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Palin parent-child interaction therapy with children with autism spectrum disorder and stuttering

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ABSTRACT

Introduction: It is estimated that 8% of children who stutter (CWS) have autism spectrum disorder (ASD) Briley & Ellis (2018). There is evidence that interventions for CWS and interventions for children with ASD can be effective, but there is little evidence to guide clinical decision making when working with CWS with a co-existing diagnosis of ASD. Palin Parent–Child Interaction (PCI) therapy Kelman & Nicholas (2020) is an evidence-based intervention for CWS, with the authors suggesting that the approach may be beneficial for CWS with ASD. The aim of this study was to examine outcomes for three CWS with ASD who received Palin PCI at a specialist centre for stuttering in London.

Method: The participants were three CWS with ASD aged 4;5, 6;7 and 7;7. Assessments were administered before therapy, and then at three, six and twelve months after therapy began. Outcome measures included stuttering frequency, child’s communication attitude, parents’ perception of the impact of stuttering on the child, the severity of stuttering and its impact on the parents, and parents’ knowledge and confidence in managing stuttering.

Results: All three children showed improvement in three or more variables. Four out of five parents reported reduced impact of stuttering on the child and themselves following therapy, and change was maintained one year post-therapy. All five parents reported increased knowledge of stuttering and confidence in managing it after therapy, and four parents maintained these changes for a year.

Conclusions: Over a one year period, these CWS with ASD who received Palin PCI showed change across multiple variables. The observed increases in parent knowledge and confidence were comparable to previously published data. These preliminary findings suggest that CWS with ASD and their parents can benefit from Palin PCI therapy and that further experimental evaluation of this approach with this client group is indicated.
communication and unusually restricted, repetitive behaviour and interests (American Psychiatric Association, 2013). It may present with or without intellectual disability, and with or without language impairment (Lai et al., 2014). Shriberg et al. (2001) first identified that stuttering and other forms of dysfluency may occur within adults with ASD. A strong genetic component is evident for both ASD and stuttering (Tick et al., 2016; Yairi & Ambrose, 2013). Subsequent studies have identified stuttering in preschoolers with ASD (Plexico et al., 2010), school-aged children with ASD (Scott et al., 2014; Sisskin, 2006; Sisskin & Wasilus, 2014), and adolescents with ASD (Sisskin, 2006).

1.2. Co-occurrence of stuttering and ASD

The only available estimates for the prevalence of stuttering amongst the population with ASD are based on data from the United States National Health Survey. Data reported from 1997 to 2005 indicates that 15.6% of children with ASD also stuttered compared to 1.6% of the total population sample of children aged 3 to 17 years (Boyle et al., 2009). Data collected from the same US National Health Survey from 2010 to 2015 found that 8% of CWS were reported to have ASD (Briley & Ellis, 2018), compared with 1.7% in the wider population (Baio et al., 2014). Since this population sample included CWS aged 3 to 17 years old, it is possible that more than 8% of CWS have ASD given that ASD is often not diagnosed until school age (Daniels & Mandell, 2014). Smith et al. (2017) also found a significantly higher percentage of children with ASD in their group of Australian children who had a persistent stutter (20%) compared to children with recovered stuttering status (4.3%) and children who never stuttered (5.9%). In summary, there is a higher incidence of stuttering in the ASD population, and a higher incidence of ASD in the stuttering population than would be expected if there were no association between the conditions.

1.3. Impact of stuttering and ASD on children

The experience of stuttering can negatively impact a child’s attitude to speaking (Vanryckeghem & Brusen, 2007). From three years old, CWS can demonstrate more negative attitudes towards speaking than children who do not stutter (CWNS) and as a group their communication attitude becomes increasingly negative with age (Clark et al., 2012; Vanryckeghem et al., 2005). A negative communication attitude may result in CWS avoiding speaking and this behaviour would appear to have its roots in the preschool years (Langevin et al., 2009). While the impact of stuttering on children with ASD is not known, there is evidence that some CWS with ASD present with affective and cognitive components of stuttering (Scott, 2018).

Furthermore, CWS and children with ASD are both populations at risk of developing anxiety disorders (Iverach et al., 2016; van Steensel et al., 2011), and it is therefore possible that this clinical subpopulation of CWS with ASD are at even greater risk of experiencing anxiety disorders. The community cohort study of school-age children in Australia suggested that the association between anxiety and stuttering was more pronounced in CWS with ASD and/or intellectual disability (Smith et al., 2017).

There are additional social consequences of being a child with a communication disorder. From preschool age, children may choose to communicate less with children with a communication impairment (Gertner et al., 1994; Langevin et al., 2009). Nearly a third of parents report that their preschool child who stutters has experienced teasing (Langevin et al., 2010). Given that children with ASD have inherent difficulties with social interaction, and experience high rates of bullying (Rowley et al., 2012), the risk of teasing and social isolation in CWS with ASD may be further elevated.

1.4. Impact of stuttering and ASD on parents

Stuttering can have a significant impact on parents (Millard & Davis, 2016; Plexico & Burrus, 2012). Over 90% of parents of preschool CWS report being negatively affected by their child’s stuttering (Langevin et al., 2010). Parents have reported fear that their child will have negative experiences or live a restrictive lifestyle as a result of stuttering and expressed uncertainty about how best to support them (Plexico & Burrus, 2012).

The variety of difficulties that a child with ASD may present with are associated with decreased parenting self-efficacy, increased parenting stress and reduced overall family wellbeing, compared with parents of both typically developing children and children with other developmental disorders (Kast & Van Hecke, 2012).

