Rapid Syllable Transition Treatment for Childhood Apraxia of Speech: Exploring Treatment Efficacy in Three Service-Delivery Contexts

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Supervisor's Statement

This is to certify that the thesis entitled 'Rapid Syllable Transition Treatments for Childhood Apraxia of Speech: Exploring Treatment Efficacy in Three Service-Delivery Contexts' submitted by Donna Claire Thomas in fulfilment of the requirements for the degree of Doctor of Philosophy is in a form ready for examination. It does not exceed the word limit prescribed for the degree.

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Statement of Authorship

This is to certify that the content of this thesis is my own work. This thesis has not been submitted for any other degree at The University of Sydney or elsewhere. No other person's work has been used without due acknowledgement in the thesis. Approval for these studies was given by The University of Sydney Human Ethics Committee (Reference numbers: 2012/710; 2012/2824; 2014/080).

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Publications and Presentations Arising From This Thesis

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Notes on Style

Spelling

This thesis includes publications; therefore, four chapters previously published as journal articles appear here in the style of the journals in which they were published. These journal articles form Chapters 3, 4, 5 and 7 of this thesis.

- Chapter 3, journal article 1, entitled 'Rapid Syllable Transitions (ReST) treatment for childhood apraxia of speech: The effect of lower dose-frequency', uses American English in accordance with the instructions for authors for *the Journal of Communication Disorders.*
- Chapter 4, journal article 2, entitled 'Telehealth delivery of Rapid Syllable Transition (ReST) treatment for childhood apraxia of speech', uses British English in accordance with the instructions for authors for the *International Journal of Language and Communication Disorders.*
- Chapter 5, journal article 3, entitled 'Combined clinician–parent delivery of rapid syllable transition (ReST) treatment for childhood apraxia of speech' uses British English in accordance with the instructions for authors for the *International Journal of Speech-Language Pathology.*
- Chapter 7, journal article 4, entitled 'Parent experiences of variations in service delivery of Rapid Syllable Transition (ReST) treatment for childhood apraxia of speech' uses British English in accordance with the instructions for authors for *Developmental Neurorehabilitation.*

The remainder of the thesis uses Australian English.

Style and terminology

The journal articles included in this thesis (i.e., Chapters 3, 4, 5 and 7) are in the styles specified in the relevant journal guidelines.

Unless otherwise indicated, the terms ‘speech pathologist’ and ‘speech pathology’ are used within the thesis to describe the professional worker and the profession respectively, as these are the terms used in Australia. Alternative terms are used when international research is described. The term ‘speech-language pathologist’ (SLP) is used in Table 2.1 because the majority of the articles in the table are North American and use that term.

The term ‘childhood apraxia of speech’ (CAS) is used within this thesis. Despite some theoretical differences between the terms, there is little clinical difference between children with CAS and those with ‘developmental verbal dyspraxia’ (DVD).

List of Shortened Forms

AAC	Augmentative and alternative communication
CAS	Childhood apraxia of speech
CELF-4	Clinical Evaluation of Language Fundamentals—fourth edition
CELF-P2	Clinical Evaluation of Language Fundamentals—Preschool-2
DTTC	Dynamic temporal and tactile cueing
E^3BP	Evidence-based practice, considering external evidence, internal evidence and client and/or carer preference
EBP	Evidence-based practice
EPG	Electropalatography
ES	Effect size
ICF-CY	International Classification of Functioning—Children and Youth
IMI	Integrated multimodal intervention
IPA	Integrated phonological awareness
MIT	Melodic intonation therapy
NDIS	National Disability Insurance Scheme
NDP-3	Nuffield Dyspraxia Programme – third edition
PPVT-4	Peabody Picture Vocabulary Test—fourth edition
PROMPT	Prompts for Restructuring Oral Muscular Phonetic Targets
ReST	Rapid syllable transition
SCED	Single-case experimental design
SGD	Speech-generating device
SLP	Speech-language pathologist
SSD	Speech sound disorder
SLTA	Speech-language therapy assistant
TCM	Touch cue method
UK	United Kingdom
US	Ultrasound
vs.	Versus

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Abstract

Many children are unable to access speech pathology treatment at the recommended intensity. To address this problem, clinicians use a range of strategies: modifying treatment intensity, mode or delivery agent.

Accessing speech pathology treatment for children with childhood apraxia of speech (CAS) is particularly difficult because treatment should be delivered face-to-face by a clinician 3–5 times per week. One relatively new treatment for CAS, rapid syllable transition (ReST) treatment has desmonstrated significant treatment and generalisation effects when delivered instensively, face-to-face, by a clinician.

This thesis uses three separate single-case experimental studies to investigate the efficacy of ReST treatment when provided via alternative service-delivery approaches. Lower dose-frequency, telehealth delivery, and a combined clinician–parent delivery model were explored. The studies showed that both lower dose-frequency and telehealth delivery were efficacious. Combined clinician–parent delivery was efficacious for fewer than half the children.

Parental experiences of telehealth and of the combined clinician–parent delivery models were investigated qualitatively. The parents reported positive experiences of telehealth, finding it convenient and time-efficient. They had concerns about the combined clinician-parent delivery model, reporting discomfort in the role of therapist, and low levels of confidence and competence in delivering treatment.

This thesis supports implementation of both lower dose-frequency and telehealth delivery of ReST treatment. Despite the intuitive appeal of parent-delivered treatment for overcoming barriers to access, this thesis does not support clinical application of parent-delivered ReST treatment. This thesis argues for further investigation of intensity variables in CAS treatment and methods for improving parent-

delivered treatment efficacy, and the need to advocate on behalf of clients to ensure sufficient service provision.

Chapter 1: Speech Pathology Service Delivery for Childhood Speech Impairments

For most people, speech is taken for granted, a means of quickly and efficiently communicating. Their speech develops with little conscious effort, such that by school-age they are speaking clearly and intelligibly and can participate in a range of social and academic endeavours (McLeod & Baker, 2017). However, for some people, speech does not develop effortlessly. Their speech is less clear than that of their peers, and they may have difficulty communicating with unfamiliar people and participating in life activities. The potential ramifications for these people extend beyond communication to educational attainment, economic achievement and psychosocial adjustment across the lifespan.

1.1 The context

1.1.1 Speech sound disorders need treatment

Children who have difficulty producing clear and intelligible speech are a heterogeneous group. Their speech difficulty can result from a number of impairments, some of which have a known origin, such as a structural or neurological difficulty, but many have no known origin. The umbrella term 'speech sound disorder' (SSD) can be used to describe all childhood speech production difficulties, regardless of origin (Shriberg et al., 2010). The term is most frequently used for articulation and phonological difficulties, but it can also include motor speech impairments, such as childhood apraxia of speech (CAS) and dysarthria (McLeod & Baker, 2017). The literature examining the consequences of speech impairment is more extensive for SSD than for specific impairments, such as CAS; therefore, this chapter initially explores the implications of SSD in general, rather than CAS specifically.

Although the reported prevalence of SSD varies (McLeod & Baker, 2017), SSD is reasonably common in childhood. Prevalence estimates for SSD in childhood range from 2.3% to 24.6% (Law, Dennis, & Charlton, 2017), with Australian population samples indicating that approximately three in 20 preschool-aged children have speech sound difficulties (McLeod, Harrison, McAllister, & McCormack, 2013). Even though not all children with SSD are referred

for speech pathology treatment, children with SSD make up a large proportion of speech pathology caseloads, with Australian speech pathologists reporting that nearly half their caseload consists of children with SSD (McLeod & Baker, 2014).

Having SSD during the preschool years affects more than communication. It places children at increased risk of other communication difficulties, such as language impairments and literacy difficulties (Eadie et al., 2015), and can cause difficulty in interpersonal interactions (McCormack, McLeod, McAllister, & Harrison, 2010). Compared with peers without SSD, preschool children with SSD are likely to have more difficulty with interpersonal interactions (McCormack, McLeod, Harrison, & McAllister, 2010) and poorer pre-literacy skills (Larrivee & Catts, 1999). Preschool-aged children with SSD have more preserved self-esteem than school-aged children with the condition (McCormack, McLeod, McAllister, et al., 2010). This is partly because preschool aged children attribute communication breakdown to their communication partner not listening properly, rather than to they themselves not speaking properly (McCormack, McLeod, McAllister, et al., 2010).

The outward signs of SSD resolve for approximately half the diagnosed children by the time they enter school (Shriberg, Kwiatkowski, & Gruber, 1994). The children least likely to have their speech errors resolve are those with sound distortions (Gruber, 1999), such as is common in CAS and dysarthria. It is this group of children, with unresolved SSD at school entry, who are most affected by the reported educational, vocational and social consequences of SSD (Shriberg, Gruber, & Kwiatkowski, 1994). However, even children who have no outward signs of speech impairments at school entry often show significant differences later in life from those who never had SSD (Felsenfeld, Broen, & McGue, 1992; Johnson, Beitchman, & Brownlie, 2010; Lewis et al., 2015).

The consequences of having SSD become more pronounced during the school years. At this stage, children with unresolved SSD frequently experience difficulties with educational attainment and social and psychosocial adjustment. Academically, they perform lower than their peers in reading,

writing, mathematics and general school achievement (Anthony et al., 2011). These children require more remedial support and more of their classroom teacher's time (Daniel & McLeod, 2017; Felsenfeld et al., 1992), and they participate less than their peers in group discussions and oral presentations (Daniel & McLeod, 2017; McAllister, McCormack, McLeod, & Harrison, 2011).They typically complete fewer years of schooling than other children (Felsenfeld et al., 1992). Children with SSD have poorer peer relationships, are more likely to be lonely, and more frequently experience bullying than peers without SSD (McAllister et al., 2011). At this stage of life, they have sufficient cognitive skills to identify their role in communication breakdown, and this contributes to feelings of low self-esteem and sadness (McLeod, 2006). In a longitudinal study of 170 children with speech and language difficulties at age 4–6 and 146 unaffected siblings, Lewis et al. (2015) revealed that the difficulties associated with having childhood SSD persisted through to adolescence, both for those whose SSD appeared to have resolved and for those with ongoing SSD. Adolescents with no outward signs of SSD had poorer skills than their unaffected siblings in producing polysyllabic words, reading and spelling. Those with persisting SSD had difficulties with polysyllabic words, reading and spelling in addition to speech and prosodic errors in conversational speech, poor non-word repetition and limited vocabulary (Lewis et al., 2015).

Adults with SSD are likely to have significant psychosocial adjustment difficulties and lower vocational outcomes than peers without SSD. The social interaction difficulties that have plagued this group since preschool persist (McCormack, McLeod, McAllister, et al., 2010) and may become more serious, in some cases leading to significant mental health conditions (Carrigg, Baker, Parry, & Ballard, 2015). This may be partly due to other people's negative perceptions of people with SSD, such as regarding them as less intelligent and less employable than people without SSD (Allard & Williams, 2008; Silverman & Falk, 1992; Silverman & Paulus, 1989). Adults who had childhood SSD are less likely to complete a university degree (Felsenfeld, Broen, & McGue, 1994) and are more likely to have semiskilled or unskilled jobs (Felsenfeld et al., 1992) and be in a lower socioeconomic group (Johnson et al., 2010) than their unaffected siblings and

peers. They are also more likely to pass on a communication difficulty to their children than are adults with no history of SSD (Felsenfeld et al., 1994).

Given the serious implications of SSD throughout the lifespan, it is vital that people with these conditions receive timely, effective and sufficient treatment.

1.1.2 Treatment is efficacious

Speech pathology treatment for children with SSD has been found to be effective (Baker & McLeod, 2011; (Law, Garrett, & Nye, 2004). A meta-analysis of 33 intervention studies for children with speech and language impairments revealed that intervention produces significantly greater improvements in speech production than no intervention (Law et al., 2004). In a seminal paper, Baker and McLeod (2011) conducted a narrative review of all studies on SSD interventions published between 1979 and 2009. The review covered 134 intervention studies across 46 distinct intervention approaches. Baker and McLeod concluded that it is better for children with SSD to have intervention than no intervention. Not only does intervention improve children's speech production, but it also helps ameliorate the associated educational, social and psychosocial difficulties (Baker & McLeod, 2011).

1.1.3 Not all children can access treatment

Unfortunately, not all children receive the necessary speech pathology treatment. Parents report a lack of speech pathology services (McAllister et al., 2011; O'Callaghan, McAllister, & Wilson, 2005) and long waiting lists for those services that are available (Hussain & Tait, 2015; Ruggero, McCabe, Ballard, & Munro, 2012). Demand for services has increased in the last decade due to factors such as the increased scope of speech pathology practice and greater community awareness of speech pathology services (SPA 2014a).

When children do receive treatment, there may be fewer sessions across a shorter time period than desired by parents (Ruggero et al., 2012). Parents frequently report that that their child would

have benefited from more treatment sessions and a shorter gap between sessions (Carroll, 2010; Glogowska & Campbell, 2000; Keilmann, Braun, & Napiontek, 2004). The limited service provision leads to dissatisfaction for parents; for example, only 25% of parents of children with communication difficulties in the United Kingdom (UK) were satisfied with the service their child received (Paradice & Adewusi, 2002). Speech pathologists report having large caseloads (Lim, McCabe, & Purcell, 2017; McLaughlin, Lincoln, & Adamson, 2008), which, combined with limited services, leads speech pathologists to 'ration' their services (Rvachew & Rafaat, 2014). Parents believe that the various systems used to decide who receives services leads to the service a child receives depending less on the child's need than on luck and the parent's ability to advocate on the child's behalf (Paradice & Adewusi, 2002). The phrase 'postcode lottery' has been used by parents (Bercow, 2008, p. 129) to describe these highly variable services.

The difficulty in ensuring children receive enough therapy is of concern to speech pathologists internationally (Keilmann et al., 2004; Kenny & Lincoln, 2012; Lim et al., 2017). For example, a questionnaire study of German logopaedists revealed that they considered their routine service provision sub-optimal and they wanted to provide treatment more frequently (Keilmann et al., 2004). Due to the pressure of large caseloads, American speech-language pathologists (SLPs) made decisions about session length and frequency that were inconsistent with what they considered best practice (Brandel & Loeb, 2011). Australian and Canadian speech pathologists reported feeling an ethical tension between their desire to provide effective services and the reality of large caseload sizes (Kenny & Lincoln, 2012; Lim et al., 2017), leading Australian speech pathologists to use metaphors such as 'scales' and 'war' to describe caseload management (Kenny & Lincoln, 2012).

The ethical tension experienced by speech pathologists is borne out by the data regarding recommended treatment and actual provided treatment. Children frequently receive less treatment than is recommended to improve their speech skills. For example, the most common frequency of paediatric speech pathology sessions in Australia is one to two times per month (Ruggero et

al., 2012), even though the evidence suggests that a frequency of two or more sessions per week is most effective (Baker & McLeod, 2011; Murray, McCabe, & Ballard, 2014). The discrepancy between community need and the availability of speech pathology services was of such concern that it was investigated by national governments in several counties (e.g., Australia (Speech Pathology Australia, 2014a; England, Bercow, 2008). Such reports make for sober reading, with the UK report concluding that the provision of services for children and young people with speech, language and communication needs were highly unsatisfactory and characterised by inconsistency, inequity and variability (Bercow, 2008).

1.1.4 Funding for speech pathology services

In Australia, there is a mixed pattern of public and private funding for speech pathology services. Publicly funded services for children are available through community health services and hospitals, primarily funded by the respective state health departments. In some states, children may receive speech pathology services through their school, with the funding then provided by the relevant state education department.

The Australian Government also funds programs for children with additional needs (e.g. Better Start, Australian Government, National Disability Insurance Scheme [NDIS], Australian Government). These programs are designed to enable parents to engage the services and provider(s) deemed most appropriate to meet their child's needs. Although these programs provide families with greater control over the services they access, children with SSDs in the absence of other disabilities are typically not eligible for funding under these programs.

In addition to the funding programs for people with more global developmental challenges, financial rebates are also available for people with isolated communication difficulties. These rebates assist with the cost of a limited number of private speech pathology sessions (or other allied health sessions) under specific Australian Government initiatives (e.g. Medicare Chronic Disease Managment, Australian Government, 2014) and/or through private health insurance

providers. Private health insurance covers for-profit and not-for-profit services and provides a rebate for a percentage of the cost of therapy, with small annual limits for allied health services. In the Australian model, private speech pathology practitioners provide services under a fee-for-service model; however, for children without government funding or private health insurance cover, parents are responsible for payment of fees in their entirety.

Within the Australian healthcare system, there are two broad reasons why children do not always access the recommended speech pathology services. The first is perceived need, and the second is the existence of barriers that reduce access to services.

1.1.5 Perceived need

To access speech pathology services, families must perceive a need for intervention. The health belief model (Carpenter, 2010; Rosenstock, 1990) describes the factors that influence whether someone seeks help for a health condition, such as a speech impairment. In this model, the client's perceptions of susceptibility and severity of the health condition combined with their perceptions about the benefits and negative consequences of the treatment influence the likelihood that they will seek treatment. This has been shown to be true for parents seeking assistance from a speech pathologist for childhood speech and language impairments (McAllister et al., 2011; McCormack, McAllister, McLeod, & Harrison, 2012; Skeat et al., 2014). For example, in a study of nearly 800 Australian 4-year-old children, only one-third of the 208 children with clinically significant communication difficulties received a speech pathology assessment in the following 12 months (Skeat et al., 2014). Within the sample, both under-servicing (not receiving help despite a clinical need) and over-servicing (seeking help in the absence of need) were present, with parent level of concern the most significant factor affecting help-seeking (Skeat et al., 2014).

1.2 Access barriers

There are three main barriers to services; these are (a) structural, (b) geographical, and (c) financial (Verdon, Wilson, Smith-Tamaray, & McAllister, 2011). These barriers are not mutually exclusive, and it is possible for interactions to occur between the barriers to accessing service.

1.2.1 Structural barriers

When parents do perceive a need for speech pathology services, they attempt to access appropriate providers. At this point, some families encounter structural barriers, such as organisational-level policies regarding eligibility for services (Carrillo et al., 2011). These policies perform a gatekeeper function, controlling the number of clients accessing the service. For people attempting to access publicly funded speech pathology services, specific requirements must be met regarding type and severity of communication impairment, presence or absence of concomitant conditions, referral agent, age and residential address (Skeat et al., 2014; Speech Pathology Australia, 2014a). Once families have successfully navigated these policies to gain access to a service with a speech pathologist, there may be further policies within the service itself regarding eligibility for ongoing treatment. These may include limits on the number of treatment sessions, enforced therapy 'blocks and breaks' and/or the application of age-related discharge criteria (Baker, 2010; Lim et al., 2017; Ruggero et al., 2012; Speech Pathology Australia, 2014a). These types of policies serve as a structural barrier to sufficient speech pathology intervention (Carrillo et al., 2011).

Most publicly funded services have waiting lists for both assessment and treatment, and sometimes, by the time a client comes to the top of the waiting list, they are too old to receive services (Ruggero et al., 2012; Speech Pathology Australia, 2014a). In other circumstances, an initial speech pathology assessment may reveal a concomitant condition that makes the client ineligible for treatment from that particular service; they then need to find another provider and possibly endure another waiting list (Speech Pathology Australia, 2014a). To further complicate

matters, most of the policies that serve as structural barriers are developed at a local, rather than state or national, level (Lim et al., 2017), making it harder for families to understand and navigate access to appropriate and adequate speech pathology services.

1.2.2 Geographical barriers

In a large, sparsely populated country such as Australia or Canada, the geographical barriers to accessing services are an issue. Children in rural and remote locations have more difficulty accessing speech pathology than children in metropolitan areas (McAllister et al., 2011; O'Callaghan, McAllister, et al., 2005; Verdon et al., 2011; Wilson, Lincoln, & Onslow, 2002). Several factors contribute to these access difficulties. These include the geographical distribution of speech pathologists, lack of publicly funded speech pathologists visiting or residing in certain areas, travel time and distance for families or clinicians, and limited private therapy options (McAllister et al., 2011).

Internationally, rural and remote areas have proportionally fewer speech pathologists than metropolitan areas. In Australia in 2014, the major cities had 25.9 speech pathologists per 100,000 population; inner regional and outer regional areas had 20.5 and 16.9 respectively; and remote and very remote areas had 12.7 and 5.9 respectively (Health Workforce Australia, 2014). Although 30% of the Australian population live in rural communities, only 4.5% of the of speech pathologists in Australia provide services in those areas (Lambier & Atherton, 2003). Not only are there proportionally fewer speech pathologists in rural areas, but in some cases these areas have the greatest need (McCormack & Verdon, 2015; O'Callaghan, McAllister, et al., 2005).

The distance to travel to services is a much greater consideration for rural families than for metropolitan clients. It is not uncommon for rural families to travel hundreds of kilometres to access speech pathology services (McAllister et al., 2011; Ruggero et al., 2012; Speech Pathology Australia, 2014a). The concept of distance decay (Eyles & Woods, 2014) is relevant for rural families accessing speech pathology. The term ‘distance decay’ denotes the relationship between

the distance to be travelled and the likelihood of someone accessing the service. The further someone needs to travel to access the service, the less likely they are to do so (Eyles & Woods, 2014). Distance decay has underpinned investigations of the maximum practical travel distance to access speech pathology services. The maximum distance proposed for accessing fortnightly service was 65 km (Wilson et al., 2002), and 50 km for a weekly service (Verdon et al., 2011). Using 50 km as the maximum travel distance for a weekly service, Verdon et al. (2011) determined that 30% of locations in Australia's two most populous states, New South Wales and Victoria, were outside the practical travelling distance for weekly speech pathology treatments. Although the distance to services is shorter in metropolitan areas, the travel time can sometimes exceed that which is practical for weekly sessions, meaning that geographical barriers can also affect people residing in metropolitan areas.

Travelling to speech pathology requires family members to spend time away from work or other duties (Theodoros, 2008; Wilson et al., 2002), and in some cases this becomes so burdensome that families elect to prioritise the needs of the family over the person requiring speech pathology (McAllister et al., 2011). Travelling to and from speech pathology appointments also comes at significant financial cost to families (McAllister et al., 2011; O'Callaghan, McAllister, et al., 2005; Wilson et al., 2002) and contributes to fatigue and inattention during treatment sessions (Dew et al., 2012). Families who live in remote areas also report hazards associated with travelling, which are unique to their geographical area, such as wildlife on the road or extreme weather conditions (Hussain & Tait, 2015).

Lastly, with regard to geographical barriers, there are fewer choices of speech pathology services in rural areas than metropolitan areas, in both the public and private sectors (O'Callaghan, McCallister, & Wilson, 2005; Speech Pathology Australia, 2014a; Wilson et al., 2002). Specialist services such as those for people with hearing impairment, autism or cerebral palsy are typically only available in metropolitan areas or large regional centres. Similarly, private speech pathology providers are mostly located in metropolitan areas and infrequently accessed by people living in

regional centres or rural and remote locations (Ruggero et al., 2012; Speech Pathology Australia, 2014a).

1.2.3 Financial barriers

Accessing a private practitioner's services may enable families to bypass structural barriers to speech pathology. However, such therapy comes at a financial cost. The government and private insurance rebates that partially offset the costs associated with private therapy for some eligible clients are limited.

Funding rebate systems vary internationally. However, because this thesis explores service delivery in Australia, the following section focuses on the financial cost of accessing speech pathology in Australia. Some services are fully government funded (e.g., NDIS), some are government subsidised (e.g., Chronic Disease Management), and others are funded by clients, potentially with partial rebates from private health insurers. One feature common to government subsidies and private health insurance rebates is that these sources of funding are limited and may not cover as many sessions as the client desires or is recommended (Speech Pathology Australia, 2014a). The burden of funding private speech pathology sessions has a disproportionate effect on people from lower socioeconomic areas, and, unsurprisingly, people from lower socioeconomic areas attend private speech pathology less frequently than those from higher socioeconomic areas (Ruggero et al., 2012; Speech Pathology Australia, 2014a).

