. Individuals perceive and evaluate situations differently and this perception, combined with the resources available to control the situation, determines how stressful an individual finds a situation (Lovallo, 2005).

The aim of the present study is to examine the diurnal cortisol curve in adults with CDB and an ID. No prior research results have been published about the cortisol curve in people with deafblindness, though other groups have been studied. For instance, the diurnal cortisol variation was smaller for blind individuals than for control subjects (Sack, Lewy, Blood, Keith, & Nakagawa, 1992). Sterkenburg, Janssen, Blankenstein, and Schuengel (2008) found a relatively flat cortisol curve in adolescents with an intellectual and visual disability who showed challenging behaviour; their cortisol levels were also lower in the morning and higher in the evening compared to a normative sample. In a group of children with severe and profound intellectual disabilities, Terstegen et al. (2003) found rather normal cortisol variation but one-third of the participants lacked a circadian rhythm. In sum, studies among persons with disabilities have shown some deviations of the diurnal cortisol curve as compared to control groups, but the findings do not show a distinct, constant pattern.

There is reason to believe that the diurnal cortisol curve of adults with CDB and an ID will differ from that of individuals without disabilities. First, individuals with deafblindness experience a lack of light and sound. Lack of light is known to affect circadian rhythms, for example, production of melatonin (Arendt, 2000). Second, because of the absence of well-functioning vision and hearing, daily life may be less predictable and controllable. This is known to lead to a rise in cortisol levels (Dickerson & Kemeny, 2004; Kirschbaum & Hellhammer, 1994). Third, many adults with

CDB and an ID have a history of social deprivation as a result of, amongst other things, an atypical communication pattern and the experience of stressful events such as early separations from their primary caregivers because of hospital visits or entering an institution (Van Dijk, 1982; Van Dijk & Janssen, 1993). The exposure to such stressors may be expected to further contribute to deviancies in the diurnal cortisol curve in these individuals.

The present study was aimed at determining the diurnal cortisol curve of six adults with CDB and an ID, and comparing them to a control group consisting of 40 typical adults. First, we hypothesized that the cortisol curve of the participants with CDB and an ID would be lower and flatter than the curve of the control group. Second, we expected to gain more insights into individual variations between the curves of the participants and into the stress physiology of this unique population in general.

2.2 Materials and methods

2.2.1 Setting and participants[2]

The assessments were conducted at a setting for individuals with intellectual and sensory disabilities in the Netherlands. The study followed the tenets of the World Medical Association Declaration of Helsinki on Ethical Principles for Medical Research Involving Human Subjects, and was approved by the board of the setting. Written consent was obtained from the participants' legal representatives.

Initially, seven adults with CDB and an ID (four males, three females) participated in this study. One of them, a woman, rejected the saliva sampling; as a result, too little saliva was collected to analyse her samples, and she was excluded from the study. During the saliva collection, the average age of the six remaining participants was 42 years (ranging from 37 to 48 years). They had been living at the setting for 27 to 38 years. At

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[2] Some minor textual changes were made as to the original publication, for sake of protecting the participants' identity.

the time of the study, they lived in group homes together with three to six others, with one or two staff.

The degree of sensory disabilities differed. Table 1 gives an overview of the relevant sample characteristics. The information was collected from the participants' files. As a child, all participants experienced life events related to maternal separations and housing changes. In the participants' files, two to five maternal separations during the first five years of life are mentioned, mainly for hospital visits. All participants were living in an institution before the age of 11.

Table 1

Participant characteristics

Participant name (fictitious)	Age	Diagnosis	Vision[a] in childhood	Present vision	Hearing[b] in childhood (age[c])	Present hearing
Tina	37	Unknown	Congenital buphthalmus; eyes removed at age 3	Totally blind	Moderate hearing loss (age 4)	Moderate/severe hearing loss
George	48	Congenital Rubella	Blind	Blind	Hearing loss (age 6)	Profound hearing loss
Romy	48	Unknown	Bilateral cataract	Blind	Hearing loss	Severe hearing loss
Charles	40	Aumorosis congenital von Leber	Blind	Blind	Unclear	Profound hearing loss
Harold	37	Congenital Rubella	Bilateral cataract	Totally blind	No reaction to sounds (age 3 months)	Hearing loss; unclear whether there is residual hearing
Richard	39	Congenital Rubella	Bilateral cataract	Blind	Hearing loss (age 3½)	Profound hearing loss

[a] Totally blind means blind without light perception; blind means blind with light perception.

[b] Moderate hearing loss: 41-60dB; Severe hearing loss: 61-80dB; Profound hearing loss: ≥81dB.

[c] Information about hearing was first found in the file for this age.

2.2.2 Cortisol assessment in saliva

The use of saliva to determine cortisol levels has some important advantages: it is non-invasive, it is not painful and the samples can be collected at the participants' home (Clements, 2012). Repeated measurement in the daily environment can provide a good estimate of the participants' basal levels and diurnal variation (Nicolson, 2007).

Saliva was collected using a sterile cotton roll, which was swabbed along the person's inner cheek and under the tongue to absorb saliva. It was stored in a Salivette in a refrigerator. Within one week, the samples were transported to a freezer, in which they were stored frozen at -20°C until analysis. Salivary cortisol was measured by radioimmunoassay (RIA) after extraction with dichloromethane and subsequent paper chromatography (Meulenberg, Ross, Swinkels, & Benraad, 1987). The samples were analysed at the Department of Laboratory Medicine, Radboud University Nijmegen Medical Centre, in the Netherlands.

Saliva samples from the participants with CDB and an ID were collected on seven days over a four-week period. On each day, five samples were collected at the following moments: just before getting up (mean (M) 7.56 h), before breakfast (M 8.48 h), before lunch (M 12.03 h), before dinner (M 16.52 h) and before going to bed (M 21.28 h). The saliva samples were collected by those caregivers who usually worked with the participants. The caregivers were asked to refrain from giving coffee, cola or orange juice to the participants within half an hour before the sample was collected. Caregivers also refrained from brushing the participants' teeth within 30 minutes before sampling.

2.2.3 Control group

The control group was created by combining two control groups from a study of Eijsbouts et al. (2008), both consisting of 20 typical adults, into one new control group of 40 adults. Inclusion criteria were self-reported health, age between 18 and 47 years, normal physical examination and normal electrocardiogram. Exclusion criteria were pregnancy, an abnormal medical history or physical examination, medication use, endocrine diseases and psychiatric diseases. This control group was chosen because their samples were analysed with the same method and at the same laboratory as the study group's samples. The saliva samples were collected from the control group at 4.00 h, 8.00 h, 12.00 h, 16.00 h, 20.00 h and 24.00 h. The 4.00 h samples are not included in the results.

2.2.4 Statistics

For each of the participants with CDB and an ID who had sampling moments with at least five values, outliers were calculated with the formulas Q1 - 1,5 × IQR and Q3 + 1,5 × IQR (Moore, McCabe, & Craig, 2009). Outliers could be calculated for 16 out of the 30 sampling moments (53%) and four outliers were excluded from analysis. The mean diurnal cortisol curve was then calculated for the six participants as a group and for each of the six participants individually, and compared to the curve of the control group. Using the caregivers' notes, the mean time of sampling was calculated for every sampling moment and rounded up to a whole hour.

The standard deviation was calculated for each sampling moment. For the participants with CDB and an ID the individuals' mean cortisol levels were used in this calculation.

2.3 Results

Thirty-five saliva samples were collected from each of the six participants with CDB and an ID. The number of useful saliva samples (> 0.05 ml) per participant varied from 12 to 34 (34% to 97%). Table 2 gives an overview of the number of useful samples for each participant at the five sampling moments (range one to seven). In total, 65 percent of all saliva samples could be analysed. The high percentage of missing samples can be explained in large part by the fact that the sampling process was not sufficiently adapted to the specific target group.

Table 2

Number of successful saliva samples per participant

Sample	Before getting up	Before breakfast	Before lunch	Before dinner	Before going to bed	Total
Tina	3	4	5	2	4	18
George	3	7	5	6	6	27
Romy	2	7	6	6	5	26
Charles	7	6	7	7	7	34
Harold	2	3	4	6	5	20
Richard	1	2	4	2	3	12

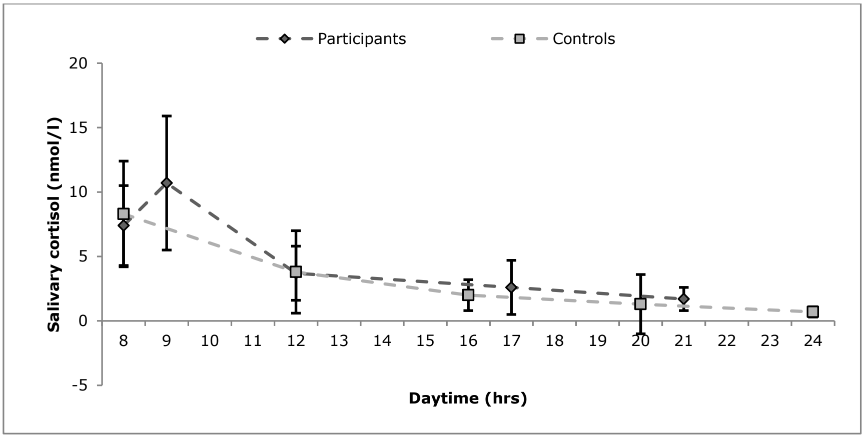

Figure 1. Cortisol curve of the participants compared to the cortisol curve of the controls.

The participants’ mean diurnal cortisol curve largely resembles that of the control group, except for 9.00 h (as presented in Figure 1). At this point, a peak can be seen in the participants’ cortisol curve, but not in that of the control group because no sample was taken from them at that time. Inter-individual variation can be seen in the individual participants’ curves (as presented in Figure 2). The curves of George, Charles, Harold and Richard show a peak at the second collection moment; Romy’s cortisol curve lies above the control group’s cortisol curve and Richard’s cortisol curve lies below the control group’s curve. Both these curves appear somewhat flattened.

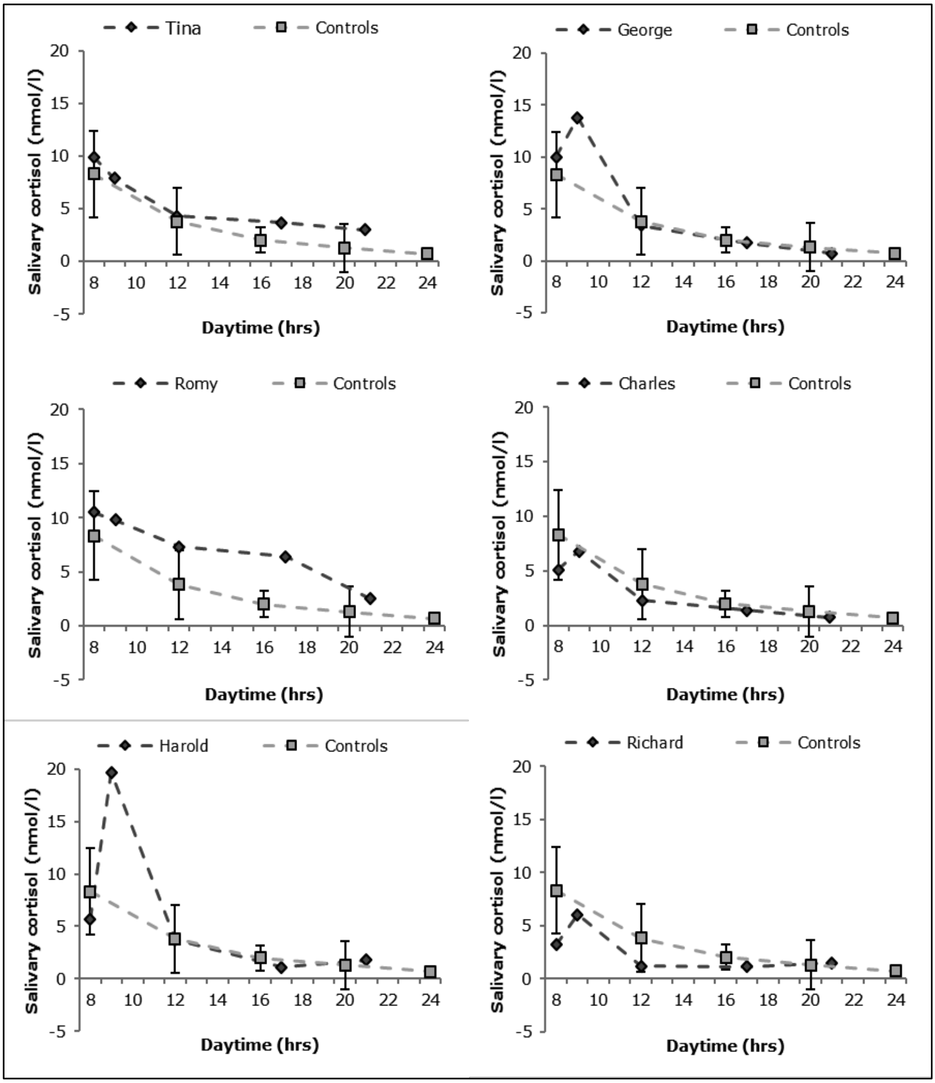

Figure 2. Individual cortisol curves of the six participants compared to the cortisol curve of the controls.

2.4 Discussion

This study compared the diurnal cortisol curves of six adults with CDB and an ID with the curve of a control group of 40 typical adults. The participants with CDB and an ID have been, and still are, at great risk for experiencing toxic stress. Our hypothesis was that the cortisol curve of these participants would be lower and flatter than the cortisol curve of the controls. Remarkably, the mean cortisol curve of the participants with CDB and an ID was rather normal, in contradiction with earlier findings showing that stress in early life often leads to changes in the diurnal cortisol curve (Heim et al., 2000; Heim, Newport, Mletzko, Miller, & Nemeroff, 2008). The fact that despite the presence of deprivation and disability, as a group the participants with CDB and an ID have a normal diurnal cortisol curve is nonetheless a very positive finding that speaks to their resilience.

The results show a substantial but normal individual variation among the six participants. Richard’s and Romy’s curves lie respectively below and above the curve of the control group – this is the case for part of their curves even when considering the SD’s of the control group – and appear slightly flattened, in line with the findings of Sack et al. (1992) and Sterkenburg et al. (2008) by persons with a visual disability. All participants have experienced potentially stressful maternal separations and went to live in an institution before the age of 11. However, we do not know the impact of these life events in terms of stressfulness for each participant. According to Lovallo (2005), the stressfulness of a specific event is determined by the individual’s appraisal of the event and by the resources available to control it. An individual can also have a genetic vulnerability to stress (Nicolson, 2007). Differences in the stress that participants experienced, in combination with differences in the developmental course of their disabilities, may very well cause the individual variation in cortisol curves. This may also be influenced by temperamental characteristics (Nelson et al., 2011). The participants also may have developed coping mechanisms to control stress, such as stereotypical behaviour like rocking. Moreover, the quality of past and present caregiving, the access to support from caring persons and individuals' well-being are factors that probably also influence their diurnal cortisol curve. Unfortunately, the difficulties in communicating with the participants in the present study make it only possible to guess at the underlying reasons for the individual differences found.

The cortisol levels of our participants and the control group were assessed using the exact same method in the same laboratory, so differences in the results are not due to different methods or facilities. However, a limitation of the present study is that the participants' saliva samples were not collected at exactly the same moments as the control group's samples. This may well explain the presence of a distinct peak at 9.00 h in the cortisol curve of the group with CDB and an ID and the absence of a peak in the morning in the control group's curve. Normally, there is a rapid increase in cortisol levels after awakening, the cortisol awakening response (CAR). This physiological response produces a peak in cortisol at around 30 minutes post-awakening (Wilhelm, Born, Kudielka, Schlotz, & Wüst, 2007). At least two samples in the morning are needed to see this peak in the diurnal cortisol curve. The peak is missing in the curve of the control group, because only one sample was taken in the morning. Two saliva samples were taken in the morning from the participants with CDB and an ID: just before getting up and before breakfast. The peak in their mean cortisol curve at 09.00 h may be due to this sample coinciding with their CAR. The peak is missing for Tina and Romy (see Figure 2), possibly because they already had been lying awake in their beds. In such a case, the sampling would be too late to capture the CAR. One participant, Harold, had a high peak at the second collection moment. An explanation, apart from the CAR, may be that he is rather obsessive about food. The second collection moment is prior to breakfast and was therefore possibly a stressful moment for him.

The number of participants in the study was relatively small, a problem inherent to the low incidence and complexity of the target group. Collecting enough saliva from the participants for scientific research was a challenge, as no more than 137 of the 210 intended samples could be collected. This is in line with an earlier observation made by Sterkenburg et al. (2008). The following recommendations can be made for collecting saliva from participants with an ID and additional disabilities, both in future studies and for medical reasons: a) give extensive instructions about the saliva sampling to caregivers (e.g. an instruction movie, a team meeting); b) take enough time for the collection; c) communicate with the participants in a way that is adapted to their ability to understand what is going to happen so that they will not be frightened (e.g. let them touch a cotton roll before collecting the saliva); and d) make adjustments to the sampling procedure (e.g. collect in two phases with a pause in between).

Despite the limitations, the present study offers a good base for future studies to elaborate on. The results give an interesting insight into the stress physiology of adults with CDB and an ID. It remains partly unclear how exposure to many risk factors for stress results in a normal cortisol curve. Factors like genetic vulnerability, coping mechanisms and temperament seem to play a role. It is also possible that what seems stressful for most people (e.g. a lack of sensory information) might not be experienced as stressful for someone born without vision and hearing. To a person who is born deafblind, a world without good information from vision and hearing is normal instead of deviant. This information is not perceived as lacking because it is not part of their reality at all. One might also adapt to living in unpredictable, uncontrollable and often novel situations from birth. The fact that the participants are all adults and have had time to adjust to their circumstances may also partially explain the positive results. In most of the studies that found deviant cortisol curves, the participants were children. The role of the ID on the results needs to be further clarified.

The results of this study are positive. They show that the presence of unfavourable circumstances do not have to lead to deviances in the cortisol curve. This speaks for the resilience of the participants in this study, but also raises questions about the underlying reasons for the individual differences. Although as a group the participants with CDB and an ID had a normal cortisol curve, two of them did have deviancies in the cortisol curve.

Deviancies in the individual cortisol curves of the participants with CDB and an ID may be caused by a combination of factors. Some of these factors cannot be influenced but other factors, aimed at increasing their current well-being, can be influenced by, for example, decreasing the number of unpredictable and uncontrollable events in daily life, or training caregivers to be sensitive and responsive partners during daily interactions. This underscores the need of a specific approach for the target group.

The present study has shown that life events alone are not reliable predictors of cortisol levels. Future research is needed to discover whether factors such as adaptation to life events are better predictors.

