ADVANCING SPOKEN AND WRITTEN
LANGUAGE DEVELOPMENT IN CHILDREN
WITH CHILDHOOD APRAXIA OF SPEECH

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The material presented in this thesis is the original work of the candidate except as acknowledged in the text, and has not been previously submitted, either in part or in whole, for a degree at this or any other University.
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ABSTRACT

Children with childhood apraxia of speech (CAS) are likely to experience severe and persistent spoken and written language disorder. There is a scarcity of intervention research, however, investigating techniques to improve the speech and literacy outcomes of this population. The series of 5 experiments reported in this thesis investigated phonological awareness and early reading development in children with CAS and trialled a new intervention designed to advance the spoken and written language development of those affected.

In the first experiment (presented in Chapter 2), a comparison of 12 children with CAS, 12 children with inconsistent speech disorder (ISD), and 12 children with typical speech-language development (TD) revealed that children with CAS may be particularly susceptible to phonological awareness and reading deficits. There was no difference in the articulatory consistency and speech severity of the CAS and ISD groups, and no difference in the receptive vocabulary of the CAS, ISD, and TD groups. The children with CAS exhibited poorer phonological awareness scores than the comparison groups and had a greater percentage of participants performing below the expected range for their age on letter knowledge, real word decoding, and phonological awareness normative measures. The children with CAS and ISD performed inferiorly than the children with TD on a receptive phonological representation task. The results showed that the children with CAS had a representational component to their disorder that needed to be addressed in intervention.
In the second experiment (presented in Chapter 3), a follow-up pilot study was conducted to examine the long-term effects of a previously conducted intensive integrated phonological awareness programme (7 hours of intervention over 3 weeks) on 2 children with CAS. The children aged 7;3 and 8;3 at follow-up assessment had previously responded positively to the intervention. Results showed that the children were able to maintain their high accuracy in targeted speech repeated measures over the follow-up period. One child was also able to maintain her high accuracy in phonological awareness repeated measures. The children performed superiorly on a standardised phonological awareness measure at follow-up than at pre-intervention. Non-word reading ability showed a sharp increase during the intervention period, while minimal gains were made in this measure over the follow-up period. The findings suggested that an integrated intervention was a potential therapeutic approach for children with CAS.

In the third experiment (presented in Chapter 4), the effectiveness of an integrated phonological awareness programme was evaluated for the 12 children (identified in the first experiment) aged 4 to 7 years with CAS. A controlled multiple single-subject design with repeated measures was employed to analyse change in trained and untrained speech and phoneme segmentation targets. A comparative group design was used to evaluate the phonological awareness, reading, and spelling development of the children with CAS compared to their peers with TD over the intervention. The children participated in two 6-week intervention blocks (2-sessions per week) separated by a 6-week withdrawal block. Seven children with CAS made significant gains in their production of trained and untrained speech words with 7 of these children demonstrating transfer of skills to connected speech for at least one target. Ten children showed significant gains in
phoneme awareness, and 8 of these children demonstrated transfer of skills to novel phoneme awareness tasks. As a group, the children with CAS demonstrated accelerated development over the intervention period in letter knowledge, phonological awareness, word decoding, and spelling ability compared to their peers with typical development.

In the fourth experiment (presented in Chapter 5), the speech, phonological awareness, reading, and spelling skills of children with CAS and TD were re-evaluated 6-months following completion of the intervention programme. A measure of reading accuracy and reading comprehension in a text reading task was administered to the children with CAS. There was no difference in the performance of the children with CAS in post-intervention and follow-up assessments. The children with CAS and children with TD presented with similar relative change in phonological awareness, reading, and decoding measures over the follow-up period. The connected reading performance of children with CAS mirrored their phonological awareness and decoding skills. The findings demonstrated that children with CAS were able to maintain gains achieved during the intervention but may need further support to promote sustained development in written language.

In the fifth experiment (presented in Chapter 6), the long-term effects of the integrated phonological awareness programme for identical twin boys who participated in the research intervention at pre-school were examined. The study examined Theo and Jamie’s spoken language, phonological awareness, reading, and spelling development during their first year of schooling. The results pointed to the benefit of providing phonological awareness within a preventative framework for children with CAS. Theo and Jamie experienced continued growth in speech and phonological awareness skills.
They exhibited age-appropriate reading and spelling development during their first year of formal literacy instruction.

It was concluded from this series of experiments that children with CAS are particularly vulnerable to phonological awareness and early reading difficulty, and that an integrated phonological awareness intervention is an effective means of developing speech, phonological awareness, reading, and spelling skills in most children with CAS. The intervention appears to target processes underlying spoken and written language development in this population. The results are discussed within a phonological representation deficit hypothesis of CAS and clinical implications of the findings are highlighted.
PUBLICATIONS ARISING FROM THIS THESIS


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CHAPTER 1

LITERATURE REVIEW

Introduction

The link between spoken and written language impairment has received widespread research attention in recent years. Childhood apraxia of speech (CAS), however, is one form of speech disorder where the literature has centred almost exclusively on the spoken language aspects of this impairment. Consequently, strategies to facilitate the reading and spelling development of children with CAS are untested. A wider intervention focus on developing processes underlying successful literacy development and speech production skills may be one means of enhancing the academic, social, and vocational outcomes of those affected. This thesis examines both the spoken and early written language development of 12 children with CAS and investigates these children’s responsiveness to a new intervention specifically designed to simultaneously enhance speech, reading, and spelling development.

1.1 What is Childhood Apraxia of Speech?

CAS is “a neurological childhood (pediatric) speech sound disorder in which the precision and consistency of movements underlying speech are impaired in the absence of neuromuscular deficits (e.g., abnormal reflexes, abnormal tone)” (American Speech-Language-Hearing Association, 2007). There are widespread non-speech, speech,
suprasegmental, and written language characteristics associated with the disorder including oral apraxia, limited phonemic repertoires, inconsistent speech errors, speech sequencing difficulties, vowel errors, prosodic deficits, and phonological awareness difficulties, amongst other symptoms (B. L. Davis, Jakielski, & Marquardt, 1998; Lewis, Freebairn, Hansen, Iyengar, & Taylor, 2004). CAS is associated with persistent spoken and written language deficits that are comparatively resilient to speech and language therapy approaches (Lewis, Freebairn, Hansen, Iyengar et al., 2004; Pannbacker, 1988).

CAS was originally identified when a group of children with speech characteristics analogous to adult onset apraxia of speech (AOS) were described. Although CAS takes its name from AOS, there are important distinctions between the acquired and developmental forms of the disorder including:

(a) Children with CAS must develop language amidst a disordered system rather than suffering neurological insult after linguistic development is completed;

(b) No particular neurological deficit has been associated with CAS; and

(c) Symptoms in CAS are more widespread than AOS including written language, morphological, and syntactic deficits (Velleman & Strand, 1994).

Epidemiological data for CAS is limited due to the use of clinical rather than population samples in the calculation of figures. Prevalence data indicate that CAS is a rare disorder occurring in one to two children per thousand (Shriberg, Aram, & Kwiatkowski, 1997a). Gender ratio analysis indicates CAS is more prevalent in males than females (Hall, Jordan, & Robin, 1993; Lewis, Freebairn, Hansen, Taylor et al., 2004). Hall et al. (1993) found that 74% of 229 cases of CAS reported in the literature were males. Lewis, Freebairn, Hansen, Taylor et al. (2004) reported a 2:1 ratio of males
to females in their sample of 22 children aged 3 to 10 years. The authors attributed the lower ratio than that reported in the Hall et al. (1993) study to the rigorous protocol used to diagnosis CAS in their sample which likely included a higher proportion of children with more severe forms of the disorder.

CAS appears to be a heritable condition with a large percentage of the family members of those affected experiencing speech, language, and/or literacy disorders (Lewis, Freebairn, Hansen, Taylor et al., 2004; Thoonen, Maassen, Gabreels, Schreuder, & de Swart, 1997). Lewis, Freebairn, Hansen, Taylor et al. (2004) found that 86% of 22 participants with CAS had at least one nuclear family member with a speech and/or language disorder, while 59% of participants had at least one parent affected with a speech and/or language disorder. Thoonen et al. (1997) reported a family history of speech and/or language disorder in 6 of 11 children with CAS. There is, however, a low aggregation of CAS amongst family members of children with CAS. Lewis, Freebain, Hansen, Taylor et al. (2004) interpreted the familial aggregation of CAS with other speech-language disorders (rather than with CAS itself) as opposing a unique genetic cause of CAS. Rather, the authors posited that children with CAS hold extra risk genes for a general ‘verbal trait disorder’.

Further demonstration of the heritability of CAS has been provided by the examination of the ‘K.E. family’, a family with approximately 50% of its members exhibiting CAS (Hurst, Baraitser, Auger, Graham, & Norell, 1990). Oral apraxia, speech errors, phonological processing, non-verbal intelligence, and syntactic deficits have been reported in those affected (Watkins, Dronkers, & Vargha-Khadem, 2002). Genetic investigation of the family has identified a mutation at chromosome 7q31 (also called the
FOXP2 gene) (Lai, Fisher, Hurst, Vargha-Khadem, & Monaco, 2001). Although research on the ‘K.E. family’ has wide scientific benefit, the application of findings of this group to the general CAS population may be limited given failure to implicate FOXP2 mutations in other children with CAS (American Speech-Language-Hearing Association, 2007).

1.1.1 Symptoms of CAS

The following section describes the diverse motor, speech, and prosodic symptoms reported in children with CAS. Readers are referred to Sections 1.3 and 1.5.1 for discussion of the phonological awareness, literacy, and language deficits associated with the disorder.

Motor symptoms in CAS

Oral apraxia and limb apraxia have been reported in children with CAS. Features of oral apraxia reported in the population include impaired voluntary oro-motor control in isolated and sequenced oral movements (B. L. Davis et al., 1998; McCabe, Rosenthal, & McLeod, 1998). For example, the child with CAS may have difficulty elevating his/her tongue in response to a verbal command, but have no difficulty with spontaneous tongue movement. Oral apraxia is one feature of CAS that is not commonly reported in other developmental speech and language disorders. Earlier intervention approaches for CAS thus tended to focus on remediation of oral-motor control deficits (Pannbacker, 1988). There is now general consensus in the literature, however, that non-verbal oral motor exercises are contraindicated in treatment approaches aiming to improve speech production in children with CAS (Forrest, 2002; Lof, 2003).
A history of feeding difficulties and low muscle tone is also reported in some children with CAS (Stackhouse, 1992). Children with CAS are more likely than the general population to be diagnosed with co-morbid limb apraxia and commonly exhibit fine and/or gross motor control difficulties (e.g. Dewey, Roy, Square-Storer, & Hayden, 1988).

Speech production symptoms in CAS

Verbal apraxia is one of the most commonly cited features of CAS. Characteristics of verbal apraxia include slow and/or mis-sequenced performance in diadochokinetic tasks and articulatory groping (silent posturing for articulatory position) during speech (B. L. Davis et al., 1998; McCabe et al., 1998; Nijland et al., 2002; Thoonen et al., 1997).

There are a myriad of other speech production deficits that have been identified in children with CAS, all of which are also evidenced by children with other types of speech disorder. Speech symptoms include a limited phonemic repertoire, vowel errors, unusual or atypical speech errors, inconsistent speech errors, sound sequencing errors, particular difficulty in the production of multi-syllabic words or connected speech, and phonotactic deficits (B. L. Davis et al., 1998; McCabe et al., 1998; Shriberg et al., 1997a). The speech production skills of children with CAS are reported to improve when producing automatic (or over-learned) phrases rather than spontaneous or imitated speech (e.g., Ozanne, 1995). CAS is also generally associated with severe and persistent speech disorder that is resistant to speech and language therapy (Pannbacker, 1988). A child with CAS who exhibited mild speech difficulty has been reported in the literature, but the articulatory deficit was
persistent and required therapy until the sixth grade to work on the /t/ sound. (Hall, 1989).

**Prosodic symptoms in CAS**

The prosodic symptom most commonly reported in children with CAS is difficulty with lexical and sentential stress production (Odell & Shriberg, 2001; Shriberg et al., 1997a; Shriberg, Aram, & Kwiatkowski, 1997b, 1997c; Shriberg, Campbell et al., 2003; Shriberg, Green, Campbell, McSweeney, & Scheer, 2003; Shriberg & McSweeney, 2002). The stress production of children with CAS is routinely described as an ‘excess and equal’ stress pattern, which listeners may perceive as a monotonous or robotic sounding voice (Shriberg et al., 1997a). Difficulties with volume, rate, and resonance control have also been reported in children with CAS (McCabe et al., 1998; Ozanne, 1995).

Prosodic disturbances in CAS may be an important diagnostic indicator of the disorder. Shriberg et al. (1997a, 1997b, 1997c) reported that around 50% of three different samples of children with CAS exhibited an excess and equal stress pattern. Further, excess and equal stress was the only characteristic that consistently differentiated children with CAS from children with other types of speech disorder. An excess and equal stress pattern has also differentiated children with CAS and adults with AOS (Odell & Shriberg, 2001). Velleman and Shriberg (1999) reported that children with CAS exhibited delayed use of stress patterns. For example, the children utilised the weak syllable deletion error pattern later in development than children with speech disorder. Consistent with this research, atypical prosody (particularly stress errors) is an important
distinguishing characteristic of CAS included within all the CAS diagnostic models discussed in Section 1.2.1.

1.1.2 Theoretical perspectives

Part of the controversy surrounding CAS is due to differing perspectives on the underlying impairment in the disorder. Traditional theoretical viewpoints have been polarised into motoric versus linguistic perspectives. Current theoretical perspectives on the disorder, however, are more adequately divided into those emphasising speech motor control processes in the disorder and those emphasizing representational and speech motor control involvement (American Speech-Language-Hearing Association, 2007). Despite the proposal of multiple theoretical accounts of CAS, there is limited evidence supporting or refuting these hypotheses.

Motor speech control perspectives

The conventional perspective on the underlying cause of CAS is that its symptoms stem from a core deficit in speech motor control processes. Many definitions of CAS implicate speech motor control deficits in those affected (Shriberg et al., 1997a). Proponents of the motoric perspective do not refute the wider linguistic deficits that have been described in CAS (e.g., syntactic and morphological deficits, speech perception difficulties, language impairment, and phonological awareness deficits) but view these difficulties as co-occurring or consequent of speech motor control involvement. Robin (1992) argued that CAS is a motor control deficit for volitional speech and refuted any phonological component to the disorder. The author concluded that all speech and non-speech characteristics of CAS could be explained by disrupted motor control. Similarly,
Hall (1992) characterised the underlying impairment in CAS as a deficit in speech motor programming.

A series of studies investigating possible prosodic markers of CAS concluded that stress and timing deficits in the speech production of children with CAS are due to pre-speech motor programming difficulties (Odell & Shriberg, 2001; Shriberg, Campbell et al., 2003; Shriberg, Green et al., 2003; Shriberg & McSweeney, 2002). It is proposed that the complex movements needed to achieve linguistic stress are disrupted by speech motor control deficits (Shriberg, Campbell et al., 2003). The authors dismissed the view that the prosodic difficulties were due to cognitive-linguistic deficits in the representation of stress assignment, as misplaced stress included excess stress on a usually stressed syllable rather than excess stress on the usually unstressed syllable. That is, children realised that a particular syllable needed to be stressed, but motor control difficulties meant that excess stress was placed on that usually stressed syllable.

Finally, disrupted sensorimotor feedback systems in children with CAS have been attributed to speech motor control deficits. Odell and Shriberg (2001) posited that children with CAS may be unable to monitor motor performance during speech production and/or respond to sensorimotor feedback. The authors concluded that such deficits may render children with CAS incapable of adjusting inaccurate movement patterns during speech production.

Although it is clear that children with CAS exhibit difficulties planning and controlling speech movements, there is limited evidence that the underlying impairment in the disorder is motoric. In line with the dominant view of CAS as a motor programming disorder, treatment approaches for CAS generally use principles of motor
learning to improve speech planning and production (Pannbacker, 1988). Data supporting the effectiveness of such techniques for children with CAS is weak. Although case study and single-subject investigations show preliminary support (Pannbacker, 1988; Strand & Debertine, 2000; Strand, Stoeckel, & Baas, 2006), comprehensive evaluation of the effectiveness of motor programming approaches for children with CAS has not been conducted. Rigorous testing of motor based interventions for CAS is needed to develop theoretical understanding of the disorder and to guide clinical practice.

Anecdotal evidence suggests that motor based techniques may not ameliorate the underlying impairment in the disorder. Children with CAS are notorious for their slow progress in therapy. Campbell (1999) reported that children with CAS require 81% more therapy than children with phonological impairment to produce a functional change in speech production. Resistance to speech and language therapy intervention has also been used as a diagnostic criterion for the disorder (Pannbacker, 1988). The failure of children with CAS to respond quickly to therapy is typically used as evidence of the severity and complexity of the disorder. It is plausible, however, that conventional intervention approaches for the population are not targeting the underlying deficit in the disorder.

A motor programming perspective appears incompatible with the language symptoms in CAS and the genetic aggregation of the disorder with other developmental speech and language disorders (Lewis, Freebairn, Hansen, Iyengar et al., 2004; Lewis, Freebairn, Hansen, Taylor et al., 2004). Proponents of the motor perspective of CAS have argued that language symptoms reported in the disorder are consequences of motor deficits rather than signs of linguistic impairment (Hall, 1992). However, Lewis Freebairn, Hansen, Iyengar et al. (2004) reported expressive and receptive language
impairment in children with CAS that became more marked over time despite gains in speech production accuracy. Further, children with CAS typically have a family history of developmental speech and language disorders rather than of CAS itself, suggesting that the disorder may not have a unique underlying cause.

**Combined representational and motor speech control perspectives**

The representational and speech motor control perspective of CAS has been developed in an effort to more parsimoniously account for the speech, language, and motor symptoms in the disorder. Unlike the motor speech control perspective, these views conceptualise the more linguistic features in CAS as part of the core difficulty in the disorder.

Phonological awareness difficulties in CAS have prompted the development of the phonological representation deficit hypothesis of the disorder (Marion, Sussman, & Marquardt, 1993; Marquardt, Sussman, Snow, & Jacks, 2002). Further discussion regarding the phonological awareness deficits of children with CAS can be viewed in Section 1.5.1 below. A phonological representation is an abstract concept describing the storage of speech sound information in long term memory (Elbro, Borstrom, & Petersen, 1998). Recent research indicates access to well-specified phonological representations is important for speech accuracy and phonological awareness ability (Rvachew, 2006; Sutherland & Gillon, 2007). The phonological representation deficit hypothesis for CAS forwards that indistinct phonological representations and/or impaired access to phonological representations underlie the speech, language, and literacy difficulties associated with CAS. Specifically, children are thought to lack quality phonological representations from which to direct motor performance (Marion et al., 1993). That is, a
precise and/or consistent motor programme cannot be formed, as it is derived from an incomplete phonological template (Marquardt, Jacks, & Davis, 2004).

Similarly, Velleman and Strand (1994) highlight the causative role of representational deficits in CAS. The authors postulated that children with CAS have difficulty with the hierarchical representation of linguistic components that impairs the organisation of language sub-units (e.g., phonemes) into larger units (e.g., syllables). The hierarchical deficit is proposed to impact all levels of language organisation (i.e., morphemes into words, word stress into phrasal stress etc.). The deficit would thus have a widespread impact on the linguistic system and explain the diverse segmental and suprasegmental characteristics of CAS. For example, deficits in the hierarchical representation of language would explain the prosodic difficulties, morphological deficits, and phonotactic limitations observed in CAS.

Combined representational and motor speech control perspectives of CAS provide an attractive alternative to purely motor based accounts of the disorder, as they are able to account for the widespread symptoms in the disorder. For example, poor performance of children with CAS on phonological awareness tasks can be explained by representational deficits. The perspective also appears to fit with the familial aggregation of CAS with other developmental speech and language disorders. Phonological representation deficits are also reported in children with speech disorder and dyslexia (Carroll & Snowling, 2004; Elbro et al., 1998; Rvachew, 2006; Sutherland & Gillon, 2007).

A focus on the language and representational deficits in CAS, however, is relatively recent, and the perspective is not well scrutinised. There are no intervention studies that have trialled the effectiveness of an approach that develops representational
skills alongside building motor control for speech in children with CAS. Further, comprehensive evaluation of the phonological awareness and language skills of children with CAS has not been conducted. An evaluation of interventions aligned with a representative perspective of CAS and a more thorough examination of the non-motor symptoms of the disorder is needed to develop this theoretical viewpoint.

1.2 Differential diagnosis of CAS

The differential diagnosis of CAS from other speech and language disorders has been the centre of much controversy (e.g. B. L. Davis et al., 1998; Shriberg et al., 1997b, 1997c; Shriberg, Campbell et al., 2003; Shriberg, Green et al., 2003). Although some measures may distinguish a portion of children with CAS (e.g., Shriberg, Green et al., 2003), no single characteristic has been identified to differentiate CAS from other developmental speech and language disorders. Failure to find a unique diagnostic indicator in CAS has prompted some researchers to question CAS as an exclusive clinical entity (Guyette & Diedrich, 1981). Currently, there is consensus in the literature that CAS does exist, and that the disorder presents a ‘symptom complex’ rather than a ‘unitary disorder’ (e.g., Shriberg et al., 1997a). A ‘symptom complex’ is a disorder that is diagnosed by a pattern of symptoms indicative of a common underlying cause. Such disorders can not be diagnosed by one characteristic in isolation, meaning that different children may have different presentations of the same disorder. Contrastively, diagnosis of a ‘unitary disorder’ always relies on the presence of the same symptom/s. There is ongoing debate and investigation into the identification of the set of symptoms indicative of the underlying cause in CAS, and convergence amongst researchers on behavioural markers of the disorder has not been reached.
1.2.1 CAS diagnostic models

CAS diagnosis is typically achieved via a diagnostic checklist which lists symptoms associated with the disorder. The checklists usually include a wide range of non-speech, speech, and prosodic symptoms. Consistent with the position of CAS as a ‘symptom complex’, not all symptoms are necessary for CAS diagnosis. There are a number of limitations in the use of diagnostic checklists. Firstly, many models do not specify the number of symptoms needed to receive a CAS diagnosis making it difficult to determine when a positive diagnosis is warranted. Secondly, the listed characteristics are not weighted for importance, meaning that items less specific to CAS such as ‘decreased intelligibility’ and ‘limited phonemic repertoire’ are given equal weight to more hallmark features of CAS such as ‘vowel errors’ and ‘inconsistent speech errors.’ Finally, the psychometric properties of diagnostic checklists have been questioned (Shriberg, Campbell et al., 2003).

There is no universally accepted method of CAS diagnosis, and differential diagnosis for this disorder will likely remain tentative until a genetic marker for CAS can be identified (American Speech-Language-Hearing Association, 2007). Current CAS diagnostic models typically include a relatively small number of symptoms thought to be critical for differential diagnosis. Ozanne (1995; 2005) proposed that a child with CAS should exhibit deficits in three levels of the speech processing chain (i.e., phonological planning, phonetic programme assembly, and motor programming) to be diagnosed with CAS. That is, deficit in one or two levels of the model is insufficient to diagnose the disorder. Table 1 links each level of impairment in Ozanne’s (1995; 2005) model to its
associated characteristics, so that the areas of assessment needed for diagnosis can be identified.

Shriberg, Campbell *et al.* (2003) identified five segmental and three suprasegmental symptoms important for differential diagnosis of CAS. Segmental symptoms included articulatory struggle or groping during speech, metathetic speech errors, inconsistent speech errors on repeated productions of the same word, increased speech omission errors compared to the overall severity of the speech disorder, and vowel errors. Surpasegmental symptoms included inconsistent stress production, inconsistent timing, and inconsistent resonance.

Davis *et al.* (1998) reported eleven characteristics that were used to differentially diagnose CAS from other speech sound disorders. However, the overlap between many of their differential characteristics and common symptoms of severe speech disorder prompted the researchers to give particular weight to four of their identified symptoms (i.e., restricted phonemic inventory, vowel errors, inconsistent speech errors, and phonotactic deficits) for differential diagnosis of CAS from other speech sound disorders.
Table 1. Levels of impairment and their associated characteristics in the Ozanne diagnostic model

<table>
<thead>
<tr>
<th>Level</th>
<th>Characteristics</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Phonological</strong></td>
<td>Vowel errors, greater errors on multisyllabic words and during connected speech, poor phonotactics, inconsistent speech errors, sound sequencing errors, prosodic deficits</td>
</tr>
<tr>
<td><strong>planning</strong></td>
<td></td>
</tr>
<tr>
<td><strong>Phonetic</strong></td>
<td>Articulatory groping during speech production, high rates of consonant deletion, voluntary speech performance poorer than involuntary speech performance</td>
</tr>
<tr>
<td><strong>programming</strong></td>
<td></td>
</tr>
<tr>
<td><strong>Motor</strong></td>
<td>Slow diadochokinetic rates, sequencing errors during diadochokinetic tasks, voicing errors, resonance inconsistencies</td>
</tr>
</tbody>
</table>

Descriptive analysis of the above diagnostic models (shown in Table 2) revealed a strong correspondence between the three methods. Although the symptoms vary slightly, each model includes deficits within the levels critical for diagnosis identified by Ozanne (1995, 2005) (i.e., the phonological planning, phonetic programming, and motor programming levels.). Guidance in the number of symptoms needed for diagnosis in these contemporary models, however, is still lacking. It is important that the use of these diagnostic models to differentiate CAS from other developmental speech and language disorders is replicated so that the psycholinguistic profiles of these disorders can be compared.
Table 2. Comparison of three diagnostic models for CAS

<table>
<thead>
<tr>
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</thead>
<tbody>
<tr>
<td>Errors on more complex structures</td>
<td>+</td>
<td>-</td>
<td>+</td>
</tr>
<tr>
<td>Vowel errors</td>
<td>+</td>
<td>+</td>
<td>+</td>
</tr>
<tr>
<td>Poor phonotactics</td>
<td>+</td>
<td>+</td>
<td>+</td>
</tr>
<tr>
<td>Inconsistent speech errors</td>
<td>+</td>
<td>+</td>
<td>+</td>
</tr>
<tr>
<td>Speech sequencing errors</td>
<td>+</td>
<td>+</td>
<td>-</td>
</tr>
<tr>
<td>Prosodic disturbances</td>
<td>+</td>
<td>+</td>
<td>+</td>
</tr>
<tr>
<td>Groping during speech production</td>
<td>+</td>
<td>+</td>
<td>-</td>
</tr>
<tr>
<td>Frequent omission errors</td>
<td>+</td>
<td>+</td>
<td>+</td>
</tr>
<tr>
<td>Difficult in imitation of speech</td>
<td>+</td>
<td>-</td>
<td>+</td>
</tr>
<tr>
<td>DDK deficits</td>
<td>+</td>
<td>-</td>
<td>+</td>
</tr>
<tr>
<td>Voicing errors</td>
<td>+</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Timing inconsistencies</td>
<td>-</td>
<td>+</td>
<td>-</td>
</tr>
<tr>
<td>Resonance inconsistencies</td>
<td>+</td>
<td>+</td>
<td>-</td>
</tr>
<tr>
<td>Expressive language &lt; receptive</td>
<td>+</td>
<td>-</td>
<td>+</td>
</tr>
</tbody>
</table>

*Note.* CAS = childhood apraxia of speech, + characteristic is included in the diagnostic model, - = characteristic is not included in the diagnostic model, DDK = diadochokinesis.

1.2.2 Importance of a developmental perspective in diagnosis

Diagnosis of CAS is further complicated by the changing set of symptoms associated with the disorder over time. Lewis, Freebain, Hansen, Iyengar *et al.* (2004) compared the linguistic profiles of children with CAS, children with an isolated speech
disorder, and children with a combined speech-language impairment at age 4 to 6 years and again at age 8 to 10 years. The researchers reported that the characteristics of CAS changed over time with speech sound production errors dominating the linguistic profile at preschool age, while language impairment, literacy deficits, and prosodic difficulties dominated at school age. Further, the children with CAS had poorer speech and language outcomes than both comparison groups at school-age. The changing characteristics of CAS over time require a developmental perspective to be included in the assessment and intervention of this population (Stackhouse & Snowling, 1992b).

Lewis, Freebain, Hansen, Iyengar et al.’s (2004) findings highlighted the difficulties in diagnosing young children with CAS. Normative speech and language testing could not differentiate the linguistic profiles of the children with CAS and children with combined speech-language impairment at preschool age. The researchers concluded that many of the characteristics considered hallmark symptoms of CAS (e.g., prosodic disturbances, sequencing errors in multisyllabic words) are unable to be assessed in children with very limited verbal output. For example, it is difficult to detect sound sequencing errors when a child’s speech production is limited to consonant-vowel (CV) productions. Similarly, it is difficult to detect prosodic disturbances in a child limited to CV productions. Diagnosis of young children or children with severely limited production skills should thus be applied cautiously (B. L. Davis & Velleman, 2000; Lewis, Freebairn, Hansen, Iyengar et al., 2004).

1.2.3 Differential diagnosis in clinical practice

Examination of the characteristics used by speech-language therapists (SLTs) to diagnose CAS has revealed that there is little consistency across clinicians in the method
employed. Forrest (2003) surveyed the diagnostic criteria used by 75 SLTs to identify CAS. The therapists were asked to name three essential symptoms for CAS diagnosis. The responses identified 50 essential characteristics of CAS across the 75 SLTs. The six most commonly cited symptoms (making up just over half of responses) were inconsistent speech errors, oral-motor impairment, articulatory groping, particular difficulty with voluntary speech, increased speech difficulty in more complex linguistic environments, and sound sequencing errors.

Davis et al. (1998) described their proposed 11 symptoms of CAS to SLTs in an effort to increase the appropriateness of referrals for their investigation. Twenty-two children were referred for participation in the study, but independent evaluation by the researchers revealed only four of these children matched their diagnostic criteria. The inconsistent methods used to identify CAS across clinicians is unsurprising given the lack of consensus amongst researchers regarding diagnostic criteria, the multiple symptoms associated with the disorder, and the changing face of CAS over time.

1.2.4 Methodological considerations

The lack of a universal method of CAS diagnosis described above has limited our understanding of the disorder. Just as there is inconsistency amongst SLTs in diagnostic methods (B. L. Davis et al., 1998; Forrest, 2003), a wide range of inclusionary criteria for the CAS group have been utilised across studies. Some investigations have relied on clinical referral alone, while others have used varying sets of symptoms. Consequently, there is uncertainty that participants with CAS across studies represent the same clinical group. It is critical that future investigations of CAS describe inclusionary criteria adequately to enable replication of findings. The employment of consistent inclusionary
criteria across studies will also allow a more valid comparison of results across different investigations.

1.3 Relationship between CAS and reading and spelling disorders

Recent research has elucidated that children with CAS are likely to experience written language disorder in addition to their significant spoken language difficulties. Stackhouse and Snowling (1992a) evaluated the literacy skills of two children with CAS in a longitudinal evaluation. The children were aged 10;7 (years;months) and 11;0 at initial assessment and 14;5 and 15;0 at follow-up assessment. Despite speech and language therapy support and participation in a phonics teaching programme throughout the study, the children with CAS presented with ongoing severe reading and spelling deficits. Reading age equivalence scores ranged between 7 to 8 years at initial and follow-up assessments, while spelling age equivalence scores ranged between 6 to 8 years at initial and follow-up assessments. The children also experienced continued difficulty grasping letter-sound correspondences.

The reading strategies employed by the children with CAS were evaluated in a decoding task consisting of phonetically regular words (e.g., lime), irregular words (e.g., flood), and non-words (e.g., plood) and compared to the performance of reading age matched controls with typical development. The children with CAS did not show a ‘regularity effect’ in their responses (i.e., the children were not more accurate at reading the phonetically consistent real words compared to the other words). The lack of a regularity effect in a child’s reading responses is typically interpreted as a difficulty in using phonological strategies in decoding (i.e., ‘sounding out’). The children with CAS also showed severe deficits in non-word reading, further indicating a difficulty in
employing a phonological or ‘sounding out’ strategy in reading. In contrast, the comparison group showed a regularity effect (i.e., they were comparatively more successful at decoding phonetically regular words), indicating the use of a phonological reading strategy. A comparison of the reading errors between the groups revealed that the children with CAS were more likely to use ‘lexicalisation’ errors (i.e., a real word error, such as reading ‘organ’ as ‘orange’). The researchers concluded that children with CAS use lexical strategies rather than utilising letter-sound correspondences in the reading process.

The spelling strategies employed by the children with CAS were evaluated in an informal spelling task consisting of common one, two, and three syllable words. Descriptive analysis of the children’s errors revealed a predominance of non-phonetic spelling errors in comparison to the reading age matched controls. This result suggests difficulty in using phonological information in the spelling process. The researchers concluded that children with CAS exhibited severe and persistent written language difficulties, and that the disordered phonological systems of the children were inadequate to support reading and spelling acquisition. Further, the children continued to experience severe reading and spelling difficulty despite gains in speech production skills over the study. The findings of the study are limited, however, by the use of a case study design and the lack of a comparison group with speech disorder.

Lewis, Freebairn, Hansen, Iyengar et al. (2004) extended Stackhouse and Snowling’s (1992a) findings by comparing the written language development of a larger number of children with CAS to children with other types of speech-language impairment. The researchers examined decoding, spelling, and reading comprehension in
their longitudinal investigation of children with CAS ($n = 10$), children with an isolated speech disorder ($n = 15$), and children with a combined speech-language impairment ($n = 14$). At school-age the children with CAS exhibited non-word and real word decoding, spelling, and reading comprehension deficits on normative measures. The children with CAS performed more poorly on all reading and spelling measures than the children with isolated speech disorder. Further, the children with CAS presented with a more severe spelling deficit than children with combined speech-language impairment. Consistent with Stackhouse and Snowling’s (1992a) findings, the children with CAS exhibited written language deficits at school-age despite large gains in speech production skills over the study.

The Lewis, Freebaim, Hansen, Iynegar et al. (2004) findings suggested that children with CAS may experience more severe written language outcomes that children with other types of speech and/or language disorders. However, the children with CAS exhibited lower percent consonant correct (PCC) scores and receptive language ability than the two comparison groups. It is therefore unclear whether the inferior reading and spelling performance of the CAS group was due to the nature and/or severity of the speech disorder or the inferior receptive and expressive language ability of this group. It is important that the reading and spelling skills of children with CAS are compared to children with speech and/or language disorders with similar language skills and speech severity, so the relationship between CAS and literacy deficits can be isolated.

Although comprehensive evaluation of the written language outcomes of children with CAS has not been conducted, the poor reading and spelling outcomes reported in the above investigations are unacceptable given the escalating negative consequences of
reading and spelling failure (e.g., Stanovich, 1986). The primary focus of the CAS literature has centred on diagnostic issues and speech production deficits in the disorder with limited attention given to the written language outcomes of the group. Exclusive attention on speech characteristics in CAS (without looking at spoken language) is incompatible with the close relationship observed between speech-language disorder and literacy impairment (Catts, Fey, Zhang, & Tomblin, 2001; Johnson et al., 1999; Larrivee & Catts, 1999; Lewis & Freebairn, 1992). Children with a history of speech and language impairment are at high risk for reading and spelling difficulty (Catts et al., 2001). Consistent with this strong association, the scope of practice of the American Speech Hearing Association (2001) and Royal College of Speech and Language Therapists (2005) have mandated that SLTs have the responsibility to facilitate both written and spoken language development in their clinical work. Consequently, it is critical that strategies to improve the reading and spelling outcomes for children with CAS are investigated.

1.4 Models of word recognition, spelling and reading development

It is useful to examine models of reading and spelling development, so that the possible processes underlying literacy deficits in CAS can be understood. The following sections describe two models of word recognition, the dual route and connectionist models, and stage models of reading and spelling development. An overview of skills required for comprehending connected text is also provided. Finally, the influence of phonological awareness on reading and spelling development is discussed within the framework of the presented models.
1.4.1 Dual-route models of word recognition

A dual route model of single word recognition was first proposed by Coltheart (1978). Within the model the meaning of a written word can be accessed via one of two independent routes: the phonological or visual route. The phonological route requires the use of a series of mental operations to access the meaning of the written word. The first step (called ‘graphemic parsing’) involves mentally dividing the letters in the word into those letter or letters that represent a single phoneme. For example, the digraph ‘ch’ would be divided from the rest of the word ‘chin’, as it represents a single phoneme. The second step requires that the graphemes identified in the ‘graphemic parsing’ process (e.g., ‘ch’, ‘i’, and ‘n’ in the example above) are translated into their corresponding phoneme (e.g., ‘ch’ is translated to its corresponding sound /ʧ/). Finally, the phonemes identified within the word must be joined together into a phonological representation which can be used to access the semantic representation (or meaning) of the word.

As the phonological route relies solely on letter-sound correspondences and phonological information, the model considers this route incapable of decoding phonetically irregular words. For example, if the word ‘sign’ was read using the phonological route, the word would be read aloud incorrectly as /ʃɪŋ/. The visual route is thus considered necessary when faced with the task of reading an irregular word. The visual route uses the shape and orthographic information in the written word to access an orthographic representation (i.e., representation of what the written word looks like) of the item in memory. The orthographic representation is then used to access the semantic representation (or meaning) of the word.
Readers are thought to switch between each route of word recognition in response to the nature of the reading task. The phonological route is thought to be reserved for reading novel words or words encountered infrequently by the reader. The visual route is reserved for decoding familiar words (i.e., items for which the reader has developed and stored a strong orthographic representation) and irregular words. The dual route model thus places limited importance on a phonologically based reading strategy in word recognition. Within the model, successful reading is possible by visual and orthographic skills alone (i.e., via the independent use of the visual route).

The limited value given to phonological skills within the dual route model has been criticised. Ehri (1992) argued that most irregular words are only partially irregular. For example, in the word ‘sign’, the only irregular component is the letter ‘g’. Ehri (1992) thus proposed a ‘modified dual route’ model that incorporates a phonological route and a visual-phonological route (rather than a visual route). The visual-phonological route enables the reader to use the consistent letter-sound correspondences within a word to help access the orthographic representation of the item. That is, some degree of phonological information is used in the reading of regular, irregular, familiar, and unfamiliar words. The modified model decreases memory demands needed in the visual route, as phonological information is used to cue the retrieval of the orthographic representation of the written word.