In summary, CWS and children with ASD share a number of characteristics. They are at greater risk of experiencing anxiety disorders (Iverach et al., 2016; van Steensel et al., 2011) and difficulties with peer relationships (Gertner et al., 1994; Langevin et al., 2009; Langevin et al., 2010; Rowley et al., 2012). These conditions each have an impact on the wider family, parents in particular (Millard & Davis, 2016; Plexico & Burrus, 2012; Kast & Van Hecke, 2012). While the cumulative effect of being a child with ASD who stutters is not known, it is clear that these conditions co-occur at a higher than expected frequency and a number of these children present at speech and language therapy clinics for therapy. Therefore, there is a clear need for effective intervention approaches.

1.5. Evidence-based interventions for children with ASD

There has been considerable interest in early intervention programmes for children with ASD. A systematic review of interventions for preschool children with ASD identified 48 randomised control trials (RCTs) evaluating 32 different models of intervention (French & Kennedy, 2018). The ultimate aim of therapy for young children with ASD is to develop their spontaneous functional communication skills.

Only two studies identified in French and Kennedy’s systematic review (2018) met criteria for low risk of bias and demonstrated
treatment effects at long-term follow up. Both were RCTs of the Preschool Autism Communication Therapy programme (PACT; Green et al., 2010; Green et al., 2015). PACT aims to increase the child’s communication initiation and language by adapting parents’ interaction style to the child’s strengths and needs. Video reflection is used as a review tool. PACT has six stages that follow a developmental trajectory, starting with establishing joint attention and developing parental sensitivity and synchronicity. Subsequent stages include establishing routines and increasing the child’s communicative functions. Parents who received PACT therapy demonstrated more sensitive responses to their children’s communication, and this led to the child initiating communication more frequently (Green et al., 2010).

1.6. Palin parent-child interaction therapy for CWS

There is a body of evidence supporting a range of interventions for pre-school CWS, including Palin PCI (Kelman & Nicholas, 2020). This is a multifactorial approach supported by multiple single-subject replicated studies (Millard et al., 20082009) and a larger cohort pre-post intervention study that included children with co-existing developmental disorders (Millard et al., 2018). Palin PCI seeks to establish the building blocks for confident and competent communication whether or not the child continues to stutter (Kelman & Nicholas, 2020). The aim of Palin PCI is threefold: to enhance the child’s fluency and ability to speak without struggle; to reduce the impact of stuttering on the child and parents; and to increase the parents’ knowledge about stuttering and their confidence in managing it. Palin PCI involves six indirect therapy sessions in clinic followed by a six-week consolidation period and, if necessary, further indirect therapy or more direct therapy sessions with the child. There are three aspects to the programme: Interaction Strategies; Family Strategies; and, Child Strategies, details of which can all be found in the manual (Kelman & Nicholas, 2020).

Interaction Strategies: Using video as a feedback tool, parents identify interaction strategies they are already using that support their child’s fluency and confident communication. Parents are encouraged to focus on using these interaction strategies during ‘Special Time’ with their child at home, three to five times a week. In Special Time, the parents play with their child for five minutes, letting the child choose the activity and giving the child their uninterrupted attention. Strategies might include: increasing pausing; following the child’s lead in play; or, reducing rate of speech.

Family Strategies: These are introduced to build the child’s confidence and to foster a positive attitude to communication in the child and a non-judgmental and accepting attitude to stuttering in the home. Strategies might include: the use of praise; behaviour management strategies; turn taking in the family; or, talking openly about stuttering.

Child Strategies: While the aims are largely achieved with the indirect therapy components alone (Millard et al., 20082009), child strategies may be introduced when children are struggling to manage the moment of stuttering or trying to make changes to their speech. These might include activities which educate the child about talking and stuttering, encourage openness and desensitisation to stuttering, as well as speech strategies, such as increased pausing or reduced rate, aimed at reducing struggling behaviours.

1.7. Fluency interventions for clients who stutter with ASD

There is little published literature documenting fluency interventions for CWS with ASD. The existing literature explores the presence of atypical dysfluencies among CWS with ASD (Healey et al., 2015; MacFarlane et al., 2017; Plexico et al., 2010; Scott et al., 2014; Sisskin, 2006). Whereas in typical developmental stuttering, dysfluencies occur primarily at the beginning of words or occur over the entire word, atypical dysfluencies occur at the end of words, for example, final syllable repetition. The literature shows that children with ASD can present with typical and/or atypical stuttering behaviours, with varying levels of awareness of their stuttering.

Given the lack of empirical evidence to support decision making, clinicians supporting CWS with ASD have reported being unsure which fluency interventions are appropriate (Scott et al., 2015). CWS with co-existing conditions have either been excluded from RCTs that require homogenous groups (de Sonneville Koedoot et al., 2015), or their individual profiles and outcomes have not been reported in group studies (Millard et al., 2018). As far as these authors have been able to ascertain, there is only one single-case study investigating the effectiveness of a fluency intervention for an adult with ASD and one single-case report for a child with ASD.

Brundage et al. (2013) examined the effectiveness of a modified version of the Fluency Rules Programme (Runyan & Runyan, 2007) in decreasing stuttering in a 21-year-old male with ASD. The participant presented with typical stuttering and was unaware of his stuttering at his evaluation prior to the study. The treatment phase involved the participant being taught three speaking rules: speak slowly; say each word only once; and say each sound only once. These rules were applied to utterances of increasing length and complexity. The primary outcome measure was the percentage of stuttered words, which reduced from 14.57% to 3% after the treatment phases. The study suggests that the fluency rules programme has promise in reducing stuttering frequency in adults when ASD is co-occurring.

