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## Research Report

# Video feedback intervention: a case series in the context of childhood hearing impairment

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### Abstract

*Background:* Recent research shows that parental sensitivity can explain a significant and unique amount of growth in speech and language outcomes in children with cochlear implants. In this intervention study we explored the impact of an intervention designed to support parental sensitivity on children's communication development.

*Aims:* This study tests the effect of a complex intervention in the context of childhood hearing impairment using a case study design of three families. Propositions for each case were made using parental report of the child's development in an attempt to identify change in outcome measurements that were not likely to be due to general development in the child or a halo effect from the intervention.

*Methods and Results:* Multiple pre- and post-intervention measures were taken. Outcome measures were mother-child contingencies to vocal utterances, emotional availability and an assessment of early communication in the child. Results for each case showed that improvements in some outcome measurements were found after the intervention and were maintained at follow-up.

*Conclusions & Implications:* Taking account of developmental change in intervention studies with children is challenging. Single-subject intervention studies can be designed to allow research interventions to be tailored to meet families' specific needs. Video interaction guidance may support pre-linguistic communicative development in children with hearing impairment.

*Keywords:* mother-child interaction, contingency, video feedback, childhood deafness, speech development, intervention design.

### What this paper adds?

This paper describes the first research findings from the application of video interaction guidance in the context of childhood hearing impairment. The findings suggest that the intervention is acceptable to parents prior to the implant, immediately post implant and after one year of implantation.

### Introduction

This paper reports the findings of an intervention study in the context of childhood hearing impairment with a series of three cases. The intervention study was designed to provide support to parents who had hearing-impaired children at the pre-linguistic stage of language

development. The trial was designed to test theoretical propositions about the centrality of parental responsiveness in the scaffolding of early speech and language development in the child. Basic scientific knowledge from speech science highlights the role of parental contingency in the development of pre-linguistic speech

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skills (Goldstein and Schwade 2008) and psychological science highlights the role of parental attunement and scaffolding in general child development (Vygotsky 1978). Contingency of parental behaviour can be understood as the sequence and timing of parent's behaviours within which the parent responds to the child's cues. It is considered to be a property of a wider parenting construct referred to as maternal responsiveness or maternal sensitivity (Shin *et al.* 2006). The role of contingency of parental behaviour has a strong presence in developmental psychological science (Jaffe *et al.* 2001). In relation to speech science, Goldstein and colleagues have a programme of experimental speech science exploring the impact of parental contingency on the development of infant babbling (Goldstein and Schwade 2008). On a more practical footing, the ecological validity of the concept of parental contingency is demonstrated by its increasing use to evaluate generic parent intervention programmes (Rodriguez *et al.* 2010). Basic science from health services research shows that the provision of family-centred services is associated with more parental satisfaction with services (King *et al.* 2004). The involvement of families in services has been highlighted as desirable in the field of childhood hearing impairment (DesJardin 2003, Moeller 2000). Author James proposed (2011a) that in the context of children with speech and language needs, working on family-centred goals in an intervention could enhance the likelihood of parents finding the intervention more satisfactory. Together, these concepts from psychological science, speech science and health services research informed the design of the intervention study which was, family-centred, being tailored around the families' goals for change, and designed to enhance attunement and responsiveness of the parent to the child's communicative initiatives. The three cases reported on in this paper were purposefully selected to demonstrate their maximal diversity in relation to the timing of their child's cochlear implantation (pre-implant, immediately post-implant, over 1 year post-implant).

Parental sensitivity underpins the quality of the parent-child interactions and these early conversations are causally related to the child's development (Crittenden and Bonvillian 1984). Maternal sensitivity has been described by Shin *et al.* (2008) as comprising four attributes of the mother: (1) her ability to respond dynamically to the infant's needs; (2) reciprocal behaviour where the mother adapts her responses so that the exchanges between the mother and infant are mutually satisfying; (3) contingent on the infant's behaviour, the mother sets up patterns of association or causation for the infant by consistently responding to the infant's cues in ways from which the child is able to learn; and (2) the overall quality of the maternal behaviour shows adaptability to the child's expression of its internal state. Maternal sensitiv-

ity is vulnerable to a range of psychological and social factors including maternal stress, depression and anxiety (Shin *et al.* 2008). However, according to self-efficacy theory (Bandura 1986), the parent who believes he/she is effective in their parenting role will be more successful in it, and this may override the impact of stress which has been associated with parenting a child with hearing impairment (Hintmier 2006). Working through parents to enhance children's language development has been normalized in UK practice (James 2011a) largely through the embedding of the Hanen Parent Programme (HPP) (Pepper and Weitzman 2004). This routine uptake of parent-child interaction therapy has not been based on particularly strong evidence; for example, there is no meta-analysis to demonstrate the efficacy of the intervention compared with other interventions. However, despite this, the practice of working through parents to support child language development is routine in speech and language therapy clinics in the UK and it has been the subject of research enquiry in the field of childhood hearing impairment.

DesJardin and Eisenberg's (2007) exploration of the relationship between parental involvement, self-efficacy and quality of parental language sought to understand the impact of parental reciprocity in their study of speech and language in children with cochlear implants. In this study the experimenters looked for correlations between standardized questionnaires of self-efficacy and numerically derived summaries of child language behaviour (i.e. the average number of words the child produced in an utterance) and maternal strategies thought to scaffold child language (such as talking in parallel to what the child is doing or playing with). They failed to find a relationship between their self-rated measure of self-efficacy and maternal interactional input to the child. However, there are a number of reasons why this study was not a good test of the link between self-efficacy, linguistic behaviour of the mother and language development of the child. Firstly, the children in the study had an average age of 4 years 9 months. It is possible that at this age the associative link between maternal sensitivity and linguistic skills in the child is difficult to find and that by this time in development the adaptations of the mother's behaviour to the hearing impairment had changed the nature of the associations that might typically be predicted between maternal sensitivity and child language. Secondly, the basic design of the study was correlational so the data from the mother-child dyads were separated and averaged in to two groups (mothers and children). We would argue that the contingent patterned response within a mother-child dyad is a critical mechanism for development and therefore the analysis should be based on patterns in timing and sequence of behaviour between mother and child.