1.4.2 Connectionist models of word recognition

Consistent with the modified dual route model, connectionist models of word recognition also highlight the critical component that phonological reading strategies play in word recognition. Connectionist models are developed from the simulation of the
reading acquisition process in computer programmes (Seidenberg & McClelland, 1989). Unlike dual route models, connectionist models posit that readers use phonological, orthographic, and semantic knowledge in an integrated manner during all reading tasks. That is, readers use any cues available to them to assist in decoding the written word rather than choosing one of two routes to access the meaning of the written word.

A connectionist model suggests that the connection between the written word and its spoken form is learned gradually during reading acquisition. For example, a beginning reader attempting to read the word ‘foot’ may stimulate all phonological representations that begin with a /f/ sound in an attempt to read the written word. As the child’s letter-sound knowledge improves and he/she can use more phonological information during reading, the word ‘foot’ will stimulate only those phonological representations that have a similar orthographic representation to the target (e.g., fat, fit). With repeated exposure to the written word the connections between the orthographic, phonological, and semantic representations of the item will be reinforced, and the correct phonological representation (and thus meaning of the word) is likely to be accessed quickly in reading.

Connectionist models have also provided insight into the disruption caused to word recognition by phonological processing deficits. Harm and Seidenberg (1999) impaired a phonological unit in their connectionist model which disrupted the computer’s ability to use phonological decoding strategies and read untrained words. A severe impairment placed on the phonological unit within the model also disrupted the computer’s ability to read both irregular and regular words, suggesting that phonological processing deficits will have widespread impact on the reading ability of those affected.
Although the simulation of the processes of reading development and reading disorder achieved within connectionist models is attractive, they have encountered some criticism (Coltheart, 2006; Coltheart, Curtis, Atkins, & Haller, 1993). Coltheart (2006) argued that some aspects of reading behaviour could not be explained by parallel processing (e.g., processing all the letters in a word simultaneously in a decoding attempt) that is implicated in connectionist models and necessitate serial processing (e.g., processing one letter at a time moving from left to right) as included in dual route models; it was further argued that connectionist models do not adequately simulate reading acquisition in children, as the computers require greater exposure to written-spoken pairs than experienced by children in early reading encounters.

1.4.3 Stage models of word recognition and spelling development

Stage models depict the different strategies used in reading and spelling attempts in the progression to decoding and encoding mastery. Although the stages are presented as mutually exclusive within these models, it is more likely that the one child uses a variety of decoding and encoding strategies at a given time depending on their familiarity with a given word (Treiman & Bourassa, 2000). Each stage more likely represents the strategy most commonly employed by the child in the reading and spelling process (Treiman & Bourassa, 2000).

Stage model of word recognition development

Stage models illustrating word recognition development generally include logographic, phonetic, and orthographic phases (Frith, 1985). During the logographic phase, word recognition is achieved by using prominent visual cues in the written word.
and/or the environment in which the word is written to attain its meaning. The child is unable to use the systematic relations between letters and sounds to assist word recognition during the logographic stage.

During the alphabetic stage, the reader begins to use grapheme-phoneme correspondences in reading attempts. In the initial phases of the alphabetic stage, the child is able to utilize simple letter-sound correspondences (e.g. t, s etc.) and can decode one or two letters in the written word. For example, early in the alphabetic stage a child may read the word ‘said’ as ‘sad’. The child’s ability to decode more complex grapheme-phoneme relationships gradually improves over the alphabetic period.

During the orthographic stage, children learn to read bound morphemes (e.g. ed, ing, tion) as whole units rather than converting each letter within the morpheme into its constituent phoneme one at a time. The recognition of a morpheme as a whole unit allows greater fluency in reading. Entry into the orthographic stage is dependent on repeated exposure to frequently used letter patterns.

Stage model of spelling development

Spelling, like reading, is also largely reliant on the development of phonological strategies rather than visual memory skills (Ehri, 2000). The stages depicting spelling development are thus closely aligned with those included in stage models of word recognition presented above.

Ehri (2000) proposed four stages of spelling development; pre-communicative, semi-phonetic, phonetic, and the consolidated alphabetic stage. During the pre-communicative stage, writing attempts include the random use of letters and letter like
symbols. There is no correspondence between the letters written down and the phonology of the word.

The semi-phonetic stage marks the beginning of using the systematic connection between letters and sounds in the spelling attempt. Initially the child will represent the prominent sounds within the word. For example, ‘kangaroo’ may be written as ‘kroo’.

During the phonetic stage, the child is able to represent all sounds within the word with a corresponding letter. However, the child may have difficulty using more complex orthographic rules. For example, ‘have’ may be written as ‘hav’.

During the consolidated alphabetic stage, the child is able to use the more complex orthographic connections and apply morphologic knowledge to the spelling process. For example, the child is able to use the letter combination ‘ed’ to mark the past tense bound morpheme in the word ‘jumped’, even though /t/ is the final sound in the word.

1.4.4 Reading connected text

It is important to examine the skills involved in reading connected text, as the desired outcome of skilled reading is much more than decoding one word at a time. In the “simple view of reading” (Hoover & Gough, 1990), connected reading is dependent upon word recognition and listening comprehension. Word recognition must be accurate and fluent to enable reading comprehension (Perfetti, 1985). Laborious decoding will obstruct an individual’s ability to focus on the meaning of the word, sentence, or story. Listening comprehension difficulties such as poor vocabulary, morpho-syntactic deficits, difficulty making inferences, poor figurative language, and weaknesses in real and
written narratives will also disrupt connected text reading (Cain, 2003; Cain, Oakhill, Barnes, & Bryant, 2001; Cain, Oakhill, & Elbro, 2003; Nation, Clarke, Marshall, & Durand, 2004; Nation & Snowling, 1998; Oakhill, 1984).

1.4.5 Phonological awareness development

The preceding models have identified the central importance of using a phonological or alphabetic strategy in the reading and spelling process. The use of an alphabetic strategy is largely dependent on phonological awareness skills (in combination with letter-sound translation skills). Phonological awareness is the ability to reflect on and manipulate the sound structure of language (Gillon, 2004) and is one of the strongest predictors of early reading and spelling ability (Bradley & Bryant, 1983; Torgesen, Wagner, & Rashotte, 1994). The phonological awareness construct is generally divided into three levels of awareness: syllable, onset-rime, and phoneme. Syllable awareness can be measured by syllable segmentation tasks (e.g., show me the syllables in hospital). Onset-rime can be measured by rhyme generation tasks (e.g., name all the words you can think of that rhyme with cat) or rhyme identification tasks (e.g., which word is the odd one out – cat, mat, sun, bat?). Phoneme awareness can be measured by a range of activities including phoneme identity tasks (e.g., what’s the first/last/middle sound in cat?, which word is the odd one out – cat, bag, sit, light), phoneme segmentation and blending tasks (e.g., show me the sounds in cat, join these sounds together: /k/ /æ/ /t/), and phoneme manipulation tasks (e.g., what’s cat without the /k/ sound?).

Phonological awareness development begins in the preschool years and progresses sequentially from the awareness of bigger sound units within the word to the awareness of smaller sound units within a word (i.e., from syllable to onset-rime to
phoneme awareness) (Lonigan, Burgess, Anthony, & Barker, 1998). Most 4 year old children have acquired syllable awareness and some aspects of onset-rime awareness (Lonigan et al., 1998). Phoneme awareness, however, is mastered over a more protracted period. The ability to perform less complex phoneme awareness tasks (e.g., initial phoneme identity skills) emerges from around 4 to 5 years of age, but acquisition of more complex phoneme awareness abilities (e.g., phoneme segmenting, blending and manipulation) are gradually mastered in the early school years (Dodd & Gillon, 2001; Hesketh, Dima, & Nelson, 2007).

Phoneme awareness is generally considered more important for reading and spelling development than others levels of phonological awareness (Hulme et al., 2002; Muter, Hulme, Snowling, & Taylor, 1997; Muter, Hulme, Snowling, & Stevenson, 2004; Muter & Snowling, 1998). Muter et al. (2004) examined the relationship between rhyme awareness, phoneme awareness, letter knowledge, grammatical skills, vocabulary, and reading skills over the first two years of schooling in 90 children aged 4;2 to 5;2 (years;months). The authors concluded that phoneme awareness and letter knowledge were powerful predictors of word recognition ability, but that rhyme awareness was a comparatively weak predictor of word recognition ability. Similar findings have been reported in other studies investigating the relative importance of phoneme and rhyme awareness for literacy development (Hulme et al., 2002; Muter et al., 1997; Muter & Snowling, 1998).

Phonological awareness intervention studies focusing at the rhyme level also provide evidence against a powerful contribution from rhyme awareness to reading and spelling development. Nancollis, Lawrie, and Dodd (2005) evaluated the effects of a
phonological awareness intervention that targeted syllable and rhyme awareness on the phonological awareness, reading, and spelling development of 99 children from low socioeconomic backgrounds. A group of 114 children who did not receive phonological awareness intervention \((n = 114)\) were employed as a retrospective control group. The effectiveness of the intervention was minimal with treatment effects evident for rhyme awareness and non-word spelling ability alone. In fact, the control group presented with superior phoneme segmentation ability than the intervention group. Hatcher, Hulme, and Snowling (2004) examined the effectiveness of a phonics based intervention approach that was coupled with either phoneme awareness or rhyme awareness training in improving the reading outcomes of children at risk for reading delay. The researchers reported that children involved in the intervention that incorporated phoneme awareness (rather than rhyme awareness) training had superior reading outcomes. Rhyme awareness appears to be associated with some aspects of early reading and spelling performance (probably via its stimulation of early phoneme awareness development), but this association weakens in the early school years (Duncan & Johnston, 1999; Muter et al., 1997).

Phoneme awareness development is also facilitated by the reciprocal relationship between phoneme awareness and literacy acquisition (Burgess & Lonigan, 1998; Hogan, Catts, & Little, 2005; Perfetti, Beck, Ball, & Hughes, 1987). That is, just as phoneme awareness influences reading and spelling, participation in literacy instruction influences the development of more sophisticated phoneme awareness skills. Hogan et al. (2005) examined the phonological awareness, non-word, and real word decoding development of 570 children from kindergarten to fourth grade. The researchers concluded that
kindergarten phonological awareness performance predicted reading performance in second grade, whereas second grade reading ability predicted phonological awareness performance in fourth grade. The results thus indicate a bidirectional pathway between phoneme awareness and written language development.

Some authors, however, have questioned the reciprocal relationship between phoneme awareness and reading development (Castles & Coltheart, 2004; Morais, Cary, Alegria, & Bertelson, 1979). Castles and Coltheart (2004), in a review of the causal connection between phoneme awareness and reading, reported that phoneme awareness skills were only exhibited by children who also had acquired some letter-sound knowledge. The authors also cited evidence that beginning readers use orthographic strategies (i.e., thinking about how a word is spelt) to complete phoneme awareness tasks (Tunmer & Nesdale, 1985). The authors thus concluded that phoneme awareness is not a separable skill from letter-sound knowledge, and that the construct can only develop as a result of exposure to letter-sound relationships.

Recent research has provided convincing evidence that phoneme awareness is an independent contributor to reading development. There is data demonstrating that young children can perform phoneme awareness tasks prior to demonstrating letter-sound knowledge. Muter et al. (2004) identified eight children aged 4;2 to 5;2 in their sample of without any letter-sound knowledge. Four of these children showed some level of phoneme identity skill. Hulme, Caravolas, Malkova, and Brigstocke (2005) reported that young children can identify a phoneme within a word when they are unaware of that phoneme’s corresponding grapheme. Overall the data support an interdependent relationship between phoneme awareness and reading ability.
Phonological awareness and phonological representations

A further contributor to phonological awareness development is the underlying phonological representations of vocabulary items. A phonological representation is an abstract cognitive construct that is thought to hold phonological information about words in long-term memory (Elbro et al., 1998). As highlighted in the section on theoretical perspectives of CAS, phonological representations are also thought to influence speech production ability (Stackhouse & Wells, 1997). Phonological representations become more segmental over time as a response to vocabulary growth (Fowler, 1991; Metsala & Walley, 1998). Words are initially stored as whole units with limited phonological detail and become increasingly more segmental with development, so that words with similar phonological structures within the lexicon can be differentiated. For example, a segmental (or detailed) representation includes all phonemes within the word, depicts each phoneme as an individual unit, and places the phonemes in the correct order. The progression from holistic to segmental phonological representations mirrors the progression from awareness of bigger (e.g. syllable) to smaller (e.g. phoneme) units within spoken words in phonological awareness development. It is hypothesised that the ability to reflect on a word’s sound structure is dependent on access to a segmental phonological representation.

Investigations examining the phonological representation skills of children with reading disorder support the importance of access to quality phonological representations for phonological awareness development (Carroll & Snowling, 2004; Elbro et al., 1998; Swan & Goswami, 1997). Swan and Goswami (1997) investigated the phonological representation and phonological awareness skills of children with dyslexia, ‘garden
variety' reading disorder (i.e., poor readers with intelligence below the expected range), reading-age matched controls, and children with typical development. The authors found that children with reading disorder performed more poorly on phonological representation tasks. Elbro et al. (1998) examined the relationship between phonological representation and later phonological awareness in 49 Danish children at genetic risk of reading disorder and 42 Danish children with typical speech and language development. The children were assessed at 6 years of age (one year before formal literacy instructions begins in Denmark) and again at 8 years of age. The authors reported that the children’s performance on the phonological representation task at the initial assessment predicted their phonological awareness performance at age 8 years. Similarly, Carroll and Snowling (2004) reported that children at genetic risk for of reading disorder performed poorly in comparison to children with typical development on phonological representation measures.

1.4.6 The importance of phonological awareness in models of word recognition and spelling

It is useful to turn back to the word recognition and spelling models presented in the current section to exemplify the critical role that phonological awareness plays in literacy development. Any element of the reading and spelling models that utilise a phonological (or alphabetic) strategy is grounded in phonological awareness. The ability to map phonemes onto graphemes (or vice versa) is dependent on phoneme awareness. That is, using letter-sound knowledge in the reading and spelling process requires that a word can be analysed at the phoneme level first. If a child can not isolate a phoneme
within a word, then he/she will find it impossible to link that phoneme with its corresponding grapheme.

The importance of a phonological strategy for decoding and encoding is emphasised in all of the reviewed models. The modified dual route and connectionist models necessitate a phonological decoding strategy within all reading contexts. Phonological awareness thus plays a critical role in reading regular, irregular, novel, and familiar words. Similarly, the development of phonological reading and spelling strategies is needed to move beyond the logographic and pre-communicative phases in stage models of reading and spelling. Finally, as a significant proportion of reading comprehension is largely dependent on fluent and accurate decoding (Hoover & Gough, 1990; Perfetti, 1985), a child with phonological awareness difficulties is at risk for reading difficulties at a text level.

1.5 Why are children with CAS at risk of reading and spelling disorder?

The models of reading and spelling development reviewed above have highlighted the foundational role that oral language skills play in written language development. There is now a large body of research indicating that children with a history of speech and/or language disorder are at heightened risk of literacy impairment (Bird, Bishop, & Freeman, 1995; Bishop & Adams, 1990; Catts, 1993; Nathan, Stackhouse, Goulandris, & Snowling, 2004; Raitano, Pennington, Tunick, Boada, & Shriberg, 2004; Rvachew, Ohberg, Grawburg, & Heyding, 2003; Snowling, Bishop, & Stothard, 2000).

Although children with speech disorder (or a history of speech disorder) are, as a group, likely to experience written language difficulty, some children within the population demonstrate typical reading and spelling development. Investigations have
thus focused on delineating which children with speech disorder (or a history of speech disorder) are likely to go on to experience reading and spelling difficulties. Bishop and Adams (1990) put forward the ‘critical age hypothesis’ stating that children who continue to exhibit speech disorder at the onset of formal literacy instruction are likely to experience reading disorder. Children whose speech disorder had resolved at the onset of formal literacy instruction, however, were considered at minimal risk of reading disorder.

Nathan et al. (2004) tested the ‘critical age hypothesis’ in a longitudinal investigation of the relative risk of written language disorder in children with an isolated speech disorder ($n = 19$), children with a combined speech-language impairment ($n = 19$), and children with typical development ($n = 19$). The groups were matched for age, socioeconomic status, and non-verbal intelligence and had an average age of 4;7 at initial assessment and 6;9 at final assessment. Children with combined speech-language impairment generally had poorer phonological awareness, reading, and spelling outcomes than the other groups. Children exhibiting persistent speech disorder (i.e. continued to present with speech disorder at final assessment) also presented with high rates of phonological awareness, reading, and spelling deficits. Phoneme awareness ability was a unique predictor of reading and spelling performance. The authors presented a ‘modified critical age hypothesis’ to explain their findings, which states that children with speech disorder at preschool will be at low risk for later reading and spelling disorder if their speech disorder is resolved at the onset of formal literacy instruction and they exhibit typical phonological awareness skills.
Children with CAS present with a number of risk factors for reading and spelling difficulty included within the modified critical age hypothesis. The following section reviews the risk factors children with CAS exhibit for written language disorder.

1.5.1 Phonological awareness deficits in children with CAS

The reading and spelling difficulties experienced by children with CAS appear to be largely attributed to phonological awareness deficits. Phonological awareness difficulties at the phoneme, rhyme, and syllable levels are reported in children with CAS (Marion et al., 1993; Marquardt et al., 2002; Stackhouse & Snowling, 1992a). Stackhouse and Snowling (1992a) reported severe rhyme and phoneme awareness deficits in their longitudinal investigation of two children with CAS. Marion et al. (1993) examined the rhyme awareness of four children with CAS and four children with no history of speech or language disorder aged 5 to 7 years. Rhyme awareness was assessed via a rhyme generation task (i.e., the child was asked to name as many words as possible that rhymed with a target word within 30 seconds) and rhyme identification tasks. The rhyme identification tasks included a ‘rhyming pairs’ task, where the children were required to choose one of two words that best rhymed with a target word, and a ‘serial rhyme pairs’ task, where the children were required to choose which words (out of a total of ten items) rhymed with a target word.

The children with CAS presented with a severe deficit in rhyme awareness compared to their peers with typical development. In the rhyme generation task, the children with CAS were unable to name any real words that rhymed with a target word in comparison to an average of seven rhyming words produced by the children with typical development. The speech production difficulties of the CAS group, however, likely
inhibited their performance on this expressive phonological awareness task. Rhyming
deficits were also evident in the receptive based tasks with children with CAS scoring an
average of 45% and 55% correct versus 95% and 97% correct achieved by the children
with typical development in the ‘rhyming pairs’ and ‘serial rhyme pairs’ task
respectively. However, the groups were not matched for language ability, making it
difficult to determine if the poor rhyme awareness of the CAS group was representative
of lower language skills or their CAS diagnosis.

Marquardt et al. (2002) examined the syllable awareness of three children with
CAS and three children with typical speech and language development aged 6 to 8 years.
The groups were matched for age and gender, but were not matched for receptive
language ability. The syllable awareness task involved the children tapping out the
constituent syllables in 40 words of one to four syllables. Novel tasks (intra-syllabic
position and intra-syllabic structure) were also administered. Examination of the novel
tasks revealed that completion of the tasks required phoneme rather than syllable
awareness. In the ‘intra-syllabic position’ task, CVC words were represented with three
coloured blocks, and children were required to indicate which block (i.e., which sound)
changed when a pair of words that differed by one phoneme alone were presented (e.g.,
pan-can, bet-bat, fan-fat). In the ‘intra-syllabic structure’ task, black blocks were used to
represent consonant sounds and white blocks were used to represent vowel sounds. The
examiner placed down a white block to represent the vowel sound in a word (e.g., when
working on the word ‘low’, the examiner would place down a white block and explain
that it represented the /oʊ/ sound in the word ‘low’). The child was then required to
arrange the black blocks around the vowel sound to represent the phonological structure
of the word. The children with CAS performed poorly in comparison to the children with CAS on the syllable awareness and novel phoneme awareness tasks.

The poor performance of children with CAS on receptive phonological awareness tasks has challenged the conceptualisation of CAS as a purely motor based speech impairment (Marion et al., 1993; Marquardt et al., 2002). Marion et al. (1993) and Marquardt et al. (2002) concluded that phonological awareness deficits in children with CAS may be consequent of a deficit in the phonological representation system rather than an isolated speech motor control deficit (as described in the theoretical perspectives of CAS presented in section 1.1.2). Further investigation, however, is required to assess the phonological awareness skills of a larger number of children with CAS, and to compare their phonological awareness ability to children with equivalent receptive vocabulary and children with other types of speech disorder.

1.5.2 Nature of the speech disorder in children with CAS

Children with CAS usually experience persistent speech production deficits. A longitudinal evaluation of speech production in three children with CAS aged 4;6 to 7;5 (presented as three separate studies) revealed high rates of inconsistent speech errors, vowel errors, syllable omissions, consonant deletion, and consonant substitution up to age 7;5 (B. L. Davis, Jacks, & Marquardt, 2005; Jacks, Marquardt, & Davis, 2006; Marquardt et al., 2004). Shriberg et al. (1997a) examined the speech production of younger (aged 4;10 to 7;0) and older children (aged 7;1 to 14;11) with CAS. The authors reported that the older group exhibited fewer speech production errors than the younger group, but continued to use speech error patterns such as palatal fronting, deaffrication, cluster reduction, and liquid simplification. Stackhouse and Snowling (1992a) reported that two
children with CAS continued to exhibit speech production difficulty at 15 and 16 years of age despite their speech deficits becoming milder over time. Lewis, Freebairn, Hansen, Iyengar et al. (2004) reported that children with CAS (n = 10) exhibited speech errors in multi-syllabic words and during connected speech at 8 to 10 years. Thus, although the speech production difficulties of children with CAS lessen over time, the majority of children with the disorder continue to exhibit some speech errors well into their school-age or adolescent years.

Children with speech disorder who exhibit atypical speech errors (i.e., speech errors not usually seen in typical phonological development) are also more likely to experience poor phonological awareness and literacy outcomes (Dodd, 1995; Leitao & Fletcher, 2004; Leitao, Hogben, & Fletcher, 1997; Rvachew, Chiang, & Evans, 2007). Dodd (1995) concluded that children with a phonological disorder (characterised by consistent use of developmental and non-developmental phonological error patterns) were more likely to experience phonological awareness and reading delay in comparison to children with phonological delay (characterised by the use of developmental speech error patterns alone), inconsistent speech disorder (characterised by inconsistent use of phonological error patterns), and children with typical speech and language development. The relatively inferior literacy outcomes of children with consistent speech disorder have been replicated in later studies (Leitao & Fletcher, 2004; Leitao et al., 1997). Rvachew et al. (2007) explored the association between speech error type and phonological awareness ability in 58 children with speech disorder. The children were assessed during their pre-kindergarten and kindergarten year. The researchers reported that atypical sound
substitutions and a high level of syllable structure errors (e.g., consonant deletions and syllable reduction) were associated with poorer phonological awareness performance.

Children with CAS are likely to present with atypical speech errors and high rates of syllable structure errors that may place those affected at further risk for literacy deficits. McCabe, Rosenthal, and McLeod (1998) conducted a retrospective analysis of clinical files to isolate possible features of CAS that differentiated the disorder from other speech sound disorders. The authors reported that unusual sound substitutions were one factor that separated children the two groups. Lewis, Freebairn, Hansen, Iyengar et al. (2004) reported atypical speech errors in children with CAS. Further, comparatively high rates of syllable structure or consonant omission errors have been reported in multiple studies of children with CAS (Jacks et al., 2006; Lewis, Freebairn, Hansen, Iyengar et al., 2004; Shriberg et al., 1997a). The three differential diagnosis models highlighted in Section 2.1 all include either phonotactic difficulties (i.e., difficulty maintaining the syllable structure of an articulated word) or consonant deletions as distinctive features of the disorder (B. L. Davis et al., 1998; Ozanne, 1995; Shriberg, Campbell et al., 2003). In summary, atypical speech errors, high rates of syllable structure errors, and persistent speech difficulty all contribute to the vulnerability of children with CAS to phonological awareness, reading, and spelling disorder.

1.5.3 Language impairment in children with CAS

As described in the summary of the ‘modified critical age hypothesis’ (Nathan et al., 2004), language impairment in a child with speech disorder increases the likelihood that the child will develop phonological awareness, reading, and spelling deficits. Children with CAS present with language difficulties that can not be explained by their
restricted speech development alone. Ekelman and Aram (1983) examined the expressive syntactic performance within a conversational language sample of eight children with CAS aged 4;4 to 11;11 (with an average age of 8;8). The authors reported a range of syntactic deficits including inflectional morpheme omission or substitution, pronoun errors, and poor sentence structure due to copula or auxiliary errors. The children’s morphological skills and sentence structure were poor in comparison to their mean length of utterance (MLU), suggesting a disordered rather than delayed syntactic profile. A number of the children’s syntactic errors appeared unrelated to speech production skills. For example, children commonly had difficulty marking the third person singular morpheme with a final /s/ sound but could produce a final /s/ sound when marking the plural morpheme. The researchers concluded that children with CAS present with co-morbid speech production and syntactic deficits.

Lewis, Freebairn, Hansen, Iyengar et al. (2004) reported ongoing expressive and receptive language impairment in children with CAS that did not lessen as the children’s speech production skills improved over time. In fact, the language deficits of children with CAS became more pronounced over time. At preschool age the language profile of children with CAS could not be distinguished from a group of children with combined speech-language impairment on standardized measures. However, the children with CAS exhibited more severe language deficits at school-age. The researchers concluded that language impairment was a core symptom of CAS.

Language impairment in CAS will also inhibit the reading comprehension skills of those affected. As outlined in the ‘simple view of reading’ described in Section 1.4.4
(Hoover & Gough, 1990), successful connected reading is dependent upon fluent, accurate decoding and language comprehension.

1.5.4 Usual intervention practices for children with CAS

The usual speech and language therapy intervention practices utilised for children with CAS may also increase this group’s risk of reading and spelling disorder. The traditional view of CAS as a disorder of speech motor control has meant that treatment approaches for the population have focused exclusively on improving oral motor control and/or speech production ability in those affected (American Speech-Language-Hearing Association, 2007; Pannbacker, 1988). There has been no attempt to examine approaches designed to facilitate this group’s phonological awareness, reading, and spelling development.

Speech interventions for CAS generally include techniques based on imitation (Strand & Debertine, 2000; Strand et al., 2006), gestural and tactile cueing for articulatory position (Hayden & Square, 1994), and the use of melodic patterns to aid articulation (Helfrich-Miller, 1994). Traditional intervention approaches for CAS share some common intervention principals including: (1) use of multimodal cueing (auditory, visual, tacticle) for articulatory placement; (2) use of drill based exercises; (3) use of imitation, and (4) systematic progression from simple to more complex sound production (Marquardt & Sussman, 1991). However, the treatment approaches described for CAS are generally untested or evaluated by studies incorporating limited experimental control such as case studies (Pannbacker, 1988).

Dynamic temporal and tactile cueing (DTTC) is one approach based on traditional CAS intervention principles that has recently been evaluated via a multiple single-subject
design for three participants (Strand et al., 2006). DTTC shapes movement for speech production moving systematically from the most supported to most independent productions. Initially, the child produces a target utterance slowly and in chorus with the SLT. Tactile and gestural cues are provided if required to achieve correct articulatory placement in the production attempt. The child is required to produce the utterance at a faster rate and more independently as his/her accuracy increases. The approach also incorporates principles of motor learning emphasizing multiple, repeated trials of intervention targets that are presented in frequent, short intervention sessions (Strand et al., 2006).

Strand et al. (2006) examined the DTTC approach on four males aged 5 to 6 years with CAS in a single-subject design with repeated measures. The participants were described as ‘essentially non-verbal’ (two to four consonants in each respective phonemic inventory) and had received two to four years of therapy before the study. Intervention scheduling included two 30-minute sessions per day for five days a week over six weeks, with the children participating in 43, 46, 50 and 38 sessions respectively. Five or six target utterances that were meaningful to the child were drilled intensively before targeting another utterance. Untrained utterances were also selected as a control measure. Three children improved production of target utterances shortly following the onset of treatment. The fourth child, who also exhibited behavioural difficulties throughout the intervention, did not improve production of the trained utterances.

Although the DTTC approach is notable for the demonstration of its effectiveness for three children with severely limited speech production using a controlled research design, there appear to be some limitations to the approach. The intervention used
extremely intensive treatment scheduling and intervention targets were limited to a small number of utterances. Further, the remediation of single utterances without the demonstration of carryover to similar utterances suggests inefficiencies in the treatment approach. Finally, the approach targeted speech production skills alone without building skills underlying reading and spelling success. The lack of an evidence base to direct intervention practices may also contribute to the persistent nature of the speech production difficulties reported in children with CAS.

1.5.5 Summary of risk factors for reading and spelling disorder in CAS

Children with CAS present with multiple risk factors for reading and spelling disorder. These children are likely to exhibit all three factors outlined in the ‘modified critical age hypothesis’ that increase the probability that a child with speech disorder will experience written language difficulties. Those affected generally experience severe and persistent phonological awareness deficits, speech production difficulties that persist beyond age 7, and co-morbid language impairment. The presence of unusual speech errors and relatively high levels of syllable structure errors in the disorder create additional risks for the population. Finally, as CAS intervention practices focus solely on speech output, it is unlikely that participation in speech and language therapy will provide protection from the children’s predisposition for literacy disorder.

The ‘modified critical age hypothesis’ can also be used as a framework to develop intervention approaches that may improve the written language outcomes for children with CAS. According to the theory, speech production and phonological awareness are critical components for reading and spelling success in children with speech disorder. Although data evaluating the effectiveness of interventions designed to improve speech
production in children with CAS are limited, the literature suggests that speech deficits in
the population do minimise over time despite ongoing deficits in phonological awareness,
reading, and spelling development. Consequently, it is essential that approaches designed
to improve the phonological awareness of children with CAS are rigorously evaluated.

1.6 Phonological awareness intervention

Phonological awareness intervention is a successful method of enhancing reading
and spelling development for a wide variety of populations. Those benefiting from the
approach include young children at risk of reading disorder due to low socioeconomic
status or poor phonological awareness skills (Blachman, Ball, Black, & Tangel, 1994;
Torgesen et al., 1999), older children with reading disorder (Gillon & Dodd, 1995; Lovett
& Steinbach, 1997), and children with speech and language disorders (Gillon, 2000,
intervention facilitates reading and spelling development.

Phonological awareness intervention that incorporates letter-sound knowledge and
focuses primarily on phoneme awareness has stronger benefits for the reading and
spelling process. Teaching phonological awareness and letter-sound knowledge
concurrently allows the link between the phonological and orthographic form of words to
be demonstrated explicitly (Hatcher, Hulme, & Ellis, 1994). The child is thus better
equipped to use his/her increased sound awareness during the reading and spelling
process.

It is also critical that phonological awareness intervention focuses predominantly
at the phoneme (rather than the syllable and onset-rime) levels (Hatcher et al., 2004;
Nancollis et al., 2005). Phonological awareness at the phoneme level has the closest
association with reading and spelling success (see Section 1.4.5 for review). It follows that intervention that focuses on this level will better equip a child to use his/her improved speech sound awareness to assist in the reading and spelling process.

1.6.1 Integrated phonological awareness intervention

An integrated phonological awareness intervention that interweaves speech, letter knowledge, and phonological awareness goals may be a promising intervention approach for children with CAS. An integrated phonological awareness programme has proven effective in simultaneously developing speech, phonological awareness, letter knowledge, reading and spelling skills in school-aged and pre-school children with speech disorder (Gillon, 2000, 2002, 2005).

Gillon (2000) evaluated the effectiveness of an integrated treatment approach for children with CAS in a controlled design. Children aged between 5 and 7 years with speech disorder participated in either 20 hours of phonological awareness intervention ($n = 23$), 20 hours of traditional speech production therapy ($n = 23$), or a control condition that included minimal intervention ($n = 15$). The literacy development of 30 children with typical speech and language development was also followed throughout the intervention. The integrated intervention provided feedback regarding speech production within phonological awareness activities. Results showed that children in the phonological awareness intervention exhibited more advanced phonological awareness, decoding, and reading comprehension ability than both comparison groups immediately following the intervention. There was no difference in the spoken language outcomes of children receiving the traditional speech therapy and phonological awareness interventions. Thus,
the dual focus on phonological awareness and speech production did not detract from the children’s spoken language gains.

Long-term positive effects of the integrated phonological programme were also evident. Gillon (2002) re-assessed the speech and reading performance of children involved in the study eleven months following the completion of the intervention. An evaluation of the children’s spelling development was also completed. The findings showed that children who participated in the phonological awareness intervention experienced continued accelerated development in phonological awareness and literacy skills in relation to the comparison groups. Most of the children who received the phonological awareness intervention exhibited age-appropriate or advanced reading skills and showed strong spelling skills.

An integrated phonological awareness programme has also been successful at facilitating spoken and written language development of children aged 3 and 4 years with moderate or severe speech disorder (Gillon, 2005). Twelve children participated in an average of 23.5 sessions (one group and one individual session per week), where speech production practice was incorporated into explicit phoneme awareness and letter-sound knowledge activities. The effects of the programme were monitored until the child’s first or second year of school. Findings indicated that children who received the phonological awareness intervention experienced accelerated growth in phonological awareness and letter knowledge skills in comparison to their peers with typical speech and language development. Examination of the group’s early academic performance revealed reading skills within or above the expected range for all children. Gains in speech production were not compromised in the children who received the integrated intervention.
In contrast to the Gillon (2000, 2002, 2005) results, Hesketh, Adams, Nightingale, and Hall (2000) found no difference in the phonological awareness outcomes of 61 children with speech disorder aged 3;6 to 5;0 participating in ten weekly sessions of either a ‘metaphonological’ or ‘articulation-based’ intervention. Results showed that participation in the integrated phonological awareness intervention provided no advantage for phonological awareness development. The inability of the metaphonogical intervention to facilitate phonological awareness development is likely due to the nature of the intervention. Explicit phonological awareness tasks (i.e., rhyme awareness, phoneme identity, and phoneme segmentation) were limited to the first four sessions of the programme. The remaining sessions employed modified tasks from the Metaphon approach for speech production remediation (Dean, Howell, Waters, & Reid, 1995). Although phonological contrast approaches (such as the Metaphon approach) to speech remediation are effective in improving speech production, they are not successful at stimulating phonological awareness development (e.g. Gillon, 2000). The four sessions spent on explicit phonological awareness tasks (as opposed to the 20 hours used in the Gillon study) likely explains the contrasting results of the studies.

Pilot investigation of the effectiveness of an integrated phonological awareness approach for children with CAS showed that the approach may also be useful for this population. Moriarty and Gillon (2006) trialled an integrated phonological awareness approach on three children with a confirmed CAS diagnosis aged 6 and 7 years using a controlled multiple single subject design with repeated measures. The children received approximately seven hours of the research intervention in individual sessions over three weeks. The words used in the phonological awareness activities were based on the child’s
targeted speech error pattern. The approach incorporated targeted speech production practice into the phonological awareness activities and used letters and phonological cues to cue speech production. For example, if a child said ‘top’ instead of ‘stop’ (in response to a stimulus card with a picture cue and word written underneath in large clear font), the SLT would point to the letter s at the start of the word stop and say “Stop starts with a /s/ sound. See the letter s that makes the /s/ sound. Say stop again with a /s/ sound at that start.” Two children significantly improved their targeted speech skills. The children, however, did not improve on control speech measures (an untargeted phonological error pattern) over the intervention. All children significantly improved their phonological awareness skills following the intervention with two children transferring these skills to an untrained non-word reading task.

Although the results of the pilot are encouraging, further investigation is required to fully examine the usefulness of the programme for children with CAS. It is necessary to establish if the effects of the intervention noted immediately post-intervention in the two participants are able to be maintained over time. Further, the impact of an integrated programme on speech accuracy in connected speech should be documented. It is also important to trial the approach on a larger number of children across an expanded intervention period to allow closer examination of treatment effects.

**Theoretical foundations of the use of an integrated phonological awareness approach for children with CAS**

It is useful to consider the theoretical foundations of an integrated phonological awareness approach, so that the potential application of the approach for children with CAS can be explored. An integrated approach to intervention for children with CAS is
consistent with hypothetical accounts of CAS identifying poor phonological representation and motor programmes as the underlying deficit in the disorder (Marquardt et al., 2004; Marquardt et al., 2002). Recent research indicates access to well-specified phonological representations is important for both speech accuracy and phonological awareness ability (Rvachew, 2006; Sutherland & Gillon, 2007).

Rvachew (2006) explored variables that may contribute to poor phonological awareness skills in 47 4- and 5- year old children with speech disorder. The author reported that speech perception was related to phonological awareness and speech development. There was no relationship, however, between speech accuracy and phonological awareness. Similar results were reported by Rvachew and Grawburg (2006). The speech perception task employed in the above studies, which required children to judge whether words were articulated correctly or incorrectly, can also be conceptualised as a phonological representation measure. Similar judgement tasks have been utilised as a receptive phonological representation measure for children with speech disorder (see Sutherland & Gillon, 2005). The results also provide support for other studies concluding that a sole clinical focus on speech accuracy will not have positive effects on phonological awareness development (Gillon, 2000; Nathan et al., 2004).

Sutherland and Gillon (2007) examined the longitudinal relationship between phonological representation, speech production, phonological awareness, and reading in nine children aged 3;9 – 5;3 with moderate to severe speech disorder. Their performance on the above measures was compared to 17 children with typical speech and language development. The authors reported that the children with speech disorder exhibited inferior phonological representation ability when compared to their typically developing
peers throughout the 12-month study. There was a moderate correlation between the phonological representation and phonological awareness skills of the children. The authors concluded that unspecified phonological representations contribute to speech and phonological awareness difficulties in children with speech disorder.

An integrated phonological awareness intervention thus focuses on improving the accessibility and specificity of phonological representations to promote speech accuracy and phonological awareness ability. Targeted speech production practice is also included within the approach. Performing phonological awareness tasks on a target speech word may increase the specificity of the phonological representation of that word and provide a more stable motor program to direct speech production.

