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Research Article

Palin Parent–Child Interaction Therapy: The Bigger Picture

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Purpose: Palin Parent–Child Interaction therapy (Kelman & Nicholas, 2008) is an evidence-based intervention for young children who stutter. The evidence consists of multiple single-subject replicated studies, and this demonstrates that the intervention is effective. The aim of this study was to enhance the evidence base by exploring the effectiveness of the therapy with a large cohort of children who stutter.
Method: Children and parents completed a range of assessments at 4 time points: start of therapy and then 3, 6, and 12 months later. The following variables were included: stuttering frequency, child's communication attitude, parents' perception of the impact of the stuttering on the child, the severity of stuttering and its impact on the parents, and their knowledge of stuttering and confidence in managing it. Hierarchical multiple regression analyses were conducted to explore whether the variables are predictive for the outcome "parent knowledge and confidence." In addition, we sought a preliminary view of factors associated with outcome level by separating children into 2 groups according to response to treatment (more successful and less successful).
Results: The results demonstrated a significant improvement in all variables, and this improvement was maintained for 1 year posttreatment. Measures collected 3 months after the start of therapy showed significant improvement in child attitude to communication, parents' knowledge and confidence in how to manage stuttering, and mothers' ratings of stuttering severity and impact the child's stuttering has on the mothers. By 6 months after therapy onset, there was a significant reduction in stuttering frequency and fathers' perception of severity and their worry about it. Furthermore, these improvements were maintained 1 year posttherapy. Several variables predicted parents' knowledge and confidence 6 months after the start of therapy. Finally, those who made greater improvements had mothers who were more negative in their ratings of severity and worry, and had less knowledge and confidence at the start of therapy. There were no differences between the groups on a range of other variables.
Conclusions: The results demonstrate that, over a year, children who attend a course of Palin Parent–Child Interaction show reduced stuttering frequency and a more positive attitude to speech. In addition, parents observe these improvements in the child, feel more confident in managing the stuttering, and are less worried about it. The different times at which specific variables significantly improved provides insight to a process of change over time. Results suggest that parents' ability to notice positive change in fluency and the impact that these observations have on both the child and the family are linked to their confidence in how to support the child. The preliminary findings with regard to response to treatment suggest that children can benefit from this program even with factors that might be predicted to reduce therapy success.

Although most stuttering in young children resolves without intervention (Yairi & Ambrose, 2013), the experience of stuttering can have a negative impact on a child. Young children who stutter (CWS) have been shown to have a more negative view of themselves as communicators, view speaking as something that is difficult, and can be less competent communicators as a result of stuttering (Clark, Conture, Frankel, & Walden, 2012; Langevin, Packman, & Onslow, 2009, 2010; Vanryckeghem, Brutten, & Hernandez, 2005). As experiences with chronic stuttering increase with age, so too do negative thoughts and emotions about stuttering and being a person who stutters (Vanryckeghem & Brutten, 1996). Even young CWS are often viewed more negatively by their peers and may be subject to teasing and social rejection (Ezrati-Vinacour, Platzky, & Yairi, 2001; Langevin et al., 2009).

Stuttering may affect not only the child but also the parents. Parents of young CWS express frequent worry about the child's stuttering and report a lack of knowledge about

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how to support their child (Langevin et al., 2010; Millard & Davis, 2016; Plexico & Burrus, 2012). They are concerned about their child's future and fear that stuttering will prevent the child from reaching his or her potential (Millard & Davis, 2016; Plexico & Burrus, 2012). Naturally, parents try to support the child; however, when attempts to do so are unsuccessful, the parent–child relationship may suffer (Lau, Beilby, Byrnes, & Hennessey, 2012).

The potential negative consequences of stuttering into adolescence and adulthood may have their roots in early experiences with the disorder. Adults who stutter have a higher incidence of social anxiety than their typically fluent peers, and such anxiety frequently emerges in the school age years (Iverach et al., 2016; McAllister, Kelman, & Millard, 2015). There are certain physiological and environmental factors that are considered to be risk factors for social anxiety, and these, along with the negative experiences of stuttering, negative responses of others, and the potential limiting effect that stuttering can have on social interactions, may explain why CWS may be at risk for anxiety disorders (Iverach et al., 2016).

Given the short-term impact on well-being for both children and parents and the potential for long-term negative consequences of stuttering for both social participation and mental health, the probability of unassisted recovery should not be the primary consideration when deciding whether therapy is warranted or recommended. If the stuttering is having a negative impact on the child's well-being, ability to communicate, or perception of himself or herself as a competent communicator, then therapy is indicated. If parents are worried and requesting help to support their child, then therapy is indicated. Therapy should not be withheld or reserved for cases when the stuttering is firmly established and clearly persistent. To meet these needs, therapists need access to efficacious and effective interventions that are directed at supporting both the child and the parents.

Interventions for Young CWS

Compared with the school and teenage years, there are more evidence-based options for intervention with preschool children that are explicitly described and available for use. For example, there are therapies that focus on indirect methods, whereby parents make environmental changes and modifications to their verbal interactions with the child that facilitate fluency; direct methods, in which the child learns to make changes in speaking; and combined approaches, where both indirect and direct methods are included to varying degrees.

Gottwald (2010) describes a multidimensional early intervention program for preschool CWS that is based on the Demands and Capacities Model of stuttering onset and development. This approach incorporates weekly individual direct therapy for the child, introducing a slower speaking rate, and reducing struggle. The program also includes weekly family therapy sessions focusing on modifying environmental factors to support the child's development of speech fluency. The average length of therapy is 12–14 sessions, with dismissal indicated when children demonstrate fewer than 3% stuttered syllables and when parents say they are comfortable managing the child's fluency needs on their own.

Richels and Conture (2007) describe a program for preschool CWS, consisting of a minimum of 12 sessions of group therapy that includes initial, transfer, and maintenance programs. Group activities focus on modeling and practicing slow utterances of increasing length and complexity and appropriate turn-taking. Parents participate in a parent group where fluency facilitating strategies are demonstrated and discussed.

Yaruss and Reardon-Reeves (2017) describe a family-focused treatment approach for early childhood stuttering, designed to help parents create a supportive environment, in addition to directly addressing stuttering and teaching the child to make intentional changes in their speech production to enhance the likelihood of more fluent speech. This approach particularly focuses on helping children develop healthy, appropriate attitudes toward their communication abilities as a way to help them achieve improved speech fluency in the context of strong self-esteem and positive thoughts and feelings about speaking. Yaruss, Coleman, and Hammer (2006) evaluated this approach with 17 children by assessing percentage of syllables stuttered (%SS) and parent report and including follow-up of between 1 and 3 years. All 17 children demonstrated significant gains in their fluency and maintained these increases at long-term follow-up.