## Experimental design

In this study we used a case series design with multiple pre- and post-assessments to explore the efficacy of a psychological intervention that has a strong evidence base in the psychological literature with capacity to produce effects on all aspects of child development (see below). Ideally we wanted to use an *N*-of-1 design with multiple pre- and post-baseline measures; however, given the exploratory nature of the trial, the timelines in which the project had to be completed and the acceptability of attending the research laboratory for multiple assessment visits, this design was not feasible. We attempted to understand the significance of any change arising after the intervention in the context of the child's overall development by taking measures of the child's general development. In addition, enhanced outcomes after intervention could be due to a halo effect, i.e. the positive enhancement of uncontrolled novelty. Attributing enhanced post-intervention outcome to the specific intervention might be an error since the enhancement could be simply due to being involved in a research programme. The halo effect, if present, is thought to decay rapidly over time and account for less than 1% of variance in an effect after 8 weeks (Clark and Sugrue 1991). Follow-up assessments that occur at least 8 weeks after the intervention will avoid miss-attribution of a change in baseline measures to a generalized halo effect.

### *The intervention*

The intervention used in the current study, Video Interaction Guidance (VIG), has an established evidence base in the general field of paediatrics (Fukkink 2008). It significantly enhances parental attitude, parental behaviour and child development in families where there are significant attachment difficulties (Juffer *et al.* 2005) and it does this in three to four sessions. In the video-based intervention the practitioner takes a brief (15–20 min) video film of a typical interaction between the mother and child. The practitioner then analyses the film using a framework based on a set of principles of the behaviours that support attunement. From this analysis, the practitioner selects a few brief clips to present to the parent and they discuss and reflect upon these clips. The use of video technology within parent interventions is not especially new, but what is unique about this approach is that it only draws attention to successful elements of communication, and promotes reflection on why these elements were successful. The parents specify the goals for the intervention work and this goal helps the practitioner decide how to make the films and which clips to select (Kennedy *et al.* 2011). To date, there has been a descriptive case study of the approach within the context of childhood hearing impairment (James 2011b).

### *Outcome measures*

During assessment of the quality of parent–child interactions, it is important to consider the bidirectional influence that both members of the dyad exert on one another. The Emotional Availability Scales (Biringen 2008) provide a holistic measure of the dynamic relational synchronicity between a mother and child, and have been used to examine differences in interactions with typically and atypically developing infants (e.g. Lam and Kitamura 2010, Pressman *et al.* 1999). Studies with hearing-impaired children report that hearing-impaired infants show reduced responsiveness and involvement of their hearing parents during social interactions when compared with normal hearing peers with hearing parents (Lam and Kitamura 2010, Pressman *et al.* 1998). Furthermore, hearing mothers with hearing-impaired children have been identified as being less able to regulate negative emotions and are more likely to provide added structure to their child's play compared with hearing parents with hearing children (Pipp-Siegel *et al.* 1998). There is research evidence that the Emotional Availability Scales are sensitive to changes in the parent–child emotional connection following an intervention (Biringen *et al.* 2012), and they provide a reliable measure in the context of childhood hearing impairment.

The Tait assessment was developed to assess pre-verbal communication in children with cochlear implants (Tait *et al.* 2007) and it has shown to be predictive of later speech and language development (Tait and Lutman 1997, Tait *et al.* 2001a). The Tait assessment provides a definition for coding the child's gestural and vocal turns, autonomy in communication, and also codes to describe turn transitions and turn responses. Autonomy was defined by Tait as when the child's initiative changes the predictable course of events (Tait *et al.* 2001a). This could be when the child changes the focus on the communication or the play by introducing a new item or idea using gesture, vocalization or a mixture of the two. The presence of autonomy during pre-linguistic development has been shown to be highly predictive of later speech and language development (Tait and Lutman 1997) in deaf children. The Tait assessment has been shown to be reliable with high inter- and intra-judge agreement on selected interaction episodes (Tait 1993). The predictive power and the reliability of the coding framework makes it a good selection as an outcome measure; however, the focus only on the child's behaviour during interaction shows that the fundamental premise of the measure, whilst related topically, is not theoretically closely aligned to the theoretical premise of the current intervention study where the reciprocal contingency between mother–child is considered the foundation for later learning.

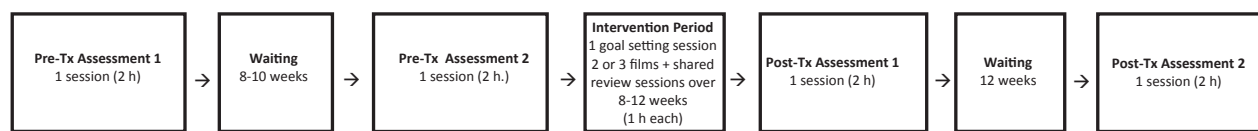


Figure 1. The study design timeline from pre-treatment (Tx) 1 to post-Tx 2, all laboratory assessments and intervention sessions were interspersed with waiting periods. Assessments were conducted at each assessment session; free play; Tait communicative analysis; emotional availability; contingency analysis; Vineland scales; narrative interview.

In order to measure maternal contingency a new experimental outcome measure using micro-analysis of dyadic interaction between mother and child was developed (for the full method, see James *et al.* 2012). The mother's response to the child's vocal behaviour and the child's response to the mother's vocal behaviour was analysed using behavioural coding of mother–children interaction during naturalistic play. Video recordings were coded as a continuous variable in INTERACT software version 9 (Mangold 2008) to extract the frequency, onset, offset and duration of each mother–child dyad's eye gaze and vocal behaviours. Videos were coded frame by frame in the format 25 frames/s. The first 9 min of free play were coded. Coding was shared by two coders (Wadnerkar-Kamble and Lam-Casettari). Each mother–child dyad was coded independently for each behaviour, giving four codes per dyad: eye gaze of the child towards the mother coded as 'child look'; child's vocalizations coded as 'child verbal'; eye gaze of the mother towards the child coded as 'mother look'; and mother's speech coded as 'mother verbal'. Contingency indices (Joint Frequency and Logg odds ratio) were computed for these variables using sequential analysis in Generalized Sequential Querier Software (Bakeman and Quera 2011). These analyses provide information on the likelihood of the contingency of behaviour within the dyad to be due to chance. See Chorney *et al.* (2010) for further explanation of methodology for sequential analysis.