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Chapter 3

Effects of changes in life circumstances on interaction and communication in adults with congenital deafblindness and an intellectual disability

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Abstract

This study examined the effects of the first 2 years of the ongoing Project CHANGE, aimed at improving interaction and communication in adults with congenital deafblindness (CDB) and an intellectual disability (ID). Six adults with no history of deafblind education participated in this study. These first 2 years of CHANGE involved an improvement in living conditions, including a transition to a new group home and interactions with specialized caregivers. Video recordings and file information were used to measure interaction and communication during baseline (T0) and two subsequent periods (T1 and T2). The results show an improvement in interaction: attention by the caregiver, confirmation by the caregiver, and affective involvement all improved. An improvement was also shown in the participants' level of expressive communication and in the variety of their communicative behaviors. The improvements over time emphasize the need for a specific approach for persons with CDB and an ID.

3.1 Introduction

Frequent and affectively pleasant interaction with others is essential for satisfying the fundamental and pervasive human need to belong (Baumeister & Leary, 1995), for quality of life (Prain, McVilly, & Ramcharan, 2012b), and for development (Dammeyer, 2010). Empirical research has shown that such interaction fosters well-being and improves the functioning of persons of all ages (Diamond & Aspinwall, 2003; Sroufe, 1995; Trevarthen & Aitken, 2001). However, establishing harmonious social interactions and subsequent communication is challenging for persons with congenital deafblindness (CDB) and their communication partners (Bruce, 2005; Dalby et al., 2009; Damen, Janssen, Huisman, Ruijssenaars, & Schuengel, 2014; Dammeyer, 2010; Hart, 2010; Martens, Janssen, Ruijssenaars, & Riksen-Walraven, 2014). Both the persons with CDB and the communication partners lack skills for establishing interpersonal contact and for exchanging thoughts and emotions (Martens et al., 2014). The interactive and communicative signals given by persons with deafblindness are often hard to interpret and unfold at a slow pace (Janssen, Riksen-Walraven, & Van Dijk, 2003a). Communication is complicated further if someone has both a sensory and an intellectual disability (ID) (Evenhuis, Sjoukes, Koot, & Kooijman, 2009).

Here, interaction is defined as the process in which two individuals mutually influence each other's behavior (Bjerkan, 1996; Janssen, Riksen-Walraven, & Van Dijk, 2003b). Communication is defined as a more complex form of interaction, with meaning being transmitted by expressions that are perceived and interpreted by the communication partner (Janssen et al., 2003b). Information is conveyed to others, and information from others is received and interpreted (Rowland, 2009). For persons with CDB and an ID, specialized knowledge and training of communication partners is needed to realize harmonious interactions and communication.

In the Netherlands, many adults with CDB live in group homes for persons with an ID. Adults with CDB living in settings for persons with an

ID often have little experience with harmonious interactions. Often, staff has no special knowledge of sensory impairments.

Persons with deafblindness need highly specialized care and rehabilitation to stimulate communication and development. A clearly structured environment, individual programs to enhance communication, and predictability of activities of daily life are some recommendations from the literature (Evenhuis, Theunissen, Denkers, Verschuure, & Kemme, 2001; Fellinger, Holzinger, Dirmhirn, & Goldberg, 2009). In a world that emphasizes sight and hearing, persons with CDB miss out on incidental learning opportunities (Bruce, 2002; Bruce, Godbold, & Naponelli-Gold, 2004; McInnes, 1999). When no special attention is given to the deafblindness, this may result in, for example, a limited development and deviant behavior. Several authors even mention the risk of an incorrect ID diagnosis in the case of deafblindness (McInnes, 1999; Narayan & Bruce, 2006).

As earlier studies have shown, interaction with children with deafblindness or adults with intellectual and visual disabilities can be improved by training their communication partners (e.g. Damen, Kef, Worm, Janssen & Schuengel, 2011; Janssen et al., 2003a). The aim of this study was to examine whether an improvement in living conditions, including interactions with specialized and trained communication partners, can improve interaction and communication in adults with CDB and an ID who have never received deafblind education.

The participants in this study underwent a pervasive change in living conditions and social experiences, which we labeled “Project CHANGE”. Here, the effects of the first 2 years of the project were examined. The 2-year period started with the participants moving from group homes for persons with a visual disability and an ID to a group home for persons with CDB and an ID, in which communication partners (caregivers) were specially trained. The relevant changes are described in more detail in section “Methods”. The expectation was that the changes would lead to increased opportunities for experiencing harmonious interactions and that this would bring about an improvement in the interaction and communication between the participants and their communication partners.

This study is the first to examine the effects of a pervasive change in both living conditions and social experiences in adults with CDB and an ID on interaction and communication.

3.2 Methods

3.2.1 Participants and setting

The study was conducted at a setting for persons with intellectual and sensory disabilities in the Netherlands. Six adults with CDB and an ID participated in the study: four males and two females. Their average starting age was 42 years, ranging from 37 to 48 years of age. The degree of intellectual and sensory disability, and the interaction and communication strategies used, varied. Table 1 provides an overview of relevant participant characteristics. None of the participants received deafblind education before or was educated with a consistent approach geared at persons with CDB.

The study was approved by the board of the residential setting. Informed consent was obtained from the participants' legal representatives. To protect the participants' identities, fictitious names are used in this article.

3.2.2 Project CHANGE

Project CHANGE began with the transition of the participants from a group home with four to seven residents with a visual disability and an ID to a twin house with eight residents with CDB and an ID, divided into two groups of four residents. During most of the day, the staff-client ratio was 1:2. At the start of the project, the staff comprised 15 caregivers, two educational psychologists, and a coordinator. One participant (Harold) knew none of the caregivers at the new setting. Four participants (Richard, George, Tina, and Romy) were acquainted with one of the caregivers. The sixth participant (Charles) knew two of the caregivers. Both the caregivers and the coordinator interacted directly with the participants. The daily care and

Table 1

Participant characteristics at the start of the study

Participant	Age (years)	SRZ score[a]	Interaction and communication[b]	Visual disability[c]	Hearing disability[d]
Richard	39	4	Interaction categories attention, confirmation, affective involvement; objects of reference; tactile signs (after 9-month extension of number of tactile signs)	Blind	Profound hearing loss
George	48	4	Interaction categories attention, confirmation, affective involvement; objects of reference; tactile signs (after 9-month extension of number of tactile signs)	Blind	Profound hearing loss
Tina	37	4	Interaction categories attention, confirmation, affective involvement; objects of reference	Totally blind	Moderate/severe hearing loss
Harold	37	4	Interaction categories attention, confirmation, affective involvement; objects of reference; tactile signs (after 9-month extension of number of tactile signs)	Totally blind	Hearing loss; unclear whether there is residual hearing
Romy	48	4	Interaction categories attention, confirmation, affective involvement	Blind	Severe hearing loss
Charles	40	3	Interaction categories attention, confirmation, affective involvement; objects of reference (after a year)	Blind	Profound hearing loss

[a] A social competence SRZ score of 3 or 4 is appropriate in case of a profound intellectual disability (Kraijer, Kema, & De Bildt, 2004).

[b] Interaction and communication from the moment of the transition to the new group home.

[c] Totally blind: no light perception; blind: blind with light perception.

[d] Moderate hearing loss: 41-60dB; severe hearing loss: 61-80dB; profound hearing loss: ≥81dB.

activities were offered by the same caregivers. The caregivers, both before and after the transition of the participants, differed with regard to their age, educational background, and the years of working experience. All of them were qualified for working in care settings and had some experience in working with persons with an ID and a visual disability.

Prior to the participants' move to the new group home, all staff were trained to adjust their approach to the specific needs of the participants. They followed a 4-day training course on interaction and communication, consisting of, amongst others, a brief introduction on the basic principles of interaction with persons with CDB, information on normal language development and language development in persons with CDB, individual communication plans, and on bodily contact. This training was provided by two specialists from a center of expertise on deafblindness, one of them being the second author. Information about the exact content of the training is available from the first author. In addition, the educational psychologists and the coordinator followed a 2-day training in video analysis, preparing them for on-the-job coaching of the caregivers. Ten months after the start of the project, they had a follow-up training. Nine months after the move, all staff received a half-day of instruction on tactile sign language.

Starting after the transition and continuing for 2.2 years, caregivers were coached on-the-job individually by the coordinator, and caregivers and the coordinator were coached individually using video analysis by the educational psychologists. Both forms of individual coaching were offered multiple times a week. The number of individual coaching sessions caregivers had per month was dependent on their working schedule. Once a month, video analysis was done with the whole team. A video was reviewed by the team, reflections were made, and new ways of doing things were suggested. The main focus in all coaching was on developing the interaction skills of the caregivers and on recognizing the expressions of the participants.

3.2.3 General procedure

Interaction and communication were measured using video recordings and file information during three periods: T0 was the 13-month baseline period before the transition. T1 and T2 were 6-month periods starting 3 and 20 months after the move, respectively. Figure 1 gives a timeline of the different events.

3.2.4 Measures

3.2.4.1 Interaction.

Video recordings. Video recordings were regularly made to gain insight into the daily life of the participants. The number and duration of the recordings were among others dependent on the variety of activities, also including interactive and communicative situations. To examine the effects of Project CHANGE on interaction, 5-min video fragments were selected for T0, T1, and T2. For the sake of comparability within and between participants, we selected video fragments recorded in the same situation, that is, meals. Meals were chosen because they were fixed moments in the daily schedule for all participants, during which there had to be contact between participant and caregiver. Before and shortly after the transition of the participants, contacts between participants and caregivers were often rare in other situations, due to, for example, lack of time and many practical tasks for the caregivers during this period.

Because of this choice for the meal situation, at T2 a fragment could be selected for only three out of six participants. For Harold, we had to use a fragment for T1 that fell shortly outside the T1 period. Because no major interventions have been done since T1, the expectation is that this will not influence the outcomes. It should also be noted that it was not per definition with the same caregiver with whom the participants were observed during the meals at T0, T1, and T2. For that reason, we could not examine the role of individual caregiver characteristics in the change of participant-caregiver interaction over time.

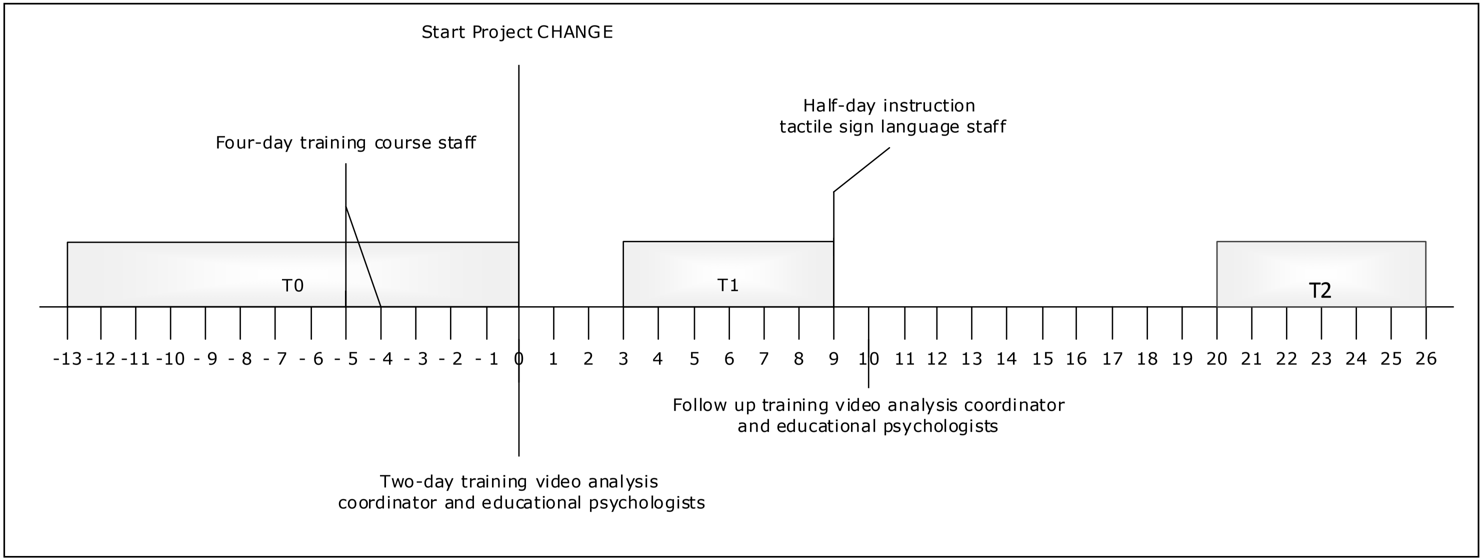

Figure 1. Timeline.

Interaction categories. The following four categories of interactive behavior (adopted from Janssen et al., 2003b) were used to measure the quality of interactions:

1a. Attention by the caregiver: focus on the interaction partner, content of the interaction, people, or objects within the interaction context, for example, looking in the direction of the participant.

1b. Attention by the participant: see definition 1a, for example, establishing physical contact with the caregiver.

2. Confirmation by the caregiver: clear acknowledgement that an initiative has been noticed and recognized, for example, by repeating the initiative of the participant in a way the participant can perceive.

3. Affective involvement: mutual sharing of emotions between caregiver and participant, for example, laughing together, in a way the participant can perceive.

Observation procedure and scoring. Each video was coded independently by two observers on a specially designed observation form. Samples of 10-sec intervals were used. Per interval, attention by the participant, confirmation by the caregiver, and affective involvement were coded 1 if observed at least once and coded 0 if not observed. Attention by the caregiver was coded 1 if observed during the main part of the interval and otherwise coded 0. For each category, an interval was excluded from coding if the information needed was lacking (e.g. if the caregiver was not visible, attention could not be observed). An overall score, reflecting the occurrence of a certain behavior in the 5-min episode, was calculated as the percentage of coded 10-sec intervals during which that behavior was observed.

Three trained observers, bachelor's students in special needs education, coded the videos. To control for observer drift, the videos were presented at random to the observers. Definitions of the categories were read prior to each observation session.

Inter-rater reliability. Prior to the formal data collection, the observers were trained with the intention of reaching 80% inter-rater reliability for the occurrence of the interactive behaviors for three training videos per participant. The training videos were not used for the actual coding.

Following Prain, McVilly, and Ramcharan (2012a), inter-rater reliability for the actual research recordings was calculated in two ways. The percentage agreement across measurements and participants was calculated by dividing the number of agreements on occurrences and non-occurrences by the total number of agreements and disagreements, and multiplying the resultant number by 100. The results were good: 99.1% for confirmation by caregiver and affective involvement, 94.7% for attention by caregiver, and 91.0% for attention by participant. The Kappa inter-rater reliability coefficient was also good: .85 for affective involvement, .85 for attention by caregiver, .83 for confirmation by caregiver, and .81 for attention by participant.

3.2.4.2 Communication.

Communication Matrix. To assess the expressive communication skills of the participants, an adapted version of the Communication Matrix of Rowland (2004) was used. Table 2 presents the protocol for this adapted version.

The Communication Matrix is suitable for assessing adults, even if they are deafblind, and can be completed using observation (Rowland, 2009). The Communication Matrix distinguishes four communicative functions (*refuse*, *obtain*, *social interactions*, and *provide or seek information*) and seven levels of communication for each of the functions, ranging from *pre-intentional behavior* (0-3 months in the normal population) to *language* (24+ months) (Rowland, 2011; Rowland & Fried-Oken, 2010). Each separate function can be expressed by someone on more than one level of communication. The method yields a differentiated profile of the person assessed, representing both the level and the variety of expressive communication skills for all four communicative functions.

In addition to the level and function of the various communication skills, the matrix differentiates between emerging and mastered communicative skills, as testified by, among others, the variety of contexts in which a skill is witnessed (Rowland & Fried-Oken, 2010). This differentiation was not included in the adapted version used here.

Observation procedure and scoring. The Communication Matrix was completed retrospectively following the protocol presented in Table 2. It was completed separately for T0, T1, and T2, on the basis of videos and file information on communication. The videos contained a variety of situations which included communication, like meals and day activities. File information consisted of (1) a profile of communication, containing information about the participants’ communication toward the caregivers, as well as the caregivers’ communication toward the participants; (2) an Individual Support Plan with relevant information about the participant, like vision, hearing, ID, additional disabilities such as epilepsy, and aims for support; (3) a multidisciplinary report, containing the most important information from the previous year; and (4) information from observation scales, like the Social Competence Scale (SRZ) (Kraijer et al., 2004) (included in Table 1). The communication skills observed at T0 and T1 were included in the scores for subsequent periods, on the assumption (from clinical experience) that expressive communication skills are stable over time.

For each participant, both the level and variety of expressive communication skills were assessed. The highest level of communication was represented in scores for T0, T1, and T2 and for the increase from T0 to T2. One score was assigned for each separate communicative function (*refuse*, *obtain*, *social interactions*, and *provide or seek information*).

The Communication Matrix distinguishes 80 communication skills across levels and functions. The variety of communication skills was calculated by assigning 1 point per communication skill, with a maximum of 80 points. The percent equivalent was calculated by dividing the total number of points by the maximum number of points (80), and multiplying the resultant number by 100. Improvement in variety of communication from T0 to T2 was expressed as a percentage gain score, which was calculated by dividing T2 - T0 by the T0 score, multiplied by 100 (Rowland & Schweigert, 2005).

Table 2

Protocol for completing the Communication Matrix retrospectively (Bloeming-Wolbrink, Van den Bovenkamp, & Janssen, 2010)

Step	Description
1	Get familiar with the Communication Matrix. This includes the definitions of the four communicative functions, the seven levels of communication, and the communication skills.
2	Gather all files and video recordings for a specific research period.
3	Examine all files kept by the setting for information on communication skills. If a communication skill is identified, allocate it to a function (refuse, obtain, social interactions, or provide or seek information) and a level. Fill in the Communication Matrix, and note from which part of the files the information was extracted.
4	Examine the video recordings and identify communication skills of the participant. If a communication skill is identified, allocate it to a function (refuse, obtain, social interactions, or provide or seek information) and a level. Fill in the Communication Matrix. Note all necessary characteristics to locate the specific fragment in which the communication skill was displayed.
5	Repeat steps 2, 3 and 4 for each research period so that matrices for all research periods are completed.

Inter-rater reliability. A master’s student in special needs education completed the Communication Matrices after receiving instructions from the first author, who also corrected the matrices.

3.3 Results

3.3.1 Interaction

Table 3 summarizes the occurrence of the interaction categories at T0, T1, and T2 for all six participants.

Table 3

Occurrence (%) of the interaction categories at T0, T1, and T2 for the six participants

Participant	Attention by caregiver			Attention by participant			Confirmation by caregiver			Affective involvement		
	T0	T1	T2	T0	T1	T2	T0	T1	T2	T0	T1	T2
Richard	0	67.9	67.9	53.3	63.3	78.6	0	0	0	0	0	0
George	46.7	100	90	93.1	60	53.3	0	10	16.7	0	16.7	23.3
Tina	95.2	60	100	78.6	56.7	66.7	0	0	0	0	0	0
Harold	51.7	75.9	-[a]	33.3	63.3	-	0	3.3	-	0	0	-
Romy	92.3	96.2	-	40	46.7	-	0	0	-	0	0	-
Charles	79.2	100	-	83.3	66.7	-	0	10	-	0	10	-

[a] No relevant data available in this research period.