Sisskin and Wasilus (2014) published a case report of a 7-year-old boy with traits of ASD who had stuttered since 4 years of age. He presented with atypical dysfluencies which included word-final repetitions. He was formally diagnosed with Asperger syndrome after therapy. He scored 1.66 on the Overall Assessment of the Speaker’s Experience of Stuttering (OASES) (Varuss & Quesal, 2016), indicating a mild/moderate life impact. The aim of the therapy was to reduce the frequency of his atypical dysfluencies. Thirteen treatment sessions incorporated the identification and correction stages of block modification (Van Riper, 1973), with teaching strategies adjusted for the learning style of children with ASD. Post-treatment measures indicated the atypical dysfluencies reduced from 12.7% syllables stuttered (SS) averaged over two baseline measures to an average of 1.45% SS over three post-treatment measures. The authors report the child’s stuttering reduced almost entirely through the identification and self-monitoring stage of block modification and remained under 2% SS for the following two years. There was no change on the OASES, indicating that increased fluency did not reduce the impact of stuttering on the child. The authors reflected that both the nature of the child’s stuttering and his
quick response to treatment suggest this case does not reflect typical developmental stuttering.

Both of these studies have focused on speech modification strategies, with stuttering frequency as the primary outcome measure. For preschool CWS, there is evidence that both behavioural approaches, such as the Lidcombe Programme (Jones et al., 2005) and multifactorial approaches, such as Restart DCM (de Sonneville Koedoot et al., 2015) and Palin-PCI are efficacious. However, there is no published empirical evidence to support that any of these widely implemented published programmes are effective for CWS with ASD.

1.8. The current study

Given the lack of representation in research, there is a need to build evidence to support dysfluency interventions for CWS with ASD. Current interventions for children with ASD have many goals and methods in common with Palin PCI, an intervention that the authors of the programme propose to be successful based on their clinical experience (Kelman & Nicholas, 2020).

The current study explores profiles of change in three children with these co-existing conditions, over the period of a year, to see if there is evidence to support the use and further evaluation of Palin PCI, and to observe whether these children with ASD make changes that are similar to those reported in the efficacy data for Palin PCI (Millard et al., 2018).

The current study is a preliminary investigation of retrospective clinical data and represents the first step in the research process (low level 4 evidence; OCEBM Levels of Evidence Working Group, Howick et al., 2011). Building the evidence to inform practice begins with clinical reports and case studies of interventions showing potential therapeutic gains, indicating that further investigation is worthwhile. Case studies provide a level of detail which is valuable in relating individual profiles with intervention effects. This is important given the heterogeneity of this clinical subpopulation, which encompasses children with ASD with a broad range of needs.

2. Method

Clinical data at a specialist centre for CWS in London was explored retrospectively to examine outcomes for CWS with ASD. Clinical data at this centre are routinely collected and stored in accordance with national data management governance guidelines. Children with a recorded diagnosis of ASD were identified on the database and those who had assessment scores recorded for at least four time points were extracted from the dataset and analysed. This study received ethical approval from City University of London [ETH1920 0232].

2.1. Participants

The participants in this study are three CWS with a diagnosis of ASD who attended this centre for specialist dysfluency assessment and intervention. All three children qualified for a diagnosis of stuttering at initial assessment as per the criteria described by Onslow (2021); the children presented with unambiguous stuttering moments, moments that, to an observer, are clearly stuttering and not normal disfluency. Stuttering moments included whole-word and part-word repetitions, blocks and prolongations, with or without tension. Observations were made by speech and language therapists specialising in stuttering and supported by parents’ descriptions of stuttering behaviours beyond the clinic. No atypical dysfluencies were observed or reported in the speech of any of the children included in this study. In addition to fluency assessments, clinicians completed a language evaluation at the time of initial assessment, by administering the core subtests of the Clinical Evaluation of Language Fundamentals-Fourth Edition (CELF-4; Semel et al., 2003) or Clinical Evaluation of Language Fundamentals-Preschool-Second Edition (CELF-P2; Wilg et al., 2006), appropriate for the child’s age. The purpose of formal language assessment is to provide insight into the child’s functional language skills. Further subtests of the CELF-4 or CELF-P2 may be administered based on the child’s presentation and their attention capability. Normative data for the CELF-4 and CELF-P2 are based on a monolingual English population sample and a scaled score of 8-12 is within the average range. For the two bilingual children in this study, formal language assessment in English was used to gain an understanding of the child’s functional language skills in their English speaking environments, including their school and wider society. Standard scores were not intended to be used to make a diagnosis of developmental language disorder for these bilingual children.

As per the findings by Coulter et al. (2009), a child’s language skills are described as ‘mismatched’ if there are dissociations between subcomponents of language, for example, average or above average expressive language skills and below average receptive skills. Participant characteristics are reported in Table 1.