A direct method of therapy for early stuttering was also reported by Trajkovski et al. (2009), using syllable-timed speech (STS) with three preschool children who stutter. STS involves speaking with minimal differentiation in linguistic stress across syllables and is achieved by saying each syllable in time to a rhythmic beat. The therapist modeled STS to the parent and child at near-normal speech rate and with normal intonation. The parent was then required to model STS to the child at home, to have practice sessions with the child four to six times per day, and to encourage the child to use STS during everyday conversations, occasionally praising the child for using STS. A mean of 8.6 clinic visits enabled the children to decrease their stuttering to below 1% syllables spoken, although no long-term follow-up data were reported.

There are two programs that are supported by randomized controlled trial evidence, the Lidcombe Program (Jones et al., 2005) and RESTART-DCM (de Sonneville-Koedoot, Stolk, Rietveld, & Franken, 2015). The Lidcombe Program is based on operant principles, with parents reinforcing fluent speech through the use of verbal contingencies for both stutter-free speech and moments of unambiguous stuttering. These are presented to the child during structured speech and natural speaking situations. Parents rate stuttering severity within and beyond the clinic on a scale from 0 to 9 (where 0 = *no stuttering* and 9 = *extremely severe stuttering*), and the treatment goal is to reach and maintain scores of 0–1 (Packman et al., 2016).

The RESTART-DCM therapy (Franken & Putker-de Bruijn, 2007) is based on the Demands and Capacity model proposed by Starkweather and Gottwald (1990). The aim of this approach is to reduce internal and external motoric, linguistic, emotional, and cognitive demands and, in doing so, enable the child to speak fluently as a result of "matching" his or her communication abilities to demands at the moment. The child's speech motor, linguistic, emotional, and cognitive capacities may be explicitly targeted, reinforced, and developed. A randomized controlled trial comparing RESTART-DCM and the Lidcombe Program yielded no significant difference in outcomes (de Sonneville-Koedoot et al., 2015).

Within all intervention programs, the specific mechanisms for change and the essential treatment components that underlie them are unclear. In the past, a number of authors have proposed the possibility that it is the similarities, rather than the differences, between programs that might explain their equivalent outcomes (Duncan, Miller, Wampold, & Hubble, 2010; Onslow & Millard, 2012; Zebrowski, 2007). This can certainly be argued for programs for preschoolers, and such "common factors" might include establishing a strong therapeutic alliance, having one-to-one parent–child communication time, having time when fluency is enhanced, increasing parents' knowledge about stuttering, increasing parents' confidence, reducing parental anxiety, and training therapists and increasing their confidence to support families.

Palin Parent–Child Interaction

Palin Parent–Child Interaction (Palin PCI) therapy (Kelman & Nicholas, 2008) is a program for CWS who are under 7 years old. Palin PCI involves both parents and the child and is based on a detailed assessment of the child's stuttering (amount, type, and impact) using qualitative and quantitative measures, including a child interview. Parents complete rating scales and provide a detailed case history to determine factors that may be relevant in the onset, development, and impact of stuttering on the child and family. This assessment identifies factors that will determine which interaction, family, and child strategies will form the family's individualized therapy program. Therapy aims to build the child's fluency and confidence to become a competent communicator as well as develop the parents' knowledge and confidence about managing their child's stuttering and their response to it.

Therapy is delivered through an initial block of six in-clinic sessions, followed by a 6-week home consolidation period where the parents continue to implement the strategies in the home setting. Interaction strategies are introduced within 5-min "Special Times," which are video-recorded in the clinic and then practiced up to five times per week at home. Parents watch the video recordings of themselves to identify helpful interaction strategies they are already using and explore ways to increase their use of these to support the child's fluency and communication success. Interaction strategies may include following a child's lead in play, using pausing and matching a child's rate of speech, and monitoring the complexity of language during play. Family strategies include confidence building, managing children's emotions, and family turn-taking in both play and conversation. Throughout, the child and parents are encouraged to talk openly about stuttering, to become more desensitized to stuttering, and to consider fluency as only one aspect of the ability to communicate and participate in speaking situations. At the end of the first therapy phase of six clinic visits and a 6-week consolidation period, the child's progress is reassessed and, if the outcomes are satisfactory, parents are encouraged to continue to implement the strategies with the therapist monitoring progress at three further review appointments during a 1-year period. If at any point there are concerns about the child's fluency, further therapy sessions are arranged that may include child strategies, a direct component introducing pausing and/or rate reduction to support fluency. Children typically do not require the direct component of the program as fluency and communicative competence and confidence often improve with the indirect component alone.

Evidence for Palin PCI

There are two published studies that report the efficacy of Palin PCI (Millard, Edwards, & Cook, 2009; Millard, Nicholas, & Cook, 2008). Both these studies are single-subject studies, replicated with six children in each study. In both of these studies, the participants had been stuttering for more than 12 months and were therefore considered by the authors to be less likely to recover naturally. Furthermore, the design of the studies, which includes a multiple-measure baseline before the introduction of therapy, allowed the pretherapy trajectory of stuttering behavior to be monitored. In all cases, there was no trend in the baseline data, confirming that stuttering was not decreasing before the onset of the intervention. The second important feature of these studies was the method of analysis. Cumulative sum analysis was used (Montgomery, 1997), which enables the researcher to determine whether observed change is over and above the natural fluctuation that would be expected. This is important given the variable nature of stuttering in young children, and the results showed that the frequency of stuttering reduced compared with the baseline, even when the variability of the stuttering was taken into account. The results showed that the frequency of stuttering significantly reduced in 10 of the 12 children and, for eight, this was during the indirect therapy phase (within 12 weeks of therapy starting).

The internal validity of each of these single-subject studies is high, and the replication across 12 participants increases the external validity (Zhan & Ottenbacher, 2001). However, these are not group data, and it is not possible therefore to make judgments about the likely outcome of Palin PCI for the population of CWS under 7 years old. With this in mind, the aim of this study was to explore the effectiveness of Palin PCI with a group of children who received therapy.

Method

This study was conducted at a specialist center for CWS in London (Michael Palin Centre) and is the center where the therapy program was developed. All children under the age of 7 years, who participated in Palin PCI during a 2-year period, were included.

Participants

Fifty-five children (38 male, 17 female) completed 1 year of data collection during the period of the study. The mean age at the start of data collection was 52.8 months (range = 30–84 months, *SD* = 14.01 months, median = 49 months). Mean interval between onset of stuttering and initial data collection was 19.59 months (range = 3–77 months, *SD* = 17.27 months, median = 15 months). There were data regarding language development based on the Clinical Evaluation of Language Fundamentals–Preschool for 45 children. Of these, 13 achieved scores within the expected range across the subtests, 18 scored above expected levels, 13 scored below, and 11 exhibited mismatched or dissociated (Anderson, Pellowski, & Conture, 2005) profiles in which one or two subtest scores fell below those for other subtests. Details of family history of stuttering were obtained through parent report. There were similar numbers of children who had mothers or fathers who stuttered: 14.8% had mothers who had experienced stuttering (n = 8, four persistent and four recovered), and 14.8% had fathers who had experienced stuttering (n = 8, four persistent and four recovered). Beyond parental family history, 36.4% had blood relatives with a history of stuttering (n = 20; 11 persistent, five recovered, and four mixed outcomes). Sixteen children were identified as having English as an additional language, although the age at which English was introduced, the degree of exposure, and the relative competency compared with the home language were not available.