An integrated phonological awareness approach is also consistent with recent research highlighting the importance of visual-verbal paired associated learning (i.e. the ability to learn that a particular shape corresponds with a particular verbal response) and phoneme awareness for reading development (Hulme, Goetz, Gooch, Adams, & Snowling, 2007). Hulme et al. (2007) found that visual-verbal learning and phoneme awareness were independent correlates of reading ability in a sample of children aged 8 to 11 years with typical reading development. Visual-verbal learning was more strongly correlated with reading ability than visual-visual (i.e. learning that two shapes go together) and verbal-verbal learning (i.e. learning that two words go together). In line with the above findings, the integrated approach facilitates the development of visual-verbal learning (e.g. letter-sound knowledge training, using letters to prompt speech production) and phoneme awareness.
The theoretical foundations of an integrated phonological awareness approach provide a strong rationale for its use with children with CAS. The technique is aligned with a recent theoretical perspective of CAS (i.e., the phonological representation deficit hypothesis). The intervention also incorporates visual-verbal learning with phonological awareness training that has shown to be beneficial for reading development. Integrated phonological awareness intervention may thus have the potential to improve speech production, phonological awareness, reading, and spelling outcomes for children with CAS.

1.7 Summary and thesis aims

Preliminary studies show that children with CAS experience high rates of severe and persistent written language disorder. Those affected may be more likely than children with other speech and language impairments to experience phonological awareness, reading, and spelling failure. Comprehensive evaluation of the written language skills of children with CAS, however, has yet to be conducted. It is also unknown if children with CAS remain at elevated risk for written language disorder when they are compared to children with speech disorder with comparable receptive vocabulary and speech severity.

Consistent with reading and spelling models, the poor reading and spelling outcomes of children with CAS appear to be grounded in phonological awareness deficits. Although phonological awareness intervention has proven effective for other populations of children with speech disorder, their efficacy for children with CAS is untested. Current intervention approaches for children with CAS tend to focus exclusively on speech and have not been rigorously evaluated.
An integrated phonological awareness programme that concurrently targets phonological awareness, speech production, and letter knowledge may be a promising method of improving the spoken and written outcomes of children with CAS. The approach is consistent with a current theoretical view of CAS and models of reading development.

The primary aims of this thesis are to:

1. Describe the phonological awareness and early reading development of children with CAS.
2. Evaluate the effectiveness of an integrated phonological awareness programme to advance spoken and written language skills for children with CAS.

The following questions must be addressed in order to accomplish this objective:

1. What are the phonological awareness, reading, and spelling skills of children with CAS compared to children with other types of speech disorder who have similar speech severity and children with typical development?
2. What are the immediate effects of an integrated phonological awareness programme on the phonological awareness, letter knowledge, reading, spelling and speech sound developments of children with CAS?
3. What are the longer term effects of an integrated phonological awareness programme for children with CAS?

Chapter 2 will describe the phonological awareness, letter knowledge, and decoding skills of children with CAS. These skills will be compared to a group of
children with inconsistent speech disorder with similar receptive vocabulary and speech severity levels to the children with CAS. Chapter 3 will present the longitudinal results of a controlled pilot study examining the potential benefit of an integrated phonological awareness programme for two children with CAS. Chapter 4 will examine the effectiveness of an integrated phonological awareness programme in more detail on a larger number of children with CAS. The study will explore the speech, phonological awareness, reading, and spelling skills of children who participated in the programme at an individual (i.e., multiple single-study design) and group level (in comparison to children with typical speech-language development). Chapter 5 will present a six month follow-up study of the effects of the programme. Chapter 6 includes a longitudinal case study of identical twin boys involved in the research intervention prior to the commencement of formal schooling.
CHAPTER 2

INVESTIGATING PHONOLOGICAL AWARENESS AND EARLY READING DEVELOPMENT IN CHILDREN WITH CHILDHOOD APRAXIA OF SPEECH

2.1. Introduction

The literature review identified that children with childhood apraxia of speech (CAS) may exhibit more severe written language and phonological awareness deficits than children with other types of developmental speech and language disorders (Lewis, Freebairn, Hansen, Iyengar et al., 2004). These results suggest that the symptoms that comprise CAS interfere with reading, spelling and phonological awareness development to a greater extent than heterogeneous groups of speech-language disorders. One aspect of CAS that is not typically associated with other speech and language impairment is speech motor planning difficulty. The complex nature of CAS makes it difficult to determine the influence of speech motor control impairment on reading development, as other aspects associated with the disorder such as receptive vocabulary deficits, severe phonological speech errors are also associated with reading delay (Lewis, Freebairn, Hansen, Iyengar et al., 2004). The current study therefore aimed to extend previous research by comparing phonological awareness and early reading development in children with CAS to children who exhibit similar types of speech and language errors.
but do not display speech motor control difficulties. This comparison will help to identify any negative influence of the motor control difficulty characteristic of CAS on processes underlying reading development.

Inconsistent speech errors have been identified as one of the hallmark characteristics of CAS (B. L. Davis et al., 1998; Marquardt et al., 2004), but inconsistent speech errors can also occur in the absence of speech motor control difficulties (Dodd, Leahy, & Hambly, 1989; Holm, Farrier, & Dodd, in press). An inconsistent speech error (also termed token to token inconsistency) occurs when a child produces different misarticulations, upon repeated production, of the same word. For example, in three trials of naming the picture ‘shark’, the child may produce /gak/, /sak/, and /sat/. Under Dodd’s (1995) classification system, inconsistency is evaluated by asking children to name the same 25 words on three occasions within a session. An inconsistent production is marked if the child produces at least two different forms for a word across the three trials. Children who exhibit 40% or greater inconsistency across the 25 items are considered to have ‘inconsistent’ speech errors. Inconsistent speech errors indicate pervasive speech processing difficulties that restrict categorical development of new phonemes (Forrest, Dinnsen, & Elbert, 1997; Williams & Stackhouse, 2000). Forrest, Elbert, and Dinnsen (2000) argued that the presence of inconsistent speech errors increases the likelihood that a child will experience persistent speech disorder.

Children with inconsistent speech errors, with or without speech motor control difficulties, appear to have written language deficits (Holm et al., in press; Lewis, Freebairn, Hansen, Iyengar et al., 2004). Poor phonological awareness knowledge is likely to negatively influence these children’s written language development. Stackhouse
and Snowling (1992a) showed impaired phoneme identification, rhyme generation, and rhyme identification skills in two children aged 10;7 and 11 years with CAS at initial assessment and at follow-up four years later. Marion, Sussman, and Marquardt (1993) found severe deficits in rhyme generation and rhyme detection in four children aged 5 to 7 years with CAS compared to their peers with typical speech and language development. Similarly, Marquardt, Sussman, Snow, and Jacks (2002) found that the three children with CAS in their sample (aged 6 to 7 years) had severe deficits on a syllable segmentation task, and novel phoneme identity and phoneme manipulation tasks.

Holm et al. (in press) compared the syllable segmentation, rhyme awareness, and alliteration awareness skills of children with inconsistent speech disorder without speech motor control difficulties \( n = 15 \) with other groups of speech disorder namely, phonological delay \( n = 46 \), consistent phonological disorder \( n = 17 \), and a control group of children with typical speech development \( n = 15 \). Children with inconsistent speech disorder performed poorly on the syllable segmentation task alone. The authors also compared the performance of children with a history of inconsistent speech disorder \( n = 9 \) and children with a history of typical speech-language development in their third year of schooling on spelling, reading, phoneme awareness, and rhyme awareness tasks. The children with a history of inconsistent speech disorder demonstrated similar performance to that of their peers with typical speech and language development on the phoneme awareness, rhyme awareness and reading tasks, but performed less well on the spelling task.

The literature review highlighted the importance of segmental phonological representations in speech and phonological awareness development. Recent advances
have been made in the development of assessment measures of children’s phonological representations that do not require verbal out. Such measures are particularly useful in the assessment of children with speech disorder. Sutherland and Gillon (2005) trialled three receptive phonological representation measures (phonological representation judgement, non-word learning task and receptive gating tasks) on 9 children with moderate-severe speech disorder and 17 children with typical speech-language development aged 3 to 5 years. In the phonological representation judgement task children were asked to indicate the ‘correctness’ of the pronunciation of familiar multi-syllabic words with and without vowel errors by pointing to a happy face or a red cross. In the non-word learning task participants were taught a non-word as the name of a picture. Following structured teaching of the non-word and its referent, children were asked to judge the correctness of the non-word’s pronunciation with or without production errors (pointing to a green tick or a red cross to indicate correctness). The authors reported that the phonological representation judgement and non-word learning tasks were appropriate measures that distinguished between groups of children with and without speech disorder and were moderately correlated with phonological awareness skills. This study utilises the phonological representation judgement task for children with CAS and children with inconsistent speech disorder without speech motor control difficulty to identify possible phonological representation deficits in these population groups.

Although evidence (Lewis, Freebairn, Hansen, Iyengar et al., 2004; Marion et al., 1993; Marquardt et al., 2002) suggests that children with CAS are more likely to experience phoneme awareness and reading difficulty than children with inconsistent speech disorder, a direct comparison of these groups has not been conducted. Such a
comparison, particularly when there is no significant difference in speech severity and receptive vocabulary ability in the groups, will provide insight into how the presence of speech motor control difficulties in CAS may impact reading and phonological awareness development. Further, an evaluation of the phonological representation skills of the groups will give insight into processes underlying each group’s phonological awareness abilities.

The following hypotheses were tested:

1. That children with CAS will perform significantly below their peers with typical development (and similar receptive vocabulary) on all measures of phonologic awareness and reading.

2. That children with CAS will perform more poorly than children with inconsistent speech disorder on phonological awareness, letter knowledge, and reading tasks (despite both groups exhibiting similar performance on speech and receptive vocabulary measures).

3. That children with CAS will perform more poorly than children with inconsistent speech disorder on a receptive phonological representation judgement task.

2.2. Method

2.2.1. Participant selection process

Following Human Ethics\(^1\) approval to conduct the study and relevant approval from the New Zealand Ministry of Education, speech-language therapists (SLTs) employed by the New Zealand Ministry of Education participated in a day-long study.

\(^1\) The University of Canterbury’s Human Ethics Committee approved the study and standard procedures regarding anonymity of participants and parental permission for entry into the study were followed.
workshop regarding CAS where an assessment battery for the diagnosis of the disorder was described. The workshop was presented in six areas across the country. The assessment battery was based on Ozanne’s (1995, 2005) model (i.e., children must display impairment in the phonological planning, phonetic programme assembly and motor execution levels of speech production to be diagnosed with the disorder). Ozanne’s diagnostic model was selected as the literature reviewed showed that this framework encompassed the differential diagnostic symptoms used in other contemporary models of CAS diagnosis (Shriberg, Campbell et al., 2003; Davis et al., 1998).

Following the workshop, SLTs administered the assessment battery described in the workshop to children on their caseload aged 4 to 7 years with suspected CAS and who had no history of sensory, cognitive or neurological impairment. Children were assessed in a quiet room at their school or home over two sessions of one hour. The Olympus DS-2 digital voice recorder (with inbuilt stereo microphone) was used to record the assessment sessions. SLTs completed the initial online transcription. Assessment results were then forwarded to the researcher who reviewed all audio files (collected by the SLTs) following the assessment sessions to ensure consistent transcription and recording of assessment data. Further information regarding data verification by an independent transcriber can be viewed in the reliability section below.

The battery included the following assessments:

- Peabody Picture Vocabulary Test - III (PPVT-III) (Dunn & Dunn, 1997). This test is a measure of receptive vocabulary. Children are required to point to one of four pictures that correspond with a particular vocabulary item. A standard score was collected from this assessment
• Bankson-Bernthal Test of Phonology (BBTOP (Bankson & Bernthal, 1990). This is a single word articulation test consisting of 80 words of one to three syllables. All responses were transcribed via broad transcription. If a spontaneous response could not be elicited by the picture, delayed imitation was employed to gather a response from the child. All responses were transcribed via broad transcription. If a spontaneous response could not be elicited by the picture, delayed imitation was employed to gather a response from the child.

• Diagnostic Evaluation of Articulation and Phonology (DEAP) (Dodd, Hua, Crosbie, Holm, & Ozanne, 2006). The DEAP consists of four subtests (articulation assessment, phonology assessment, oro-motor assessment and inconsistency assessment. The oro-motor and inconsistency subtests were administered.

  o The oro-motor subtest consists of three components: isolated movements (e.g. lateral tongue movement), sequenced movements (e.g. blow and then put your tongue up) and diadochokinetic testing (elicit multiple trials of ‘pat-a-cake’ which was rated on sound sequencing, intelligibility and fluency). A standard score is calculated for each component (i.e. isolated movements, sequenced movements and diadochokinetic testing) to indicate performance within/below the expected range.

  o The test-retest reliability of the isolated movements, sequenced movements, diadochokinetic subtests have a Pearson correlation of 0.81, 0.67, and 0.60 respectively (Dodd et al., 2006). The inter-rater
The reliability of the diadochokinetic subtest has a Pearson correlation of 0.39. The agreement percentage on standard scores for the isolated movements and sequenced movements subtests range from 60% to 100%.

- In the inconsistency subtest, participants are required to articulate a set of 25 words three times within a session with an activity between each trial. A production is marked as inconsistent if the same item is articulated differently on two or three of the three trials. Children who are 40% and over inconsistent (i.e. produce an inconsistent error on ten or more of the 25 items) are deemed to have inconsistent production. All responses were transcribed via broad transcription. If a spontaneous response could not be elicited by the picture, delayed imitation was employed to gather a response from the child.

- Multi-syllabic real and non-word repetition task (Larrivee & Catts, 1999) to evaluate single-word articulation in spontaneous versus imitation contexts.

- Personal narratives were collected following a standardized protocol (Westerveld & Gillon, 2002) to allow informal evaluation of prosodic features (stress, loudness, resonance, pitch) during connected speech; to compare the child’s speech abilities in connected versus single-word contexts and to evaluate the presence of articulatory groping during connected speech.

Forty-four children were assessed for participation in the project by 20 SLTs and the first author with 12 children receiving a positive CAS diagnosis and a further 12 children matching an inconsistent speech disorder profile (Dodd, 2005). Two of the
twelve children in the CAS group were identical twins. No other children in the sample were siblings. The inclusionary criteria for each group are presented below:

**CAS group and Inconsistent Speech Disorder (ISD) group**

- Standard score (SS) below 1.5 standard deviations of the mean (i.e. SS below 77) on the BBTOP (Bankson & Berenthal, 1990). This criterion was used so that children with moderate to severe speech production difficulties were included in the study.

- Scored 40% or greater inconsistency on the inconsistency subtest of the DEAP (i.e. a whole word inconsistency measure) (Dodd et al., 2006). This criterion was used as inconsistent speech is a hallmark characteristic of CAS and ISD.

- Achieved a standard score within 1.5 standard deviations of the mean (i.e. a SS above 77) on the PPVT-III (Dunn & Dunn, 1997). This criterion was used so that the poor phonological awareness of participants could not be attributed to significant receptive vocabulary deficits.

**CAS group**

- a) Achieved standard scores below 8 on all three oro-motor sub-tests of the DEAP (i.e. isolated movements, sequenced movements and diadochokinetic) or

(b) Achieved a standard score below 8 on the diadochokinetic subtest and presence of articulatory groping during connected speech (i.e., noted during the child’s personal narrative production). Oral (along with verbal) apraxia is present in only a portion of children diagnosed with CAS (Davis et al., 1998).

**ISD group**
Achieved a standard score of 8 and above on all three sub-tests of the DEAP (Dodd et al., 2006) and did not exhibit articulatory groping during speech production.

A cross-comparison of the linguistic profiles of each child in the CAS and ISD group to the group of CAS differential diagnostic characteristics identified by Davis et al. (1998) is presented in Tables 3 and 4 below. The comparison shows that CAS diagnosis via the two methods corresponded well.

Negative diagnoses

Twenty children were assessed as for the study but did not match criteria for either group. Of those children, nine children achieved a PPVT (Dunn & Dunn, 1997) standard score below 77, five children achieved a standard score within 1.5 standard deviations of the mean on the BBTOP (Bankson & Bernthal, 1990), and six children were under 40% inconsistent on the inconsistency subtest of the DEAP (Dodd et al., 2006).
Table 3. Comparison of CAS group to Davis et al. (1998) diagnostic symptoms.

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<th>Characteristic</th>
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<tr>
<td>Limited phonemic repertoire</td>
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<td>Omission errors</td>
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<td>Vowel errors</td>
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<td>More errors on longer units</td>
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<td>Uses simple syllable shapes</td>
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<td>Diadochokinetic difficulties</td>
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**Note.** CAS = childhood apraxia of speech, + = characteristic present, - = characteristic absent, n = characteristic unable to be assessed (due to lack of production of multisyllabic words and/or connected speech).
Table 4. Comparison of ISD group to Davis et al. (1998) diagnostic symptoms.

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**Note.** CAS = childhood apraxia of speech, + = characteristic present, - = characteristic absent, n = characteristic unable to be assessed (due to lack of production of multisyllabic words and/or connected speech).

**Children with typical speech and language development (TD)**

A further group of 12 children with typical speech-language development were included as a peer comparison group. Teachers from two primary schools and one kindergarten in middle socioeconomic areas were asked to refer monolingual children of New Zealand-European descent with average literacy/language skills and no history of speech or language disorder. Participants in the comparison group exhibited speech and
language skills within the expected range (as measured by the PPVT-III and BBTOP). The first 12 children referred that met the inclusionary criteria for were included in this group. Assessment data for the three groups can be viewed in Table 5.

Demographic information

All children were from middle socioeconomic areas and were monolingual English speakers of New Zealand-European descent. Socioeconomic status was measured according to the ‘decile’ ranking given to the school. In New Zealand a decile ranking indicates the degree to which a school obtains its pupils from low socioeconomic areas. Decile one schools are the 10% of schools with the greatest percentage of pupils from low socioeconomic areas, while decile ten schools are the 10% of schools with the lowest percentage of pupils from low socioeconomic areas (Ministry of Education, 2007). The decile ranking of participants in the study ranged from four to seven indicating middle socioeconomic status. There were nine males and three female participants in each group and all children were aged 4 to 8 years.

Percent Consonants Correct (PCC) and Percent Vowels Correct (PVC) analysis

PCC and PVC analyses were completed by analysing participants’ responses from the BBTOP and the first trial of the 25-word consistency test (giving a sample of 105 words) with Computerized Profiling software (PROPH, Long & Fey, 2005). Further detail from the PROPH analysis for the children with CAS can be viewed in Chapter 4.

Assessment data for the three groups (CAS, ISD and TD) is presented in Table 3 below. A single-factor analysis of variance (ANOVA) revealed no significant group effect in age \([F (2, 33) = 2.37, p = 0.109]\) or receptive vocabulary \([F (2, 33) = 3.04, p = 0.062]\). Further analyses revealed a significant group effect for PCC \([F (2, 33) = 28.6, p < \)
0.001], inconsistency \[F (2,33) = 71.6, p < 0.001\] and PVC \[F (2,33) = 9.86, p < 0.001\] scores. Post-hoc pair-wise comparisons using Bonferroni tests showed the TD group had a higher PCC and more consistent speech productions than the CAS and ISD groups. The TD group had significantly higher PVC scores than the CAS group but not the ISD group. The only significant difference between the CAS and inconsistent groups was oro-motor performance \[t (22) = -4.267, p < 0.001\]. Oro-motor performance was not calculated for the TD group. The effect size index \(f\) (appropriate for ANOVA, Portney & Watkins, 2000) was also calculated for all statistically significant comparisons and can be viewed in Table 5. The effect size was interpreted as follows: 0.10 – small; 0.25 – medium and 0.40 and above – large (Portney & Watkins, 2000).
Table 5. Assessment data for the three groups

<table>
<thead>
<tr>
<th>Group</th>
<th>Mean</th>
<th>SD</th>
<th>Mean</th>
<th>SD</th>
<th>Mean</th>
<th>SD</th>
<th>F</th>
<th>p</th>
<th>f</th>
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</thead>
<tbody>
<tr>
<td></td>
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<td></td>
<td></td>
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<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>CAS (n = 12)</td>
<td>68.5</td>
<td>17.1</td>
<td>61.2</td>
<td>8.2</td>
<td>73.4</td>
<td>15.3</td>
<td>2.37</td>
<td>0.11</td>
<td></td>
</tr>
<tr>
<td>ISD (n = 12)</td>
<td>61.2</td>
<td>8.2</td>
<td>97.1</td>
<td>8.6</td>
<td>97.2</td>
<td>8.5</td>
<td>0.06</td>
<td>0.42</td>
<td></td>
</tr>
<tr>
<td>TD (n = 12)</td>
<td>73.4</td>
<td>15.3</td>
<td>79.2</td>
<td>8.5</td>
<td>15.3</td>
<td>0.2</td>
<td>9.86</td>
<td>&lt;0.00</td>
<td></td>
</tr>
</tbody>
</table>

Note. 1 = Peabody Picture Vocabulary Test – Third Edition (Dunn & Dunn, 1997), 2 Percent Consonants Correct, 3 Percent Vowels Correct, 4 Diagnostic Evaluation of Articulation and Phonology (Dodd et al., 2006), a** TD group performance significantly below comparison groups (p < .05), c** significant difference between CAS and ISD groups (p < .05).

2.1.1. Procedure

The participants from the three groups were administered the following phonological awareness, letter-sound knowledge, and reading tasks. All assessments were conducted individually in a quiet room in the children’s home or school.
Standardised phonological awareness assessment

- The Test of Phonological Awareness (TOPA, Torgesen & Bryant, 1994) (TOPA) was used for children aged five to eight years ($n = 7$ for CAS group, $n = 7$ for ISD group, $n = 8$ for TD group). This assessment provides a receptive measure of phoneme awareness. Responses to test items are made by participants marking one of four picture boxes. The test has two forms: (1) a kindergarten version for children aged 5 and 6 years that measures initial phoneme identity and (2) an elementary version for children aged 7 and 8 years that measures final phoneme identity. The TOPA has strong psychometric properties with internal consistency reliability, test-retest reliability, and inter-scorer reliability of $r = 0.80$ or above across all ages. Standard scores were collected for analysis.

- Preschool Inventory of Phonological Awareness (PIPA) (Dodd, Crosbie, MacIntosh, Teitzel, & Ozanne, 2000) was used for children aged 4 years ($n = 5$ for CAS group, $n = 5$ for ISD group, $n = 4$ for TD group). The rhyme awareness, alliteration awareness and phoneme identity sub-tests were administered. The standard scores from each subtest were added to form a composite score for analysis. The administered subtests have strong psychometric properties with test-retest reliabilities of $r = 0.870$, $0.803$ and $0.949$ respectively and internal consistency reliability coefficients from $r = 0.83 – 0.92$.

Letter-sound knowledge assessment
The Letter Knowledge task from the PIPA (Dodd et al., 2000) was administered to all children. This task requires the child to name the sound associated with 32 letters, digraphs, blends and vowels. Raw scores (out of a total of 32) were collected for analysis.

Phonological representation assessment

A phonological representation judgement task (Sutherland & Gillon, 2005) was administered to all participants. This receptive task requires the child to judge the correctness of 25 multi-syllabic words articulated by a New Zealand speaker by pointing to a tick or a check. Some items were produced correctly and others were produced with vowel errors. Five training items were included to familiarise the participants with the task. This task has high internal consistency with a coefficient alpha of 0.835 (Sutherland and Gillon, 2005). Full details about the development of this task may be viewed in Sutherland and Gillon (2005). Raw scores (out of 25) were collected for analysis.

Additionally the following measures were administered to children aged 6 years and older (n = 6 for CAS group, n = 5 for ISD group, n = 6 for TD group). As New Zealand children begin formal education at five years, all children who completed the reading measures had received at least one year of literacy instruction.

Word decoding assessment

Burt single-word reading test: New Zealand revision (Gilmore, Croft, & Reid, 1981). This task requires children to read a series of real words that are graduated for difficulty. Raw and age-equivalence scores were collected from
this assessment. Raw scores were used for data analysis as this assessment does not present standard scores. This assessment provides normative data from six to thirteen years.

- Informal non-word reading task (Calder, 1992). This test requires children to decode 30 non-words. The percent phonemes read correctly were calculated for analysis. For example, if a child read ‘vab’ as ‘vob’, he/she would be awarded 2/3 phonemes read correctly. A full list of the items in the task can be viewed in Appendix A.

2.1.2. **Reliability**

**Speech data**

In addition to the researcher reviewing all online transcriptions completed by the SLTs, transcription reliability was determined for a second independent transcription conducted by an experienced SLP on eight participants (just over 20% of participants). Mean inter-rater agreement, based on the number of phonemes transcribed identically, was 88.2% with a range of 78.4% to 97.3%.

**Non-word reading data**

An independent reviewer analysed and scored the non-word reading responses of eight participants (just over 20% of participants). Mean inter-rater agreement, based on the number of phonemes transcribed identically was 93.4% with a range of 87.7% to 100%.

2.3. **Results**
Multiple ANOVAs were employed to compare each group’s performance on phonological awareness, reading and phonological representation measures. The effect size index $f$ was also calculated for all statistically significant comparisons (Portney & Watkins, 2000).

**Phonological awareness**

An ANOVA performed on TOPA scores (used for participants aged 5 years and older) revealed a significant group effect \[ F(2,21) = 11.69, p < 0.001; \text{large effect size } f = 0.88 \]. 

*Post-hoc* pair-wise comparisons using Bonferroni tests revealed the TD group had significantly higher scores than the ISD and CAS groups. Further, the ISD group had significantly higher scores than the CAS group.

An ANOVA performed on PIPA scores (used for participants aged 4 years) revealed a significant group effect \[ F(2,10) = 6.456, p = 0.016; \text{large effect size } f = 1.14 \]. 

*Post-hoc* pair-wise comparisons using Bonferroni tests revealed the TD and ISD groups had significantly higher scores than the CAS group. There was no significant difference between the PIPA performance of the TD and ISD groups.

**Letter knowledge**

An ANOVA performed on letter knowledge raw scores revealed a significant group effect \[ F(2,33) = 3.827, p = 0.032; \text{large effect size } f = 0.49 \]. 

*Post-hoc* pair-wise comparisons using Bonferroni tests revealed the TD group had significantly higher scores than the CAS and ISD groups in the letter knowledge task. There was no significant difference between the performance of the CAS and ISD groups.

**Phonological representation**
An ANOVA performed on phonological representation scores revealed a significant group effect \( F(2,33) = 25.838, p < 0.001; \) large effect size \( f = 1.25 \). *Post-hoc* pair-wise comparisons using Bonferroni tests revealed the TD group had significantly higher scores than the CAS and ISD groups. There was no significant difference between the performance of the CAS and ISD groups.

**Non-word reading**

The Burt reading and non-word reading results were analysed qualitatively due to the small number of participants aged six years and above in each group. The five children with CAS aged 6-years or over (i.e., had received at least one year of formal reading instruction) were compared to the four children with ISD who had received a year of schooling and five children with TD. The CAS group appeared to have difficulty with the non-word reading task in comparison to children in the TD and ISD groups. The range of percent phonemes correct (PPC) achieved by the CAS group was 0–38% compared to 15-52% and 43-81% achieved by the ISD and TD groups respectively. Four of the five participants in the CAS group achieved a PPC score below 13%. This result should be interpreted cautiously, however, as participants in the ISD group are generally younger than the CAS and TD groups.

**Comparison to norms**

To control for the use of raw scores in the reading and letter knowledge measures, participants’ scores were compared to the normative data. The percentage of participants in each group that were performing within or above normal limits on the phonological awareness, letter knowledge, and Burt word-reading assessments are presented in Figure 1 (below). A standard score of 85 or above on the TOPA or standard scores over 7 on at
least two of the three administered subtests of the PIPA was the criterion for phonological awareness within/above normal limits. A standard score over 7 on the letter-knowledge subtest was the criterion for letter-knowledge skills within/above normal limits. The letter knowledge sub-test is normed for children aged three to six years so only 8, 12 and 8 children from the CAS, ISD and TD groups who were within this age range were compared to the norms respectively. An age equivalence score within/above that expected for a child’s age was the criterion for single word reading (Burt assessment) within/above normal limits. The Burt word reading test is normed for children aged six to 12 years so only 5, 4, and 7 children for the CAS, ISD and TD groups who were within this age range were compared to the norms respectively.

These results show that the CAS group have fewer participants performing within/above normal limits than the ISD and TD groups in phonological awareness, letter knowledge and word reading. The ISD group also has a greater proportion of participants than the TD group performing below normal limits in all areas. Two children in the TD group performed below normal limits on the letter knowledge task. These children named all letter names correctly, but had difficulty labelling the corresponding sound especially for digraph, clusters and vowels.
Figure 1. Percentage of children in each group within or above normal limits in phonological awareness, letter knowledge and word reading ability.

Note. PA = phonological awareness \((n = 36)\) (Preschool and Primary Inventory of Phonological Awareness (PIPA) performance for children aged 4 years; Test of Phonological Awareness (TOPA) performance for children aged 5 to 8 years), LK = letter-sound knowledge \((n = 36)\); Burt = Burt word reading test \((n = 20)\); CAS = Childhood Apraxia of Speech group; ISD = inconsistent speech disorder group; TD = typically developing group

2.4. Discussion

This study compared the phonological awareness, letter knowledge, reading, and phonological representation ability of children with CAS, children with inconsistent speech disorder, and children with typical speech-language development. All groups
exhibited comparable receptive vocabulary ability, while the children with CAS and inconsistent speech disorder also exhibited similar speech severity.

The first hypothesis tested was that children with CAS will perform significantly below their peers with typical development (and similar receptive vocabulary) on all measures of phonologic awareness and reading. The data supported this hypothesis. The TD group outperformed the CAS group in all phonological awareness and reading measures.

The second hypothesis tested was that children with CAS would perform more poorly than children with inconsistent speech disorder on phonological awareness, letter knowledge, and reading tasks (despite both groups exhibiting comparable speech severity and receptive vocabulary levels). This hypothesis was supported partially by the data. The children with CAS exhibited poorer phonological awareness scores than the inconsistent speech disordered group, but no difference was found between the groups on the letter knowledge and reading measures.

The finding that children with CAS performed more poorly than the inconsistent group on the phonological awareness measures supports previous research indicating that children with CAS are likely to experience more severe phonological awareness deficits than children with other types of speech-language impairment (Lewis et al., 2004). The finding also extends this research by demonstrating that children with CAS continue to perform more poorly despite the comparison group presenting with similar speech error types, receptive vocabulary ability, and speech severity. The result is further consistent with previous research demonstrating that children with inconsistent speech disorder have an isolated difficulty in syllable awareness (Holm et al., in press), with this group
performing well on the receptive phonological awareness measures used in the current study that did not contain a syllable awareness component.

The real word and non-word reading performance of the groups was analysed qualitatively due to the small number of participants over 6 years in each group. The analysis indicated that participants in the CAS group performed inferiorly on the reading tasks in comparison to children of a similar age with inconsistent speech disorder and typical speech-language development. The CAS group appeared to have particular difficulty with the non-word reading task. This finding is expected given the group’s poor phonological awareness, as non-word reading requires the use of phonological information in the reading process (Gillon, 2004). Results also indicated that the children with inconsistent speech disorder performed more poorly on the reading tasks than the children with typical development. These results must be interpreted cautiously however as raw scores were compared for the reading measures (rather than the standard scores compared for the phonological awareness measures). Although there was no significant difference in the ages of the groups, the CAS group had more 7 year old participants than the inconsistent group, meaning that the raw scores may not have provided a fair comparison of the reading abilities of both groups given the variability in exposure to formal reading instruction.

The finding that a higher percentage of children in the CAS group performed below the expected range for their age when compared to normative data than the inconsistent and control groups on all measures is consistent with previous investigations indicating that children with CAS are likely to experience severe phonological awareness and reading deficits (Lewis, Freebairn, Hansen, Iyengar et al., 2004; Marion et al., 1993;
Marquardt et al., 2002; Stackhouse & Snowling, 1992a). The findings from this study confirm that children with CAS are more likely to experience deficits in phonological awareness and reading than children with other speech-language disorders (Lewis, Freebairn, Hansen, Iyengar et al., 2004). The finding extends previous research by demonstrating that a greater proportion of children with CAS perform below typical range on letter knowledge and reading measures than a speech disordered comparison group despite the comparison group presenting with similar speech error types, receptive vocabulary ability, and speech severity.

Despite the inconsistent group having a greater proportion of children than the CAS group performing within the expected range on the phonological awareness and reading measures, this group had a greater proportion of children underperforming than the control group. This result conflicts with previous findings that children with inconsistent disorder are at risk for spelling rather than reading difficulties (Dodd, 1995; Holm et al., in press). However, the reading and letter knowledge measures required verbal output which may have confounded the results in these tasks given the multiple speech errors exhibited by the CAS and inconsistent groups.

The second hypothesis tested was that children with CAS would perform more poorly than children with inconsistent speech disorder and children with typical speech-language development on a phonological representation judgement task. The data did not support the hypothesis with both the CAS and inconsistent groups performing below the control group on this task. The inferior performance of the CAS group is consistent with previous research that concluded children with CAS have deficiencies in their
phonological representational systems, due to this population’s poor performance on receptive phonological tasks (Marion et al., 1993; Marquardt et al., 2002).

One explanation for the unexpected result is that children with inconsistent speech disorder have a deficit in their phonological representation system. Alternatively, the poor performance of the CAS and inconsistent groups may be due to different causes. Dodd (1995) argued that children with inconsistent speech disorder have an isolated deficit in phonological assembly (the creation of phonological plans for speech production). Phonological assembly deficits are hypothesised to disrupt the translation of words into phonological working memory. It follows that the inconsistent group’s performance on the phonological representation judgment task may be impeded, as they are required to compare the clinician’s pronunciation to a potentially distorted assembled target in working memory. Thus, difficulty with the task could stem from underspecified phonological representations of words in long term memory or impaired phonological assembly for working memory.

Phonological awareness findings in the current study support differing causes of impaired performance in the phonological representation judgement task for the experimental groups. The performance of the CAS group was inferior to that of the inconsistent group on the receptive phonological awareness measure, whereas the performance of the two groups was comparable on the phonological representation task. The completion of phonological awareness tasks is thought to necessitate reflection on the phonological representation of a word in long term memory (Elbro et al., 1998). Thus, the phonological awareness results indicated that the group with inconsistent speech disorder presented with stronger representational abilities than the CAS group. In
contrast, there was no difference in the groups’ performance on the phonological representation judgement task, which may indicate that the inconsistent group’s performance was hindered by poor phonological assembly while the CAS group’s performance was hindered by their underspecified representational systems.

The above findings must be interpreted with caution, however, as the phonological representation judgement task employed was developed for children aged 4 to 5 years (Sutherland and Gillon, 2006). The task may not have been sufficiently sensitive to detect differences in the phonological representation abilities of the CAS and inconsistent speech disordered groups. Further, the raw scores gathered from the phonological representation judgement task are subject to the same possible age effects outlined for the reading and letter knowledge measures. There is a need for future research that explores the development and assessment of phonological representation of older children with and without speech disorder.

The current study identified 12 children with CAS according to a current published diagnostic model and compared their phonological awareness and early reading development to children with inconsistent speech disorder. The findings confirmed the presence of reading, spelling, phonological awareness, and phonological representation deficits in children with CAS. The development of an intervention to improve the written language outcomes of those affected is required, so the spiralling negative effects of literacy disorder can be prevented. The following chapter presents the longitudinal results of a pilot study investigating the effectiveness of an intervention for children with CAS designed to remediate processes underlying spoken and written achievement in those affected.
CHAPTER 3

A FOLLOW UP PILOT STUDY EXAMINING TREATMENT EFFECTIVENESS FOR TWO CHILDREN WITH
CHILDHOOD APRAXIA OF SPEECH

3.1 Introduction

The study presented in Chapter 1 confirmed that children with childhood apraxia of speech (CAS) are particularly susceptible to phonological awareness, reading, and spelling deficits. As outlined in the literature review, a pilot study revealed that an integrated phonological awareness programme was effective in enhancing both the spoken and written language development of two of the three participants with CAS (Moriarty & Gillon, 2006). However, the pilot study only evaluated the effects of the programme immediately post-intervention. It is therefore unknown whether the intervention had any lasting impact on these children’s speech, phonological awareness, and reading skills.

The present experiment monitored the long-term effects (12 months post-intervention) for the two children who participated in the pilot study who demonstrated significant improvement immediately following the intensive three week intervention.
The following hypotheses were tested:

1. The children would maintain the post-intervention accuracy score (percent phonemes correct, PPC) for target speech items at the follow-up assessment.
2. The children would improve the accuracy (PPC) of control speech items over the follow-up period.
3. The children would maintain the post-intervention accuracy score (percent correct) for trained and untrained phoneme segmentation measures at the follow-up assessment.
4. The children would decrease the difference between their chronological age and age equivalence score on normative decoding and phonological awareness tasks from post-intervention to follow-up assessment.
5. The children improve their accuracy in a non-word reading task during the follow-up period.

3.2 Method

3.2.1 Participants

Two children (Derek$^2$ and Katie) participated in the follow up study. Derek was aged 8;3 at follow-up assessment and Katie was aged 7;3. The children were diagnosed with CAS via Ozanne’s (1995, 2005) diagnostic model prior to participation in the study. A summary of the children’s assessment data at pre-intervention can be viewed in Table 6.

$^2$ Pseudonyms are used in this paper.
Suppression of the s-cluster reduction speech error pattern was targeted during the intervention for both children. Derek’s control speech error pattern (i.e. the speech error pattern that was monitored but not treated during intervention) was velar fronting. Katie’s control speech error pattern was r-cluster reduction. Both children significantly improved the accuracy of their targeted speech words. Katie also exhibited a small improvement in the accuracy of control words. Descriptive analysis showed the improvement was due to more accurate vowel production at post-intervention rather than suppression of the r-cluster speech error pattern. Derek did not improve in the production of his control words. The children improved in their phoneme segmentation and manipulation of trained and untrained words and in an untrained non-word reading task.
<table>
<thead>
<tr>
<th></th>
<th>Derek</th>
<th>Katie</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Pre-intervention age (years;months)</strong></td>
<td>7;3</td>
<td>6;3</td>
</tr>
<tr>
<td><strong>Non-verbal intelligence: TONI quotient</strong></td>
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<td>115</td>
</tr>
<tr>
<td><strong>Language</strong></td>
<td></td>
<td></td>
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<td>PPVT quotient</td>
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<td>106</td>
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<tr>
<td>CELF (4) receptive language score (standard score)</td>
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<tr>
<td>CELF (4) expressive language score (standard score)</td>
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<td>80</td>
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<tr>
<td><strong>Speech</strong></td>
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<td>Percent vowels correct (PVC)</td>
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<td>Inconsistency percentage (DEAP)</td>
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<td>52</td>
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<td>Normal</td>
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<tr>
<td>Diadochokinetic sequencing</td>
<td>Reduced</td>
<td>Reduced</td>
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<td>4;0 – 4;5</td>
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<td>Letter knowledge (raw score out of 32)</td>
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<td>24</td>
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<tr>
<td>Non-word reading (percent phonemes correct)</td>
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<td>44.9</td>
</tr>
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<td>Word decoding*</td>
<td>6;3 – 6;9</td>
<td>5;9 – 6;3</td>
</tr>
</tbody>
</table>
**Table 6 continued.**

**Note.** TONI = Test of Non-Verbal Intelligence (Brown, Sherbenou, & Johnsen, 1990), PPVT = Peabody Picture Vocabulary Test (Dunn & Dunn, 1997), CELF = Clinical Evaluation of Language Fundamentals (Semel, Wiig, & Secord, 2003), DEAP = Diagnostic Evaluation of Articulation and Phonology (Dodd *et al.*, 2006) *Burt Word Reading Test (Gilmore *et al.*, 1981) age equivalent band.