Child 1, aged 4;5 at initial assessment, had been stuttering for two years at the time of initial assessment. He was bilingual. At home, he was exposed to two languages, his mother’s first language and English. At school and outside of home, he was exposed to English. His mother had sufficient English to conduct therapy sessions in English without any support from an interpreter. There was no family history of stuttering. His CELF-P2 subtest scores indicated mismatched language skills (Sentence Structure = 9; Word Structure = 9;

Table 1

<table>
<thead>
<tr>
<th>Child</th>
<th>Age at initial assessment (years; months)</th>
<th>Sex</th>
<th>Time since onset of stuttering (months)</th>
<th>Family history of stuttering</th>
<th>Bilingual</th>
<th>Previous Therapy</th>
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<td>1</td>
<td>4;5</td>
<td>Male</td>
<td>24</td>
<td>No</td>
<td>Yes</td>
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</tr>
<tr>
<td>2</td>
<td>6;7</td>
<td>Male</td>
<td>10-12</td>
<td>Yes</td>
<td>No</td>
<td>Yes</td>
</tr>
<tr>
<td>3</td>
<td>7;7</td>
<td>Male</td>
<td>42</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
</tr>
</tbody>
</table>
Concepts and Following Directions = 4; Expressive Vocabulary = 5). Before the start of therapy, his stuttering frequency was 5.7% SS (averaged over two baseline measures). He attended therapy with his mother.

Child 2, aged 6;7 at initial assessment, had been stuttering for 10–12 months. He had a family history of persistent stuttering. He had received therapy for dysfluency from his local SLT service prior to referral to this centre. His scores on the CELF-P2 revealed his receptive language was below the average range for his age (Sentence Structure = 6; Concepts and Following Directions = 6; Word Classes – Receptive = 6), while his expressive language was borderline (Expressive Vocabulary = 7; Word Structure = 7; Recalling Sentences = 7). In addition to stuttering, he presented with articulation difficulties, notably lateralisation of the speech sounds /s/ and /z/ and reduced /s/ clusters. His connected speech was described as ‘often unintelligible out of context’. While attending Palin PCI therapy, Child 2 continued to receive articulation therapy from his local SLT service. He had a history of middle ear infections but no hearing difficulties at the time of assessment and therapy. Before the start of therapy, his stuttering frequency was 1.5% SS (averaged over two baseline measures). Both parents attended the course of therapy.

Child 3, aged 7;7 at initial assessment, had been stuttering for 3.5 years. He was bilingual. At home, he was exposed to two languages, his parents’ first language and English. At school and outside of home, he was exposed to English. Both parents had sufficient English to conduct therapy sessions in English without any support from an interpreter. He had a family history of persistent stuttering and had previously received therapy for dysfluency from his local SLT service before being referred to this centre. His CELF-4 subtest scores indicated mismatched language skills; Concepts and Following Directions = 6; Formulated Sentences = 13; Word Structure = 9; Recalling Sentences = 6. The average range is 8-12, a score of 7 is 1SD below the mean and 13 is 1SD above. In addition to stuttering, he presented with a lisp. Before the start of therapy, his stuttering frequency was 17% SS. Both parents attended the course of therapy.

2.2. Outcome measures

The following assessments are part of a comprehensive assessment battery completed by all children and their parents attending this centre, for the purposes of clinical decision making.

Stuttering frequency

Stuttering frequency was derived through analysis of a conversational speech sample elicited through a picture description task using ‘What’s wrong’ cards (LDA Publishing, 1999). The speech sample were analysed by Speech and Language Therapists, or a Research Assistant trained in the procedure, to determine the percentage syllables stuttered (%SS). Whole-word and part-word repetitions, blocks and prolongations were included in the counts.

Child’s perception of stuttering and communication

Self-evaluation measures were given to each child depending on age and ability. Child 1 and Child 2 completed the preschool version of the Communication Attitude Test (KiddyCAT; Vanryckeghem & Brutten, 2007). The KiddyCAT is a standardised 12-item, binary response questionnaire, designed to assess preschool-age children’s attitudes towards their communication. It is suitable for children aged 3-6 years old. Higher scores reflect a more negative attitude to speaking. For the children in this study, communication attitude may be affected by both their stuttering and communication difficulties related to their ASD.

Child 2 turned 7 years old over the course of therapy and therefore completed the CAT for the final two assessment time points. The CAT (Brutten & Vanryckeghem, 2007) has 33 statements that have to be marked as true or false. Example statements include: ‘I do not talk right’; and ‘I do not mind asking the teacher a question in class’. Like the KiddyCAT, higher scores reflect a more negative attitude to speaking, which again may be impacted by both stuttering and ASD.

Child 3 completed both the CAT and the Overall Assessment of the Speaker’s Experience of Stuttering for school-age children (OASES-S; Yaruss & Quesal, 2016). The OASES is a standardised self-report questionnaire, for CWS aged 7-12 years old, assessing the child’s reaction to stuttering, impact on daily communication and impact on quality of life, with higher scores reflecting a higher impact on the child.

Parents’ perception of stuttering severity and impact on the family

The Palin Parent Rating Scale (Palin PRS; Millard & Davis, 2016) was used to evaluate stuttering impact and severity from the parents’ perspective. The Palin PRS is a standardised measure of three constructs (hereafter referred to as factors): the impact of stuttering on the child (Factor 1); the severity of stuttering and its impact on the parents (Factor 2); and the parents’ knowledge about stuttering and their confidence in managing it (Factor 3). The Palin PRS is a 19-item questionnaire which parents complete separately online.

Outcome measures were obtained at the following time points:

• T0 = initial assessment
• T1 = pre-therapy assessment
• T2 = three months after the start of therapy
• T3 = six months after the start of therapy
• T4 = twelve months after the start of therapy

All three children have pre-therapy measures (T1). Child 1 and Child 2 have T0 and T1 scores because their initial assessment (T0) took place more than 12 weeks before therapy started, and therefore the assessment measures are repeated before the start of therapy.
As such, these two children have a no-intervention baseline period.