#### *Analysis of the experimental hypothesis*

If the intervention is successful in enhancing maternal sensitivity, then this should be observable when coding for emotional availability in the mother. The impact on the child should be observable in his/her early communicative behaviour, in particular the communicative intent and autonomy of the child should increase as the mother makes more contingent responses to the child-initiated cues. The concept of communicative autonomy in the child is of particular relevance in the field of childhood hearing impairment since studies using the Tait analysis have shown that communicative autonomy (whether signed or spoken) is predictive of later speech–language development (Tait and Lutman 1997). In addition, the mother and child should be-

come more attuned to each other so the number of contingent episodes, where mother and child's vocal responses occur within a timed window in direct sequence with each other, should increase after the intervention and be maintained at follow-up. If enhancements in child outcome measures are found at times of reported static or declining developmental profile, then the effect is unlikely to be caused by general development in the child. If enhancements are maintained at the final post-intervention follow-up which occurs at least 8 weeks after the intervention, then the effect is unlikely to be due to a halo effect.

## **Method**

### *Overview of the study*

This paper presents preliminary single-case results on three parent–child dyads. Each case was tested using a repeated design to measure change across multiple pre- and post-intervention assessments. Families had two pre-intervention assessments interspersed by an 8–10-week waiting period followed by the intervention. All families came for a post-intervention assessment 2 weeks after completing the intervention phase and a follow-up assessment sometime later. The total duration of time in the study was approximately 7 months (figure 1).

Training in the intervention approach is accredited in the UK by the Community of Practice of Associated Video Interaction Guidance Practitioners. The first named author of this paper is a fully accredited practitioner and an advanced national supervisor in VIG. She was the intervention provider for all families in this study. For more details on the intervention philosophy and phases of training and baselines for accreditation, see Kennedy *et al.* (2011). The intervention protocol recommended a total of seven sessions. These consisted of a goal-setting session, three filming sessions and three review sessions. The intervention approach, which is family centred, is based on co-construction of the intervention design with the achievement of the parents' goals for change being the priority against which work is designed and delivered. This means that if families wanted more than three films or fewer than three films, then this would be discussed and agreed against the

Table 1. Demographic variables for each child

Characteristics	Case A	Case B	Case C
Sex	Female	Male	Male
Age at first visit	1,11 years	3,10 years	9 months
Age at second visit	2,01 years	4,01 years	11 months
Age at third visit	2,07 years	4,02 years	1,04 years
Age at fourth visit	3,00 years	4,08 years	1,10 years
Hearing loss	Profound	Profound	Profound
Prostheses status	CIs implanted 2 months prior to entering the study. Progressing to a regular CI wearer	Initially bilateral hearing aids were worn with no effect. Profound hearing loss was confirmed and bilateral CIs were implanted at 2.5 years. Regular CI user for more than 12 months prior to entering the study	Bilateral hearing aids worn at pre-intervention 1, pre-intervention 2 and post-intervention 1 baselines. Fitted with a CI 5 months prior to his second post-intervention baseline. Regular CI wearer
Birth order	Second born	Twin brother	Third born
Additional medical complications	None	Premature with associated motor delays and diagnosis of cerebral palsy, visual impairment, and global developmental delay	Glue ear requiring grommets

Note: CI, cochlear implant.

families' own belief about the achievement of their goal for change (detail for each family is provided below).

To collect information on parental factors and child language development, the test battery included an initial case history interview to collect demographic information and a narrative interview in which parents were asked open-ended questions about their functioning as a family unit. Parent-child dyads were also video recorded in a 20-min free-play interaction, which provided information for the subsequent coding of mother-child interaction at the macro-level for Emotional availability (Biringen 2008), and the micro-level for behavioural analysis using frame-by-frame coding, and vocal development using Tait assessment (Tait 1993; Tait *et al.* 2001b). Child development was assessed using Vineland Adaptive Behavior Scales (Sparrow *et al.* 2008).

### Participants

Families were recruited on a first-come, first-recruited basis from Specialist Speech and Language Therapy (Primary Care) and Specialist Teachers of the Deaf for children with hearing impairment. Written informed consent was obtained from each parent prior to starting the study. The study was reviewed and approved by Derbyshire Research Ethics Committee and Nottingham University Hospitals NHS Trust Research and Development department. All families were reported to be in good health at the time of filming. All mothers were hearing and all children were congenitally profoundly deaf and pre-lingual. Details on the family dyads are provided in table 1.

### Procedure

The intervention timeline is shown in figure 1. Baseline measurements were conducted in the laboratory. The pre-intervention waiting period was a time when no measurements or input was provided to the family from the research unit. The intervention sessions were all conducted at the family home.

For the baseline measurements families were video recorded during an unstructured<sup>1</sup> free-play interaction in the Child and Family Lab at Nottingham Biomedical Research Unit in Hearing. To minimize the obtrusiveness of the equipment, the free-play recordings were taken via three wall-mounted hard-drive video cameras disguised with fluffy toys (so that only the lens was showing). Two cameras (Canon Legria HV40) were remote controlled by an experimenter in an adjacent room. A third camera also recorded a wide-screen angle of the room from a side view (Canon Legria HFS200). For the free play mothers were instructed to play and spend time with their child as they normally would at home using any of the toys available for approximately 25 min. They were allowed to take a break if required. Following the test session, the recordings were converted into a 20-min movie in MPEG4 format at 25 frames/s.

In the intervention phase the first session involved setting a family-centred goal for change and then a discussion of how to approach the first film (where the film would be taken, what activity would be filmed and with whom). The film recording and editing are integral

<sup>1</sup>Unlike a face-to-face interaction or structured play, the mother-child pair is free to choose their play activity and move about in the room.