For all families, there are indirect financial considerations associated with attending speech pathology appointments. Indirect costs arise due to missing work or other roles to attend appointments, and these costs can place significant pressure on families (Dew et al., 2012). For some families, these indirect costs mean that difficult decisions must be made about the priority of speech pathology intervention in relation to other family needs.

1.3 Strategies for ensuring equitable access

Within the confines of finite resources, speech pathologists strive for efficient, efficacious and equitable management of clients (Kenny & Lincoln, 2012; Lim et al., 2017). This leads them to explore creative ways of providing speech pathology services (Cirrin et al., 2010; Lim et al., 2017; Schooling, Venediktov, & Leech, 2010), such as changing the agent delivering the treatment or the mode of treatment. 'Service delivery' is the term used to describe how clinical services are organised from an activities perspective and covers aspects such as the frequency of appointments, the mode of delivery and the provider of intervention. Some of the variations in service delivery that may enhance access to speech pathology are explored in sections 1.3.1 - 1.3.3 below.

1.3.1 Manipulating dose parameters

One way for speech pathologists to provide services for more people is to modify dose parameters. These modifications are grouped into two broad categories: direct modifications to dose parameters, such as providing less-frequent sessions than recommended or desired, and indirect modifications, such as having administrative policies that enable 'gatekeeper' functions thereby limiting the number of clients receiving a service and/or the amount of service they receive. The gatekeeper function within speech pathology is also a type of structural barrier (see 'Structural barriers' earlier).

1.3.1.1 Direct dose-parameter modifications

In order to understand direct modifications to dose parameters, it is useful to explore the concept of treatment intensity. Treatment intensity has increasingly become of interest to speech pathologists (Baker, 2012; Kaipa & Peterson, 2016; Schmitt, Justice, & Logan, 2017) as awareness has grown of the critical effect and sometimes complex role that treatment intensity has on intervention outcomes (Kleim, 2013; Schmitt et al., 2017). Although the profession is still becoming familiar with the vocabulary associated with treatment intensity, a helpful framework

for understanding treatment intensity was proposed by Warren, Fey, and Yoder (2007). Their framework includes five intensity components: (a) dose, (b) dose form, (c) dose-frequency, (d) total intervention duration, and (e) cumulative intervention intensity. Dose is a measure of how many times the active ingredient of the treatment is delivered per session (e.g., the number of productions by a client). Dose form is the type of activity through which the dose is delivered. Dose form considers both context (e.g., individual or group) and activity type (e.g., drill play or recasts of grammatical errors). Dose-frequency is a measure of the frequency of the therapy and may be measured in number of sessions per week, month or school term (e.g., twice-weekly). Total intervention duration is a measure of the length of the treatment in weeks, months or school terms (e.g., 8 months). Cumulative intervention intensity is a measure of the overall intensity of the treatment and is calculated using the other intensity variables, as follows:

Cumulative intervention intensity = dose x dose-frequency x total intervention duration.

For example, the cumulative intervention intensity of a treatment where the dose form is speech production trials, with 70 trials per session across two sessions per week for 10 weeks would be 70 x 2 x 10 = 1,400 production trials.

Speech pathologists report that they modify aspects of intensity to manage large caseloads (Lim et al., 2017). At some level, it is easy to understand the appeal of such modifications. If a clinician provides shorter sessions, thereby decreasing dose, there is time to schedule more sessions each day and treat more people. Similarly, if the dose form is changed to group rather than individual treatment, more people receive treatment each session. Alternatively, discharging clients after a set number of sessions (i.e., reducing total intervention duration) enables new clients to receive treatment. If these types of modifications can be made without compromising treatment efficacy, speech pathologists can explore creative service-delivery solutions for higher numbers of clients. However, if modifying intensity variables leads to lower treatment efficacy, it may instead be a false economy.

1.3.1.2 Indirect dose-parameter modifications

Some speech pathologists indirectly reduce the intensity of intervention through service-level approaches to caseload management (e.g. Little & Grasselli, 2013; Pertile & Page, 2003). These approaches aim to improve client throughput but often function as a structural barrier (see 'Structural barriers'), thereby reducing total intervention intensity. One of these service-level approaches, the Essence model (Little & Grasselli, 2013), is used in a specific geographical area of Australia. Under this model, parents are responsible for initiating all contact with clinicians, and clinicians have predetermined appointment slots available in their diaries for specific appointment types (e.g., assessment or therapy). Parents are permitted to book appointments at predetermined times only. If the predetermined appointment slots are unsuitable for families, they need to contact the speech pathologist in 8–12 weeks, during the next available appointment window. Unsurprisingly, this system decreases the number of children attending appointments. For example, during a 6-month trial of the essence model, one quarter of the 125 children whose parent initiated a speech pathology referral did not receive therapy due to parent failure to either book or attend an appointment (Little & Grasselli, 2013). It is true that these types of approaches decrease waiting times, but they achieve this through their gatekeeper function and, in many cases, create barriers for clients attempting to access services (see 'Structural barriers' earlier).

1.3.1.3 Use in speech pathology

The modification of intensity variables is common in routine speech pathology practice. This section discusses how these variables can differ in routine practice compared with in research studies and in evidence-based practice (EBP) recommendations. The dose in routine practice (i.e. non-research) treatment sessions is often lower than the EBP recommendation, due to either shorter sessions or a lower number of the active ingredient within each session. For example, in paediatric stuttering treatment, sessions are more often 30–45 minutes rather than the hour recommended in the literature that informs EBP recommendations (O'Brian et al., 2013 cf. Onslow, Packman & Harrison, 2003). Both reduced session length and reduced number of trials

serve to decrease the treatment dose. Treatment provided outside research contexts generally has a lower dose-frequency than that employed in research trials. For example, outside research contexts, paediatric SSD therapy sessions are usually delivered once or twice per month rather than the recommended twice-weekly (Ruggero et al., 2012 cf. Baker & McLeod, 2014).

Dose form is another variable that clinicians vary as a way to manage large caseloads, by providing group rather than individual sessions (Lim et al., 2017; McAllister et al., 2011). However, as the limited information about the efficacy of group versus individual treatment is equivocal (see 'Efficacy' below), it is difficult to say whether modifications to dose form align with EBP recommendations. About one-third of Australian speech pathologists use group treatment as the dose form for paediatric SSD (McLeod & Baker, 2014). Both individual and group treatment are used in the United States of America, with the group form more common for children in K-12 and the individual form more common for pre-K (Mullen & Schooling, 2010). Stand-alone dose forms, such as computer-based applications, have been explored in speech pathology (e.g. Nordness & Beukelman, 2010; Shahin et al., 2015) but have not been part of routine service delivery (McLeod & Baker, 2014).

1.3.1.4 Efficacy

A small but growing number of research articles have investigated the impact of intensity variables on treatment outcomes. In general, a higher dose, higher dose-frequency and higher cumulative intervention intensity are all associated with stronger treatment outcomes (Kaipa & Peterson, 2016; Schooling et al., 2010; Wambaugh, Nessler, Cameron, & Mauszycki, 2013). However, recent research has indicated there may be an interaction between the variables of dose and dose-frequency, at least for childhood language disorders (Schmitt et al., 2017). For childhood language impairments, low-frequency treatment is more effective when there is a high dose within each session, and high-frequency treatment is more effective when there is a low dose in each session (Schmitt et al., 2017). This type of analysis of the interactions between dose

variables has not been conducted in SSD; therefore, it is unclear whether similar effects would be obtained.

There are limitations in the literature investigating dose parameters for SSD treatment. In a recent systematic review of studies investigating intensity variables in treatments for speech impairments, fewer than half the studies accounted for all intensity variables (Kaipa & Peterson, 2016). Kaipa and Peterson (2016) concluded that it was therefore not possible to confidently determine the effect of any one variable. For example, Namasivayam et al. (2015) investigated the relative effectiveness of weekly treatment, compared with twice-weekly treatment, for children with CAS. Thirty-seven children were allocated to one of two treatment groups for 10 weeks of treatment. One group received treatment once per week, the other twice per week. The authors reported that 'only the higher intensity treatment (2x/week) led to significantly better outcomes for articulation and functional communication compared with 1x/week' (Namasivayam et al., 2015, p. 529). Although this study appeared to investigate dose-frequency (weekly vs. twice-weekly therapy), the children in the twice-weekly group had a greater cumulative intervention intensity because they had 20 sessions compared with the weekly group's 10 sessions. The failure to control for cumulative intervention intensity means that it was impossible to separate the impact of dose-frequency and cumulative intervention intensity within the study. The higher dose-frequency may have been responsible for the improvement, but it could be that the greater number of sessions (i.e., cumulative intervention intensity) was responsible. Failure to control for all the intensity variables was also evident in other service-delivery investigations for SSD (Edeal & Gildersleeve-Neumann, 2011; Eiserman, McCoun, & Escobar, 1990).

Although there is limited research examining the impact of dose form (group vs. individual context), it appears that this variable has little impact on the efficacy of treatment (Cirrin et al., 2010). A systematic review of service-delivery variables for preschool-aged children with speech or language impairments (Schooling et al., 2010) found six studies addressed the effect of individual versus group treatment. The vast majority of effect sizes (17/20) indicated no clinically

significant difference between group and individual treatment. Of the three clinically significant differences, two favoured individual treatment, and one favoured group intervention. The review by Schooling et al., (2010) was limited in application to children with SSD because it included both speech and language treatments, and the service-delivery variables may differentially affect these two impairments.

Modifying dose parameters is one way speech pathologists attempt to improve access to speech pathology for their clients. Such modifications may include reducing the dose-frequency, dose amount, cumulative intervention intensity and/ or changing the dose form.

1.3.2 Modifying mode of treatment

Another way that speech pathologists attempt to improve access to speech pathology is by utilising alternative modes of treatment. The traditional mode for speech pathology services has been individual face-to-face mode, where one client meets with one clinician in the same room for the consultation. Alternative modes may vary the number of clients, such as in group treatment (discussed more fully in 'Dose parameters' above). Other variations in mode include less-traditional clinician roles, such as occur with transdisciplinary treatment, where multiple allied health inputs are provided by one clinician, or with multidisciplinary treatment, where several allied health providers provide services to the same client. One popular modification of treatment mode is the use of technology to allow the client and clinician to connect, such as is used in telehealth treatment. The following section discusses the use of various telehealth modes of treatment.

1.3.2.1 Telehealth

The last decade has seen a significant increase in research and practice demonstrating the use of telehealth modality for speech pathology services. Telehealth is 'the application of telecommunications technology to deliver clinical services at a distance, by linking clinician to client, caregiver, or any person(s) responsible for delivering care to the client, for the purposes of

assessment, intervention, consultation and/or supervision' (Speech Pathology Australia, 2014b, p. 4). Telehealth is variously known as telespeech, telecare, telerehabilitation and telepractice (Mashima & Doarn, 2008; Theodoros, 2013). Telehealth fits within the broader category of e-health, which encompasses all electronic processes and communication technologies that support healthcare, such as electronic medical records, stand-alone technology-based therapy, and digital collection of data for assessment and treatment (Speech Pathology Australia, 2014b).

Telehealth applications can be synchronous, where information is sent and received in real time such as videoconferencing, or asynchronous (i.e., store and forward) such as email and purpose-built computer applications. Although various telehealth platforms are available, such as teleconference, video, email and videoconference, this thesis focuses mostly on videoconferencing. During a videoconference, visual and auditory information is transferred in real time via the Internet. Videoconferencing enables the clinician to directly conduct activities with the client and provide feedback in real time despite significant physical distance between the client and the clinician (Theodoros, 2008). During a speech pathology videoconference, the speech pathologist is most commonly located at their workplace (e.g., clinic or hospital) and the client is located at home or an accessible place in the community (e.g., school or community centre; Hill & Miller, 2012).

The main advantage of telehealth for service delivery is the ability to address the geographical barrier to accessing services that disproportionately affects people living in rural and remote locations (American Speech Language Hearing Association, 2005; Lowe, O'Brian, & Onslow, 2014; Speech Pathology Australia, 2014b; Theodoros, 2011). Services provided by telehealth decrease clients' travel time and associated costs (Mashima & Doarn, 2008) and increase convenience (Hill & Miller, 2012; Theodoros, 2013). Telehealth also saves clinicians' time, as it can eliminate the need for travel to outlying areas (Hill & Miller, 2012). Although many videoconferencing studies use custom-built systems, it is possible to use personal computers or

tablets with inbuilt webcams, and for families with existing Internet connections, set-up costs are low (Speech Pathology Australia, 2014b).

The benefits of telehealth are not limited to clients in rural and remote areas. Telehealth has application for metropolitan clients who find it difficult to attend face-to-face sessions (Lowe et al., 2014; Theodoros, 2013). It can enable metropolitan clients to receive weekly services that would otherwise have been impractical due to lengthy travel time resulting from traffic congestion (Verdon et al., 2011). In many cases, clients prefer telehealth to the face-to face modality due to the convenience, time savings and reduction in transport costs (Mashima & Doarn, 2008; Theodoros, 2011). Telehealth treatment can have stronger generalisation gains than traditional face-to-face clinic-based treatment (Burgess et al., 1999; Mashima & Doarn, 2008; Theodoros, 2011).

1.3.2.2 Use in speech pathology

Telehealth is used in many countries around the world, including the United States of America, Canada, Greece, Ireland, the UK and Japan, for a variety of impairments, from neurogenic communication disorders and paediatric speech and language impairments to dysphagia and stuttering (Molini-Avejonas, Rondon-Melo, Amato, & Samelli, 2015). Consequently, many speech pathology professional organisations have published position statements regarding telehealth (e.g. American Speech Language Hearing Association, 2005; Canadian Associaion of Speech Language Pathologists and Audiologists, 2006; Speech Pathology Australia, 2014b).

Telehealth is more commonly used in rural areas than metropolitan areas (Molini-Avejonas et al., 2015), and most speech pathologists who use telehealth are based in regional or rural areas (Hill & Miller, 2012). However, despite Australia's vast rural landscape, telehealth use is not universal, with only 13% of Australian allied health professionals using telehealth at the beginning of the decade (Australian Government Department of Health, 2012). Even when telehealth is used, it is generally only for a small proportion (e.g., 0%–30% of the caseload; Hill & Miller, 2012).

Historically, telehealth has been used more with paediatric clients than with adult clients and in treatment more than in assessment sessions (Hill & Miller, 2012).

1.3.2.3 Efficacy

In general, telehealth has similar efficacy to that of face-to-face treatment (Speech Pathology Australia, 2014b). There has been a steady increase in investigations of telehealth efficacy since the first two comprehensive reviews of the literature reported that telehealth holds great potential for application in speech pathology assessment and treatment (Theodoros, 2008, 13 studies: Mashima & Doarn, 2008, 40 studies). The most recent comprehensive literature reviews (Molini-Avejonas et al., 2015, 103 studies; Speech Pathology Australia, 2014b, 77 studies) revealed more extensive research across the breadth of speech pathology range of practice areas for both assessment and treatment. Speech Pathology Australia (2014b) reported that personal-computer-based (PC-based) videoconferencing is valid and reliable for the assessment and treatment of childhood speech and language, acquired neurological impairments and hearing impairment and for the treatment of stuttering, provided the technical capabilities of the signal permit high audio and visual quality. Videoconferencing was also reported efficacious for the assessment and treatment of dysphagia, voice, craniofacial and head and neck disorders; however, this was demonstrated by researchers using custom-built systems with features and technical capabilities beyond that of PC-based systems (Speech Pathology Australia, 2014b). No research has been published investigating the efficacy of videoconferencing for treatment of children with CAS.

1.3.2.4 Other considerations

Telehealth service delivery requires specific hardware, software, connectivity and clinician and client skills and attitudes (Keck & Doarn, 2014). Custom-built videoconferencing systems, although technically superior, are expensive and not widely available. Even PC-based systems have minimum specifications (Keck & Doarn, 2014), and not all homes have access to sufficient or sufficiently powered hardware. Although free or low-cost videoconferencing software is

available via programs such as Skype, clinicians have concerns about security, privacy, reliability and confidentiality, and many workplaces prohibit use of these programs for clinical services (Hill & Miller, 2012; May & Erickson, 2014). Although Internet connectivity in Australia is relatively good, and improving year on year (Akamai, 2016), access to high speed Internet is not universal (Australian Government Department of Health, 2012), and poor telecommunication connectivity is one of the main barriers to telehealth use (Behl & Kahn, 2015; Hill & Miller, 2012). There is limited evidence supporting the use of PC-based video conferencing for conditions that require high fidelity audio signal, such as speech and voice (Keck & Doarn, 2014; Speech Pathology Australia, 2014b) and, by extension, CAS. For families, telehealth treatment can be more expensive than the equivalent traditional service, as there are limits on financial reimbursements for telehealth services under some government schemes (e.g., NDIS) and private health insurers.

In addition to hardware and software requirements, effective telehealth delivery requires specific clinician skills and attitudes (Hines, Lincoln, Ramsden, Martinovich & Fairweather, 2015; Wade & Eliott, 2012). Clinicians may require training to develop technical proficiency with the specific telehealth system and may need technical support during the implementation of telehealth treatment (May & Erickson, 2014; Speech Pathology Australia, 2014b). Some assessment and treatment materials require modification for use in telehealth modality (Hill & Miller, 2012). Lastly, it has been argued that the clinician's positive attitude to telehealth is integral to the success of telehealth treatment (May & Erickson, 2014; Wade & Eliott, 2012).

1.3.3 Changing treatment delivery agent

Although speech pathology intervention has traditionally been delivered by a speech pathologist, it is possible for the treatment to be delivered in part, or in the majority, by a trained caregiver, teacher, speech-language therapy assistant (SLTA), allied health assistant or even a computer (SLTA, Law et al., 2017). These alternative treatment delivery agents enable the client to potentially receive higher treatment intensity than would be available from the speech pathologist

alone (Baker & McLeod, 2011; Law et al., 2017; Lim et al., 2017). When the alternative delivery agent provides treatment in a natural setting, such as the client's home or classroom, generalisation can be enhanced (Roberts & Kaiser, 2011). For service providers, there is a potential cost saving associated with employing a SLTA rather than a speech pathologist, and no salary costs are associated with service provided by a caregiver or teacher. Even when the time required to train parents is considered, parent-delivered treatment is more cost effective than treatment delivered by a speech pathologist (e.g. Barnett, Escobar, & Ravsten, 1988).

Despite the prevalence of non-speech pathologist involvement in treatment (see 'Use in speech pathology' below), there are significant gaps in the detail provided in the literature about the nature of this involvement (Sugden, Baker, Munro, & Williams, 2016). There are three broad purposes this involvement may take: to facilitate generalisation (e.g. PROMPT, Dale & Hayden, 2013), to supplement the intervention provided by the speech pathologist (e.g. PACT, Bowen, 2010) and to be the predominant intervention agent (e.g. Onslow, Packman, & Harrison, 2003, Eiserman et al., 1990). Each of these three broad types of involvement involves myriad tasks, methods and levels of intensity, resulting in almost limitless permutations of involvement by other intervention agents (Sugden et al., 2016). For the purposes of this thesis, another agent is considered the treatment delivery agent when they (a) establish a situation that allows for administration of a therapeutic dose and (b) respond to the client in such a way as to encourage higher accuracy with the targeted skill (e.g., provide accurate feedback on productions).

1.3.3.1 Use in speech pathology

Speech pathologists, internationally, involve other agents in the delivery of treatment (e.g. UK, Joffe & Pring, 2008; Germany, Keilmann et al., 2004; South Africa, Pascoe et al., 2010; Australia, Watts Pappas et al., 2008). More than 85% of Australian paediatric speech pathologists provide therapy via parents, and more than a third provide therapy via teachers and teacher's aides (Baker & McLeod, 2011). Involvement of other agents occurs across client age groups and impairments (e.g. Onslow et al., 2003; Roberts & Kaiser, 2011; Togher, Power,

Rietdijk, McDonald, & Tate, 2012). As discussed, there is a lack of clarity regarding the nature of this involvement. For example, when investigating the nature of parent involvement, questionnaire studies about routine service delivery have variously asked respondents to indicate whether they use 'parent involvement' (Joffe & Pring, 2008, p. 159), whether they 'give exercises' (Keilmann et al., 2004, p. 54) and whether they 'incorporate home programmes' (Pascoe et al., 2010, p. 75).

1.3.3.2 Efficacy

As one of the foci of this thesis is caregiver-provided treatment, this section predominantly focuses on the efficacy of parents as treatment delivery agents. In the literature, there are frequent statements about the importance of caregiver involvement for the success of treatment, particularly for paediatric clients (e.g. Glogowska, Campbell, Peters, Roulstone, & Enderby, 2002; Law, Zeng, Lindsay, & Beecham, 2012; Roberts & Kaiser, 2011). However, the specific nature of caregiver involvement can vary markedly between studies and between clinical populations. Although clinic sessions supplemented with caregiver home practice are effective for many communication impairments—for example, childhood language impairment(e.g. childhood language impairment, Roberts & Kaiser, 2011; paediatric speech sound disorder, Bowen & Cupples, 1999)—it can be difficult to determine the relative contribution made by the caregiver agent to the outcome. The reason for this difficulty is twofold. Firstly, information about the tasks performed by the caregiver and about the frequency and fidelity of the tasks is limited (Kaderavek & Justice, 2010; Sugden et al., 2016). Secondly, comparisons between participants receiving the same intensity of speech-pathologist-delivered treatment with caregiver involvement and without caregiver involvement are rare.

The difficulty in determining the relative contribution of the intervention agent to the treatment outcome was demonstrated by Eiserman et al. (1990) in their study of the cost effectiveness of service delivery for preschool-aged children with speech impairments. Eiserman and colleagues compared parent-delivered and therapist-delivered treatment for preschool-aged children with

speech and language impairments. The children were allocated to therapy either in a pair with a speech pathologist at the clinic (1-hour treatment, one session per week) or individually at home with a trained parent (20–30 minutes, four times per week) for 7 months, using traditional articulation therapy. Following treatment, no significant differences were found between the two treatment conditions on the speech measures, although the parent-delivered condition resulted in better performance on the expressive language measures. However, the numerous differences between the two groups, such as setting (clinic vs. home), context (group vs. individual), intervention agent (clinician vs. parent), dose-frequency (once per week vs. four times per week) and cumulative intervention intensity (1,680 mins in the clinic vs. 2,240–4,460 mins at home) make it impossible to determine which feature or features were responsible for the treatment outcome.

It is easier to determine the efficacy of caregiver-delivered intervention when it is delivered exclusively by the caregiver and compared with either no treatment or treatment by a speech pathologist. Although these types of studies are scant (see Sugden et al., 2016, for a review), the outcomes are generally positive. A meta-analysis of caregiver-provided therapy in allied health found that caregiver-provided therapy was better than no therapy for stuttering and for expressive language (Lawler, Taylor, & Shields, 2013). Similarly, childhood SSD treatment provided by trained caregivers is better than no treatment (e.g. Broen & Westman, 1990). The limited investigations of the efficacy of caregiver-provided treatment compared with clinician-provided treatment indicate that treatment has similar levels of effectiveness for expressive language and SSDs (Lawler et al., 2013). There is no research investigating the outcomes of caregiver-provided treatment for CAS.

1.3.3.3 Other considerations

Parents prefer treatment to be delivered by a clinician than by another provider. They consider the speech pathologist to be the expert, and they expect the clinician to carry out the intervention (Carroll, 2010; Glogowska et al., 2002; Hayhow, 2009). For example, only 4% of Australian

parents wanted their child to be treated using parent training or a home program (Ruggero et al., 2012). Some parents place less value on intervention than their speech pathologists do, due to their differing perspectives regarding speech and language development, the child's difficulties and the need for intervention (Carroll, 2010; Davies, Marshall, Brown, & Goldbart, 2016; McAllister et al., 2011), as predicted by the health belief model (Carpenter, 2010). There may be practical reasons for parents not being able to complete intervention with their children, such as difficulty ensuring child compliance and not having sufficient time to implement the treatment (McAllister et al., 2011). Indeed, clinicians identify that limited parent engagement in the therapeutic process affects the delivery of home-based speech practice (Lim et al., 2017), and clinicians would like parents to be more involved (Baker & McLeod, 2011; Lim et al., 2017).