3. Results

3.1. Stuttering frequency

The stuttering frequency of each child over time is presented in Fig. 1. Child 1 showed a small increase in stuttering frequency over the baseline phase. After therapy, there is a reduction in stuttering frequency, most evident at six months. At one year post-therapy, there is a small increase in stuttering frequency, but all three post-therapy measures are lower than the highest baseline measure. Child 2 showed minimal change in stuttering frequency over a one year period. Child 3 shows a steady reduction in stuttering frequency over time.

3.2. Children’s attitude to communication

Fig. 2 shows Child 1’s scores on the KiddyCAT over time. Higher scores on the KiddyCAT reflect a more negative attitude to speaking. The baseline trend suggests an increasingly negative communication attitude pre-therapy, with improved communication attitude from three months after therapy started. No change between T3 and T4 shows improvement was maintained at one year post-therapy.

Fig. 3 presents Child 3’s scores on the CAT and OASES over time. Higher scores on the CAT reflect a more negative attitude to speaking. Visual analysis shows Child 3 developed a more negative attitude towards speaking over time. Higher scores on the OASES reflect a higher impact of stuttering on the child. Visual analysis shows that the impact of stuttering on Child 3 increased after therapy but reduced to baseline level at six months after therapy started. At one year post-therapy, stuttering still had a moderate/severe impact on Child 3.

Child 2 completed the KiddyCAT at initial assessment (T0; score 5/12) and pre-therapy assessment (T1; score 3/12). As explained above, Child 2 turned 7 years old during the course of therapy and therefore completed the CAT at six months post-therapy (T3; score 12/33) and one year post therapy (T4; score 3/33). As these are different assessments, any interpretation would be speculative.

3.3. Parents’ perception of the impact of stuttering on the child

Factor 1 of the Palin PRS explores parents’ views of how much stuttering impacts the child. Higher scores indicate less impact. Fig. 4 presents the parent’s scores over time. The weighted standard score on the y axis is indicated on the left and the degree of impact is indicated by the shading and category labels on the right. Child 1’s mother considered the impact of stuttering on him to be moderate over the baseline period. Three months after the start of therapy, the impact reduced to low and this was maintained at six months and one year post-therapy. For Child 2’s parents, visual analysis shows both parents considered the impact of stuttering on their son reduced over time. Child 3’s father consistently reported higher scores than the mother, indicating that he considered stuttering to have less of an impact on their son, and from his perspective, the impact reduced over time. From the mother’s perspective, stuttering continued to have a high impact on their son.

Fig. 1. Frequency of syllables stuttered over time
Fig. 2. Child 1’s attitude to communication over time

Fig. 3. Child 3’s attitude to communication over time
3.4. Parents’ perception of stuttering severity and the impact that stuttering has on them

Factor 2 of the Palin PRS explores the parents’ perception of the severity of the child’s stuttering and the impact that it has on the parents in terms of worry and anxiety. Higher scores reflect a more desirable circumstance, so the higher the score, the less severe the stuttering and impact it is having on the parents. Fig. 5 shows the parents’ scores over time. The weighted standard score is indicated on the left y axis and the degree of impact is indicated by the shading and category labels on the right. For Child 1’s Mother, visual analysis shows that stuttering severity and impact on the mother improved before therapy began. Further improvement was made
immediately after therapy (T2). Change was not maintained at six months, however, at one year gains were recovered.

For Child 2’s parents, the stuttering severity and impact on them was low over the baseline phase. From the father’s perspective, the child’s stuttering severity and impact on the father reduced to very low after therapy. From the mother’s perspective, the stuttering severity and the impact that it had on her remained low at one year after therapy.

Child 3’s father consistently reported their child’s stuttering as less severe than the mother, and the impact on him was less than on the mother. From the father’s perspective, the stuttering severity and impact on him reduced over time. From the mother’s perspective, the stuttering severity and the impact that it had on her remained very high at three months after therapy started, high at six months and very high at one year.

3.5. Parents’ knowledge about stuttering and confidence in managing it

Factor 3 on the PalinPRS measures the parents’ knowledge about stuttering and their confidence in supporting their child. Higher scores reflect greater knowledge and confidence. Fig. 6 shows the parents’ scores over time. The standard score is indicated on the left and the level of knowledge and confidence is indicated by the shading and category labels on the right.

Child 1’s Mother reported low knowledge about stuttering and confidence in managing it over the baseline period. Six months after therapy, she reported a moderate level of knowledge and confidence, and one year after therapy, she reported very high knowledge and confidence.

Both Child 2’s parents’ knowledge about stuttering and confidence in managing it improved over the baseline period and increased further at six months after therapy started. Gains were maintained at one year post-therapy.

Child 3’s mother reported a high level of knowledge and confidence before therapy started. Her knowledge and confidence increased after therapy, but gains were not maintained over time. Her confidence reduced at six months post-therapy. Child 1’s father started with a very low level of knowledge and confidence, which increased over time and was maintained at one year after therapy started.

4. Discussion

This study explored change over a one year period in three CWS with ASD who received Palin PCI therapy for stuttering at a specialist centre in London. Despite a large proportion of the literature describing CWS with ASD exhibiting atypical dysfluencies, these three cases demonstrate that not all CWS with ASD have atypical dysfluencies. These three children presented with typical stuttering behaviours, including whole-word and part-word repetitions, blocks and prolongations in the word initial position. Change in the overt stuttering behaviours was measured by the clinician recording %SS. Change to the impact of stuttering on the child and parents was
considered from the perspectives of the child and the parents through self-report questionnaires.