Before the start of therapy, stuttering frequency data were available for 48 of the children. Stuttering ranged from 0% to 36.6% stuttered syllables (M = 6.76, SD = 6.55, median = 5.45). Those who did not stutter in the speech sample obtained were observed and confirmed to be stuttering by parents and speech-language pathologists (SLPs) during other activities. In terms of distribution, 16 were mild (< 3%); 13, moderate (3%–7%); 15, severe (7%–14%); and 4, very severe (≥ 14%).

Outcome Measures

Stuttering Frequency

A sample of speech was video-recorded while the child described a series of "What's wrong?" pictures with the SLPs in the clinic. These were analyzed by SLPs or an administrator trained in the procedure, to determine the %SS in the sample. Whole- and part-word repetitions, prolongations, and blocking of sounds were included in the counts.

Child's Attitude to Speaking

Child's attitude to speaking was assessed using the KiddyCAT (Vanryckeghem & Brutten, 2007). The core construct of this test is a rating the child assigns to the ease or difficulty of speaking (Clark et al., 2012).

Parents' Perception of Stuttering and the Degree of Impact

The Palin Parent Rating Scale[1] (PRS; Millard & Davis, 2016) was completed by parents. This is a standardized measure comprising three constructs, hereafter referred to as factors: Factor 1, the impact of stuttering on the child; Factor 2, the severity of the stuttering and impact on the parents; and Factor 3, parents' knowledge of stuttering and confidence in how to support the child. Internal consistency for each of the scales was shown previously using Cronbach's alpha (Factor 1: α = .865; Factor 2: α = .863; and Factor 3: α = .838), demonstrating the reliability of the measure (Millard & Davis, 2016).

Outcome measures were obtained at the following four time points (T1–T4):

- T1 = pretherapy
- T2 = 3 months after the start of therapy
- T3 = 6 months after the start of therapy
- T4 = 12 months after the start of therapy

Analyses

A repeated-measures analysis of variance (ANOVA) was conducted to explore change over time in each of the variables. Post hoc comparisons with Bonferroni corrections for multiple comparisons were conducted. Hierarchical regression analysis was used to explore whether certain variables were predictive of selected outcome measures.

Response to Intervention

The individual variability in response to intervention and change over time prompted us to consider whether there were differences between those children at either end of a range of outcomes (i.e., "most" and "least" successful over time). We considered each of the variables (KiddyCAT; %SS and the Palin PRS scores) and determined for each whether there was a change between T1 and the final data collection point that was completed for that variable and child. Not all participants contributed data for all variables at T1. Those children with a change in two of five variables were considered to have the "most successful" outcome. This does not mean that therapy for children with a change in fewer variables was "unsuccessful"; it is well known that a significant change in a single factor can be important or large enough to be meaningful for the child or parents. The following criteria were based on practice-based evidence

[1]https://www.palinprs.org.uk/secure/pprs_connect.php

of treatment outcomes and applied to each variable as reasonable cutoffs for a clinically significant change:

Stuttering frequency: Because of the range of stuttering frequency at the beginning of therapy, we adopted a criterion of 50% reduction from pretherapy to posttherapy for a clinically significant change.

KiddyCAT: A reduction to 0 or 1 was considered a successful outcome.

Palin PRS: We considered a positive shift from one category to another to be clinically significant (e.g., a reduction from moderate to low impact on the child for Factor 1). We used mothers' ratings rather than fathers' because there were more of those available at T1. There was one participant who had father ratings but not mother ratings at T1 and so was included.

Results

As is routinely the case when collecting clinical data, there were missing data points across participants, variables, and time points (see Table 1). In making decisions about the best analysis procedures for incomplete data sets, we first visually inspected the data to determine any patterns across measures and time points (T1–T4). As shown in Table 1, the number of observations decreases from T1 to T3, followed by an increase at T4. This pattern suggests that the missing data cannot be solely accounted for by a single factor such as attrition due to absence, nonresponsiveness to treatment, or a positive outcome. However, it is not possible to state with confidence that the data are missing at random. With this in mind, we decided to use single imputation, the most basic form of handling missing data in clinical research. A single imputation of the series mean to replace missing data was carried out, in which the missing values were replaced by the mean value for that variable obtained from all participants (Dziura, Post, Zhao, Zhixuan, & Peduzzi, 2013). Because repeated measurements from an individual or a group of individuals tend to be correlated, this method allows all available data from each child and parent who participated in the study to be included.

Data are reported in Table 1. The summary of findings for each of the variables reported below focuses on the post hoc pairwise comparisons (Bonferroni corrected) to understand how each variable changed over time.

Stuttering Frequency

Figure 1 shows the average stuttering frequency for each data collection point over time. Visual analysis suggests a steady reduction in stuttering frequency across time, which was supported overall by the repeated-measures ANOVA, $F(3, 162) = 8.2$, $p < .001$. Mauchly's test demonstrated sphericity was not violated, $\chi^2(5) = 41$, $p < .001$. Pairwise comparisons with Bonferroni corrections revealed a significant reduction in stuttering frequency between T1 and T3 ($p = .057$) and between T1 and T4 ($p < .001$) as well as Time Points 2 and 3 with T4 ($p = .019$ and $p = .025$, respectively).

Child's Attitude to Communication

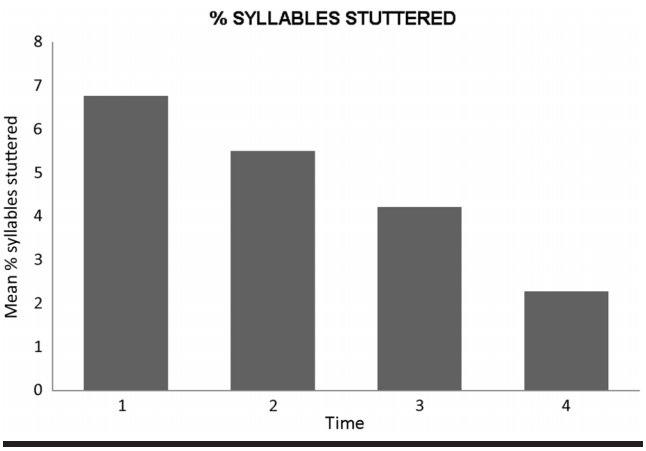
Results from the KiddyCAT are shown in Figure 2. Mauchly's test demonstrated that sphericity is not violated, $\chi^2(5) = 30.48$, $p < .001$. The repeated-measures ANOVA demonstrated a significant effect of time, $F(3, 162) = 73.4$, $p < .001$, with pairwise comparisons demonstrating significant reductions in scores (or decreasing negative thoughts about speaking and stuttering). The reduction in CAT scores between T1 and all other time points was significant at $p < .001$. There were significant reductions between T2

Table 1. Mean scores and standard deviations for each time point.