The time required to train another intervention agent must be balanced against the potential benefits. Clinicians in the UK spend more time on training parents and other professionals than they do on providing direct treatment (Pring, Flood, Dodd, & Joffe, 2012). Therefore, to determine whether time spent on parent training is time well spent, it would be necessary to first understand the effectiveness of parent training on treatment outcomes. Another factor to consider regarding parent-delivered treatment is whether parents deliver the intervention accurately (i.e., with high treatment fidelity). High treatment fidelity is associated with the strongest treatment outcomes (Kaderavek & Justice, 2010). Lastly, there are indications that some treatments or conditions may be too complex for someone other than a speech pathologist (Sugden et al., 2016). For example, it is not yet known whether it will be appropriate for parents to deliver therapy to children with CAS, a highly complex condition.

1.4 Stakeholder perspectives

1.4.1 Families

This chapter has outlined several strategies that could potentially improve access to speech pathology services. However, these strategies are not equally well accepted by families. Parents

desire frequent treatment (Glogowska et al., 2002) and would like more therapy than their child routinely receives (Boyle, McCartney, Forbes, & O'Hare, 2007; Glogowska & Campbell, 2000). Strategies that reduce the frequency of the sessions in order to 'stretch out' treatment may not be well received by parents. Parents repeatedly report a preference for individual treatment rather than group treatment (McAllister et al., 2011; Watts Pappas, McLeod, McAllister, & McKinnon, 2008). Although caregivers are initially sceptical about telehealth treatment, those who have experienced it report very positive experiences (e.g. Lincoln, Hines, Fairweather, Ramsden, & Martinovich, 2014; Molini-Avejonas et al., 2015). Regarding delivery agent, parents repeatedly express a desire for the treatment to be implemented by the speech pathologist rather than anyone else, and they are reluctant to deliver the treatment with their child (Carroll, 2010; O'Callaghan, McCallister, et al., 2005; Ruggero et al., 2012).

1.4.2 Speech pathologists

There is limited research investigating the impact of service-delivery models on clinicians. What is known is that the struggle between caseload demands and limited resources is a frequent cause of anxiety and internal conflict for speech pathologists (Kenny & Lincoln, 2012; Lim et al., 2017) and that speech pathologists try to resolve this tension through creative approaches to service delivery (Lim et al., 2017).

1.4.3 Service providers

Cost effectiveness and efficiency of service are important for service providers. There is relatively little research examining cost-benefit of either telehealth (Mashima & Doarn, 2008) or modification of dose parameters (Baker, 2012; Tindall, 2013). Treatments delivered by alternative delivery agents are appealing to service providers, as they are associated with potential reductions in staffing costs, and there have been some investigations of cost-benefit in this area (e.g. Boyle et al., 2007; Eiserman et al., 1990). Cost-benefit analyses of treatments delivered by non-speech pathologists have considered service-delivery approaches either individually or in

combination. For example, Boyle et al. (2007) investigated the cost effectiveness of direct and indirect (i.e., provided by a SLTA) treatment in individual and group contexts for school-aged children with language impairments. There was no difference between direct and indirect treatment in regard to treatment effectiveness; however, the most cost-effective treatment was delivered by the SLTA and even more so when in a group context (Boyle et al., 2007). Although further research is needed on the efficacy and cost effectiveness of service-delivery approaches, such research is beyond the scope of this thesis.

1.5 The challenges

Despite the pragmatic appeal of modifying service-delivery approaches as a means of overcoming access barriers, two main challenges remain. The first is to determine whether these service-delivery approaches are efficacious for specific impairments and treatments. The second is to determine the acceptability of the approaches for families and whether the approaches have any unintended consequences. Each of these challenges is explored briefly below.

1.5.1 Efficacy

It is essential to determine whether treatment continues to be efficacious when the service delivery is modified. As noted by Brumbaugh and Smit (2013), service delivery can have a significant effect on the efficacy of a treatment, and speech pathologists need 'to consider the differences between the conditions under which they provide services and the conditions under which the intervention was found to be efficacious'(Brumbaugh & Smit, 2013, p. 308). For example, despite evidence that language therapy is effective (Law et al., 2004), when it is provided for only 6 hours across a year it has no greater effect than no therapy (Glogowska, Roulstone, Enderby, & Peters, 2000). Further, there may be an interaction between service-delivery approach and impairment, with dose-parameter modification producing differing results for various impairments (Allen, 2013; Smith-Lock et al., 2013). This has led to repeated calls for service-delivery models to be tailored to the specific impairment and treatment (Law & Conti-Ramsden, 2000; Nippold, 2012).

1.5.2 Parent experiences

To deliver effective treatment, speech pathologists need to consider more than treatment efficacy. They need to also consider the three elements of evidenced-based practice: the best available evidence regarding efficacy, the preferences of a fully informed client, and their own clinical skills and experience (Dollaghan, 2007). The first step in understanding clients' preferences is to understand their experience of treatments and service-delivery approaches.

Chapter 2: Childhood Apraxia of Speech

Chapter 1 described the significant psychosocial, educational and vocational impacts of SSD, and established the need for intervention. It also described service-delivery strategies that may help to overcome barriers that reduce opportunities for children with SSD to access speech pathology services. Importantly, Chapter 1 explored emerging indications that some service-delivery approaches may be more suitable for some clients and some impairments than for others. This chapter explores treatments for CAS, a specific type of SSD. This chapter has three goals. Firstly, to describe the nature of the impairment; secondly, to report on the existing treatment for CAS; and, thirdly, to explore the service-delivery methods used for CAS treatments. For the purposes of this thesis, CAS has been used as the name of the disorder, although it is known as developmental verbal dyspraxia in the UK (Royal College of Speech & Language Therapists, 2011).

2.1 Nature of the impairment

2.1.1 Classification and definition

As discussed in Chapter 1, CAS is a SSD (American Psychiatric Association, 2013). Although CAS fits into the broad category of SSD, SSD is not a homogeneous category. Many classification systems have been proposed for SSDs (e.g. Dodd, 2005; Duffy, 2005), but one commonly used is the Speech Disorders Classification System (SDCS, Shriberg et al., 2010). According to the SDCS, CAS belongs to the class 'motor speech disorder' and the subtype 'motor speech disorder—childhood apraxia of speech' (Mabie & Shriberg, 2017).

For decades, there was contention regarding the existence, nature, and diagnostic features of CAS (American Speech Language Hearing Association, 2007b; Forrest, 2003; Ozanne, 1995). The publication of a position statement and accompanying technical report by the American Speech Language Hearing Association (American Speech Language Hearing Association, 2007a) increased consensus regarding the nature of CAS. In these documents, CAS was defined as a 'neurological childhood (pediatric) speech sound disorder in which the precision and consistency

of movements underlying speech are impaired' (American Speech Language Hearing Association, 2007b, p. 3). The difficulty that children have with achieving accuracy in speech movements results in errors with speech sounds and prosody (American Speech Language Hearing Association, 2007b). In the UK, a similar position statement was released by the Royal College of Speech and Language Therapists (Royal College of Speech & Language Therapists, 2011).

Three key features indicate the motor planning and programming difficulties central to CAS (American Speech Language Hearing Association, 2007b). These are (a) inconsistent errors on consonants and vowels in repeated productions of words, (b) lengthened and disrupted coarticulatory transitions, and (c) inappropriate prosody, particularly with lexical or phrasal stress (American Speech Language Hearing Association, 2007b). Although these features were not listed as being sufficient for diagnosis of CAS, in the ensuing decade they have regularly been used as minimum diagnostic criteria in treatment research (e.g. Namasivayam et al., 2015; Skelton & Hagopian, 2014).

2.1.2 Prevalence

CAS is an uncommon SSD, reported to occur in one or two children per thousand (American Speech Language Hearing Association, 2007b). By one older estimate, approximately 3%–5% of children with SSD have CAS (Shriberg, 1994).

2.1.3 Associated difficulties

Children with CAS have difficulties beyond speech production. Many researchers and clinicians report broader communication difficulties in children diagnosed with CAS (American Speech Language Hearing Association, 2007b). What is unclear is whether these difficulties are a central component of the impairment, sequelae, or co-occurring conditions (See American Speech Language Hearing Association, 2007b). For example, children with CAS are likely to have difficulty encoding auditory information and with auditory memory (Shriberg, Lohmeier, Strand,

& Jakielski, 2012). They are also reported to have poor skills with linguistic processing (Maassen, 2002) and to rely more on auditory feedback than other children their age (Iuzzini-Seigel, Hogan, Guarino, & Green, 2015).

Children with CAS are more likely than typically developing children to have language difficulties. They frequently have poor expressive language skills (Lewis, Freebairn, Hansen, Iyengar, & Taylor, 2004; Ozanne, 1995; Velleman, Strand, Bernthal, & Bankson, 1994) and can have poor receptive language (Hall, Jordan, & Robin, 2007) and/or poor receptive vocabulary (Carrigg, Parry, Baker, Shriberg, & Ballard, 2016). Although there is debate about whether these language difficulties are a central component of CAS, a result of the speech motor impairment, or independent co-occurring conditions (See American Speech Language Hearing Association, 2007b), the fact remains that many children with CAS have language challenges.

Children with CAS frequently experience learning and literacy impairments. Compared with typically developing peers, children with CAS have more difficulty with spelling (Snowling & Stackhouse, 1983), phonological awareness (Gillon & Moriarty, 2007), literacy (Carrigg et al., 2016; Moriarty & Gillon, 2006) and verbal intelligence (Carrigg et al., 2016). They are likely to need additional support at school (Carrigg et al., 2015; Fish, 2015; Hall et al., 2007; Lewis, Freebairn, Hansen, Iyengar, et al., 2004) and frequently report continuing difficulties into adulthood (Carrigg et al., 2016; McCabe, Preston, Murray, Bricker & Morgan, 2017).

Lastly, people with CAS may also have social and psychosocial difficulties. Although the research in this area is more limited for CAS than for children with SSDs more broadly, the indications are that the same challenges are experienced, with potentially greater severity. For example, McCormack et al. (2012) reported the reflections of a young man, aged 17, growing up with CAS. He told of the challenges of not being understood, the difficulty in finding friends who would take the time to understand him, and the experiences of being bullied. Carrigg et al. (2015) reported the experiences of another young man with apraxia, named BJ, who had persistent speech difficulties. Despite average intelligence, BJ's speech was so unintelligible that he used an

electronic device to communicate. During BJ's school years, he became more withdrawn and refused to speak outside the family home. He experienced anxiety and depression, engaged in self harm and reported suicidal thoughts, and at the age of 12, BJ was admitted to an inpatient mental health service.

In summary, CAS is an uncommon and lifelong impairment affecting multiple communication, academic and psychosocial domains. As with other SSDs, CAS requires treatment. The lifelong experience of CAS is exemplified in the following words from BJ:

> My communication disorder has had a significant and profound impact on my life. It has affected my ability to interact with the people around me, my education, and my mental health. I am unable to do many things because of my communication. Growing up I often felt left out because I wasn't able to talk with other people, I wasn't able to tell other people my thoughts or if I needed something. It was heartbreaking because I knew what I wanted to say, but I couldn't say it. I still feel deeply sad about not talking to others. It was extremely hard for me to make friends because of my communication. When I was at school I can remember spending every lunch time sitting by myself because no one will even try to talk to me. I was constantly bullied because of the way I talk, this really impacted (Carrigg et al., 2015, p. 46).

2.2 Principles underpinning intervention

Selecting an appropriate treatment for children with CAS can be complex. There are many factors to consider, such as the weight of the supporting evidence and the goals of the intervention. These factors are considered below.

2.2.1 Evidence-based practice

EBP had its origins in medicine as a construct for evaluating the trustworthiness of claims regarding assessment and intervention approaches (Vallino-Napoli & Reilly, 2004). EBP was

originally described as 'the conscientious, explicit and judicious use of current best evidence in making decisions about the care of individual patients' (Sackett, Rosenberg, Gray, Haynes, & Richardson, 1996, p. 71). In considering how to apply the concept of EBP to communication disorders, Dollaghan (2007) modified the EBP framework to consider three sources of evidence: (a) external evidence from systematic research, (b) internal evidence from the clinician's expertise, and (c) the client's preferences, and she termed the construct E^3BP. The following section focuses on the first of these three areas: external evidence from systematic research. Parents' preferences are discussed in Chapter 7; internal evidence is beyond the scope of this thesis, but is discussed in Chapter 8 as a direction for future research.

2.2.2 Focus of the intervention

Another important principle to consider in selecting treatment is the goal of the intervention. The World Health Organization's International Classification of Functioning, Disability and Health—Child and Youth (ICF-CY, World Health Organization, 2007) has two main classifications of goals. These are (a) Body Structures and Functions, and (b) Activities and Participation. The ICF-CY provides a framework for describing goals, with reference to an individual's abilities and limitations, in terms of anatomical structures, functional use of the structures, engagement in activities, and participation in society. The ICF-CY framework has been used for classifying speech pathology interventions (Cunningham et al., 2017; McLeod, 2004; O'Halloran & Larkins, 2008) and is used within this thesis as a means of classifying CAS treatments. Moving beyond ICF-CY classification, treatments are then classified according to the focus of the intervention—that is, into treatments with a motor, linguistic, or multimodal focus. Some of these categories are further subdivided; for example, biofeedback is a subcategory of motor treatments.

CAS treatments with published efficacy investigations are summarised in Table 2.1. In the table, each treatment is listed along with ICF-CY classification, focus of intervention, studies of efficacy, number of participants demonstrating treatment effect, and service-delivery information.

The service-delivery information covers intervention variables, mode of intervention, and treatment agent.

2.3 Treatment approaches

2.3.1 Treatments addressing Body Structures and Functions

In classifying speech production, Body Structures refers to the anatomy of the vocal tract, respiratory system, ear and nervous system (World Health Organization, 2007). Body Functions are the physiological functions of body systems (World Health Organization, 2007) and include speech production, quality, rhythm and intelligibility (Cunningham et al., 2017). Speech pathology treatments rarely address Body Structures. The majority of CAS treatments address limitations of Body Functions (see Table 2.1). Treatments addressing Body Functions can be further classified into categories of motor and linguistic foci, and are described below (see also Table 2.1).

2.3.1.1 Motor intervention

Motor interventions for CAS aim to improve the movement aspects of speech production. There are theoretical indications that a motor speech impairment such as CAS is likely to respond well to treatment that uses motor learning principles (e.g., Maas et al., 2008; Schmidt & Lee, 2011) and /or neural plasticity principles (Kleim, 2013). Treatments using these principles frequently employ a high number of trials and/or frequent sessions across an extended period of time with the aim of enhancing true learning rather than short term performance (Kleim, 2013; Maas, 2008). The majority of CAS treatments address motor skills (Murray et al., 2014, see also Table 2.1), and many of these are underpinned by motor learning and/ or neural plasticity principles (see Maas, Gildersleeve-Neumann, Jakielski, & Stoeckel, 2014 for a review). There is diversity in the motor treatment approaches, with motor interventions employing techniques ranging from visualisation of the articulators, through biofeedback, to implementation of tactile cues, and imitation of non-words. However, all treatments within this category share a common goal: to improve the motor act of speech production.

Table 2.1: CAS treatments, including ICF domain, communicative focus, published efficacy, and service delivery

ICF focus	Domain	Treatment	Partic. with Tx effect	Published outcomes (design)	Intensity variables						Rx mode	Rx agent	Home practice?
					Dose form	Dose amount	Dose-frequency	Intervention duration	Session length	Cumul-ative intensity[a]			
Body Structure and Function	Function—Motor	Concurrent treatment	3/3	Skelton & Hagopian, 2014 (Multiple BL)	Indiv.	101–104 trials/sess.	2x week	12–28 sess.	30 min	2,040 trials	FTF	SLP	NS
		DuBard approach	12/12	Martin et al., 2015 (AB)	Class	NS	Each school day	11 months	School day	–	FTF	SLP (teacher)	NS
		DTTC/ Integral stimulation	15/17	Baas et al., 2008 (Multiple BL)	Indiv.	NS	10x week (Blk 1); 4x week (Blk 2); 1x week (Blk 3)	6 weeks–8 months	NS	–	FTF	SLP	Yes^
				Edeal & Gildersleeve-Neuman, 2011 (AB)	Indiv.	130–170 prod./sess.	2–3x week	5 weeks (1 child); 10 weeks (1 child)	40–50 min	2,812 prod.	FTF	SLP	Yes^
				Gildersleeve-Neumann & Goldstein, 2015 (Multiple BL)	Indiv.	40–100 prod./sess.	2–3x week	8 weeks	50 min	1,400 prod.	FTF	SLP student	NS
				Maas et al., 2012 (Multiple BL)	Indiv.	46–88 prod./sess.	3x week	2x 4-week blks	60 min	603 prod.	FTF	SLP student	NS
				Maas & Farinella, 2012 (Multiple BL)	Indiv.	NS	3x week	2x 4-week blks	NS	–	FTF	SLP student	NS
				Strand et al., 2006 (Multiple BL)	Indiv.	≤ 150 prod./sess.	10x week	4–6 weeks	30 min	1,350 prod.	FTF	SLP	Yes, 5 min 2x day
				Strand & Debertine, 2000 (Multiple BL)	Indiv.	NS	4x week	Approx. 34 sess.	30 min	–	FTF	SLP	NS

ICF focus	Domain	Treatment	Partic. with Tx effect	Published outcomes (design)	Intensity variables						Rx mode	Rx agent	Home practice?
					Dose form	Dose amount	Dose-frequency	Intervention duration	Session length	Cumul-ative intensity[a]			
Body Structure and Functions	Function—Motor	Nuffield Dyspraxia Programme—third edition (NPD-3)	Sig. group effect	Murray et al., 2015 (RCT)	Indiv.	101 prod./sess.	4x week	3 weeks	60 min	1,212 trials	FTF	SLP student	No
		Prompts for Restructuring Oral Muscular Phonetic Targets (PROMPT)	2/4*+group effect	Dale & Hayden, 2013 (ABB and ACB)	Indiv.	45–60 prod./sess.	2x week	8 weeks	50 min	848 prod.	FTF	SLP	Yes^
				Namasivayam et al., 2015 (Quasi-experimental study)	Indiv.	NS	1 or 2x week	10 weeks	NS	–	FTF	SLP	Yes, 126–1,897 min/blk
		Rapid Syllable Transition (ReST)	7/7 + sig. group effect	Ballard et al., 2010 (Multiple BL)	Indiv.	100–120 trials/sess.	4x week	3 weeks	60 min	1,320 trials	FTF	SLP student	NS
				McCabe et al., 2014 (Multiple BL)	Indiv.	100 trials/sess.	4x week	3 weeks	60 min	1,200 trials	FTF	SLP student	No
				Murray et al., 2015 (RCT)	Indiv.	100 trials/sess.	4x week	3 weeks	60 min	1,200 trials	FTF	SLP student	No
		Melodic intonation therapy (MIT)	4/4	Krauss & Galloway, 1982 (ABACA)	Indiv.	NS	2x week	2 months	NS	–	FTF	SLP	NS
				Helfrich-Miller, 1994 (Case study)	Indiv.	NS	NS	NS	NS	–	FTF	NS	NS
	Function—Motor (Combined Tx)	MIT and TCM	1/1	Martikainenen & Koprilahti, 2011 (ABACA)	Indiv.	NS	3x week	12 weeks	30 min	–	FTF	NS[@]	No
		Stimulability + Core Vocabulary	4/4	Iuzzini & Forrest, 2010 (Multiple BL)	Indiv.	NS	2x week	10 weeks	55 min	–	FTF	NS[@]	NS[7]
		NDP-3, MIT and multisensory	1/1	Singh & Trivedi, 2016 (Case study)	Indiv.	NS	2x week	7 months (3 x 18-week blks)	60 min	–	FTF	NS[@]	Yes[!]

ICF focus	Domain	Treatment	Partic. with Tx effect	Published outcomes (design)	Intensity variables						Rx mode	Rx agent	Home practice?
					Dose form	Dose amount	Dose-frequency	Intervention duration	Session length	Cumul-ative intensity[a]			
Body structure and functions	Function—Motor (biofeedback)	Electro-palatography (EPG)	3/3	Carter & Edwards, 2004 (AB)	Indiv.	NS	1 x week	10 weeks	30 min	–	FTF	SLP	NS
				Lundeborg & Edwards, 2007 (Multiple BL)	Indiv.	NS	Daily	30 weeks (3x 5-week blks Rx)	15–20 min	–	FTF	Parent [#]	NS[*]
		Ultrasound (US)	8/12	Preston et al., 2013 (Multiple BL)	Indiv.	150 US trials/sess.	2 x week	18 sess.	60 min	2,700 trials	FTF	SLP or SLP student	NS
				Preston, Leece et al., 2016 (Case series)	Indiv.	NS	5x week	2 weeks	120 min	–	FTF	SLP	NS
				Preston, Maas et al., 2016 (Multiple BL)	Indiv.	210 prod./sess.	2x week	14 sess.	60 min	2,940 prod.	FTF	SLP	No
	Function—Linguistic	Integrated Phonological Awareness (IPA)	11/15	Moriaty & Gillon, 2006 (Multiple BL)	Indiv.	NS	3 x week	3 weeks	45 min	–	FTF	SLP	No
				McNeill et al., 2009a (Case series)	Indiv.	≥ 15 prod./ activity	2 x week	12 weeks	45 min	360 prod.	FTF	SLP or student	NS
				McNeill et al., 2009b *subset of 2009a (Quasi-experimental design)	Indiv.	≥ 15 prod./ activity	2x week	12 weeks	45 min	–	FTF	SLP or SLP student	No
				McNeill et al., *subset of 2009a (Multiple BL)	Indiv.	≥ 15 prod./ activity	2x week	12 weeks	45 min	–	FTF	SLP or SLP student	NS

ICF focus	Domain	Treatment	Partic. with Tx effect	Published outcomes (design)	Intensity variables						Rx mode	Rx agent	Home practice?
					Dose form	Dose amount	Dose-frequency	Intervention duration	Session length	Cumul-ative intensity[a]			
Activities and participation	Activities	Aided AAC modelling with picture symbols	4/4	Binger et al., 2011 (Multiple probe)	Indiv.	≥ 10 models/ sess.	NS	7 sess.	15 min	–	FTF	SLP	NS
				Binger et al., 2008 (Multiple probe)	Indiv.	≥ 6 models/ sess.	NS	11 sess.	10 min	–	FTF	Parent	NS
				Binger & Light, 2007 (Multiple probe)	Indiv.	≥ 30 models/ sess.	1–3x week	15 sess.	15 min	450 models	FTF	SLP	NS
		Speech-generating device	3/3	Bornman et al., 2001 (AB)	Indiv.	NS	1–2x week	2 weeks	60 min	–	FTF	Parent	Yes^
				Harris, Doyle et al., 1996 (Multiple BL)	Indiv.	100–150 utt./sess.	2x week	4 months (22 sess.)	45 min	2,750 utt.	FTF	SLP and comp.	Yes^
				Lüke, 2016 (AB)	Indiv.	NS	1x week[b]	50 sess.	45 min	–	FTF	SLP	Yes[!]
	Participation	Partners in Augmentative Communication Training	1/1	Culp, 1989 (AB)	Indiv.	NS	3 days	3 days	1 day	–	FTF	SLP trained parent	Yes
		Integrated multimodal intervention	3/3	King et al., 2013 (Multiple probe)	Indiv.	≤ 120 reps/ sess.	2x week	9–14 weeks	35–45 min	2,760 reps	FTF	NS[@]	NS

ICF focus	Domain	Treatment	Partic. with Tx effect	Published outcomes (design)	Intensity variables						Rx mode	Rx agent	Home practice?
					Dose form	Dose amount	Dose-frequency	Intervention duration	Session length	Cumul-ative intensity[a]			
		Eclectic (signing, communication board, and various speech approaches)	5/5	Watson & Leahy, 1995 (Case study)	Indiv.	NS	Fortnightly	2 years	2 hr	–	FTF	SLP student	Yes^
				Tierney et al., 2016 (Case study)	Indiv.	NS	2–3x week	18 months	45–60 min	–	FTF	SLP	Yes^
				Cumley & Swanson, 1999 (Case study)	Indiv.	NS	Daily (*n* = 1); 2–3x week (*n* = 1); NS (*n* = 1)	6 months (*n* = 1); NS (*n* = 2)	1 hr (*n* = 1); NS (*n* = 2)	–	FTF	SLP	Yes^ (n = 1); NS (n=2)

Note. Partic. = participants; Tx = treatment; Rx = therapy; Multiple BL = multiple baseline; indiv. = individual; / = per; sess. = session; min = minutes; FTF = face-to-face; SLP = speech-language pathologist; NS = not stated; – = not possible to calculate; DTTC = dynamic temporal and tactile cueing; blk = block; AB = treatment + follow-up; ^ = no specific details provided; prod = productions; approx. = approximately; sig. = significant; RCT = randomised controlled trial; ABB = baseline, treatment, treatment; ACB = baseline, treatment 1, treatment 2; @ = not stated, assumed to be SLP; ABACA = baseline, treatment 1, withdrawal, treatment 2, withdrawal; ! = home practice was reported as 'intensive' without further detail; # = home practice was for generalising treatment targets; * = no details regarding home practice outside of the parent-delivered therapy sessions; US = ultrasound; utt. = utterances; comp. = computer; reps = repetitions. [1] Cumulative intervention intensity = dose x dose-frequency x total intervention duration (Warren et al., 2007); average used when a range for dose, frequency or duration were reported. [b] Sessions were between 2–28 days apart.