4.1. Summary of findings

Child 1 had mild stuttering at baseline. His stuttering frequency reduced six months after therapy started. He reported a more positive communication attitude at three months after therapy and change was maintained at one year post-therapy. From his mother’s perspective, the impact of stuttering on her son reduced after therapy and change was maintained over time. From her perspective, the stuttering severity and impact on her reduced over the baseline, and three months after therapy and remained very low at one year. Her knowledge and confidence managing stuttering increased six months after therapy started and change was maintained at one year post-therapy.

Child 2 had mild stuttering at baseline and showed little change in stuttering frequency over time. Child 2’s self-report measures were not consistent across all time points and therefore could not be compared. Both parents reported that the impact of stuttering on their son reduced over time. The parents’ perception of stuttering severity and impact on them was low at baseline and remained low over the period of a year. Both parents’ knowledge about stuttering and confidence in managing it increased over time and changes were maintained at one year post-therapy.

Child 3 had moderate-severe stuttering at baseline and his stuttering frequency reduced over time. Stuttering had a high impact on Child 3 at baseline, and sadly, continued to have a high impact on him, and his mother. The father’s perspective of change was more positive. The father reported very low knowledge and confidence about managing his son’s stuttering, and this increased over time to a high level of knowledge and confidence. The mother started with a high level of knowledge about stuttering and confidence in managing it and her knowledge and confidence increased further immediately after therapy but then reduced over time.

By having a baseline for two of the children (Child 1 and Child 2), we are acknowledging that there is a role for natural change to occur. Though developmental changes may have occurred between the two baseline points, we would argue that the results still indicate that Palin PCI therapy can be useful. The child’s initial assessment begins the therapeutic process, with parents being informed about stuttering, their child’s strengths and needs and what is likely to be helpful for their child. It may also be that improved changes in scores reflect natural improvement. Either way, the introduction of therapy advances the trajectory of positive change, suggesting that the therapy itself is adding to the process. Further experimental investigation would be necessary to examine this further.

4.2. Stuttering frequency

Child 1 and Child 3 demonstrated reduced stuttering frequency over the period of a year. A reduction of 50% could be considered to be clinically significant change (Millard et al., 2018), and therefore the reduction for Child 1 is nearly clinically significant and the reduction for Child 3 is considerable. For Child 2, there was minimal change in stuttering frequency. A reduction in %SS would have been hard to detect with low baseline measures (average = 1.45%SS). These scores highlight the need to obtain measures in addition to stuttering frequency, since frequency is not necessarily linked to the degree of impact the stuttering has, as evidenced by Child 3.

With regards to latency of change, stuttering frequency reduced three months after therapy started for Child 3 and six months after therapy started for Child 1. Previous research demonstrated stuttering frequency reduces within three months of therapy (Millard et al., 2008, 2009). However, in both of these studies, speech samples were recorded at home. A more recent cohort study, using clinic-based speech samples, as this study did, found that reduced stuttering frequency was observable six months after therapy started (Millard et al., 2018). The authors suggested that it takes time for the reduction in stuttering frequency at home (where therapy takes place) to be reflected in clinic speech samples. Consistent with these findings, Child 1’s mother reported reduced stuttering severity on the Palin PRS at three months, before stuttering frequency reduced on clinic measures at six months.

There is no research exploring the impact of stuttering on children with ASD. The majority of children who will resolve the stuttering without intervention will do so within approximately 12-18 months of onset (Yairi & Ambrose, 2013). At 10–12 months post onset, Child 2 is still within this ‘recovery window’. However, there is no change in his stuttering during the baseline phase, suggesting that this process was not underway, or was underway to a limited extent, prior to the introduction of the therapy. The change in data post therapy would suggest that the introduction of therapy either enhanced or was in addition to, any natural reduction that may have been taking place.

It is important to consider that fluency on a picture description task in clinic may not be representative of the child’s fluency in spontaneous speech with familiar people in familiar environments. This may account for the differences observed between parents’ ratings of stuttering severity on the Palin PRS and stuttering frequency measured in clinic for Child 1 and Child 3. Picture description tasks, especially ‘what’s wrong?’ cards which require verbal reasoning skills, may be more linguistically and cognitively demanding, particularly for these children with ASD and this may cause increased dysfluency (Yaruss, 1999). Indeed, Shriberg et al. (2001) drew on the ‘limited capacity system’ proposed by Paul (1992) to argue that the social and pragmatic demands of conversation contributed to a resource drain, resulting in increased dysfluency for people with ASD.

4.3. Impact of stuttering on the child

There is no research exploring the impact of stuttering on children with ASD. A feature of ASD is a person’s lack of awareness and insight into their communication difficulties (Frith. and Happé, 1999). As with all of the characteristics of ASD, the degree to which this is an issue will vary. There will be children who are more aware and children who are less aware of their stuttering and communication strengths and needs. In the limited literature, there are reports that clients with ASD lack awareness of their dysfluency.
difficulties beyond the child was evident in both factors of the Palin PRS post-therapy. Six months after therapy started, both variables showed a temporary frequency. Child 3 presented with rigid thinking as a trait of his ASD, so although there was a decrease in %SS, there was little change to his communication attitude, as despite the reduction he still viewed himself as a child who stuttered.