Attention by caregiver. The occurrence of attention by the caregiver had improved at T1 as compared to T0 for five participants; for Tina it decreased. At T2, the occurrence of attention by caregiver was higher as compared to T0 for Richard, George, and Tina.

Attention by participant. Comparison of the occurrence of attention by the participant at T1 versus T0 yields an inconsistent picture: an improvement for Richard, Harold, and Romy, but a decrease for the others. The results at T2 are inconsistent as well: an improvement for Richard and a decrease for Tina and George when compared to T0.

Confirmation by caregiver. Confirmation by the caregiver did not occur for any of the participants at T0. At T1, confirmation occurred for George, Harold, and Charles. At T2, confirmation further improved only for George. No confirmation was observed for Richard and Tina at T2.

Affective involvement. Affective involvement did not occur at T0 for any of the participants, but at T1 it occurred for George and Charles. At T2, affective involvement was still higher as compared to T0 for George. No affective involvement occurred at T2 for Richard or Tina.

3.3.2 Communication

Table 4 presents the highest level of communication by the six participants for the functions *refuse*, *obtain*, and *social interactions* at T0, T1, and T2, as well as the increase in levels from T0 to T2. The communicative function *provide or seek information* did not occur.

Table 4

Level of communication for the functions refuse, obtain, and social interactions at T0, T1, and T2 for the six participants

		T0	T1	T2	Gain T0-T2[a]
Richard	Refuse	IV	IV	IV	0
	Obtain	IV	VI	VI	2
	Social	III	III	III	0
George	Refuse	III	III	III	0
	Obtain	I	III	III	2
	Social	II	II	III	1
Tina	Refuse	III	III	III	0
	Obtain	II	III	III	1
	Social	III	III	III	0
Harold	Refuse	II	II	II	0
	Obtain	IV	VI	VI	2
	Social	III	III	III	0
Romy	Refuse	III	IV	IV	1
	Obtain	III	III	III	0
	Social	III	III	III	0
Charles	Refuse	II	III	III	1
	Obtain	II	II	II	0
	Social	II	II	II	0

[a] Gain score represents the increase in levels from T0 to T2.

Two participants, Romy and Charles, showed a higher level of communication at T2 compared to T0 for the function *refuse*. The other four participants obtained a higher level of communication at T2 for the function *obtain*. For George, the level of communication at T2 was higher than at T0 for the function *social interactions*. The level of communication increased for all participants in at least one communicative function.

Table 5 presents the variety of communication skills assessed at T0, T1, and T2 for all participants, and the percentage gain from T0 to T2.

Table 5

Variety in communication skills (%)[a] at T0, T1, and T2 for the six participants

Participant	T0	T1	T2	% Gain T0-T2[b]
Richard	18.8	21.3	23.8	26.7
George	7.5	11.3	13.8	83.3
Tina	11.3	12.5	13.8	22.2
Harold	15	16.3	17.5	16.7
Romy	10	11.3	11.3	12.5
Charles	8.8	10	10	14.3

[a] Computed as the total number of points (1 point for each communication skill possessed) divided by the maximum number of points (80), and multiplying the resultant number by 100.

[b] Computed as T2 - T0 divided by T0, multiplied by 100.

The variety of communication skills (a percentage score) improved for all participants. The gain ranged from 12.5% to 83.3%. This means that the number of communication skills the participants possess increased, regardless of the level and function of communication. Tina, for instance, possessed no communication skills for the function *obtain* at level 3 at T0, one (requests more object) at T1, and another one (requests new object) at T2. At T1 she asked for the next piece of bread by holding up her hand. At T2 she asked for a drink by holding her hand in the position for drinking, while eating porridge. George possessed the communication skills belonging to levels 1 and 2 of the Communication Matrix for the function *social interactions* at T0 and T1. At T2, he had acquired a communication skill at level 3 for this function, "shows affection". This meant that George "expressed interest in other people" at T0, for example, by touching the caregiver during shaving. At T2, he "showed affection", for example, by sitting next to the caregiver while smiling.

3.4 Discussion

This study examined the effects of the first 2 years of participation in Project CHANGE on the interaction between participants with CDB and an ID and their caregivers and on the participants' expressive communication skills. In the first 2 years of Project CHANGE, the participants underwent a pervasive change in living conditions and social experiences, starting with a transition to a new group home. Here, they came to live with other persons with deafblindness. Staff was trained and coached in offering the specialized care needed to the participants. The environment was clearly structured, individual programs to enhance communication were followed, and activities of daily life were made predictable, following recommendations from the literature (Evenhuis et al., 2001; Fellinger et al., 2009). Overall, the conclusion is that the changes have resulted in an improvement in interaction and communication. Positive results are found in the occurrence of attention by caregiver, confirmation by caregiver, and affective involvement, as well as in the level of expressive communication on three functions of communication and in the variety of communication skills across levels and functions possessed by the participant. The results for attention by participant, however, are not clear.

Attention by the caregiver is part of, but also a prerequisite for, harmonious interactions with persons with CDB and an ID. The improvement in attention has probably laid the foundation for further harmonious interactions. Confirmation by the caregiver is also an important ingredient of harmonious interactions. The confirmation that an initiative has been noticed and recognized does not come naturally in interaction with persons with CDB and an ID. The highly insufficient confirmation by the caregiver at T0 emphasizes the need for special training or coaching. Such training and coaching can indeed be effective, as demonstrated by the occurrence of confirmation by the caregiver for George, Harold, and Charles at T1 and for George at T2. The same can be said for affective involvement or the mutual sharing of emotions. Affective involvement was lacking entirely at T0, which stresses the need for special training aimed at fostering this important aspect and outcome of harmonious interaction. Again, after training and coaching (T1), affective involvement did occur for George and Charles. At T2, affective involvement occurred for George.

The varying results for attention given by the participant are puzzling, possibly because the improved attention by the caregiver resulted in their being more observant of the participants’ signals. If a caregiver observes the person with CDB and an ID regulating intensity, and respects this, the attention paid by the participant might wane. In this study, regulation of intensity was not included as a variable. Because we found no prior studies that report on this relationship, we cannot draw any conclusions about this here. Research on the relationship between the attention by the caregiver and regulation of intensity by the person with CDB and an ID is much needed.

Expressive communication skills of the participants also improved, both in the level of communication for the functions *refuse*, *obtain*, and *social interactions*, and in the variety of communicative behaviors. It implies that the participants learned to express themselves better, having a broader range of communicative behaviors at their command, and achieving a higher level of communication. The communicative function *provide or seek information* did not occur in this study. It might be that more attention for this function is needed in training and coaching for developing this function (Damen et al., 2014), but in this study this was not the aim.

As stated in "Introduction" section, harmonious interaction is important for the well-being and functioning of persons of all ages (Diamond & Aspinwall, 2003; Sroufe, 1995; Trevarthen & Aitken, 2001) and is essential for satisfying the human need to belong (Baumeister & Leary, 1995), quality of life (Prain et al., 2012b), and development (Dammeyer, 2010). Our study shows improved interaction and communication with persons with CDB and an ID in adulthood, even when no special attention was given to the deafblindness in their youth, which is a promising result for all persons with CDB and an ID living in similar settings. It is recommended to teach professionals on deafblindness and its consequences for daily life. A second recommendation is to make regular use of video analysis to improve the interaction skills of caregivers and to recognize and interpret interactive and communicative expressions of the persons with CDB and an ID.

3.4.1 Limitations and directions for future research

The first 2 years of Project CHANGE included many changes for the participants (new home, smaller living group, new approach). Our results suggest that these changes have led to an improvement in the interaction and communication (which we used as outcome measures). It may be assumed that the interaction training for the caregivers has contributed most to this effect, but it cannot be excluded that other changes in living conditions have also contributed, for example, by improving the mood of the participants, which, in turn, may have made it more easy to establish interaction and communication. Given that we focused specifically on the effects of CHANGE on interaction and communication and did not include other outcome measures, we cannot draw any valid conclusions regarding the possible effect of other changes in living conditions beyond the interaction training.

The video recordings used to observe interaction were selected to include comparable situations at the different time periods for each participant. However, different caregivers were included in the videos, which may have influenced the results, as interaction and communication skills, age, level of education, and years of experience will vary between caregivers. Since different caregivers were observed while interacting with the participants at T0, T1, and T2, it was not possible to control for caregiver characteristics in this study; only the effects across caregivers could be examined. For future studies examining participant-caregiver interactions over time, however, we recommend to examine the role of caregiver characteristics. It would be interesting, for example, to examine the role of the length of the relationship between participant and caregiver in interventions to improve interaction and communication.

An evaluation of the caregiver’s perception and professional development is lacking in this study. Since this could have added important information, our recommendation for future studies is to include a systematic evaluation by the caregivers.

Measuring communication retrospectively has its limitations. The results are confined to communicative behaviors recorded on videos or in files: greater variety of communicative behaviors or a higher level of communication might well be witnessed in everyday life. Besides that, the participants' files contained mainly general information. This made it difficult to allocate the information to the specific communicative behaviors in the Communication Matrix.

As this study underlines, adults with CDB and an ID do require a special approach when it comes to interaction and communication. The average age of the participants was 42 years, and they had never been offered an approach designed to meet their specific combination of disabilities. Far into adulthood, they entered the Project CHANGE, specially designed to improve their interaction and communication. The results show that a tailored approach can be fruitful, regardless of their age. It is remarkable that the observed improvements already took place during the first 2 years, with minimal training and on-the-job coaching of the involved staff.

After the first 2 years, Project CHANGE has continued, with more specific training and on-the-job coaching being provided to caregivers. Future research will show whether this has led to further improvements in interaction and communication in persons with CDB and an ID.

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Chapter 4

Effects of an intervention program on interaction and bodily emotional traces in adults with congenital deafblindness and an intellectual disability

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Abstract

Interaction with people with congenital deafblindness (CDB) and an intellectual disability (ID), and recognition of their often unconventional expressions, is complex. In this study, the effects of a two-phase intervention program intended to foster harmonious interaction and the use and recognition of expressions based on a bodily emotional trace (BET) were examined. Five adults with CDB and an ID, and ten of their caregivers participated in the study. A multiple-baseline design was used. Target behaviours were attention by caregiver, attention by participant, confirmation by caregiver, mutual affective involvement, quality of interaction, participant expressions based on a BET, and participant expressions based on a BET recognised by the caregiver. Overall, the results tend to be positive. However, some of the changes were minimal and the results varied considerably among participants. The results of this study suggest that it is possible to foster harmonious interaction and the use and recognition of expressions based on a BET in adults with CDB and an ID.

4.1 Introduction

Having harmonious interactions with others fosters well-being and improves the functioning of people of all ages (Diamond & Aspinwall, 2003; Sroufe, 1995; Trevarthen & Aitken, 2001). Infants have biologically founded capacities for social interaction (Aitken & Trevarthen, 1997), which is defined as the process through which two individuals mutually influence each other’s behaviour (Bjerkan, 1996; Janssen, Riksen-Walraven, & Van Dijk, 2003a).

From childhood on, vision and hearing play important roles in social interaction, as is evident in eye-gaze, smiling, and reactions to acoustic signals similar to the human voice (Pérez-Pereira & Conti-Ramsden, 1999). Vision and hearing cannot be addressed in the same way in interactions with people with congenital deafblindness (CDB). It is therefore necessary to address other senses, like the tactile modality.

People with CDB mainly experience the world through their proximal senses, often resulting in expressions that are atypical for seeing and hearing people. These expressions can be stereotypical and idiosyncratic (Bjerkan, 1996), and difficult for their communication partners to understand. Expressions made by people who have an intellectual disability (ID) in addition to CDB are often even more idiosyncratic and subtle, and are not always used consistently. This makes it very hard to notice and interpret these expressions, complicating interaction and subsequent qualitative communication. Qualitative communication is assumed to be based on harmonious interactions.

Moreover, communication is facilitated by a history of shared experiences. Adults with CDB and an ID who live in residential settings meet many different communication partners, which severely limits the construction of a history of shared experiences to communicate about. In short: establishing harmonious interaction and subsequent qualitative communication with people with CDB and an ID is very difficult (Bloeming-Wolbrink, Janssen, Ruijssenaars, Menke, & Riksen-Walraven, 2015; Bruce, 2005; Dalby et al., 2009; Damen, Janssen, Huisman, Ruijssenaars, &

Schuengel, 2014; Dammeyer, 2010; Hart, 2010; Janssen et al., 2003a; Martens, Janssen, Ruijssenaars, & Riksen-Walraven, 2014). Communication partners (e.g. caregivers, parents, teachers) need high levels of sensitivity, special insights, and considerable skills to establish harmonious interaction and qualitative communication with people with CDB and an ID (Janssen, Riksen-Walraven, & Van Dijk, 2003b).

Several intervention studies have shown that coaching communication partners has positive effects on interaction and/or communication with people with CDB (e.g. Damen, Janssen, Ruijssenaars, & Schuengel, 2015; Janssen et al., 2003b; Martens, Janssen, Ruijssenaars, Huisman, & Riksen-Walraven, 2014a; Martens, Janssen, Ruijssenaars, Huisman, & Riksen-Walraven, 2014b). Consequently, it is worthwhile to also put efforts into coaching communication partners to improve their interaction and communication with people with CDB and an ID.

People with CDB mainly experience the world through bodily-tactile experiences. When they think about these experiences, their thoughts are often reflected in bodily expressions such as touching a location on the body, making a movement, or replicating a sensation. Such bodily expressions of memories are based on a bodily emotional trace (BET). These expressions are typically neither social nor communicative when they emerge (Daelman et al., 2001), but they do show that something is going on in the mind of the person with CDB, and they may give important insights into how that person experiences the world and reflects on earlier experiences (Daelman, 2003).

Recognising these idiosyncratic expressions is useful for the partner in the meaning-making process and is thus valuable for communication, in which information is conveyed to and received and interpreted from others (Rowland, 2009). According to Johnson (2008), the human “mind” cannot exist apart from embodiment. What and how humans experience, feel, think, value, and act is shaped by embodiment (Johnson, 2008). This is very clear in people with CDB and an ID. Training their communication partners to recognise and use BETs might provide insights into how a person with CDB and an ID reflects upon the world, thus helping the communication partners recognise and add meaning to idiosyncratic expressions.

The present study constitutes the second part of the broader Project CHANGE, an ongoing project that aims to improve interaction and communication in adults with CDB and an ID. The first part of Project CHANGE (reported in an earlier paper; Bloeming-Wolbrink et al., 2015) focused on the effects of a general improvement in the life circumstances of adults with CDB and an ID and the general training and coaching given to their caregivers on the quality of their interaction and expressive communication.

The goal of the present study – the second part of Project CHANGE – was to examine the effects of a two-phase intervention program for caregivers working with adults with CDB and an ID. It is specifically aimed at fostering harmonious interactions (Phase I) and the use and recognition of participant expressions based on a BET (Phase II). During Phase I, it was attempted to improve the interactive competence of the adults with CDB by coaching their caregivers to respond appropriately to their interactive behaviours. During Phase II it was attempted to improve the use and recognition of participants' expressions based on a BET by coaching their caregivers to introduce something new in a routine situation and to strengthen the experience of the participant (e.g. by repeating parts of the experience).

The present study aims to answer three research questions:

1) To what extent does the intervention increase the quality of interaction and, more particularly, the occurrence of attention by the caregiver, attention by the participant, confirmation by the caregiver, and affective involvement between participant and caregiver?
2) To what extent does the intervention increase the number of expressions based on a BET made by the participant and the recognition of these expressions by the caregiver?
3) To what extent do the effects of the intervention endure after completion of the intervention?

4.2 Method

4.2.1 Participants and setting

The study was conducted at a facility for people with intellectual and sensory disabilities in the Netherlands. Five adults with CDB and an ID (“participants”) took part in the study: four men and one woman. They differed with regard to age and the degree of sensory disability. Table 1 presents an overview of relevant participant characteristics.

Table 1

Participant characteristics at the start of the intervention program

Participant code	Gender	Age (years)	SRZ score[a]	Visual disability[b]	Hearing disability[c]
P1	Female	42	3-	Totally blind	Moderate/severe hearing loss
P2	Male	53	3-	Blind	Profound hearing loss
P3	Male	42	3	Totally blind	Hearing loss; unclear whether there is residual hearing
P4	Male	18	3-	Blind	Profound hearing loss
P5	Male	43	3	Blind	Profound hearing loss

[a] SRZ POP-EM score of 3 or 3- is appropriate in the case of a profound intellectual disability.

[b] Totally blind: no light perception; Blind: blind with light perception.

[c] Moderate hearing loss: 41-60dB; Severe hearing loss: 61-80dB; Profound hearing loss: ≥81dB.

Prior to the start of Project CHANGE, none of the participants had received deafblind education or been educated with a consistent approach that took their deafblindness into account. The main focus in their education had been on the ID.

Ten caregivers (all female) were included in the study. Their ages ranged from 20 to 55 years (M = 36.4). Four caregivers had a Bachelor’s degree, and the other six caregivers had had vocational training. They had 2 to 28 years (M = 12.6) of experience in working with persons with ID, and 1 to 17 years (M = 4.6) of experience in working with persons with deafblindness.

Each participant formed pairs, at random, with two caregivers, resulting in ten pairs. Two pairs were excluded from analysis, due to illness of the caregiver and the poor quality of video recordings. Thus, results are presented for three participants with two caregivers (A and B) and for two participants with one caregiver.

The study was approved by the board of the facility. Written informed consent was obtained from the participants' legal representatives. Verbal informed consent was obtained from the caregivers. To protect the participants' identities, each participant is identified by a number in this article.

4.2.2 Research design

This study followed a multiple-baseline design across subjects (see Figure 1). The baseline becomes longer for each consecutive participant, with a minimum length of four baseline sessions (Barlow, Nock, & Hersen, 2009) and a maximum of 12. The two-phase intervention program had a total duration of 15 weeks: 9 for Phase I (interaction training) and 6 for Phase II (BET training). These two phases were complementary. The design was time-based: Phase II began directly after finishing Phase I, regardless of the Phase I results. The study ended with follow-up sessions at 3, 7, and 11 weeks after the end of the intervention program.

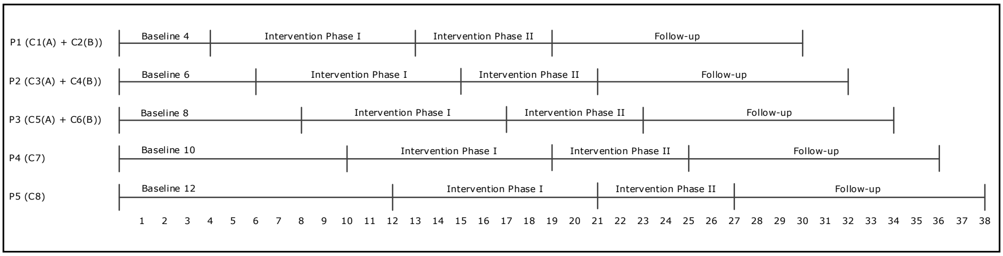

Figure 1. Duration of the effect study (in weeks) per participant and caregiver.