Dynamic temporal and tactile cueing and integral stimulation

Dynamic temporal and tactile cueing (DTTC) is based on integral stimulation treatment, which was initially developed for use with adults with acquired apraxia of speech (Strand & Skinder, 1999). Integral stimulation involves a series of steps, gradually decreasing the speaker's reliance on the adult's model (Strand & Skinder, 1999). Early reports of this treatment with children used the term 'integral stimulation' (Strand & Debertine, 2000); however, most recent reports have used the term 'DTTC' (e.g. Baas, Strand, Elmer, & Barbaresi, 2008; Maas, Butalla, & Farinella, 2012). DTTC is based on the premise that the underlying difficulty in CAS is a disorder of movement required for speech (i.e., it is a disorder of praxis; Strand & McCauley, 2008). DTTC aims to improve the child's accuracy with the timing and degree of articulator movements. The treatment is dynamic because the clinician varies the degree of cueing following each trial. It aims to reduce the child's reliance on temporal and tactile cues by gradually increasing the time between the clinician's model and child's production and by decreasing the amount of touch cues for the articulators (Strand, Stoeckel, & Baas, 2006).

The efficacy of integral stimulation therapies, including DTTC, for CAS has been demonstrated in seven single-case experimental design (SCED) studies (see Table 2.1). SCED studies may include multiple baseline design, ABAB (A= baseline phase, B=treatment phase), and alternating treatment design. Across all DTTC studies, 17 children received treatment, with 14 demonstrating treatment effect. Some children also generalised to untreated words and phrases, and some children maintained their treatment gain for up to 4 weeks post-treatment. Although DTTC was initially used with a small set of functional stimuli (e.g. Strand & Debertine, 2000), the three later studies used larger stimuli sets and targeted specific sounds rather than functional phrases (Edeal & Gildersleeve-Neumann, 2011; Maas et al., 2012; Maas & Farinella, 2012). These studies still demonstrated a treatment effect for most participants; however, the effect sizes were more modest than those from studies with both functional stimuli and more intense service delivery (Strand & Debertine, 2000; Strand et al., 2006).

DTTC is generally delivered at a higher intensity than other CAS treatments. In the research, it is reported as having been delivered in intensive blocks with up to 10 sessions per week for 6 consecutive weeks (Baas et al., 2008; Strand & Debertine, 2000), and even the lowest intensity reported was two to three times per week for 5 weeks (Edeal & Gildersleeve-Neumann, 2011). All published reports indicate that DTTC was provided individually, face-to-face, by a speech pathologist or speech pathology student. Four of the seven published DTTC treatment reports involved parents in practising speech at home, but limited details of the nature of this practice were provided (Baas et al., 2008; Edeal & Gildersleeve-Neumann, 2011; Strand & Debertine, 2000; Strand et al., 2006).

Concurrent treatment

Concurrent treatment (Skelton & Hagopian, 2014) aims to improve the articulation of specific error sounds. Concurrent treatment uses the levels and tasks within a traditional articulation hierarchy (Van Riper, 1972) but presents the levels to children randomly rather than hierarchically. There are two phases in concurrent treatment (a) establishment of the sound in simple words, and (b) random variable practice of the sound in syllables, words, phrases, sentences and storytelling. Concurrent treatment demonstrated efficacy for three children with CAS aged 4–6 years in a SCED study (Skelton & Hagopian, 2014). All three children showed improvement with the targeted words, untreated words and three-word phrases.

Concurrent treatment is delivered individually, face-to-face, by a speech pathologist in two 30-minute sessions per week, with approximately 100 trials per session.

DuBard Association Method

The goal of the DuBard Association Method is to improve speech intelligibility. It teaches accurate production of individual phonemes, with a series of steps gradually moving to more complex linguistic units (Martin et al., 2016). Tasks are organised hierarchically so that the children experience high levels of success. This approach uses visual, auditory, kinaesthetic and

tactile cues. In order to deliver the DuBard Association Method, a clinician needs to attend an advanced university course or equivalent, consisting of 40 hours of training (Martin et al., 2016). A single pre–post investigation of 12 children aged 3–10 years with CAS and comorbid conditions indicated that 2 years of treatment resulted in significant gains in articulation (Martin et al., 2016). As the investigation did not use an experimental design and did not control for maturation, it is not possible to conclude that the children's improvements were due to the treatment.

Within the published study, the DuBard Association Method was delivered within a special education setting across all subjects for the entire school day, each and every school day for 2 school years. The delivery agent was an SLP, who was also the class teacher.

Rapid Syllable Transition Treatment

The goal of ReST treatment is to improve the motor planning and programming deficits in CAS that cause prosody and speech sound errors (McCabe, Murray, Thomas, & Evans, 2017). It addresses sound consistency, rapid and fluent transitions between syllables and lexical stress (Murray, McCabe, & Ballard, 2012). McCabe et al., (2017) claim that ReST treatment is designed to explicitly treat the three core features of CAS (American Speech Language Hearing Association, 2007b). ReST treatment uses pseudo-word stimuli, as these are thought to enable the child to learn new motor plans and programs without the interference of old, incorrect plans and programs (Murray et al., 2012). The treatment employs motor-learning principles thought to facilitate long-term skill retention and generalisation (Maas et al., 2008; Schmidt & Lee, 2011), such as pre-practice and practice phases within each session, random presentation of stimuli, and a predominance of delayed low-frequency feedback about whether or not the item was correct (McCabe et al., 2017). The efficacy of ReST treatment has been evaluated for 21 children aged 4–12 years, across one randomised controlled trial (Murray, McCabe, & Ballard, 2015) and two published SCED studies (Ballard, Robin, McCabe, & McDonald, 2010; McCabe, Macdonald-D'Silva, van Rees, Ballard, & Arciuli, 2014). There was a significant treatment effect for ReST in the randomised controlled trial and for all seven participants in the published SCED studies,

with generalisation to untreated items and maintenance of gain for 4 months post-treatment (Murray et al., 2015).

All ReST treatment studies have employed individual treatment, delivered face-to-face by a speech pathology student across four 1-hour sessions per week for 3 consecutive weeks, with no home practice.

Prompts for Restructuring Oral Muscular Performance Targets

Prompts for Restructuring Oral Muscular Performance Targets (PROMPT) aims for the speaker to control and organise articulator movement patterns, using tactile-kinaesthetic cues (Hayden, 2006). PROMPT is 'based explicitly on a hierarchical interdependent bottom-up model of speech motor control and development, the Motor Speech Hierarchy' (Dale & Hayden, 2013, p. 645). In order to deliver PROMPT treatment, clinicians require training consisting of a minimum of a 3-day workshop delivered by a PROMPT-certified trainer (The PROMPT Instutute, n.d.). Although PROMPT goals address organisation of movement, they also target language and social functions (Dale & Hayden, 2013).

Positive effects have been shown for children with CAS following PROMPT treatment. Using a pre–post study, Dale and Hayden (2013) demonstrated improvements in articulation test scores, intelligibility and production of untreated words for four children with CAS, aged 3–4 years after 16 sessions of PROMPT treatment. A similar study indicated that children with CAS, aged 2–4 years improved following treatment very similar to PROMPT delivered by non-PROMPT-certified clinicians, with greater improvement by children who had treatment twice per week (20 sessions in total) than once per week (10 sessions in total; Namasivayam et al., 2015).

The service delivery for published CAS PROMPT treatment is two sessions per week, across a minimum of 8 weeks. Session length is approximately 50 minutes, with 45–60 speech production trials per session. Treatment is delivered face-to-face by a speech pathologist, with home practice of the speech targets in functional contexts.

Nuffield Dyspraxia Programme—third edition

The Nuffield Dyspraxia Programme—third edition (NDP-3) aims to improve children's speech production by developing their motor planning and programming (Williams & Stephens, 2004). It is based on a psycholinguistic framework and teaches skills in a hierarchical manner, beginning with isolated phonemes and phoneme sequences and then moving to simple words and lastly to sentences, with success required at foundation levels before progressing to more complex tasks (Williams & Stephens, 2004). Three goals are selected for each child, with at least one targeting new phonotactic structures and at least one targeting sounds (Murray et al., 2012). Each goal is targeted for a short block within each session, with immediate detailed feedback provided on each attempt of each word. The NDP-3 is one of only two CAS treatments that have been evaluated in a randomised controlled trial (RCT, Murray et al., 2015). Thirteen children aged 4–12 years demonstrated large improvement with the treated goals following intense NDP-3 treatment, 4 days per week for 3 consecutive weeks, with significant generalisation to untreated real words (Murray et al., 2015). The participants did not maintain all the reported treatment gain, with small losses from 1 week to 1 month post-treatment, and again from 1 month to 4 months. Murray et al. (2015) hypothesised that the large treatment gain associated with subsequent small loss following treatment withdrawal may be due to the treatment's use of motor-learning principles that facilitate skill acquisition rather than retention (i.e., immediate, detailed feedback on all production attempts; see Maas et al., 2008 for a review of the application of motor-learning principles in speech pathology).

The NDP-3 was evaluated in an intense clinic-based service delivery with no home practice (Murray et al., 2015); however, the recommended service delivery is one or two sessions per week, with regular home practice (Williams & Stephens, 2004). The NDP-3 has also been evaluated as a combined treatment with melodic intonation therapy (MIT) and multimodal cueing (see 'Combined Treatments' below).

Melodic Intonation Therapy

MIT uses prosodic cueing to facilitate spontaneous speech production (Helfrich-Miller, 1994). MIT was initially developed for use with adult patients with neurogenic communication impairments, such as aphasia (Helfrich-Miller, 1994). During MIT, simple phrases are intoned (i.e., produced with a simple melody). The melody aims to follow the natural prosodic contours of the phrase in terms of rhythm, stress and inflection, using only a few notes (Helfrich-Miller, 1994). Over time, the child becomes less dependent on the melodic cues and learns to produce the phrase spontaneously. MIT has been investigated in the treatment of four children with CAS, across two separate case studies (Helfrich-Miller, 1994; Krauss & Galloway, 1982). Although the children showed improvement with speech and imitation following treatment, limitations in study design and reporting mean it is not possible to definitively conclude that the improvements were a result of MIT. MIT was also combined with another treatment in two studies (Martikainen & Korpilahti, 2011; Singh & Trivedi, 2016- see 'Combined approaches' below).

MIT is typically delivered individually by a speech pathologist. Limited details about the intensity variables were provided by Helfrich-Miller (1994). Krauss and Galloway (1982) provided treatment twice-weekly for 2 months, with MIT used for 20% of the session. No information was provided about home practice in either study.

Combined approaches

There are several reports of combined treatments for CAS. It is not immediately clear why individual researchers used combined treatments. It may be that the nature of CAS, with deficits in several areas, is thought to benefit from combined treatments given that most treatments aim to treat only one facet of the impairment. Indeed, combined treatments are not dissimilar to routine clinical practice for many speech impairments, where clinicians report using eclectic treatments (Glogowska et al., 2000; Joffe & Pring, 2008; McLeod & Baker, 2014).

Stimulability with modified Core Vocabulary

This combined approach aimed to improve the consistency of speech productions and expand the child's phonological repertoire (Iuzzini & Forrest, 2010). This treatment directly targeted one of the key difficulties associated with CAS: speech inconsistency (American Speech Language Hearing Association, 2007b). Using a SCED approach, Iuzzini and Forrest (2010) investigated the efficacy of this approach with four children with CAS aged 3–6 years. During each 55-minute session, the child received 10 minutes of stimulability training (Miccio & Elbert, 1996) and 45 minutes of core vocabulary treatment (Dodd, Holm, Crosbie, & McIntosh, 2006), with the core vocabulary words containing complex phonological targets. Following 10 weeks of twice-weekly treatment, all four children expanded their phonetic inventory, and three of the four showed improvement in their speech consistency.

The treatment was delivered individually by an SLP. No information was provided about the number of trials per session or whether home activities were included.

Melodic Intonation Therapy and Touch Cue Method

This combined treatment employed prosodic cues through the application of MIT (Helfrich-Miller, 1994) and tactile cues via the touch cue method (TCM, Bashir, Grahamjones, & Bostwick, 1984) to improve speech sound production and decrease sound sequencing errors. The efficacy of the treatment for improving the speech production of a 4-year-old child with CAS was evaluated (Martikainen & Korpilahti, 2011). The child received 6 weeks of MIT, followed by a 6-week break and then 6 weeks of TCM using a SCED approach. Although the study used an experimental design, it is not possible to conclude that the child's improvements were due to the treatment because the greatest improvement was made during a no-treatment period and the rate of change during the following treatment phase was no greater than during the no-treatment phase.

This combined treatment was delivered individually, face-to-face, across three 30-minute sessions per week. Although the treatment agent was not stated, the assumption is that it was an SLP.

Nuffield Dyspraxia Programme, Melodic Intonation Therapy and Multisensory Cues

The final combined approach used the NDP-3 (NDP-3, Williams & Stephens, 2004), MIT (Helfrich-Miller, 1994) and multisensory cues to improve automaticity and flexibility of articulation (Singh & Trivedi, 2016). This treatment was very similar to the standard NDP-3 (see Nuffield Dyspraxia Program—third edition, above) in terms of goal selection and progression criteria but had the addition of tactile-kinaesthetic and prosodic cues. The authors reported using a SCED approach to investigate the efficacy of the combined treatment for an 8-year-old girl. However, insufficient detail was reported about the child's baseline performance, treatment-phase performance and follow-up performance for the study to be considered a SCED; therefore, in this thesis, it has been classified as a case study. The authors reported that the child improved her automaticity and flexibility with articulation, but limitations in the reporting of data meant it was not possible to evaluate the timing, degree and rate of change within the baseline and treatment phases.

The combined treatment was delivered face-to-face in 60-minute sessions, twice-weekly, across three 18-week blocks. Although the treatment agent was not stated, it was assumed to be an SLP.

Biofeedback

Biofeedback treatments use instrumental means to provide feedback on aspects of physiological functioning (Preston, Brick, & Landi, 2013). Biofeedback techniques such as electropalatography (EPG) and ultrasound provide feedback on the position and movement of the articulators, particularly the tongue, during speech. EPG registers contact between the tongue and areas of the palate via a custom-designed dental plate with inbuilt electrodes, whereas ultrasound uses reflected sound waves via a transducer held below the chin to determine the position of the tongue.

It has been hypothesised that the feedback provided by biofeedback systems compensate for the poor feed-forward mechanism in children with CAS by providing 'knowledge of an individual's performance that can be used to update, modify, and stabilise motor plans for speech' (Preston et al., 2013, p. 628).

Most biofeedback treatments target sound production, but there are many ways the treatments are structured to achieve this broad goal. In some treatments, biofeedback is used for an entire treatment session (Carter & Edwards, 2004), in others for only part of a session (e.g. Preston et al., 2013). The stimuli, feedback type and schedule vary between the treatments using biofeedback. For example, Preston, Maas, Whittle, Leece, and McCabe (2016) used biofeedback via ultrasound in approximately half of each treatment session to target the production of two sound sequences per child (e.g., VC, /ir/; CC, /fl/) in linguistic structures up to sentences. In contrast, Carter and Edwards (2004) used biofeedback via EPG for the whole of every session in a treatment block, targeting all the child's errored consonants (up to 16 error patterns) with up to half the block spent practising the position for the consonant silently.

Electropalatography

Carter and Edwards (2004) demonstrated improvement in consonant production following 10 sessions of EPG treatment for two children with CAS. The study was a pre–post design, with no measures of generalisation or maintenance. EPG was also investigated in a case study of a hybrid treatment approach combining EPG and oral stimulation (Lundeborg & McAllister, 2007). Although the child's percentage of consonants correct improved following the EPG phase of the treatment, the lack of experimental control prevents analysis to determine whether it was the treatment that was responsible for the improvement in speech (Lundeborg & McAllister, 2007).

Ultrasound

Ultrasound has demonstrated mixed efficacy for children with CAS. In a SCED study, twice-weekly ultrasound treatment resulted in all six CAS participants showing improvement with at

least two of their four targeted sounds (Preston et al., 2013). These participants also generalised to untreated exemplars of the sounds and maintained their gains for 2 months post-treatment (Preston et al., 2013). However, when the same treatment was delivered via an intensive format with 2.5 hours of treatment daily for 5 consecutive days across 2 weeks, the treatment was only effective for acquisition, maintenance and generalisation for one of three participants (Preston, Leece, & Maas, 2016). One participant showed generalisation but limited maintenance, and the other showed no evidence of generalisation or maintenance of treatment effect. Limited efficacy was shown for three participants with CAS receiving ultrasound treatment for the articulation of /r/ in twice-weekly sessions (Preston, Maas, et al., 2016). Although one of the three participants improved their production in treatment sessions, none of the three generalised their skill to untreated words, nor maintained the skill post-treatment.

A variety of service-delivery approaches have been reported for biofeedback treatments. Most are delivered by a speech pathologist in a clinic-based context, although one hybrid EPG treatment was delivered by a trained parent at home (Lundeborg & McAllister, 2007). Dose variables have varied, with session length ranging from 30 minutes to 120 minutes, and dose-frequency ranging from once per week to five times per week. The studies that reported a dose per session reported a high dose of at least 150 trials per session. All biofeedback studies used face-to-face delivery. There was no reference to home practice in biofeedback treatment apart from the parent-delivered EPG treatment (Lundeborg & McAllister, 2007).

2.3.1.2 Linguistic intervention

The second group of speech pathology treatments addressing Body Functions (World Health Organization, 2007) are those addressing linguistic-based goals. The treatments aim to improve the language-based skills of children with CAS, such as to improve phonological awareness skills or extend sentence length. In some cases the treatments address what is perceived to be the underlying cause of CAS (e.g. Integrated Phonological Awareness, McNeill, Gillon, & Dodd,

2009a aims to treat impaired phonological plans) while others address language difficulties that are secondary to the motor impairment (e.g. aided AAC modelling, Binger & Light, 2007).

Integrated Phonological Awareness

Integrated phonological awareness (IPA) treatment aims to improve phonological awareness and improve children's speech production (McNeill et al., 2009a). The treatment aims to improve the stability of phonological plans (Marquardt, Jacks, & Davis, 2004) by performing phonological awareness tasks on specific speech targets (McNeill et al., 2009a). By stabilising the phonological plan, IPA treatment aims to improve the child's speech, phonological awareness and literacy skills (McNeill, Gillon, & Dodd, 2010). IPA treatment includes phonological awareness tasks (phoneme segmentation, phoneme manipulation, and letter knowledge) using target words containing a phonological error evident in the child's speech (e.g., /st/ clusters; (Moriarty & Gillon, 2006). The child is asked to produce their target words and to use them in phonological awareness activities.

Four published studies using a SCED approach report the outcomes of IPA treatment for children with CAS (McNeill et al., 2009a; McNeill, Gillon, & Dodd, 2009b; McNeill et al., 2010; Moriarty & Gillon, 2006). One of these studies reports treatment outcomes for three children (Moriarty & Gillon, 2006), while the remaining three studies (McNeill et al., 2009a, 2009b, 2010) report different aspects of the treatment outcome for the same 12 children. Twelve of the 15 children who received IPA treatment improved their speech production, and, as a group, all children improved their phonological awareness skills (McNeill, Gillon, & Dodd, 2009c; Moriarty & Gillon, 2006).

IPA is delivered by a speech pathologist or a trained speech pathology student, individually, in 45-minute sessions, either two or three times per week across a period of 3–12 weeks. There is no information about whether home practice is provided for children receiving IPA treatment.

2.3.2 Treatments addressing Activity and Participation

Treatments that focus on Activities and Participation aim to improve a person's communication skills and involvement in life (ICF-CY, World Health Organization, 2007). This may include the activities of understanding and using language and of using non-verbal communication (Cunningham et al., 2017). It may include participation in interpersonal interactions, school activities and family relationships (World Health Organization, 2007). A description of the treatments addressing Activity and Participation follows (see also Table 2.1).

2.3.2.1 Activity level

All published CAS treatments addressing activity goals use a multimodal approach (see Table 2.1). The basic premise of multimodal intervention is that communication can be achieved via a variety of modalities, not exclusively speech (Beukelman & Mirenda, 2005). Multimodal intervention may include manual signing, picture-based communication boards and books, voice-output communication devices and speech.

Aided Augmentative and Alternative Communication modelling with picture symbols

Aided Augmentative and Alternative Communication (AAC) modelling uses language enrichment techniques, such as modelling, recasts, extensions and expansions (Fey, 1986) within a multimodal context. Treatments in this category aim to extend the length and complexity of symbol communication. Several well-designed SCED studies have demonstrated the efficacy of aided AAC modelling on the linguistic outcomes for children with CAS (Binger, Kent-Walsh, Berens, Del Campo, & Rivera, 2008; Binger & Light, 2007; Binger, Maguire-Marshall, & Kent-Walsh, 2011). Specifically, following treatment, preschool-aged children with CAS and developmental delay spontaneously used multi-symbol messages (Binger & Light, 2007), parents effectively used AAC modelling (Binger et al., 2008), and children began to use symbols for grammatical morphemes in AAC multi-symbol messages (Binger et al., 2011)

Aided AAC modelling was typically delivered in short sessions of 10–15 minutes, one to three times per week for 7–11 sessions (Binger et al., 2011).

Speech-generating device

Other multimodal treatments addressing linguistic skills within the activity domain use speech-generating devices (SGDs). There are two categories of CAS treatments using SGDs. The first uses an SGD to teach children to segment utterances, based on the theoretical understanding that to produce multi-symbol messages one first needs to know how to separate grammatical constituents (e.g. Harris, Doyle, & Haaf, 1996). The second category uses an SGD to provide language modelling (e.g. Lüke, 2016) in a similar way to aided AAC modelling with picture symbols, as described above. In a SCED study, a 5-year-old girl with CAS and non-verbal skills within the normal range demonstrated improvement with her ability to use an SGD to identify linguistic segments and use more expressive language following treatment (Harris et al., 1996). Two observational case studies suggest positive expressive language outcomes for children with CAS following language enrichment treatment using an SGD (Bornman, Alant, & Meiring, 2001; Lüke, 2016).

The service-delivery for multimodal treatments with a linguistic focus was typically an individual dose form, administered face-to-face by an SLP. Dose-frequency was once or twice per week with sessions of 45–60 minutes. Unlike some other CAS treatments, multimodal treatment with a linguistic focus commonly employed home practice. No detail about the amount, frequency or nature of the home practice was provided in any of the studies.