Table 2. Details of each families film sessions

	Case A	Case B	Case C
Number of sessions	Three film and shared review sessions	Two film and shared review sessions	Three film and shared review sessions
Parent's predefined goal for the video sessions	'When I'm following the advice of the speech and language therapist I want to use the film to find out whether it has an impact'	'erm I'd like to help with ( <i>child's name</i> ) eye contact and from there get a sort of mirroring each other a little bit' 'just to help him get a little bit more out of it . . . so he can do more, and we could do more'	'For my other family members to feel more confident when they communicate with ( <i>child's name</i> ) using voice and signs and their bodies and their facial expressions'
Parent's descriptor of level achieved in relation to their goal	'yeah we are'	'it's all there, he's copying sounds, he's learning even when I'm not there'	'got there'
Video participants	1. Mother and child 2. Father and child 3. Mother, maternal grandmother and child	1. Mother and child 2. Mother and child 3. n.a.	1. Mother and child 2. Paternal grandmother and child 3. Maternal grandmother and child
Duration of intervention period (weeks)	21	6	16
Additional notes	Mother and father were both present at all filming sessions, and both mother and father participated in all video review sessions together	Mother said that she did not need a third session because she had achieved her goal by the completion of the second film and shared review session. Mother attended all video reviews	Mother was present at the first and third filming session, and she participated in those video review sessions. The paternal grandmother participated in the second video review session

components of the intervention. All films were taken by the first named author and the editing of each film was also undertaken by her. The second session was when the first film was taken. This was followed by a third session where edited segments showing highly successful moments were shared in a review with the families. At the end of each shared review the need for another filming session was discussed and if deemed desirable then the details of the subsequent film were agreed. All families wanted a second film and shared review. Two families decided to have a third film and shared review. The families' goals for the interventions and the particular ways in which the intervention protocol varied for each family are shown in table 2.

#### *Emotional Availability (EA) measure*

The EA Scales, Infancy/Early Childhood version, 4th edition (Biringen 2008) was used to score the quality of interactive congruence shown by the mother-child dyads during the full 20-min free-play session. The EA scales consist of six dimensions, of which four are used to score the caregiver's EA toward the child: maternal sensitivity, maternal structuring, maternal non-intrusiveness and maternal non-hostility; and two are used to score the child's EA toward the caregiver: child responsiveness and child involvement. A direct global score is

rated separately for parent and child dimensions using a seven-point scale. Inter-rater reliability was obtained by a coder who was blind to the occasion of measurement of four randomly selected videos.<sup>2</sup> Both coders were certified reliable in using the 4th edition EA scales following distance training by the scales' author. Intra-class correlations showed 0.84 agreement for maternal sensitivity and 0.89 agreement for maternal structuring (due to a 0.5 difference in ratings for one video), 1.00 (exact) agreement for child responsiveness and 1.00 (exact) agreement for child involvement.

#### *Tait analysis framework*

The Tait framework (Tait *et al.* 2007) was used to assess the pre-verbal vocalizations produced by the child during a 2-min purposefully selected segment from the 20-min free play. The Tait protocol for the purposeful selection requires that the 2-min selection contains the most successful sequences of communication between adult and child. In line with this, the entire recording of the free-play session was viewed, around two or three

<sup>2</sup>The four videos represented one-third of the video sample, and were randomly selected to ensure that a minimum of one video for each family, and one session from each time point (Tx 1, Tx 2, Tx 3 and Tx 4) was used to obtain reliability.

times until the most successful sequence of communication was identified. This section of footage was used to assess the pre-verbal behaviours of the child which is based on a detailed classification of the child's turns. The Tait framework categorizes the child's communicative behaviours into four categories: (1) eye contact, (2) turn-taking, (3) autonomy and (4) auditory processing; and a percentage of the total proportion of time the child is scored within each category is determined. In this study each turn was coded as either a gestural turn, a vocal turn, a gesturally autonomous turn, a vocally autonomous turn, a non-looking vocal turn or a non-response. The total number of turns was used to calculate a percentage of each of these turn types. This synthesis of the data varies from the protocol of previous research reports on the Tait, but the coding for analysis of each turn was not changed. One file was coded by two coders and inter-rater reliability was around 60% of the main differences concerned with the rating of non-response. After discussion a further two files were coded and the inter-rater reliability was 85% and 87%.

#### *Frame-by-frame behavioural coding*

Video recordings were coded as a continuous variable in INTERACT software (Mangold 2008) to extract the frequency, onset, offset and duration of each mother-child dyads' vocal behaviours. Gaze behaviour was also coded. Videos were coded frame by frame in the format 25 frames/s. Coding was shared by two coders (Wadnerkar-Kamble and Lam-Cassettari). The first 9 min from each free-play session was selected for behavioural coding. Inter-rater reliability was tested using a third coder to blind code 20% of each case (a randomly selected 4-min segment). There was a good agreement between the first/second and the third coder as indicated by the Kappa coefficient (Cohen's Kappa), for case A,  $\kappa = 0.95$  for child and 0.83 for mother; for case B,  $\kappa = 1.00$  for child and 1.00 for mother; and for case C,  $\kappa = 1.00$  for child and 0.95 for mother. The analysis is based on a probability statistic to identify the significance of co-occurrences of behaviour within a specified time window (onset of a mother's vocal behaviour within 3 s of the onset of the child's vocal behaviour, and vice versa). The 3-s window was chosen because it was in line with previous research in the area (see detailed methods in James *et al.* 2012). The log-odds ratio was used to identify if sequences of vocal behaviour were significantly co-occurring given the overall pattern of co-occurrence in the data.