4.2.3 Intervention program

The intervention program consists of Phase I: interaction training, and Phase II: BET training. The outline of the coaching in Phase I was based on the diagnostic intervention model CONTACT, which is used to foster harmonious interactions between children with deafblindness and their educators. The model is based on theoretical aspects from attachment and intersubjectivity theories (see Janssen et al., 2003a). The intervention program applied in this study adds a second Phase, a BET training, based on theoretical aspects of embodiment theory (Johnson, 2008). Phase II of the intervention program focused on increasing the use and recognition of participant expressions based on a BET.

The intervention program, consisting of seven subsequent steps (Phase I: step 1-4, Phase II: step 5-7), was applied by a coach (the first author). As shown in Figure 2, the intervention program included five 45-minute coaching sessions registered in a protocol, resulting in a total of 3 hours and 45 minutes of information transfer and video-feedback per caregiver.

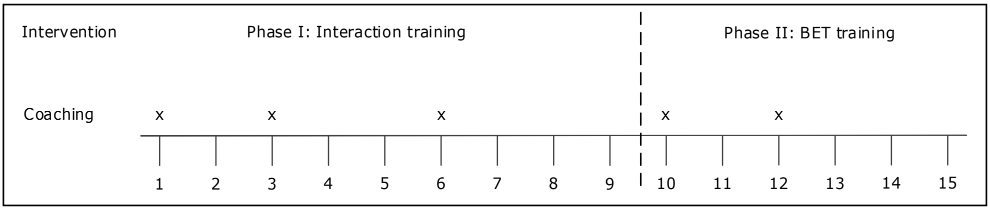

Figure 2. Coaching sessions across weeks during the intervention program.

The intervention protocol[4]:

1. *Determining the question.* The caregivers requested coaching for questions concerning interaction.
2. *Clarifying the question.* The coach consulted the caregivers in order to specify the questions for interaction training. Examples include: how can I recognise the participant's initiatives and confirm these?; how can I share emotions with the participant in a tactile way?
3. *Interaction analysis.* The coach analysed video recordings of interaction to translate the questions for interaction training into

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[4] The exact content of the intervention program is available from the first author.

intervention aims for Phase I of the intervention. Example: the caregiver confirms the participant’s initiative by repeating it tactilely.

4. *Implementing intervention focusing on interaction.* The three coaching sessions included information transfer and video-feedback. The intervention aims, other aspects of interaction, and the interaction context were addressed. Examples of aspects of the interaction context discussed include: changing the caregiver’s position during a foot massage to make the participant more comfortable or changing the caregiver’s position while baking cookies so that she could observe the participant’s face.
5. *BET analysis.* The coach analysed video recordings for preparing step 6. The focus was on possibilities for introducing something new in the routine situation, since this seems to evoke BETs and expressions based on a BET (Bloeming-Wolbrink, 2007). Looking for ways to strengthen the experience for the participant, for example repeating parts of the experience, was another focus in the analysis.
6. *Implementing intervention focusing on BETs.* The two coaching sessions included information transfer and video-feedback. The definition and importance of the BET concept, and ways to strengthen an experience were discussed in the first coaching session of Phase II, illustrated with video-clips of persons with CDB. It was discussed which new topic could be introduced in the routine situation. Example: intentionally leaving the butter out while baking cookies. In the last coaching session video-feedback was used to observe expressions based on a BET, and to decide whether changes in the new topic were needed (e.g. to clarify the situation for the participant or to arouse the participant’s interest).
7. *Evaluation.* The intervention is evaluated with regard to the intervention aims set and the satisfaction of the caregivers with the intervention process.

4.2.4 Data collection

To examine the effects of the intervention program on interaction and BETs, 20-minute video recordings were made weekly during baseline and the intervention phases. During follow-up, they were made 3, 7, and 11 weeks after the end of the intervention. Throughout the study, each participant-caregiver pair was recorded during the same activity, an activity in which both parties felt comfortable (based on the caregiver's impression), and which guaranteed that it could always be performed (e.g. baking cookies, interacting on the waterbed).

The recordings of each pair were all made on the same day of the week and at the same time of day, based on the caregiver's working schedule and the time of the day the activity could be performed. Caregivers' coaching sessions were pre-planned for weeks 1, 3, 6, 10, and 12 of the 15-week intervention program (see Figure 2). In case of unanticipated circumstances, like illness, recordings or coaching sessions were rescheduled.

4.2.5 Instruments and measures

4.2.5.1 Interaction: coding.

Observation categories. To measure the effect of the intervention program on interaction, four categories of interactive behaviour were used (adopted from Janssen et al., 2003a):

1a) Attention by caregiver: focus on interaction partner, interaction content, people and/or objects within the interaction context;

1b) Attention by participant (see 1a);

2) Confirmation by caregiver: clear acknowledgement that an initiative has been noticed and recognised;

3) Affective involvement: mutual sharing of emotions between caregiver and participant.

Observation procedure and scoring. Five-minute fragments (4-12 for baseline, 15 for intervention, 3 for follow-up) with a fixed starting point per pair were selected from the 20-minute recordings. The fixed starting point was chosen to maximise the comparability of the fragments. Examples of fixed starting points are offering the pastry bowl to the participant and taking off the participant's shoes.

Observers coded the videos on an observation form with 10-second intervals. If attention by participant and caregiver was observed for ≥ 6 seconds during the interval, it was coded 1; otherwise, it was coded 0. Confirmation by caregiver and affective involvement were coded 1 if observed at least once during an interval; if not, they were coded 0. For each category, an interval was excluded from coding if the information needed to code that category was lacking (e.g. if a face was not visible, affective involvement could not be observed). The occurrence of the categories of interactive behaviour per five-minute fragment was expressed in a number, calculated by counting the number of intervals with code 1. The maximum score was 30. In case of intervals excluded from coding, the number of intervals with code 1 was divided by the total number of intervals with code 1 and 0, and multiplied by 30.

Five trained observers, current or graduated Bachelor's and Master's students in the social sciences, coded the videos. To control for observer drift, the videos were coded at random by the observers. Prior to each observation session, each observer read the definition of the categories and the operationalisation of the participant's interactive behaviour. The operationalisation of the participant's interactive behaviour was based on videotaped observations of various interaction situations.

Inter-rater reliability. Prior to formal data collection, the observers were trained to reach 80% inter-rater reliability for the occurrence of the interactive behaviours for three training videos per participant-caregiver pair. The training videos were not used for the actual coding. Two observers were trained for each participant.

When coding the research recordings, the first observer coded all of a participant's recordings; the second observer coded ≥25% of them (Barlow et al., 2009), which were selected at random. Inter-rater reliability was calculated using percentage agreement and Kappa, following Prain, McVilly, and Ramcharan (2012). Percentage agreement was calculated by dividing the number of agreements on occurrences and non-occurrences by the total number of agreements and disagreements, and multiplying the result by 100 (Brown & Snell, 1993).

Inter-rater reliability was calculated across measurements and participants. The percentage agreement and Kappa scores were 96.8% and .84 for attention by caregiver, 91.4% and .83 for attention by participant, 97.0% and .74 for confirmation by caregiver, and 99.4% and .44 for affective involvement. The Kappa score for affective involvement implies that the results for this category should be interpreted with great caution.

4.2.5.2 Interaction: focus group.

To qualitatively measure the effect of the intervention program, the quality of interaction in the participant-caregiver pairs was discussed in three focus group meetings, with the first author as moderator. Four people participated in the focus group: a professor in Special Needs Education with a focus on deafblindness (the second author), another professor in Special Needs Education (the third author), and two Master's students in Special Needs Education. For each participant-caregiver pair, four coded five-minute fragments were selected: one for baseline, two for the ends of the intervention phases, and one for follow-up. The fragments were offered to the focus group members in a random order per participant-caregiver pair, in order to avoid observer drift.

The discussion of the quality of interaction for each pair started with a short introduction of the participant and the activity. The four fragments were watched with short breaks in between, during which the focus group members were asked to react in a few words about the quality of interaction. After finishing all fragments of a pair, a longer discussion began about the quality of interaction in general and different aspects of it: togetherness, turn taking, closeness, initiatives, and answers.

Subjective judgement. After finishing the discussion for a participant-caregiver pair, the four focus group members were asked to make independent judgements about the quality of interaction in each fragment. Judgements were made on a scale ranging from 1 (not good) to 4 (very good). Focus group members were also asked to rank the quality of interaction in the four fragments. The individual scores and rankings were combined into an overall score and ranking for each fragment, by summing the individual scores or rankings and dividing the total number by four.

Content analysis. Focus group meetings were audiotaped and then transcribed. Eleven important concepts (attention, attunement, answers, involvement, communication, contact, initiatives, interaction, reacting, togetherness, and closeness) were included in the analysis. These concepts were derived from the categories of interactive behaviour (Janssen et al., 2003a), the questions asked by the moderator, and concepts often mentioned by the focus group members. Inclusion criteria for the concepts were identified. Information about the inclusion criteria per concept is available from the first author.

Semantic content analysis was performed, in which concepts are classified according to their meaning. Semantic content analysis has three forms, one of which (assertion analysis) was performed here. Assertion analysis includes both the frequency of naming the concepts and the nature of the context in which the concepts are used (positive, neutral, negative) (Janis, 1965, in Stewart, Shamdasani, & Rook, 2007). A Master's student performed the analysis and coded the concepts in the transcriptions. Each concept received a positive (1), negative (-1), or neutral (0) characterisation, resulting in a score that reflected the judgement of the quality of interaction for each fragment.

4.2.5.3 Bodily emotional traces.

Observation categories and criteria. The effect of the intervention program on the use and recognition of participants' expressions based on BETs was measured by coding the expressions the participants made and coding whether the caregiver noticed them. Three criteria were set to identify these expressions: it is an expression of thinking; it is judged to be referring to an earlier experience in a bodily way; and, when making the expression, the person is not paying attention to the surroundings

(Stellingwerf, 2012). These criteria were operationalised for each participant, meaning that concrete behaviours were described.

Observation procedure and scoring. For each participant, one participant-caregiver pair was included in the analysis. The pairs were selected at random. Six 10-minute fragments were selected from the 20-minute recordings: one from baseline, two from each of the intervention phases, and one from follow-up. The 10-minute length was chosen because it was expected that expressions based on a BET would occur often enough in this period to make it possible to compare the number of expressions in the different phases. If possible, the caregiver's introduction of the new topic was taken as the starting point of the 10-minute analysis. When necessary, in order to analyse 10 minutes of the recordings, the starting point was set before the introduction of the new topic.

Two observers independently coded each fragment on a specially designed observation form. Samples of 15-second intervals were used. Per interval, expressions based on a BET were coded 1 if observed at least once, and coded 0 if not observed. Expressions continuing in subsequent intervals were coded 1 in the first interval in which they were observed, and coded 0 in the subsequent intervals, to avoid counting a single expression twice. A note was made on the observation form in these cases. In the case of a code 1, the expression based on a BET was described on the observation form.

After the observers finished independently coding a participant's six fragments, they discussed their scores and arrived at a consensus score. An overall score, reflecting the occurrence of expressions based on a BET in the 10-minute episode, was calculated by summing the number of intervals coded 1.

For each 15-second interval coded 1, the observer noted whether or not the caregiver recognised the expression. The observer carefully described the behaviour of the caregiver that made her believe the caregiver did or did not recognise the expression. A percentage score was calculated by dividing the number of expressions recognised by the caregiver by the total number of expressions in the fragment, multiplied by 100%.

Two trained observers, one current and one graduated Master’s student in Special Needs Education, coded the videos. To control for observer drift, the videos were presented at random to the observers. Prior to each observation session, the observers read the manual for the participant they observed. The manual consisted of the three criteria for identifying expressions based on a BET and the operationalisation of these criteria (i.e. the description of concrete behaviours by the participant). Examples for the criterion “it is an expression of thinking” are tapping with the finger right beside the eye, freezing, or turning the head backwards. Examples for the criterion “it is judged to be referring to an earlier experience in a bodily way” are making a movement that resembles the movement in an earlier experience or touching a location on the body that was touched during the earlier experience. Examples for the criterion “the person is not paying attention to the surroundings” are the participant closing his or her hand or not responding to questions.

Inter-rater reliability. Prior to formal data collection, the observers were trained until 80% inter-rater reliability was reached for the occurrence of expressions based on a BET for three training videos per participant. The training videos were not used for the actual coding. Following the recommendation of Prain et al. (2012), we chose to use consensus coding because the participants’ expressions were expected to be highly idiosyncratic and context dependent. The observers coded the research videos independently. During scoring, the observers noted the reasons when coding 1. After the independent coding, the observers compared and discussed their scores and their justifications for them. This discussion resulted in a consensus score.

Inter-rater reliability for the actual research recordings was calculated using percentage agreement and Kappa. Inter-rater reliability was calculated across measurements and dyads for the occurrence of expressions based on a BET, between observers 1 and 2 (99.6% and .90), between observer 1 and consensus score (99.5% and .87), and between observer 2 and consensus score (99.9% and .98). Both percentage agreement and Kappa were good, even when comparing observers 1 and 2.

4.2.6 Social validity

To ensure the clinical significance of the intervention program, the participating caregivers were actively involved in setting and evaluating their own intervention aims for interaction and in choosing which activity to perform with the participants. After finishing the study, they were asked to rate components of the intervention program on a 5-point scale, on an adapted version of the Social Validity Scale (Bloeming-Wolbrink & Janssen, 2010, following Seys, 1987). Components were subjective effectiveness (e.g. "the idea of coaching caregivers in order to improve the interaction between participants and caregivers is very bad/ bad/ sufficient/ good/ very good"), effectiveness of single components of the intervention (e.g. "recognising expressions based on a BET was very hard/ hard/ well performable/ good/ very good"), and workability (e.g. "I found that the presence of a video camera in the group home was very bad/ bad/ sufficient/ good/ very good"). All the caregivers (n=8) returned the questionnaire.

4.2.7 Data analysis

Given the relatively small number of observations and the serial dependencies in the data, statistical tests such as time series methods are not feasible. Therefore, descriptive and visual analysis were used as the most important analysis method in this study. This method is commonly used in single subject experimental research (Horner, Carr, Halle, Odom, & Wolery, 2005; Nourbakhsh & Ottenbacher, 1994).

4.3 Results

4.3.1 Effects on interaction: coding

Table 2 presents an overview of the mean occurrence, range, and standard deviation of the categories of interactive behaviour (i.e. attention by caregiver and affective involvement) across the four phases of the study for each participant. Figures 3 and 4 present the results for the categories of interactive behaviour, attention by participant, and confirmation by participant.[5] The maximum score was 30, representing the total number of intervals in a five-minute fragment.

Attention by caregiver. For participants 1, 3, and 4, the mean occurrence of attention by caregiver was already high at baseline (28.8, 29.8, and 29.9, respectively), making only small improvements possible (see Table 2). For participants 2 and 5, however, the mean occurrence of attention by caregiver at baseline was 20.8 and 14.3, respectively. Large improvements for these participants were found, with a mean occurrence of 26.7 and 17.4, respectively, in Phase I, and a mean occurrence of 26.6 and 29.3, respectively, in Phase II. Also, a smaller range in scores was found during intervention for these participants, also reflected in a lower standard deviation.

Attention by participant. The line graphs in the right panel of Figure 3 demonstrate a large variability between the participants. For participant 4, the occurrence was already high during baseline and stayed high with a few dips in Phase II and the follow-up phase. Figure 3 shows that occurrence increased for participants 2 and 5 during Phase I, but slightly decreased during Phase II, with a downward trend line. Occurrence during follow-up was variable: participant 5 had an upward trend line and participant 2 had a slightly downward trend line. Participant 1 had a downward trend line in the scores for Phase I and a strong upward trend line for Phase II because

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[5] More detailed information about the results for the categories of interactive behaviour attention by participant and confirmation by participant is available from the first author.

of a high peak in session 19. The occurrence of attention by the participant declined in the follow-up with a slight upward trend line.

Confirmation by caregiver. Figure 4 shows the large variability within the different phases per participant and between the individual participants. For participant 4, confirmation by the caregiver occurred relatively more often than for the other participants. Remarkably, participant 4 started Phase I with a very high increase in the occurrence of confirmation, but the trend line unfortunately lowered during this phase. In the other phases, there was not much difference from the baseline.

The other participants had upward or steady trend lines in Phase I. In Phase II, the occurrence of confirmation was lower than in Phase I for all cases except participant 2. An upward trend line was also observed for participant 3. For participants 1 and 2, occurrence was higher during follow-up than during the other phases (unfortunately with a downward trend for participant 2). For the other three participants, occurrence during follow-up was higher than the occurrence in Phase II. A peak was found in session 24 for participant 3, but unfortunately a downward trend line for participants 3 and 4 was found.

Affective involvement. No affective involvement occurred for participant 5 (see Table 2). For participant 4, affective involvement only occurred during follow-up. The other three participants were observed with two caregivers. However, no affective involvement was observed with one of the caregivers. With the other caregiver, a more regular occurrence of affective involvement was observed in three of the four study phases. The highest occurrence per participant was found at follow-up and Phase II.