### 3.2.2 Intervention

As described in Chapter 1, the children received 7 hours of an integrated phonological awareness programme (Gillon & McNeill, 2006) administered by the author to each child individually over a three week period. The intervention aimed to improve speech production, letter knowledge and phoneme awareness simultaneously. The children continued to receive their regular class programme during the three week period of intervention, but received no other forms of intervention.

Following the intervention, Derek received speech-language therapy support once every three to four weeks during the school year administered by his local speech-language therapist. The therapist administered her own intervention which generally followed a traditional approach to speech production and focused on improving his receptive and expressive language skills. Katie did not receive any further speech-language therapy support following the intervention programme. The children were not involved in any specialist reading programmes over the follow-up period.

### 3.2.3 Procedure
The children were re-assessed in a quiet withdrawal setting at school 12 months following the pilot study. The Olympus DS-2 digital voice recorder (with inbuilt stereo microphone) was used to record the sessions for data verification and reliability purposes. The following assessments were re-administered at follow-up:

**Repeated measure assessments**

The same speech and phonological awareness assessment probes used in the pilot study (Moriarty & Gillon, 2006) were re-administered at follow-up assessment. During the pilot study each speech target and control word, and the phoneme segmentation and manipulation stimulus items were probed three times over a two-week period at baseline and post-intervention assessment. This procedure was consistent with the repeated measures subject design of the intervention pilot study. However, the assessment probes were only administered once at the follow-up assessment.

**Speech probes**

The speech probe consisted of 12 trained and 12 control words (pertaining to each child’s trained and control speech error pattern). The percent phonemes correct (PPC) was calculated for these items.

**Phoneme awareness probes**

The phoneme segmentation probe consisted of 10 items where the child was required to segment the probe word into its constituent phonemes using coloured blocks. For example, the word ‘back’ would be segmented into three different phonemes /b ae k/, represented by three different block colours. The PPC was calculated for trained and untrained items. For example, if the child segmented the word ‘stop’ into two sounds, he/she would be awarded two out of a possible four points. The stimulus words for the
trained phoneme segmentation probes were taken from the children’s target speech probes (i.e., s-cluster words). The stimulus words for the untrained phoneme segmentation probes were taken from the children’s control speech probes (i.e., words containing the /k/ sound for Derek, r-cluster words for Katie).

The phoneme manipulation probes required each child to manipulate letter tiles to demonstrate sound changes within a word. For example, a child would change the word ‘stop’ to ‘step’ by swapping an ‘o’ letter tile with an ‘e’ letter tile. A percentage correct (out of 9 items) was calculated for the task. The chain of words used for the trained phoneme manipulation were based around words from the children’s target speech production probes and were thus included within the intervention sessions. The chain of words used for the untrained phoneme manipulation probes included words from the children’s control speech production probes and were thus not included in the intervention sessions.

Children were given practice items before administration of the phoneme awareness probes to ensure the task was understood.

Pre-intervention and follow-up measures

The measures listed below were administered pre-intervention and at follow-up (except for the non-word reading task, which was also administered immediately post-intervention). More detailed description of these assessments is provided in Chapter 2.

*Standardised decoding measure*

The Burt Word Reading Test– New Zealand Revision (Gilmore et al., 1981) was used to measure real word decoding. An age equivalent band and raw scores were collected from this assessment.
Standardised phonological awareness measure

The Phonological Awareness Skills Program Test (Rosner, 1999) was administered. In this assessment, children are required to delete syllables or phonemes from a target word. For example, ‘say cowboy without the boy’. An age equivalence band and raw scores (i.e., how many items were answered correctly) were collected from this assessment.

Informal non-word reading task

An informal non-word reading task (adapted from Calder, 1992) was used to measure non-word decoding (See Appendix A). The children were required to decode three sets of ten non-words of Consonant-Vowel (CV), CVC or CCVC structure. The percentage of phonemes correctly decoded for this task was calculated.

Supplementary follow-up measure

The following measure was administered at follow-up alone:

Standardised connected reading measure

The Neale Analysis of Reading Ability – Third Edition (Neale, 1999) was administered at follow-up to evaluate the children’s reading accuracy and comprehension in connected text. In this assessment children are required to read a short passage and then answer some comprehension questions about that passage. Percentiles were collected from the accuracy and comprehension subtests.

3.2.4 Data reliability

An independent SLT transcribed all follow-up speech and phoneme segmentation probes. Inter-rater agreement (based on the number of phonemes transcribed identically by the initial and independent analysis) was 91.7% and 97.3% respectively. Any
differences between the transcribed and the reviewed responses were resolved by the reviewers reaching consensus following repeatedly listening to recordings.

An independent SLT analysed and scored the non-word reading responses of both participants. Mean inter-rater agreement (based on the number of phonemes transcribed identically by the initial and independent analysis) was 92.3%. Any differences between the transcribed and the reviewed responses were resolved by the reviewers reaching consensus following repeatedly listening to recordings.

3.3 Results

3.3.1 Speech and phoneme awareness probes

The children’s performance in the speech probes at baseline, post-intervention, and follow-up are presented in Table 7. The results show that Derek’s speech production for both target and control items were similar to that achieved at post-intervention. Derek’s follow-up score for his control speech measure was not significantly different from the baseline phase of the intervention project. Katie maintained 100% accuracy in s-cluster production from post-intervention to follow-up and also showed improvement in r-cluster production over the follow-up period.
Table 7. Performance on speech production measures (percent phonemes correct).

<table>
<thead>
<tr>
<th></th>
<th>Baseline M (SD)</th>
<th>Est 2SD</th>
<th>Post-Intervention M (SD)</th>
<th>Follow-up M (SD)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Derek</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Target Speech</td>
<td>52.7 (4.6)</td>
<td>43.5 – 61.9</td>
<td>92.0 (0.0)*</td>
<td>91.7</td>
</tr>
<tr>
<td>Control Speech</td>
<td>56.3 (4.0)</td>
<td>48.3 – 64.3</td>
<td>57.0 (3.5)</td>
<td>62.5</td>
</tr>
<tr>
<td><strong>Katie</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Target Speech</td>
<td>60.3 (4.6)</td>
<td>51.1 – 69.5</td>
<td>100.0 (0.0)*</td>
<td>100.0</td>
</tr>
<tr>
<td>Control Speech</td>
<td>58.3 (1.2)</td>
<td>55.9 – 60.7</td>
<td>64.0 (1.7)*</td>
<td>89.6</td>
</tr>
</tbody>
</table>

**Note.** 1Base line phase and post-intervention scores are the mean scores (M) and standard deviations (SD) from three assessment trials over a two-week period, Est 2SD = Estimated range of two standard deviation band method (post-intervention scores must be above the estimated range to be considered significant), 2 one probe was administered at follow-up, all figures indicate percent phonemes correct (PPC), * = significant change.

The children’s performances on the phoneme awareness probes at baseline, post-intervention and follow-up are presented in Table 8. Derek failed to maintain the percentage achieved immediately post-intervention. However, all of his follow-up scores were significantly greater than that achieved in the baseline phase of the intervention project. Katie achieved similar scores at post-intervention and follow-up assessments.
### Table 8. Phoneme Awareness Results (percent correct).

<table>
<thead>
<tr>
<th></th>
<th>Baseline¹</th>
<th>Est 2SD</th>
<th>Post-Intervention¹</th>
<th>Follow-up²</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>M (SD)</td>
<td></td>
<td>M (SD)</td>
<td></td>
</tr>
<tr>
<td>Derek</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Trained PS</td>
<td>0.0 (0.0)</td>
<td>0.0 – 0.0</td>
<td>91.7* (2.1)</td>
<td>31.3</td>
</tr>
<tr>
<td>Untrained PS</td>
<td>0.0 (0.0)</td>
<td>0.0 – 0.0</td>
<td>71.1* (4.6)</td>
<td>32.5</td>
</tr>
<tr>
<td>Trained PM</td>
<td>22.2 (0.0)</td>
<td>22.2 – 22.2</td>
<td>93.3* (6.4)</td>
<td>55.6</td>
</tr>
<tr>
<td>Untrained PM</td>
<td>14.8 (6.4)</td>
<td>2.0 – 27.6</td>
<td>74.1* (6.4)</td>
<td>55.6</td>
</tr>
<tr>
<td>Katie</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Trained PS</td>
<td>46.7 (2.9)</td>
<td>40.9 – 52.5</td>
<td>98.4* (4.3)</td>
<td>100.0</td>
</tr>
<tr>
<td>Untrained PS</td>
<td>45.0 (4.3)</td>
<td>36.4 – 53.6</td>
<td>80.0* (1.4)</td>
<td>92.5</td>
</tr>
<tr>
<td>Trained PM</td>
<td>25.9 (6.4)</td>
<td>13.1 – 38.7</td>
<td>92.6* (6.4)</td>
<td>88.9</td>
</tr>
<tr>
<td>Untrained PM</td>
<td>37.0 (6.4)</td>
<td>24.2 – 49.8</td>
<td>81.5* (6.4)</td>
<td>77.8</td>
</tr>
</tbody>
</table>

**Note.** ¹Base line phase and post-intervention scores are the mean scores (M) and standard deviations (SD) from three assessment trials over a two week period, Est 2SD = Estimated range of two standard deviation band method (post-intervention scores must be above the estimated range to be significant), ²one probe administered at follow-up, all figures indicate percent correct, PS = phoneme segmentation, PM = phoneme manipulation, * = significant change.

### 3.3.2 Phonological awareness, decoding, and connected reading measures

The children’s performance at baseline and follow-up assessment on the Burt Word Reading Test (Gilmore *et. al*, 1981) and Phonological Awareness Skills Program
(Rosner, 1999) is presented in Table 9. Derek continued to perform below the expected range on the decoding and phonological awareness measures at follow-up assessment. However, his age-equivalence score for the phonological awareness measure increased by two and a half years from pre-intervention to follow-up. Katie performed within normal limits on the decoding and phonological awareness measures at follow-up, despite exhibiting phonological awareness deficits at pre-intervention.

The children’s performance in the informal non-word reading task (Calder, 1992) at baseline, post-intervention, and follow-up is presented in Table 10. Both children continued to improve their non-word reading performance over the follow-up period. Descriptive analysis indicates that the gains achieved in the intervention period are more marked than that achieved over the follow-up period.

Table 9. Pre and Follow-up Phonological Awareness and Reading Performance

<table>
<thead>
<tr>
<th></th>
<th>Burt Reading Test</th>
<th>PASP</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Pre-intervention</td>
<td>Follow-up</td>
</tr>
<tr>
<td></td>
<td>Raw</td>
<td>AE</td>
</tr>
<tr>
<td><strong>Derek</strong></td>
<td>27</td>
<td>6;9–7;0</td>
</tr>
<tr>
<td><strong>Katie</strong></td>
<td>13</td>
<td>6;0–6;3*</td>
</tr>
</tbody>
</table>

Note. PASP = Phonological Awareness Skills Program (Rosner, 1992), Burt Raw = raw scores (number of words read correctly), PASP Raw = raw score (number of items correct), AE = age-equivalence scores reported (years;months), * = within normal limits
Table 10. Pre, Post and Follow-up Performance on the Non-word Reading Test.

<table>
<thead>
<tr>
<th></th>
<th>Pre-Intervention</th>
<th>Post-Intervention</th>
<th>Follow-up</th>
</tr>
</thead>
<tbody>
<tr>
<td>Derek</td>
<td>7.1%</td>
<td>55.7%</td>
<td>68.0%</td>
</tr>
<tr>
<td>Katie</td>
<td>44.9%</td>
<td>72.2%</td>
<td>79.4%</td>
</tr>
</tbody>
</table>

Note. All scores represent the percentage of phonemes correctly decoded.

Analysis of the children’s performance on the Neale Analysis of Reading Ability (Neale, 1999) at follow-up assessment revealed that both children performed within the expected range on the accuracy measure, but below the expected range on the comprehension measure. Derek achieved an accuracy percentile rank of 23 and a comprehension percentile rank of 7. Katie achieved an accuracy percentile rank of 42 and a comprehension percentile rank of 3.

3.4 Discussion

This follow-up study evaluated the long-term effects of a short and intensive integrated phonological awareness programme on the speech, phonological awareness, and reading skills of two children with CAS. The intervention consisted of just less than seven hours of treatment administered individually over a three-week period administered by the author. The two children demonstrated significant positive treatment effects immediately post-intervention and the follow-up evaluation reported in this chapter examined the children’s performance on speech, reading, and phonological awareness measures 12 months after post-intervention assessment.
The first hypothesis tested was that the Derek and Katie would maintain the post-intervention high accuracy score (PPC) (92%, and 100% correct respectively) for target speech items at the follow-up assessment. This hypothesis was supported by the data. The children scored within 1% of their post-intervention PPC at follow-up for trained speech items. The findings indicated that a long-term change in speech production for children with CAS can be achieved over a relatively short treatment period. The results are in contrast to reports that children with CAS demonstrate slow progress in therapy (Campbell, 1999).

The second hypothesis tested was that the children would improve the accuracy (PPC) of control speech items over the intervention period. The hypothesis was partially supported by the data. Katie demonstrated gains in the accuracy of the control speech measure with her PCC for improving from 64% to 90% over the follow-up period. These results are consistent with reports demonstrating that the speech production deficits of children with CAS minimise over time (Lewis, Freebairn, Hansen, Iyengar et al., 2004; Stackhouse & Snowling, 1992a).

Derek’s production of words containing velar sounds showed a modest gain from 57% to 62% over the follow-up period. Closer analysis demonstrated that Derek’s accuracy in the control speech measure at follow-up was not significantly higher than that achieved in the baseline phase of the intervention study. Derek’s results highlight the effectiveness of the research intervention. He was able to make significant gains in target speech production over a three week intervention period which were maintained over a 12 month follow-up period. However, he was unable to significantly improve the
accuracy of the control speech measure over the 12 month follow-period despite other types of therapy intervention.

The third hypothesis tested was that the children would maintain the post-intervention accuracy score (percent correct) for trained and untrained phoneme segmentation measures at the follow-up assessment. This hypothesis was supported partially by the data. Katie maintained the accuracy achieved in these measures from post-intervention to follow-up assessments. Derek’s follow-up performance in trained and untrained phoneme segmentation probes fell by 61% and 38% respectively, and his follow-up performance in trained and untrained phoneme manipulation probes fell by 37% and 38% respectively. Further analysis revealed, however, that Derek’s follow-up scores in the phoneme awareness measures were significantly above those achieved in the baseline phase of the intervention study (despite his difficulty maintaining his post-intervention performance).

Derek’s difficulty in maintaining phoneme awareness gains may be due to the severe nature of his phonological awareness impairment. Prior to the participation in the intervention, he achieved an age-equivalence score of 4;0 – 4;5 in a standardised phonological awareness measure and was unable to use a phonological strategy in decoding. These results suggest that a longer intervention period may have been necessary for Derek to maintain accelerated growth in phoneme awareness.

The fourth hypothesis tested was that the children would decrease the difference between their chronological age and age equivalence score on normative decoding and phonological awareness tasks from post-intervention to follow-up assessments. The data supported this hypothesis for the phonological awareness measure alone. The
discrepancy between Derek’s age equivalence score and chronological age decreased by two years in the phonological awareness measure from pre-intervention to follow-up. Katie performed within the expected range on the phonological awareness measure at follow-up (despite demonstrating a severe deficit of two years below her chronological age at pre-intervention). The discrepancy between Derek’s age equivalence score and chronological age for the decoding measure was comparable at pre-intervention and follow-up. Katie continued to decode within the expected range for her age at follow-up. These findings suggest that a short period of integrated phonological awareness intervention can have a positive long-term influence on the phonological awareness ability of children with CAS. However, a longer intervention period may be required for the intervention effects to transfer to real word decoding.

The fifth hypothesis tested was that the children would continue to improve in the accuracy on a non-word reading task during the follow-up period. This hypothesis was supported by the data with both children achieving a higher PPC score on the task at follow-up than post-intervention assessment. Descriptive analysis, however, revealed that the children’s gains in non-word reading performance were more marked during the three week intervention phase than the 12 month follow-up phase. Derek improved his non-word reading performance by 49% over the intervention period and 12% over the 12 month follow-up period. Katie improved her non-word reading performance by 27% over the intervention period and 7% over the 12 month follow-up period. The findings suggest that an integrated phonological awareness intervention is a successful means of teaching a phonological reading strategy for children with CAS.
Finally, the children’s performance in the standardised connected reading measure revealed that they were able to use their increased decoding skills when reading. Both children demonstrated reading accuracy within the expected range for their age at follow-up assessment, although their reading comprehension performance remained below average. Derek’s persistent comprehension difficulties are likely to be associated with his significant language deficits (he scored below the average range on all three receptive language subtests of the CELF-4 prior to intervention). Consequently, he may have been able to decode accurately, but not understand the content of the story. Katie did not exhibit a notable receptive language deficit at pre-intervention (scoring below the expected range on only one receptive subtest of the CELF-4 at pre-intervention). Katie may have benefited from language-based therapies such as narrative or story structure interventions (Westerveld & Gillon, in press) to help develop her comprehension of connected text.

Overall the findings indicated that an integrated phonological awareness programme is a promising method of producing long-term speech, phonological awareness, and decoding gains in children with CAS. The results add further evidence that speech production goals can be successfully targeted alongside phonological awareness goals within therapy programmes (Gillon, 2000, 2002, 2005).

3.4.1 Limitations and future directions

The generalisation of the findings is limited due to the exploratory nature of the study. It is important that the results are replicated on a larger number of children across a longer intervention period so that the treatment effects can be more carefully evaluated.
The findings are also limited by the confinement of speech measures to one context, namely single word production within the therapy setting. Measures were taken within a single context due to the brevity of the intervention programme and generalization of speech goals was not expected within this timeframe. However, given that children with CAS are expected to make more speech errors in connected speech than in a single word production context (Ozanne, 1995), it is important that future investigations measure the impact of an integrated phonological awareness programme on the spontaneous and connected speech production of children with CAS. It is also critical to extend the evaluation of treatment effects from a reading to spelling context as previous research has indicated that the spelling skills of children with CAS may be more compromised than their word decoding ability (Lewis, Freebairn, Hansen, Iyengar et al., 2004; Stackhouse & Snowling, 1992a). The intervention study reported in Chapter 4 addresses these limitations.
CHAPTER 4

THE EFFECTIVENESS OF AN INTEGRATED PHONOLOGICAL AWARENESS APPROACH FOR CHILDREN WITH CHILDHOOD APRAXIA OF SPEECH

4.1 Introduction

Children with childhood apraxia of speech (CAS) are frequently reported to respond slowly to speech-language therapy intervention. Indeed, slow progress in therapy has been described as a characteristic symptom of this disorder (Ekelman & Aram, 1983; Pannbacker, 1988). It is critical, therefore, that new treatment approaches for children with CAS are investigated using controlled research designs to explore whether these children have potential for more rapid change in speech and language development under differing treatment conditions. The study reported in this chapter examines the effectiveness of a novel integrated phonological awareness intervention, which was previously piloted (see Chapter 3), for 12 children diagnosed with CAS.

More common therapy interventions for children with CAS such as motor based intervention approaches (see Chapter 1 for a review) have a number of limitations including:

1. Failure to facilitate phonological awareness and literacy development;
2. Failure to produce rapid changes in children’s speech production;
3. Failure to consider the multiple levels of breakdown associated with childhood apraxia of speech; and

4. Failure to be based upon controlled intervention studies demonstrating their treatment efficacy.

The few studies that have attempted to demonstrate speech treatment effectiveness for children with CAS have been limited by methodological weaknesses where the description of CAS diagnosis is often not described well enough to allow replication or uncontrolled study designs have been employed (Stackhouse, 1992). This study aims to address previous criticism of intervention designs for children with CAS and to examine treatment effectiveness not only for speech production, but to explore intervention effects on reading and spelling development. The study adopts an integrated model to facilitating phonological awareness, accuracy of speech production, and word decoding ability concurrently.

An integrated phonological awareness intervention is consistent with theoretical perspectives that emphasise phonological representational deficits and motor symptoms in CAS (Marion et al., 1993; Marquardt et al., 2002). The experiment presented in Chapter 2 confirmed a representational component to the disorder, with the children with CAS performing more poorly than their typically developing peers on a receptive measure of phonological representation. The importance of phonological representations for phonological awareness and speech production development has also been highlighted for children with other types of developmental speech disorders (Sutherland & Gillon, 2007, Rvachew, 2006).
Phonological awareness intervention is proposed to increase the accuracy and segmental nature of a child’s underlying phonological representation of words. Preliminary research has examined the impact of including intervention activities that increase the accuracy of underlying phonological representations within speech production intervention. Rvachew, Nowak and Cloutier (2003) compared the benefit of adding weekly 15 minute sessions of either speech perception or vocabulary training to a 16-week therapy programme for children with speech disorder. The speech perception component of the programme involved learning to identify correct and incorrect pronunciations of commonly misarticulated words (thought to promote the accuracy of the phonological representation of the training items) and some phonological awareness tasks (i.e., letter-sound knowledge and onset-rime awareness). Children whose intervention included the speech perception component responded more favourably to the speech intervention, confirming the links between phonological representation and speech production ability. However, the two treatments did not produce a differential effect on phonological awareness abilities. This unexpected result is likely due to the brevity of the phonological awareness component of the experimental intervention, which made up only a portion of the weekly 15-minute sessions of speech perception training over the 16-week intervention. The results highlighted the effects of therapeutic frameworks that include a focus on representational processes on speech production and phonological awareness skills. However, it is also critical to examine the impact of phonological awareness therapy on phonological representation skills.

The current study was designed to extend the findings of the pilot study reported in Chapter 3 with regard to the effectiveness of an integrated phonological awareness
intervention for children with CAS. A controlled single-subject research design and
detailed description of participants with CAS was included to address the methodological
weaknesses commonly found in CAS studies. The impact of the integrated phonological
awareness programme on performance in a phonological representation task was also
explored.

The following hypotheses were tested:

That an integrated phonological awareness intervention will:

1. Improve the accuracy (percent phonemes correct, PPC) of speech production of
   trained and untrained words containing target speech error patterns of children
   with CAS.

2. Improve the accuracy (PPC) of spontaneous production of words containing a
target speech error pattern during connected speech of children with CAS.

3. Increase the phonological awareness of trained and untrained words containing a
target speech error pattern of children with CAS.

4. Increase the letter-sound knowledge of children with CAS; real word and non-
   word decoding and spelling ability of school-aged children with CAS.

5. Improve performance in a phonological representation task of children with CAS.

6. That the CAS group will demonstrate more rapid growth in phonological
   awareness, letter knowledge, spelling, and reading over the intervention period
   than children with typical development not receiving the intervention.
Method

Research design

The study employed a controlled multiple single-subject design with repeated measures to evaluate the effectiveness of the intervention for each participant. A comparative group design (children with CAS compared to children with typical speech and language development) was used to explore whether children with CAS made accelerated growth towards the level of their peers during the intervention period.

Participants

The participants included the same 12 children with CAS and 12 children with typical speech and language development (TD) aged 4 to 7 years described in Chapter 2. Parents of these children provided written permission for the children’s participation in the intervention study as required by the University of Canterbury Human Ethics approval process. The assessment data for the CAS and TD groups can be viewed in Table 11.

Presentation of the detailed speech analysis for the CAS group is presented in Tables 12 to 14.
Table 11. Assessment data for the CAS and TD groups

<table>
<thead>
<tr>
<th></th>
<th>CAS</th>
<th></th>
<th>TD</th>
<th></th>
<th>T</th>
<th>p</th>
<th>Cohen’s d</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>(n = 12)</td>
<td></td>
<td>(n = 12)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Age (months)</td>
<td>Mean 68.5 SD 17.2</td>
<td>Mean 73.4 SD 15.1</td>
<td>T -0.752</td>
<td>p 0.460</td>
<td>Cohen’s d 0.31</td>
<td></td>
<td></td>
</tr>
<tr>
<td>PPVT-III¹</td>
<td>Mean 91.3 SD 6.8</td>
<td>Mean 97.2 SD 8.5</td>
<td>T -1.864</td>
<td>p 0.076</td>
<td>Cohen’s d 0.76</td>
<td></td>
<td></td>
</tr>
<tr>
<td>PCC²</td>
<td>Mean 51.6 SD 23.4</td>
<td>Mean 97.6 SD 1.9</td>
<td>T -7.041</td>
<td>p 0.000**</td>
<td>Cohen’s d 2.87</td>
<td></td>
<td></td>
</tr>
<tr>
<td>PVC³</td>
<td>Mean 83.7 SD 14.7</td>
<td>Mean 99.9 SD 0.2</td>
<td>T -3.793</td>
<td>p 0.003*</td>
<td>Cohen’s d 1.55</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Inconsistency% (DEAP⁴)</td>
<td>Mean 58.9 SD 18.2</td>
<td>Mean 2.0 SD 4.0</td>
<td>T 10.648</td>
<td>p 0.000**</td>
<td>Cohen’s d 4.34</td>
<td></td>
<td></td>
</tr>
<tr>
<td>CELF-4 receptive score⁵</td>
<td>Mean 90.9 SD 13.9</td>
<td>Mean 98.7 SD 5.9</td>
<td>T -1.775</td>
<td>p 0.090</td>
<td>Cohen’s d 0.72</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Note. CAS = childhood apraxia of speech, TD = typically developing, ¹Peabody Picture Vocabulary Test – III, ²Percent consonants correct, ³Percent vowels correct, ⁴DEAP; Diagnostic Evaluation of Articulation and Phonology Speech Inconsistency subtest, ⁵CELF-4 receptive score = Clinical Evaluation of Language Fundamentals receptive standard score (Semel et al., 2003).

* The TD group performance was significantly better than the CAS group (p < .05), **The TD group performance was significantly better than the CAS group (p < .001)
Table 12. Percent of each sound class produced correctly for children with CAS

<table>
<thead>
<tr>
<th>Child’s age</th>
<th>P1</th>
<th>P2</th>
<th>P3</th>
<th>P4</th>
<th>P5</th>
<th>P6</th>
<th>P7</th>
<th>P8</th>
<th>P9</th>
<th>P10</th>
<th>P11</th>
<th>P12</th>
</tr>
</thead>
<tbody>
<tr>
<td>Stops</td>
<td>85.0</td>
<td>72.5</td>
<td>73.2</td>
<td>75.2</td>
<td>64.5</td>
<td>63.6</td>
<td>33.3</td>
<td>93.6</td>
<td>94.4</td>
<td>47.2</td>
<td>22.2</td>
<td>30.0</td>
</tr>
<tr>
<td>Nasals</td>
<td>79.2</td>
<td>83.2</td>
<td>72.9</td>
<td>75.1</td>
<td>90.0</td>
<td>45.5</td>
<td>38.9</td>
<td>84.2</td>
<td>100</td>
<td>68.4</td>
<td>14.3</td>
<td>25.0</td>
</tr>
<tr>
<td>Fricatives</td>
<td>72.5</td>
<td>17.1</td>
<td>34.5</td>
<td>38.7</td>
<td>68.3</td>
<td>75.0</td>
<td>0.0</td>
<td>79.1</td>
<td>41.5</td>
<td>27.3</td>
<td>4.2</td>
<td>28.6</td>
</tr>
<tr>
<td>Affricates</td>
<td>100</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
<td>84.6</td>
<td>28.6</td>
<td>0.0</td>
<td>71.4</td>
<td>92.3</td>
<td>22.3</td>
<td>0.0</td>
<td>0.0</td>
</tr>
<tr>
<td>Glides</td>
<td>57.1</td>
<td>48.4</td>
<td>45.2</td>
<td>47.5</td>
<td>100</td>
<td>40.0</td>
<td>60.0</td>
<td>80.6</td>
<td>66.7</td>
<td>11.2</td>
<td>0.0</td>
<td>0.0</td>
</tr>
<tr>
<td>Liquids</td>
<td>43.5</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
<td>76.2</td>
<td>66.7</td>
<td>13.3</td>
<td>90.0</td>
<td>94.4</td>
<td>42.9</td>
<td>0.0</td>
<td>0.0</td>
</tr>
<tr>
<td>Clusters</td>
<td>24.2</td>
<td>24.1</td>
<td>12.1</td>
<td>13.4</td>
<td>62.1</td>
<td>15.4</td>
<td>0.0</td>
<td>60.0</td>
<td>46.4</td>
<td>23.3</td>
<td>0.0</td>
<td>14.3</td>
</tr>
</tbody>
</table>

Note. CAS = childhood apraxia of speech, age is presented in months, P = participant, analysis from Computerized Profiling (Long & Fey, 2005).

Table 13. The percent consonants correct (PCC) for the early 8 sounds, middle 8 sounds, late 8 sounds, and total consonants for children with CAS.

<table>
<thead>
<tr>
<th></th>
<th>P1</th>
<th>P2</th>
<th>P3</th>
<th>P4</th>
<th>P5</th>
<th>P6</th>
<th>P7</th>
<th>P8</th>
<th>P9</th>
<th>P10</th>
<th>P11</th>
<th>P12</th>
</tr>
</thead>
<tbody>
<tr>
<td>Early  8</td>
<td>77.8</td>
<td>80.8</td>
<td>80.3</td>
<td>83.1</td>
<td>89.3</td>
<td>70.0</td>
<td>52.9</td>
<td>83.8</td>
<td>91.7</td>
<td>72.3</td>
<td>25.0</td>
<td>38.1</td>
</tr>
<tr>
<td>Middle 8</td>
<td>80.4</td>
<td>47.3</td>
<td>48.7</td>
<td>52.0</td>
<td>60.6</td>
<td>47.9</td>
<td>2.3</td>
<td>86.2</td>
<td>85.4</td>
<td>14.8</td>
<td>4.0</td>
<td>17.6</td>
</tr>
<tr>
<td>Late 8</td>
<td>50.6</td>
<td>31.7</td>
<td>25.9</td>
<td>26.9</td>
<td>67.2</td>
<td>57.1</td>
<td>5.0</td>
<td>73.1</td>
<td>46.9</td>
<td>38.1</td>
<td>13.0</td>
<td>18.5</td>
</tr>
<tr>
<td>Total</td>
<td>69.5</td>
<td>54.3</td>
<td>52.5</td>
<td>54.8</td>
<td>71.7</td>
<td>57.5</td>
<td>19.2</td>
<td>81.7</td>
<td>76.4</td>
<td>42.3</td>
<td>13.9</td>
<td>24.6</td>
</tr>
</tbody>
</table>

Note. CAS = childhood apraxia of speech, P = participant, early ‘8’ = early developing sounds, middle ‘8’ = middle developing sounds, late ‘8’ = late developing sounds, total = total percent consonants correct, analysis from Computerized Profiling (Long & Fey, 2005).
Table 14. Percent of substitutions and omissions errors for children with CAS.

<table>
<thead>
<tr>
<th></th>
<th>P1</th>
<th>P2</th>
<th>P3</th>
<th>P4</th>
<th>P5</th>
<th>P6</th>
<th>P7</th>
<th>P8</th>
<th>P9</th>
<th>P10</th>
<th>P11</th>
<th>P12</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sub</td>
<td>75.0</td>
<td>67.0</td>
<td>67.4</td>
<td>69.3</td>
<td>85.0</td>
<td>67.4</td>
<td>38.3</td>
<td>62.2</td>
<td>76.1</td>
<td>71.1</td>
<td>18.2</td>
<td>20.5</td>
</tr>
<tr>
<td>Omission</td>
<td>25.0</td>
<td>33.0</td>
<td>32.6</td>
<td>30.7</td>
<td>15.0</td>
<td>32.6</td>
<td>61.7</td>
<td>37.8</td>
<td>23.9</td>
<td>28.9</td>
<td>81.8</td>
<td>79.5</td>
</tr>
</tbody>
</table>

Note. CAS = childhood apraxia of speech, P = participant, sub = substitution error, omission = omission error, analysis from Computerized Profiling (Long & Fey, 2005).

Procedure

Probes for repeated measures

Repeated assessment measures for speech production and phonological awareness were used to establish a stable baseline for each child prior to the implementation of the intervention. The measures where repeated on three occasions over a two week period. A stable baseline was able to be demonstrated for all participants from these three assessments. The measures were also re-administered on three occasions post-intervention to evaluate intervention effects. Figure 2 depicts these repeated measures in relation to the intervention.

The probes pre- and post-intervention were selected according to each child’s speech PROPH (Long & Fey, 2005) assessment analysis. One phonological error pattern was chosen to be targeted in each intervention block for each child. Selection of each child’s target phonological error patterns was made according to Hodson’s potential targets for therapy in a cycles approach (Hodson & Paden, 1991). The speech probes consisted of 10 trained words and 5 untrained words for each phonological process. Trained words consisted of items containing the target phonological error pattern that
were used as stimuli during intervention sessions. Untrained words consisted of items containing the target phonological process that were not used as stimuli during intervention sessions (i.e., a generalisation measure). The trained and untrained words were similar in phonological structure (i.e., had the same balance of Consonant (C) Vowel (V), CVC or CCVC structures).

Different phoneme awareness tasks (phoneme segmentation and initial phoneme identity) were probed for children aged 4 years and children aged 5 to 7 years to ensure the tasks were developmentally appropriate. The phoneme segmentation probe (for participants aged 5 to 7 years) consisted of 10 trained and 5 untrained items where the child was required to segment the probe word into its constituent phonemes using coloured blocks. For example, the word ‘back’ is segmented into three different phonemes represented by three different block colours. All the stimulus words for this phoneme segmentation task were taken from the child’s target speech production words.

The initial phoneme identity probes (for participants aged 4 years) consisted of seven trained and five untrained words. The child was required to select one out of three words that started with a target sound that corresponded with the child’s target phonological process. An example of one child’s target phonological error patterns and the words used for his trained and untrained speech and phonological awareness probes can be viewed in Appendix B.

Speech probes were recorded via broad phonetic transcription and a percent phonemes correct (PPC) score was calculated for trained and untrained items. A PPC was used instead of a PCC score, as all children had consonant and vowel errors. The number of phonemes produced correctly in each of the 30 trained and 15 untrained
probes pre and post-intervention were also recorded for a paired \textit{t-test} analysis. Distortion errors were not counted as an ‘incorrect’ production. A total PPC was calculated for the trained and untrained phoneme segmentation probes. For example, if the child segmented the word ‘ten’ as /t/ / n/, two out of the three phonemes in the word would be counted as correct. The number of phonemes segmented correctly in each of the 30 trained and 15 untrained probes pre and post-intervention were also recorded for a paired \textit{t-test} analysis. The percentage of correct responses was calculated for trained and untrained phoneme identity probes.

\textbf{Control probe}

The children’s oral narrative performance in the assessment battery (Westerveld \& Gillon, 2002) was also used to select a control probe for the intervention and to monitor the intervention’s effect on the children’s speech accuracy during connected speech. A language error that was inappropriate for the child’s age (e.g., pronoun error, morphological error) was selected as a control probe from the Systematic Analysis of Language Transcripts (SALT) analysis of each child’s personal narrative production measures. The control probe was utilized to ensure that any change in the children’s speech production and phoneme awareness skills could not be attributed to maturation alone. Care was taken to ensure that the language skill selected for the control probe was not due to the child’s speech production errors. For example, if the child had difficulty producing the /s/ sound, then absence of the plural morpheme -s would not be chosen as a control probe. The personal narrative was administered on one occasion pre and post-intervention. Table 15 demonstrates the target phonological error patterns and control probes selected for each child.
The personal narrative production was also used to evaluate the transfer of improved speech production to a connected and spontaneous speaking context. The post-intervention narrative samples were analyzed to detect whether children were suppressing target phonological error patterns. Children were required to produce at least 90% of words containing target sounds correctly to evidence transferral of speech goals to a connected context.
Table 15. Target phonological speech error pattern and control probes for the CAS group.

<table>
<thead>
<tr>
<th>P</th>
<th>Age (months)</th>
<th>Target 1</th>
<th>Target 2</th>
<th>Control language probe</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>87</td>
<td>S-cluster reduction</td>
<td>Tri-cluster reduction</td>
<td>Pronoun ‘we’</td>
</tr>
<tr>
<td>2</td>
<td>64</td>
<td>Velar fronting</td>
<td>S-cluster reduction</td>
<td>Pronoun ‘I’</td>
</tr>
<tr>
<td>3</td>
<td>53</td>
<td>Velar fronting</td>
<td>S-cluster reduction</td>
<td>Pronoun ‘I’</td>
</tr>
<tr>
<td>4</td>
<td>53</td>
<td>Velar fronting</td>
<td>S-cluster reduction</td>
<td>Pronoun ‘I’</td>
</tr>
<tr>
<td>5</td>
<td>90</td>
<td>Velar fronting</td>
<td>Palatal fronting</td>
<td>Past tense</td>
</tr>
<tr>
<td>6</td>
<td>87</td>
<td>Velar fronting</td>
<td>S-cluster reduction</td>
<td>Pronoun ‘I’</td>
</tr>
<tr>
<td>7</td>
<td>51</td>
<td>Final consonant</td>
<td>S-cluster reduction</td>
<td>Pronoun ‘I’ deletion</td>
</tr>
<tr>
<td>8</td>
<td>92</td>
<td>Syllable reduction</td>
<td>S-cluster reduction</td>
<td>Pronoun ‘we’</td>
</tr>
<tr>
<td>9</td>
<td>79</td>
<td>S-cluster reduction</td>
<td>Palatal fronting</td>
<td>Pronoun ‘we’</td>
</tr>
<tr>
<td>10</td>
<td>55</td>
<td>S-cluster reduction</td>
<td>Velar fronting</td>
<td>Pronoun ‘I’</td>
</tr>
<tr>
<td>11</td>
<td>51</td>
<td>Initial bilabials</td>
<td>Final consonant</td>
<td>Pronoun ‘I’ deletion</td>
</tr>
<tr>
<td>12</td>
<td>60</td>
<td>Final consonant</td>
<td>S-cluster reduction</td>
<td>Pronoun ‘I’ deletion</td>
</tr>
</tbody>
</table>

Note. CAS = childhood apraxia of speech, P = participant, target 1 = phonological error pattern targeted in the first intervention block, target 2 = phonological error pattern targeted in the second intervention block.
Pre and post-intervention measures

The following measures were administered pre and post-intervention to the CAS and TD groups. Detailed description of these measures is provided in Chapter 2.