#### *Developmental Quotient Measure*

Vineland Adaptive Behavior Scales (Sparrow *et al.* 2008) were used to evaluate each child's personal and social development. The instrument assesses four broad domains

of development: communication, daily living skills, socialization and motor skills according to the child's age level. The communication domain assesses receptive, expressive and written skills. The daily living skills assesses personal, domestic and community skills. Socialization assesses the child's interpersonal skills, socialization during play and leisure, and coping skills. Gross and fine motor skills are assessed by the motor skills domain. The expanded interview form was administered to each parent, at each visit, in a semi-structured interview format. The *adaptive behaviour composite* score was calculated by combining the scores across each of the four domains to provide a total score of the child's general level of adaptive functioning. The Vineland scales shows good internal consistency and test-retest reliability (Sparrow *et al.* 2008). It has also been used reliably with individuals with autism, other developmental delays (De Bildt *et al.* 2005) and with deaf children (Altepeter *et al.* 1986).

#### *Blinding*

In drug trials the experimental blinding of the participants and the researchers is an important design feature that avoids the pitfalls associated with incorrect attribution of change to a specific drug therapy. In this design it was not possible to blind anyone to their participation in the intervention study. In this study coders were blind to the assessment session they were coding for outcome measurement and disclosure of the experimental hypotheses and specific outcome measures were not given in detail to parents or recruiters. In addition, the interventionist was not present for any of the baseline measurements and the researchers taking the baseline measurements were not involved in any of the intervention sessions. The recordings of mother-child play taken at baseline measures were allocated random numbers so that when data for the outcome measures were coded from these sessions, it was not obvious which session the data were coming from. Data coding for the outcome measures were deliberately staggered and delayed so that coders were as naive to the session the data had come from as possible.

#### *Approach to analysis*

In this study we charted the general developmental quotient using the Vineland Adaptive Behavior Scales to identify fluctuations in general development across the four assessment sessions for each child. The developmental trajectory was plotted for each child so specific changes in outcome measurements could be assessed against the background context of the child's general development. It was reasoned that post-intervention enhancements in outcome measures that took place against a backdrop of declining or static general development



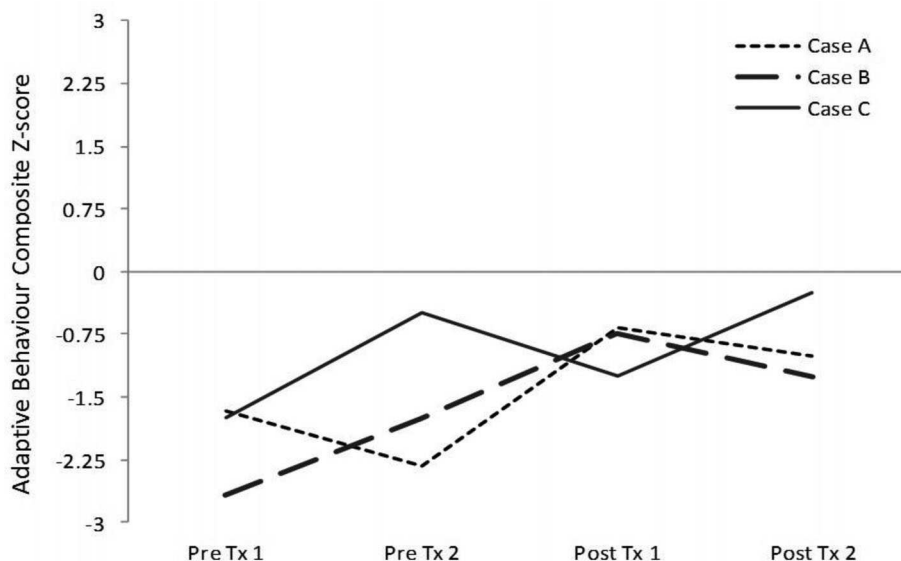


Figure 2. Vineland adaptive behaviour composite score calculated as a Z-score for cases A–C from pre- to post-treatment (Tx).

were not likely to be attributable to general development in the child.

In this developmental context with the experimental hypothesis underpinning this study it was not desirable for any improvements in baseline states to return to pre-intervention levels after the treatment. In order to test the effect of the intervention, the pattern of change in the outcome measures would ideally show stable baselines before the intervention, and enhanced outcome measures at the post-intervention assessment, with these gains maintained at the follow-up assessment. The analysis is based on a pattern analysis of graphed data of the outcome measurements against a general backdrop of the child's general development.

## Results

The developmental quotient was calculated as a Z-score from the Vineland Adaptive Behavior Composite Score for each case at each time point. Data are provided in figure 2.

Case A and case B's developmental trajectories are within the normative developmental range (using  $-1$  z-score as the lower limit of the normal range) at the first post-intervention assessment. The developmental quotients decline marginally in both cases, but are maintained within the normative range at the second post-intervention follow up. In these cases we reasoned that stable pre-intervention baseline measures, with enhanced post-intervention measures that are maintained or improved upon at post-intervention follow-up, are indicative of a change that is unlikely to be due to either general increased development or a halo effect. The spiky development observable for case C is different. It

is possible that the intervention of the cochlear implant fitting, which took place between the post-intervention follow-up assessments, could have exerted an influence on the parental reporting of child development (halo effect of the cochlear implant). For this case we reasoned that positive changes in outcome measures after the research intervention would be particularly indicative of a research intervention effect as this goes against the background context of a declining developmental pattern and comes before a potentially significant intervention of implant fitting.