Table 2

Attention by caregiver and affective involvement over time

		Attention by caregiver										
			P1			P2			P3		P4	P5
		A	B	Score	A	B	Score	A	B	Score	Score	Score
Baseline	Mean	27.8	29.8	**28.8**	26.5	15	**20.8**	29.8	29.8	**29.8**	**29.9**	**14.3**
	Range	25-30	29-30	**25-30**	25-28	6-28	**6-28**	29-30	29-30	**29-30**	**29-30**	**9-26**
	SD	2.63	0.50	**2.05**	1.22	8.29	**8.25**	0.46	0.46	**0.45**	**0.32**	**4.97**
Intervention Phase I	Mean	29.6	29.7	**29.6**	25.2	28.2	**26.7**	30	29.6	**29.8**	**29.9**	**17.4**
	Range	28-30	28-30	**28-30**	23-27	25-30	**23-30**	30	28-30	**28-30**	**29-30**	**12-25**
	SD	0.73	0.71	**0.70**	1.39	1.64	**2.14**	0	0.73	**0.55**	**0.33**	**4.00**
Intervention Phase II	Mean	30	28.7	**29.3**	25	28.2	**26.6**	30	29.3	**29.7**	**30**	**29.3**
	Range	30	22-30	**22-30**	23-27	26-29	**23-29**	30	28-30	**28-30**	**30**	**26-30**
	SD	0	3.27	**2.31**	1.67	1.17	**2.15**	0	0.82	**0.65**	**0**	**1.63**
Follow-Up	Mean	30	28	**29**	24.7	29	**26.8**	30	29	**29.5**	**30**	**24**
	Range	30	27-29	**27-30**	23-27	28-30	**23-30**	30	27-30	**27-30**	**30**	**22-26**
	SD	0	1.00	**1.26**	2.08	1.00	**2.79**	0	1.73	**1.22**	**0**	**2.00**
		Affective involvement										
			P1			P2			P3		P4	P5
		A	B	Score	A	B	Score	A	B	Score	Score	Score
Baseline	Mean	0	0.3	**0.1**	0	0	**0**	0.1	0	**0.1**	**0**	**0**
	Range	0	0-1	**0-1**	0	0	**0**	0-1	0	**0-1**	**0**	**0**
	SD	0	0.50	**0.35**	0	0	**0**	0.35	0	**0.25**	**0**	**0**
Intervention Phase I	Mean	0	0.1	**0.1**	0	0.2	**0.1**	0.2	0	**0.1**	**0**	**0**
	Range	0	0-1	**0-1**	0	0-1	**0-1**	0-1	0	**0-1**	**0**	**0**
	SD	0	0.33	**0.24**	0	0.44	**0.32**	0.44	0	**0.32**	**0**	**0**
Intervention Phase II	Mean	0	0.3	**0.2**	0	0.2	**0.1**	0	0	**0**	**0**	**0**
	Range	0	0-1	**0-1**	0	0-1	**0-1**	0	0	**0**	**0**	**0**
	SD	0	0.52	**0.39**	0	0.41	**0.29**	0	0	**0**	**0**	**0**
Follow-Up	Mean	0	0	**0**	0	0.7	**0.3**	0.3	0	**0.2**	**0.7**	**0**
	Range	0	0	**0**	0	0-1	**0-1**	0-1	0	**0-1**	**0-2**	**0**
	SD	0	0	**0**	0	0.58	**0.52**	0.58	0	**0.41**	**1.15**	**0**

Note. SD = standard deviation.

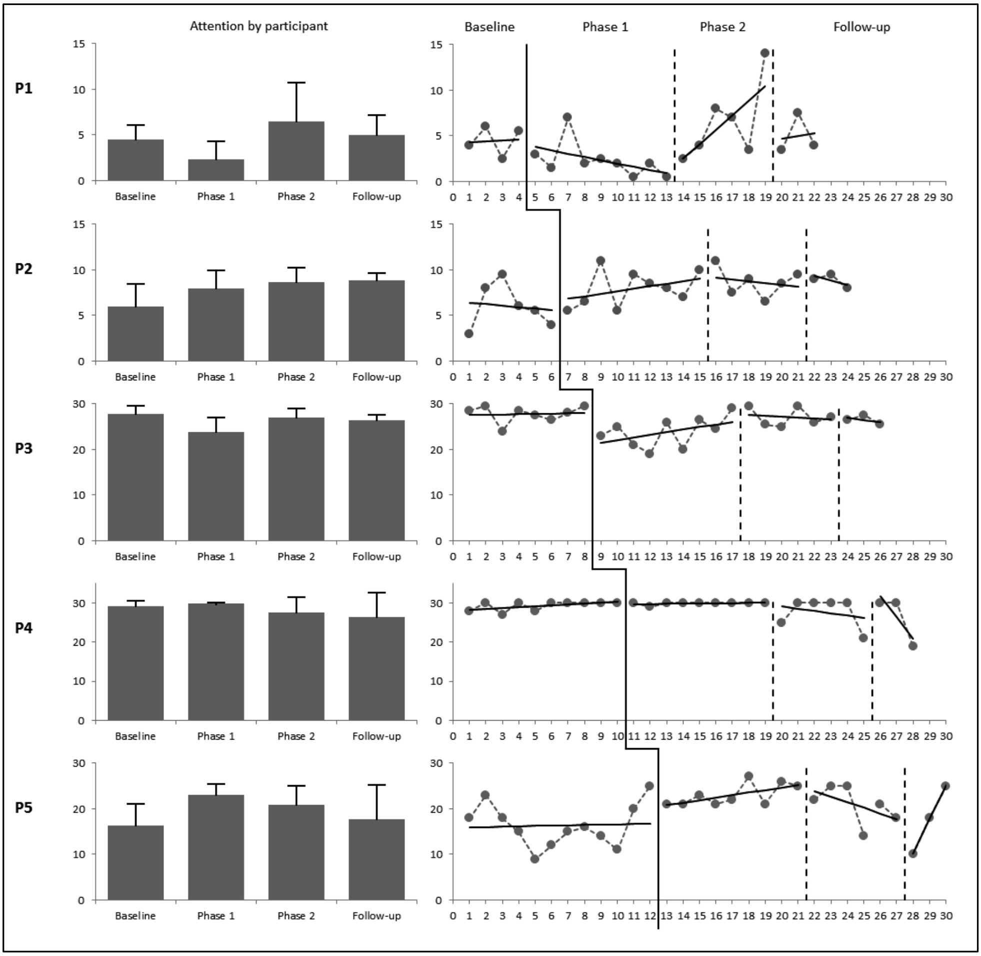

Figure 3. Attention by participant. Mean occurrence (number) across the entire phase for each of the four intervention conditions (left) and occurrence (number) during the separate observation sessions (right).

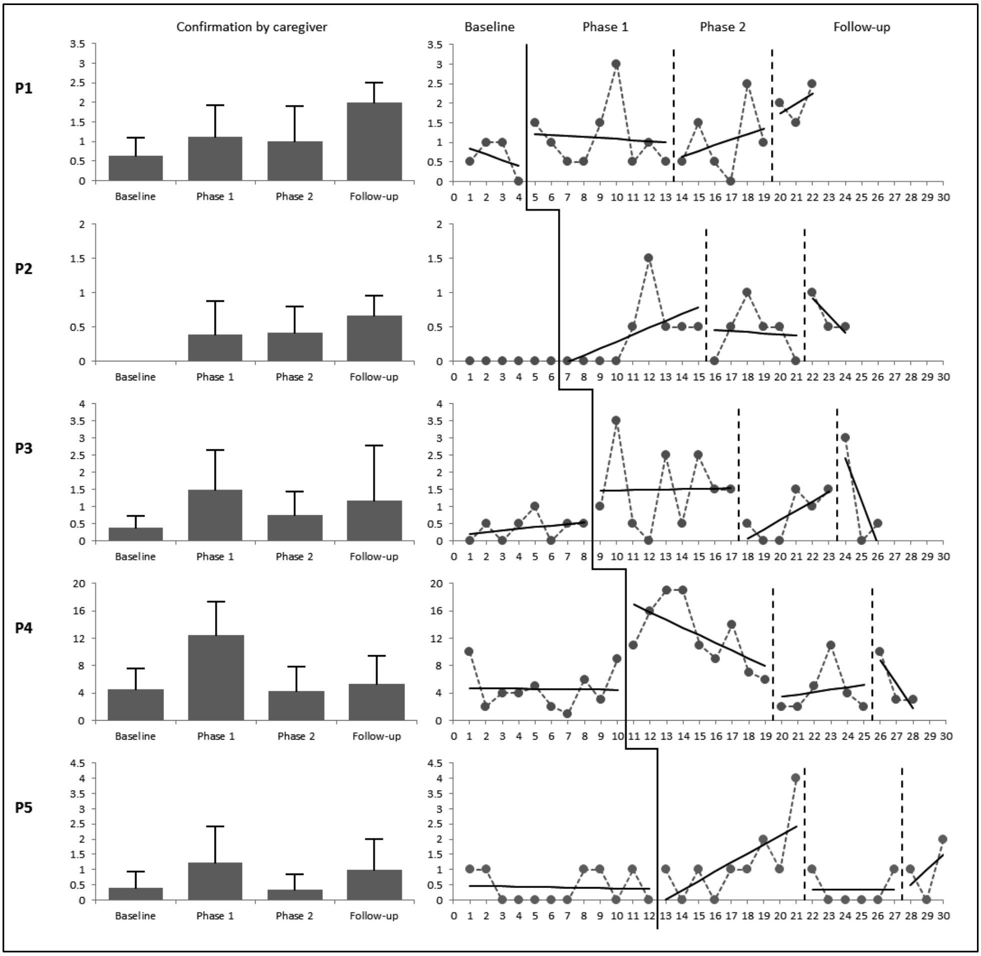

Figure 4. Confirmation by caregiver. Mean occurrence (number) across the entire phase for each of the four intervention conditions (left) and occurrence (number) during the separate observation sessions (right).

4.3.2 Effects on interaction: focus group

Table 3 presents an overview of the focus group results, including the subjective judgement of quality of interaction and ranking of the study phases, as well as the results of the content analysis. Both the scores per participant and the mean scores across participants are presented.

Subjective judgement. The mean subjective judgement scores in the right-hand column of Table 3 show that the quality of interaction was judged highest at Phase I, followed by Phase II and follow-up. The mean ranking of videos with regard to quality of interaction, in the right-hand column of Table 3, follows the same pattern.

For participants 1 and 2, the quality of interaction was subjectively judged to be highest during Phase II. For participants 3, 4, and 5, the quality of interaction was judged to be highest during Phase I. The ranking of videos with regard to quality of interaction found the same results.

Content analysis. The results of the content analysis (see Table 3) suggest that the quality of interaction was highest during Phase I, followed by Phase II. The quality of interaction was lower during follow-up than during the intervention, but higher than during baseline. Participants 1 and 2 had the best results during Phase II, whereas the best results for participants 3, 4, and 5 were found during Phase I. This corresponds with the results from the subjective judgement.

As can be seen in Table 3, participants 2 and 5 had negative scores across the study (except for Phase II for participant 2). This means that the discussion about the quality of interaction included more negative than positive characterisations.

Table 3

Focus group results for subjective judgement and content analysis

Subjective judgement: mean judgement (1=not good; 4=very good)

	P1			P2			P3			P4	P5	Mean P1-P5
	A	B	**Mean score P1**	A	B	**Mean score P2**	A	B	**Mean score P3**	**Score**	**Score**	**Score**
Baseline	1.25	1	**1.13**	1	1	**1**	1.75	2.25	**2**	**2.25**	**1**	**1.48**
Intervention Phase I	1	1.75	**1.38**	1	2	**1.50**	2	2.75	**2.38**	**3.75**	**2.25**	**2.25**
Intervention Phase II	2	2.50	**2.25**	1.50	3	**2.25**	1.25	2.75	**2**	**2.50**	**1.50**	**2.10**
Follow-up	1.25	1.75	**1.50**	1	2.25	**1.63**	1	2.50	**1.75**	**3**	**1.25**	**1.83**

Subjective judgement: mean ranking (1=highest quality of interaction; 4=lowest quality of interaction)

	P1			P2			P3			P4	P5	Mean P1-P5
	A	B	**Mean score P1**	A	B	**Mean score P2**	A	B	**Mean score P3**	**Score**	**Score**	**Score**
Baseline	2.50	4	**3.25**	4	4	**4**	2.25	3.25	**2.75**	**3.75**	**3.75**	**3.50**
Intervention Phase I	4	2.50	**3.25**	2.25	2.50	**2.38**	1.25	2.25	**1.75**	**1**	**1**	**1.88**
Intervention Phase II	1	1	**1**	1	1	**1**	3.25	1.50	**2.38**	**3.25**	**2.25**	**1.98**
Follow-up	2.50	2.50	**2.50**	2.75	2.50	**2.63**	3.25	3	**3.13**	**2**	**3**	**2.65**

Content analysis (number of positive characterisations – number of negative characterisations)

	P1			P2			P3			P4	P5	Mean P1-P5
	A	B	**Mean score P1**	A	B	**Mean score P2**	A	B	**Mean score P3**	**Score**	**Score**	**Score**
Baseline	-8	10	**1**	-26	-5	**-15.5**	-2	29	**13.5**	**40**	**-22**	**3.4**
Intervention Phase I	-7	15	**4**	-26	11	**-7.5**	5	38	**21.5**	**44**	**-1**	**12.2**
Intervention Phase II	2	28	**15**	-17	22	**2.5**	-4	45	**20.5**	**37**	**-15**	**12.0**
Follow-up	-2	12	**5**	-27	18	**-4.5**	-2	32	**15**	**37**	**-15**	**7.5**

4.3.3 Effects on bodily emotional traces

Table 4 shows the number of expressions based on a BET that were observed for the five participants. For baseline and follow-up, the absolute number is presented. For Phases I and II, the mean number of the two recordings included in the analysis is presented. Table 4 also presents the percentage of expressions based on a BET that were noticed by the caregiver.

Table 4

Mean number of expressions based on a BET and percentage recognised by the caregiver

		P1	P2	P3	P4	P5	Mean P1-P5
Baseline	Expressions	0	0	2	0	0	0.4
	Recognised (%)	-	-	0	-	-	0
Intervention Phase I	Expressions	0.5	1.5	1	0	1	0.8
	Recognised (%)	0	0	0	-	0	0
Intervention Phase II	Expressions	0.5	1	2.5	2	0	1.2
	Recognised (%)	0	0	60	75	-	50
Follow-up	Expressions	0	3	2	0	0	1
	Recognised (%)	-	0	100	-	-	40

Overall, a small number of expressions based on a BET was found across the participants and study phases (*n*=27). Across participants, the occurrence of expressions based on a BET was lowest at baseline and highest during Phase II. The differences between phases were small. Expressions based on a BET occurred for four participants during Phases I and II, for two participants during follow-up, and for one participant at baseline.

Before the start of BET training, the caregivers recognised none of the expressions. During Phase II and follow-up (after the start of BET training), caregivers recognised the expressions in 50% (6 out of 12) and 40% (2 out of 5) of the cases, respectively.

4.3.4 Social validity

Overall, caregivers positively evaluated the intervention study. Coaching caregivers with the aim of improving interaction between caregivers and people with CDB and an ID was evaluated as “good” by three caregivers and “very good” by five (possible answers: very bad/ bad/ sufficient/ good/ very good). The caregiver’s role in setting the intervention aims and choosing the activity being recorded was evaluated as “good” by seven caregivers and as “very good” by one (possible answers: very bad/ bad/ sufficient/ good/ very good).

Different parts of the intervention were evaluated as more or less well performable. For instance, affective involvement was evaluated as “difficult” by five caregivers, whereas confirmation was evaluated as “difficult” by only one caregiver. Caregivers differed in how they evaluated the difficulty of recognising expressions based on a BET, ranging from “very difficult” to “very easy” (possible answers: very difficult/ difficult/ well performable/ easy/ very easy). Individual coaching with video analysis was evaluated as “effective” by three caregivers and as “very effective” by five (possible answers: not effective at all/ not effective/ somewhat effective/ effective/ very effective). Finally, seven caregivers indicated that the change in their communicative skills towards the person with CDB and an ID was “positive”, while one caregiver evaluated that change as “somewhat positive”. Six caregivers indicated that the change in participants’ communicative skills was “somewhat positive”, while two evaluated it as “positive” (possible answers: very negative/ negative/ somewhat positive/ positive/ very positive).

4.4 Discussion

This study examined the effects of a two-phase intervention program aimed at fostering harmonious interactions and increasing the use and recognition of participant expressions based on a BET. Three research questions were formulated. With regard to the first research question – to what extent does the intervention increase the quality of interaction and, more particularly, the occurrence of attention by the caregiver, attention by the participant, confirmation by the caregiver, and affective involvement between participant and caregiver? – the occurrence of all target behaviours across participants increased during the intervention. However, some of the changes were minimal and considerable variation among participants was found.

With regard to the second research question – to what extent does the intervention increase the number of expressions based on a BET made by the participant and the recognition of these expressions by the caregiver? – the number of participant expressions based on a BET increased after the interaction training, and increased further after the BET training. Caregivers' recognition of these expressions only emerged after the BET training.

The answer to research question 3 – to what extent do the effects of the intervention endure after completion of the intervention? – is not so clear. During follow-up, overall the occurrence of the target behaviours was higher than at baseline, but lower than during the intervention phase with the highest score. The only exception is affective involvement, which increased in occurrence from intervention to follow-up for three participants.

Large variations were observed in the occurrence of attention by participant, regardless of the caregiver involved. Participant 1 had a large peak in session 19. In this session she played with a rope, which kept her attention. After throwing the rope away, she and the caregiver worked together to get it back. The caregiver gestured about it. Apparently, this situation caught her attention. No general conclusions can be drawn about the cause of the variation in the occurrence of attention by participant. It would be interesting for future studies to include more participant characteristics (i.e. temperament, interests, activities that are motivating, alertness, and time needed for regulation of intensity) to shed more light on this question.

Confirmation by the caregiver occurred more often after intervention. Although the intervention appears to be effective for increasing the occurrence of confirmation by caregiver, there is still much room for improvement (Table 2). The mean occurrence of confirmation by caregiver was remarkably high for participant 4. Across the study, the results on interaction for this participant-caregiver pair were good, which is a possible explanation for the high score on confirmation. It would be interesting to add initiatives taken by the participant as a target behaviour in future studies, since these initiatives give caregivers opportunities for confirmations.

The occurrence of affective involvement was low across the entire study, with a small improvement after intervention. However, the finding that it is possible to increase the occurrence of affective involvement is in line with the results of two earlier studies that aimed to increase affective involvement in interaction with people with CDB (Martens et al., 2014a; Martens et al., 2014b). Together, these three studies show that it is possible to improve the occurrence of affective involvement by coaching caregivers, despite the fact that very specific competences are needed for sharing emotions in the case of deafblindness.

The focus group results confirm that the intervention program led to an improvement in the quality of interaction. However, even after the intervention, the quality of interaction was subjectively judged by a focus group to be “not good” (1) and “moderate” (2) on a 4-point scale ranging from 1 (“not good”) to 4 (“very good”), after discussing the quality of interaction in general and different aspects of it (e.g. togetherness, turn taking). Also, there were still negative scores in the content analysis for participants 2 and 5 after the intervention; in these cases, the discussion about quality of interaction included more negative than positive characterisations. So, despite the increase in quality of interaction, caregivers need more coaching and training to attain a good or very good quality of interaction. Interestingly, participants 2 and 5 had the lowest mean occurrence of attention by caregiver at baseline and a negative score in the content analysis, suggesting a relationship between attention by caregiver and quality of interaction.

The results indicate that most participants regularly made expressions based on a BET. However, they also clearly indicate that caregivers require coaching to recognise these expressions: the caregivers recognised none of the participants' expressions before they received the BET training. After receiving just two 45-minute training sessions, the caregivers recognised up to half of all expressions based on a BET. However, two caregivers still did not recognise any expression based on a BET after the training. One of the caregivers who did recognise them after the training evaluated this as very difficult. The number of expressions based on a BET increased after the coaching. Caution is needed in interpreting this result, since the total number of expressions across participants and phases of the study was small. Overall, caregivers might profit from an extension of BET training, giving them more time to become familiar with the concept and to reflect upon themselves.