Variable	T1	T2	T3	T4
% Stuttered syllables	6.76 (6.11) (n = 48)	5.5 (7.61) (n = 31)	4.21 (4.33) (n = 21)	2.28 (2.5) (n = 35)
KiddyCAT	4.64 (1.33) (n = 11)	2.91 (1.60) (n = 11)	1.6 (0.82) (n = 10)	2.00 (1.44) (n = 23)
Mothers' Palin PRS Factor 1	5.26 (1.11) (n = 49)	5.15 (0.93) (n = 29)	5.69 (0.67) (n = 23)	5.65 (0.74) (n = 32)
Fathers' Palin PRS Factor 1	5.31 (0.94) (n = 41)	5.54 (0.73) (n = 25)	5.62 (0.61) (n = 17)	6.0 (0.53) (n = 25)
Mothers' Palin PRS Factor 2	3.55 (1.36) (n = 49)	4.36 (1.14) (n = 29)	5.05 (0.88) (n = 23)	5.06 (1.07) (n = 32)
Fathers' Palin PRS Factor 2	3.84 (1.18) (n = 41)	4.33 (0.91) (n = 25)	4.67 (0.75) (n = 17)	5.6 (0.66) (n = 25)
Mothers' Palin PRS Factor 3	4.42 (1.74) (n = 49)	5.74 (1.04) (n = 29)	5.91 (0.82) (n = 23)	6.08 (0.91) (n = 32)
Fathers' Palin PRS Factor 3	3.8 (1.46) (n = 41)	5.53 (0.96) (n = 25)	5.66 (4.8) (n = 17)	6.47 (0.6) (n = 25)

Note. T1 = pretherapy; T2 = 3 months after the start of therapy; T3 = 6 months after the start of therapy; T4 = 12 months after the start of therapy; PRS = Parent Rating Scale.

Figure 1. Frequency of syllables stuttered over time.



and all other time points (T2 and T3: $p < .001$; T2 and T4: $p = .021$). There was no significant difference in KiddyCAT scores between T3 and T4 ($p = .426$).

Parents' Perception of the Impact of Stuttering on the Child

The Palin PRS Factor 1 explores parents' views of how much stuttering impacts the child. Higher scores indicate less impact. Scores obtained by mothers over time are presented in Figure 3, and fathers' scores are in Figure 4. Sphericity was not violated for mothers' scores, $\chi^2(5) = 13.37$, $p = .02$, but was for fathers', $\chi^2(5) = 5.44$, $p = .365$, and so Greenhouse–Geisser results are reported for fathers. There was a significant effect of time for both mothers, $F(3, 162) = 4.18$, $p < .001$, and fathers, $F(2.8, 131.5) = 11.44$, $p < .001$. Pairwise comparisons with Bonferroni correction revealed a significant reduction in impact between T1 and T3 ($p = .017$), T2 and T3 ($p = .01$), and T2 and T4 ($p = .009$) for mothers, whereas fathers demonstrated a significant reduction between each time point and T4 (T1 and T2: $p < .001$; T3: $p = .007$).

Figure 2. Children's attitude to communication over time (KiddyCAT).

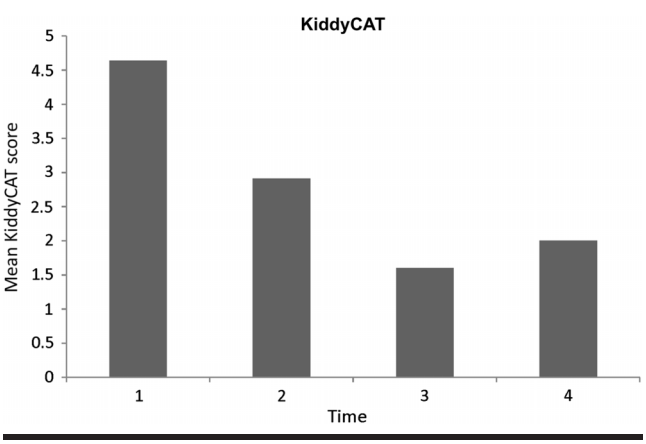
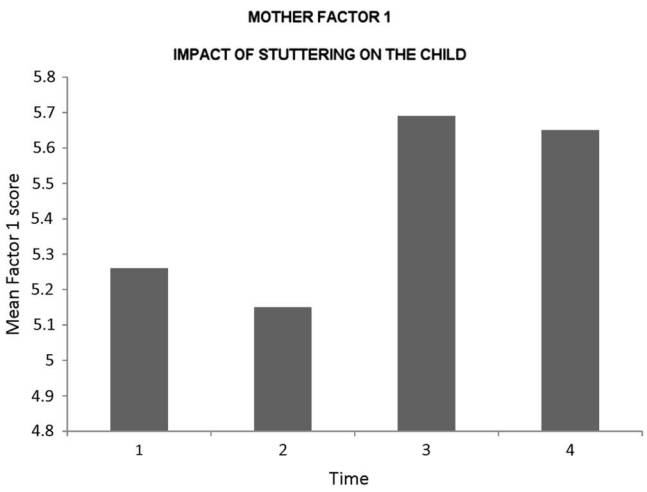


Figure 3. Mothers' perception of the impact of stuttering on the child.



Parents' Perceptions of Stuttering Severity and the Impact That It Has on Them

Factor 2 of the Palin PRS is made up of questions that explore the parents' view of the severity of the child's stuttering and the impact that it has on them in terms of worry and anxiety. Mothers' scores are shown in Figure 5, and fathers' scores are in Figure 6. The higher the score, the less severe the stuttering and impact on the parents. Mauchly's test showed that sphericity was not violated for mothers, $\chi^2(5) = 17$, $p = .005$, but was for fathers, $\chi^2(5) = 5.21$, $p = .391$, and so Greenhouse–Geisser results are reported for fathers. There was a significant effect of time for both mothers, $F(3, 162) = 28.08$, $p < .001$, and fathers, $F(2.8, 153.3) = 50.18$, $p < .001$. For mothers, there was a significant reduction in severity of stuttering and impact on the mothers between T1 compared with T2 ($p = .003$), T3 ($p < .001$), and T4 ($p < .001$) and between

Figure 4. Fathers' perception of the impact of stuttering on the child.