2.3.2.2 Participation level

Treatments that focus on participation aim to enhance a person's ability to participate fully in society (ICF-CY, World Health Organisation, 2007). These types of goals aim to improve the person's use of their present communication skills rather than to improve the linguistic or motoric complexity.

Partners in Augmentative Communication Training

Rather than addressing linguistic skills, some multimodal treatments aim to improve the child's participation in life activities. One such treatment is Partners in Augmentative Communication Training (PACT)[1]. PACT is a training program for parents to learn how to facilitate their child's communication by using an AAC device and to maximise opportunities for the child to participate in communicative interactions (Culp, 1989). The only evidence for PACT comes from a non-experimental pre–post study of an 8-year-old child with CAS (Culp, 1989). The authors report that 2 months after the 3-day PACT training, the child's mother decreased her domination of the interaction, and the child improved her speech intelligibility (Culp, 1989). As with all non-experimental case studies, it is difficult to definitively attribute the changes in the child's communication to the intervention. The service delivery for PACT is a 3-day face-to-face parent-training session run by a speech pathologist, with implementation of the techniques in naturalistic contexts by the trained parent.

Integrated Multimodal Intervention

Integrated Multimodal Intervention (IMI) also aims to facilitate the child's participation in meaningful social interaction through AAC, while also improving the use and accuracy of verbal speech (King, Hengst, & deThorne, 2013). The second aim of IMI is consistent with a focus on Body Functions; however, for the purposes of this thesis, it has been classified as a Participation treatment, based on its primary goal of improving participation in social interaction. IMI is delivered in naturalistic contexts and uses five key techniques: (a) augmented input via AAC; (b) multiple language and AAC models; (c) naturalistic milieu techniques; (d) verbal praise for correct productions; and (e) correction procedures, such as placement cues and requests for imitation (King et al., 2013). The efficacy of IMI was investigated for three children aged 4–9

[1] Two speech pathology treatments use the acronym PACT: (a) Partners in Augmentative Communication Training (Culp, 1989) and (b) Parents and Children Together (Bowen & Cupples, 1999). This section of the thesis refers to Partners in Communication Training (Culp, 1989).

years with CAS in a SCED study (King et al., 2013). Each of the participants increased their participation in communication, as well as their speech production. IMI was delivered face-to-face, twice-weekly across 9–14 weeks. Although the intervention agent was not stated, it was assumed to be a speech pathologist.

Eclectic approaches

Eclectic treatments use a broad and diverse range of approaches and techniques, and are commonly employed during 'business as usual' treatment of paediatric clients (Joffe & Pring, 2008; Roulstone, 2015). Three case studies of multimodal treatment for children with CAS employed an eclectic approach and reported successful outcomes with speech production and social participation following many months, or years, of treatment (Cumley & Swanson, 1999; Tierney, Pitterle, Kurtz, Nakhla, & Todorow, 2016; Watson & Leahy, 1995). For example, Cumley and Swanson (1999) reported that, for an 8-year-old girl with mild cognitive delay, language delay and CAS, daily therapy across a period of 6 months using a combination of cycles approach (Hodson & Paden, 1991), cued speech (Klick, 1994), touch cues (Bashir et al., 1984), mirror work, self-monitoring, picture communication boards, remnant books and a voice-output communication device resulted in a greater willingness to communicate with others. Similarly, Tierney et al. (2016) reported that an eclectic multimodal approach across a period of 18 months for a 3-year-old boy reduced frustration and enabled him to expand his social interaction prior to the emergence of speech. Although case reports such as these provide information about a child's communication over time, the lack of experimental control means it is not possible to attribute change in communication to the intervention approach.

These case studies employed differing service delivery within and across studies. The sessions ranged from daily (Cumley & Swanson, 1999) to fortnightly (Watson & Leahy, 1995), in the clinic (Tierney et al., 2016) or in a combination of clinic and home (Watson & Leahy, 1995) across a period of 6 months to 2 years.

2.4 Service delivery in childhood apraxia of speech treatments

Although a variety of approaches to service delivery were evident with CAS treatments, some trends were observed. The following section discusses these general trends for the service-delivery modifications outlined in Chapter 1: intensity variables, treatment mode, and treatment delivery agent. These trends in service delivery are interpreted with reference to treatment efficacy and expert recommendations.

2.4.1 Intensity variables

Intensity variables are poorly described in the CAS literature. Of the 40 treatment studies reviewed in this chapter—representing 19 separate treatments—only 16 (40%) provided sufficient detail for calculating cumulative intervention intensity (Warren et al., 2007, See Table 2.1). Motor speech treatments generally used a higher dose per session at an average of 110 trials per session (*SD* = 44) compared with 43 trials per session for linguistic treatments (*SD* = 39; see Table 2.1).

More therapy, more frequently, across a longer time period has long been recommended for children with CAS (American Speech Language Hearing Association, 2007b; Hall, Jordan, & Robin, 1993; Skinder-Meredith, 2001). The benefit of a higher number of production trials was confirmed empirically by Edeal and Gildersleeve-Neumann (2011), who demonstrated that for two children with CAS a dose of 100 productions in a 15-minute block was more effective than a dose of 30–40 productions in the same time period. Although most CAS treatments are delivered with high dose-frequency, the empirical benefit of high dose-frequency has not been conclusively demonstrated for CAS. The dose-frequency of treatments examined in this chapter ranged from one to 10 sessions per week, with an average of 3.5 (*SD* = 2.2; See Table 2.1). Namasivayam and colleagues (2015) attempted to investigate the effect of various dose-frequencies for CAS. Unfortunately, they did not control the cumulative intervention intensity; the children who had twice-weekly therapy received 20 sessions, whereas those who received weekly therapy had 10

sessions. It was therefore not possible to conclude that the difference between the two groups was due to the frequency of the sessions rather than to the total number of sessions provided. There are indications that a higher dose-frequency may be more effective in CAS; most treatments use a high dose-frequency, and greater treatment effect was shown in DTTC treatment when delivered across 10 sessions per week (Strand & Debertine, 2000) than three sessions per week (Maas et al., 2012; Maas & Farinella, 2012).

2.4.2 Dose form

Treatment for CAS has almost exclusively been delivered individually in a face-to-face context. Of the 40 treatment studies examined, only one used a group (whole-class) treatment context (Martin et al., 2016), and one used a combination of individual and group sessions (Watson & Leahy, 1995). All published investigations of CAS treatment reviewed in this chapter used face-to-face delivery (see Table 2.1). Given the emergence of telehealth treatment modality across most other domains of speech pathology (Molini-Avejonas et al., 2015; Speech Pathology Australia, 2014b), it is surprising there are no investigations of the efficacy of this approach for CAS.

2.4.3 Delivery agent

In the 40 CAS treatment studies investigated in this chapter, the most frequent delivery agents were an SLP or an SLP student (see Table 2.1). Parents were the primary intervention agent in four of the 40 studies examined (10%). Of these four studies, three were multimodal treatments (Binger et al., 2008; Bornman et al., 2001; Culp, 1989), and one was a biofeedback treatment (Lundeborg & McAllister, 2007). There were no investigations of parent delivery of motor-based treatments that require parents to use auditory perception of their child's productions or to provide cues to improve the child's production attempts. Given that parent delivery of treatment may enable children with CAS to receive a higher intensity of treatment than would otherwise be

possible and may minimise the geographical and financial barriers associated with treatment, it would be beneficial to investigate the efficacy of parent-delivered CAS motor-based treatment.

2.4.4 Empirical investigations of service-delivery approaches

This thesis so far has summarised the serious communication difficulties affecting children with CAS. Although effective treatments exist for these children, they are typically delivered in high-frequency, face-to-face contexts by a speech pathology clinician. However, as a result of structural, geographical and financial barriers, children with CAS are frequently unable to access high-frequency, face-to-face, clinician-delivered treatment. Modifying the service-delivery approach could ease the burden of barriers to treatment access for children with CAS. However, there is scant information available about the impact of such modifications to service delivery for children with CAS.

ReST is one of only two CAS treatments whose efficacy is supported by evidence from a randomised control trial. Like most CAS treatments, ReST aims to address motor skills in the ICF domain of Body Function. ReST treatment is claimed to remediate the planning and programming impairment identified in the American Speech Language Hearing Association (2007a) consensus definition (McCabe et al., 2017). To date, all investigations of ReST treatment have used only one service-delivery approach: face-to-face delivery, four times per week, by a speech pathology clinician.

The following chapters seek to address the service-delivery gap in the CAS literature by reporting the efficacy of ReST treatment using service-delivery approaches that modify one of (a) intensity of sessions, (b) treatment mode, or (c) delivery agent. Chapter 3 reports on an investigation of intensity modification—the use of lower dose-frequency; Chapter 4 reports on mode modification via telehealth treatment; and Chapter 5 reports on delivery-agent modification via a combined clinician–parent delivery model. In each chapter, only the variable of interest was modified. In all cases, children received twelve 1-hour sessions of ReST treatment, with 100 treatment trials

per session, to facilitate comparison across studies and with the existing literature on ReST treatment (McCabe et al., 2014; McCabe, McDonald-D'Silva, van Rees, Arciuli, & Ballard, 2010).

Chapter 3: Intensity Modification—Low Dose-Frequency Rapid Syllable Transition Treatment

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I declare that I made the following contribution to the study:

- Conception of the research question in collaboration with the other authors
- Design of the study with the other authors
- Collection of data
- Data entry and analysis
- Writing of the first draft of the manuscript
- Journal revisions and resubmission

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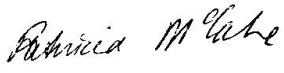
Sign: 

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Rapid Syllable Transitions (ReST) treatment for Childhood Apraxia of Speech: The effect of lower dose-Frequency

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ABSTRACT

This study investigated the effectiveness of twice-weekly Rapid Syllable Transitions (ReST) treatment for Childhood Apraxia of Speech (CAS). ReST is an effective treatment at a frequency of four sessions a week for three consecutive weeks. In this study we used a multiple-baselines across participants design to examine treatment efficacy for four children with CAS, aged four to eight years, who received ReST treatment twice a week for six weeks. The children's ability to acquire new skills, generalize these skills to untreated items and maintain the skills after treatment was examined. All four children improved their production of the target items. Two of the four children generalized the treatment effects to similar untreated pseudo words and all children generalized to untreated real words. During the maintenance phase, all four participants maintained their skills to four months post-treatment, with a stable rather than rising profile. This study shows that ReST treatment delivered twice-weekly results in significant retention of treatment effects to four months post-treatment and generalization to untrained but related speech behaviors. Compared to ReST therapy four times per week, the twice-weekly frequency produces similar treatment gains but no ongoing improvement after the cessation of treatment. This implies that there may be a small but significant benefit of four times weekly therapy compared with twice-weekly ReST therapy.

Learning outcomes: Readers will be able to define dose–frequency, and describe how this relates to overall intervention intensity. Readers will be able to explain the acquisition, generalization and maintenance effects in the study and describe how these compare to higher dose frequency treatments. Readers will recognize that the current findings give preliminary support for high dose–frequency CAS treatment.

1. Introduction

Childhood Apraxia of Speech (CAS) is a complex speech disorder that affects movement control required for accurate articulation of speech sounds and production of prosody (American Speech-Language-Hearing Association, 2007). There is general agreement that children with CAS are perceived to have inconsistent phonetic errors in words over multiple productions, lengthened and disrupted co-articulatory transitions, and more equal stress in multisyllabic words and phrases (American Speech-Language-Hearing Association, 2007). CAS is a persistent speech impairment (Lewis, Freebairn, Hansen,

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Iyengar, & Taylor, 2004) and children with CAS are reported to have slow response to therapy (Aram & Nation, 1982; Hall, Hardy, & LaVelle, 1990).

One intervention approach that has been shown effective for CAS is Rapid Syllable Transitions (ReST) treatment (Murray, McCabe, & Ballard, 2012b). ReST uses a high dose–frequency (4 sessions a week for 3 weeks) to target the key problems of CAS (Murray, McCabe, & Ballard, 2012c). However, high dose–frequency protocols are often impractical in clinical settings due to various logistical challenges. The purpose of the current study was to evaluate the effectiveness of a lower dose–frequency of ReST (2 sessions a week for 6 weeks) that may be more feasible for implementation in clinical environments.

1.1. Treatments for Childhood Apraxia of Speech

Although a number of treatments exist for CAS, (see Murray, McCabe & Ballard, 2014) there is a paucity of high-level evidence supporting treatment efficacy (Morgan & Vogel, 2008; Watts, 2009). Most of the evidence supporting interventions for CAS comes from case studies and case-series designs, which are limited in their application to the population as a whole (Kazdin, 2011). To date, only one randomized controlled trial (RCT) has been conducted in this field (Murray et al., 2012c). The RCT compared the efficacy of the Nuffield Dyspraxia Programme–Third Edition (Nuffield) and Rapid Syllable Transition Treatment (ReST). Both treatment approaches resulted in significant gains on treated speech behaviors and generalization of treatment effects to untreated behaviors. Murray et al. suggests that, with intensive treatment, both approaches stimulated significant change in speech skills but the ReST treatment resulted in stronger retention of skill long-term.

ReST is based on principles of motor learning derived primarily from studies of limb motor-learning (Schmidt & Lee, 2011) and the protocol has been described in detail previously (Murray et al., 2012b). ReST treatment consists of high intensity practice of randomly presented pseudo words, with varying phonetic structure and lexical stress. Using pseudo words enables the children to practice motor planning and programming on word-like forms without interference from previously incorrectly learned plans. During practice, only delayed low-frequency 'knowledge of results' feedback is provided to combine active learning through self-evaluation. These various principles of practice and feedback structure tend to generate stronger retention and generalization of trained skills for both limb and speech motor learning (Maas et al., 2008; Schmidt & Lee, 2011). Efficacy of ReST has also been demonstrated in a number of single-case design studies (Ballard, Robin, McCabe, & McDonald, 2010; McCabe, Macdonald-D'Silva, Van Rees, Ballard & Arciuli, 2010). All studies to date have employed a therapy regimen of 1 h a day for 4 days a week over 3 weeks. However, it is not currently known whether this high dose–frequency is necessary for efficacy.

1.2. Intensity of treatment

Speech–language pathology treatment is generally recommended at a higher frequency and for a longer duration for CAS than for other speech disorders (American Speech-Language-Hearing Association, 2007; Hall, Jordan, & Robin, 1993; Skinder-Meredith, 2001). Many treatments have demonstrated some level of efficacy in high intensity formats (see Murray et al., 2014 for a review), however it is not yet known whether comparable gains can be achieved for similar amounts of therapy delivered with lower frequency (e.g. twice-weekly for 6 weeks).

In order to provide efficient and effective speech–language pathology services for children in general, and those with CAS specifically, it is essential to determine optimal treatment intensity (Baker, 2012). As noted by Law, Zeng, Lindsay, and Beecham (2012) providing too many treatment sessions wastes scarce resources, but providing too few sessions risks diluting the intervention's effect. In the past few years speech–language pathologists have become increasingly aware of the importance of controlling a treatment's intensity (e.g. Baker, 2012; Ukrainetz, 2009).

To facilitate comparisons of intensity across intervention approaches, Warren, Fey, and Yoder (2007) proposed a formula for calculating intervention intensity. The formula takes into account (a) dose—the 'active ingredient' in the treatment such as the number of productions by a client or the number of teaching episodes per session, (b) dose form—the type of activity in which the dose is delivered including the context (e.g. individual vs. group) as well as the type of activity (e.g. focused practice on a motor skill vs. incidental practice during play), (c) dose–frequency—the frequency of the therapy, normally measured in number of sessions per week, month or school term, (d) total intervention duration – the length of the treatment, generally in weeks or months, to determine the cumulative intervention intensity – a product of dose, dose–frequency, and total intervention duration. For example, in previous studies of the ReST treatment (Ballard et al., 2010; Murray et al., 2012c), participants produced 100 responses per session, had four sessions per week across a three-week period for a cumulative intervention intensity of $100 \times 4 \times 3 = 1200$ trials.

A greater amount of therapy ('cumulative intervention intensity') is generally associated with superior outcomes for motor learning tasks (Schmidt & Lee, 2011) and for a range of speech and language impairments. This was demonstrated in children with CAS by Namasivayam (2013), who showed that twenty sessions of 'specialized motor treatment' across 10 weeks resulted in stronger gains than ten sessions across the same period.

1.2.1. Dose–frequency

Even when the *amount* of therapy ('cumulative intervention intensity') is constant, the spacing of the sessions ('dose–frequency') can affect the treatment outcome, (e.g. Allen, 2013; Barratt, Littlejohns, & Thompson, 1992). There has been

very little research into the effect of dose–frequency for pediatric speech impairments, and none that we are aware of that investigates dose–frequency in pediatric motor speech disorders. In phonological treatment, a multiple oppositions approach (Williams, 2000) was more efficacious at a high dose–frequency (i.e. 3 sessions per week for 8 weeks) than a low dose–frequency (i.e. 1 session per week for 24 weeks) (Allen, 2013).

In acquired motor speech disorders, dose–frequency has been reported to have no impact on treatment efficacy. There was no difference in the outcome of Lee Silverman Voice Treatment (LSVT®) for adults with Parkinson's disease when delivered 4 times per week for 4 weeks compared with 2 sessions a week for 8 weeks (Spielman, Ramig, Mahler, Halpern, & Gavin, 2007; Wohlert, 2004). Similarly, dose–frequency had no significant effect on the outcome of sound production treatment (SPT) for adults with apraxia of speech (AOS) when delivered 16 sessions per week for 1 week or 3 sessions per week for 5.5 weeks (Wambaugh, Nessler, Cameron, & Mauszycki, 2013). However, compared to traditional practice, frequency conditions in these three studies were relatively high and a low frequency condition (e.g. one session per week) was not included.

In summary, although high intensity treatments for CAS are efficacious, we do not currently know whether high intensity is essential as there has been no research into the impact of dose–frequency for CAS or other pediatric motor speech impairments. Within adult motor speech impairments, dose–frequency appears to have no effect when comparing dose–frequencies of two or more sessions per week. However, high dose–frequency phonology treatment was more efficacious than a low dose–frequency treatment (Allen, 2013), suggesting that dose–frequency has different effects across linguistic and motor domains or in children versus adults.

1.3. Service delivery

High dose–frequency treatments are demanding in terms of clinician time, family commitment, and client energy and attention. Children are often able to access only limited service, particularly within public health settings (Baker & McLeod, 2011; Bercow, 2008; Ruggero, McCabe, Ballard, & Munro, 2012) and the frequency of sessions provided is often less than desired by parents (Bercow, 2008; Ruggero et al., 2012). Children in Germany, Australia and Canada most typically receive therapy once a week (Ruggero et al., 2012) while those in the US most typically receive therapy twice a week (Mullen & Schooling, 2010). As most children are unable to access the high frequency sessions recommended for CAS and used in ReST, it is important to determine whether the efficacy of ReST treatment is affected by dose–frequency.

1.4. Purpose

The present study aimed to investigate the effect of a dose–frequency of twice a week on the efficacy of ReST intervention for CAS, while maintaining the cumulative intervention intensity of previous ReST studies (1200 practice trials over 12 sessions). We hypothesized that low-frequency ReST treatment would result in:

(a) Improved segmental and prosodic accuracy for treated pseudo words.
(b) Improved segmental and prosodic accuracy for untreated but related pseudo words and real words.
(c) Maintenance of gains at 1 week, 4 weeks and 4 months post-treatment.
(d) No change in articulation accuracy on the Goldman–Fristoe Test of Articulation–2 (Goldman & Fristoe, 2000).

2. Method

The research project was granted ethical approval by The University of Sydney Human Ethics Committee—approval number 15069.

2.1. Participants

Four monolingual Australian English speaking children with a diagnosis of CAS aged 4:8 (years:months) to 8:0 participated in this study. Each child had participated in the treatment study of Murray et al. (2012c) 9 to 13 months earlier, but had not received ReST treatment. The children were recruited via circulation of flyers on the university campus and electronically to speech–language pathologists and parents interested in CAS treatment research. Inclusion criteria were (a) a consensus diagnosis of CAS (see below), (b) normal receptive vocabulary, hearing acuity, and oral structure. The tests used and the participants' performance are shown in Table 1.

The diagnosis of CAS was made by the first two authors based on the consensus core perceptual features of CAS; inconsistent errors on repeated productions of the same words, difficulty transitioning between syllables within words and prosodic difficulties (American Speech-Language-Hearing Association, 2007). Operationally, these perceptual features were defined following Murray, McCabe, and Ballard (2012a) (1) inconsistency score of at least 40% on the Inconsistency subtest of the Diagnostic Evaluation of Articulation and Phonology (DEAP, Dodd, Zhu, Crosbie, Holm & Ozanne, 2006), (2) a minimum of 12 words with perceptually identified syllable segregations in the 50 word Test of

Table 1
Participants' initial assessment results.

Test	F1			M1			M2			F2		
	Value	PR	Result	Value	PR	Result	Value	PR	Result	Value	PR	Result
CELF P2 or 4[1]												
Receptive language index	132	98	>NL	115	84	WNL	70	2	<NL severe	103	58	WNL
Expressive language index	91	27	WNL	98	45	WNL	45	>0.1	<NL severe	68	2	<NL severe
Peabody picture vocabulary test[2]	105	63	WNL	121	92	>NL	93	32	WNL	94	34	WNL
GFTA 2[3]	93	17	WNL	97	22	WNL	<40	<1	<NL severe	55	1	<NL severe
DEAP inconsistency assessment[4]	40	–	Inconsistent	48	–	Inconsistent	60	–	Inconsistent	88	–	Inconsistent
Test of polysyllables[5]												
% Consonants correct	77	–	–	81	–	–	54	–	–	26	–	–
% Vowels correct	91	–	–	86	–	–	71	–	–	59	–	–
% Phonemes correct	83	–	–	83	–	–	61	–	–	40	–	–
Stress pattern errors[6]	11	–	–	14	–	–	24	–	–	34	–	–
Syllable segregations[7]	21	–	–	12	–	–	30	–	–	23	–	–
Oral and motor speech protocol[8]												
Structure	24	–	WNL	24	–	WNL	22	^	–	24	–	WNL
Function	105	–	<NL severe	99	–	<NL moderate	88	^	–	71	–	<NL severe
Observations	Strength and range of movement WNL. Inco-ordination during DDK tasks			Difficulty co-ordinating movements and imitating multisyllabic words			Slow speech rate, inconsistent hypernasality, reduced intonation			Inconsistent hypernasality. Reduced loudness. Mild reduction in rate and range of bilabial and labiodental consonants consistent with mild bilateral flaccid dysarthria		

[1] Dependent on age, participants had either Clinical Evaluation of Language Fundamentals–Fourth Edition [CELF 4], (Semel et al., 2006); or Clinical Evaluation of Language Fundamentals–Preschool Second Edition [CELF–P2] (Wiig et al., 2006).
[2] Dunn and Dunn (2007).
[3] GFTA 2 = Goldman Fristoe Test of Articulation - Second Edition (Goldman & Fristoe, 2000).
[4] DEAP = Diagnostic Evaluation of Articulation and Phonology (Dodd et al., 2006).
[5] Gozzard et al. (2006).
[6] Identified perceptually as a mismatch between the target stress pattern and the stress pattern produced.
[7] Identified perceptually as an absence of smooth joining of the syllables within a word.
[8] (Robbins & Klee, 1987), PR = Percentile Rank, WNL = Within Normal Limits, NL = Normal Limits.
^ = outside of age range for normative scores.

Polysyllables (Gozzard, Baker, & McCabe, 2006) and (3) a minimum of 7 words with perceptually identified stress pattern errors in the Test of Polysyllables (Gozzard et al., 2006).

None of the participants had any neurological diagnoses, however F2 had a mild bilateral flaccid dysarthria of no known origin. All of the children had previously received SLP services, but had no outside speech therapy during the 6-month period of treatment trial.

2.2. Design

A multiple-baseline across participants and behaviors design (Kazdin, 2011) was used in this study. Production of treated and untreated items was assessed between 3 and 6 times during the baseline phase, and probed immediately prior to treatment sessions three, seven and eleven and at one day, one week, four weeks and four months post-treatment.