### *Emotional availability (EA)*

Scores for EA for two maternal scales (sensitivity and structuring) and two child scales (responsiveness and involvement) are provided for all cases at all time points in figure 3. At pre-intervention, all cases show a moderate level of maternal EA, with scores ranging from 5.0 to 5.5, and moderate levels of child EA with scores ranging from 4.5 to 5.5. At post-intervention, maternal EA scores show maintenance or slight improvements ranging from 5.5 to 7.0, and child EA scores show improvement with a range of 5.0–6.0. Specifically, case A shows a trend for the child to be more responsive and more involved in the interaction at the post-intervention follow-up. Case B shows a trend for the child to be more responsive and involved at the post-intervention assessment, and this is maintained at the second follow-up. Case C shows a stable baseline in the mother's sensitivity and structuring prior to the intervention and an increase in this outcome after the intervention. Furthermore, the child shows increased responsiveness and involvement after the research intervention and these gains were

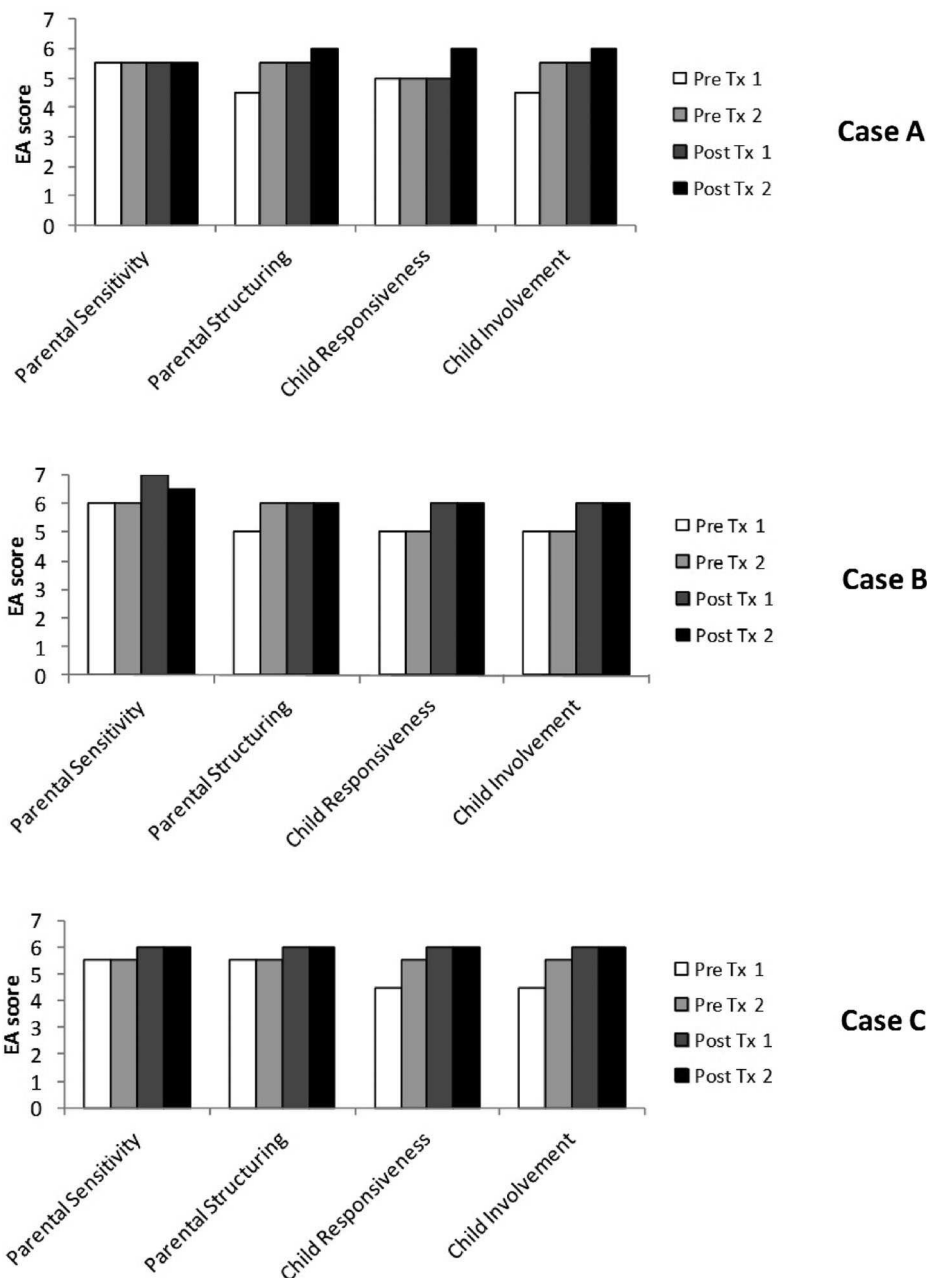


Figure 3. Emotional availability ratings from pre- to post-treatment for cases A–C on two parental dimensions (sensitivity and structuring) and two child dimensions (responsiveness and involvement) using the Infancy to Early Childhood Emotional Availability Scales, 4th edition.

maintained at follow-up. All cases show enhanced scores, particularly child scores, after the research intervention.

*Tait analysis*

The percentage of autonomy (gestural and vocal) and non-response as a percentage of the total number of child turns (gestural, vocal, non-looking vocal turns) are provided in figure 4 for all cases at all time points. The pattern that emerges for each case is stable baselines be-

fore intervention and an eradication of non-responses after the intervention, which is maintained at follow-up in all cases. The trend for increased autonomy in the early communicative repertoires is also evident in all three cases, in particular each child showed increased vocal autonomy. In case C, this occurs after the research intervention and before the intervention of the cochlear implant. The results from the Tait assessment show that the children become more involved in the conversations with their mothers after the intervention and they are