It is also possible that some children with ASD might have heightened awareness of some aspects of stuttering (for example, tension) compared to CWS who do not have ASD. While Child 2 came into the age range eligible to complete both the CAT and the OASES, he was only able to complete the CAT. There is a clinical need for self-report outcome measures to be validated with this clinical subpopulation.

4.4. Child’s communication attitude

The CAT and KiddyCAT reveal the child’s attitude to communication, which will be affected by both ASD and stuttering, but do not measure the relative contributions of each.

The improvements demonstrated for Child 1 are clinically important given that research shows communication attitude worsens with age (Vanryckeghem & Bruttin, 1997). Child 1’s communication attitude improved three months after therapy started, before his stuttering frequency decreased. This is in line with research that shows children’s communication attitude can improve even when stuttering frequency does not significantly reduce (Millard et al., 2018). Child 3 reported an increasingly negative attitude to communication over time. His scores on the CAT are considerably higher than CWS his age (mean [SD] = 12.09 (6.4)) (Bruttin & Vanryckeghem, 2007). His scores on the OASES changed little over time. Child 3 presented with rigid thinking as a trait of his ASD, so although there was a decrease in %SS, there was little change to his communication attitude, as despite the reduction he still viewed himself as a child who stuttered.

It is important to consider that communication difficulties related to ASD may impact a child’s responses on a self-report questionnaire. Some children with ASD may have reduced awareness and insight into their difficulties and that may impact their responses. It is also possible that some children with ASD might have heightened awareness of some aspects of stuttering (for example, tension) compared to CWS who do not have ASD. While Child 2 came into the age range eligible to complete both the CAT and the OASES, he was only able to complete the CAT. There is a clinical need for self-report outcome measures to be validated with this clinical subpopulation.

4.5. Impact of stuttering on the parents

Factor 2 of the Palin PRS reflects parents’ perceptions of the stuttering severity as well as parents’ worry about stuttering. This makes it somewhat troublesome to interpret which aspect of that component is being measured or changing. The reason that they are combined is a result of the psychometric evaluation of the tool, which determined that these elements were highly correlated and measuring the same construct (Millard & Davis, 2016). Previous research shows that parental worry and anxiety is one of the first changes observed from therapy (Millard et al., 2018). For the children in this study, there was evidence that parental worry can reduce alongside therapy based on the Palin PRS Factor 2 scores. This was observed in the parents of each child.

The long-term change differed for each case. Child 2’s parents perceived and maintained changes that reflected their child’s stuttering frequency. This might suggest that their Palin PRS Factor 2 scores were more influenced by the questions relating to stuttering severity than worry about stuttering, or may suggest that their level of worry is closely tied to the child’s stuttering frequency.

Child 1’s mother perceived changes in a manner that reflected her perceptions of the impact of stuttering on her son. Improvement was evident in both factors of the Palin PRS post-therapy. Six months after therapy started, both variables showed a temporary reduction and then gains were recovered at one year post-therapy.

The children of Child 3 showed different profiles with regard to this variable, whereas the parents of Child 2 were consistent between them. Child 3’s father perceived changes in a manner that was consistent with a positive impact of therapy and change was maintained over the long-term. Child 3’s mother perceived changes consistent with her perception of the impact of stuttering on her son. This suggests that her worry is closely tied to her son’s level of worry. There is no research exploring the impact of stuttering on parents of CWS with ASD. The Palin PRS asks about stuttering specifically. It is possible that both the child and the mother’s anxiety is related to difficulties beyond the child’s fluency. Aspects of communication are interlinked; the impact of ASD and the impact of stuttering on overall communication are not independent and it is likely that one may influence the other.

4.6. Parents’ knowledge about stuttering and confidence in managing it

Previous research shows parental confidence is one of the first changes to occur with Palin PCI (Millard et al., 2018). In this study, all five parents reported increased knowledge about stuttering and confidence in managing it after therapy, in line with previous research. Change was evident at three months (Child 3’s parents) and six months (Child 1’s mother; Child 2’s parents). Four parents maintained improved knowledge and confidence at one year post-therapy. These findings are clinically important given that parental self-efficacy may be particularly affected by having a child with ASD (Karst & Van Hecke, 2012).

Child 2’s parents’ knowledge and confidence increased despite minimal change in their child’s stuttering frequency, in line with previous research (Millard et al., 2018). These parents showed increased knowledge and confidence over the baseline. This may be the result of the assessment where parents are informed about stuttering, the child’s strengths and needs and what is likely to be helpful for their child. It is also possible that this increase in knowledge and confidence could be due to other information or support that the parents have sought out after the initial assessment, for example, reading materials or online resources.

The long-term change differed for the parents of Child 3. The father’s knowledge and confidence increased steadily over time, with the biggest improvement seen immediately after therapy. The mother started with a high level of knowledge and confidence, perhaps
because of previous therapy, and though her confidence increased immediately after therapy, it reduced after therapy finished. This suggests that therapy supported her confidence and when therapy finished, her confidence reduced.

Whilst the Palin PRS is specific to stuttering, many questions may be influenced by a child’s ASD over and above the influence of stuttering, for example: ‘How confident is your child in speaking situations?’; ‘How well can your child express how he feels?’; ‘How much does your child struggle when speaking?’ One of the outcomes for this study is the need for outcome measures, including the Palin PRS, to be validated on this clinical subpopulation.

4.7. Response to intervention: theoretical underpinnings

Palin PCI is a child-centred, parent-implemented approach which involves the clinician and parents working together to identify strategies that facilitate fluency and effective communication. Given the focus is not solely on increasing fluency, but also on parents supporting their child to be a confident and competent communicator, Palin PCI may be particularly accessible to CWS with ASD.