2.3. Probe stimuli

A probe list with 120 items was selected to allow for (1) analysis of treatment effect, (2) generalization to related, but untreated items, and (3) generalization to real words. The probe stimuli included pseudo word strings with strong-weak (SW) stress patterns (e.g. /kabəfi/ and weak-strong (WS) patterns (e.g. /dəbəfi/). The consonants selected for the pseudo word stimuli were /k/,/b/,/d/ and /f/; as these represent different place, manner and voicing conditions (Ballard, 2001; Ballard, Maas, & Robin, 2007). The vowels used in the pseudo word strings were /ə/,/a/,/i/, and /ə/.

Participants M1, F1 and M2 had the same probe stimuli: 20 SW and 20 WS 3 syllable (CVCVCV) pseudo words, of which 10 SW and 10 WS items were randomly selected for treatment and the other 10 were kept to assess generalization to untreated items. The 10 treatment items of each set were randomly selected and each participant had a different randomly selected 10 items for each set. The probe set included 20 carrier phrases (e.g. I want a _____) with the 3 syllable strings (10 treated items and 10 untreated) and 20 two syllable real words (10 SW and 10 WS), 20 three syllable real words (10 SW and 10 WS) and 20 four syllable real words (10 SW and 10 WS).

Due to the severity of F2's speech difficulties, her treatment stimuli were two syllable pseudo words, and her probe list contained 20 SW and 20 WS two syllable (CVCV) pseudo words, with 10 SW and 10 WS items randomly selected for treatment and 10 of each kept to assess generalization to untreated items. Her probe list also included 20 carrier phrases with the two syllable (CVCV) strings (10 treated items and 10 untreated) and 20 two syllable real words (10 SW and 10 WS) and 20 three syllable real words (10 SW and 10 WS). Single syllable real words were not included due to the absence of stress contrast within these words.

2.4. Equipment

All sessions were recorded using a digital voice recorder and video recorded with a Cinde 88 audio-visual—system for later reliability, fidelity and student training purposes. Additionally the baseline and probe sessions were recorded with an AKG C520 headset microphone at 5 cm mouth-to-microphone distance and Roland Quad Capture UA-55.

2.5. Procedure

The first author carried out eligibility assessment and baseline probes. Trained graduate speech–language pathology students conducted the treatment under the supervision of the first and second authors, who are both qualified speech–language pathologists. The participants were randomly assigned to a student, who treated each child for the duration of the treatment phase. Clinicians who were unfamiliar with the participant conducted each probe session, and no child saw the same clinician twice for a probe assessment.

2.5.1. Baseline and probe sessions

The procedure for the baseline and the probe sessions was identical. Each child imitated all items on their probe list. The probe items were presented in one of three randomized orders, randomly selected. The participants viewed a Powerpoint slide show, with the orthography for each pseudo word item and orthography plus picture for each real word item. To ensure consistency of the pseudo word models, the slide for each probe item included a prerecorded sound file by a native Australian English female speaker. The Goldman-Fristoe Test of Articulation–2 (GFTA-2) was conducted prior to baseline testing and at 1 month post-treatment as a measure of experimental control. Although it could be argued that change on the GFTA-2 could be expected as a result of the feedback on speech sounds, we predicted ReST treatment would not improve *articulation* of specific sounds and use of phonological processes in simple words.

2.5.2. Treatment

The Rapid Syllable Transitions Treatment (ReST) was used, following Murray et al. (2012b). Consistent with the Principles of Motor Learning Approach (Schmidt & Lee, 2011), each session began with approximately 10 min of pre-practice to explain the task and ensure the participants had a reference of correctness for the target stimuli. The participant attempted production of the target stimuli and was given immediate feedback for each production about the

elements that were correct and the changes that were required to make the production more accurate (i.e. Knowledge of Performance, or KP, feedback). Participants were given cues and support to improve their production including: modeling, phonetic placement cues, segregation of syllables and rejoining, visual representation of relative syllable length, and reduced speed of production. Once five items were produced with modeling and shaping, the participant moved into the practice phase. The pre-practice phase lasted for up to 25 min in sessions 1 and 2 and approximately 10 min in sessions 3–12.

In the practice phase, each participant completed 100 trials: 5 trials each of the 20 treated items, in random order. The items were presented in written form, each on an individual PowerPoint slide. Knowledge of results (KR) feedback was provided on approximately 50% of the items, beginning with 100% feedback on the first 10 items, 90% of the next 10 items and fading to 10% feedback on the final 10 items. KR feedback was provided after a delay of 3–5 s. After every 20 trial items, a 2-min rest break was provided.

Clinicians phonemically transcribed the participant's response online and coded whether the target stress pattern was produced and syllable transitions were fluent/without hesitation or segregation. If all elements were scored as accurate, the item was counted as correct. Once a participant achieved $\geq$80% correct on two consecutive practice sessions, the client was moved to the next most complex treatment level. If a participant achieved $<$5% correct in the practice phase of two consecutive sessions the goals were made less complex. Participant M1 met the progression criteria in session 8 and was treated on pseudo words at the end of carrier phrases (e.g. ''she has a big /bəfadi/'' or ''there's a /kədəfi/'') from session 9. Participant F2 obtained less than 5% correct in her first two sessions and from session 3 was required to meet only two of the three criteria for a correct response (i.e. correct sounds and stress pattern) on her treatment items.

2.5.3. Data analysis

Each probe item and treatment item was perceptually judged as correct or incorrect on the accuracy of the phonemes, stress pattern, and fluency of syllable transitions. To avoid confounds from increasing familiarity with the child's speech, each examiner conducted, transcribed, and scored only one probe assessment with each child. Intra-and inter-rater reliability was conducted on all probe assessments and treatment sessions.

Data for each participant were graphed for visual analysis. Prior to calculation of statistics, all data were screened by testing autocorrelations of the residuals, as the presence of autocorrelation would prohibit the use of parametric statistics. In all cases the correlation between residuals between adjacent time points (lag 1 autocorrelation) was nonsignificant. For each participant, ANOVAs and Helmert planned orthogonal contrasts were performed across phases (i.e. baseline, treatment, follow-up) within behaviors (i.e. treated pseudo words, untreated pseudo words, and untreated real words with the same number of syllables, fewer syllables and more syllables). Contrasts were a) average performance in the baseline phase compared with later phases (i.e. treatment and follow-up pooled) within participants, and b) average performance in treatment phase compared with the follow-up phase within participants. Effect sizes were calculated using the protocol advocated by Beeson and Robey (2006): d_2 = (mean score post-treatment – mean score pre-treatment/pooled standard deviation). To further examine the performance of the participants within the maintenance phase, post hoc planned orthogonal contrasts were performed on the data points within the maintenance phase, comparing performance at (a) one day post-treatment with later points (i.e. one week-, one month-, and four months post-treatment combined), (b) one-week post-treatment with later points (i.e. one-month-, and four-months-post-treatment combined) and (c) one month post-treatment with four months post-treatment.

The Standard Score for the 'Sounds in Words' subtest of the Goldman-Fristoe Test of Articulation–2 (Goldman & Fristoe, 2000) at pre-and post-treatment were compared to determine whether the post-treatment score was within the 95% confidence interval of the pre-treatment score.

2.5.4. Reliability

A randomly selected 20% of each probe session was analyzed for inter-and intra-rater reliability of phonemic transcription and scoring of articulation accuracy, stress pattern and fluency of syllable transitions. Intra-rater and inter-rater point-to-point agreement for phonemic transcription of responses to experimental probes was 89.6% (SD = 4.06) and 85.8% (SD = 4.12), respectively. Average intra-rater and inter-rater agreement for judgments of accuracy, stress, and fluency were 93% (SD = 3.19) and 89% (SD = 2.49), respectively.

2.5.5. Treatment fidelity

Treatment fidelity, between the clinician and the first author, was calculated for a random 20% of each session on the accuracy of the model provided by the clinician, the number of trials given feedback, the accuracy of the feedback, the type of feedback (i.e. KP in pre-practice and KR in practice) and the delay of feedback. Average fidelity was 94.43% (SD = 9.8).

3. Results

Individual results are presented for each child, followed by post hoc group evaluations of performance within the maintenance phase. The results of the statistical analyses are shown in Table 2.

Table 2
Planned contrasts and effect sizes.

	Word set	Effect size	BL vs. T and FU		T vs. FU		Direction of change T to FU
		d_2 =	t =	p =	t =	p =	
F1	Treated non-words	8.973	4.27	0.004**	1.16	0.279	Stable
	Untreated non-words	3.918	2.99	0.02*	2.99	0.055	Stable
	Untreated real words-2 syllables[1]						
	Untreated real words-3 syllables[1]						
	Untreated real words-4 syllables	6.445	3.98	0.005**	2.39	0.049*	Up
M1	Treated non-words	6.574	6.00	0.001**	0.94	0.379	Stable
	Untreated non-words	2.391	2.56	0.038*	0.33	0.750	Stable
	Treated non-words in phrases	2.752	0.38	0.005*	0.10	0.504	Stable
	Untreated non-words in phrases	0.240	1.56	0.16	1.69	0.128	Stable
	Untreated real words-2 syllables	1.876	4.17	0.004**	1.85	0.109	Stable
	Untreated real words-3 syllables	10.678	18.15	<0.001**	4.91	0.002*	Down
	Untreated real words-4 syllables	4.522	5.50	0.001**	0.09	0.930	Stable
M2	Treated non-words	4.459	2.41	0.48*	0.94	0.372	Stable
	Untreated non-words	1.386	1.29	0.257	0.23	0.800	Stable
	Untreated real words-2 syllables	0.805	5.04	0.002**	2.13	0.070	Stable
	Untreated real words-3 syllables	3.681	2.83	0.024*	1.96	0.093	Stable
	Untreated real words-4 syllables	2.449	39.52	0.006**	2.70	0.025*	Down
F2	Treated non-words	2.449	5.34	0.001**	1.22	0.267	Stable
	Untreated non-words	1.044	1.22	0.257	0.27	0.800	Stable
	Untreated real words-2 syllables	1.876	3.04	0.024*	1.96	0.013*	Down
	Untreated real words-3 syllables	5.196	41.76	0.004**	0.42	0.668	Stable

Effect size = Cohen's d_2 using weighted averages of baseline and follow up variances formula (Beeson & Robey, 2006). BL = baseline phase, T = treatment phase, FU = follow-up phase.
Helmert planned orthogonal contrasts and effect sizes across all participants and all conditions.
[1] F1's real words-2 syllables and real words-3 syllables are not reported because the data showed autocorrelation.
* Significant at 0.05.
** Significant at 0.01.

3.1. Effects of treatment

For F1, during the baseline phase her percent accuracy with the to-be-treated items ranged from 0% to 15% with no trend toward improvement (see Fig. 1, panel A). During the treatment phase her treated pseudo words improved, with accuracy of 20% to 90%. Planned contrasts confirmed that the improvement from baseline to the later phases (treatment and follow-up) was significant.

For M1, during the baseline phase his percent accuracy with the to-be-treated items ranged from 20 to 45% and although there was variability there was no trend toward improvement (see Fig. 2, panel A). During the treatment phase, percent accuracy ranged from 55 to 80% resulting in a significant difference between baseline performance and later phase performance indicative of a significant treatment effect. As M1 reached the a priori criterion of 80% accuracy on treated behaviors over two consecutive sessions in the eighth treatment session, the therapy target changed from single pseudo words to pseudo words in carrier phrases for remaining treatment sessions. As can be seen (see Fig. 2, panel B), during baseline his percent accuracy on treated pseudo words in carrier phrases ranged between 0 and 20% with no trend toward improvement. In probes 5 and 6, performance started to improve, suggesting generalization of treatment effects (see below). Performance in probe 7 did not show any further improvement.

For M2, percent accuracy with the to-be treated items during baseline was 0–5% (see Fig. 3, panel A). Within the treatment phase his percent accuracy improved to 35%, resulting in a significant difference between the baseline phase and the later phases.

For F2, performance during the baseline phase was stable at 0% correct for the to-be-treated pseudo words (see Fig. 4, panel A). During the treatment phase these pseudo words ranged in accuracy from 20 to 30% correct, representing significant improvement from baseline.

3.2. Generalization of treatment effects

F1 generalized her skills to similar but untreated pseudo words, as shown in Fig. 1, panel A. During baseline her accuracy with these items ranged from 5% to 15% with no trend toward improvement, and during treatment performance improved from 20% to 45%. Planned contrasts confirmed that her accuracy was significantly better in the treatment and follow-up phases than the baseline phase. Production of real words also improved; during baseline, there was a trend toward improvement for accuracy of 2 syllable real words from 25 to 60% and for 3 syllable real words from 20 to 40% and these items were therefore not analyzed further. Four syllable words however were stable in baseline with percent accuracy between 10 and 15, improving during treatment from 10 to 50%, with planned contrasts confirming that the change from baseline to later phases was significant.

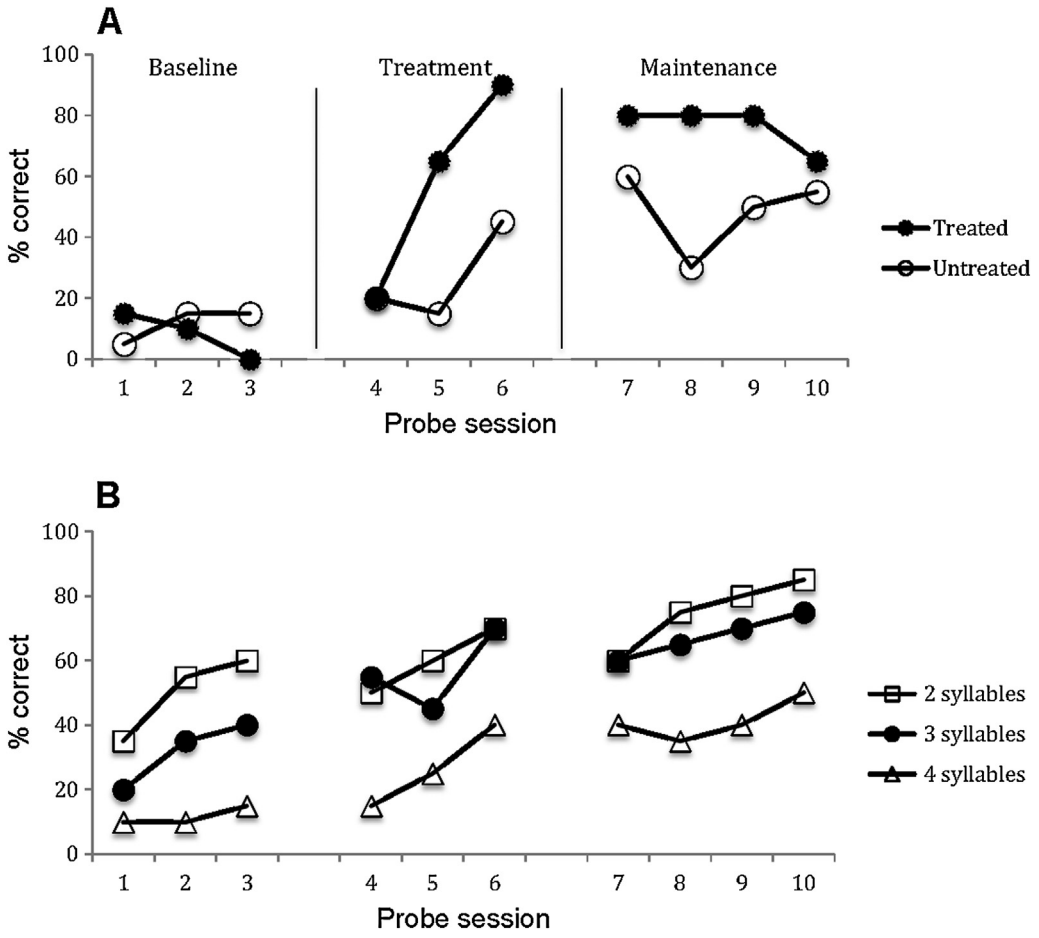

Fig. 1. F1 results. Panel A—treated and untreated pseudo words. Panel B—untreated real words.

M1 generalized his skill to similar, but untreated pseudo words. In baseline, his accuracy with these items ranged from 30 to 45% (see Fig. 2, panel A) with no clear trend toward improvement and during the treatment phase his performance improved significantly to between 55% and 85% accuracy. M1 also generalized his skill to real words with two-, three- and four-syllables (Fig. 2, panel C). During baseline his percent accuracy was 40–60%, 40–45% and 10–25%, respectively, with no trend toward improvement for any of these real-word lengths. During the treatment phase he achieved accuracy scores of 80–85% for 2 and 3 syllable real words and 35–50% for 4 syllable real words. As treatment shifted to treated pseudo words in carrier phrases after probe 5, performance with treated words in carrier phrases in probe 6 was compared to the first five probes to establish presence of a generalization effect. No significant difference was shown for either treated or untreated pseudo words in carrier phrases between the first 5 probe sessions and the subsequent baselines, however there was a significant difference between his performance on treated pseudo words in carrier phrases between the first three probes and subsequent probe sessions, suggesting generalization to treated carrier phrases once treatment began on the pseudo words (Fig. 2, panel B). No significant difference was shown between untreated pseudo words in carrier phrases between the first three probe sessions and subsequent probe sessions.

M2 appeared to show a trend toward improvement in the treatment and follow-up phases for similar-but—untreated pseudo words this change was not statistically significant. M2 had significant generalization to real words with two-, three- and four-syllables, as shown in Fig. 3, panel 2. With regard to two syllable real words, M2 had baseline accuracy of 5–20% with no trend toward improvement, followed by treatment phase accuracy of 40–70%, with significantly better performance in the treatment and follow-up phases than the baseline. With three syllable real-words, M2 had baseline accuracy of 0–5% with no trend toward improvement and treatment phase accuracy of 0–15% with significantly better performance in treatment and follow-up phases relative to baseline. With four syllable real words, M2 had zero accuracy in baseline and 0–15% during treatment with planned contrasts confirming that performance in treatment and maintenance phases was significantly better than the baseline phase.

Although Fig. 4, panel A shows F2 had a general improvement with untreated pseudo words in the treatment and follow-up phases, this change was not significant. She did however show significant generalization to untreated real words with two

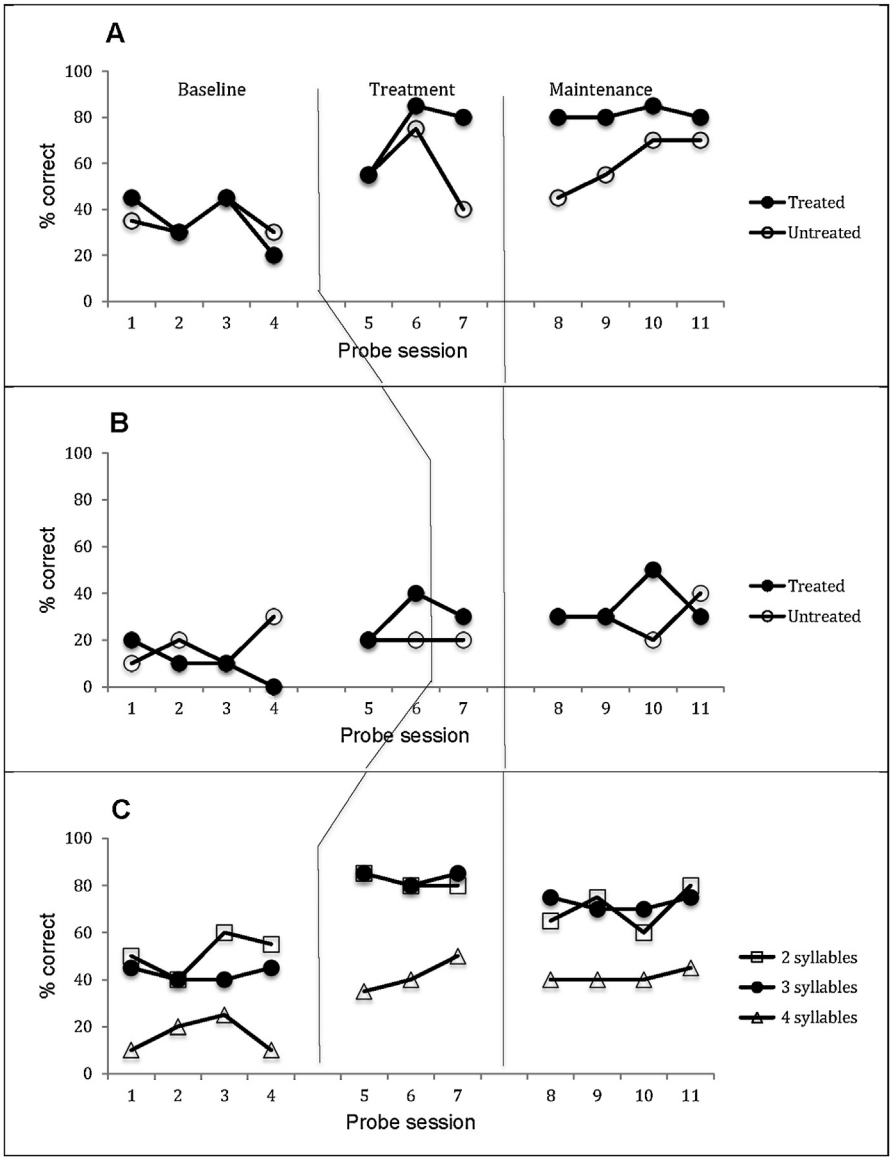

Fig. 2. M1 results. Panel A—treated and untreated pseudo words. Panel B—treated and untreated pseudo words in carrier phrases. Panel C—untreated real words.

and three syllables, as shown in Fig. 4, panel B. After baseline percent accuracy of 0–10 for two syllables with no trend toward improvement and flat 0% for three syllable real words, F2 had accuracy of 10–20% and 5–10% with these items respectively in the treatment phase.

3.3. Maintenance of treatment effects

For F1, at all four follow-up points (one day, one week, one month and four months) her percent accuracy was higher than baseline levels for treated pseudo words, similar but untreated pseudo words and untreated real words (Fig. 1, panels A and B). Planned contrasts revealed no significant difference between performance in the treatment and maintenance phases with treated items or untreated pseudo words, and a significant improvement in her performance with 4 syllable real words supporting maintenance of effects to 4 months post-treatment.

For M1, at all four follow-up points his percentage accuracy with treated pseudo words, sentences with pseudo words and untreated real words with 3 and 4 syllables was higher than baseline levels (Fig. 2, panels, A–C). Three of the four follow-up points were higher than baseline levels for untreated pseudo word and real words with two syllables. Planned contrasts

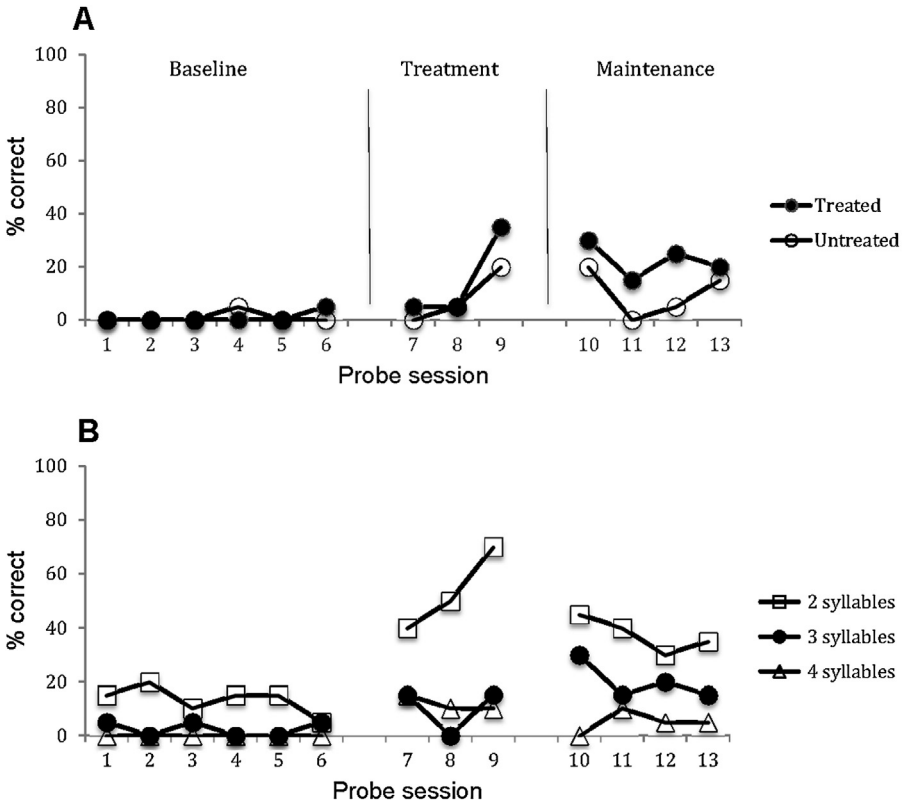

Fig. 3. M2 results. Panel A—treated and untreated pseudo words. Panel B—untreated real words.

revealed no significant difference between his performance in the treatment and maintenance phases with treated items or untreated items, sentences with treated pseudo words, two syllable real-words or 4 syllable real-words, indicating maintenance of skill until four months post-treatment. M1 had a significant decline in his accuracy of production of three syllable real-words in the follow-up phase, relative to the treatment phase.