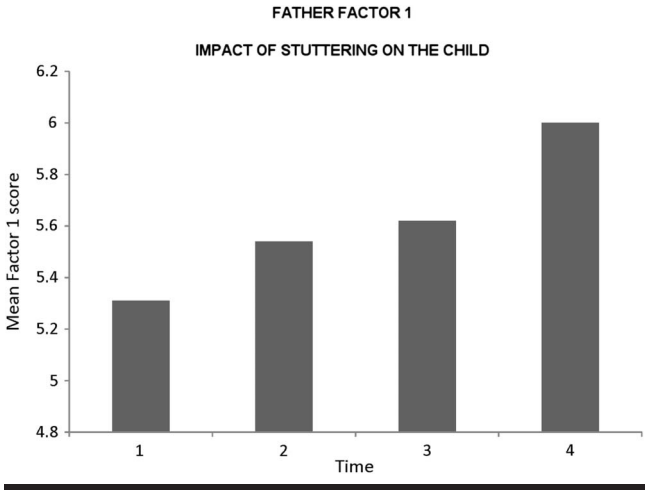


Figure 5. Mothers' perception of the severity of stuttering and the impact that it has for them.

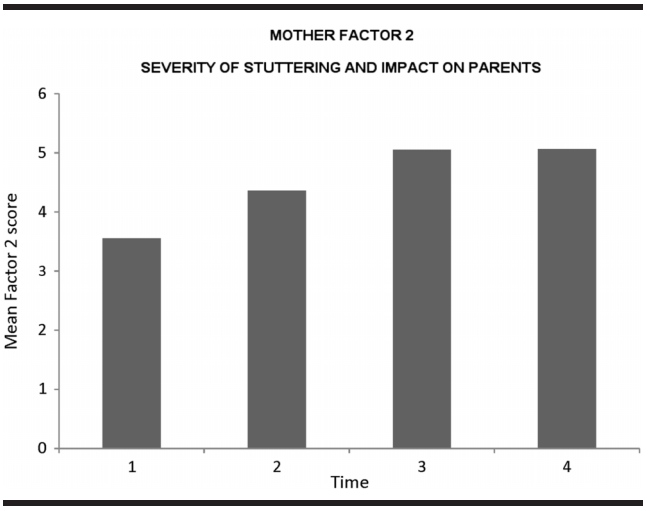
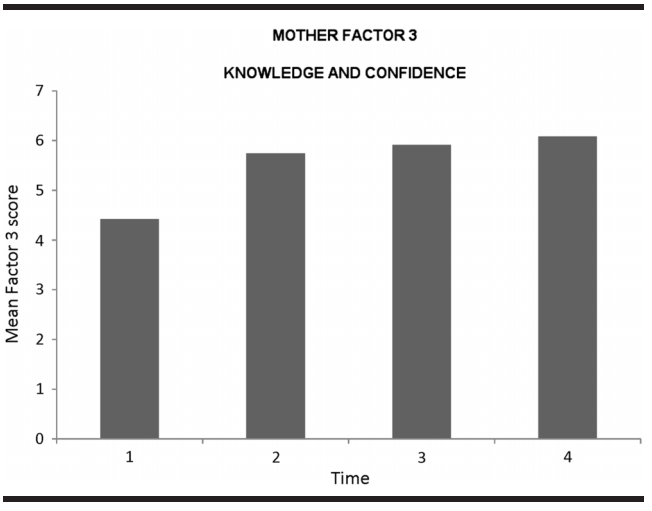


Figure 7. Mothers' knowledge about stuttering and confidence in helping their children.



T2 compared with T3 ($p = .001$) and T4 ($p = .003$). There was a significant improvement in fathers' scores between T1 and T2 ($p = .022$), T3 ($p < .001$), and T4 ($p < .001$), with a significant change also observable between T2 ($p < .001$) and T3 ($p < .001$). There was no significant difference in either group's perceptions of stuttering severity and impact when comparing scores at T3 and T4, indicating that these improvements were maintained.

Parents' Knowledge About Stuttering and Confidence in Managing It

Factor 3 on the Palin PRS measures the parents' knowledge about stuttering and their confidence in supporting their child (see Figures 7 and 8). Higher scores reflect greater parental knowledge about stuttering and how to support the child. Mauchly's test demonstrated that sphericity was not violated for either mothers, $\chi^2(5) = 47.76$, $p < .001$, or fathers, $\chi^2(5) = 42.17$, $p < .001$. There was a significant effect of time for mothers, $F(3, 162) = 31.78$, $p < .001$, and fathers, $F(3, 162) = 87.71$, $p < .001$. For mothers, the post hoc pairwise comparison demonstrated a significant increase in confidence between T1 and all other time points ($p < .001$), with no other significant time point comparisons. This indicates that mothers became more knowledgeable and confident early in the therapy process, and this is maintained throughout the year. Fathers also demonstrate a significant increase between T1 and all other data points ($p < .001$). For fathers, there were also significant differences between T2 and T3 compared with T4 ($p < .001$), suggesting that the initial improvement was maintained from T2 to T3, and increases by T4, in the absence of therapy.

Figure 6. Fathers' perception of the severity of stuttering and the impact that it has for them.

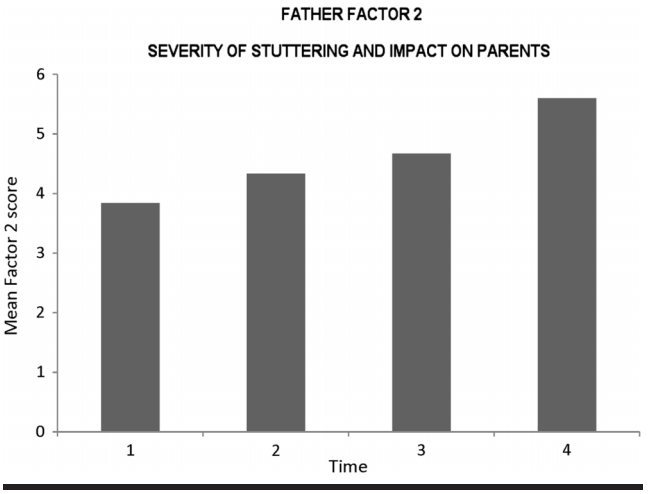
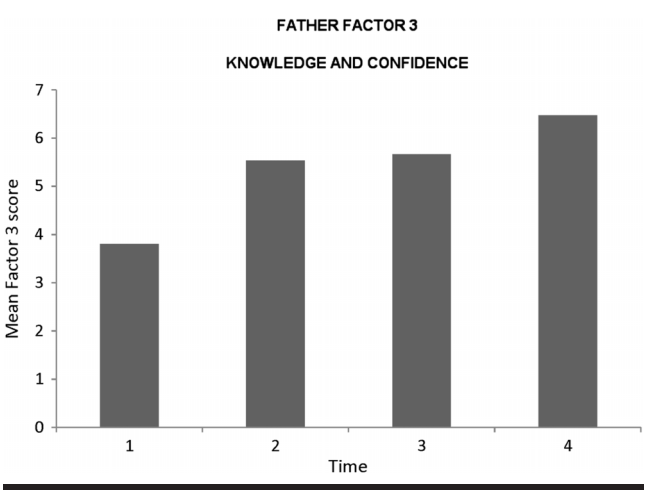


Figure 8. Fathers' knowledge about stuttering and confidence in supporting their children.