While the effectiveness of PCI therapy has been demonstrated with both CWS and children with ASD, the specific components critical for change have not been isolated (Millard et al., 2008, 2009; Lieneman et al., 2017; Zlomke & Jeter, 2020). There are commonalities across the methods used with the different client groups which might explain why Palin PCI appears to have been helpful for the children in this study. Some of the same interaction strategies contained in Palin PCI are utilised in parent-mediated therapies for children with ASD. For example, PACT (Green et al., 2010) also endorses the interaction strategies of following the child’s lead, giving the child time, and modifying parent’s language input. The overlap would indicate that Palin PCI may have additional benefits for CWS with ASD.

4.8. Clinical implications

Given that there is a higher incidence of stuttering in the ASD population (Boulet et al., 2009), and a higher incidence of ASD in the stuttering population than would be expected if there were no association between the conditions (Briley & Ellis, 2018), clinicians must consider all aspects of speech, language and communication when assessing children. The variability in age at which children are identified and diagnosed with ASD means that children may present with stuttering and begin treatment for stuttering before they are diagnosed with ASD, as in the case reported by Siskin and Wasilus (2014). The average age for stuttering onset is approximately 36 months (Yairi & Ambrose, 2013). The median age for diagnosis of ASD in the UK is 55 months (Brett et al., 2016), however diagnosis may occur later into the school years, or indeed adulthood. This has implications for initial dysfluency assessment and management. If CWS present with undiagnosed ASD, onward referral to a paediatrician for developmental assessment should be discussed with parents. As Palin PCI has a number of elements that are similar to therapy for children with ASD, for children who present with needs associated with ASD but are not yet diagnosed, Palin PCI may support parents to increase their confidence in managing the child’s communication overall, not just in relation to stuttering.

4.9. Delivering Palin PCI for CWS with ASD

The same Palin PCI assessment approach is used for CWS with additional needs, considering the child’s speech, language and communication skills, stuttering profile, the child’s awareness of their stuttering and the impact on the child and parents (Kelman & Nicholas, 2020). Clinicians consider the same question for all clients; ‘What does this child need?’ This enables clinicians and parents to prioritise which components of the programme are likely to be most helpful for the individual child to become a confident and competent communicator (Kelman & Nicholas, 2020).

In delivering Palin PCI for CWS with ASD, clinicians may consider making reasonable adjustments to support the child’s emotional regulation and sensory needs, for example, through ensuring consistency in the clinic environment (Kelman & Nicholas, 2020). Parents may need to be supported to identify appropriate toys for special times and interaction strategies should support the child’s engagement and reciprocal communication (Kelman & Nicholas, 2020). Aspects of Palin PCI can be differentiated, e.g. turn taking can be introduced in play before applying the concept to conversation. Praise needs to be adapted to the child’s cognitive and linguistic level, incorporating visual materials if this is beneficial (Kelman & Nicholas, 2020).

4.10. Limitations and recommendations for future research

This study of retrospective clinical data suggests that further experimental designs would be of value to investigate the effectiveness of Palin PCI for CWS with ASD. This study did not have the level of experimental control to infer a causal relationship between the therapy and the positive changes for the children and their parents, but does show that outcomes for these children improved over a period of time when they received therapy.

As with all single-case reports, external validity is limited. Any number of variables described for these children, including their ASD profile, language abilities and bilingualism, could influence therapy outcomes. The purpose of case studies is not to generalise beyond the individual cases but to gather information about responses to intervention. No attempt is made to suggest that these findings are applicable to all CWS with ASD, or beyond this context. All three children were treated at a specialist centre for stuttering, where staff have specialist skills and experience managing referrals of complex cases. Therefore, outcomes may differ in other settings.

An additional limitation is that there are no fidelity data reported for the implementation of Palin PCI. While the therapy was
carried out by expert Palin PCI therapists, and the children’s clinical records indicated that Palin PCI was being delivered as expected by the programme, no treatment fidelity analysis was conducted. Within the Palin PCI programme, there is an expectation that the content of therapy will be adapted to meet the child’s individual needs. In these cases, it is anticipated that the therapy targets chosen by the family and therapist would be relevant not only for the child’s stuttering but also the child’s ASD. That said, the absence of treatment fidelity data means that we cannot make an absolute judgement about the degree to which the therapists conformed to the Palin PCI programme. Treatment fidelity will be an important aspect for a larger scale study.

Further research is required to validate the assessment measures on this clinical subpopulation and assess the range of factors that may affect change. These factors may include the child’s severity of needs related to ASD, child’s language level and families’ experiences of previous therapy. Given the commonalities between the parent-child interaction strategies endorsed in Palin PCI and preschool therapies for children with ASD, future research may explore the extent to which results extend beyond the behaviours measured, specifically, to language and ASD-focused outcome measures.

4.11. Conclusions

The findings indicate that CWS with ASD and their parents can benefit from Palin PCI. All three children showed improvement in three or more variables. Four out of five parents reported reduced impact of stuttering on the child and on themselves after therapy and change was maintained one year after therapy. All five parents reported increased knowledge of stuttering and confidence in managing therapy and change was maintained one year after therapy for four parents. The findings indicate that further investigations of Palin PCI therapy with CWS with ASD are merited, and outcome measurement tools need to be validated with this clinical subpopulation.

CRediT author statement

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References