M2 maintained his treatment gains to four months post-treatment for treated items and untreated real words with 2, 3 and 4 syllables, with percent accuracy at all four follow-up points above baseline levels (Fig. 3, panels A and B). Planned contrasts revealed no significant difference between accuracy scores in the treatment and follow-up phases for treated items and untreated real-words with two syllables and three syllables. Although M2 had percent accuracy with four syllable real words above baseline levels in the maintenance phase, it was significantly lower in the maintenance phase than the treatment phase.

F2 maintained her treatment gains to four months post-treatment for treated items and untreated real words with 3 syllables, with percent accuracy at all 4 follow-up points above baseline levels (Fig. 4, panels A and B). Planned contrasts revealed no significant difference between accuracy scores in the treatment and follow-up phases for treated items and untreated real-words with 3 syllables. She did not maintain her treatment gain with untreated real words with two syllables, with no follow-up points above baseline levels and significantly poorer performance in the follow-up phase than the treatment phase.

In order to monitor the participants' progress at different time points *within* the maintenance phase, the data for the four participants were grouped and Helmert Planned Orthogonal Contrasts performed. The results indicated there was no significant difference in performance at any maintenance points for treated and untreated pseudo words or real words at any level of complexity.

3.4. Control behavior

All four participants had standard scores for the 'Sounds in Words' subtest of the Goldman–Fristoe Test of Articulation–2 (Goldman & Fristoe, 2000) within the 95% confidence interval of their pre-test Standard Score, indicating no significant difference in their articulation of consonants in simple words. The pre-test and post-test scores with the 95% confidence interval for pre-test score respectively for each participant were F1: 92, 89 (85–99), M1: 97, 95 (90–104), M2: 40, 40 (29–51), F2: 55, 58 (49–61).

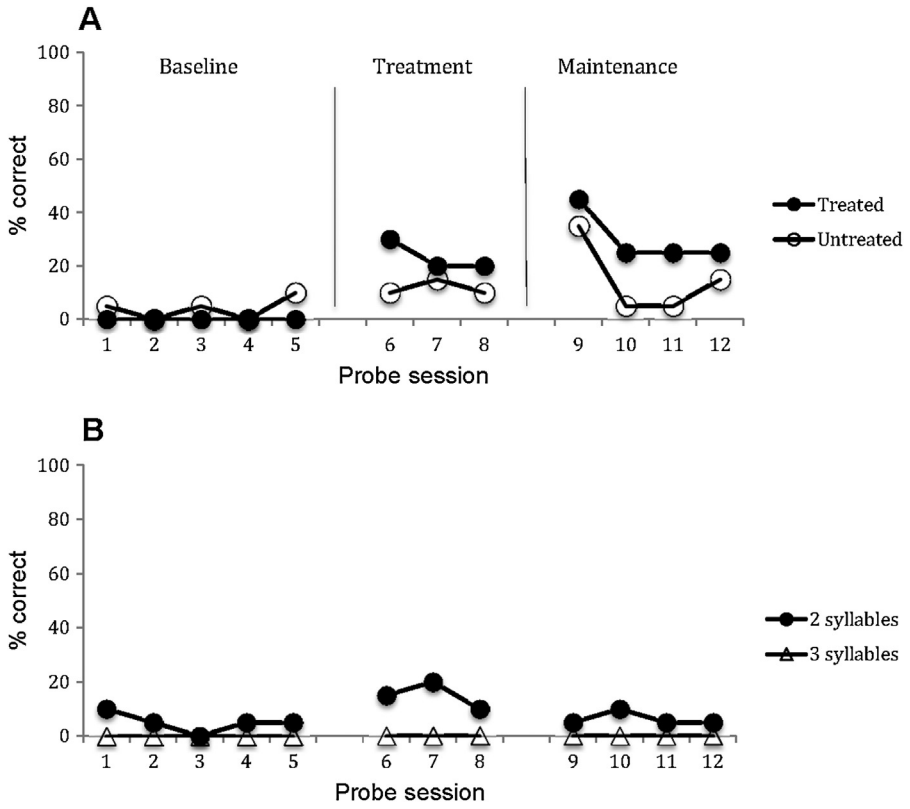

Fig. 4. F1 results. Panel A—treated and untreated pseudo words. Panel B—untreated real words.

4. Discussion

This study aimed to determine the efficacy of ReST treatment provided twice-weekly for children with CAS. We hypothesized that twice-weekly treatment would result in (a) significant skill acquisition, (b) significant generalization to related but untreated behaviors and (c) maintenance of treatment and generalization effects. Hypotheses were supported with all four children showing positive gains that were maintained to 4 months post-treatment. Furthermore, experimental control was demonstrated with no change in general articulatory abilities from baseline to 1 month post-treatment, as examined with the GFTA-2.

Low dose–frequency ReST treatment (twice per week) had similar effects to high dose–frequency treatment (Ballard et al., 2010; Murray et al., 2012c). Both dose frequencies produced significant acquisition of speech skills, with large effect sizes. Here, the two children with relatively better speech initially also showed significant generalization to untreated exemplars of the treated pseudo word forms. The twice-weekly dose produced significant transfer to untreated real words, similar to Murray et al. (2012c). Although the children in this study maintained their gains to four months post-treatment, Murray et al. (2012c) reported significant ongoing improvement from 1 to 4 months post-treatment. It is unclear whether this difference in the maintenance profiles across the two studies is due to the different dose–frequencies or some other factor. Experts have long advocated the use of high frequency treatment for children with CAS (American Speech-Language-Hearing Association, 2007; Hall et al., 1993; Skinder-Meredith, 2001). However, the current study and Murray's earlier one differed in participant characteristics with two children in the current study having concomitant language impairment, which may have interfered with their performance trajectory over the longer term.

The extant studies of dose–frequency in speech treatments suggest a minimum of two sessions per week to stimulate acquisition, generalization, and maintenance of treated skills. For example, Allen (2013) reported that therapy delivered three times weekly was more effective than therapy once a week. However, studies comparing frequencies of two or more sessions a week have tended to report no differences in gains (Spielman et al., 2007; Wambaugh et al., 2013). The current study did not directly compare the effects of different dose frequencies but these findings in combination with those of Murray et al. (2012c) suggest support for a relatively higher frequency of treatment. Further investigation is necessary to determine whether the strength of the ReST treatment effect increases in a linear fashion with increasing session frequency or if the twice-weekly threshold applies with limited additional gains for higher frequencies.

Two of the four children (M2 and F2) demonstrated weaker patterns of generalization with improvements in untreated real words but not in the untreated pseudo words. It may be that real words provide more opportunities for incidental practice, which facilitated generalization. Also, while M2 and F2 showed clear improvement in treated pseudo words during the treatment phase, their performance did not reach the criterion of 80% accuracy within the twelve sessions. Consistent with their slower response to the treatment, M2 and F2 had more severe speech difficulties than M1 and F1 (Ballard et al., 2010; Maas & Farinella, 2012), as well as concomitant language difficulties and, in the case of F2, dysarthria.

It could be argued that the functional goal in a treatment targeting pseudo words is generalization to real word stimuli. However, the lack of generalization to pseudo words in these children is worthy of examination. The primary goal of ReST is to improve the ability to rapidly and smoothly transition between varied sounds and syllables, not simply to learn the motor plan/program for a small set of words. A range of phonemes, syllables and syllable sequences are used in random order to encourage practice in retrieval of syllabic motor plans, fluent (i.e. non-segmented) concatenation of syllables, and manipulation of prosodic contour over the sequence. As such, the lack of generalization to untreated but similar pseudo words suggests that these skills were not sufficiently mastered by these children. If one views learning of pseudo words as simulating the processes children must use when encountering novel words, then this lack of generalization to pseudo words warrants further discussion.

Perhaps children with more severe speech impairment benefit from greater cumulative intervention intensity (i.e. more therapy). M2 and F2 were making progress in treatment and we could hypothesize that had treatment continued, it is likely that they would have continued to improve with possible transfer to untreated pseudo words. Provision of ongoing therapy was not possible in this context, as we wanted to maintain cumulative intensity to facilitate comparison with other ReST studies. However in a clinical setting it may be appropriate to continue treatment until children received a higher percentage correct on probes of performance. In both this study and Ballard et al. (2010) children with approximately 80% correct in treatment had superior generalization or maintenance to those whose accuracy did not reach this level.

More frequent sessions may be beneficial for children with severe difficulties. High dose–frequency ReST had better maintenance than low dose–frequency, and it may be that a high dose–frequency it is also more beneficial for children with severe speech difficulties. High dose–frequency and high cumulative intervention intensity is used with positive effects for children with severe difficulties in the Dynamic Temporal and Tactile Cueing (DTTC) approach (Baas, Strand, Elmer, & Barbaresi, 2008; Strand, Stoekel, & Baas, 2006).

All children maintained their gains with treated items to four months post-treatment and this was also the case for the majority of behaviors tested for generalization. This was a particularly strong finding, given the relatively low level of accuracy with treated items achieved by M2 and F2 and replicates findings from an earlier unpublished ReST study (Staples, McCabe, Ballard, & Robin, 2008). Motor speech interventions employing principles of motor learning typically achieve positive maintenance effects (see Maas et al., 2008; Bislick, Weir, Spencer, Kendall, & Yorkston, 2012 for reviews). However, even in studies employing these principles, lower levels of accuracy or stability in treatment is often associated with weaker maintenance or loss of skill post-treatment (Maas, Butalla, & Farinella, 2012; Wambaugh et al., 2013).

4.1. Limitations and future directions

It would be beneficial to do a direct comparison of a range of dose frequencies in a larger study. In this study, the small sample made it difficult to interpret variations in response that may relate to patient variables and may be washed out in group studies. Larger samples will permit analyses of main effects as well as exploration of covariates such as severity of motor speech disorder and/or concomitant language impairment. Future research could investigate the implications of modifying the use of different motor learning principles on acquisition, generalization and maintenance for children with severe difficulties.

4.2. Summary

In four children with a range of CAS severity, twice-weekly ReST therapy resulted in significant improvement in segmental accuracy, segmentation of syllables, and lexical stress production for treated pseudo words, as well as generalization of treatment effects to untreated items and maintenance of these effects to four months post-treatment. Compared to four-times weekly ReST, the results are similar for acquisition and generalization, but less positive for maintenance; the participants in this study had stable, rather than improving performance in maintenance. If high dose–frequency treatment is not available in a given setting, the findings reported here provide preliminary support for clinicians proceeding with twice-weekly ReST intervention.

Declaration of interest

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The authors have no financial or nonfinancial relationships to disclose.

Appendix A. Continuing education questions

CEU Questions

1. According to Warren et al. (2007) dose–frequency is a measure of the number of
   a. trials produced per session multiplied by the number of sessions per week
   b. sessions per day or per week
   c. trials produced per session
   d. sessions per day or week multiplied by the number of trials
2. In the practice phase, ReST uses which of the following motor learning principles
   a. low frequency feedback
   b. random presentation of items
   c. delayed feedback
   d. knowledge of performance feedback
   e. a, b & c
3. The results of this study indicate that 12 sessions of ReST treatment is
   a. much more effective four times a week
   b. much more effective twice a week
   c. similar when provided twice weekly and four-times weekly, with stronger maintenance following four times weekly treatment
   d. similar when provided twice weekly and four-times weekly
4. Children with severe speech difficulties had _____than children with mild speech difficulties
   a. lower accuracy with treated items
   b. poorer generalization to untreated pseudo words
   c. poorer maintenance
   d. a and b
   e. b and c
5. Future research is needed to determine
   a. whether treatment effects change systematically with dose - frequency
   b. which pseudo words should be treated in ReST
   c. whether the patterns observed in this study are evident in larger group studies
   d. b and c
   e. a and c

References

Allen, M. M. (2013). Intervention efficacy and intensity for children with speech sound disorder. *Journal of Speech, Language, and Hearing Research, 56*, 865–877. http://dx.doi.org/10.1044/1092-4388(2012/11-0076)

American Speech-Language-Hearing Association (2007). *Childhood apraxia of speech [technical report]*. American Speech-Language-Hearing Association Retrieved 18th of November, 2013. From http://www.asha.org/policy/PS2007-00277.htm

Aram, D. M., & Nation, J. E. (1982). *Child language disorders*. St. Louis. MI Mosby.

Baas, B. S., Strand, E. A., & Elmer Barbaresi, W. J. (2008). Treatment of severe childhood apraxia of speech in a 12-year-old male with CHARGE association. *Journal of Medical Speech-Language Pathology, 16*(4).

Baker, E. (2012). Optimal intervention intensity. *International Journal of Speech-Language Pathology, 14*(5), 401–409.

Baker, E., & McLeod, S. (2011). Evidence-based practice for children with speech sound disorders: Part 1 Narrative review. *Language, Speech, and Hearing Services in Schools, 42*(2), 102.

Ballard, K. J. (2001). Response generalization in apraxia of speech treatments: Taking another look. *Journal of Communication Disorders, 34*(1–2), 3–20.

Ballard, K. J., Maas, E., & Robin, D. A. (2007). Treating control of voicing in apraxia of speech with variable practice. *Aphasiology, 21*(12), 1195–1217.

Ballard, K. J., Robin, D. A., McCabe, P., & McDonald, J. (2010). A treatment for dysprosody in childhood apraxia of speech. *Journal of Speech Language & Hearing Research, 53*(5), 1227–1245.

Barratt, J., Littlejohns, P., & Thompson, J. (1992). Trial of intensive compared with weekly speech therapy in preschool children. *Archives of Disease in Childhood, 67*(1), 106–108.

Beeson, P. M., & Robey, R. R. (2006). Evaluating single-subject treatment research: Lessons learned from the aphasia literature. *Neuropsychology Review, 16*(4), 161–169. http://dx.doi.org/10.1007/s11065-006-9013-7

Bercow, J. (2008). *The Bercow report: A review of services for children and young people (0–19) with speech, language and communication needs*. Retrieved 18th of November, 2013. From http://dera.ioe.ac.uk/8405/1/7771-dcsf-bercow.pdf

Bislick, L. P., Weir, P. C., Spencer, K., Kendall, D., & Yorkston, K. M. (2012). Do principles of motor learning enhance retention and transfer of speech skills? A systematic review. *Aphasiology, 26*(5), 709–728. http://dx.doi.org/10.1080/02687038.2012.676888

Dodd, B., Zhu, H., Crosbie, S., Holm, A., & Ozanne, A. (2006). *Diagnostic evaluation of articulation and phonology (DEAP)*. Psychology Corporation.

Dunn, L. M., & Dunn, D. M. (2007). *Peabody picture vocabulary test, (PPVT-4)*. Minneapolis, MN: Pearson Assessments.

Goldman, R., & Fristoe, M. (2000). *Goldman–Fristoe test of articulation 2 (GFTA-2)*. Minneapolis, MN: Pearson Assessments.

Gozzard, H., Baker, E., & McCabe, P. (2006). Children's productions of polysyllables. *ACQuiring Knowledge in Speech, Language and Hearing, 8*, 113–116.

Hall, P. K., Hardy, J. C., & LaVelle, W. E. (1990). A Child with signs of developmental apraxia of speech with whom a palatal lift prosthesis was used to manage palatal dysfunction. *Journal of Speech and Hearing Disorders, 55*(3), 454.

Hall, P. K., Jordan, L. S., & Robin, D. A. (1993). *Developmental apraxia of speech: Theory and clinical practice*. Austin, TX: Pro-ed.

Kazdin, A. E. (2011). *Single-case research designs: Methods for clinical and applied settings* (2nd ed.). New York, NY: Oxford University Press.

Law, J., Zeng, B., Lindsay, G., & Beecham, J. (2012). Cost-effectiveness of interventions for children with speech, language and communication needs (SLCN): A review using the Drummond and Jefferson (1996) 'Referee's Checklist'. *International Journal of Language & Communication Disorders, 47*(1), 1–10. http://dx.doi.org/10.1111/j. 1460-6984.2011.00084.x

Lewis, B. A., Freebairn, L. A., Hansen, A. J., Iyengar, S. K., & Taylor, H. G. (2004). School-age follow-up of children with childhood apraxia of speech. *Language, Speech & Hearing Services in the Schools, 35*(2), 122–140.

Maas, E., Butalla, C., & Farinella, K. (2012). Feedback frequency in treatment for childhood apraxia of speech. *American Journal of Speech-Language Pathology, 21*(3), 239–257.

Maas, E., & Farinella, K. A. (2012). Random versus blocked practice in treatment for childhood apraxia of speech. *Journal of Speech, Language, and Hearing Research, 55*(2), 561–578.

Maas, E., Robin, D. A., Austermann Hula, S. N., Freedman, S. E., Wulf, G., Ballard, K. J., & Schmidt, R. A. (2008). Principles of motor learning in treatment of motor speech disorders. *American Journal of Speech-Language Pathology, 17*(3), 277–298.

McCabe, P., Macdonald-Da'Silva, A., Van Rees, L., Ballard, K., & Arciuli, J. (2010). Using orthographic cues to improve speech production in children with and without childhood apraxia of speech. *Paper presented to the motor speech conference*.

Morgan, A. T., & Vogel, A. P. (2008). Intervention for childhood apraxia of speech. *Cochrane Database of Systematic Reviews, 3*, CD006278. http://dx.doi.org/10.1002/14651858.CD006278.pub2

Mullen, R., & Schooling, T. (2010). The national outcomes measurement system for pediatric speech-language pathology. *Language, Speech, and Hearing Services in Schools, 41*(1), 44.

Murray, E., McCabe, P., & Ballard, K. J. (2012a). Childhood apraxia of speech (CAS): What do kids with and without CAS look like? *Paper presented at the speech pathology Australia conference*.

Murray, E., McCabe, P., & Ballard, K. J. (2012b). A comparison of two treatments for childhood apraxia of speech: Methods and treatment protocol for a parallel group randomised control trial. *BMC Pediatrics, 12*(1), 112.

Murray, E., McCabe, P., & Ballard, K. J. (2012c). The first randomised controlled trial for childhood apraxia of speech. *Paper presented at the speech pathology Australia national conference*.

Murray, E., McCabe, P., & Ballard, K. J. (2014). A systematic review of treatment outcomes for children with childhood apraxia of speech. *American Journal of Speech Language Pathology*.

Namasivayam, A. (2013). *The relationship between treatment intensity & treatment outcomes for children with apraxia of speech*. Retrieved 18th of November, 2013. From http://apraxia-kids.blogspot.com.au/2013/01/the-relationship-between-treatment.html

Robbins, J., & Klee, T. (1987). Clinical assessment of oropharyngeal motor development in young children. *Journal of Speech and Hearing Disorders, 52*(3), 271.

Ruggero, L., McCabe, P., Ballard, K. J., & Munro, N. (2012). Paediatric speech–language pathology service delivery: An exploratory survey of Australian parents. *International Journal of Speech-Language Pathology, 14*(4), 338–350.

Schmidt, R. A., & Lee, T. (2011). *Motor control and learning, a behavioral emphasis* (5th ed.). Champaign, IL: Human Kinetics.

Semel, E., Wiig, E., & Secord, W. (2006). *Clinical evaluation of language fundamentals fourth edition, Australian standardised edition (CELF-4 Australian)*. Sydney, Australia: Pearson Inc.

Skinder-Meredith, A. (2001). Differential diagnosis: Developmental apraxia of speech and phonologic delay. *Augmentative Communication News, 14*, 5–8.

Spielman, J., Ramig, L. O., Mahler, L., Halpern, A., & Gavin, W. J. (2007). Effects of an extended version of the Lee Silverman voice treatment on voice and speech in Parkinson's disease. *American Journal of Speech-Language Pathology, 16*(2), 95.

Staples, T., McCabe, P., Ballard, K. J., & Robin, D. A. (2008). Childhood apraxia of speech: Treatment outcomes at 6 months of an intervention incorporating principles of motor learning. *Paper presented at the joint New Zealand Speech–Language Therapy Association/Speech Pathology Australia conference*.

Ukrainetz, T. A. (2009). Foreword. *Topics in Language Disorders, 29*(4), 291–293. 210.1097/TLD.1090b1013e3181c97821.

Wambaugh, J. L., Nessler, C., Cameron, R., & Mauszycki, S. C. (2013). Treatment for acquired apraxia of speech: Examination of treatment intensity and practice schedule. *American Journal of Speech-Language Pathology, 22*(1), 84–102. http://dx.doi.org/10.1044/1058-0360(2012/12-0025)

Wiig, E. H., Secord, W., & Semel, E. M. (2004). *CELF preschool 2: Clinical evaluation of language fundamentals preschool—second edition, Australian edition (CELF P-2 Australian)*. Sydney, Australia: Harcourt Assessment.

Warren, S. F., Fey, M. E., & Yoder, P. J. (2007). Differential treatment intensity research: A missing link to creating optimally effective communication interventions. *Mental Retardation & Developmental Disabilities Research Reviews, 13*(1), 70–77.

Watts, C. R. (2009). Lack of randomized controlled trials prohibits analysis of effectiveness for the treatment of childhood apraxia of speech: A systematic review of quasi-experimental group designs and single-subject experimental designs is needed. *Evidence-Based Communication Assessment and Intervention, 3*(1), 8–10.

Williams, A. L. (2000). Multiple oppositions: Case studies of variables in phonological intervention. *American Journal of Speech-Language Pathology, 9*(4), 289.

Wohlert, A. (2004). Service delivery variables and outcomes of treatment for hypokinetic dysarthria in Parkinson disease. *Journal of Medical Speech-Language Pathology, 12*(4), 235–239.

Chapter 4: Mode Modification—Telehealth Delivery of Rapid Syllable Transition Treatment

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I declare that I made the following contribution to the study:

- Conception of the research question
- Design of the study with the other authors
- Collection of data
- Data entry and analysis
- Writing of the first draft of the manuscript
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Research Report

Telehealth delivery of Rapid Syllable Transitions (ReST) treatment for childhood apraxia of speech

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Abstract

Background: Rapid Syllable Transitions (ReST) treatment uses pseudo-word targets with varying lexical stress to target simultaneously articulation, prosodic accuracy and coarticulatory transitions in childhood apraxia of speech (CAS). The treatment is efficacious for the acquisition of imitated pseudo-words, and generalization of skill to untreated pseudo-words and real words. Despite the growing popularity of telehealth as a method of service delivery, there is no research into the efficacy of telehealth treatments for CAS. Telehealth service delivery is associated with compromised audio and visual signal transmission that may affect the efficacy of treatment.
Aims: To conduct a phase 1 efficacy study of telehealth delivery of ReST treatment for CAS, and to discuss the efficacy with reference to face-to-face ReST treatment.
Methods & Procedures: Using a multiple baseline across participants design, five children aged 5–11 years with CAS received ReST treatment four times a week for 3 weeks via video conferencing with Adobe Connect. The children's ability to imitate new pseudo-words, generalize the skills to untreated pseudo-words and real word items, and maintain the skills following treatment were assessed. Both visual and statistical analyses were utilized.
Outcomes & Results: All five children significantly improved with their production of the imitated treated pseudo-word items and significantly generalized to similar untreated pseudo-words and real words. Additionally, two of the children showed significant generalization to imitated phrases with the treatment items. Four of the children maintained their treatment gains up to 4 months post-treatment. Telehealth delivery produced similar acquisition of pseudo-words and generalization to untreated behaviours as face-to-face delivery; however, in the 4 months following treatment, the children showed stable rather than improving speech skills. The intra- and inter-judge reliability was similar in telehealth delivery for face-to-face delivery. Caregivers and clinicians were satisfied with the telehealth treatment.
Conclusions & Implications: This phase 1 study provides promising indications of the efficacy of ReST treatment when delivered four times per week via telehealth, and warrants further large-scale investigation.