Speech production measures (CAS group only)

- The items from the BBTOP (Bankson & Bernthal, 1990) and first trial of the inconsistency subtest of the DEAP (Dodd et al., 2006) were analysed to gain a PCC and PVC score.
- An inconsistency percentage (DEAP, Dodd et al, 2006) was calculated.

Standardized phonological awareness measures

- The rhyme awareness, alliteration awareness, and phoneme identity subtests of the Preschool and Primary Inventory of Phonological Awareness (PIPA) (Dodd et al., 2000) were administered to children aged 4 years \( (n = 5 \text{ for CAS group, } n = 4 \text{ for TD group}) \). The standard scores from these subtests were combined into a composite score for data analysis.
- The Test of Phonological Awareness (TOPA) (Torgesen & Bryant, 1994) was administered to children aged 5 to 7 years \( (n = 7 \text{ for CAS group, } n = 8 \text{ for TD group}) \). Raw scores were collected for data analysis while standard scores were used to determine performance within/below the expected range.

Letter knowledge measure

- The letter-sound knowledge subtest of the PIPA (Dodd et al., 2000) was administered to all participants. Standard scores were collected for children 6 years and under (to match the normative population) to determine
performance within/below the expected range. Raw scores (out of 30) were collected for all participants for data analysis.

Additionally the following reading and spelling measures were administered to participants aged 6 years and older. As New Zealand children begin formal education at 5 years, all children who participated in the reading and spelling measures had completed at least one year of formal literacy instruction.

**Word decoding measures**

- The Burt Word Reading Test– New Zealand Revision (Gilmore *et al.*, 1981) was used to measure real word decoding. Age equivalence bands were collected from this assessment to denote functioning below/within the expected age range. Raw scores (number of words read correctly) were collected for data analysis.

- An informal non-word reading task (adapted from Calder, 1992) was used to measure non-word decoding (see Appendix A). Children were required to decode three sets of ten non-words of CV, CVC or CCVC structure. The percentage of phonemes correctly decoded for this task was calculated.

**Spelling measure**

- An informal spelling task was used to measure spelling ability. Children were required to spell ten common words from one to three syllables. The items used in the spelling task are taken from the words sampled in the inconsistency subtest of the DEAP (Dodd *et al.*, 2006). Responses were analysed for the number of graphemes represented correctly per attempt and
combined into a percent graphemes correct (PGC) score. For example, if a child spelt the word ‘chips’ as tips, they would be awarded three out of a possible four marks. Responses were also classified according to Ehri’s (2000) classification of spelling stages (i.e., pre-communicative, semi-phonetic, phonetic, and consolidated).

Intervention

The first author instructed three speech-language therapists (SLTs) and two senior student SLTs under clinical supervision in the implementation of the intervention for five of the participants. The first author administered the research intervention to the seven remaining participants. The content and materials used in intervention were standardised across SLTs. All materials and the instruction manual were provided to the SLTs (Gillon & McNeill, 2006). The SLTs and student SLTs watched (and had continued access to) a demonstration video. To ensure treatment fidelity SLTs were required to fill out a session completion worksheet after each session. SLTs were also required to elicit a minimum of 15 elicited productions of any trained speech target words in each activity. Productions were not required to be produced correctly to be counted as elicitations. Sessions were audio or videotaped for later independent analysis of treatment fidelity.

The research intervention had three aims:

1. Reduce target speech error patterns at the single word level and in connected speech
2. Improve phoneme awareness
3. Improve letter-sound knowledge and/or word decoding.
Each child participated in 24 individual 45-minute intervention sessions over 18 weeks. The intervention scheduling was as follows: Intervention block one (12 sessions over 6 weeks, 2 sessions per week), followed by a 6-week withdrawal block without speech-language intervention, followed by a second intervention block (12 sessions over 6 weeks, 2 sessions per week). The timing of the intervention blocks in relation to the pre and post assessment phases is depicted in Figure 2. Intervention sessions were conducted in a quiet room in the child’s home for the five preschool participants and in the child’s school for the other seven participants.

**Figure 2.** Framework of the intervention phases in relation to baseline assessment phases that were implemented for each of participant with CAS.
Structure of the sessions

All sessions included the following types of phonological awareness tasks (and are described in detail in Gillon & McNeill, 2006). Tasks were presented as game activities with colourful pictures and toys. Examples of some materials that were used in the phonological awareness activities can be viewed in Appendix C. Examples of the activities are provided as follows:

- **Letter-sound knowledge example:**
  
  SLT sets up three posters displaying a target letter (m, t, s).
  
  SLT: “Drive your motorbike to the letter ‘c’ that makes a /k/ sound. Great work! You have found the letter ‘c’ that makes a /k/ sound. Let’s say the sound together” (pointing to the letter).
  
  Child and SLT: “/k/”

- **Phoneme identity example:**
  
  SLT: “Let’s find all the toys that start with a /b/ sound”
  
  Child finds a ball.
  
  SLT: “Well done. You found a ball that starts with a /b/ sound.” (SLT then introduces a picture card of a ball with the word ball written underneath in large clear font). Look at the word ball. It starts with a /b/ sound (pointing to the letter ‘b’ at the start of the word ball). “Let’s read the word together before we put the card in our /b/ toy box. It says….” (prompting for the target word ball).
  
  Child: “all”
SLT: When you say ‘all, I can’t hear the /b/ sound at the start’ (pointing to the letter ‘b’). “Try and say ‘ball’ again with a ‘b’ sound at the start”.

Child: “ball”

- Segmentation and blending example:
  - Onset-rime level for 4 year old participants
  - Phoneme level for 5 to 7 year old participants

Phoneme level example: SLT and child played bingo together using bingo boards and bingo cards prepared for the child’s target speech process. For example, if the target speech process was the correct production of s clusters, the “s cluster bingo board” which had six pictures to represent the following words: star, stop, spoon, spit, slot, slap was used. The child was required to pick a picture card from a card pile which contained the six target speech words plus distracter items. Each time a new word was picked up from the card pile the child was required to segment the word into phonemes (e.g., /s/-/t/-/a/ = 3 phonemes) and then articulate the word together (i.e., speech practice). The SLT modelled and prompted for the correct production of the s cluster as necessary. The child then placed the picture card on his/her board if it matched a picture card on the bingo board.

- Phoneme manipulation with letter (grapheme) blocks

Blocks (each representing one grapheme) are used to track sound changes in words.

SLT: “If this says stop, show me step”.

Child: Exchanges the ‘o’ letter block for the ‘e’ letter block.
SLT: “And it says…”

Child: “Step”.

SLT: “Great talking”.

The words used in the phonological awareness activities were the children’s trained speech probe words. For example, if the child was working on suppressing the s-cluster reduction error pattern, he/she would be required to segment words that contained an s-cluster. Targeted speech production practice was required in all activities. SLTs were required to elicit a minimum of 15 (correct or incorrect) productions of target speech words in each activity. If a speech production error occurred in the activities, the SLT assisted the child to identify the error and then utilized a letter block as a prompt for speech production.

**Reliability and treatment fidelity**

**Assessment data**

In addition to the first author reviewing the online transcription completed by the SLTs, transcription reliability was determined for a second independent transcription conducted by an experienced SLT for five participants (just over 20% of participants). Mean inter-rater agreement (based on the percentage of phonemes transcribed identically) was 92.31% with a range of 79.8% to 100.0%.

**Speech probes**

Twenty percent of the baseline and post-intervention speech probes were randomly chosen for re-transcription by an independent SLT. Analysis showed 86.21% inter-rater agreement for baseline repeated measures and 89.87% agreement for post-
intervention repeated measures (based on the percentage of phonemes transcribed identically).

**Phoneme awareness probes**

Twenty percent of the baseline and post-intervention phoneme segmentation probes were randomly chosen for re-marking by an independent reviewer. Analysis showed 93.5% inter-rater agreement for baseline repeated measures and 95.3% agreement for post-intervention repeated measures (based on the percentage of responses marked identically as ‘correct’ or ‘incorrect’ by the examiners).

**Non-word reading data**

An independent reviewer analysed and scored the non-word reading responses of five participants (just over 20% of participants). Mean inter-rater agreement (based on the percentage of sounds transcribed identically) was 94.7% with a range of 88.2% to 100%.

**Spelling data**

An independent reviewer analysed and scored the pre and post-intervention spelling responses of five children (nearly 40% of the children that completed the spelling task). Mean point-by-point inter-rater agreement for analysis of spelling stage was 96.0% with a range of 95.0% to 100.0%. Mean inter-rater agreement for GPC was 93.2% with a range of 88.4% to 100.0%.

**Treatment fidelity**

Twenty-nine sessions, (just over 20% of the total sessions), were randomly chosen for evaluation of treatment fidelity. An independent SLT was familiarized with the intervention activities. The SLT then listened to the audiotape and recorded the presence
or absence of the following important treatment components: letter-sound knowledge, phoneme identity, segmentation, blending, and manipulation activities. A minimum of 15 elicited productions of trained words were required in each activity. The sessions were required to exclude words from the participant’s ‘untrained’ items. Cues were required to be given when the child produced a speech production error of a trained word. The majority of cues for speech production were required to be limited to those giving information about the phonological structure of the word or those helping the child to use graphemes to direct speech production. Analysis showed that adherence to the above treatment fidelity measures was 96.6% in the sampled sessions.

Results

Repeated measures

The two standard deviation band method was used to identify if variation between baseline and post-intervention phases were indicative of significant improvement across the phases (Portney & Watkins, 2000). This method involves calculating the mean and standard deviation of the baseline phase. The mean and two standard deviations above and below the mean of the baseline phase are then plotted along the baseline and post-intervention phases. If the data points in the intervention phase fall outside the banded area, the change in performance is considered significant (Portney & Watkins, 2000).

Paired t-tests were also used to evaluate change in the speech and phonological probes over the intervention period. As parametric statistics are contraindicated in single-subject research designs when data is serially dependent (Brossart, Parker, Olson, & Lakshmi, 2006; Kazdin, 1982), an autocorrelation analysis was performed on all data to ensure the test was appropriate. A paired t-test was not performed on comparisons that
included serially dependent data. All data points pre and post-intervention were compared for the paired \textit{t-test} (rather than being cumulated into 3 data points pre and post-intervention as used in the two standard deviation band method).

**Speech production**

An example of the graphs used to analyse the trained and untrained speech production gains of one child (Participant 9) via the two standard deviation band method is presented in Figures 3 to 6. The individual graphs demonstrating speech production changes in trained and untrained items for all participants can be viewed in Appendix D. However, given the number of graphs needed for this analysis (four per child), the results for the trained and untrained speech production probes of all participants are summarised in Table 16. The findings show that 7 of the 12 children showed significant improvement in trained and untrained speech probes for both target phonological processes. A further three children demonstrated improvement in two trained and one untrained speech probe. One child demonstrated improvement in the trained and untrained production of one phonological process only. One child did not improve speech production in any of the trained and untrained measures.

The number of children that transferred speech production gains to the connected speaking context immediately post-intervention is also presented in Table 16.
**Figure 3.** Pre and post-intervention performance for trained speech probes during block one (s-cluster reduction) for participant 9.

Post-intervention performance must be above the two standard deviation band to denote significant improvement.
Figure 4. Pre and post-intervention performance for untrained speech probes during block one (s-cluster reduction) for participant 9.

Post-intervention performance must be above the two standard deviation band to denote significant improvement (in this example, the standard deviation of the pre-intervention probes was zero).
Figure 5. Pre and post-intervention performance for trained speech probes during block two (palatal fronting) for participant 9.

Post-intervention performance must be above the two standard deviation band to denote significant improvement.
Figure 6. Pre and post-intervention performance for untrained speech probes during block two (palatal fronting) for participant 9.

Post-intervention performance must be above the two standard deviation band to denote significant improvement.
Table 16. Change in speech production probes for all participants

<table>
<thead>
<tr>
<th>P</th>
<th>Pre</th>
<th>Est 2SD</th>
<th>Post</th>
<th>T</th>
<th>p</th>
<th>d</th>
<th>CS?</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>P1: T1</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>U1</td>
<td>61.2 (3.0)</td>
<td>55.2 – 67.2</td>
<td>82.5 (8.0)</td>
<td>-2.10</td>
<td>0.054</td>
<td>0.76</td>
<td></td>
</tr>
<tr>
<td>T2</td>
<td>60.4 (0.0)</td>
<td>60.4 – 60.4</td>
<td>90.3 (3.2)</td>
<td>-6.50</td>
<td>0.00**</td>
<td>1.68</td>
<td>No</td>
</tr>
<tr>
<td>U2</td>
<td>54.2 (0.0)</td>
<td>54.2 – 54.2</td>
<td>93.1 (2.4)</td>
<td>-5.55</td>
<td>0.00**</td>
<td>2.02</td>
<td></td>
</tr>
<tr>
<td><strong>P2: T1</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>U1</td>
<td>56.2 (8.0)</td>
<td>40.2 – 72.2</td>
<td>86.0 (3.1)</td>
<td>na</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>T2</td>
<td>50.0 (2.6)</td>
<td>44.8 – 55.2</td>
<td>92.1 (2.6)</td>
<td>-13.0</td>
<td>0.00**</td>
<td>3.35</td>
<td>Yes</td>
</tr>
<tr>
<td>U2</td>
<td>40.0 (3.1)</td>
<td>33.8 – 46.2</td>
<td>89.5 (0.0)</td>
<td>-14.0</td>
<td>0.00**</td>
<td>5.12</td>
<td></td>
</tr>
<tr>
<td><strong>P3: T1</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>U1</td>
<td>50.0 (7.1)</td>
<td>35.8 – 64.2</td>
<td>97.6 (4.1)</td>
<td>-1.79</td>
<td>0.00**</td>
<td>2.31</td>
<td></td>
</tr>
<tr>
<td>T2</td>
<td>59.6 (1.5)</td>
<td>56.6 – 62.6</td>
<td>91.2 (1.5)</td>
<td>na</td>
<td></td>
<td></td>
<td>No</td>
</tr>
<tr>
<td>U2</td>
<td>61.4 (6.1)</td>
<td>49.2 – 73.6</td>
<td>96.5 (6.1)</td>
<td>-7.14</td>
<td>0.00**</td>
<td>2.60</td>
<td></td>
</tr>
<tr>
<td><strong>P4: T1</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>U1</td>
<td>60.0 (0.0)</td>
<td>60.0 – 60.0</td>
<td>91.0 (3.5)</td>
<td>-14.0</td>
<td>0.00**</td>
<td>5.11</td>
<td></td>
</tr>
<tr>
<td>T2</td>
<td>53.0 (5.3)</td>
<td>42.4 – 63.6</td>
<td>94.0 (1.7)</td>
<td>-10.6</td>
<td>0.00**</td>
<td>3.48</td>
<td>No</td>
</tr>
<tr>
<td>U2</td>
<td>54.3 (6.4)</td>
<td>41.6 – 67.0</td>
<td>91.0 (8.2)</td>
<td>-7.4</td>
<td>0.00**</td>
<td>2.73</td>
<td></td>
</tr>
<tr>
<td><strong>P5: T1</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>U1</td>
<td>61.7 (4.0)</td>
<td>53.6 – 61.7</td>
<td>97.7 (4.0)</td>
<td>-10.2</td>
<td>0.00**</td>
<td>3.75</td>
<td></td>
</tr>
<tr>
<td>T2</td>
<td>69.1 (0.0)</td>
<td>69.1 – 69.1</td>
<td>95.7 (5.1)</td>
<td>-7.4</td>
<td>0.00**</td>
<td>1.91</td>
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<td>10.0 – 50.0</td>
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<td>64.5 (3.9)</td>
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<td>16.6 – 72.2</td>
<td>44.4 (7.7)</td>
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<td>47.8 – 69.8</td>
<td>79.8 (1.6)*</td>
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**Note.** Pre and post scores are the mean and standard deviation of the three baseline and post-intervention phases, P = Participant; Est = Estimated, 2SD = two standard deviation band method, *\(^1\) = significant change via 2 standard deviation band method (must be above estimated ranges for significance), CS = transfer to connected speech; T = trained probe, U = Untrained probe, * = significant change \((p < .05)\), ** = significant change \((p < .001)\), na = *t-test* not used due to serially dependency of data, scores are percent phonemes correct.

**Phonological awareness**

An example of the graphs used to analyse the trained and untrained phonological awareness gains of one child (Participant 9, P9) via the two standard deviation band method is presented in Figures 7 to 10. The results for trained and untrained phonological awareness probes for all children are summarised in Table 17. The results show that 8 out of the 12 children improved in both their trained and untrained phonological awareness probes for both targets (participants 2, 3, 4, 5, 6, 8, 9, and 12). One child
improved in one trained phonological awareness probe alone (participant 7). Three children did not improve their phonological awareness scores (participants 1, 10, and 11). The individual graphs demonstrating change in phoneme awareness over the intervention period can be viewed in Appendix E.

**Figure 7.** Pre and post-intervention performance for trained phoneme segmentation probes during block one (s-cluster reduction) for participant 9.

Post-intervention performance must be above the two standard deviation band to denote significant improvement (in this example, the standard deviation of the pre-intervention probes was zero).
Figure 8. Pre and post-intervention performance for untrained phoneme segmentation probes during block one (s-cluster reduction) for participant 9.

Post-intervention performance must be above the two standard deviation band to denote significant improvement (in this example, the standard deviation of the pre-intervention probes was zero).
Figure 9. Pre and post-intervention performance for trained phoneme segmentation probes during block two (palatal fronting) for participant 9.

Post-intervention performance must be above the two standard deviation band to denote significant improvement (in this example, the standard deviation of the pre-intervention probes was zero).
Figure 10. Pre and post-intervention performance for untrained phoneme segmentation probes during block two (palatal fronting) for participant 9. Post-intervention performance must be above the two standard deviation band to denote significant improvement (in this example, the standard deviation of the pre-intervention probes was zero).
Table 17. Change in phonological awareness probes for all participants

<table>
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<tr>
<th>Participant</th>
<th>Pre</th>
<th>Est 2SD</th>
<th>Post</th>
<th>T</th>
<th>p</th>
<th>d</th>
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<td>P1: Trained 1</td>
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<td>73.7 (7.0)</td>
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<tr>
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<td>61.4 (16.0)</td>
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<td>51.7 – 102.5</td>
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<td>-1.5</td>
<td>0.15</td>
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<td>68.0 – 68.0</td>
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<td>-1.2</td>
<td>0.25</td>
<td>0.44</td>
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<td>10.5 – 10.5</td>
<td>88.6 (1.6)*</td>
<td>-17.0</td>
<td>0.00**</td>
<td>4.40</td>
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<tr>
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<td>7.0 (2.9)</td>
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<td>13.3 (11.5)</td>
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<td>100.0 (0.0)*</td>
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<td>Untrained 2</td>
<td>P12: Trained 1</td>
<td>Untrained 1</td>
<td>Trained 2</td>
</tr>
<tr>
<td>---------------</td>
<td>-------------</td>
<td>-----------</td>
<td>-------------</td>
<td>---------------</td>
<td>-------------</td>
<td>-----------</td>
</tr>
<tr>
<td>28.6 (0.0)</td>
<td>40.0 (0.0)</td>
<td>33.6 (8.1)</td>
<td>13.3 (11.5)</td>
<td>25.5 (4.0)</td>
<td>31.1 (3.8)</td>
<td>30.7 (4.0)</td>
</tr>
<tr>
<td>28.6 – 28.6</td>
<td>40.0 – 40.0</td>
<td>17.4 – 49.8</td>
<td>17.5 – 33.5</td>
<td>17.5 – 33.5</td>
<td>23.5 – 38.7</td>
<td>22.7 – 38.7</td>
</tr>
<tr>
<td>57.1 (14.3)*</td>
<td>80.0 (20.0)*</td>
<td>33.6 (8.1)</td>
<td>46.7 (11.5)*</td>
<td>63.2 (2.7)*</td>
<td>82.2 (15.4)*</td>
<td>57.9 (2.6)*</td>
</tr>
<tr>
<td>-1.7</td>
<td>-1.5</td>
<td>0.00</td>
<td>-1.8</td>
<td>-8.5</td>
<td>-8.0</td>
<td>-2.4</td>
</tr>
<tr>
<td>0.11</td>
<td>0.27</td>
<td>1.00</td>
<td>0.10</td>
<td>0.00**</td>
<td>0.00**</td>
<td>0.03*</td>
</tr>
<tr>
<td>0.51</td>
<td>0.42</td>
<td>0.00</td>
<td>0.65</td>
<td>2.19</td>
<td>2.91</td>
<td>1.91</td>
</tr>
</tbody>
</table>

**Note.** Pre and post scores are the mean and standard deviation of the three baseline and post-intervention phases, Est = Estimated, 2SD = two standard deviation band method, *\(^1\) = significant change via 2 standard deviation band method (must be above estimated ranges for significance), * = significant change in paired t-test \((p < .05)\), ** = significant change in paired t-test \((p < .001)\), na = paired t-test not used due to serially dependency of data.

**Pre and post measures**

A paired t-test was used to evaluate change over the intervention period in the pre and post measures for the CAS group. The results which are presented in Table 18 indicate that the CAS group made a significant improvement in all the pre-post measures except for the Burt reading test over the intervention period. There was, however, a large effect size for the Burt reading measure indicating a clinically significant effect.
Table 18. Change in pre and post measures for the CAS group

<table>
<thead>
<tr>
<th></th>
<th>Pre Mean (SD)</th>
<th>Post Mean (SD)</th>
<th>T</th>
<th>p</th>
<th>d</th>
</tr>
</thead>
<tbody>
<tr>
<td>PCC (n = 12)</td>
<td>51.6 (22.5)</td>
<td>64.0 (20.5)</td>
<td>-8.1</td>
<td>0.000**</td>
<td>2.33</td>
</tr>
<tr>
<td>PVC (n = 12)</td>
<td>83.7 (14.9)</td>
<td>92.9 (9.4)</td>
<td>-2.4</td>
<td>0.0340*</td>
<td>0.70</td>
</tr>
<tr>
<td>Inconsistency% (n = 12)</td>
<td>58.7 (18.1)</td>
<td>43.0 (21.8)</td>
<td>4.0</td>
<td>0.002*</td>
<td>1.16</td>
</tr>
<tr>
<td>TOPA (n = 7)</td>
<td>6.3 (3.1)</td>
<td>11.1 (2.6)</td>
<td>-3.8</td>
<td>0.009*</td>
<td>2.0</td>
</tr>
<tr>
<td>LK (n = 12)</td>
<td>8.3 (9.6)</td>
<td>16.8 (9.6)</td>
<td>-4.9</td>
<td>0.000**</td>
<td>1.41</td>
</tr>
<tr>
<td>Burt Reading (n = 6)</td>
<td>14.2 (10.8)</td>
<td>23.0 (7.8)</td>
<td>-2.2</td>
<td>0.078</td>
<td>0.90</td>
</tr>
<tr>
<td>NWR (n = 6)</td>
<td>8.9 (15.0)</td>
<td>49.3 (19.5)</td>
<td>-8.1</td>
<td>0.000**</td>
<td>3.30</td>
</tr>
<tr>
<td>Spell (PGC) (n = 5)</td>
<td>20.1 (12.8)</td>
<td>51.7 (9.2)</td>
<td>-5.0</td>
<td>0.007*</td>
<td>2.25</td>
</tr>
<tr>
<td>PhonRep (n = 12)</td>
<td>16.7 (4.8)</td>
<td>20.0 (5.3)</td>
<td>-2.7</td>
<td>0.021*</td>
<td>0.78</td>
</tr>
</tbody>
</table>

*Note.* CAS = childhood apraxia of speech, TOPA = Test of Phonological Awareness (Torgesen & Bryant, 1994), LK = letter-sound knowledge, NWR = non-word reading, PGC = percent graphemes correct, PhonRep = phonological representation task.

** = significant difference (p < .001); * = significant difference (p < .05)

Individual Descriptive Analysis

Descriptive analysis at an individual level was undertaken to examine any relationships between phonological representation and performance in phoneme awareness and speech probes, non-word reading, and spelling performance. The data revealed that seven children (participants 2, 3, 4, 5, 8, 9, and 12) showed gains in all speech probes, all phoneme awareness probes, and phonological representation ability.
The school-aged children in this group (i.e., participants 2, 5, 8, and 9) also demonstrated large gains in non-word reading and spelling performance. Participant 1 and Participant 11 demonstrated inconsistent (i.e. not all probes showed a significant increase) or absent gains in speech and phoneme awareness probes and no improvement on the phonological representation task. The school-aged child in this group (participant 1) demonstrated gains in non-word reading (from 0% to 29% phonemes correct) and spelling performance (from 10% to 34% graphemes correct). Participant 6, who made speech gains in the first target phonological error pattern only, made overall gains in phoneme awareness probes and no gain in the phonological representation task. This participant showed improvement in non-word reading (from 3% to 29% phonemes correct) and minimal gain in spelling (from 35% to 42% graphemes correct). Participant 7 made overall improvements in speech probes but no improvement in phoneme awareness probes or in the phonological representation task. Participant 10 made speech and phonological representation gains without gains in phonological awareness ability. However, closer inspection of the data show that this child was performing at near-ceiling on phoneme awareness probes at pre-intervention making it difficult to demonstrate gains in the phoneme awareness probes.

**Comparison to children with typical development**

A *t-test* with Bonferroni correction was used to compare relative changes over the intervention period in the pre and post measures for the CAS and TD groups. To control for initial differences in scores between the groups, proportional change scores (calculated as time2 - time1 / time1) were compared. Results, shown in Table 19, indicate that the CAS group experienced accelerated growth in phonological awareness,
letter knowledge, non-word reading and spelling measures compared to the TD group over the intervention period.

### Table 19. Relative change in the pre-post measures for CAS and TD groups

<table>
<thead>
<tr>
<th></th>
<th>CAS Mean</th>
<th>SD Mean</th>
<th>TD Mean</th>
<th>SD Mean</th>
<th>T</th>
<th>p</th>
<th>d</th>
</tr>
</thead>
<tbody>
<tr>
<td>TOPA</td>
<td>1.14</td>
<td>(1.08)</td>
<td>0.09</td>
<td>(0.07)</td>
<td>2.78</td>
<td>0.016*</td>
<td>1.44</td>
</tr>
<tr>
<td>Letter</td>
<td>3.79</td>
<td>(3.41)</td>
<td>0.59</td>
<td>(0.94)</td>
<td>3.14</td>
<td>0.028*</td>
<td>1.28</td>
</tr>
<tr>
<td>Burt</td>
<td>2.11</td>
<td>(3.51)</td>
<td>0.07</td>
<td>(0.07)</td>
<td>1.67</td>
<td>0.725</td>
<td>0.68</td>
</tr>
<tr>
<td>NWR</td>
<td>24.75</td>
<td>(24.54)</td>
<td>0.14</td>
<td>(0.10)</td>
<td>2.88</td>
<td>0.014*</td>
<td>1.18</td>
</tr>
<tr>
<td>Spelling</td>
<td>2.52</td>
<td>(1.90)</td>
<td>0.24</td>
<td>(0.36)</td>
<td>3.39</td>
<td>0.036*</td>
<td>1.38</td>
</tr>
<tr>
<td>PhonRep</td>
<td>0.24</td>
<td>(0.30)</td>
<td>0.04</td>
<td>(0.10)</td>
<td>2.23</td>
<td>0.218</td>
<td>0.91</td>
</tr>
</tbody>
</table>

**Note.** TOPA = Test of Phonological Awareness; NWR = non-word reading task; PhonRep = phonological representation task.

* = significant difference (p < .05)

#### Spelling performance

The spelling of the CAS group showed a marked improvement from pre to post-intervention. Figures 11 to 15 show the spelling performance of the five children from the CAS group who completed the test per and post-intervention. These spelling words were not directly targeted in therapy. Comparison of the children’s spelling attempts to Ehri’s (2000) stage model of spelling development showed that the majority of children progressed in the sophistication of the principal spelling strategy employed. Participant 1
Participant 5 (Figure 12) shifted from predominantly semi-phonetic spelling attempts at pre-intervention to predominantly phonetic spelling attempts at post-intervention. Participant 6 (Figure 13) did not show steady progression in spelling development, and included predominantly semi-phonetic and phonetic spelling attempts at pre and post-intervention. Participant 8 (Figure 14) shifted from predominantly pre-communicative and semi-phonetic spelling attempts at pre-intervention to predominantly phonetic spelling attempts at post-intervention. Participant 9 (Figure 15) shifted from predominantly pre-communicative and semi-phonetic spelling attempts at pre-intervention to predominantly phonetic spelling attempts at post-intervention.

Figures 16 and 17 show the spelling performance of two children from the TD group over the intervention period. The child displayed in Figure 16 was at the consolidated stage of spelling development at both assessment points. The child displayed in Figure 17 was primarily at the phonetic stage of spelling development at both assessment points. These figures demonstrate the severe spelling deficit that the children with CAS presented with in comparison to their peers with typical speech and language development. Further, the figures demonstrate the large qualitative gains made by the children with CAS in comparison to their typically developing peers over the intervention period.
<table>
<thead>
<tr>
<th>Target</th>
<th>Pre (age 7;3)</th>
<th>Spelling stage</th>
<th>Post (aged 7;8)</th>
<th>Spelling stage</th>
</tr>
</thead>
<tbody>
<tr>
<td>Rain</td>
<td>Phonetic</td>
<td>ran</td>
<td>Phonetic</td>
<td></td>
</tr>
<tr>
<td>Kangaroo</td>
<td>Pre-communicative</td>
<td>karo</td>
<td>Semi-phonetic</td>
<td></td>
</tr>
<tr>
<td>Girl</td>
<td>Pre-communicative</td>
<td>gir</td>
<td>Semi-phonetic</td>
<td></td>
</tr>
<tr>
<td>Shark</td>
<td>Pre-communicative</td>
<td>hac</td>
<td>Phonetic</td>
<td></td>
</tr>
<tr>
<td>Dinosaur</td>
<td>Pre-communicative</td>
<td>din</td>
<td>Semi-phonetic</td>
<td></td>
</tr>
<tr>
<td>Teeth</td>
<td>Phonetic</td>
<td>df</td>
<td>Phonetic</td>
<td></td>
</tr>
<tr>
<td>Fish</td>
<td>Semi-phonetic</td>
<td>Fraser</td>
<td>Semi-phonetic</td>
<td></td>
</tr>
<tr>
<td>Chips</td>
<td>Pre-communicative</td>
<td>hir</td>
<td>Pre-communicative</td>
<td></td>
</tr>
<tr>
<td>Bridge</td>
<td>Pre-communicative</td>
<td>brin</td>
<td>Semi-phonetic</td>
<td></td>
</tr>
<tr>
<td>Cake</td>
<td>Pre-communicative</td>
<td>kwait</td>
<td>Phonetic</td>
<td></td>
</tr>
</tbody>
</table>

**Figure 11.** Spelling performance at pre-intervention and post-intervention for participant 1. Spelling items were not directly trained in therapy.
<table>
<thead>
<tr>
<th>Target</th>
<th>Pre (aged 7;6)</th>
<th>Spelling Stage</th>
<th>Post (aged 7;11)</th>
<th>Spelling Stage</th>
</tr>
</thead>
<tbody>
<tr>
<td>Rain</td>
<td>ran</td>
<td>Phonetic</td>
<td>ran</td>
<td>Phonetic</td>
</tr>
<tr>
<td>Kangaroo</td>
<td>semi-phonetic</td>
<td>semi-phonetic</td>
<td>Kangaroo</td>
<td>semi-phonetic</td>
</tr>
<tr>
<td>Girl</td>
<td>semi-phonetic</td>
<td>semi-phonetic</td>
<td>gorl</td>
<td>Phonetic</td>
</tr>
<tr>
<td>Shark</td>
<td>semi-phonetic</td>
<td>semi-phonetic</td>
<td>Shark</td>
<td>consolidated</td>
</tr>
<tr>
<td>Dinosaur</td>
<td>semi-phonetic</td>
<td>semi-phonetic</td>
<td>dinosaur</td>
<td>Phonetic</td>
</tr>
<tr>
<td>Teeth</td>
<td>semi-phonetic</td>
<td>semi-phonetic</td>
<td>Tof</td>
<td>Phonetic</td>
</tr>
<tr>
<td>Fish</td>
<td>phonetic</td>
<td>phonetic</td>
<td>Fish</td>
<td>consolidated</td>
</tr>
<tr>
<td>Chips</td>
<td>semi-phonetic</td>
<td>semi-phonetic</td>
<td>Chish</td>
<td>semi-phonetic</td>
</tr>
<tr>
<td>Bridge</td>
<td>semi-phonetic</td>
<td>semi-phonetic</td>
<td>Bridge</td>
<td>Phonetic</td>
</tr>
<tr>
<td>Cake</td>
<td>semi-phonetic</td>
<td>semi-phonetic</td>
<td>cake</td>
<td>Phonetic</td>
</tr>
</tbody>
</table>

**Figure 12.** Spelling performance at pre-intervention and post-intervention for participant 5. Spelling items were not directly trained in therapy.
<table>
<thead>
<tr>
<th>Target</th>
<th>Pre</th>
<th>Spelling Stage</th>
<th>Post</th>
<th>Spelling Stage</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>(aged 7;3)</td>
<td></td>
<td>(aged 7;8)</td>
<td></td>
</tr>
<tr>
<td>Rain</td>
<td>![Rain]</td>
<td>Phonetic</td>
<td>![Rain]</td>
<td>Consolidated</td>
</tr>
<tr>
<td>Kangaroo</td>
<td>![Kangaroo]</td>
<td>Semi-phonetic</td>
<td>![Kangaroo]</td>
<td>Pre-communicative</td>
</tr>
<tr>
<td>Girl</td>
<td>![Girl]</td>
<td>Semi-phonetic</td>
<td>![Girl]</td>
<td>Semi-phonetic</td>
</tr>
<tr>
<td>Shark</td>
<td>![Shark]</td>
<td>Phonetic</td>
<td>![Shark]</td>
<td>Phonetic</td>
</tr>
<tr>
<td>Dinosaur</td>
<td>![Dinosaur]</td>
<td>Pre-communicative</td>
<td>![Dinosaur]</td>
<td>Semi-phonetic</td>
</tr>
<tr>
<td>Teeth</td>
<td>![Teeth]</td>
<td>Pre-communicative</td>
<td>![Teeth]</td>
<td>Consolidated</td>
</tr>
<tr>
<td>Fish</td>
<td>![Fish]</td>
<td>Consolidated</td>
<td>![Fish]</td>
<td>Phonetic</td>
</tr>
<tr>
<td>Chips</td>
<td>![Chips]</td>
<td>Pre-communicative</td>
<td>![Chips]</td>
<td>Pre-communicative</td>
</tr>
<tr>
<td>Bridge</td>
<td>![Bridge]</td>
<td>Pre-communicative</td>
<td>![Bridge]</td>
<td>Pre-communicative</td>
</tr>
<tr>
<td>Cake</td>
<td>![Cake]</td>
<td>Semi-phonetic</td>
<td>![Cake]</td>
<td>Consolidated</td>
</tr>
</tbody>
</table>

**Figure 13.** Spelling performance at pre-intervention and post-intervention for participant 6. Spelling items were not directly trained in therapy.
<table>
<thead>
<tr>
<th>Target</th>
<th>Pre Spelling stage</th>
<th>Post Spelling stage</th>
</tr>
</thead>
<tbody>
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<td>Rain</td>
<td>Semi-phonetic</td>
<td>Phonetic</td>
</tr>
<tr>
<td>Kangaroo</td>
<td>Pre-communicative</td>
<td>Semi-phonetic</td>
</tr>
<tr>
<td>Girl</td>
<td>Pre-communicative</td>
<td>Phonetic</td>
</tr>
<tr>
<td>Shark</td>
<td>Semi-phonetic</td>
<td>Phonetic</td>
</tr>
<tr>
<td>Dinosaur</td>
<td>Pre-communicative</td>
<td>Semi-phonetic</td>
</tr>
<tr>
<td>Teeth</td>
<td>Semi-phonetic</td>
<td>Phonetic</td>
</tr>
<tr>
<td>Fish</td>
<td>Semi-phonetic</td>
<td>Consolidated</td>
</tr>
<tr>
<td>Chips</td>
<td>Pre-communicative</td>
<td>Consolidated</td>
</tr>
<tr>
<td>Bridge</td>
<td>Pre-communicative</td>
<td>Semi-phonetic</td>
</tr>
<tr>
<td>Cake</td>
<td>Semi-phonetic</td>
<td>Phonetic</td>
</tr>
</tbody>
</table>

Figure 14. Spelling performance at pre-intervention and post-intervention for participant 8. Spelling items were not directly trained in therapy.
<table>
<thead>
<tr>
<th>Target</th>
<th>Pre (age 6;7)</th>
<th>Spelling stage</th>
<th>Post (aged 6;11)</th>
<th>Spelling stage</th>
</tr>
</thead>
<tbody>
<tr>
<td>Rain</td>
<td></td>
<td>Semi-phonetic</td>
<td></td>
<td>Phonetic</td>
</tr>
<tr>
<td>Kangaroo</td>
<td></td>
<td>Semi-phonetic</td>
<td></td>
<td>Phonetic</td>
</tr>
<tr>
<td>Girl</td>
<td></td>
<td>Pre-communicative</td>
<td></td>
<td>Phonetic</td>
</tr>
<tr>
<td>Shark</td>
<td></td>
<td>Pre-communicative</td>
<td></td>
<td>Phonetic</td>
</tr>
<tr>
<td>Dinosaur</td>
<td></td>
<td>Semi-phonetic</td>
<td></td>
<td>Phonetic</td>
</tr>
<tr>
<td>Teeth</td>
<td></td>
<td>Semi-phonetic</td>
<td></td>
<td>Phonetic</td>
</tr>
<tr>
<td>Fish</td>
<td></td>
<td>Semi-phonetic</td>
<td></td>
<td>Consolidated</td>
</tr>
<tr>
<td>Chips</td>
<td></td>
<td>Pre-communicative</td>
<td></td>
<td>Semi-phonetic</td>
</tr>
<tr>
<td>Bridge</td>
<td></td>
<td>Pre-communicative</td>
<td></td>
<td>Semi-phonetic</td>
</tr>
<tr>
<td>Cake</td>
<td></td>
<td>Pre-communicative</td>
<td></td>
<td>Phonetic</td>
</tr>
</tbody>
</table>

**Figure 15.** Spelling performance at pre-intervention and post-intervention for participant 9. Spelling items were not directly trained in therapy.
<table>
<thead>
<tr>
<th>Target</th>
<th>Pre</th>
<th>Spelling</th>
<th>Post</th>
<th>Spelling</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>(aged 7;3)</td>
<td>Stage</td>
<td>(aged 7;8)</td>
<td>Stage</td>
</tr>
<tr>
<td>Rain</td>
<td></td>
<td>Consolidated</td>
<td>Rain</td>
<td>Consolidated</td>
</tr>
<tr>
<td>Kangaroo</td>
<td></td>
<td>Consolidated</td>
<td>Kangaroo</td>
<td>Phonetic</td>
</tr>
<tr>
<td>Girl</td>
<td></td>
<td>Consolidated</td>
<td>Girl</td>
<td>Consolidated</td>
</tr>
<tr>
<td>Shark</td>
<td></td>
<td>Consolidated</td>
<td>Shark</td>
<td>Consolidated</td>
</tr>
<tr>
<td>Dinosaur</td>
<td></td>
<td>Phonetic</td>
<td>Dinosaur</td>
<td>Semi-phonetic</td>
</tr>
<tr>
<td>Teeth</td>
<td></td>
<td>Semi-phonetic</td>
<td>Teeth</td>
<td>Consolidated</td>
</tr>
<tr>
<td>Fish</td>
<td></td>
<td>Consolidated</td>
<td>Fish</td>
<td>Consolidated</td>
</tr>
<tr>
<td>Chips</td>
<td></td>
<td>Consolidated</td>
<td>Chips</td>
<td>Consolidated</td>
</tr>
<tr>
<td>Bridge</td>
<td></td>
<td>Semi-phonetic</td>
<td>Bridge</td>
<td>Phonetic</td>
</tr>
<tr>
<td>Cake</td>
<td></td>
<td>Consolidated</td>
<td>Cake</td>
<td>Consolidated</td>
</tr>
</tbody>
</table>

**Figure 16.** Spelling performance at pre-intervention and post-intervention for a child from the typically developing group.
<table>
<thead>
<tr>
<th>Target</th>
<th>Pre Spelling Stage</th>
<th>Post Spelling Stage</th>
</tr>
</thead>
<tbody>
<tr>
<td>(aged 6;6)</td>
<td>(aged 6;10)</td>
<td></td>
</tr>
<tr>
<td><strong>Rain</strong></td>
<td>Phonetic</td>
<td>Phonetic</td>
</tr>
<tr>
<td>Kangaroo</td>
<td>Semi-phonetic</td>
<td>Semi-phonetic</td>
</tr>
<tr>
<td><strong>Girl</strong></td>
<td>Phonetic</td>
<td>Consolidated</td>
</tr>
<tr>
<td><strong>Shark</strong></td>
<td>Phonetic</td>
<td>Phonetic</td>
</tr>
<tr>
<td><strong>Dinosaur</strong></td>
<td>Phonetic</td>
<td>Phonetic</td>
</tr>
<tr>
<td><strong>Teeth</strong></td>
<td>Phonetic</td>
<td>Consolidated</td>
</tr>
<tr>
<td><strong>Fish</strong></td>
<td>Consolidated</td>
<td>Consolidated</td>
</tr>
<tr>
<td><strong>Chips</strong></td>
<td>Consolidated</td>
<td>Phonetic</td>
</tr>
<tr>
<td><strong>Bridge</strong></td>
<td>Phonetic</td>
<td>Semi-phonetic</td>
</tr>
<tr>
<td><strong>Cake</strong></td>
<td>Consolidated</td>
<td>Consolidated</td>
</tr>
</tbody>
</table>

**Figure 17.** Spelling performance at pre-intervention and post-intervention for a child from the typically developing group.
Discussion

This study employed a controlled multiple single subject design to evaluate the effectiveness of an integrated phonological awareness intervention for 12 children with CAS aged 4 to 8 years. The intervention aimed to improve speech, letter sound and/or word decoding knowledge, and phoneme awareness skills. The effects of the intervention on the children’s speech, reading, and spelling ability were monitored over the intervention period. The above skills were also monitored for a group of 12 children with typical speech and language development (who did not participate in therapy) over the intervention period. The therapy was administered in two 6-week therapy blocks (two individual sessions per week) with one 6-week withdrawal block between the two treatment blocks.