When looking at the overall results for the individual participants, it seems that the effect of the intervention program was highest for participants 2 and 5. A possible reason is the baseline score for these participants, leaving room for improvement. Subjective judgement of the quality of interaction by the focus group members was "not good" for these participants. The occurrence of attention by caregiver at baseline was low when compared to the other participants. The intervention program turned out to be effective for improving these scores for participants 2 and 5. Overall, according to the subjective judgement of the members of the focus group, further improvement of the interaction is possible for all participants.

4.4.1 Limitations and directions for future research

A multiple-baseline design was used to draw conclusions about the effects of the intervention program across participants. However, since the intervention program was applied to relatively few participants, the generalisability of the findings to other people with CDB and an ID is restricted. Replication of the intervention is recommended (Barlow et al., 2009).

The use of a two-phase intervention program limits the possibilities to draw conclusions regarding the exact effects of the two separate phases of the intervention (i.e. the interaction training and the BET training). Keeping this limitation in mind, the results seem to suggest that the two training phases strengthen each other. For instance, after the BET training, the occurrence of attention by caregiver and attention by participant increased. An explanation for the increase in attention by caregiver might be that the BET training teaches caregivers to be very observant, leading to more attention. An explanation for the increase in attention by participant might be that the introduction of a new element in a routine situation causes the participant to be attentive. Also, although the BET training was Phase II of the intervention program, the occurrence of participant expressions based on a BET already increased after the interaction training (Phase I). This might be because a good quality of interaction forms a good basis for the occurrence of participant expressions based on a BET (Bloeming-Wolbrink, 2007; Daelman, 2003). However, the recognition of participant expressions based on a BET did not occur until the start of the BET training. So it seems that caregivers require specific knowledge to recognise these expressions.

The results provide some evidence for the surplus value of the two-phase intervention program. The interaction training and the BET training seem to strengthen each other. However, some caution is needed since the occurrence of confirmation by caregiver decreased from Phase I to Phase II. It could be that giving confirmation is more difficult than, for example, giving attention. Combined with a difficult subject like BETs, caregivers probably need more time to internalise the use of all these elements.

Working with a focus group was added as a tool to evaluate interaction quality, next to the observation of categories of interactive behaviour. Both the coding of observation categories and the judgements of focus group members provided the same pattern of results. A valuable contribution of the focus group is that it not only provided information about the effect of the intervention program on the frequency of interactive behaviours, but also about the quality of interaction as judged by the focus group members. As the results show, an increase in the occurrence of categories of interactive behaviour does not necessarily mean that the quality of interaction is good. So the focus group results provide a very valuable context for interpreting the meaning of the coding results.

In this study, expressions were coded as expressions based on BETs when three criteria were met. One of the criteria was that the person is not paying attention to the surroundings, assuming that expressions based on BETs are neither social nor communicative when they emerge. But what if a person is very expressive and focused on other persons? It might be that this expression then develops while interacting with others. To further explore the BET concept, it would be interesting for future studies to code all expressions of thinking that are judged to refer to earlier experiences in a bodily way, also when the person is paying attention to their surroundings.

4.4.2 Implications for practice

The results of this study show that this intervention, aimed at improving caregivers' competencies and skills, had a positive effect on their interaction with people with CDB and an ID. The use of video-feedback is a powerful coaching tool, also for helping caregivers recognize expressions based on a BET. Prior to the BET training, none of the expressions based on a BET was recognized by the caregiver. After only two coaching sessions, the caregivers did recognize expressions based on a BET. This stresses the importance of coaching on the topic BETs in daily practice.

The coaching was effective and was evaluated positively by the caregivers. The fact that there is still much room for further improvement in interaction illustrates the need for continuous coaching of caregivers. This need is further stressed by the fact that the improvements after intervention were not stable; during follow-up, the occurrence of most target behaviours was higher than at baseline, but lower than during the intervention phase with the highest score. Only the results for affective involvement showed an improvement from intervention to follow-up for the majority of participants.

This study was the first to examine the effects of BET training on the use and recognition of participants' expressions based on a BET. Expressions based on a BET are bodily expressions such as touching a location on the body, making a movement, or replicating a sensation. Since expressions of persons with CDB and an ID are often idiosyncratic and subtle, and are not always used consistently, reaching qualitative communication is difficult. Recognition of expressions based on a BET is useful in the meaning-making process and in reaching qualitative communication, since recognising these expressions helps the communication partners recognise bodily, idiosyncratic expressions. When making an expression based on a BET, the intention of the person with CDB and an ID is not communicative; the expressions can be seen as inner speech. However, by confirming the expression, the communication partner might start a communicative sequence. Also, the expressions reflect which elements of activities left impressions on the person with CDB and an ID, which gives caregivers information about what activities to offer and how to offer them. Offering activities in such a way that they are interesting and meaningful to the person with CDB and an ID helps in reaching harmonious interaction.

The two-phase intervention program presented here improved interaction and helped caregivers recognise the participants' unconventional expressions. This brought sighted and hearing people one step closer to the world of people with CDB and an ID, and subsequently closer to future harmonious interaction and qualitative communication.

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Chapter 5

Summary, conclusions, and general discussion

In this thesis, we addressed the problems and possibilities in establishing harmonious interactions and subsequent qualitative communication faced by adults with congenital deafblindness (CDB) and an intellectual disability (ID) and their communication partners. Seven adults with CDB and an ID (participants), living in a residential setting for persons with an ID and a visual disability, were followed for a duration of 6;6 years while taking part in Project CHANGE, a Project with the overall aim to improve interaction and communication. Before the start of Project CHANGE, the participants did not receive deafblind education and/or the approach of the participants was not (continuously) adjusted to their deafblindness. With the start of Project CHANGE, the focus in the approach of communication partners shifted from the ID to the CDB.

Project CHANGE included two subsequent interventions aiming to improve the living conditions and social experiences of the participants. For the participants, the first intervention (Part 1 of Project CHANGE) started with a transition to new group homes specifically for persons with deafblindness, and implied pervasive changes in living conditions, and interactions with specialized caregivers. The second intervention (Part 2 of Project CHANGE) consisted of an intervention program on interaction and the use and recognition of participant expressions based on a bodily emotional trace (BET). Of the total number of seven participants in Project CHANGE, four were included in both Parts of the Project, two were included only in Part 1 of the Project, and one was included only in Part 2 of the Project.

The main aim of the research project reported in this thesis was to evaluate if it is possible to improve interaction and communication with adults with CDB and an ID. Since the participants' living conditions and life history contained many factors that could lead to early and chronic stress, for instance unpredictability and uncontrollability of daily life, early separations from the primary caregiver because of hospital visits, and communication difficulties, we were also interested in the stress physiology of the participants. The diurnal cortisol curve was assessed to collect information about this. This was done directly prior to the participants' transition to the new group homes specifically for persons with deafblindness.

In addition to a general introduction (Chapter 1), the thesis includes three empirical studies (Chapters 2, 3, and 4). In this concluding final chapter (Chapter 5), we summarize and reflect upon the results of the three empirical studies.

5.1 Summary of the three studies

Study 1. The objective of the first study (Chapter 2) was to assess the diurnal cortisol curve of six adults with CDB and an ID. All participants lived in a residential setting for persons with intellectual and visual disabilities. Here, they lived in different group homes. They did not receive (continuously) an approach adjusted to their deafblindness.

In the report of this study in Chapter 2, the various challenges adults with CDB and an ID face that may cause severe and chronic stress, are described. These include sensory deprivation, separations from primary caregivers, and difficulties in communication from birth on. Salivary cortisol can be used as a biological indicator of stress. Cortisol levels show a circadian rhythm: the diurnal cortisol curve. In healthy individuals, levels peak in the early morning and are lowest around midnight. It is explained how chronic and high stress can lead to deviancies in the diurnal cortisol curve, and how this, in turn, can have negative effects on mental and physical health. No prior research results had been published about the diurnal cortisol curve in persons with deafblindness, but our hypothesis was that the diurnal cortisol curve of adults with CDB and an ID would be deviant.

The diurnal cortisol curve of the six participants was determined by taking saliva samples five times a day during seven days within a four-week period. It was compared to the diurnal cortisol curve of a control group of 40 typical adults. We expected the participants' curve to be lower and flatter when compared to the curve of the controls. However, in spite of all past and present stress factors, the diurnal cortisol curve of the participants as a group was rather normal when compared to the controls. The individual diurnal cortisol curves appear slightly flattened for two of the participants. Possible explanations for inter-individual variation in the diurnal cortisol curves of the six participants were discussed, such as the individual’s appraisal of life events in terms of stressfulness, differences in genetic vulnerability to stress, and temperamental characteristics. The fact that the diurnal cortisol curve was rather normal, speaks to the resilience of the participants. Coping mechanisms to control stress may play a role, as well as the quality of past and present caregiving.

Finally, the limitations of the study were discussed, resulting in recommendations for future studies. One limitation was that the participants’ saliva samples were not collected at exactly the same moments as the control groups’ samples. Another limitation was the challenge in collecting enough saliva for analysis from the participants. Suggestions were made, both for future studies and medical reasons. To end with, suggestions for future research for gaining more insights in stress physiology in persons with CDB and an ID were made.

Study 2. The second study (Chapter 3) reports the effects of the first 2;2 years of Project CHANGE (Part 1) on interaction and expressive communication in six adults with CDB and an ID (participants) with no history of deafblind education and/or a (continuous) approach adjusted to the deafblindness prior to the start of this Project. The participants in this study were the same as the six participants in study 1 (Chapter 2).

Interaction with others is important for well-being and functioning. The challenges in reaching harmonious interaction and communication for persons with CDB and an ID are described, as well as the difficulties that are faced by adults with CDB who live in group homes for persons with an ID. Among these challenges are the difficulty in interpreting the interactive and communicative signals given by the person with deafblindness, and the fact that staff does not always have special knowledge of sensory impairments. The possible consequences when no special attention is given to the deafblindness are described, such as a limited development, deviant behavior or even an incorrect ID diagnosis.

Earlier studies have shown that it is possible to improve interaction with children with deafblindness or adults with intellectual and visual disabilities by training their communication partners. With our study we wanted to examine whether an improvement in living conditions could improve interaction and communication in adults with CDB and an ID who did not receive deafblind education in the past.

Part 1 of Project CHANGE involved an improvement of living conditions, including a transition to new group homes for persons with CDB and an ID, and interactions with communication partners (caregivers) who were specially trained. Prior to the participants' transition to the new group homes, the caregivers who went to work with them followed a 4-day training course on interaction and communication with a focus on CDB. Nine months after the transition the caregivers received a half-day instruction on tactile sign language. Across the 2;2 years of Part 1 of Project CHANGE the caregivers received individual coaching on-the-job, individual coaching using video analysis, and monthly video analysis with the whole team.

Video recordings and file information were used to measure interaction and communication during baseline (T0) and two subsequent periods (T1 and T2). Baseline was the 13-month period before the transition. T1 and T2 were 6-month periods starting 3 and 20 months after the transition, respectively. Interaction was examined using 5-min video fragments, coding four categories of interactive behavior: attention by the caregiver, attention by the participant, confirmation by the caregiver, and affective involvement. The level of expressive communication skills of the participants and the variety of their communicative behaviors were assessed using an adapted version of the Communication Matrix. The Communication Matrix was completed on the basis of video recordings and file information.

Overall, the results showed an improvement for the categories of interactive behavior attention by the caregiver, confirmation by the caregiver, and affective involvement, as well as for the partcipants' level of expressive communication and in the variety of their communicative behaviors. The improvements did not apply to all participants for all outcome measures. For instance, confirmation by the caregiver and affective involvement did not occur throughout the study for three and four participants, respectively. The results for attention by the participant yielded an inconsistent picture. Both improvements and decreases were found.

In the discussion section some limitations of the study are mentioned, like difficulties in determining the contribution each change in the 2;2 year period made to the results, and the limitations of measuring communication retrospectively. To end with, it is stated that the improvement over time in interaction and expressive communication is a promising result for all persons with CDB and an ID in similar settings. Recommendations for practice are made, like teaching professionals on deafblindness and its consequences for daily life, and making regular use of video analysis to improve interaction skills of caregivers and to recognize and interpret interactive and communicative expressions of the persons with CDB and an ID.

Study 3. The third study (Chapter 4) reports the effects of Part 2 of Project CHANGE, an intervention program on interaction and the use and recognition of participant expressions based on a BET. The study included five participants, all adults with CDB and an ID who did not (continuously) receive an approach adjusted to their deafblindness in the past. Four of them took part in study 1 (Chapter 2) and 2 (Chapter 3) as well, the fifth participant only took part in this study.

The report of this study starts with explaining the complexity of interaction and communication with persons with CDB and an ID. Experiencing the world through the proximal senses often results in expressions that are atypical for seeing and hearing people. Also, meeting many different communication partners limits the construction of a history of shared experiences to communicate about. It is stated that communication partners need high levels of sensitivity, special insights, and considerable skills to establish harmonious interaction and qualitative communication with persons with CDB and an ID.

The concept "expressions based on a BET" is introduced: bodily expressions of memories, like touching a location on the body, making a movement, or replicating a sensation. These expressions give insights into how a person experiences the world and reflects on earlier experiences, and are thus useful in the meaning-making process and valuable for communication.

Five adult participants with CDB and an ID and eight of their caregivers took part in a two-phase intervention program on interaction and the use and recognition of participant expressions based on a BET. The intervention program had a total duration of 15 weeks: 9 weeks for Phase I (interaction training) and 6 weeks for Phase II (BET training). Phase II started directly after Phase I. The focus of Phase I of the intervention program was on interaction. Each caregiver had three video-feedback sessions. The focus of Phase II of the intervention program was on the use and recognition of expressions based on a BET. Each caregiver had two coaching sessions during this Phase; one for transfer of theory and discussing possible interventions to evoke expressions based on a BET and one video-feedback session. The intervention was preceded by a baseline and followed by a follow-up. The study followed a multiple-baseline design across subjects.

To examine the effects of the intervention program, a 20-minute video recording was made weekly for each participant-caregiver couple during baseline and intervention. Three video recordings were made during a follow-up period: 3, 7, and 11 weeks after the end of the intervention. Target behaviors were: attention by caregiver; attention by participant; confirmation by caregiver; affective involvement; quality of interaction; participant expressions based on a BET; and participant expressions based on a BET recognised by the caregiver. Analysis was done by way of coding the presence of target behaviors in video recordings and by judging quality of interaction by a focus group.

Quantitative analyses showed, that, across participants, the occurrence of attention by caregiver and confirmation by caregiver increased during the intervention. Large variability between participants was shown for attention by participant. Affective involvement occurred not often; the highest occurrence per participant was found at intervention Phase II and at follow-up.

Expressions based on a BET occurred more after the start of Phase II of the intervention program. It is only at this point, that caregivers (2 out of 5) started to recognize participant expressions based on a BET.

In addition to the quantitative analyses, quality of interaction was also judged by the four members of a focus group (two professors in Special Needs Education, and two Master's students in Special Needs Education), based on the inspection of video recordings. They were asked to judge the quality of interaction for baseline, intervention Phase I, intervention Phase II, and follow-up. In addition, they were asked to make a ranking of quality of interaction: which recording had the highest quality of interaction, the second best, etcetera. The video recordings were offered at random to them. The results showed that either intervention Phase I or intervention Phase II was judged to have the highest quality of interaction. Quality of interaction was judged lower at follow-up than at Phase I and II of the intervention, but higher than at baseline. Focus group meetings were audiotaped and transcribed. Content analysis was performed on the transcriptions. Concepts received positive, neutral or negative characterisations, resulting in a score reflecting the judgement of quality of interaction for each fragment. The results of the content analysis suggested that quality of interaction was highest during intervention Phase I, followed by intervention Phase II, follow-up, and baseline. Two participants had negative scores across the study, meaning that during the discussion by the focus group more negative than positive characterisations were given on quality of interaction.

The intervention program was evaluated positively by the caregivers.

In the discussion section some reflections on the results were made. Also, some limitations of the study were mentioned, like the relatively small number of participants that restricts the generalisability of the findings. Directions for future research were given, followed by implications for practice. The results of the study suggest that it is possible to foster harmonious interaction and the use and recognition of expressions based on a BET in adults with CDB and an ID. However, continuous coaching of caregivers is needed, since part of the improvements after intervention were not stable. Also, the results of the focus group show that more improvement of interaction is needed. Training caregivers in recognizing expressions based on a BET is possible and useful.

5.2 Conclusions

The results of this thesis can be summarized in the following four main conclusions:

1. Despite many factors present in adults with congenital deafblindness and an intellectual disability that are likely to cause severe and chronic stress, their diurnal cortisol curve is relatively normal.
2. Offering specific living conditions and communication partners specialized in interaction and communication with persons with congenital deafblindness and an intellectual disability, leads to an improvement of interaction and expressive communication in adults with congenital deafblindness and an intellectual disability.
3. The intervention program on interaction and BETs is effective in improving interaction and the use and recognition of expressions based on a BET in adults with congenital deafblindness and an intellectual disability.
4. Despite the positive results of the interventions, further improvement of interaction and communication in adults with congenital deafblindness and an intellectual disability is needed. Continuous coaching and education of communication partners is essential.

5.3 General discussion

In addition to the discussion points addressed in the three empirical papers included in this thesis (Chapters 2, 3, and 4), some new and more general points deserve attention in this final discussion section.

5.3.1 Cortisol and experienced stress: some reflections

This thesis presents the results of a longitudinal research. Data were collected over a period of 6;6 years. Analysis of the collected data was labor-intensive. As a result, papers were published with relatively large time intervals. Our study on cortisol (Chapter 2) was published in 2012. At that time, we found only three studies that assessed the diurnal cortisol curve in persons with intellectual and/or visual disabilities. As far as we know, our study was the first to assess the diurnal cortisol curve in persons with CDB and an ID. The main conclusion from our study was that the diurnal cortisol curve of the participants with CDB and an ID was relatively normal. This was a surprising finding at the time. After 2012 a few more interesting studies have been published on cortisol in persons with a visual disability, deafblindness and/or an ID. Two of these studies (Pitchford et al., 2019; Presland et al., 2013) assessed the diurnal cortisol curve of the participants, like we did in our study. Both of these studies included participants with an ID. Pitchford et al. (2019) compared the diurnal cortisol curve of 16 adolescents with Down Syndrome with the diurnal cortisol curve of 16 adolescents with typical development. No significant differences were found. Presland et al. (2013) compared the diurnal cortisol curve of 16 adults with mild or moderate ID with published literature (e.g. Edwards, Clow, Evans, & Hucklebridge, 2001; Pruessner et al., 1997). They found that the diurnal cortisol curve of individuals with an ID was qualitatively similar to that of the typical, healthy, adult population. No studies on the diurnal cortisol curve in persons with deafblindness from 2012 on could be found in literature.