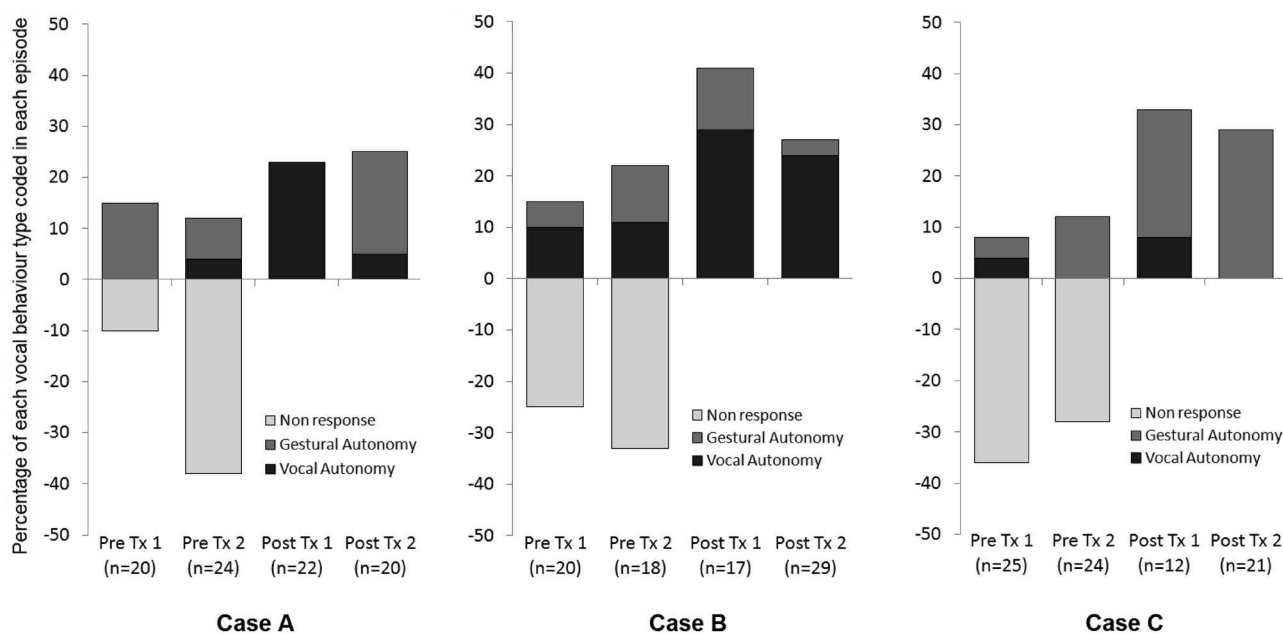


Figure 4. Proportion of vocal behaviours classified as vocal autonomy (black), gestural autonomy (grey) and non-response (white) shown as a percentage of the number ( $n$ ) of vocal turns produced for case A (left), case B (mid) and case C (right) in each coded episode using TAIT analysis, from pre- to post-treatment (Tx).

more likely to take a lead in those conversations. The findings of stability prior to the intervention and maintenance at post-intervention follow-up suggest that these enhancements in early communicative repertoire are not related to the children's general developmental trajectories and, the maintenance in change suggests they are not attributable to a potential halo effect.

### Contingency analysis

The log-odds ratio showing the likelihood of the vocal contingencies of mother to child and child to mother for all cases is provided in figure 5. The data across cases show no pattern of increased contingency in any of the cases after the intervention. What is clear is that the contingencies for the mother on the child's vocal cues and the child's on the mother's look very similar to each other, almost like a mirror image.

### Discussion

We set out to design a robust and relevant study to explore the potential of a new intervention in the context of childhood hearing impairment. There are many limitations to this study which are discussed below, but there are two findings that we think are useful to the field. Firstly, two measures (Tait and EA) appear to provide stable and sensitive measurements and could be used to assess the impact of psychosocial interventions in the field of childhood hearing impairment. These results

provide new knowledge for the field as, to date, both measures have typically been used in experimental/basic science or to measure the impact of cochlear implantation. Secondly, in line with advice about the avoidance of the misattribution of intervention effects (Adair 1984), we sought the opinion of the families themselves on the benefit of the intervention. All families reported that they had reached their goals for the intervention. This was inferred to mean that the parents were all satisfied with the intervention and provides evidence that the intervention itself is acceptable to families. In the wider study, which included 17 families in total, there was no attrition from the intervention. This finding further corroborates the acceptability of the intervention for families. A relevant and useful finding from a phase 1 study concerns the acceptability of the intervention to the population (Medical Research Council (MRC) 2008). Since the onset of the study, video interaction guidance has been highlighted in National Institute for Health and Clinical Excellence (NICE) guidance for vulnerable families: 'should consider using interactive video guidance to improve maternal sensitivity' (NICE 2012). The intervention is valued as an efficacious family intervention and this is the first study to show that it is likely to be acceptable to families in the context of childhood hearing impairment.

In the study we wanted to be able to interpret the results within the context of the child's general development. We attempted to model this using the developmental quotients from the Vineland. In clinical practice