Speech

The first hypothesis tested was that the research intervention would improve the accuracy of speech production of trained and untrained words containing target speech error patterns. This hypothesis was supported by the data for the majority of participants. Ten of the twelve participants improved the accuracy of the trained words for both targeted error patterns. Seven children also improved the accuracy of the untrained words for both targeted error patterns, while a further three children improved the accuracy of the untrained words for one targeted error pattern alone. One child improved the accuracy of trained and untrained words for one targeted error pattern alone. A final child did not improve in any of the trained or untrained items. There were no gains in the control language probe over the intervention period for participants.
The second hypothesis tested was that participants would improve the accuracy of words containing a target speech error pattern that were produced spontaneously during connected speech. This hypothesis was supported partially by the data. Nine participants generalized gains in the first speech target to the connected speaking context, whereas four participants generalized gains in the second speech target to the connected speaking context. The lower proportion of participants exhibiting generalization to connected speech for the second speech target is likely due to the administration of the connected speech probe immediately post-intervention giving less time for generalization of the second target. Follow-up testing is required to evaluate if the increased accuracy of the speech target from the second block is generalised to a connected speaking context.

The results showed that the intervention was effective in suppressing the targeted speech error patterns and that the majority of participants exhibited transfer of speech gains to untrained speech items. There was evidence of generalization of treated error patterns to the connected speaking context, (particularly targets from block one), for some participants. The gains in speech production over the intervention period could not be attributed to general maturation of the linguistic system alone as development in the control language measure was not exhibited over the intervention period.

The increased accuracy of trained speech items is consistent with that reported in the pilot study. Moriarty and Gillon (2006) reported that two of the three participants in their investigation suppressed the use of their target phonological process without change in their control probe. The results also extend the pilot results by demonstrating the generalization of speech gains to untrained items and a connected speaking context for some participants. The generalization of speech gains in the current sample is contrary to
the common notion that children with CAS struggle with transferring treatment gains to linguistic contexts and is likely due to the nature of the research intervention. The integrated approach targeted speech error patterns, rather than drilling certain words or phrases, which may have facilitated wider spread change in the children’s phonological systems. Although intervention based on speech error pattern suppression has been contraindicated by some authors for children with CAS who do not identify a phonological component to the disorder (Aram, 1984; Love & Fitzgerald, 1984), its use is appropriate if the child presents with delayed or disordered speech error patterns as exhibited by participants in the current study (Velleman & Strand, 1994).

The improvement in speech production skills achieved by the CAS group is also contrary to reports indicating that children with CAS progress slowly in therapy compared to children with other developmental speech-language disorders (Campbell, 1999; Pannbacker, 1988). The average PCC gain of 12.4% achieved by the CAS group over the four month intervention period is similar to that reported in treatment studies for children with other developmental speech sound disorders with integrated goals over similar intervention periods. For example, Gillon (2000) reported that 20 hours of integrated phonological awareness approach over 4.5 months produced an average PCC gain of 13.2% in 23 participants aged 5;6 to 7;6 with spoken language impairment. Tyler, Lewis, and Welch (2003) reported that 24 weeks of morpho-syntactic and phonological intervention (one individual and one group session per week) produced an average PCC gain of 13.1% for participants aged 3;0 – 5;11. The findings thus indicate consistency in the response to integrated interventions between CAS and children with other types of speech disorder.
Phoneme Awareness

The third hypothesis tested was that participants would increase the phonological awareness of trained and untrained words containing a target speech error pattern. The data supported the hypothesis for the majority of the participants. Eight children improved in both their trained and untrained phonological awareness probes for both targets. One child improved in one trained phonological awareness probe alone. Three children did not improve their phonological awareness scores. Improvement in phonological awareness at a group level was also evident with a mean gain of 10.8 standard scores achieved on a standardized phonological awareness measure.

The findings are consistent with results from the pilot study demonstrating it is possible to teach phoneme awareness in an integrated framework to children with CAS (Moriarty & Gillon, 2006). The results are also consistent with previous research demonstrating that phoneme identity skills can be enhanced in 4 year old children with speech impairment (Gillon, 2005; Hesketh, 2007). The single-subject design also drew attention to four participants who did not improve in all trained and untrained phonological awareness probes. Two of the children who did not increase their phonological awareness performance (participants 7 and 11) were the two youngest children in the sample, presented with the most severe speech difficulty, and showed no change in their performance in the phonological representation task over the intervention. The third pre-school aged child who did not improve in the phonological awareness probes (participant 10) performed near-ceiling on the measures at pre and post-intervention making it difficult to show a treatment effect. The lack of improvement in the phonological probes in the fourth child (Participant 1) is likely related to the
complexity of the phonological structure of his probe words (CCVC and CCCVC respectively). This participant did evidence some gains in phonological awareness skills, however, through his improved performance on the non-word reading and informal spelling tasks.

Phonological awareness ‘treatment resisters’ have been described in other phonological awareness intervention studies (Gillon, 2005; Hesketh et al., 2007). The current results suggest that resistance to phonological awareness intervention may be related to phonological representation skills. Further investigation is required to elucidate the relationship between phonological representation and phonological awareness in response to phonological awareness intervention.

**Letter Knowledge, reading, spelling, and phonological representation**

The fourth hypothesis tested was that participants would increase their letter knowledge skills. The data supported this hypothesis with the CAS group learning an average of 8.5 letter-sound combinations during the intervention. The fifth hypothesis tested was that participants would increase their real word decoding, non-word decoding, spelling, and phonological representation ability. The participants made significant gains in all areas except for real word decoding.

The non-significant change in real word decoding may be explained by the nature of the reading assessment which included many phonetically irregular items (e.g., some, one). The reading task in the intervention programme (i.e., manipulation with letter blocks) focused on decoding phonetically regular real and non-words alone. Further, the small number of children aged six years and older in the CAS sample made it more
difficult to detect statistically significant results. The non-word reading performance of the children with CAS (which improved an average of 40% phonemes correct) demonstrated that the children with CAS were able to use their increased phonological awareness skills in the reading process.

Overall the findings suggested that an integrated approach provides an effective method of targeting factors underlying poor reading and spelling skills in children with CAS such as phonological awareness and phonological representation ability. Further, the integration of letter knowledge into phoneme awareness tasks facilitated the transfer of improved phonological awareness skills to the reading and spelling process.

The fifth hypothesis tested was that the CAS group would demonstrate more rapid growth in phonological awareness, letter knowledge, spelling, non-word reading, real-word reading, and phonological representation over the intervention period than children with typical development (TD) not receiving the intervention. This hypothesis was partially supported by the data with the CAS group demonstrating more rapid growth over the intervention period in phonological awareness, letter knowledge, non-word reading, and spelling ability than the TD group. There was no difference in the rate of development in real word reading, and phonological representation between the two groups.

The ability of the integrated programme to create gains in non-word reading, spelling, phonological awareness, and letter knowledge in children with CAS at a greater rate than children with TD holds great promise for enhancing the literacy outcomes for children with CAS. It is essential, however, to establish the longer term impact of the research intervention on the spoken and written language development of children with
CAS. The longitudinal pilot study presented in Chapter 3 reported that one child (Katie) was able to maintain gains in phonological awareness over time, whereas the second child (Derek) did not maintain his post-intervention performance in phonological awareness probes at follow-up assessment 12 months post-intervention. The proceeding chapter therefore compares the CAS and TD groups’ performance on the speech, phonological awareness, decoding, and spelling measures six months following completion of the phonological awareness intervention programme described in the current chapter.
CHAPTER 5

THE LONGER TERM EFFECTS OF AN INTEGRATED PHONOLOGICAL AWARENESS INTERVENTION FOR CHILDREN WITH CHILDHOOD APRAXIA OF SPEECH

5.1 Introduction

The experiment presented in Chapter 4 demonstrated that children with childhood apraxia of speech (CAS) made significant gains in speech, phonological awareness, decoding, and spelling skills in response to an integrated phonological awareness intervention. Further, they exhibited accelerated growth in these skills compared to their peers with typical development over the intervention period.

Although the positive treatment effects from the intervention study are very encouraging, the effectiveness of an integrated phonological awareness intervention requires ongoing investigation. In particular, the longer term effects of the intervention require examination. Children with CAS are notorious for their slow progress in therapy and are reported to experience difficulty generalising intervention targets (Campbell, 1999; Velleman, 2003). It is important, therefore, to demonstrate that children with CAS are able to maintain speech, phonological awareness, reading, and spelling gains once intervention support is removed. Further, the evaluation of the effectiveness of the integrated phonological awareness approach in facilitating literacy development has been
limited to single word decoding and spelling of words presented in isolation. As one of the ultimate goals of phonological awareness intervention is to improve a child’s ability to decode and understand meaningful text, it is critical that the reading accuracy and reading comprehension at text level is examined for children who have participated in the intervention.

Gillon (2002) conducted a follow-up study to compare the phonological awareness, reading, and spelling effects of an integrated phonological awareness intervention and traditional speech-language therapy for children with spoken language impairment. The children were re-assessed 11 months following completion of their intervention. The researcher reported that children who participated in the phonological awareness intervention exhibited continued accelerated growth in phonological awareness and literacy measures in relation to the comparison group with speech disorder over the follow-up period. There was no difference in the developmental rate in target measures between the children who participated in the phonological awareness intervention and their peers with typical speech-language development over the follow-up period. Although the rate of change in targeted skills was not as extreme over the follow-up period as the intervention period, children who participated in the phonological awareness intervention continued to show gains in all measures. The Gillon (2002) study suggests that an integrated phonological awareness intervention continues to influence phonological awareness, reading, and spelling development after intervention support is removed.

The study reported in this chapter evaluated the phonological awareness, letter knowledge, decoding, and spelling development in children with CAS six months
following their participation in the integrated phonological awareness intervention. The literacy development of the group of children with typical speech and language development (TD) was also monitored over the follow-up period to compare the relative change in phonological awareness, letter knowledge, decoding, and spelling between the groups. The reading accuracy and comprehension at text level was evaluated for the CAS group.

The following hypotheses were tested:

1. Children with CAS would exhibit higher scores on speech, phonological awareness, decoding, and spelling measures at the six month follow-up assessment than those achieved at pre and post-intervention assessment.

2. Children with CAS would exhibit similar proportions of children performing below expected range on phonological awareness, letter knowledge, and decoding normative tests immediately following phonological awareness intervention and at follow-up intervention.

3. Children with CAS would not continue the accelerated relative change exhibited over the intervention period compared to their peers with typical development in phonological awareness, decoding, and spelling measures over the follow-up period.

4. Children with CAS would be able to transfer improved phonological awareness and decoding abilities to connected text reading performance.
5.2 Method

5.2.1 Participants

All children from CAS and TD group were available for re-assessment six months following completion of the intervention programme. All children had participated in two full terms of school or kindergarten in the follow-up period.

Children from the CAS group received varying levels of speech and language therapy support following participation in the research intervention. Four children resumed speech and language therapy within the public education system. One of these children received therapy sessions once every two weeks with their speech-language therapist and daily support from a teacher aide in the classroom context. The other child received two speech-language therapy intervention sessions during the six month follow up period. The intensity of intervention scheduling was unknown for the other two children who received therapy support within the public education system following the intervention. Five children were referred for further speech and language therapy intervention through the public education service immediately following the research intervention. However, they were still waiting to receive this support at the time of the six month follow-up assessment. Three children accessed weekly private speech-language therapy intervention, and one of these children also had daily teacher aide assistance in the classroom. No children were involved in any remedial reading interventions such as reading recovery during the six month follow-up period.
5.2.2 Procedure

All follow-up assessment sessions were conducted in a quiet withdrawal setting in the child’s school or home. The sessions were recorded on the Olympus DS-2 digital voice recorder (with inbuilt stereo microphone) for reliability purposes. The following measures were re-administered at follow-up.

Speech measures

Percent Consonant Corrects (PCC)

- The PCC was calculated by analyzing participants’ responses from the BBTOP (Bankson & Bernthal, 1990) and the first trial of the 25-word consistency test from the Diagnostic Evaluation of Articulation and Phonology (DEAP, Dodd et al., 2006) (giving a sample of 105 words) with Computerized Profiling software (Long & Fey, 2005).

Inconsistency percentage

- The 25-word consistency test from the DEAP (Dodd et al., 2006) was used to calculate an inconsistency percentage for each child. An inconsistent production is marked when the child produces at least two different forms of a target word across three production trials. An inconsistency percentage of 40% and above denotes inconsistent speech errors.

Standardised phonological awareness measures

- Preschool and Primary Inventory of Phonological Awareness (PIPA) (Dodd et al., 2000) for children aged 4 years at pre-intervention.
  - Standard scores from the rhyme awareness, alliteration awareness, and phoneme identity subtests were collected to determine performance
within/below the expected range. The standard scores from the three subtests were also combined into a composite score for data analysis.

- Test of Phonological Awareness (TOPA) (Torgesen & Bryant, 1994) for children aged 5 to 7 years at pre-intervention assessment.
  - This assessment measures initial and final phoneme identity awareness in a receptive task.
  - Standard scores were collected to determine performance within/below the expected range. Raw scores were collected for data analysis.

**Letter-sound Knowledge Measure**

- The letter-sound knowledge subtest of the PIPA (Dodd *et al.*, 2000) was administered to all participants.
  - Raw scores (out of 30) were recorded for data analysis. Standard scores were also recorded for children aged 6 years and younger (i.e., aged within the normative sample of the assessment) to determine performance within/below the expected range.

**Decoding Measures** (administered to participants who were aged 6:0 and older at pre-intervention)

  - Raw scores were collected for data analysis. Age equivalence scores were also recorded to determine performance within/below the expected range.

- *Non-word reading task* (adapted from Calder, 1992)
Children were required to decode three sets of ten non-words of CV, CVC or CCVC structure.

The percentage of phonemes correctly decoded for this task was calculated. For example, if the child read the word ‘tob’ as ‘top’, they would receive a score of out of a possible score of 3 for that item. See Appendix A for a full list of items included within the non-word reading task.

**Informal Spelling Measure** (administered to participants aged 6;0 and older at pre-intervention)

- Ten words from the 25 word consistency test were used to assess spelling.
  - A percent graphemes correct (PGC) figure was calculated from the children’s responses. For example, if the word ‘fish’ was spelt as ‘fis’, the child would be awarded 2 out of a possible 3 points for that item.
  - Spelling attempts were also classified according to Ehri’s (2000) stage model of spelling development (i.e., pre-communicative, semi-phonetic, phonetic, and transitional).

**Additional Follow-up Measures**

- The Neale Analysis on Reading Ability – Third Edition (Neale, 1999) was administered to children in the CAS group aged 6;0 and older at follow-up (n = 7) to evaluate the children’s reading accuracy and comprehension from connected text. Percentiles were collected from the accuracy and comprehension subtest.
• There was one child (participant 12) who was aged 6;0 at follow-up assessment, but was too young to complete reading and spelling tasks at the pre-intervention and post-intervention assessment points. The following measures were administered to participant 12 (at follow-up assessment alone) to monitor his early reading and spelling development.
  o Burt word reading test (Gilmore et al., 1981, as described above)
  o Informal non-word reading task (Calder, 1992, as described above)
  o Informal spelling task (as described above)

5.2.3 Data reliability

Speech data (PCC)

Transcription reliability was determined for a second independent transcription conducted by an experienced SLP for five participants (just over 20% of participants). Mean inter-rater agreement (based on the number of phonemes transcribed identically) was 90.7% with a range of 83.5% to 100.0%.

Non-word reading data

An independent reviewer analyzed and scored the non-word reading responses of five participants (just over 20% of participants). Mean inter-rater agreement (based on the number of phonemes transcribed identically) was 89.5% with a range of 81.2% to 100%.

Spelling data

An independent reviewer analysed all spelling responses from the CAS and TD groups and calculated a PGC score and provided descriptive analysis of the spelling stage (Ehri, 2000). Mean inter-rater agreement for PGC was 91.2% with a range of 86.7% to
100%. Mean inter-rater agreement for the identification of spelling stages was 94.7% with a range of 91.2% to 100%. Any differences between the original and reviewed responses were resolved by the reviewers reaching consensus after reviewing the spelling responses together.

5.3 Results

A one-way repeated measures ANOVA was used to compare performance in the speech, phonological awareness, decoding, and spelling measures of the CAS group over the three assessment points (pre-intervention, post-intervention, and follow-up). The time period between pre- and post-intervention assessments was approximately five months for each child and six months had lapsed between post-intervention and follow-up assessments. The analysis was used to determine if children with CAS exhibited significantly higher scores on the assessment measures at post-intervention and follow-up assessments compared to pre-intervention assessment. In circumstances where data did not meet normality or equal variance assumptions, a Friedman Repeated Measures ANOVA on ranks was employed to evaluate change over the three assessment points. Post-hoc testing (Tukey) was employed to determine which data points were significantly different from each other.

The results, which are presented in Table 20 (one way repeated measures ANOVA) and Table 18 (Friedman repeated measures ANOVA), indicate that the CAS group made a significant gain in all the pre-post measures over the intervention period, and that the CAS group did not make a significant gain in any of the measures from post-intervention to follow-up. The effect size index $f$ (appropriate for ANOVA, Portney & Watkins, 2000) was also calculated for all statistically significant comparisons and can be
viewed in Table 21. The effect size was interpreted as follows: 0.10 – small; 0.25 – medium and 0.40 and above – large (Portney & Watkins, 2000).

Table 20. Assessment data for the three assessment points (one way repeated ANOVA analysis)

<table>
<thead>
<tr>
<th></th>
<th>Pre</th>
<th>Post</th>
<th>Follow-up</th>
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<tbody>
<tr>
<td></td>
<td>Mean</td>
<td>SD</td>
<td>Mean</td>
</tr>
<tr>
<td>Letter Knowledge</td>
<td>8.3</td>
<td>9.6</td>
<td>16.8</td>
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<tr>
<td>(n = 12)</td>
<td></td>
<td></td>
<td></td>
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<tr>
<td>NWR&lt;sup&gt;1&lt;/sup&gt; (PPC&lt;sup&gt;2&lt;/sup&gt;)</td>
<td>8.9</td>
<td>15.0</td>
<td>49.3*</td>
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<tr>
<td>(n = 6)</td>
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<tr>
<td>Spelling (PGC&lt;sup&gt;3&lt;/sup&gt;)</td>
<td>20.1</td>
<td>12.8</td>
<td>51.7*</td>
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<td>(n = 6)</td>
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Note. <sup>1</sup> Non-word reading, <sup>2</sup> Percent Phonemes Correct, <sup>3</sup> Percent Graphemes Correct, * = Significant difference between pre and post-intervention score (p < .05).
Table 21. Assessment data for the three assessment points (Friedman repeated measures ANOVA on ranks)

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<th>Post</th>
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<tr>
<td></td>
<td>M¹ 25%</td>
<td>75%</td>
<td>M¹ 25%</td>
<td>75% M¹</td>
<td>25%</td>
<td>75%</td>
<td>Chi</td>
<td>P</td>
<td></td>
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<tr>
<td>PCC²</td>
<td>54.6</td>
<td>33.5</td>
<td>70.6</td>
<td>66.0</td>
<td>52.7</td>
<td>83.4</td>
<td>84.2</td>
<td>67.8</td>
<td>91.1</td>
<td>22.2</td>
<td>&lt;0.001</td>
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<td>(n = 12)</td>
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<tr>
<td>TOPA⁴</td>
<td>7.0</td>
<td>3.5</td>
<td>7.0</td>
<td>11.0*</td>
<td>9.5</td>
<td>12.8</td>
<td>12.0</td>
<td>9.0</td>
<td>13.8</td>
<td>9.5</td>
<td>0.016</td>
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<td>(n = 7)</td>
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<tr>
<td>Burt⁵</td>
<td>12.0</td>
<td>5.0</td>
<td>24.0</td>
<td>20.0*</td>
<td>18.0</td>
<td>25.0</td>
<td>24.0</td>
<td>18.0</td>
<td>36.0</td>
<td>11.3</td>
<td>0.002</td>
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<td>(n = 6)</td>
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Note. ¹ Median, ² Percent Consonants Correct, ³ Percent Vowels Correct, ⁴ Test of Phonological Awareness (Torgesen & Bryant, 1994), ⁵ Burt Word reading test raw score (Gilmore et al., 1981), Chi – chi-squared, * Significant difference between pre and post-intervention score (p < .05)

5.3.1 Comparison to norms

The proportion of participants in the CAS group performing below the expected range on the standardized phonological awareness (TOPA, PIPA), letter knowledge and word decoding (Burt) measures at pre-intervention, post-intervention and follow-up assessments was calculated to compare the proportion of children with CAS performing below the expected range at each assessment point. The results, which can be viewed in Figure 18, show that the CAS group had fewer participants performing below normal limits at the post-intervention and follow-up assessment points. However, a significant
proportion of participants continue to perform below the expected range at post-intervention and follow-up despite significant gains across the intervention period.

Figure 18. Proportion of CAS group performing below expected range in standardized phonological awareness, letter knowledge and decoding measures

Note. TOPA = Test of Phonological Awareness (Torgesen & Bryant, 1994), PIPA = Preschool and Primary Inventory of Phonological Awareness (Dodd et al., 2000), RA = rhyme awareness, AA = alliteration awareness, PI = phoneme identity, LK = letter knowledge, Burt = Burt word reading test (Gilmore et al., 1981).
5.3.2 **Comparison of CAS and TD change scores**

**TOPA**

The change scores (post-test minus pre-test, divided by pre-test to control for pre-test variability) for the TD and CAS groups were compared for the intervention (pre-post-intervention) and follow-up (post-intervention to follow-up) periods. Change scores were examined to determine if the accelerated relative change in phonological awareness, decoding, and spelling achieved by the children with CAS over the intervention period would continue over the follow-up period. A two-way repeated measures ANOVA with one factor repetition (group by intervention change and follow-up change scores) showed a significant group effect, $F(1,13) = 6.9, p = 0.021$, a significant time effect $F(1,29) = 8.3, p = 0.013$, and group x time interaction $F(1,29) = 7.8, p = 0.015$. *Post-hoc* analysis (Tukey) showed that the CAS group had significantly higher change scores than the TD group over the intervention period ($p < 0.001$). There was no difference between the change scores over the follow-up period for the CAS and TD groups ($p = 0.878$). The accelerated growth in TOPA performance of the CAS group compared to the TD group over the intervention period can be viewed in Figure 19.
Figure 19. TOPA performance (mean raw score) of the CAS and TD groups at pre-intervention, post-intervention and follow-up assessments.

Note. TOPA = Test of Phonological Awareness (Torgesen & Bryant, 1994), CAS = Childhood Apraxia of Speech group, Typical = Typically developing group.

Letter Knowledge

The change scores for the TD and CAS groups were compared for the intervention and follow-up periods. A two-way repeated measures ANOVA with one factor repetition (group by intervention change and follow-up change scores) showed a significant group effect, $F(1, 22) = 8.4, p = 0.008$, a significant time effect, $F(1, 47) = 15.9, p < 0.001$, and a significant group x time interaction, $F(1, 47) = 11.4, p = 0.003$. 
Post-hoc analysis (Tukey) showed that the CAS group had significantly higher change scores than the TD group over the intervention period \((p<0.001)\). There was no difference between the change scores over the follow-up period for the CAS and TD groups \((p = 0.670)\). The accelerated growth in letter knowledge performance of the CAS group compared to the TD group over the intervention period can be viewed in Figure 20.

![Figure 20](image-url)

**Figure 20.** Letter knowledge performance of CAS and TD groups at pre-intervention, post-intervention and follow-up assessments

**Note.** CAS = Childhood Apraxia of Speech group \((n = 12)\), Typical = typically developing group \((n = 12)\).

**Decoding**

The Burt Word Reading Test change scores for the TD and CAS groups were compared for the intervention and follow-up periods. A two-way repeated measures
ANOVA with one factor repetition (group by intervention change and follow-up change scores) showed a insignificant group effect, $F(1, 12) = 1.9, p = 0.117$, an insignificant time effect, $F(1, 27) = 2.8, p = 0.120$, and a insignificant group x time interaction, $F(1, 27) = 2.7, p = 0.125$. The performance of the CAS and TD groups across the three assessment points (pre-intervention, post-intervention, follow-up) can be viewed in Figure 21.

![Figure 21. Burt Word Reading Test (Gilmore et al., 1981) performance of CAS and TD groups at pre-intervention, post-intervention and follow-up assessments](image)

**Figure 21.** Burt Word Reading Test (Gilmore *et al.*, 1981) performance of CAS and TD groups at pre-intervention, post-intervention and follow-up assessments

**Note.** CAS = Childhood Apraxia of Speech group ($n = 6$), Typical = typically developing group ($n = 8$).

The non-word reading change scores for the TD and CAS groups were compared for the intervention and follow-up periods. A two-way repeated measures ANOVA with one factor repetition (group by intervention change and follow-up change scores) showed
a significant group effect, $F(1, 12) = 8.1, p = 0.015$, a significant time effect, $F(1, 27) = 8.5, p = 0.013$, and a significant group x time interaction, $F(1, 27) = 8.4, p = 0.013$. Post-hoc analysis (Tukey) showed that the CAS group had significantly higher change scores than the TD group over the intervention period ($p<0.001$). There was no difference between the change scores over the follow-up period for the CAS and TD groups ($p = 0.987$). The accelerated growth in letter knowledge performance of the CAS group compared to the TD group over the intervention period can be viewed in Figure 22.

**Figure 22.** Non-word reading performance of the CAS and TD groups at pre-intervention, post-intervention and follow-up assessments

**Note.** CAS = Childhood Apraxia of Speech group ($n = 6$), Typical = typically developing group ($n = 7$)
Spelling

The spelling change scores for the TD and CAS groups were compared for the intervention and follow-up periods. A two-way repeated measures ANOVA with one factor repetition (group by intervention change and follow-up change scores) showed a significant group effect, $F(1, 11) = 10.3, p = 0.008$, a significant time effect, $F(1, 25) = 16.4, p = 0.002$, and a significant group x time interaction, $F(1, 25) = 12.7, p = 0.004$. Post-hoc analysis (Tukey) showed that the CAS group had significantly higher change scores than the TD group over the intervention period ($p<0.001$). There was no difference between the change scores over the follow-up period for the CAS and TD groups ($p = 0.846$). The accelerated growth in letter knowledge performance of the CAS group compared to the TD group over the intervention period can be viewed in Figure 23.
Figure 23. Spelling performance of the CAS and TD groups at pre-intervention, post-intervention and follow-up assessments

Note. CAS = Childhood Apraxia of Speech group (n = 6), Typical = typically developing group (n = 7)

5.3.3 Additional follow-up measures

Connected reading

The performance of children in the CAS group aged 6;0 and older at follow-up on the Neale Analysis of Reading Ability (Neale, 1999) was analysed to determine if the children with CAS were able to transfer their improved phonological awareness and decoding abilities to their connected text reading performance. The results are presented in Table 22. The findings demonstrate that three children with CAS exhibited decoding accuracy and reading comprehension within the expected range on the connected reading
task, while four children with CAS exhibited decoding accuracy and reading comprehension below the expected range on the connected reading task.

Table 22. Neale Analysis of Reading Ability (Neale, 1999) performance for children in the CAS group aged 6;0 and older at follow-up assessment (n = 7)

<table>
<thead>
<tr>
<th>Participant</th>
<th>Accuracy (percentile)</th>
<th>Comprehension (percentile)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>10</td>
<td>12</td>
</tr>
<tr>
<td>2</td>
<td>47</td>
<td>52</td>
</tr>
<tr>
<td>5</td>
<td>27</td>
<td>42</td>
</tr>
<tr>
<td>6</td>
<td>11</td>
<td>19</td>
</tr>
<tr>
<td>8</td>
<td>11</td>
<td>19</td>
</tr>
<tr>
<td>9</td>
<td>11</td>
<td>19</td>
</tr>
<tr>
<td>12</td>
<td>50</td>
<td>45</td>
</tr>
</tbody>
</table>

Early reading and spelling development for Participant 12

A descriptive analysis of Participant 12’s reading and spelling performance at follow-up (aged 6;0) was conducted. Participant 12 was the only child from the CAS
group who had received the research intervention at pre-school and had participated in six months of formal schooling at the follow-up assessment. Although Participant 12 was aged 6;0 at follow-up, he had only attended school for six months, as his parents chose to delay school entry due to his unintelligibility. As shown in Table 19, Participant 12 performed within the expected range in the accuracy and comprehension subtests of the connected reading measure. He also performed within the expected range on the normative real word decoding task (reading 17 words correctly), and showed an ability to use an alphabetic reading strategy in the non-word reading task (scoring 29% phonemes correct). During the non-word reading task he consistently decoding the initial sound correctly, and the ability to decode the final sound in the word was emerging.

Participant 12’s performance on the informal spelling test is presented in Figure 24. The results show that his spelling attempts are predominately at the semi-phonetic and phonetic stages of spelling development (Ehri, 2000). Participant 12’s early reading and spelling development is consistent with his phonological awareness ability, scoring a standard score of 94 on the TOPA (Torgesen & Bryant, 1994) at follow-up assessment.
<table>
<thead>
<tr>
<th>Target</th>
<th>Follow-up</th>
<th>Spelling stage</th>
</tr>
</thead>
<tbody>
<tr>
<td>Rain</td>
<td>ran</td>
<td>Phonetic</td>
</tr>
<tr>
<td>Kangaroo</td>
<td>kooro</td>
<td>Semi-phonetic</td>
</tr>
<tr>
<td>Girl</td>
<td>goog</td>
<td>Semi-phonetic</td>
</tr>
<tr>
<td>Shark</td>
<td>sak</td>
<td>Semi-phonetic</td>
</tr>
<tr>
<td>Dinosaur</td>
<td>No response</td>
<td>No response</td>
</tr>
<tr>
<td>Teeth</td>
<td>teth</td>
<td>Phonetic</td>
</tr>
<tr>
<td>Fish</td>
<td>fosh</td>
<td>Phonetic</td>
</tr>
<tr>
<td>Chips</td>
<td>No response</td>
<td>No response</td>
</tr>
<tr>
<td>Bridge</td>
<td>balv</td>
<td>Semi-phonetic</td>
</tr>
<tr>
<td>Cake</td>
<td>kak</td>
<td>Phonetic</td>
</tr>
</tbody>
</table>

Figure 24. Spelling performance at follow-up for participant 12.

Spelling items were not directly trained in therapy.
5.4 Discussion

This study examined the longer term effects of an integrated phonological awareness program for 12 children with CAS aged 4 to 7 years with phonological awareness and speech deficits. The intervention aimed to simultaneously improve the children’s phonological awareness, letter knowledge, and speech production ability. These skills were re-assessed six months following completion of the intervention programme. The phonological awareness, decoding, and spelling development of a group of children with typical speech and language development (TD) was also monitored over the follow-up period to compare the relative change in these measures between the two groups.

The first hypothesis tested was that children with CAS would exhibit higher scores on speech, phonological awareness, decoding, and spelling measures at the six month follow-up assessment than those achieved at pre- and post-intervention assessment. This hypothesis was not supported by the data. Although there was a significant difference between the pre-and post-intervention assessment measures, there was no difference in any of the post-intervention and follow-up assessment measures for the CAS group.

These findings are inconsistent with reports showing continued growth during the intervention and follow-up period for children with spoken language impairment that have participated in an integrated phonological awareness intervention (Gillon, 2002). The children with CAS did not regress in the assessment measures, but their scores plateaued following the completion of the intervention. The failure of the children with CAS to demonstrated gains in the assessment measures over the follow-up period may be
due to the nature of the disorder. Children with CAS commonly experience difficulty generalizing treatment gains to different contexts. Velleman (2003) pinpoints the need to ‘train flexibility’ within speech intervention approaches for children with CAS. It is possible that children with CAS experience similar generalization difficulties in response to phonological awareness intervention. This would result in difficulty transferring improved phonological awareness and decoding skills from intervention sessions to the reading and spelling process at home or in the classroom. For example, children with CAS may be able to use a phonological reading strategy when supported within intervention sessions but revert to a non-phonological strategy (e.g., making whole word guesses) when reading in the classroom. In line with the self teaching hypothesis, such restricted decoding experience would inhibit the acquisition of more complex letter-sound combinations and more fluent reading (Share, 1995).

The lack of continued development may also be due to the severity of the reading and phonological awareness deficits in the children with CAS. The older children in the sample (i.e., aged 6;6 – 7;10) all achieved age equivalence scores of <6;0 in the real word decoding measure at pre-intervention. Consistent with these results, five of the seven children tested on the TOPA (Torgesen & Bryant, 1994) presented with standard scores more than 1.5 standard deviations below the mean, with the other two children demonstrating performance more than 1.0 standard deviation below the mean. The children also presented with multiple risk factors for reading delay including persistent speech disorder, expressive language impairment, and poor phonological awareness (Nathan et al., 2004) which may have contributed to the limited growth in targeted areas following the completion of the intervention.
The second hypothesis tested was that children with CAS would exhibit similar proportions of children performing below expected range on phonological awareness, letter knowledge, and decoding normative tests immediately following phonological awareness intervention and at follow-up intervention. The data confirmed these hypotheses. The results showed that the CAS group had fewer participants performing below normal limits at the post-intervention and follow-up assessment points than at pre-intervention. However, a significant proportion of participants continued to perform below the expected range in phonological awareness and real-word decoding at follow-up. These results are consistent with previous reports indicating the effectiveness of phonological awareness intervention for children with CAS (Moriarty & Gillon, 2006). The findings show that the treatment effects of an integrated phonological awareness intervention can be maintained over time by children with CAS.