Predicting Outcome

One of the primary aims of Palin PCI is to increase parents' knowledge and confidence in how to support and help their child (Palin PRS Factor 3); therefore, we were interested to know which child (stuttering severity) and parent (impact of stuttering on the child [PRS Factor 1]), and stuttering severity and impact on parents (PRS Factor 2) variables might predict the extent to which parents feel more competent and less unsure about how to effectively manage their child's stuttering overall. Interestingly, although there were increases in parents' knowledge and confidence across time and decreases in children's stuttering frequency over the same period, these two variables were not consistently correlated. To understand the seemingly complex relationships between and among the outcome measures and particularly that of stuttering severity and parents' knowledge and confidence, we conducted a hierarchical multiple regression (a) to determine whether Factors 1 and 2 predicted Factor 3 (Model 1) and (b) to examine whether stuttering frequency predicted parents' knowledge and confidence over and above Factors 1 and 2 (Model 2). We decided to examine these relationships at two time points—T1 (pretherapy) and T3 (6 months after therapy commenced).

Mothers

At T1, there was a significant correlation between Factors 1 and 2 and Factor 3 ($p = .013$ and $p < .001$, respectively). The correlation between %SS and Factor 3 was not significant ($p = .077$). Results of the regression analysis indicated that Factors 1 and 2 explained 38% of the variance in Factor 3 at T1 (with a significant change in adjusted R^2 (0–7.6; $p = .0001$). When %SS was added as a covariant (Model 2), the change in adjusted R^2 ($p = .004$) is not significant.

At T3, there was a significant correlation between Factors 1 and 2 and Factor 3 ($p < .001$). The correlation between %SS and Factor 3 was not significant ($p = .155$). In Model 1, Factors 1 and 2 again explained 38% of the variance in Factor 3 at T3 with a significant change in R^2 ($p < .05$) from 0 to 17.4. When %SS was added as a covariant, there was also a significant increase in adjusted R^2 ($p < .05$), from 3.8 to 4.5.

Fathers

At T1, there was a significant correlation between Factors 1 and 2 and Factor 3 ($p = .09$ and $p < .001$, respectively). The correlation between %SS and Factor 3 was not significant ($p = .069$). Hierarchical regression indicated that Factors 1 and 2 predicted Factor 3 at T3 with a significant increase in adjusted R^2 ($p < 05$; 0–10.6). When %SS was added as a covariant, the change in adjusted R^2 from.263 to .252 was not significant.

At T3, there was a significant correlation between Factors 1 and 2 and Factor 3 ($p < .001$), whereas the correlation between %SS and Factor 3 was not significant ($p = .115$). Hierarchical regression revealed that Factors 1 and 2 explained 41% of the variance in Factor 3 at T3 with a significant change ($p < .05$) in adjusted R^2 (from 0 to 19.43). When %SS was added as a covariant, there was a significant increase ($p < .05$) in adjusted R^2 (from .412 to .451).

The findings demonstrate that the severity of the child's stuttering, as measured by %SS, does not predict the parents' level of knowledge and confidence at T1. However, at T3 where Factors 1 and 2 account for almost half of the variance in parents' knowledge and confidence, the child's stuttering behavior significantly adds to the predictive strength of these two factors.

Response to Intervention

There were 50 children who contributed at least two variables at T1 and at least one time point posttherapy. Therefore, to be considered as a "most successful outcome," each participant needed to achieve what we deemed to be a clinically significant change in at least two of a potential five variables. Thirty-five children were considered to have a "most successful outcome" on the basis of these criteria, with 15 not demonstrating a clinically significant change in two variables (see Table 2).

Outcome Measure Variables

A two-tailed independent *t* test revealed no significant difference between the "most successful" ($M = 7.05$, $SD = 12.11$) and "less successful" ($M = 2.96$, $SD = 2.09$) groups in terms of stuttering frequency at T1, $t(26) = 0.88$, $p = .39$, although there is clearly greater variance within the "more successful" group. Neither was there a significant difference in mothers' Palin PRS Factor 1 scores between the "more successful" ($M = 5.06$, $SD = 1.29$) and "less successful" ($M = 5.71$, $p = .73$) groups, $t(47) = 0.07$, $p = .74$. However, there was a significant difference between the two groups with regard to mothers' ratings on Factor 2, $t(47) = -3.41$, $p = .001$, and Factor 3, $t(47) = -2.11$, $p = .04$, at T1, with mothers in the "most successful" group rating the stuttering as more severe and being more worried (Factor 2: $M = 3.13$, $SD = 1.32$) and having less knowledge and confidence (Factor 3: $M = 4.06$, $SD = 1.78$) at T1 compared with those who made less change (Factor 2: $M = 4.51$, $SD = 1.29$; Factor 3: $M = 5.22$, $SD = 1.78$).

Child Factors

In terms of age at the start of therapy, there was no significant difference between the "most successful" ($M = 51.31$, $SD = 12.96$) and "less successful" ($M = 55.87$, $SD = 16.57$) groups on the basis of a two-tailed independent *t* test, $t(48) = -1.46$, $p = .30$. The vast majority of both groups of children had no other diagnoses. In the "most successful group," there was one child who had dyspraxia and another child with attention-deficit/hyperactivity disorder. Proportionally, there was a small difference between the groups with regard to girls and boys. There was a slightly higher ratio in the successful group, with almost a 3:1 male-to-female ratio, compared with 2:1 in the "less

Table 2. Epidemiological data for children categorized as having more or less successful outcomes on the basis of a clinically significant change in two or more variables.