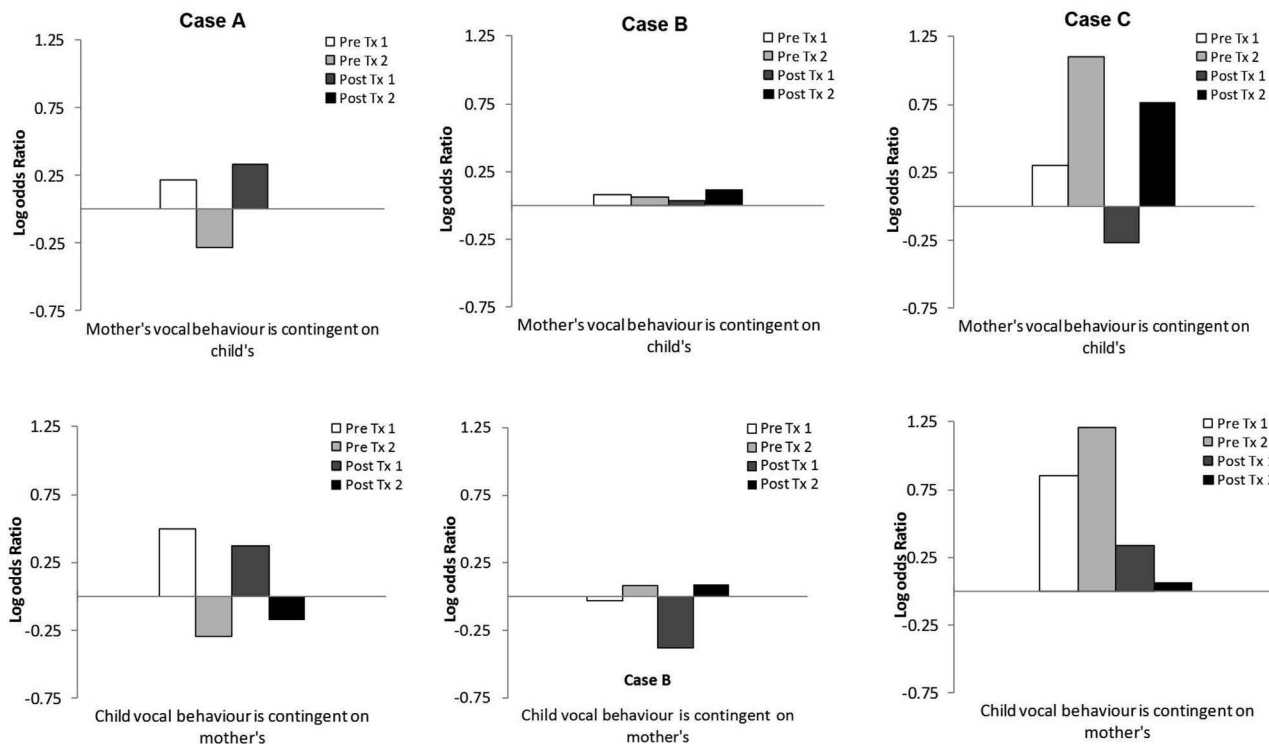


Figure 5. Log odds ratio for the mother's vocal behaviours being contingent on (occurring within 3 s) the child's vocal behaviours (top), and for the child's vocal behaviours being contingent on their mother's vocal behaviours (bottom) for case A (left), case B (mid) and case C (right) from pre- to post-treatment (Tx).

the administration of a standard test is not usually recommended within a 6-month period in order to avoid potential confounds of practice effects or boredom, etc. In this research, where we wanted to have a snapshot of the child's developmental level to interpret the results, we decided to administer the test repeatedly to check for change over time. The Vineland has good test-retest reliability (with differences across standard means that range from  $-1.3$  to  $3.1$  standard points across subscales). In addition, the test-retest reliability across interviewers is rather weaker, but in our protocol the same academic clinical psychologist administered the test, so reliability was not compromised on that basis. However, the repetition of the test at short intervals may have led to unplanned effects so that the results are not as reliable an indicator of development as we would have liked. This means that our way of using the Vineland results to understand the changes in the outcome measures may not be well justified. The desire to measure change robustly (using a reliable measure) and attribute that change, if found, to the intervention (using a valid construct that any change found could be due to maturity in development) in some ways worked against us. The principle of conducting robust examination of change mechanisms was right, but the design of the measures requires more consideration for the future.

The use of short-term longitudinal design with multiple baselines post-intervention allowed us to consider the potential impact of a halo effect because the second post-intervention assessment was administered at least 8 weeks after the intervention. The maintenance of change on the outcome measures that occurred after the intervention was sustained and this does suggest that the change was not due to a halo effect.

As well as allowing the bespoke tailoring of the research intervention, the case study design allowed us to recruit a maximally varying sample in the study. This was important in the present phase 1 clinical trial because a more generic question of an early clinical trial is to identify who the intervention might produce the most change for. It also meant that the sample reflected the variation that exists in clinical practice because it included children with additional needs and children at different trajectories with cochlear implantation. Given our hypothesis that the intervention contained elements that would enhance primal attributes of human parenting, we predicted that all cases would benefit from the intervention by showing enhancements on the outcome measures. We did find this pattern. In terms of EA, despite the finding that none of the families was seen to be at risk of problematic due to moderate to high baseline scores, all families showed stability and enhancements of

up to 1 point on each of the EA dimensions over time. This indicates that the global disposition of both mother and child was becoming more emotionally connected, reciprocal and involving of one another after the intervention period. When the Tait changes are considered the data suggest that the research intervention could have particular potential for children at the pre-implant stage (data from case C) and for children with additional complex needs (data from case B) as both of these cases showed more improvement in child outcomes than case A who had just received her implant and was not considered to have additional needs. The small number of cases presented here means that the generalizability of the study to the wider population remains a question for future research where larger datasets can be used to infer the overall size of the effect of the intervention on the main outcome measures. This work is currently underway.

The complexity inherent in the design of intervention research presents a huge challenge at all stages of the design process. A limitation of our study lies in the potential weakness of relying on visually based interpretations of results and the underdeveloped sense of what counts as a significant change in an outcome measure. In disseminating the findings from this small number of cases our intention is to model the building of the rationale for the study and in particular to draw attention to the concept of contingency and its measurement. In a previous study that explored the concept of parenting style and speech and language development in children with hearing impairment DesJardin and Eisenberg (2007) extracted behavioural data from the interactional context in which it was generated and then classified and grouped independently as mother or child behaviours. We have argued that this approach cannot provide a good test of a theory which emphasizes the social construction of speech–language that we think is built as a co-endeavour between parent and child. We therefore employed a methodology specifically to test the patterns between mother and child by assessing contingent behaviour predicated on vocal behaviour of each member of the dyad. Whilst we did not find any enhancements in the number of contingent episodes which we did predict, we were able to show how the likelihood of contingency appeared to be a property of the dyad rather than a feature of the mother or of the child. This was evident as the likelihood of contingencies to vocal utterances, which varied quite markedly in two of the cases across the play sessions, followed a similar pattern and number. In fact, the data in figure 5 of child and parent contingent responses look almost like mirror images. This finding does show that our focus on exploring interactional data from a dyadic perspective provides potential for new insight into how the basic unit of mother–child interaction should be con-

ceptualized. In future work, this experimental outcome measure will be explored further to consider changes in patterns and features within contingent episodes that might reveal further patterning within the dyad.

The imperative towards the provision of family-centred services has a robust foundation because family-centred provision provides longevity in a comprehensive array of valuable outcomes (King *et al.* 2004). In this paper we have sought to illuminate the design of a family-centred research intervention which we achieved mainly by adopting an *N*-of-1 design. In on-going work we are using qualitative methodology and applied linguistic analysis to evaluate the way in which this family-centred intervention has an impact on the family unit and the effect of the intervention on the self-efficacy of the mother. In future work we hope to triangulate the quantitative and qualitative findings to provide an example of a robust family-centred intervention research design and provide new knowledge for the academics, educators, clinicians and families in the field.

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