The results in both of the studies in persons with an ID are in line with our findings in persons with CDB and an ID: no large deviations in the diurnal cortisol curve were found, suggesting that there is no dysregulation of the HPA system. Given that there is substantial evidence that early and chronic exposure to high levels of stress leads to dysregulation of the HPA system (Fries, Hesse, Hellhammer, & Hellhammer, 2005; Gunnar & Vazquez, 2001), a simple conclusion could be that the reported findings indicate that, despite our expectations, persons with CDB and/or an ID do not have a history and presence of chronic exposure to high levels of stress. But is this the right conclusion to draw? Or should the question be whether assessing the diurnal cortisol curve is indeed a reliable measure for assessing stress in persons with disabilities? When it is taken for a fact that persons with CDB and/or and ID are indeed exposed to high levels of stress, both in the past and in the present, we have to wonder how much stress a person must experience before it is reflected in a disruption of the diurnal cortisol curve.

Two more recent studies on cortisol are worth mentioning, although they did not have the diurnal cortisol curve as outcome measure. De Vaan et al. (2018) measured the cortisol level of 46 participants with combined sensory and intellectual disabilities, part of whom had an Autism Spectrum Disorder (ASD), three times a day on two days: one day included a test situation, which was supposed to be stressful for the participants, the other day was a control day. No differences in cortisol levels were found between the two days, both for participants with and without ASD. A study in three children and youth with a visual and hearing disability (Nelson, Greenfield, Hyte, & Shaffer, 2013) measured, with one exception, no abnormally high cortisol levels when behavioral manifestations believed to be indicators of stress were seen. These two recent cortisol studies thus showed no abnormal cortisol reactions. Indirectly, this is in line with the findings in our and other studies on the diurnal cortisol curve. Interestingly, in addition to measuring cortisol levels, Nelson et al. (2013) and De Vaan et al. (2018) also looked at behavioral manifestations that may indicate experiencing stress. Nelson et al. (2013) found an abnormally high level of cortisol in one case for one participant. At that point in time, the participant did an attempt to sleep. De Vaan et al. (2018) found that cortisol levels were positively related to the presence of stereotyped and repetitive behaviors, suggesting that these behaviors may be stress reactions. Caution is needed in relating the findings of De Vaan et al. (2018) to persons with CDB and an ID, since their target group did not include persons with CDB.

Despite the suggested caution, it is of huge importance to consider passivity, sleeping, stereotyped behaviour, and repetitive behavior in persons with CDB and an ID as possible reflections of stress, or as coping mechanisms to deal with high stress, and to act on this. An early study in Rubella children (Van Dijk, 1982) concluded that some stereotyped behaviors, like light-gazing, were related to visual deprivation, whereas other behaviors, like rocking, were connected to a poor relationship with the environment as a result of visual problems and problems in the central nervous system. The rocking served to stimulate themselves.

Daily practice for communication partners in residential settings is that there is always a shortage of time. Whereas acting out behavior prompts to action, since it can harm the persons themselves or persons in their surroundings, behaviors like sleeping and repetitive behavior hold the risk of being overlooked. When this behavior is in fact an indicator of high stress or coping with high stress, it is harmful when it is not acted upon.

In sum: from the fact that the diurnal cortisol curve of the participants in our study and the other studies described is relatively normal, it cannot be concluded that adults with CDB and/or an ID do not have a history and presence of chronic exposure to high levels of stress. Future studies are needed to expand our knowledge, and to make it possible to act on this in the right way. Following Nelson et al. (2013) it is recommended to study the differences between persons with congenital and acquired deafblindness. When born deafblind, one does not know what life is like for a person without these disabilities. Circumstances that seem very stressful from the point of view of persons without these disabilities, like uncontrollability, sensory deprivation, and meeting many communication partners, are ‘normal’ for persons born with deafblindness, and, to some extent, for persons with an intellectual disability. Maybe, as a result, the factors expected to be stressful, are not that stressful for these persons. Contrary, when becoming deafblind across the life span, the reference point is different. It would be interesting to know if this is reflected in a deviant diurnal cortisol curve.

5.3.2 Some reflections on bodily emotional traces

In Chapter 4 the results of an intervention program on interaction and the use and recognition of participant expressions based on a BET were reported. The number of expressions based on a BET increased after intervention, and communication partners recognized more of these expressions. In addition, the intervention program was evaluated positive by the participating communication partners. This induces feelings of optimism. However, two points need consideration. First, the evaluation of the difficulty of the phase on BETs disclosed large variation across communication partners. For instance, recognizing expressions based on a BET was evaluated ranging from very easy to very difficult. A clear explanation for this is lacking at this point; both communication partner and participant characteristics may play a role. Second, two of the participating communication partners still did not recognize expressions based on a BET after the coaching. From a third communication partner it remains unknown if she was capable of recognizing expressions based on a BET after the training, since the participant she formed a pair with did not make any expression based on a BET. Caution is needed in drawing conclusions, since only a small number of expressions based on a BET occurred in the coded video recordings. But it does show that recognizing these kinds of expressions is not so easy for every communication partner or in any person with CDB and an ID.

This is in line with Nafstad and Rødbroe (2015) who mentioned that the low readability of the expressions of persons with CDB is an important concern regarding communication with them. Since most expressions come from the bodily-tactile modality, they are difficult to understand for the seeing and hearing partner (Forsgren, Daelman, & Hart, 2018). The bodily-tactile expressions are often highly idiosyncratic. Because of this, it is important to observe the way the persons with CDB explore, since this usually forms the background of the way they express themselves and is thus important for recognizing and confirming or answering expressions (Näslund & Kastrup Pedersen, 2019). According to Nafstad and Daelman (2017), a BET is embodied, and it is the basic potential sign component. This sign constructing activity, arising from the manner in which persons with CDB cognize their bodily-tactile impressions from interactions with the world, can be seen as an expression of strong language-making agency in persons with CDB (Forsgren et al., 2018). The task of the communication partner is to give access to situations where BETs can be formed, to situations where BETs can be revived, and to situations where BETs can be transformed into signs (Daelman et al., 2001). To see, understand, and recognize the traces when they are expressed by the person is a challenge (Vege & Van den Tillaart, 2015), in which the intervention program on interaction and the use and recognition of participant expressions based on a BET is helpful.

Therefore, for future research it is recommended to extend the second intervention phase and to evaluate the results of such an extension, both with regard to the effect on the number of participant expressions based on a BET made and recognized, and to the evaluation of the intervention through the communication partners. With regard to the extension of Phase II of the intervention program, the emphasize needs to be on video-feedback sessions, for communication partners to get familiar with being observant of the bodily-tactile way of experiencing the world by the persons with CDB and an ID, and to become observant of their bodily-tactile expressions. To foster this process, it is recommended to motivate communication partners to experience the world in a bodily-tactile way themselves as much as possible. This way, they become aware of the different way of experiencing things, and, as a consequence, how this can be reflected in bodily-tactile expressions. Of course, their experiences will never equal the experiences of the persons with CDB and an ID, but it can be helpful in understanding and being able to communicate in a bodily-tactile way.

5.3.3 The value of focus groups

The research described in this thesis is mainly quantitative. A qualitative measure has been added, namely the focus group in Chapter 4. This appeared to be a valuable complement. It offered a context for the interpretation of the quantitative results. Using our observational data, we found increases in various target behaviors across participants, but the focus group (whose members were asked to discuss the quality of interaction, to rank video recordings from baseline, intervention phase I, intervention phase II, and follow-up on the quality of interaction, and to give a judgement of quality of interaction for each separate video recording) learned us that this does not imply naturally that quality of interaction is good in all cases. These focus group results underscore that further improvement of the quality of interaction and communication is needed, for example by extending the intervention program on interaction and the use and recognition of participant expressions based on a BET, and/or through the development of new interventions. So, the combination of both quantitative measures and a qualitative measure like the focus group proved valuable in the interpretation of the results of our study. For future studies it is therefore recommended to add a focus group to the quantitative outcome measures used to assess interaction, communication, or other kinds of target behavior.

5.3.4 Limitations and strengths of the study

A limitation of this thesis is the small number of participants included. This has to do with the target group: adults with CDB and an ID who did not receive deafblind education and/or an approach (continuously) adjusted to their deafblindness. This group is relatively small and hard to find. The start of group homes specifically for persons with CDB and an ID formed a great opportunity to perform scientific research in this target group. The small number of participants in the three empirical studies (Chapter 2, 3, and 4) means that caution is needed in generalization of the results. However, despite the small number of participants, it is justified to state that the findings in this thesis are valuable in different respects. First, the study is innovative. Both assessing the diurnal cortisol curve in adults with CDB and an ID, and applying the intervention program on interaction and the use and recognition of participant expressions based on a BET had not been done before. Both offered new insights. It is recommended to replicate these studies with new participants, to examine if the results will be in line with our results. Second, the number of participants was small, but they were followed intensively and longitudinally. Many video recordings were made, resulting in a wealth of data on interactive and communicative behaviors of the participants and their communication partners. Within the two consecutive studies reported in Chapter 3 and Chapter 4 of this thesis, the participants were compared with themselves over time periods of up to 39 and 10 months, respectively. Of the total number of seven participants in these studies, four took part in all three studies. This opens possibilities to compare participants with themselves over longer periods of time, by combining data across studies. This is not strictly limited to the target behaviors that were looked at in this thesis, and may probably lead to additional valuable insights.

5.3.5 Implications for practice

Although in some cases small and not always consistent, the results of Project CHANGE tend to be positive. Project CHANGE had a duration of 5;5 years and included two main Parts, with the overall aim to improve interaction and communication with adults with CDB and an ID. For the participants, Part 1 of Project CHANGE involved an improvement of living conditions, including a transition to group homes specifically for persons with deafblindness, and interactions with communication partners who were specially trained (the results were reported in Chapter 3). Part 2 of Project

CHANGE consisted of an intervention program on interaction and the use and recognition of participant expressions based on a BET (the results were reported in Chapter 4).

The findings suggest that it is possible to improve interaction and communication with adults with CDB and an ID who have different ages and life histories, but who have in common that they did not have a history of deafblind education and/or an approach (continuously) adjusted to the deafblindness. In Chapter 1 (General introduction) it was said that the practical field recognizes the necessity of a specific approach for persons with CDB and an ID, but that scientific foundation for this assumption is lacking. This study contributes to the scientific foundation of the importance of a specific approach for persons with CDB and an ID.

Studies in the Netherlands and Austria revealed that combined sensory disabilities in persons with an ID remained unidentified in about 85% of the cases (Fellinger, Holzinger, Dirmhirn, & Goldberg, 2009; Meuwese-Jongejeugd et al., 2008). These studies, along with a third study, reported a prevalence of combined sensory disabilities in persons with an ID of 5 to 21.4% (Evenhuis, Theunissen, Denkers, Verschuure, & Kemme, 2001; Fellinger et al., 2009; Meuwese-Jongejeugd et al., 2008). This is quite a wide range, but it does imply that there are quite a lot of persons living in residential settings for persons with an ID who are deafblind, but whose deafblindness is not recognized and who, as a consequence, do not receive an approach that is fully adjusted to their deafblindness. How many of these persons are congenital deafblind is unknown, since the studies on prevalence did not distinguish between congenital and acquired deafblindness.

When the deafblindness of a person is not recognized and/or when the need of a specific approach is not acknowledged, the risks are numerous and huge. Developmental possibilities are limited, with deprivation as a result. Behavioral problems and passivity are among the possible consequences. Persons are either under- or over-asked.

Deafblindness frequently co-occurs with developmental disability (Dammeyer, 2011; Fellinger et al., 2009). In many cases it is hard to determine the degree of the ID, since part of the retardation might be the result of deprivation. So, there is an urge to ‘discover’ the adults with CDB in residential settings for persons with an ID. Once ‘discovered’, favourable living conditions and specialized communication partners should be offered to them. We now know that it is still possible to develop at least part of their potential in adulthood. It is uncertain if it is possible for the persons with deafblindness to develop their potential to its fullest, but this should not prevent us from striving to achieve the best result possible. All human beings have the right to develop themselves.

Within the intervention program on interaction and the use and recognition of participant expressions based on a BET (described in Chapter 4), the focus lays on BETs during only two coaching sessions of 45 minutes each. Despite this small investment, the program proved to be supportive in recognizing expressions based on a BET by communication partners. However, in chapter 5.3.2 we discussed both the large variation in evaluation of the difficulty of this part of the intervention program by the communication partners, and the fact that not all communication partners recognized expressions based on a BET after they received coaching on this. Therefore, it is recommended to implement the intervention program on interaction and the use and recognition of participant expressions of a BET in daily practice, and to extend the second Phase, thus offering more time for communication partners to get familiar with these kinds of expressions. Alongside of the implementation and extension of the intervention program on interaction and the use and recognition of expressions based on a BET, it is recommended to get communication partners acquainted with experiencing the world in a bodily-tactile way themselves.

A problem in recognizing expressions based on a BET by the communication partners is that they were not always part of the actual experience the expression refers to. Therefore, reporting among communication partners on experiences of the person with CDB and an ID, with attention for the main features in it, is of great importance. This means that communication partners need to share the experience with the person with CDB and an ID, be observant of the main features for the person with CDB and an ID, and report about it in order to inform other communication partners. Attention for these multiple tasks in coaching and education of communication partners is highly recommended.

The empirical studies show that it is possible to improve interaction, expressive communication, and the use and recognition of expressions based on a BET, but the results of the intervention program presented in Chapter 4 also make clear that part of the improvements do not hold on during follow-up. This is in line with findings in other studies that include coaching of communication partners from persons with deafblindness (e.g. Janssen, Riksen-Walraven, & Van Dijk, 2003; Martens, Janssen, Ruijssenaars, Huisman, & Riksen-Walraven, 2014a, 2014b, 2017). This implicates that educating and coaching communication partners of adults with CDB and an ID is important, but that offering this incidentally or during a short period of time is not enough for results to endure. Also, although the quantitative observational measurements of interaction showed that the intervention program on interaction and the use and recognition of expressions based on a BET led to an improvement of interaction, the qualitative outcomes of the focus group learned that this did not naturally mean that a good quality of interaction was the result.

So, both maintaining the improvements achieved, as well as a further improvement of interaction, communication and the use and recognition of expressions based on a BET, is needed. In order to achieve this, it is advised to extend the intervention program on interaction and the use and recognition of expressions based on a BET. Like it was said before, this extension needs to include the use of video-feedback sessions for the communication partners to become competent in observing the bodily-tactile way of exploring and experiencing by the person with CDB and an ID, and to become competent in recognizing expressions based on a BET. Also, much attention for experiencing the world in a bodily-tactile way by the communication partners is essential, in order for them to get some understanding of what interactive and communicative expressions might look like.

Unfortunately, to continue the frequency of individual coaching using video-feedback offered to communication partners during the intervention program (five 45-minute video-feedback sessions within a 15-week period), does not always seem feasible due to organizational circumstances in daily practice. So, it is necessary to look for other ways. Regular education and coaching of communication partners is needed for them being able to do the job, and to enable the persons with CDB and an ID to improve interaction and communication. When communication partners start the job, basic education on deafblindness is required, along with an introduction program that needs to include transferal of knowledge and coaching-on-the-job. Once communication partners are familiar with the basics in working with persons with CDB and an ID, deepening education is needed on a regular base, on a yearly basis at a minimum. In addition, it is recommended to make team video-feedback sessions a standard item in team meetings. Individual video-feedback has to be offered to communication partners, on a yearly base as a minimum.

To end with, the most important conclusion from this thesis is that it is possible to improve interaction, communication, and the use and recognition of participant expressions based on a BET in adults with CDB and an ID, even when conditions have not always been favourable to them. It is our duty to help them develop their potential, to whatever extent possible, every single day again!

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Samenvatting (Summary in Dutch)

Interactie, communicatie en stress bij volwassenen met congenitale doofblindheid en een verstandelijke beperking

Het hebben van harmonieuze interacties en communicatie met anderen is van groot belang voor onder andere het welbevinden en de ontwikkeling van mensen, maar is voor mensen met doofblindheid niet vanzelfsprekend. In dit proefschrift richten we ons op een zeer specifieke groep binnen de totale groep mensen met doofblindheid, namelijk volwassenen met congenitale doofblindheid en een verstandelijke beperking die in hun jeugd geen doofblindenonderwijs hebben gevolgd en/of voor wie de begeleiding niet (continu) was afgestemd op hun doofblindheid. We bespreken de problemen én mogelijkheden in interactie en communicatie die volwassenen met congenitale doofblindheid en een verstandelijke beperking en hun communicatiepartners tegenkomen. Zeven volwassenen met congenitale doofblindheid en een verstandelijke beperking, woonachtig bij een organisatie voor mensen met een verstandelijke en een visuele beperking, zijn gedurende 6;6 jaar gevolgd, terwijl zij deelnamen aan Project CHANGE. Het overkoepelende doel van CHANGE was het verbeteren van interactie en communicatie tussen volwassenen met congenitale doofblindheid en een verstandelijke beperking en hun communicatiepartners. Gedurende het project verschoof de focus in de begeleiding van de verstandelijke beperking naar de congenitale doofblindheid.

Project CHANGE bestond uit twee delen die gericht waren op het verbeteren van de leefomstandigheden en de sociale interacties van de deelnemers. Deel 1 van Project CHANGE startte met de verhuizing van de deelnemers naar nieuwe woningen specifiek voor mensen met doofblindheid. Dit deel van het Project omvatte grote veranderingen in de leefomstandigheden en interacties met gespecialiseerde begeleiders. Deel 2 van Project CHANGE bestond uit een interventieprogramma gericht op interactie en het maken en herkennen van “uitingen gebaseerd op een bodily emotional trace (BET)”. Uitingen gebaseerd op een BET zijn lichamelijke uitingen van herinneringen, zoals het aanraken van een plek op het lichaam, het maken van een beweging, of het nabootsen van een sensatie. Het herkennen van deze uitingen is belangrijk met het oog op betekenisverlening en daarmee waardevol voor de communicatie, omdat deze uitingen inzicht bieden in de wijze waarop een persoon de wereld ervaart en reflecteert op eerdere ervaringen.

Van de zeven deelnemers aan Project CHANGE hebben vier aan zowel Deel 1 als Deel 2 van het Project meegedaan, twee deelnemers deden alleen aan Deel 1 mee en één deelnemer deed alleen mee aan Deel 2.

Het hoofddoel van het onderzoek dat wordt beschreven in dit proefschrift is onderzoeken of het mogelijk is om interactie en communicatie te verbeteren met volwassenen met congenitale doofblindheid en een verstandelijke beperking bij wie de begeleiding niet (continu) afgestemd is geweest op de doofblindheid. De leefomstandigheden en levensgeschiedenis van de deelnemers bevatten veel factoren die kunnen leiden tot vroege en chronische stress, zoals onvoorspelbaarheid en gebrek aan controle in het dagelijks leven, vroege scheidingen van de primaire verzorgers als gevolg van ziekenhuisopnames, en problemen in de communicatie. Om deze reden waren we ook geïnteresseerd in de stressfysiologie van de deelnemers. De cortisol curve van de deelnemers over de dag heen is gemeten om hierover informatie te verzamelen. Dit is gedaan direct voorafgaand aan de verhuizing van de deelnemers naar de nieuwe woningen specifiek voor mensen met doofblindheid.