The third hypothesis tested was that children with CAS would not continue the accelerated comparative change exhibited over the intervention period compared to their peers with typical development in phonological awareness, decoding, and spelling measures over the follow-up period. The data confirmed this hypothesis with no difference in the change scores in the above measures between the CAS and TD groups over the follow-up period. This result is in contrast to the accelerated change in the phonological awareness, letter knowledge, non-word reading, and spelling measures experienced by the CAS group over the intervention period.

Although the results point to the positive effects of the intervention during the intervention period, it is important to develop approaches that facilitate sustained phonological awareness, reading, and spelling development for children with CAS. A
preventative intervention framework (Gillon, 2005) that facilitates speech, early phoneme awareness, and letter-sound knowledge in the preschool years may be an effective means of ensuring sustained development in literacy skills for this population. Gillon (2005) reported that children with moderate-severe speech disorder who had participated in an integrated intervention at age 3 and 4 years performed within or above the expected range in a standardised decoding task in their first or second year of schooling. The current findings also allude to the potential long-term benefits of providing an integrated intervention to children with CAS in the preschool years. The early decoding and spelling development of one child who received the research intervention prior to school entry was examined. The findings showed that this child had developed the ability to use phoneme awareness (blending and segmenting) and letter-sound knowledge in non-word decoding and spelling tasks. Further, he performed within the expected range for his age on real-word decoding and the connected reading normative assessments. The phonological awareness performance of this child at follow-up mirrored this performance, achieving a standard score of 94 on the normative phonological awareness measure. This result, however, must be interpreted cautiously, as the early reading and spelling development of other children in the CAS group who participated in the research intervention was not explored (due to their young age). Further, the two youngest children in the sample presented with the most severe speech and phoneme awareness difficulty at pre-intervention. It is important that the performance of these children is monitored over the long-term. Future investigations are needed to examine the effectiveness of an integrated phonological awareness provided within a preventative framework for a larger number of children with CAS.
The fourth hypothesis tested was that children with CAS would be able to transfer increased phonological awareness and decoding abilities to a connected reading task. This hypothesis was supported by the data with reading performance in the text reading task matching the children’s phonological awareness and decoding skills. The same three participants (in the group aged 6;0 and above at follow-up) who presented with phonological awareness skills within the expected range also performed within the expected range in the decoding sub-test of the text reading measure. Children who performed within the expected range in the accuracy component of the text reading measure also performed within the expected range on the comprehension component of the measure. Four children in this group continued to perform well below average on text reading accuracy and comprehension (in line with their phonological awareness ability).

The immediate and longer-term results of the intervention study indicated that the research intervention was effective in facilitating spoken and written language development. The data from one participant also suggested that it may be particularly beneficial for reading and spelling development to provide phonological awareness intervention prior to the onset of formal education. The impact of an integrated phonological awareness intervention within a preventative framework for literacy development for children with CAS, however, requires further investigation. The following chapter therefore presents a longitudinal case study of identical twin boys, aged 4;5 at pre-intervention and followed during their first year of formal schooling.
CHAPTER 6

A PHONOLOGICAL AWARENESS INTERVENTION CASE STUDY OF IDENTICAL TWIN BOYS WITH CHILDHOOD APRAXIA OF SPEECH (CAS)

6.1 Introduction

The experiments presented in Chapters 4 and 5 highlighted the benefit of providing an integrated phonological awareness intervention for children with CAS. Significant gains were made in all targeted areas and these improvements were maintained over a six month follow-up period. The intervention, however, was unable to stimulate sustained accelerated growth in phonological awareness and written language measures following completion of the programme for the majority of participants. The typical early literacy development of one child who received the research intervention prior to the onset of formal schooling and the relatively strong follow-up performance in phonological awareness for the children aged 4 years during intervention pointed to the value of providing phonological awareness intervention for children with CAS within a preventative framework. The current study provides a case study evaluation of identical twin boys (aged 4;5 at the start of the study) who participated in the integrated phonological awareness intervention and were monitored for one year following participation in the programme. The study thus allowed a detailed examination of the
effects of the intervention on early reading and spelling development for these two children.

Longitudinal evaluation of children with CAS is essential to delineate the developmental course of the disorder. Stackhouse (1992) argued that a developmental perspective in CAS research was necessary to understand the evolving shape of the disorder over time. The few longitudinal studies focusing on CAS have generally focused on speech, phonological awareness, and written language deficits in the disorder. Jacks et al., (2006), Davis et al., (2005), and Marquardt et al., (2004) provided a longitudinal evaluation of vowel errors, inconsistent speech production, and consonant production in three children with CAS. Stackhouse and Snowling (1992a) provided a detailed examination of the written language development of two older children with CAS. Given recent reports that language impairment may also be a key symptom in CAS (Lewis, Freebairn, Hansen, Iyengar et al., 2004; Lewis, Freebairn, Hansen, Taylor et al., 2004), it is essential to document language development in children with the disorder.

One longitudinal study that has included a focus on language impairment in CAS is Lewis, Freebairn, Hansen, Iyengar et al.’s (2004) longitudinal study of children with CAS (as compared to children with speech disorder and combined speech-language impairment). This investigation tracked performance in a global language measurement (the Test Of Language Development (TOLD, Newcomer & Hammill, 1997) at preschool assessment and the Clinical Evaluation of Language Fundamentals (CELF-4, Semel et al., 2003) at school-age assessment) across the study. Although the investigation presented valuable information regarding the severity of language impairment in children with CAS, more detailed data regarding the development of particular language skills
within the same child will give further insight into the language deficits in the disorder. Morphosyntactic production appears to be one language area where children with CAS experience particular difficulty. Ekelman and Aram (1983) reported deficits in sentence structure and morphological marker production that could not be explained by the children’s speech production difficulties. Further, the children’s morpho-syntactic deficits were more severe than predicted from their mean length of utterance (MLU). Longitudinal evaluation of the morpho-syntactic ability of children with CAS will identify if grammatical deficits persist over time in the disorder and if they minimise with improved speech production skills.

6.1.1 Evaluating the response of identical twins to the research intervention

As far as the author is aware, identical twins with CAS have not been described elsewhere in the research literature. This study thus creates a unique opportunity to examine genetic and environmental factors that may influence a child’s response to an integrated intervention. As outlined in Chapter 1, CAS is a heritable disorder with strong familial aggregation to other developmental speech and language disorders and a mutation in the FOXP2 (7q31) gene identified in one family with around half its members exhibiting symptoms consistent with CAS (Lai et al., 2001; Lewis, Freebairn, Hansen, Iyengar et al., 2004). The aggregation of CAS in identical twins (i.e., twins that share the same genetic make-up) in the current study also highlights the genetic basis to the disorder.

The combination of shared genes (and generally shared environment) means that identical twins tend to have similar speech, language, and literacy development. Bishop, North, and Donlan (1996) found that there is a 70% chance that an identical twin with an
articulation or language disorder has a co-twin that is also affected, whereas there is a 50% chance that a non-identical twin will have a co-twin that is also affected. This evidence thus highlights a strong genetic base to speech and language disorder (given identical and non-identical twins have different DNA but similar environments). Shared (e.g., parenting style) and non-shared environment (e.g., temperament, personality, different teacher) factors also have an influence on speech and language development (hence there is not a 100% correspondence between the aggregation of speech and language disorders in identical twins).

There has been limited research comparing the response of twins to speech-language therapy intervention. McMahon and Dodd (1995) evaluated the effectiveness of phonologically based speech intervention for twins with severe expressive phonological impairment (33% and 8% PCC respectively). The twins responded well to the intervention (increasing to 82% and 76% PCC respectively). The authors noted that the twin that presented with the strongest speech production skills initially responded more favourably to the intervention. These preliminary results suggest that pre-intervention severity may predict which twin will progress more rapidly in speech-language therapy.

The current investigation evaluated the long-term effects of an integrated phonological programme provided within a preventative framework for identical twin boys with CAS. In particular the study focused on the early reading and spelling development of the children. The study also examined the expressive morpho-syntactic development production of the boys within a standardised personal narrative over the 1 year and 4 month duration of the study.

The following hypotheses were tested:
1. That the children’s speech accuracy (percent consonants correct, PCC and percent vowels correct, PVC) in single word production would improve and speech inconsistency would decrease over the course of the study.

2. That the children’s phonological awareness would be within the expected range for their age following participation in the intervention.

3. That the children’s performance in a receptive phonological representation task would improve over the course of the study.

4. That the children’s reading development would be within the expected range for their age in their first year of school.

5. That the children would be able to use a phonological strategy in early writing attempts.

6. That the children would present with persistent morphosyntactic deficits in a personal narrative task at all assessment points despite gains in speech production skills.

7. That the children will progress at a similar rate in the intervention (given their shared genetic make-up, shared environment, and similar pre-intervention assessment results).

6.2 Case history

Theo and Jamie\(^3\) (identical twins) were induced at 37 weeks gestation due to their size (weighing 7 pound 2 ounces and 7 pound 11 ounces respectively). Their birth was unremarkable. They have one sister who is two years older and one sister who is two years younger than them. Both boys experienced early feeding and swallowing

\(^3\) Pseudonyms
difficulties (although these were more severe in Theo) including nasal regurgitation, difficulty transitioning to solid food, and choking. Theo underwent a videofluoroscopic evaluation of swallowing at age 3;3. The assessment showed that he used a discoordinated swallow and was at increased risk of aspiration, but that he was able to clear the bolus successfully with a flexed neck posture. At age 4 years the boys continued to have some difficulty chewing solids, but the choking and nasal regurgitation had largely resolved. Theo continued to have some difficulty controlling his drool at this age. Theo and Jamie’s early motor development was typical, crawling at around 5 to 6 months and walking at 14 to 16 months. The boys have some history of otitis media but hearing evaluations carried out at age 3;5 revealed normal hearing acuity.

Theo and Jamie’s early expressive speech and language development was slow. The boys’ mother (Kate) reported that they did not babble much as infants, and many of their communicative attempts consisted of ‘grunts’. Their first word was uttered at around 12 to 18 months. At age 2;6, Theo and Jamie used around five words and communicated primarily via gesture. The boys’ initial speech and language therapy programme focused on teaching the Makaton signing system to decrease their communication frustration and to promote expressive language development. Later therapy focused on increasing speech intelligibility by repeatedly practicing target sounds and words. By age 3;9 Theo and Jamie’s expressive vocabularies had increased to around 100 words. Their speech production was effortful and included multiple speech errors making them unintelligible to unfamiliar listeners. Theo and Jamie have no history of receptive language impairment and were diagnosed with CAS by their SLT at age 3;9.
Theo and Jamie underwent genetic assessment at age 3;10 to identify any genetic cause of their CAS. The karyotype analysis revealed a small interstitial deletion on chromosome 10 (deletion at 10q21.2 – 22.1). Kate’s karyotype analysis also showed the same deletion, but has no history of speech, language, or literacy disorder. Phenotypic evaluation noted the following characteristics in the boys: mild dysmorphia with small eyes, small ears, and slightly thickened lips, small ear pit in front of the right ear tragus, broad nose with well defined tip, mild epicanthus, mild pectus carinatum chest deformity, and mild abnormality of both maxillary incisors. Theo and Jamie presented with normal cardiorespiratory and abdominal examinations and normal genitalia. The interstitial deletion identified in Theo and Jamie is extremely rare (Doheny et al., 1997). Other cases reported in the literature presented with more severe deficits than Theo and Jamie, and generally involve developmental delay, hypotonia, heart murmur, telecanthus, broad nose, and ear abnormalities (J. Davis, Kardon, & Selman, 1982; Doheny et al., 1997; Glover, Gabarron, & Lopez-Ballester, 1987; Marks, Yu, Curry, & Zorn, 1991).

Theo and Jamie have a family history of speech and language impairment:

1. Their older sister has experienced some early reading difficulty (their younger sister, however, presented with typical language).
2. Their paternal cousin, maternal aunt, and maternal grandmother have a history of dyslexia.
3. Another maternal aunt was born with absent maxillary incisors.
4. Their maternal cousin (son of their aunt born with absent maxillary incisors) has a history of cleft lip and dyslexia.
6.2.1 *Shared and non-shared environment*

Theo and Jamie shared a very similar environment. They lived in the same home, enjoyed the same activities and attended the same kindergarten session. Theo and Jamie tended to play with each other at kindergarten.

Although Jamie and Theo’s linguistic profiles at pre-intervention assessment were very similar (see Table 23 below), Kate reported that Jamie tended to lead Theo slightly in development. For example, Jamie was nine ounces bigger than Theo at birth and had less severe feeding difficulties than his brother.

The author observed differences in personality between Theo and Jamie. Jamie tended to be more self conscious about his speech errors whereas Theo tended to be a little more outgoing.

6.3 *Method*

6.3.1 *Procedure*

Theo and Jamie were assessed on four occasions (pre-intervention, post-intervention, six months following completion of the intervention programme, and one year following completion of the intervention programme). They were aged 4;5, 4;9, 5;3, and 5;9 at each respective assessment point. Theo and Jamie started school at age 5;0.

In their initial evaluation the following assessments were administered (more detailed description of the measures can be viewed in Chapter 2):

- Peabody Picture Vocabulary Test (PPVT) (Dunn & Dunn, 1997). The standard score was collection from this evaluation.
• The three receptive subtests of the Clinical Evaluation of Language Fundamentals – Preschool (CELF-P) (Wiig, Secord, & Semel, 2004). Standard scores were collected for the subtests. Expressive language assessment was not conducted due to their speech production difficulties.

• The oro-motor and inconsistency subtests of the Diagnostic Evaluation of Articulation and Phonology (DEAP, Dodd et al., 2006). Standard scores were collected from the oro-motor subtests and the children’s inconsistency percentage was calculated.

• The items from the first trial of the inconsistency subtest and the Bankson Bernthal Test of Phonology (BBTOP) (Bankson & Bernthal, 1990) were used to gain a PCC and PVC score via Computerized Profiling (Long & Fey, 2005).

• The rhyme awareness, alliteration awareness, phoneme identity and letter knowledge subtests of the Preschool and Primary Inventory of Phonological Awareness (PIPA) (Dodd et al., 2000). Standard scores were collected for this subtest. A zero raw score equates to a standard of 7 in some subtests for 4 year olds.

• The phonological representation judgement task (Sutherland & Gillon, 2007). Raw scores (out of 25) were collected from this task.

• The language sampling protocol (Westerveld & Gillon, 2002) was used to assess expressive morpho-syntactic performance in personal narrative production. In the assessment the examiner shows the child a standard
series of photos and tells a standard story that matches each photo. After the examiner has told the story, he/she prompts the child to tell their own story relating to the picture. Responses were analysed via the Systematic Analysis of Language Transcripts (SALT) software and compared to the New Zealand database (Miller & Chapman, 2000). The children’s personal narrative production was also used to evaluate prosody and speech accuracy within a connected speaking context.

Theo and Jamie’s language profile at pre-intervention can be viewed in Table 20.
<table>
<thead>
<tr>
<th></th>
<th>Theo</th>
<th>Jamie</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Table 23. Assessment data for Theo and Jamie</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>PPVT</strong> (standard score: typical range 85 – 115)</td>
<td>89</td>
<td>91</td>
</tr>
<tr>
<td><strong>CELF – P</strong> (standard score: typical range 7 – 13)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Concepts and following directions</td>
<td>14</td>
<td>14</td>
</tr>
<tr>
<td>Basic concepts</td>
<td>9</td>
<td>11</td>
</tr>
<tr>
<td>Sentence structure</td>
<td>11</td>
<td>15</td>
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<tr>
<td><strong>DEAP oro-motor</strong> (standard score: typical range = 7 – 13)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Isolated movements</td>
<td>6</td>
<td>6</td>
</tr>
<tr>
<td>Sequenced movements</td>
<td>7</td>
<td>7</td>
</tr>
<tr>
<td>Diadochokinetatic task</td>
<td>5</td>
<td>6</td>
</tr>
<tr>
<td><strong>Speech analysis</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Early 8 sounds (percent consonants correct, PCC)</td>
<td>80.3%</td>
<td>83.1%</td>
</tr>
<tr>
<td>Middle 8 sounds (PCC)</td>
<td>48.7%</td>
<td>52.0%</td>
</tr>
<tr>
<td>Late 8 sounds (PCC)</td>
<td>25.9%</td>
<td>26.9%</td>
</tr>
<tr>
<td>Total PCC</td>
<td>52.5%</td>
<td>54.8%</td>
</tr>
<tr>
<td>Percent vowels correct (PVC)</td>
<td>82.1%</td>
<td>83.3%</td>
</tr>
<tr>
<td>Inconsistency percentage</td>
<td>64%</td>
<td>56%</td>
</tr>
<tr>
<td><strong>PIPA</strong> (standard score)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Rhyme awareness</td>
<td>7</td>
<td>6</td>
</tr>
<tr>
<td>Alliteration awareness</td>
<td>8</td>
<td>8</td>
</tr>
<tr>
<td>Phoneme identity</td>
<td>7</td>
<td>7</td>
</tr>
<tr>
<td>Letter knowledge</td>
<td>7</td>
<td>7</td>
</tr>
</tbody>
</table>
Table 23 continued

**Phonological representation judgement task** (out of 25) 14 16


**Intervention**

Theo and Jamie participated in the research intervention in individual sessions conducted by the author in a quiet room in their home. Each session was alternated so if Theo received his session before Jamie, the order would be reversed at the next session. Both boys worked on the suppression of the velar fronting phonological error pattern in the first treatment block and the s-cluster phonological error pattern in the second treatment block. Theo and Jamie responded well to the intervention, with both boys significantly improving trained and untrained performance for all speech and phonological awareness probes. By the end of the second treatment block, they had generalised their production of velar sounds to connected speech. Graphic analysis of Theo and Jamie’s performance in the probes can be viewed in Appendices D and E (Participants 3 and 4).

The following measures were re-administered at the post-intervention, first follow-up, and second follow-up assessment points:
• The BBTOP (Bankson & Bernthal, 1990) and inconsistency subtest of the DEAP (Dodd et al., 2006).

• The rhyme awareness, alliteration awareness, phoneme identity, and letter knowledge subtest of the PIPA (Dodd et al., 2000).

• The phonological representational judgement task (Sutherland & Gillon, 2007).

• The personal narrative language sample (Westerveld & Gillon, 2002).

The following measures were used to monitor early reading and spelling development:

• Descriptive analysis of early classroom writing attempts was completed at the first follow-up session.

The second follow up assessment session also included:

• Burt Word Reading Test (Gilmore et al., 1981). Raw and age equivalence scores were collected.

• Informal non-word reading task (adapted from Calder, 1992). The percent phonemes correctly decoded (PPC) was collected. A full list of the words used in this test can be viewed in Appendix A.

• Informal spelling task. The percent graphemes correct (PGC) and spelling stage was calculated (Ehri, 2000).

• Neale Analysis of Reading Ability (Neale, 1999). Percentiles from the reading accuracy and comprehension subtests were calculated.
6.3.2 Data reliability

Reliability was calculated for the personal narrative transcription and measures employed in the second follow-up session (measures employed at the other assessment points were included in the reliability figures presented in Chapters 4 and 5).

Personal narrative transcription

Two of the eight transcriptions (one for Theo and Jamie at each of the four assessment points) were randomly chosen for reliability analysis. An independent reviewer listened to the audio-file of the personal narrative while viewing the original SALT transcript. Point by point inter-rater reliability (i.e., utterance by utterance) for the first transcript was 84.0% and 80.0% for the second transcript. Any differences between the transcribed and the reviewed responses were resolved by the reviewers reaching consensus following repeatedly listening to the recordings.

Speech

The items from the BBTOP (Bankson & Bernthal, 1990) and inconsistency subtest of the DEAP (Dodd et al., 2006) in the second follow-up session were transcribed by an independent reviewer. Inter-rater agreement was 96.3% for Theo’s scores and 94.7% for Jamie’s scores. Any differences between the transcribed and the reviewed responses were resolved by the reviewers reaching consensus following repeatedly listening to recordings.

Non-word reading

An independent reviewer re-transcribed Theo and Jamie’s non-word reading attempts. Inter-rater agreement was 90.1% for Theo’s scores and 88.7% for Jamie’s
scores. Any differences between the transcribed and reviewed responses were resolved by the reviewers reaching consensus following repeatedly listening to recordings.

**Spelling**

An independent reviewer re-analysed Theo and Jamie’s spelling attempts for PGC and analysis of spelling stage. Inter-rater agreement for PGC was 96.5% for Theo and 97.2% for Jamie. Inter-rater agreement for spelling stage was 100.0% for Theo and 90.0% for Jamie. Any differences between the original and reviewed analysis were resolved by the reviewers reaching consensus following reviewing the spelling transcripts together.

### 6.4 Results

#### 6.4.1 Speech production

Theo and Jamie’s speech production accuracy (PCC, PVC) and inconsistency percentage over the four assessment points are presented in Figures 25 and 26. Their growth in the accuracy of early, middle, and late developing consonants (Shriberg, 1993) is represented in Figures 27 and 28. The figures show that Theo and Jamie became more accurate and more consistent in their speech production over the study. Their large PCC gain (particularly in later developing consonants) from the post-intervention to the first follow-up session is due to the acquisition of many fricative sounds over this period. This development may have been a consequence of targeting s-cluster reduction in the second block of intervention, as the boys’ did not produce any fricatives before this error pattern was targeted. Despite the high PCC scores that were achieved by both Theo and Jamie at the second follow-up session, they were still unintelligible at times during
connected speech. Theo and Jamie presented with effortful speech production at all assessment points. They had difficulty controlling their speech volume and presented with an excess and equal stress pattern.

Figure 25. Theo’s speech accuracy and inconsistency over the four assessment points

Note. Pre = pre-intervention, post = post-intervention, FU 1 = follow-up assessment six months following the completion of the intervention, FU 2 = follow-up assessment 12 months following completion of the intervention, PCC = percent consonants correct, PVC = percent vowels correct, inconsistency = inconsistency percentage from the inconsistency subtest of the Diagnostic Evaluation of Articulation and Phonology (DEAP, Dodd *et al.*, 2006), age represented as years;months.
Figure 26. Jamie’s speech accuracy and inconsistency over the four assessment points.

Note. Pre = pre-intervention, post = post-intervention, FU 1 = follow-up assessment six months following completion of the intervention, FU 2 = follow-up assessment 12 months following completion of the intervention, PCC = percent consonants correct, PVC = percent vowels correct, inconsistency = inconsistency percentage from the inconsistency subtest of the Diagnostic Evaluation of Articulation and Phonology (DEAP, Dodd et al., 2006), age represented as years:months.
Figure 27. Theo’s accuracy for early, middle, and late developing consonants

Note. Pre = pre-intervention, post = post-intervention, FU 1 = follow-up assessment six months following completion of the intervention, FU 2 = follow-up assessment 12 months following completion of the intervention, age represented as years;months.

Figure 28. Jamie’s accuracy for early, middle, and late developing consonants.

Note. Pre = pre-intervention, post = post-intervention, FU 1 = follow-up assessment six months following completion of the intervention, FU 2 = follow-up assessment 12 months following completion of the intervention, age represented as years;months.
6.4.2 Phonological awareness and phonological representation ability

Theo and Jamie’s performance on the PIPA (Dodd et al., 2000) is presented in Table 24. The results show that Theo and Jamie were able to maintain phonological awareness and letter knowledge performance well within or above the expected range for their age following the intervention programme.

Table 24. Theo and Jamie’s phonological awareness over the study

<table>
<thead>
<tr>
<th></th>
<th>Theo</th>
<th></th>
<th>Jamie</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>RA</td>
<td>AA</td>
<td>PI</td>
</tr>
<tr>
<td>Pre</td>
<td>7</td>
<td>8</td>
<td>7</td>
</tr>
<tr>
<td>Post</td>
<td>15</td>
<td>17</td>
<td>12</td>
</tr>
<tr>
<td>FU 1</td>
<td>11</td>
<td>13</td>
<td>11</td>
</tr>
<tr>
<td>FU 2</td>
<td>10</td>
<td>11</td>
<td>11</td>
</tr>
</tbody>
</table>

Note. RA = rhyme awareness, AA = alliteration awareness, PI = phoneme identity, LK = letter knowledge, Pre = pre-intervention, post = post-intervention, FU 1 = follow-up six months following completion of the intervention assessment 1, FU 2 = follow-up assessment 12 months following completion of the intervention, all scores are standard scores.

Phonological representation

Theo and Jamie’s performance over time on the phonological representation judgement task is displayed in Figures 29 and 30. The boys showed marked improvement in the task over the intervention period and scored at ceiling (raw score of 25) or near-ceiling level for the remainder of the study.
Figure 29. Theo’s phonological representation scores over the study.

Note. Pre = pre-intervention, post = post-intervention, FU 1 = follow-up assessment six months following completion of the intervention, FU 2 = follow-up assessment 12 months following completion of the intervention, phon rep = phonological representation task.

Figure 30. Jamie’s phonological representation scores over the study.

Note. Pre = pre-intervention, post = post-intervention, FU 1 = follow-up assessment six months following completion of the intervention, FU 2 = follow-up assessment 12 months following completion of the intervention, phon rep = phonological representation task.
6.4.3 Early reading and spelling development

Letter-sound knowledge

Theo and Jamie’s raw scores in the letter-sound knowledge subtest of the PIPA (Dodd et al., 2000) are presented in Figures 31 and 32. The boys made sustained development in letter-sound knowledge throughout the study, although Jamie showed greater improvement than Theo over the last follow-up period. The marked change in letter-sound knowledge from the post-intervention to first follow-up assessment point coincides with the onset of formal literacy instruction.

![Figure 31](image)

**Figure 31.** Theo’s letter-sound knowledge over the study.

**Note.** Pre = pre-intervention, post = post-intervention, FU 1 = follow-up assessment six months following completion of the intervention, FU 2 = follow-up assessment 12 months following completion of the intervention, raw score is out of a possible 30 points.
Figure 32. Jamie’s letter-sound knowledge over the study.

Note. Pre = pre-intervention, post = post-intervention, FU 1 = follow-up assessment six months following completion of the intervention, FU 2 = follow-up assessment 12 months following completion of the intervention first follow-up session, FU 2 = second follow-up session, raw score is out of a possible 30 points.

Early writing attempts

Samples of Theo and Jamie’s early story writing attempts from their first term of school (i.e. first ten weeks of school) were collected. Two of Theo’s stories are shown in Figure 33. These early writing attempts show an immediate use of semi-phonetic and phonetic spelling attempts (Ehri, 2000). Note the s-clusters ‘sp’ and ‘st’ are represented correctly in the spelling of ‘speedy’ and ‘monster’. Theo’s second speech target was s-cluster reduction and his trained words included four words containing the ‘st’ cluster and four words containing the ‘sp’ cluster. An example of Jamie’s early story writing is presented in Figure 34. Jamie also shows an early use of phonetic and phonetic spelling strategies (Ehri, 2000).
Figure 33. Examples of Theo’s story writing in his first school term.
Figure 34. An example of Jamie’s story writing in his first school term.

Reading assessment at age 5:9

Theo and Jamie’s performance in the real-word reading, non-word decoding, and connected reading measures at the second follow-up session are presented in Table 25. Theo and Jamie presented with decoding and reading comprehension within the expected range, and showed an ability to use an alphabetic reading strategy in the non-word reading task. Jamie achieved a higher score than Theo in the non-word reading task (49% compared to Theo’s 23%) which may be related to his superior performance on the letter-sound knowledge task at the second follow up assessment.
Table 25. Theo and Jamie’s scores in the reading measures at age 5;9.

<table>
<thead>
<tr>
<th></th>
<th>Theo</th>
<th>Jamie</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Burt Word reading</strong> (raw score)</td>
<td>7</td>
<td>9</td>
</tr>
<tr>
<td><strong>Non-word reading</strong> (percent phonemes correct)</td>
<td>23%</td>
<td>49%</td>
</tr>
<tr>
<td><strong>Neale accuracy</strong> (percentile 1)</td>
<td>45</td>
<td>45</td>
</tr>
<tr>
<td><strong>Neale comprehension</strong> (percentile 1)</td>
<td>57</td>
<td>57</td>
</tr>
</tbody>
</table>

Note. Burt = Burt word reading test (Gilmore *et al.*, 1981), Neale = Neale Analysis of Reading Ability (Neale, 1999), age = years;months.

Spelling assessment at age 5;9

Theo and Jamie’s spelling performance is presented in Figure 35. The attempts demonstrate that the boys are able to use semi-phonetic and phonetic spelling strategies (Ehri, 2000).
<table>
<thead>
<tr>
<th>Target</th>
<th>Theo</th>
<th>Spelling Stage</th>
<th>Jamie</th>
<th>Spelling Stage</th>
</tr>
</thead>
<tbody>
<tr>
<td>Rain</td>
<td>ran</td>
<td>Phonetic</td>
<td>ran</td>
<td>Phonetic</td>
</tr>
<tr>
<td>Kangaroo</td>
<td>can`foW</td>
<td>Semi-phonetic</td>
<td>cagroo</td>
<td>Semi-phonetic</td>
</tr>
<tr>
<td>Girl</td>
<td>go1</td>
<td>Phonetic</td>
<td>go1</td>
<td>Phonetic</td>
</tr>
<tr>
<td>Shark</td>
<td>tha0</td>
<td>Semi-phonetic</td>
<td>shAC</td>
<td>Phonetic</td>
</tr>
<tr>
<td>Dinosaur</td>
<td>diuso</td>
<td>Semi-phonetic</td>
<td>b9300a</td>
<td>Semi-phonetic</td>
</tr>
<tr>
<td>Teeth</td>
<td>tef</td>
<td>Phonetic</td>
<td>tef</td>
<td>Phonetic</td>
</tr>
<tr>
<td>Fish</td>
<td>pith</td>
<td>Semi-phonetic</td>
<td>fish</td>
<td>Consolidated</td>
</tr>
<tr>
<td>Chips</td>
<td>thip</td>
<td>Semi-phonetic</td>
<td>ThS</td>
<td>Semi-phonetic</td>
</tr>
<tr>
<td>Bridge</td>
<td>beth</td>
<td>Semi-phonetic</td>
<td>bi g</td>
<td>Semi-phonetic</td>
</tr>
<tr>
<td>Cake</td>
<td>cau</td>
<td>Semi-phonetic</td>
<td>cac</td>
<td>Phonetic</td>
</tr>
</tbody>
</table>

**Figure 35.** Theo and Jamie’s spelling attempts at age 5:9.

Spelling stage analysed according to Ehri, 2000
6.4.4 Expressive morphsyntactic development

Theo and Jamie’s SALT analysis from the personal narrative task (Westerveld & Gillon, 2002) across the four assessment points is summarised in Table 26. Each personal narrative was cut at 50 complete and intelligible utterances to allow a fair comparison in the measures across the assessment points. The analysis showed that the boys caught up to their peers in their mean length of utterance (MLU) over the follow-up period. There was, however, negligible development in MLU over the intervention period. Theo and Jamie presented with persistent deficits in word level errors, omitted words, and omitted bound morphemes throughout the study despite gains in intelligibility.
**Table 26. SALT analysis summary for Theo and Jamie**

<table>
<thead>
<tr>
<th></th>
<th>Theo</th>
<th>Jamie</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Pre</td>
<td>Post</td>
</tr>
<tr>
<td>MLU (morphemes)</td>
<td>2.9**</td>
<td>3.2**</td>
</tr>
<tr>
<td>TNDW</td>
<td>58**</td>
<td>79*</td>
</tr>
<tr>
<td>Intelligibility %</td>
<td>64**</td>
<td>79**</td>
</tr>
<tr>
<td>Omitted words</td>
<td>17**</td>
<td>22**</td>
</tr>
<tr>
<td>Omitted bound morphemes</td>
<td>2**</td>
<td>0</td>
</tr>
<tr>
<td>Word level errors</td>
<td>33**</td>
<td>27**</td>
</tr>
<tr>
<td>Utterance level errors</td>
<td>2</td>
<td>4**</td>
</tr>
</tbody>
</table>

**Note.** * = at least one standard deviation from the database mean, ** = at least two standard deviations from the database mean, MLU = mean length of utterance, TNDW = total number of different word roots, pre = pre-intervention assessment, post = post-intervention assessment, FU1 = first follow-up assessment (six months following completion of the intervention), FU2 = second follow-up assessment (12 months following completion of the intervention).

**Omitted words**

Descriptive analysis of Theo and Jamie’s omitted words identified that they consisted primarily of functional words. Theo and Jamie omitted determiners (a, an, the), auxiliary verbs (is, was, have, do etc.) and the word ‘to’ at high rates at each assessment point.
Bound morphemes

A breakdown of the bound morphemes that Theo and Jamie used correctly and omitted in obligatory contexts in their personal narrative production at each assessment point is presented in Tables 27 and 28. Theo had ongoing difficulty producing the past tense and using contractible forms of auxiliary and copula verbs. Jamie had ongoing difficulty in producing the past tense and did not produce a wide variety of bound morphemes in his narrative production. Theo and Jamie continued to have difficulty with bound morpheme production at the follow-up assessments, despite having an age appropriate MLU and increased speech intelligibility at this time.
Table 27. Theo’s bound morpheme use and omission in personal narrative production

<table>
<thead>
<tr>
<th>Bound Morpheme</th>
<th>Pre Used</th>
<th>Pre Omitted</th>
<th>Post Used</th>
<th>Post Omitted</th>
<th>Follow-up 1 Used</th>
<th>Follow-up 1 Omitted</th>
<th>Follow-up 2 Used</th>
<th>Follow-up 2 Omitted</th>
</tr>
</thead>
<tbody>
<tr>
<td>-ed</td>
<td>0</td>
<td>2</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>3</td>
<td>0</td>
<td>10</td>
</tr>
<tr>
<td>Plural –s</td>
<td>3</td>
<td>0</td>
<td>4</td>
<td>0</td>
<td>10</td>
<td>1</td>
<td>9</td>
<td>2</td>
</tr>
<tr>
<td>-ing</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>4</td>
<td>0</td>
</tr>
<tr>
<td>-n’t (not)</td>
<td>0</td>
<td>0</td>
<td>2</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>-s (is)</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>possessive ‘s</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>2</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Third person</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>1</td>
</tr>
</tbody>
</table>

Note. Pre = pre-intervention assessment, post = post-intervention assessment, follow-up 1 = assessment six months following completion of the intervention, follow-up 2 = assessment 12 months following completion of the intervention.
Table 28. Jamie’s bound morpheme use and omission in personal narrative production

<table>
<thead>
<tr>
<th>Bound Morpheme</th>
<th>Pre Used</th>
<th>Pre Omitted</th>
<th>Post Used</th>
<th>Post Omitted</th>
<th>Follow-up 1 Used</th>
<th>Follow-up 1 Omitted</th>
<th>Follow-up 2 Used</th>
<th>Follow-up 2 Omitted</th>
</tr>
</thead>
<tbody>
<tr>
<td>-ed</td>
<td>0</td>
<td>6</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>11</td>
<td>2</td>
<td>9</td>
</tr>
<tr>
<td>Plural –s</td>
<td>1</td>
<td>0</td>
<td>2</td>
<td>0</td>
<td>5</td>
<td>0</td>
<td>4</td>
<td>0</td>
</tr>
<tr>
<td>-ing</td>
<td>2</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>2</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>-n’t (not)</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>-‘s (is)</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>possessive ‘s</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Third person singular</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
</tbody>
</table>

Note. Pre = pre-intervention assessment, post = post-intervention assessment, follow-up 1 = assessment six months following completion of the intervention, follow-up 2 = assessment 12 months following completion of the intervention.

Word errors

The SALT analysis revealed that Theo and Jamie made a high number of word level errors in their narrative production compared to their typically developing peers. Closer inspection of the boys’ word level errors showed that the majority of these errors were due to pronoun difficulties (e.g., the boys contained to say ‘me’ for ‘I’ and ‘us’ for them throughout the study) and irregular past tense errors (e.g., saying ‘fly’ for ‘flew’,...
‘run’ for ‘ran’ etc.). These word level errors can not be explained by Theo and Jamie’s speech production difficulties.

6.5 Discussion

This study evaluated the effectiveness of an integrated phonological awareness intervention for identical twin boys with CAS. Theo and Jamie were aged 4;5 at the outset of the study and assessments were conducted immediately post-intervention (aged 4;9), six months following the completion of the programme (aged 5;3) and one year following completion of the programme (aged 5;9). The study monitored Theo and Jamie’s speech production, phonological awareness, reading, spelling and morphsyntactic development over the study.

6.5.1 Speech production

The first hypothesis tested was that Theo and Jamie’s speech accuracy (PCC, PVC) in single-word production would improve and speech inconsistency would decrease over the course of the study. This hypothesis was generally supported by the data. Theo and Jamie improved their consonant production within single words over each assessment point. Theo increased his vowel accuracy within single words over each assessment point. Jamie improved his vowel accuracy over the first three assessment points, but it dropped slightly at the final assessment. By the end of the study Theo and Jamie’s consonant and vowel production within single words was approaching 100%. Despite the high accuracy obtained in single word production, Theo and Jamie continued to be unintelligible at times during connected speech. Their speech production remained
effortful throughout the study, and they continued to have difficulty controlling their speaking volume.

Theo and Jamie’s speech production generally became more consistent throughout the study. Theo’s inconsistency percentage decreased at each successive assessment point. Jamie’s inconsistency percentage decreased over the first three assessment points, but increased slightly at the final assessment (probably due to the vowel errors noted in his speech in the session). Theo and Jamie continued to exhibit some inconsistent speech production throughout the study, both scoring 20% inconsistent productions at the final assessment point.

These results are consistent with previous research indicating that while there may be some fluctuation in speech production from session to session, children with CAS generally become more accurate and consistent in their speech production over time (B. L. Davis et al., 2005; Jacks et al., 2006; Lewis, Freebairn, Hansen, Iyengar et al., 2004; Marquardt et al., 2004; Stackhouse & Snowling, 1992b). The results are also consistent with reports indicating that children with CAS have persistent errors in connected speech production (Lewis, Freebairn, Hansen, Iyengar et al., 2004).