Variable	"Less successful" (*n* = 15)	"More successful" (*n* = 35)
Male	10 (66.7%)	26 (74.1%)
Female	5 (33.3%)	9 (25.7%)
Phonological delay/disorder		
No	9 (60%)	17 (53.1%)
Yes	6 (40%)	15 (46.9%)
Language level		
Within normal range	5 (33.3%)	8 (22.9%)
Above normal range	7 (46.7%)	10 (28.6%)
Below normal range	1 (6.7%)	9 (25.7%)
Mismatch	2 (13.3%)	8 (22.9%)
Comorbid diagnoses		
None	13 (86.7%)	29 (87.9%)
Dyspraxia		1 (3%)
ADHD		1 (3%)
Other	2 (13.3%)	2 (6%)
Maternal history of stuttering		
None	13 (86.7%)	28 (82.3%)
Persisted		4 (12.5%)
Recovered	2 (13.3%)	2 (6.3%)
Paternal history of stuttering		
None	14 (93.3%)	29 (85.3%)
Persisted		3 (9.4%)
Recovered	1(6.7%)	2 (6.3%)
English as a second language		
Yes	10 (66.6%)	24 (68.6%)
No	5 (33.3%)	11 (31.4%)
Mean age at the start of therapy	55.87 (*SD* = 16.57)	51.31 (*SD* = 12.96)

Note. ADHD = attention-deficit/hyperactivity disorder.

successful" group. This suggests that the outcome over this 1-year period was slightly less likely to be successful for girls than it was for boys.

Evidence of phonological delay or disorder was similarly prevalent in both the "most successful" and "less successful" groups. Language level was measured using the Clinical Evaluation of Language Fundamentals–Preschool. Children were categorized as within normal range, above and below according to the results of that test. Children who had a mixed profile across the subtests were categorized as "mismatched." It is interesting to note that there was a noticeably larger proportion of children with advanced language skills in the "less successful group," compared with the successful group. This does not mean that children who have advanced language skills will not improve, because we can see that there are also children in the "successful group" who were considered to have above-average language skills, and indeed there are data in Millard et al. (2009) to support this empirically. However, there was a more even spread of language ability within the "more successful" cohort.

Approximately one third of children in each group had English as an additional language, suggesting that this is not a factor that influences outcome. However, this dichotomous distinction of bilingual or not is a rather crude categorization. Bilingualism is considered to be a continuum, with exposure and use being critical factors in relation to language development (Byrd, Bedore, & Ramos, 2015). However, this finer differentiation was not collected as part of this study.

Most children in each of the groups had no family history of stuttering, but only the "more successful" group had within it parents who had persistent stuttering. One child in the "more successful" group had both a father (recovered) and a mother (persistent) who stuttered.

Discussion

Data from three perspectives contributed to the exploration into the outcomes over a 1-year period in children who received Palin PCI therapy (Kelman & Nicholas, 2008). Change was considered from the view of the child, parents, and clinician with respect to overt stuttering and the impact that it had on the child's confidence and ability to communicate and participate in speaking situations. Furthermore, parents' confidence in their ability to support the child was also examined across the therapy process and beyond. The findings demonstrate significant improvements over time for all the variables studied, although the pattern of change was not linear, with a significant change in all measures occurring at differing times in the process. That is, results indicate that some changes occur earlier and others occur later in the process and are likely to be observed at different times by children, parents, and clinicians.

Stuttering Frequency and Severity

The children were significantly more fluent at the end of the year compared with the start according to the stuttering frequency data. The temporal course of this reduction does not coincide with prior research by our group, where reductions in stuttering frequency were evident within 3 months of starting therapy for most (Millard et al., 2008, 2009). On the other hand, parents are perceiving improved fluency earlier than evidenced by the clinic frequency measures, with reductions in stuttering severity and impact on them as demonstrated by Factor 2 measurements closely after the onset of therapy (T2). There are a number of possibilities that might explain these differences. First, Factor 2 reflects the parents' observation of the child's fluency beyond the clinic, while the %SS measures were obtained from in-clinic speech samples. Furthermore, while the parents are observing spontaneous conversational speech in a typical context for the child, the in-clinic samples are recorded while the child is describing a series of pictures that depict nonsensical situations. These descriptions are arguably more cognitively and linguistically demanding than daily conversation with family and peers. For some young CWS, a more linguistically demanding task or complex utterance can result in a greater number of disfluencies (Ratner, 1995; Yaruss, 1999). Another potential explanation can be linked to the fact that Palin PCI is a home-based therapy, with parents practicing and implementing strategies at home. Therefore, generalization in this instance might be considered to take place from home to clinic, rather than from clinic to home. It would be expected that change would be first observed in the context in which it is practiced or experienced most. One further interpretation is that parents are evaluating severity on the basis of parameters that are in addition to or independent of stuttering frequency. In which case, the change in Factor 2 may be due to reduced struggle and tension. The final possibility is that change in Factor 2 is due to a reduction in parental anxiety rather than stuttering because those are both contained within Factor 2.

The findings from previous studies (Millard et al., 2008, 2009) demonstrated a significant reduction in stuttering frequency within 3 months of the start of therapy. Differences between the studies may once again be explained by the difference in speech samples (previous studies used recordings from home). It is also possible that the differences between the studies could be explained by differences in the populations being studied. The current study included all children who received the intervention, whereas the previous efficacy eliminated bilingual children, those with concomitant diagnoses, and those with developmental language delay. The current population is more consistent with a general clinical population who would receive this intervention.

Impact of Stuttering on the Child

There was a significant reduction in the KiddyCAT (Vanryckeghem & Brutten, 2007) scores at each time point compared with pretherapy levels. This demonstrated an improvement in the children's views of themselves as communicators and specifically how difficult it is for them to speak (Clark et al., 2012). No change between T3 and T4 indicated that the improvements were maintained over the long term. These improvements are clinically important given the finding that these scores typically increase with age, rather than decrease with time and age (Vanryckeghem & Brutten, 1997). It is interesting to note that this improvement occurred despite the lack of significant change in %SS and suggests that children's attitude to speaking can be improved even when stuttering does not significantly reduce. It might also suggest that there is a change in the degree of struggle and effort that is required to speak, which is not reflected in the stuttering frequency scores but is reflected in the Palin PRS Factor 2. Stuttering frequency was selected as an outcome measure, as this is the most consistently reported outcome measure in clinical trials, but it is possible that a stuttering severity score, which incorporates duration and struggle, may have revealed an earlier change in the clinical stuttering measure.

The parents' perception of the impact of stuttering on the child was measured through Factor 1 of the Palin PRS. Mothers showed significantly improved scores between the start of therapy and 6 months, whereas fathers exhibited statistically significant scores between each time point and T4. Parents were therefore noticing a reduction in the impact that the stutter has on the child between 6 and 12 months after the start of therapy. This is later than is observed by the children themselves, according to the KiddyCAT scores. This suggests that it takes longer for the parents to observe a reduced impact than the children. One possibility is that the children are considering an internal sense of the difficulty of speaking, which takes longer to translate into observable affective or behavioral reactions. Another possibility is that parents are expecting or looking for more tangible or dramatic changes to consider impact to have reduced and are less sensitive to small or subtle changes.