Naast de algemene inleiding (Hoofdstuk 1) bestaat het proefschrift uit drie empirische studies (Hoofdstuk 2, 3 en 4). In het afsluitende hoofdstuk (Hoofdstuk 5) worden de resultaten van de drie studies samengevat en wordt gereflecteerd op de resultaten.

Samenvatting van de drie studies

Studie 1. Het doel van de eerste studie (Hoofdstuk 2) was het bepalen van de cortisol curve van zes volwassenen met congenitale doofblindheid en een verstandelijke beperking. De deelnemers woonden in verschillende woningen bij een organisatie voor mensen met een verstandelijke en een visuele beperking.

In de beschrijving van deze studie worden de verschillende factoren genoemd die kunnen leiden tot ernstige en chronische stress bij volwassenen met congenitale doofblindheid en een verstandelijke beperking. Het gaat hierbij onder andere om zintuiglijke deprivatie, scheiding van primaire verzorgers en problemen in de communicatie vanaf de geboorte. Cortisol in speeksel kan worden gebruikt als biologische indicator van stress. Cortisol volgt een circadiaan ritme: de cortisol curve over de dag heen. Normaal gesproken zijn de waarden vroeg in de ochtend het hoogste en rond middernacht het laagste. Chronische en ernstige stress kan leiden tot afwijkingen in de cortisol curve en dit kan vervolgens negatieve effecten hebben op de psychische en fysieke gezondheid. Ten tijde van de studie was er nog niet gepubliceerd over de cortisol curve van mensen met doofblindheid. Onze hypothese was dat de cortisol curve van mensen met congenitale doofblindheid en een verstandelijke beperking afwijkingen zou vertonen.

De cortisol curve van de zes deelnemers werd bepaald door vijf keer per dag gedurende zeven dagen binnen een periode van vier weken een speekselmonster af te nemen. De cortisol curve van de deelnemers werd vergeleken met de cortisol curve van een controlegroep van 40 personen. De verwachting was dat de cortisol curve van de deelnemers met congenitale doofblindheid en een verstandelijke beperking lager en vlakker zou zijn in vergelijking met de cortisol curve van de controlegroep. Echter, ondanks alle aanwezige factoren die in het heden en verleden zouden hebben kunnen leiden tot chronische en ernstige stress, bleek de cortisol curve van de deelnemers als groep relatief normaal, vergeleken met de cortisol curve van de controlegroep. Bij de individuele cortisol curve van de deelnemers valt op dat deze voor twee van hen enigszins vlak lijkt. Mogelijke verklaringen voor de variatie tussen de deelnemers zijn besproken, zoals de wijze waarop de persoon gebeurtenissen beleeft in termen van stressvolheid, verschillen in genetische kwetsbaarheid voor stress en karaktereigenschappen. Het feit dat de cortisol curve van de deelnemers als groep vrij normaal bleek te zijn, zegt iets over de veerkracht van de deelnemers. Coping-mechanismen voor de omgang met stress zouden een rol kunnen spelen, evenals de kwaliteit van de huidige en vroegere ontvangen zorg.

Tenslotte zijn de beperkingen van de studie besproken, wat resulteert in aanbevelingen voor toekomstige studies. Eén van de beperkingen was dat de speekselmonsters van de deelnemers niet op exact dezelfde momenten zijn verzameld als de speekselmonsters van de controlegroep. Een andere beperking was de moeilijkheid om voldoende speeksel te verzamelen om analyse mogelijk te maken. Hiervoor zijn suggesties gedaan, zowel voor toekomstig wetenschappelijk onderzoek als voor medisch onderzoek. Tenslotte zijn suggesties voor toekomstig onderzoek gedaan om meer inzicht te krijgen in de stressfysiologie bij mensen met congenitale doofblindheid en een verstandelijke beperking.

Studie 2. Zoals eerder aangegeven bestond Project CHANGE uit 2 delen. Studie 2 (Hoofdstuk 3) richt zich op Deel 1 van het Project. Deel 1 van Project CHANGE kende een duur van 2;2 jaar en omvatte voor de deelnemers grote veranderingen in de leefomstandigheden en interacties met gespecialiseerde begeleiders. De effecten hiervan op interactie en expressieve communicatie bij zes volwassenen met congenitale doofblindheid en een verstandelijke beperking, die voorafgaand aan de start van Project CHANGE geen doofblindenonderwijs hebben gevolgd en/of voor wie de begeleiding niet (continu) was afgestemd op de doofblindheid, worden beschreven. De deelnemers in deze studie waren dezelfde als de zes deelnemers in de eerste studie (Hoofdstuk 2).

Interactie met anderen is belangrijk voor het welzijn en het functioneren van mensen. In de beschrijving van deze studie worden de uitdagingen genoemd in het bereiken van harmonieuze interactie en communicatie voor mensen met congenitale doofblindheid en een verstandelijke beperking, evenals de moeilijkheden die volwassenen met congenitale doofblindheid ervaren wanneer zij wonen in een setting die voornamelijk gericht is op mensen met een verstandelijke beperking. Enkele van deze uitdagingen zijn het feit dat medewerkers niet altijd specifieke kennis hebben over zintuiglijke beperkingen en dat het moeilijk is om interactieve en communicatieve uitingen van personen met doofblindheid te interpreteren. De mogelijke gevolgen wanneer de benadering niet wordt aangepast aan de doofblindheid worden beschreven, zoals een beperkte ontwikkeling, afwijkend gedrag en zelfs het ten onrechte diagnosticeren van een verstandelijke beperking.

Eerdere studies hebben aangetoond dat het mogelijk is om de interactie met kinderen met doofblindheid of volwassenen met een verstandelijke en een visuele beperking te verbeteren door het scholen van hun communicatiepartners. Met onze studie wilden we onderzoeken of een verbetering van leefomstandigheden zou leiden tot een verbetering van de interactie en communicatie bij volwassenen met congenitale doofblindheid en een verstandelijke beperking die in het verleden geen doofblindenonderwijs hebben gevolgd.

Deel 1 van Project CHANGE bestond uit een verbetering van de leefomstandigheden. Dit hield onder andere in dat de deelnemers verhuisden naar nieuwe woningen specifiek voor mensen met congenitale doofblindheid en een verstandelijke beperking en dat zij interacties hadden met gespecialiseerde communicatiepartners (begeleiders). Voordat de deelnemers verhuisden naar de nieuwe woningen, volgden de begeleiders die met hen gingen werken een vierdaagse cursus over interactie en communicatie, waarbij de focus lag op congenitale doofblindheid. Negen maanden na de verhuizing volgden de begeleiders een scholing van een halve dag over tactiele gebarentaal. Gedurende de duur van 2;2 jaar van Deel 1 van Project CHANGE kregen de begeleiders individuele coaching on-the-job, individuele coaching met behulp van video analyse en maandelijks video analyse met het hele team.

Om interactie en communicatie te meten tijdens de baseline (T0) en twee daaropvolgende periodes (T1 en T2), werd gebruik gemaakt van video opnames en dossierinformatie. De baseline was de periode van 13 maanden voorafgaand aan de verhuizing van de deelnemers. T1 en T2 waren periodes van 6 maanden; T1 begon 3 maanden na de verhuizing van de deelnemers, T2 begon 20 maanden na de verhuizing van de deelnemers. Om interactie te onderzoeken werden videofragmenten van 5 minuten gebruikt, waarbij vier interactiecategorieën werden gecodeerd: aandacht van de begeleider, aandacht van de deelnemer, bevestiging door de begeleider en affectieve betrokkenheid. Om het niveau van expressieve communicatievaardigheden van de deelnemers en de variatie in hun communicatieve gedragingen te bepalen, werd een aangepaste versie van de Communication Matrix gebruikt. De Communication Matrix is een instrument waarmee de expressieve communicatie in kaart wordt gebracht voor de functies iets weigeren, iets verkrijgen, sociale interactie en informatie. De communicatie wordt onderverdeeld in 7 niveaus, gebaseerd op de taalontwikkeling bij normaal functionerende kinderen tot een leeftijd van 2 jaar. De Communication Matrix kan gebruikt worden bij volwassenen. De Communication Matrix werd ingevuld op basis van video opnames en dossierinformatie.

De resultaten laten een verbetering zien voor de interactiecategorieën aandacht van de begeleider, bevestiging door de begeleider en affectieve betrokkenheid, evenals voor het niveau van expressieve communicatie van de deelnemers en de variatie in hun communicatieve gedragingen. Deze verbeteringen gelden voor de groep als geheel, maar dit betekent niet dat er voor iedere deelnemer voor elk van deze categorieën sprake is van een verbetering. Een voorbeeld hiervan is de interactiecategorie bevestiging door de begeleider; bij drie deelnemers werd gedurende de gehele studie geen bevestiging door de begeleider gezien. De resultaten voor aandacht van de deelnemer lieten een inconsistent beeld zien. Zowel verbeteringen als afnames werden gemeten.

In de discussie zijn enkele beperkingen van de studie besproken, zoals de onmogelijkheid om de bijdrage van iedere individuele verandering in de periode van 2;2 jaar van Deel 1 van Project CHANGE op de resultaten te bepalen, en de beperkingen die het achteraf meten van communicatie met zich meebrengt. Er is aangegeven dat de verbetering in interactie en expressieve communicatie een veelbelovend resultaat is voor alle personen met congenitale doofblindheid en een verstandelijke beperking in vergelijkbare omstandigheden. Tenslotte zijn aanbevelingen voor de praktijk gedaan. Eén van de aanbevelingen betreft het scholen van professionals in kennis over doofblindheid en de gevolgen hiervan voor het dagelijks leven. Een andere aanbeveling betreft het regelmatig gebruik maken van video analyse om de interactie vaardigheden van begeleiders te verbeteren en om interactieve en communicatieve uitingen van personen met congenitale doofblindheid en een verstandelijke beperking te leren herkennen en interpreteren.

Studie 3. De derde studie (Hoofdstuk 4) rapporteert de effecten van Deel 2 van Project CHANGE, een interventieprogramma gericht op interactie en het maken en herkennen van uitingen gebaseerd op een bodily emotional trace (BET). Vijf deelnemers, allen volwassenen met congenitale doofblindheid en een verstandelijke beperking voor wie de benadering in het verleden niet (continu) was afgestemd op de doofblindheid, namen deel aan de studie. Vier van hen namen ook deel aan studie 1 (Hoofdstuk 2) en aan studie 2 (Hoofdstuk 3), de vijfde deelnemer nam alleen deel aan deze studie.

De rapportage van deze studie begint met een uitleg van de complexiteit van interactie en communicatie met personen met congenitale doofblindheid en een verstandelijke beperking. Het ervaren van de wereld middels de nabijheidszintuigen resulteert vaak in uitingen die atypisch zijn voor personen die zien en horen. Daarnaast beperkt de omgang met veel verschillende communicatiepartners de opbouw van een geschiedenis van gedeelde ervaringen om over te kunnen communiceren. Aangegeven wordt dat communicatiepartners een hoge mate van sensitiviteit, speciale inzichten en aanzienlijke vaardigheden nodig hebben om harmonieuze interactie en goede communicatie met personen met congenitale doofblindheid en een verstandelijke beperking te bewerkstelligen. Het concept “uitingen gebaseerd op een BET” is geïntroduceerd: lichamelijke uitingen van herinneringen, zoals het aanraken van een plek op het lichaam, het maken van een beweging, of het nabootsen van een sensatie. Deze uitingen bieden inzicht in de wijze waarop een persoon de wereld ervaart en reflecteert op eerdere ervaringen, zijn van belang in het proces van betekenisverlening en zijn daarmee waardevol voor communicatie.

Het doel van onze studie was het onderzoeken van de effecten van een interventieprogramma dat gericht was op het verbeteren van de interactie en het maken en het herkennen van uitingen gebaseerd op een BET van de deelnemers. Het interventieprogramma was gericht op communicatiepartners die werken met volwassenen met congenitale doofblindheid en een verstandelijke beperking.

Vijf deelnemers en acht van hun begeleiders namen deel aan het interventieprogramma. Het interventieprogramma bestond uit twee fasen en had een totale duur van 15 weken: negen weken voor Fase I (interactie training) en zes weken voor Fase II (BET training). Fase II volgde direct op Fase I. In Fase I van het interventieprogramma lag de focus op interactie. Iedere begeleider had drie coaching sessies met video analyse tijdens deze fase. De focus in Fase II van het interventieprogramma lag op uitingen gebaseerd op een BET. Iedere begeleider had twee coaching sessies tijdens deze fase: één voor de overdracht van theorie en het bespreken van mogelijke interventies die zouden kunnen leiden tot het maken van uitingen gebaseerd op een BET, en één sessie met video analyse. De interventie werd voorafgegaan door een baseline en gevolgd door een follow-up. In de studie werd een multiple baseline design gebruikt.

Om de effecten van het interventieprogramma te onderzoeken werd tijdens de baseline en interventie iedere week een video opname van 20 minuten gemaakt van ieder koppel deelnemer-begeleider. Tijdens de follow-up werden nog eens drie video opnames gemaakt: drie, zeven en elf weken na het einde van de interventie. Doelgedragingen waren: aandacht van de begeleider; aandacht van de deelnemer; bevestiging door de begeleider; affectieve betrokkenheid; kwaliteit van interactie; uitingen gebaseerd op een BET van de deelnemer; het herkennen van uitingen gebaseerd op een BET van de deelnemer door de begeleider. De frequentie van de doelgedragingen in de video opnames werd gecodeerd en de kwaliteit van interactie werd beoordeeld door een focusgroep.

Kwantitatieve analyses lieten, over alle deelnemers heen, een toename zien in aandacht van de begeleider en bevestigen door de begeleider. Voor aandacht van de deelnemer waren de verschillen tussen de deelnemers aanzienlijk. Affectieve betrokkenheid kwam weinig voor; bekeken per deelnemer werd dit het meest gezien tijdens Fase II van de interventie en tijdens de follow-up.

Uitingen gebaseerd op een BET kwamen vaker voor na de start van Fase II van het interventieprogramma. Pas vanaf dat moment kwam herkenning van deze uitingen door begeleiders voor (bij twee van de vijf begeleiders).

In aanvulling op de kwantitatieve analyses werd de kwaliteit van interactie beoordeeld door de vier leden van een focusgroep (twee hoogleraren in de orthopedagogiek en twee master studenten orthopedagogiek), waarbij zij zich baseerden op video opnames. Er werd hen gevraagd om de kwaliteit van interactie te beoordelen voor de baseline, interventie Fase I, interventie Fase II en follow-up. Daarnaast werd hen gevraagd om een rangorde aan te brengen in de kwaliteit van interactie: welke opname kende de hoogste kwaliteit van interactie, welke daarna, etc. De video opnames werden in willekeurige volgorde aangeboden. De resultaten lieten zien dat de kwaliteit van interactie het hoogste werd beoordeeld in interventie Fase I of interventie Fase II. De kwaliteit van interactie in follow-up werd lager beoordeeld dan in Fase I en Fase II van de interventie, maar hoger dan in de baseline. De bijeenkomsten van de focusgroep werden opgenomen (audio) en uitgeschreven. Content analyse werd uitgevoerd op de transcripties. Concepten werden positief, neutraal of negatief gekarakteriseerd, wat resulteerde in een score die de beoordeling van de kwaliteit van interactie voor ieder fragment weergeeft. De resultaten van de content analyse suggereerden dat de kwaliteit van interactie het hoogste was tijdens Fase I van de interventie, gevolgd door respectievelijk interventie Fase II, follow-up en de baseline. Twee deelnemers hadden negatieve scores gedurende de studie, wat inhoudt dat in de focusgroep de kwaliteit van interactie meer negatief dan positief werd gekarakteriseerd.

Door de begeleiders werd het interventieprogramma positief geëvalueerd.

In de discussie van Hoofdstuk 4 wordt gereflecteerd op de resultaten. Daarnaast worden enkele beperkingen van de studie besproken, zoals het relatief kleine aantal deelnemers wat de mogelijkheden om de bevindingen te generaliseren beperkt. Er worden suggesties gedaan voor toekomstig onderzoek, gevolgd door aanbevelingen voor de praktijk. De resultaten van de studie duiden erop dat het mogelijk is om harmonieuze interacties en het maken en herkennen van uitingen gebaseerd op een BET te verbeteren bij volwassenen met congenitale doofblindheid en een verstandelijke beperking. Echter, continue coaching van begeleiders is nodig, want een deel van de verbeteringen na de interventie bleken niet stabiel. Daarnaast laten de resultaten van de focusgroep zien dat een verdere verbetering van interactie nodig is. Het scholen van begeleiders in het herkennen van uitingen gebaseerd op een BET is mogelijk en zinvol.

Conclusies

De resultaten van dit proefschrift kunnen worden samengevat in de volgende vier hoofdconclusies:

1. Ondanks de aanwezigheid van meerdere factoren die kunnen leiden tot ernstige en chronische stress bij volwassenen met congenitale doofblindheid en een verstandelijke beperking, is hun cortisol curve relatief normaal.
2. Het bieden van specifieke leefomstandigheden en communicatiepartners die gespecialiseerd zijn in interactie en communicatie met mensen met congenitale doofblindheid en een verstandelijke beperking, leidt tot een verbetering van interactie en expressieve communicatie bij volwassenen met congenitale doofblindheid en een verstandelijke beperking.
3. Het interventieprogramma 'Interactie en BETs' is effectief gebleken in het verbeteren van interactie en het maken en herkennen van uitingen gebaseerd op een BET bij volwassenen met congenitale doofblindheid en een verstandelijke beperking.

4. Ondanks de positieve resultaten van de interventies is een verdere verbetering van interactie en communicatie bij volwassenen met congenitale doofblindheid en een verstandelijke beperking nodig. Continue coaching en scholing van communicatiepartners is essentieel.

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Curriculum Vitae

Kitty Bloeming-Wolbrink werd op 2 februari 1975 geboren te Groningen. Na het behalen van haar diploma orthopedagogiek aan de Rijksuniversiteit Groningen ging ze in 1998 als groepsleidster werken met mensen met een verstandelijke beperking bij Nieuw Woelwijck te Sappemeer. In 2001 ging zij als groepsleidster, en later als zorgcoördinator, werken met mensen met een verstandelijke en een visuele beperking bij De Brink, wat later Koninklijke Visio ging heten. Vanaf 2002 combineerde zij het werken als groepsleidster met de functie van ambulant begeleider. In 2007 studeerde zij cum laude af aan de Rijksuniversiteit Groningen binnen de internationale master 'Communication and congenital deafblindness'. Na afronding van deze opleiding is zij gaan werken als orthopedagoog. In 2009 is zij gestart met haar promotieonderzoek 'Interactie, communicatie en stress bij volwassenen met congenitale doofblindheid en een verstandelijke beperking'. Het werken aan het promotieonderzoek heeft zij gedurende de gehele periode gecombineerd met de functie van orthopedagoog bij Koninklijke Visio.

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