Theo and Jamie made the largest gains in speech accuracy over the intervention period and the first follow-up period. Examination of the data showed that the speech gains over the intervention period were largely due to more accurate production of the ‘middle 8’ sounds (Shriberg, 1993). This improvement is reflective of Theo and Jamie’s acquisition of velar sounds (their first targeted phonological error patterns). Theo and Jamie also produced s-clusters correctly within the post-intervention assessment, but still continued to produce singleton /s/ sounds as a cluster at this time. The speech gains over
the first follow-up period were largely due to more accurate production of the ‘late 8’ sounds (Shriberg, 1993). This gain is reflective of Theo and Jamie’s generalisation of s-cluster production to /s/ singleton production and the acquisition of further fricative sounds. Hodson (2006) highlighted the potential of targeting s-cluster production to incite widespread change within a child’s phonological system. The results indicated that children with CAS may be able to generalise targeted phonological error patterns, providing evidence for the use of a phonological treatment approach for those affected providing their assessment data indicate phonological process usage.

6.5.2 Phonological awareness and phonological representation ability

The second hypothesis tested was that the children’s phonological awareness would be within the expected range for their age following participation in the intervention. This hypothesis was supported by the data with Theo and Jamie performing within or above the expected range on the phonological awareness measure following participation in the intervention. These findings are consistent with data showing the long-term positive effects of an integrated phonological awareness approach provided within a preventative framework for preschool children with moderate-severe speech disorder (Gillon, 2005). The results are in conflict with the minimal phonological awareness gains reported in other phonological awareness intervention studies for children with speech disorder that have devoted less therapy time to phonological awareness and/or used a less direct teaching approach (Hesketh et al., 2000; Rvachew, Nowak, & Cloutier, 2004). The results thus highlighted the importance of providing intensive and explicit phonological awareness training when attempting to facilitate its development in children with speech disorder.
The third hypothesis tested was that Theo and Jamie would improve their performance in a receptive phonological representation task over the study. This hypothesis was partially supported by the data. Theo and Jamie improved from near chance level performance (14 and 16 out of 25 respectively) at pre-intervention to near ceiling level (24 out of 25) at post-intervention. Theo and Jamie continued to perform near ceiling level throughout the remainder of the study. Thus, the phonological representation task was not sensitive enough to determine if Theo and Jamie continued to develop these skills following participation in the programme. Theo and Jamie’s phonological awareness and speech development throughout the study suggested that their phonological representation skills became more sophisticated throughout the study.

Sutherland and Gillon (2007) conducted a longitudinal evaluation of the phonological representation skills of children with moderate-severe speech disorder (utilising the same phonological representation judgement task that was employed in the current study as one of their measures). Although the children with speech disorder presented with inferior phonological representation skills than children with typical development, they exhibited growth in phonological representation ability throughout the study. The Sutherland and Gillon (2007) study, however, increased the complexity of the stimuli employed in the phonological representation over time. It is important that developmentally appropriate phonological representation tasks are used in future investigations to gauge the impact of phonological awareness intervention on phonological representation ability. The correspondence between Theo and Jamie’s development in phonological awareness and phonological representation skills over the
intervention period supports the link between these two variables (Elbro et al., 1998; Rvachew, 2006; Sutherland & Gillon, 2007).

6.5.3 Reading and spelling development

The fourth hypothesis tested was that Theo and Jamie’s early reading development would be within the expected range for their age. The data supported this hypothesis with Theo and Jamie achieving age-appropriate scores on the normative decoding and reading comprehension measures at the second follow-up assessment. Theo and Jamie’s performance in the non-word reading task also showed the ability to use an alphabetic strategy in their early reading attempts.

The fifth hypothesis tested was that Theo and Jamie would be able to use a phonological strategy in early writing attempts. The data supported this hypothesis. Theo and Jamie showed phonetic and semi-phonetic spelling strategies in their story writing in their first term of school. Further, their performance in the spelling task revealed the utilisation of phonetic and semi-phonetic spelling strategies. These results are consistent with the typical reading outcomes achieved by children with speech disorder that have participated in an integrated phonological awareness programme in their preschool years (Gillon, 2005).

The sixth hypothesis tested was that Theo and Jamie would present with persistent morpho-syntactic deficits in a personal narrative task at all assessment points despite gains in speech production skills. The data supported this hypothesis. Although Theo and Jamie’s MLU had increased to the level of their typically developing peers by the final assessment, they continued to omit more words and bound morphemes in obligatory contexts and produce more word level errors than their peers. Theo and Jamie presented
with multiple pronoun errors (e.g., saying ‘me’ for ‘I’) and did not mark the irregular or regular past tense. Theo and Jamie’s expressive morpho-syntactic skills are in contrast to their receptive grammatical abilities, achieving a standard score of 11 and 15 respectively on the sentence structure subtest of the CELF-P (Wiig et al., 2004) at pre-intervention.

Ekelman and Aram (1983) also reported that children with CAS have morphological and sentence structure deficits that were incongruent with the children’s speech production ability or MLU. Further, in Chapter 4 all children in the CAS group presented with either a pronoun error or omitted bound morpheme that was inappropriate for their age that could be employed as a control language probe. These findings suggest that children with CAS are likely to experience persistent expressive syntactic deficits irrespective of speech accuracy or receptive language skills. The results are further consistent with Lewis, Freebairn, Hansen, Iyengar et al.’s (2004) study that demonstrated that language deficits in children with CAS persist regardless of gains in speech accuracy.

It appears that the ongoing expressive syntactic difficulties may need to be incorporated into therapeutic strategies for children with CAS. Other investigations examining treatment options for preschool children with co-morbid phonological and morpho-syntactic deficits have reported that phonological interventions do not provoke a cross domain treatment effect in morpho-syntactic skills (Tyler, Lewis, Haskill, & Tolbert, 2002). A focus on the morphological development of children with CAS is particularly essential given the importance of morphological skills for reading comprehension and later decoding and spelling development (Muter et al., 2004; Singson, Mahony, & Mann, 2000).
6.5.4 Response to the intervention

The seventh hypothesis tested was that Theo and Jamie would progress at a similar rate in the intervention. The intervention data largely supported this hypothesis. Theo and Jamie made almost identical gains in speech, phonological awareness, and phonological representation ability throughout the study. Some differences were noted in Theo and Jamie’s assessment profile at the final assessment session. Jamie performed superiorly to Theo on the letter-sound knowledge task at the final assessment point, despite the boys showing similar scores in the first three assessment sessions. Despite Theo and Jamie’s similar performance on the standardised phonological awareness measure at final assessment, Jamie achieved a higher score on the non-word reading task and slightly stronger spelling score at the final assessment. The discrepancy in the scores on this task may have been reflective of Theo’s slightly better letter-sound knowledge at the final assessment point.

These results are consistent with the shared genetic and environmental influences on speech and language development. Theo and Jamie have identical DNA and participated in the same intervention, using the same resources, and working with the same SLT. The slightly stronger performance by Theo on letter knowledge, non-word reading, and spelling at the final assessment point might be due to his slightly stronger speech and language skills at pre-intervention (McMahon & Dodd, 1995). Jamie exhibited slightly better speech accuracy (PCC of 54.8% versus 52.5%), receptive vocabulary (PPVT standard score of 91 versus 89), and more consistent speech prior to the commencement of the intervention (56% inconsistent versus 64% inconsistent).
The current study demonstrated the positive speech, phonological awareness, and early literacy development of identical twins with CAS who participated in an integrated phonological awareness intervention at preschool age. Theo and Jamie had ongoing expressive morpho-syntactic deficits despite strong receptive language and exhibiting a positive response to the intervention. It is important to monitor the long-term literacy development of Theo and Jamie to identify any negative impact of their syntactic deficit on later written language development.
CHAPTER 7

SUMMARY OF RESULTS AND GENERAL DISCUSSION

7.1 Introduction

Children with childhood apraxia of speech (CAS) frequently experience severe and persistent spoken and written language disorder. Whether these children are at greater risk for reading and spelling difficulty than children with other speech-language impairment, however, is unresolved. Further, treatment efficacy data to advance spoken and written language development for this population is sparse. Although differing approaches to improve speech production in children with CAS have been described in the literature, there is little evidence from controlled research designs to validate their effectiveness. Intervention research examining the effectiveness of approaches to enhance written language development in children with CAS has not previously been undertaken. This thesis addressed these areas of research need. The development of linguistic areas known to influence later written language development, namely phonological awareness, letter knowledge, and early decoding ability, was examined in 12 children aged 4 to 7 years with CAS. The effectiveness of a new intervention designed to simultaneously facilitate speech, phonological awareness, reading, and spelling development for these children was explored. Two broad research questions were addressed in this thesis:
1. How do the phonological awareness, reading, and spelling skills of children with CAS compare to children with other speech disorder and children with typical speech and language development?

2. What are the immediate and longer term effects of an integrated phonological awareness programme on the phonological awareness, speech, reading, and spelling development of children with CAS?

The following section describes the research methodology employed in a series of five experiments designed to answer these research questions.

7.2 Research methodology

7.2.1 Experiment 1: Investigating phonological awareness and early reading development in children with CAS

This study identified 12 children with CAS aged 4 to 7 years according to a current published diagnostic model of CAS. The phonological awareness, phonological representation, and early reading skills of these children were compared to children with inconsistent speech disorder and children with typical speech-language development. Children with inconsistent speech disorder presented with similar speech severity, inconsistency of speech production, and receptive vocabulary ability to children with CAS. Consequently, it was possible to determine whether the phonological awareness and early reading development of children with CAS was significantly delayed compared to their peers with typical development, and whether children with CAS were more disposed to written language disorder than children with another type of speech disorder.
7.2.2 Experiment 2: Follow-up pilot study

This study monitored the long-term effects of an intensive integrated phonological awareness programme (seven hours of intervention over three weeks) on two children with CAS aged 6;3 and 7;3 at the outset of the study. The investigation examined the children’s speech, phonological awareness, and decoding development 12 months following participation in the pilot intervention (Moriarty & Gillon, 2006). The exploratory study aimed to ascertain whether an integrated phonological awareness programme was a potential successful treatment for children with CAS.

7.2.3 Experiment 3: Intervention study

The effectiveness of an integrated phonological awareness approach in facilitating speech, phonological awareness, reading, and spelling for the 12 children with CAS identified in the first experiment was evaluated. The participants thus had a confirmed CAS diagnosis and the inclusionary criteria for the CAS group were described explicitly to allow study replication. The study employed a controlled multiple single-subject design with repeated measures to analyse change in trained and untrained speech and phoneme segmentation targets. An expressive morpho-syntactic error that was not age appropriate (e.g., pronoun error, bound morpheme error) was selected as a control probe. The treatment design thus permitted evaluation of the children’s ability to generalise speech and phoneme awareness targets and to exclude general maturation as a cause of any treatment effects. The study also used a comparative group design to evaluate the phonological awareness, reading and spelling development of the group of children with CAS compared to their peers with typical development over the intervention time period.
Speech-language therapists (SLTs) or senior speech-language therapy students delivered the intervention to the children with CAS on an individual basis. The treatment schedule included two six week treatment blocks (each including two 45-minute sessions per week) with a six week withdrawal block between the treatment blocks. Changes in the speech accuracy and consistency, phonological awareness, decoding, and spelling skills were analysed at both an individual and group level for the children with CAS.

7.2.4 Experiment 4: Follow-up study 6 months post intervention

The follow-up investigation re-evaluated the speech, phonological awareness, reading, and spelling skills of children with CAS six months following completion of the intervention programme. A measure of reading accuracy and reading comprehension in a text reading task was administered to the children with CAS. The children with typical speech and language development were also re-assessed at this time. The study aimed to establish if children with CAS could maintain any gains in spoken and written language ability achieved in the intervention, and if continued improvement in their phonological awareness, reading, and spelling skills relative to their peers with typical development was evident once intervention support had been removed.

7.2.5 Experiment 5: Case study of twin boys with CAS

This study evaluated the long-term effects of the integrated phonological awareness programme for identical twin boys who participated in the research intervention. These children were selected for more in-depth investigation for two reasons:
1. The development of identical twins that both have CAS has not previously been described in the literature; and

2. These two children received the phonological awareness intervention just prior to their commencement of formal schooling. Thus, the effects of the intervention on facilitating early literacy development could be carefully examined.

The study aimed to examine Theo and Jamie’s spoken language, phonological awareness, reading, and spelling development during their first year of schooling (i.e., six month and twelve month assessments post-intervention).

It will be argued from the findings reported in these experiments that children with CAS are at particular risk for persistent difficulties in phonological awareness, reading, and spelling and that an integrated phonological awareness intervention is effective in simultaneously advancing aspects of these children’s spoken and written language development.

7.3 Children with CAS: At risk for written language disorder

Evidence to support the argument that children with CAS are at particular risk for written language difficulties is provided from two sources: comparison with other groups and follow-up intervention effects.

7.3.1 Comparison with other groups

The first source of evidence that children with CAS are at particular risk is provided from the comparative group study reported in Experiment 1. A comparison of
the phonological awareness, letter knowledge, and reading skills of children with CAS, children with inconsistent speech disorder, and children with typical development revealed that children with CAS are particularly susceptible to phonological awareness and written language difficulties. The children with CAS exhibited poorer phonological awareness than the children with inconsistent speech disorder and typical development. Further, evaluation of the normative assessments revealed that children with CAS had a greater percentage of participants performing below the expected range for their age on letter knowledge, real word decoding, and phonological awareness normative measures. In addition, the school aged children with CAS showed significantly inferior performance than their peers with typical development on the non-word decoding measure. All of their scores on this task fell below the range of scores exhibited by their peers with typical development, indicating a severe difficulty in decoding novel words.

Performance on phonological awareness and letter knowledge are powerful predictors of early literacy success, and the ability to use phonological information in the decoding process and reading performance itself is a strong predictor of later reading success (Hogan et al., 2005). It can be concluded, therefore, that as all 12 children with CAS in this study demonstrated poor phonological awareness, and that the school aged children in the study also demonstrated poor word decoding of both real and non-words, children with CAS are at high risk for persistent written language difficulties.

A comparison of the three groups’ performance in a receptive phonological representation task indicated that the two groups with speech disorder performed inferiorly to the children with typical development. There was no difference, however, in the scores of the children with CAS and children with inconsistent speech disorder on the
phonological representation task. The differing profiles of the children with CAS and children with inconsistent speech disorder in the study suggested that the poor performance of the children with inconsistent speech disorder in the phonological representation task may have been due to an impairment in phonological assembly rather than a deficit in phonological representation ability (Dodd, 1995). The pattern of results from the children with CAS, however, suggested a deficit in phonological representation ability.

These findings provide preliminary support for the phonological representation deficit hypothesis of CAS (Marquardt et al., 2002; Marion et al., 1993). The assessment findings showed that children with CAS exhibited poor phonological awareness in comparison to children with speech disorder and children with typical development who had comparable receptive vocabulary ability. The results thus extend previous research that reported phonological awareness deficits in children with CAS in comparison to children with typical development who were not matched for receptive vocabulary skills (Marquardt et al., 2002; Marion et al., 1993). The assessment findings also demonstrated that children with CAS perform poorly on a receptive task designed to tap phonological representation ability compared to their peers with typical speech-language development. The demonstration of impaired performance on receptive phonological awareness and phonological representation tasks is compatible with the phonological representation hypothesis of CAS. It is difficult to explain the children’s performance in these measures within a purely motoric view of the disorder.

Further investigation is required to determine whether the phonological representation skills of children with CAS are different to children with other types of
speech or language impairments. The current findings indicated that the children with CAS and children with inconsistent speech disorder presented with similar phonological representation skills. The task employed, however, may not have been sensitive enough to differentiate the performance of the children with CAS and children with inconsistent speech disorder. It is critical that tasks are developed that differentiate phonological assembly and phonological representation in children with speech disorder. Such research will help to determine if children with CAS have a particular deficit in phonological representation.

7.3.2 Follow-up intervention effects

The second source of evidence that children with CAS are at particular risk for written language difficulties is provided from the follow-up intervention study reported in Experiment 4. Although as a group the children with CAS showed rapid and accelerated development during the five month intervention period, they showed remarkably little continued growth in their reading and spelling ability in the six month period following intervention. Importantly, their skill level did not drop back to pre-intervention levels when the experimental intervention was discontinued. The lack of continued growth, however, given ongoing daily classroom instruction in reading and spelling (and additional language support for some participants) suggests these children remain at risk and may need periodic intensive interventions to help integrate newly acquired knowledge more fully into written language activities.
7.4 Phonological awareness treatment effectiveness

Evidence to support the argument that an integrated phonological awareness intervention is effective in simultaneously advancing aspects of spoken and written language development in children with CAS is provided from three main sources which are described in the following section.

7.4.1 Longitudinal pilot study

The first source of evidence of the potential benefits of an integrated approach to phonological awareness intervention for children with CAS is provided in the longitudinal pilot study reported in Experiment 2. Derek and Katie maintained their accurate (92% to 100% respectively) speech performance for their targeted phonological error pattern 12 months after their participation in a short intensive intervention period (seven hours over a three week period). Katie also improved her production of words containing her control phonological error pattern over the 12 month follow-up period, and maintained her performance in the trained and untrained phoneme awareness probes from post-intervention to follow-up assessment.

The children performed superiorly on a standardised phonological awareness measure at follow-up than at pre-intervention assessment, and their decoding accuracy performance was within the expected range for their age on a connected reading measure at follow-up assessment.

There was a limited effect of the programme, however, on performance in a normative real word decoding measure, and Derek demonstrated little transfer to untrained speech words containing his untargeted phonological error pattern at follow-up assessment. Derek’s rapid improvement in phoneme awareness during the intensive
intervention was not maintained. Further, although there was a sharp increase in the ability to use phonological information in decoding (as measured by the non-word decoding task) during the intervention period, minimal additional gains were evident on this measure over the 12 month follow-up period.

7.4.2 Effectiveness of an integrated phonological awareness approach

Experiment 3 (described in Chapter 4) provides the second and main source of evidence supporting the argument that an integrated phonological awareness intervention is an effective means of facilitating speech, phonological awareness, and written language development in children with CAS. Analysis of the speech repeated measures showed that the majority of the 12 participants improved in the accuracy of the trained and untrained words for both targeted speech error patterns. Some generalisation of speech targets to connected speech was also observed for the majority of children. There were no gains in the control language probe over the intervention period for participants, indicating that the observed improvement in speech development could be attributed to the intervention and not general maturation alone.

The ability of the intervention to provoke rapid change in the children’s speech production skills is in contrast to reports that children with CAS respond slowly to speech-language therapy approaches (Campbell, 1999; Pannbacker, 1998, Hall, 1989). Further, the transfer of speech gains to untrained words and a connected speaking context for the majority of participants is contrary to reports that children with CAS have difficulty generalising treatment goals (e.g., Velleman, 2003). Traditionally speech intervention targeting phonological error pattern suppression has been contraindicated for children with CAS due to the conventional view of CAS as a motor speech disorder (e.g.,
for review see Pannbacker, 1988). The current results support the use of a phonological intervention approach for children with CAS if the child presents with delayed or disordered phonological error patterns. It is, however, unlikely that phonologically based speech intervention will be the only intervention technique employed for a child with CAS given the diverse nature of the symptoms associated with the disorder.

Analysis of the benefits of the intervention at a group level showed that the children with CAS made a significant improvement in their speech accuracy, articulation consistency, phonological awareness, non-word reading, and spelling skills throughout the intervention. Further, the children with CAS exhibited greater relative gains in these areas compared to their peers with typical speech-language development over the intervention period.

The follow-up investigation undertaken in Experiment 4 (described in Chapter 5) indicated that children with CAS were able to maintain the gains in spoken and written language made during the intervention period. The children did not, however, continue to experience sustained accelerated growth in these skills. There was no difference in any of the speech, phonological awareness, or literacy measures in the post-intervention assessment and the follow-up assessment six months later. Further, there was no difference in the comparative change in these skills between the children with CAS and children with typical development over the follow-up period. The lack of sustained development over the follow-up period provided further evidence that the gains made over the intervention period could be attributed to the intervention and not sporadic linguistic development in the participants. It is essential, however, that strategies are
developed to ensure ongoing growth in targeted areas for children with CAS following participation in the integrated intervention.

7.4.3 Longitudinal case studies of twin boys

A third source of evidence supporting the effectiveness of the experimental intervention is provided from a longitudinal intervention case study of twin boys (Experiment 5 reported in Chapter 6). Theo and Jamie, who participated in the intervention study just prior to their commencement of formal schooling, demonstrated typical phonological awareness, reading, and spelling development throughout their first year of literacy instruction. They also demonstrated a large gain in speech accuracy and improved articulatory consistency over the study. These findings suggest that providing phonological awareness intervention to preschool children with CAS within a preventative model may be particularly beneficial in enhancing their early literacy development.

7.4.4 Summary of evidence supporting the effectiveness of the experimental intervention

The results of the intervention studies have important implications for the potential to improve the written language outcomes of children with CAS. It is possible to facilitate rapid growth in the phonological awareness, reading, and spelling skills of children with CAS. Further, the focus on phonological awareness and literacy skills is not at the expense of improving speech production skills in those affected. Indeed, the combination of representational and targeted speech practice components in the intervention may accelerate the children’s speech production development.
It can be concluded from the intervention experiments that:

1. An integrated phonological awareness intervention is an effective method of stimulating speech and phonological awareness development in most children with CAS.

2. Children with CAS are able to transfer their improved phonological awareness skills to the reading and spelling process.

3. The intervention gains from an integrated phonological awareness programme are able to be maintained over time by children with CAS.

4. Older children with CAS may need ongoing support to ensure sustained phonological awareness, reading, and spelling development following participation in the integrated programme.

7.5 Broader theoretical implications of the findings

The results from this thesis add to the research body demonstrating the positive effects of phonological awareness for reading and spelling development (Ehri et al., 2001). In line with the models of word recognition and spelling development described in Chapter 1, the children with CAS were able to use their improved phonological awareness skills to acquire an alphabetic strategy in decoding and encoding processes. There was a large gain in the children’s non-word reading performance, and the majority of children progressed to the semi-phonetic or phonetic stage of spelling development (Ehri, 2000). The gains in non-word reading and spelling were dramatic considering that the majority of the school-aged children had received at least two years of formal literacy instruction prior to participation in the programme. Despite the children’s history of severe written language impairment, they were able to make rapid gains in non-word
reading and spelling. Further, the children were able to utilise their improved decoding skills in a connected reading task.

The findings provide further evidence of the benefit of simultaneously targeting speech, phonological awareness, and letter knowledge skills for children with speech disorder (Gillon, 2000, 2002, 2005). Children with speech disorder are, as a group, at heightened risk of phonological awareness, reading, and spelling delay (Bird, Bishop, & Freeman, 1995; Nathan, Stackhouse, Goulandris, & Snowling, 2004; Raitano, Pennington, Tunick, Boada, & Shriberg, 2004; Rvachew, Ohberg, Grawburg, & Heyding, 2003). The use of integrated goals for this population is an effective and efficient means of improving the written and spoken language outcomes of children with speech disorder. It is essential, however, to ensure that the content of the integrated phonological awareness programme and the intensity of the phonological awareness intervention are appropriate. Some investigations examining the effectiveness of integrated approaches for children with speech disorder have spent less therapy time on phonological awareness activities or targeted phonological awareness skills in a less explicit manner (Hesketh, Adams, Nightingale, & Hall, 2000; Rvachew, Nowak, & Cloutier, 2004). Consequently, these integrated studies reported more muted treatment effects for phonological awareness development in children with speech disorder. It is also essential that integrated programmes retain the general treatment principles guiding the effectiveness of phonological awareness interventions. Integrated phonological awareness programmes must focus at the phoneme rather than the rhyme level to ensure the strongest impact on reading and spelling skills (Muter et al., 2004; Nancollis et al., 2005). Letter-sound training must also be included within an integrated phonological awareness approach.
These guiding principles remain applicable to children receiving the intervention within a preventative model (i.e., within the preschool years). Consistent with previous work, the current study showed that it is possible to stimulate early phoneme level skills (i.e., initial phoneme identify) and letter-sound knowledge in children as young as four years with speech disorder (Gillon, 2005; Hesketh, 2007).

The ability of an integrated phonological awareness approach to create rapid gains in children’s phonological awareness, reading, and spelling skills provides further evidence for the importance of visual-verbal paired associate learning in literacy development. Hulme et al. (2007) reported that aptitude in visual-verbal learning (i.e., learning to pair a particular shape with a particular verbal response) is a unique predictor of later reading skills. The integrated phonological awareness incorporates visual-verbal paired associated learning in its letter-sound knowledge activities and speech prompting techniques. The letter-sound activities included in the programme prompt the child to articulate the sound associated with a particular letter. When the child makes a speech error, he/she is cued by the written word to aid speech accuracy. The integrated approach thus targets the development of two unique contributors to reading success (i.e., phoneme awareness and visual-verbal paired associated learning).

7.6 Clinical implications

The clinical implications from this thesis are significant. The study provides further evidence that intervention for children with CAS should not be limited to the remediation of verbal motor control difficulties. It is important that phonological awareness and written language deficits are included within treatment approaches for children with CAS. The findings show that an integrated phonological awareness
programme is an effective means of targeting speech production, phonological awareness, letter knowledge, reading, and spelling skills in children with CAS. The results are particularly important considering previous research that has shown that speech production difficulties in CAS tend to minimise over time while language, reading, and spelling deficits persist (Lewis, Freebairn, Hansen, Iyengar et al., 2004).

The findings of the follow-up study indicate that it is critical to monitor the written and spoken language development of children with CAS following participation in an integrated phonological awareness programme. Re-assessment of the children with CAS six months following participation in the research intervention showed that many of the children did not continue to make gains in phonological awareness, speech, and literacy measures over the follow-up period. The findings suggest that older children with CAS may require ongoing speech-language therapy support to maintain growth in phonological awareness, reading, and spelling skills.

The study also provides preliminary evidence that the application of an integrated phonological awareness approach may be more effective within a preventative intervention model. Three children with CAS who received the research intervention prior to the onset of formal literacy instruction proceeded to maintain age-appropriate phonological awareness, letter knowledge, and decoding skills in their first year of school. Although the benefits of early intervention must be weighed up against the diagnostic issues around identifying children with CAS at a young age (Davis & Velleman, 2000; Lewis, Freebairn, Hansen, Iyengar, & Taylor, 2004; Velleman, 2003), providing phonological awareness intervention for children with CAS within a
preventative framework may be the most optimal use of clinical resources (e.g., Gillon, 2005).

The results also suggest that expressive morpho-syntactic deficits in children with CAS need to be targeted explicitly in intervention programmes for children with CAS. Theo and Jamie (presented in Chapter 6) exhibited persistent morpho-syntactic impairment that did not resolve in line with the children’s gains in speech production and phonological awareness. The inclusion of morpho-syntactic goals within the speech-language therapy programme of children with CAS is pertinent given the importance of morphological ability for reading comprehension and later decoding and spelling development (Muter, Hulme, Snowling, & Stevenson, 2004; Singson, Mahony, & Mann, 2000).

There has been a widespread impetus for SLPs to incorporate evidence based practice into clinical work which has been difficult to achieve for children with CAS given the lack of intervention studies for this population. The current study is a first step in demonstrating treatment effectiveness for a relatively large number of children with CAS compared to previous studies via a controlled intervention design. The data also indicated that the integrated approach is an efficient use of intervention resources, with participants making rapid gains in speech and phoneme awareness skills.

7.7 Limitations of the current research

The comparison of children with CAS and children with inconsistent speech disorder presented in Chapter 2 was limited by the use of raw scores in some analyses and the content of the phonological representation task. Although there was no significant difference in the ages of the groups, the CAS group had more 7 year old participants than
the inconsistent group. The raw scores thus may not have provided a fair comparison of the reading and letter knowledge abilities of both groups, given the variability in exposure to formal reading instruction. The phonological representation task was originally developed for children aged 4 to 5 years (Sutherland & Gillon, 2006) and may not have been sensitive enough to detect differences in the phonological representation abilities of the children with CAS and inconsistent speech disorder.

The simplicity of the stimuli used in the phonological representation task also made it difficult to track the development of phonological representational ability in children with CAS over time. It was thus difficult to examine the children’s ongoing development in phonological representation skills in response to the research intervention. For example, Theo and Jamie exhibited a large gain in the phonological representation measure over the intervention period, but performed at near-ceiling level on the task for the remainder of the study. It is important that future studies evaluating the effects of phonological awareness training on phonological representation skills incorporate developmentally appropriate tasks.

The results of the study are limited by the relatively small sample size employed. The limitation of small sample sizes in research involving children with CAS, however, is difficult to avoid given the low incidence rate of CAS when applying strict inclusionary criteria. To the author’s knowledge, the current project involved the largest number of children with CAS to date involved in an intervention study.

The findings suggest that the research intervention may not be universally beneficial for children with CAS. Although the programme was successful for the majority of participants, a significant number of children did not show improvement in all
speech and phonological awareness repeated measures. Seven out of the twelve children improved the accuracy of trained and untrained speech repeated measures for both targeted phonological error patterns. Eight out of the twelve children improved in both their trained and untrained phonological awareness repeated measures for both targets. Further investigation is required to determine the percentage of children with CAS for whom the intervention is successful.

7.8 Future directions

The findings of this thesis highlight many future directions for CAS research. It is important to compare the phonological representation and phonological awareness ability of children with CAS to children with other types of speech disorder and children with reading disorder. Such comparisons will delineate whether children with CAS present with a more severe deficit in phonological awareness or phonological representation than these groups. This analysis is important for refining the phonological representation deficit hypothesis of CAS and will provide further information regarding the association between phonological representation, phonological awareness, and speech production skills in children with developmental speech and language disorder.

The integrated phonological awareness approach for children with CAS requires ongoing investigation. It is important that the current findings are replicated on other children with CAS within a controlled research design. The current results also suggest that it may be valuable to target a greater number of speech goals within the 12 week timeframe. The main intervention study reported in Chapter 4 targeted two phonological error patterns per child over the intervention. The chosen rotation of speech targets was slower than suggested in the cycles phonological intervention approach for other types of
speech disorder (Hodson & Paden, 1991), as the literature indicated children with CAS progressed slowly in therapy (e.g., Hall, 1989). However, session worksheets demonstrated that many children in the current study suppressed target phonological process in relatively short periods (i.e., three to four weeks). Similarly, the two children involved in the pilot study reported in Chapter 2 achieved high accuracy on words containing their targeted phonological error patterns following three weeks of intervention. It is important that future research investigates whether it is possible to target a greater number of speech goals over the 12 week intervention period to further increase intervention efficiency.

It is important that future investigations focus on methods to improve the long-term effects of the integrated phonological awareness programme for older children with CAS. An extended intervention programme may create sustained growth in phonological awareness, reading, and spelling skills for older children with CAS. Further, a trained generalisation phase, where children are supported in using phonological strategies in reading and spelling within the classroom environment, may be beneficial for the long-term effects of the intervention approach for children with CAS.

The application of an integrated phonological awareness approach within a preventative framework requires additional examination. The current findings suggest that children may be more likely to demonstrate continued growth in phonological awareness following participation in the research intervention if they receive the intervention prior to school entry. Three children within the current sample who participated in the integrated approach in the preschool years presented with typical phonological awareness and literacy development in their first year of school. It is
critical, however, that the value of an integrated approach administered within the preschool years is evaluated on a larger number of children. The two youngest children in the current sample presented with the most severe speech deficit and did not improve their phonological representation ability in response to the intervention. The early reading and spelling development of these children thus needs to be carefully monitored. It is also important that the response of children with CAS to a preventative phonological awareness intervention is followed into the later school years. This is particularly important given the ongoing morpho-syntactic deficits experienced by children with CAS that may hinder reading comprehension and later decoding and spelling development. It may be necessary to introduce some form of integrated morphological awareness intervention (i.e., targeting bound morpheme use and orthographical reading and spelling techniques) in later treatment cycles for these children.

The single-subject design employed within the intervention studies drew attention to children within the sample who did not benefit from the integrated intervention. It is important to identify factors that may preclude a child from responding favourably to an integrated phonological awareness approach. It is further critical that modifications to an integrated intervention or other therapeutic techniques are trialled with children who appear resistant to the current approach. The identification of methods to improve the speech, language, and literacy outcomes of children with complex communication difficulties such as CAS is an ongoing pursuit.

The connection between spoken and written language is well established (e.g. Catts, 1993). Intervention techniques that link spoken and written goals, however, are a relatively recent achievement. The findings presented in this thesis add to the growing
body of evidence indicating the effectiveness of phonological awareness approaches in fostering literacy and speech production development for children with speech disorder (Gillon, 2000, 2002, 2005). Integrated frameworks are a valuable means of targeting multiple skills that underlie speech, reading, and spelling success. Future development of integrated approaches may thus hold the key to efficiently and effectively advancing the spoken and written language of children with diverse linguistic deficits and ensuring that they have the opportunity to meet their potential within our highly literate society.
REFERENCES


intervention with special populations (pp. 110-139). New York: Thieme Medical Publishers.


APPENDIX A

The non-word reading task (Calder, 1992)

Section 1
Vab, kos, sim, dup, mov, tob, zug, hud, tiz, sep

Section 2
Plob, bling, bruch, trock, twud, cliz, thrad, whan, gluff, swek

Section 3
Feen, poy, zie, hoob, yoat, mape, roit, gice, pute, lawp
APPENDIX B

Example demonstration of phonological error pattern targets, speech probes, and phoneme awareness probes for one child from the CAS group

**Target phonological error pattern 1:** Velar fronting (to be targeted in the first intervention block)

*Speech probes:* Child is required to name the word in response to a picture.

*Phoneme awareness probes:* Child is required to segment the above words into their constituent phonemes using coloured blocks.

<table>
<thead>
<tr>
<th>Trained probes</th>
<th>Untrained probes</th>
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<tbody>
<tr>
<td>Car</td>
<td>Cow</td>
</tr>
<tr>
<td>Coo</td>
<td>Cap</td>
</tr>
<tr>
<td>Can</td>
<td>Corn</td>
</tr>
<tr>
<td>Cup</td>
<td>Book</td>
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<tr>
<td>Coin</td>
<td>Nick</td>
</tr>
<tr>
<td>King</td>
<td></td>
</tr>
<tr>
<td>Back</td>
<td></td>
</tr>
<tr>
<td>Neck</td>
<td></td>
</tr>
<tr>
<td>Bike</td>
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<td>Pack</td>
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</tbody>
</table>

**Target phonological error pattern 2:** S-cluster reduction (to be targeted in the second intervention block)

*Speech probes:* Child is required to name the word in response to a picture

*Phoneme awareness probes:* Child is required to segment the above words into their constituent phonemes using coloured blocks.

<table>
<thead>
<tr>
<th>Trained probe</th>
<th>Untrained probe</th>
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</thead>
<tbody>
<tr>
<td>Star</td>
<td>Sleigh</td>
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<tr>
<td>Slow</td>
<td>Stem</td>
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<td>Step</td>
<td>Sleep</td>
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<tr>
<td>Stan</td>
<td>Spot</td>
</tr>
<tr>
<td>Stone</td>
<td></td>
</tr>
<tr>
<td>Slip</td>
<td></td>
</tr>
<tr>
<td>Slide</td>
<td></td>
</tr>
<tr>
<td>Spin</td>
<td></td>
</tr>
<tr>
<td>Spoon</td>
<td></td>
</tr>
<tr>
<td>Spit</td>
<td></td>
</tr>
</tbody>
</table>
APPENDIX C

Examples of some of the materials used in the phonological awareness activities

*Speech cards*

- **bee**
- **car**

Used to prompt speech (by pointing to the appropriate letter) and to play games

*Phoneme identity character*

Crunching crocodile wants to eat all the words that start with a /k/ sound

*Phoneme segmentation and blending board for s-clusters*

Target words are slot, slop, spoon, step, spit, and stop
APPENDIX D

The individual graphs demonstrating changes in trained and untrained speech repeated measures

Participant 1
Trained speech measures

Untrained speech measures

Participant 2
Trained speech measures
Untrained speech measures

\[ y = 7.9x + 40.367 \]

Probes

Percent Phonemes Correct

Pre

Post

Linear (Pre)

Participant 3

Trained speech measures

\[ y = 1.85x + 51.94 \]

Probes

Percent Phonemes Correct

Pre

Post

Linear (Pre)

Untrained speech measures

\[ y = -3.55x + 57.13 \]

Probes

Percent Phonemes Correct

Pre

Post

Linear (Pre)
**Participant 4**

*Trained speech measures*

**Untrained speech measures**

**Participant 5**

*Trained speech measures*
Untrained speech measures

Participant 6
Trained speech measures

Untrained speech measures
Participant 7
Trained speech measures

Untrained speech measures

Participant 8
Trained speech measures
Untrained speech measures

Trained speech measures

Participant 9
Participant 10

Trained speech measures

Untrained speech measures

Participant 11

Trained speech measures
Untrained speech measures

![Graph showing untrained speech measures with equations and data points]

Trained speech measures

![Graph showing trained speech measures with equations and data points]

Participant 12

Untrained speech measures

![Graph showing untrained speech measures for Participant 12 with equations and data points]
The individual graphs demonstrating changes in trained and untrained phoneme awareness repeated measures

**Participant 1**
*Trained phoneme awareness measures*

![Graph](image1)

*Untrained phoneme awareness measures*

![Graph](image2)

**Participant 2**
*Trained phoneme awareness measures*

![Graph](image3)
Untrained phoneme awareness measures

Participant 3
Trained phoneme awareness measures

Untrained phoneme awareness measures
Participant 4

*Trained phoneme awareness measures*

Untrained phoneme awareness measures

Participant 5

*Trained phoneme awareness measures*
Untrained phoneme awareness measures

![Graph](image1)

Participant 6
Trained phoneme awareness measures

![Graph](image2)

Trained phoneme awareness measures

![Graph](image3)
**Participant 7**  
*Trained phoneme awareness measures*

**Untrained phoneme awareness measures**

**Participant 8**  
*Trained phoneme awareness probes*
Untrained phoneme awareness measures

Participant 9
Trained phoneme awareness measures

Untrained phoneme awareness measures
Participant 10

Trained phoneme awareness measures

Untrained phoneme awareness measures

Participant 11

Trained phoneme awareness measures
**Untrained phoneme awareness measures**

![Graph 1](image1)

**Trained phoneme awareness measures**

![Graph 2](image2)

**Participant 12**

**Trained phoneme awareness measures**

![Graph 3](image3)

**Untrained phoneme awareness measures**

![Graph 4](image4)