Parents' Perceptions of the Severity of Stuttering and Impact That the Stuttering Has on Them

The psychometric evaluation of the Palin PRS demonstrated that the individual scales in these areas were measuring one construct and that these were highly correlated and inseparable (Millard & Davis, 2016). Mothers show an early change in their perception of the severity of stuttering and their worry about it. This reduces significantly between each of the time points until T3. There is no difference between T3 and T4, which indicates that this is maintained for the remainder of the year. Fathers also demonstrate a significant reduction in this variable early in the process, with reduction associated with the onset of therapy (T1–T2), holding steady between T2 and T4, before further improvement at T3–T4. This change is important as it reflects parents' observations of speech beyond the clinic and shows that parental worry is one of the first changes that might be expected with therapy. This is

reassuring given that a central aim for Palin PCI is that parents should feel less worried and anxious and suggests that parents have confidence in the therapy process and the potential long-term outcome for their children.

Parents' Knowledge About Stuttering and Confidence in Managing It

A small number of studies have been conducted exploring what parents experience when they have a child who stutters and identified guilt, anxiety, worry for the child's future, helplessness, and a lack of confidence in their skills (Langevin et al., 2010; Plexico & Buruss, 2012). In the development of the Palin PRS, parents were asked to consider important outcomes for therapy, and they identified that reduced parental anxiety, worry about the future, and improved knowledge about how to support the child were important gains from therapy. In this study, increases in Factor 3 were reported early in the process by both mothers and fathers. In contrast to what might be anticipated, this increase can and does take place when there is no commensurate reduction in the impact of stuttering on the child or a reduction in %SS scores.

One of the primary aims for Palin PCI is for parents to feel empowered to support their child and gain some strategies that will help promote fluency and effective communication between themselves and the child. Because this appeared to improve ahead of the other aims (increased fluency and reduced impact), it was decided to explore the relationship between these variables further, specifically whether %SS would predict Factor 3 over and above Factors 1 and 2. It was anticipated that, because improvement in Factor 3 occurred seemingly without improved %SS, there would be no relationship. Stuttering frequency was not found to be predictive for Factor 3 at T1, which means that the degree to which parents are confident in supporting their child is independent of the stuttering frequency in the clinic samples. It was predicted that, after therapy, this lack of relationship would persist; however, %SS was found to contribute to Factor 3 at T3. Therefore, parents of children who were more fluent at this point felt more confident in their skills. Given that stuttering frequency has significantly reduced by this point in time, this may be further interpreted to suggest that it is the parents' ability to notice this improvement that is important.

Response to Intervention

We were interested to explore whether there were any obvious differences between the children who did most well versus those who did least well. To be clear, the study was not designed to investigate this issue per se, and therefore some of the variables (e.g., bilingualism, presence of phonological difficulties, and language levels) were only broadly defined. It is also important to recognize that "less successful" is not the same as "unsuccessful." Children allocated to the "less successful" group may still have benefited from the intervention; it is only that they did not reach the criteria set for two of the measures. Nevertheless, the findings are interesting. Factors such as phonological delay, bilingualism, and age were included in the exploration because it was anticipated that they may have a role in outcome and did not differ between the groups. Comparing the two groups, the "more successful" group had proportionally fewer girls, more children with language delay, and more children with a parent who had a persistent stutter. This is interesting given the literature on risk of persistence factors (Yairi & Ambrose, 2013). Although we are not suggesting that all of these children are recovered, because we based our criteria on improvement in two measures, the information does provide reassurance for parents and indeed therapists that therapy can be helpful for children who might otherwise receive a more pessimistic prognosis.

It seems that mothers who are more concerned (Factor 2) and who have the least confidence (Factor 3) are more likely to have the more successful outcomes. There are a number of possible explanations for this. The first is that those with more negative ratings have more changes to make and so are more likely to make the necessary shift in two variables required by the categorization. It is also possible that those who are more worried, view the stutter as more severe, and who are not confident about what to do may engage more fully with therapy and effect the greatest change. Because engagement with therapy was not measured, it is not possible to explore this further in this instance.

These improvements have been achieved with a mean number of six sessions of clinic-based therapy. This compares favorably with the results of the RESTART trial in which children received approximately 20 sessions of intervention within each program (de Sonneville-Koedoot et al., 2015). It is important to note that Palin PCI differs from these programs in that intervention does not continue until a predetermined outcome is achieved in terms of stuttering frequency, and this may explain the difference in session numbers. A cross-comparison of treatment dose and outcome between Palin PCI and other programs would provide further insight into the relative cost-effectiveness of each. The evidence that these three programs can result in improved outcomes suggests that there are common factors that account for the improvements. Understanding the mechanisms of the relationships between the variables studied would help to unravel the areas that should be prioritized and targeted in therapy.

Limitations

The data in this study were collected from a group of children attending Palin PCI and monitored over a year. One of the limitations of this study is that there were missing data across participants, time points, and measures—a pervasive problem for clinical researchers that requires careful attention, particularly when the goal is to make every attempt to use all the data that are available (Haukoos

& Newgard, 2007). We recognize that using series mean imputation procedures can lead to valid estimates of treatment differences but, at the same time, can underestimate the variability in the data and increase the probability of a Type I error. That said, because this study was not a clinical trial, we decided to use this more straightforward process in our within-group repeated-measures analyses.

There was definite skew in the frequency data at T1, with 29 children classified as mild or moderate, compared with 19 classified as severe or very severe. There were some who did not stutter during the speech sample task, despite stuttering on other occasions. These data reflect the reality of the variability of stuttering in young children, but this causes two problems for this study. The first is that there may be overrepresentation of mild stuttering, and the second is that it is impossible to detect any reduction if they are already at base level. The absence of a control group is also a limitation, meaning that change cannot be directly attributed to the therapy program. It is important to view the findings as an insight into how a group of CWS progress over a year, when they receive Palin PCI. The large numbers in this study increase confidence that the findings are valid and likely to be the result of the intervention. The participants are drawn from one center, and it is possible that the population, although diverse, is not necessarily representative of the whole clinical population of CWS.

Finally, we are not attempting to make any strong claims about any differences between children who were considered to have a more or less successful outcome over the year. There were a number of variables that would need to be more refined, and the reliability of the data needs to be established, but the data provide an interesting insight into possible influencing variables with regard to the impact of treatment and demonstrate that the factors that are prognostic for natural recovery in a nonclinical population are not likely to be the same as those that are related to therapy outcomes in a clinical population.

Conclusions

The data demonstrated that, over a year, children who received Palin PCI therapy experienced a significant reduction in stuttering frequency and impact of stuttering. In addition, the children's perception of themselves as communicators improved and they found it easier to speak. Parents felt less worried and anxious about the stuttering and more knowledgeable and confident in how to support their child. The findings suggest that there is a process of change that occurs, with improvements in these areas occurring at different times. Improvements are maintained over the period of a year. The comparison between those who had a more successful outcome provides encouragement and reassurance that children who do not have higher risk of persistence profiles for recovery can still benefit from therapy. These findings provide further support for Palin PCI as an effective intervention for young CWS.

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