Keywords: therapy, intervention, prosody, dyspraxia, video conferencing, Adobe Connect, telespeech, telepratice.

What this paper adds?

What is already known on the subject?

Telehealth is being increasingly used for assessment and treatment of communication disorders. This service delivery method has demonstrated effectiveness for the treatment of many communication disorders, including articulation and phonology impairments, but there is no information about the efficacy of telehealth treatments for CAS. ReST treatment is efficacious for CAS when delivered face to face, producing the acquisition of new pseudo-words, generalization to untreated skills and retention of skills following treatment. This phase 1 study investigates the efficacy of ReST treatment for CAS when delivered by telehealth.

What this paper adds?

Preliminary support for the use of ReST treatment, when delivered four times per week via video conferencing. The results justify larger scale studies of this service delivery method.

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Introduction

Children with childhood apraxia of speech (CAS) have difficulty planning and programming the movements required for the production of accurate speech sounds and prosody. Their speech is often characterized by inconsistent errors, inappropriate prosody and disrupted coarticulatory transitions (American Speech–Language–Hearing Association 2007). The difficulties associated with their impairment are often persistent (Lewis *et al.* 2004) with potential effects in a range of linguistic and speech-motor domains (American Speech–Language–Hearing Association 2007). It has been argued that children with CAS require more intensive treatments than other speech-sound disorders (Maas *et al.* 2014, Murray *et al.* 2014, Namasivayam *et al.* 2015), and for a longer period (Skinder-Meredith 2001).

Although several different treatments are used for CAS, most have been investigated in case study or case-series designs and have low levels of evidence regarding their effectiveness (Murray *et al.* 2014, Maas *et al.* 2014). Rapid Syllable Transitions (ReST) is a relatively new treatment for CAS that uses pseudo-word targets with varying lexical stress patterns to target simultaneously articulatory accuracy, fluent transitions between syllables and lexical stress. ReST incorporates motor learning principles to facilitate retention and generalization of treated skills. ReST treatment has demonstrated an improvement in treated items (Ballard *et al.* 2010, Thomas *et al.* 2014), generalization of treatment effects to untreated pseudo-words (Ballard *et al.* 2010, Thomas *et al.* 2014), and to connected speech (Staples *et al.* 2008). A randomized controlled trial comparing ReST treatment with the Nuffield Dyspraxia Programme—Third Edition, demonstrated the efficacy of both treatments (Murray *et al.* 2015). Specifically, ReST treatment resulted in significant acquisition of treated pseudo-words, significant generalization of treatment effects to untreated pseudo-words and real words, and maintenance of treatment effects for 4 months post-treatment (Murray *et al.* 2015). Although typically delivered across four 1-h sessions per week for 3 weeks, ReST is also efficacious when provided across two 1-h sessions per week for 6 weeks (Thomas *et al.* 2014).

Even though effective treatments exist for CAS, many families are unable to access speech pathologists to provide the required treatment, and when treatment is received it is often less frequent and for a shorter duration than necessary (Ruggero *et al.* 2012). These access difficulties are compounded for people who need to see a specialist clinician or who live in rural and remote areas (O'Callaghan *et al.* 2005). Telehealth, with its provision of therapy services at a distance, can improve access to both high-intensity speech-pathology treatments (Mashima and Doarn 2008) and specialist clinicians. When provided in the client's home, telehealth eliminates the travel time associated with face-to-face therapy (Reynolds *et al.* 2009), and improves generalization (Theodoros 2013). Telehealth is well accepted by families (Constantinescu 2012) and in some cases is preferable for clients over face-to-face delivery (Ciccia *et al.* 2011). Although the term 'telehealth' covers all types of services mediated by technology, the focus of this article is video conferencing, which provides real-time transmission of both audio and visual information.

There is growing evidence supporting the use of video conferencing for speech pathology (for reviews, see Theodoros 2011 and Mashima and Doarn 2008). The effectiveness of video conferencing has been more widely investigated for assessments than for therapy. Video conferencing assessments produce equivalent results to face-to-face assessments in several speech and language areas, including paediatric speech-sound disorders (Eriks-Brophy *et al.* 2008, Waite *et al.* 2012). Despite the promising results from speech-pathology assessment of speech-sound disorders using video conferencing, poor inter-rater reliability has been shown between face-to-face and telehealth assessments for the identification of the presence or absence of voicing, accuracy of fricative phoneme perception, identification of phonemes without visible articulation (e.g., /ʧ/ and /l/) (Eriks-Brophy *et al.* 2008, Waite *et al.* 2006), and perception of abnormal nasal resonance in speech (Hill *et al.* 2006).

Video conferencing as a service delivery model is showing promising results for speech-pathology treatments, particularly treatments that are operationally defined. Effective treatment via video conferencing has been demonstrated for the Lidcombe Program for stuttering (O'Brian *et al.* 2014), the Camperdown Program for stuttering (Carey *et al.* 2014), and the Lee Silverman Voice Treatment (LSVT®) for patients with Parkinson's disease (Constantinescu *et al.* 2011).

Articulation impairments have been effectively treated via video conferencing. In a series of studies culminating in a randomized controlled trial, traditional articulation therapy was shown to be as effective via video conferencing as face-to-face delivery (Grogan-Johnson *et al.* 2013). The participants in Grogan-Johnson et al.'s (2013) study had articulation and phonological disorders rather than CAS (S. Grogan-Johnson, personal communication, 6 February 2015) and therefore these findings cannot necessarily be applied to children with CAS. Effective treatments for CAS often focus on prosody or speech movements (Maas *et al.* 2014, Murray *et al.* 2014) rather than targeting specific sound errors in a step-by-step progression.

There is currently no evidence for efficacy of video conferencing for CAS treatments. The compromised sound signal sometimes associated with video conferencing (Keck and Doarn 2014) may potentially

reduce the effectiveness of treatment. Given that speech pathologists have an ethical responsibility to ensure their treatments are effective and efficient (Speech Pathology Australia n.d.), it is important to investigate the efficacy of telehealth for delivering treatment for this population.

In this study we investigated the efficacy of ReST treatment for CAS via video conferencing, with the participants receiving treatment at home, using their own computers and existing Internet connection.

The hypotheses were as follows:

- ReST treatment, delivered four times a week for 3 weeks via video conferencing, will result in:
  - acquisition of targeted speech behaviours, namely accurate production of phonemes, lexical stress pattern and smooth transitions between syllables, in imitated pseudo-words, as perceived by the probe assessor;
  - generalization of this treatment effect to untreated but related imitated speech behaviours:
    - pseudo-words with the same phonemes and lexical stress patterns as treated items;
    - real words with the same number of syllables as the treated items.
  - maintenance of speech gains up to 4 months post-treatment.
- Telehealth treatment will be viewed as comparable or more desirable than intensive face-to-face clinic treatment, as measured via telephone interview with one caregiver per child, 4 weeks post-treatment.

Method

Participants

Eleven monolingual Australian English-speaking children consented to participate in the study. Six children were excluded from the study following assessment, as they did not meet the inclusion criteria defined below. Five children with a diagnosis of CAS aged 5:5 (years; months) to 11:2 completed the study.

Inclusion criteria were (1) consensus diagnosis of CAS (see below), (2) passed pure tone audiometry at 20 dB at 500, 1, 2 and 4 kHz, (3) normal receptive vocabulary (Peabody Picture Vocabulary Test—4th Edition; Dunn and Dunn 2007), and (4) normal oral structure (Oral and Speech Motor Protocol; Robbins and Klee 1987). The diagnosis of CAS was made independently by the first two authors based on the perception of the presence of core perceptual features of CAS (American Speech–Language–Hearing Association 2007) during a battery of speech production tests. There are currently no specific tests or agreed cut-off points for determining the presence of the core perceptual features (American Speech–Language–Hearing Association 2007). We chose relatively low cut-off points for each feature, as we were recruiting children up to 12 years of age and the frequency and/or severity of behaviours associated with the core perceptual features may possibly reduce as children get older. Diagnosis of CAS was given when (1) children < 11 years showed $> 40\%$ inconsistency in word production on repeated attempts during the Inconsistency subtest of the Diagnostic Evaluation of Articulation of Articulation and Phonology (DEAP; Dodd *et al.* 2006) or children aged ≥ 11 years showed $> 30\%$ inconsistency[1] over three separate administrations of 25 words from the Test of Polysyllables (Gozzard *et al.* 2006); (2) a minimum of 10 words exhibited syllable segregation within words during the Test of Polysyllables (Gozzard *et al.* 2006), indicating difficulty transitioning between syllables; and (3) a minimum of 15% stress pattern mismatches were produced on the Test of Polysyllables, and the examiners perceived abnormal prosody during conversational speech.

Two additional tests were used to provide more detail on the severity of the children's overall language and articulation skills relative to age-matched peers, but were not used to determine suitability for the study: (1) the Clinical Evaluation of Language Fundamentals–Preschool Second Edition (CELF-P2; Wiig *et al.* 2004) or the 4th Edition Australian version (CELF-4; Semel *et al.* 2006), depending on age; and (2) the Goldman–Fristoe Test of Articulation—2 (GFTA-2; Goldman and Fristoe 2000).

The children were assigned pseudonyms. Their performance on the above speech and language tests is reported in table 1.

All children had previously received speech therapy, but did not have any other speech treatment from the start of baseline testing until 1 month post-treatment. During the period between 1 and 4 months post-treatment, none of the participants received speech-sound intervention; however, Emily received therapy to improve her receptive and expressive language skills. The research project was approved by The University of Sydney Human Ethics Committee (reference number 2014/080).

Design

A multiple baseline across participants design (Kazdin 2011) was used in this study. Participants were allocated either three, four, five or six twice-weekly baseline sessions. The treatment commenced after different numbers of baseline sessions to demonstrate that change occurred following the commencement of treatment, rather than after a certain number of baseline sessions. During the treatment phase, each participant's performance was monitored three times; immediately prior to treatment sessions five and nine, and 1 day

Table 1. Participants' initial assessment results

Test	Oliver (5 years; 6 months)	Jack (11 years; 0 months)	Emily (11 years; 2 months)	Luke (5 years; 3 months)	Lachlan (7 years; 6 months)
Clinical Evaluation of Language Fundamentals—Preschool Second Edition (CELF-P2) or Clinical Evaluation of Language Fundamentals—Fourth Edition (CELF 4)[a]					
Receptive Language Index					
Standard score	79	106	88	84	75
Percentile rank	8	66	21	14	5
Interpretation	< NL mild	WNL	WNL	< NL mild	< NL moderate
Expressive Language Index					
Standard score	92	112	63	59	70
Percentile rank	30	79	1	0.3	2
Interpretation	WNL	WNL	< NL severe	< NL severe	< NL severe
Peabody Picture Vocabulary Test					
Standard score	90	90	99	88	108
Percentile rank	25	25	47	21	70
Interpretation	WNL	WNL	WNL	WNL	WNL
Goldman–Fristoe Test of Articulation					
Standard score	79	45	75	58	69
Percentile rank	11	<1	<1	2	5
Interpretation	< NL mild	< NL severe	< NL severe	< NL severe	< NL moderate
Test of Auditory Perception—Third Edition, Word Discrimination subtest					
Scaled score	9	9	12	7	10
Percentile rank	37	37	75	16	50
Interpretation	WNL	WNL	WNL	WNL	WNL
Inconsistency Assessment[b]					
% Inconsistency	68	32	44	64	48
Interpretation	Inconsistent	Inconsistent	Inconsistent	Inconsistent	Inconsistent
Test of Polysyllables					
% Consonants correct	76	84	85	36	67
% Vowels correct	67	91	88	50	74
% Phonemes correct	73	87	86	42	70
% Stress pattern errors[c]	46	26	32	77	47
% Syllable segregations[d]	20	20	25	21	22
Oral and motor speech protocol					
Structure					
Raw score	23	23	25	24	23
Interpretation	WNL	^	^	WNL	^
Function					
Raw score	94	97	102	107	104
Interpretation	< NL	^	^	< NL	^
Observations	Difficulty coordinating lip and tongue movements in non-speech and speech tasks	Reduced speaking volume, intermittent hypernasality	Inconsistent hypernasality. Loud speaking volume	Difficulty coordinating lip and tongue movements in non-speech and speech tasks	Difficulty imitating multisyllabic words. Incoordination during DDK tasks

Notes: [a]Children aged 5 years completed the Clinical Evaluation of Language Fundamentals—Preschool Second Edition (CELF-P2), those aged more than 6 years completed the Clinical Evaluation of Language Fundamentals—Fourth Edition (CELF 4); see the participants section for test references; WNL, within normal limits; NL, normal limits; DDK, Diadochokinesis.
[b]Children less than 11 years completed the Inconsistency subset of the Diagnostic Evaluation of Articulation and Phonology (Dodd *et al.* 2006) and those ≥ 11 years completed three productions of 25 words from the Test of Polysyllables (Gozzard *et al.* 2006). Inconsistent; see the participants section for further information regarding inconsistency assessment.
[c]Calculated using Profile of Phonology (PROPH) software.
[d]Percentage of words with at least one perceptually identified absence of smooth joining of the syllables; ^ = outside of age range for normative scores.

post-treatment. Each participant's performance was also monitored three times in the follow-up phase at 1 week, 4 weeks and 4 months post-treatment.

Demonstration of experimental control in multiple-baseline designs is through the replication of the treatment effect across participants, with staggered introduction of the independent variable across different time points (Kazdin 2011). Although internal validity is typically addressed through replication of the effect, research with children faces a threat to internal

validity as a result of maturation. As an additional safeguard against maturation effects we included a control behaviour to our probe stimuli for each child (see 'Probe stimuli' for details).

Probe stimuli

A 90-item probe list was created for each child to permit analysis of (1) treatment effect, (2) generalization to related, but untreated items, (3) generalization to real words with the same number of syllables as the treated items, and (4) maturational control. The probe stimuli included pseudo-word strings with strong–weak (SW) stress patterns (e.g., /dabəfi/) and weak–strong (WS) patterns (e.g., /kədɔfi/). The consonants for the pseudo-word stimuli represented different manner, place and voicing conditions, namely /d/, /k/, /f/ and /b/. The vowels selected for the pseudo-word strings were /a/, /ɔ/, /i/ and /ə/. The probe and treatment stimuli are included in appendix A.

Lachlan, Oliver, Jack and Emily's probe list consisted of 20 SW and 20 WS three-syllable (CVCVCV) pseudo-words, of which 20 (10 SW and 10 WS) were treated and 20 (10 SW and 10 WS) remained untreated, in order to assess generalization to similar but untreated items. The treated items were selected from the set of pseudo-words, and each participant had a different set of treated items. The probe list also included 20 carrier phrases (e.g., I found a _____) with the three-syllable strings to assess generalization effects to sentence level, and 20 three-syllable real words to assess generalization to real words. Additionally, each child had 10 control items, which contained an articulation error or phonological process that we hypothesized would not change during ReST treatment, as it was unrelated to treated items (e.g., a liquid when only plosives and fricatives were trained, or an inter-dental lisp when prosody and nasality were targeted), or it represented a more complex skill level than treated (e.g., clusters). Lachlan's control behaviour was production of word initial /s/ clusters, Oliver and Emily's was articulation of /r/ in initial and medial word position, and Jack's was articulation of /s/ in initial- and final-word position.

Luke's speech difficulties were more severe than the other participants and his treatment stimuli were two syllable pseudo-words. His probe list contained 20 SW and 20 WS two syllable (CVCV) pseudo-words, with 10 SW and 10 WS items randomly selected for treatment, and 10 of each kept to assess generalization to untreated items. His probe list also included 20 three syllable (CVCVCV) pseudo-words, to assess performance on more complex pseudo-words, 10 two syllable real words, and 10 three syllable real words, to assess generalization to real words. Luke's control behaviour was the production of initial /l/ clusters (/pl/, /bl/, /kl/, /fl/ and /gl/).

Figure 1. Microphone and headphone set-up

Equipment

Video conferencing was conducted using Adobe Connect, version 8, which had the function to share documents and interactive workspaces as well as transmit real-time audio and visual information. The speech-pathology clinicians used either a Dell Latitude E6320 laptop computer with an inbuilt web camera or a custom-built Bosch P8C WS desktop computer with Logitech C930e web camera. Clinicians wore a USB headset (Sennheiser PC 8 or Logitech H540). All participants used their home computer, with broadband Internet connection. Participants wore a Sennheiser PC 8 USB headset around the neck with the microphone positioned approximately 10 cm from the mouth to record sound and Yellowstone YSYROHRD headphones over the ears (figure 1). A separate headphone and microphone for participants was used to enable a 3.5-mm audio splitter to connect to the caregiver's Yellowstone YSYROHRD headphones allowing them to hear the child's session. All sessions were recorded through Adobe Connect for later assessment of treatment fidelity, scoring reliability, and for student training purposes. The sessions were also recorded at the participant's home using an Olympus VN-711PC digital voice recorder; however, all data reported here are based on the Adobe Connect recordings.

The face-to-face initial assessments were audio recorded with an AKG C520 headset microphone and Roland Quad Capture UA-55. They were video recorded using a Bosch NBN-832V-P camera, and an Electrovoice RE90HW microphone connected to a Bosch DIVAR IP 7000 2U DVD.

Procedure

The first author, a qualified speech pathologist, carried out the face-to-face eligibility assessments and video conferencing baseline probes. Jack and Oliver were

treated by qualified speech pathologists experienced in ReST treatment; while Lachlan, Luke and Emily were treated by trained speech pathology students, under the supervision of the first and second authors. The same clinician treated Emily and Lachlan. One clinician treated each child for the duration of the treatment phase.[2]

Baseline and probe sessions

Identical procedures were used for baseline and the probe sessions. The probe list items were presented in one of three randomized orders. The participants viewed a PowerPoint slide show, with the orthography for each pseudo-word item and a picture plus orthography for each real-word item and the sound file of an Australian English female speaker producing each item. As the participant viewed each slide, the parent played the sound file for the item and the child imitated the word. Imitation was used due to the non-familiarity of the pseudo-word items and to ensure consistency of procedure between pseudo- and real-word items. During the PowerPoint slide show, the clinician could see and hear the participant via the web camera and microphone, and the participant could hear the clinician, but see only the PowerPoint slide show.

Technology set-up

Prior to the baseline sessions, each participant had one or two 30-min web-conferencing familiarization sessions where the treating clinician and child talked via video conference, played interactive web-based games, and solved any technical difficulties with equipment or connectivity.

Technology rating

Following each session, the treating clinician completed a form noting any technical issues, whether the issues were resolved and the strategies employed. The clinician also marked a line on a 10-cm visual analogue scale to rate the technology in the session, from 'very poor' to 'excellent'.

Parent satisfaction

Four weeks post-treatment, telephone interviews were conducted with the treating clinicians and the parents. During the semi-structured interview, the parents and clinicians used a 10-point rating scale (e.g., 0 = not convenient at all, 10 = very convenient) to rate the convenience of the sessions, their perception of the child's motivation and their overall satisfaction with the telehealth mode of treatment.

Treatment

The ReST treatment was used, following the procedure described in Murray *et al.* (2012). However, unlike Murray *et al.* (2015), all children in this study imitated the stimulus items, while looking at the written stimulus rather than reading the items. Each session began with approximately 10 min of pre-practice to explain the task and ensure the children had a reference of correctness for the target stimuli. During pre-practice, the participants (1) viewed a card with the written pseudo-word via the webcam, (2) listened and watched the computer monitor while the clinician produced the selected written pseudo-word, from the 20 treatment items, and (3) attempted to imitate the word production.

The participants were provided with knowledge of performance (KP) feedback immediately following each production (e.g., 'That word was broken, the parts were separated. Try to join the parts together smoothly'). A variety of cueing techniques were employed such as breaking the words into syllables and rejoining, representing relative syllable duration with magnetic strips on a whiteboard, slowing overall rate of production, and cueing about correct articulator placement. Once five items were produced correctly with modelling and shaping, the participant moved into the practice phase. The pre-practice phase lasted for up to 25 min in sessions 1, 2 and in any session where a child progressed to a new level of treatment, and approximately 10 min in all other sessions.

In the practice phase, each participant aimed to complete 100 trials ($\overline{x} = 99$, SD = 9.33): five trials each of the 20 treated items, in random order. The clinician provided a live model of the item for the child to imitate during the practice trials. Knowledge of results (KR) feedback (i.e., feedback about whether the item was correct or incorrect) was provided on approximately 50% of the items after a delay of 3–5 s. After every 20 trial items, a 2-min rest break was provided.

Once a participant achieved ≥ 80% correct in two consecutive practice sessions, the client began treatment on the next, more complex treatment level (see Murray *et al.* 2012 for levels in ReST treatment). The progression criterion was met by Jack in session 5, and Emily in session 10, and these children moved to treatment on pseudo-words at the end of carrier phrases (e.g., 'She has a big /dəfabi/' or 'There's a /dəbɔfi/') from sessions 6 and 11 respectively.

Dependent measures and data analysis

The probe assessors made perceptual judgments about each probe item with regard to the accuracy of the phonemes, stress pattern and fluency of syllable transitions. Judgements were made about each construct

individually and, in order to be counted as correct, the probe item needed (1) correct sounds, (2) correct lexical stress and (3) smooth connection of the syllables. The dependent measure was the percentage of items correct (i.e., with correct sounds, lexical stress and smooth connection between the syllables). The first author conducted all baseline assessments. A rater blinded to the phase of treatment and baseline level of speech skill conducted the probe assessments. Intra- and inter-rater reliability was calculated on 20% of each baseline session, probe assessment and treatment session.

Data for each participant were graphed for visual analysis. Visual analysis consisted of examining the level, trend, variability, overlap and immediacy of effect. Visual analyses were supported with statistical analyses where possible. In order to do so, we tested each child's data for independence by preliminary analyses of variance comparing phases, recording residuals from these analyses and testing the residuals for autocorrelation. With the exception of Lachlan's untreated pseudo-words and carrier phrases, and Emily's real words, in all cases the lag 1 correlation of the residuals was non-significant, indicating no evidence that the assumption of independence was violated in most cases. Where other analysis of variance (ANOVA) assumptions were met, ANOVAs and Helmert planned orthogonal contrasts were performed for each participant to test for differences across phases (baseline, treatment, follow-up) within behaviours (treated pseudo-words, untreated pseudo-words, untreated real words, more complex pseudo-words or pseudo-words in carrier phrases and control words). In each case, the first Helmert contrast compared the average within-participant performance in the baseline phase with average performance over treatment and follow-up phases, and the second contrast compared the average within-participant performance in the treatment phase with the follow-up phase. A study-wide adjustment to the significance level, to account for multiple comparisons, was not performed. This is because the primary method of analysis was visual analysis, as is common in single-case design, with the statistical analyses used to confirm the results of visual analysis. Significance at both 0.05 and 0.01 levels are indicated in table 3, and readers are advised to use caution when interpreting significance values between 0.05 and 0.01. Where data were autocorrelated, only visual analysis was performed.

In order to test for maintenance of treatment effect within the follow-up phase, post-hoc planned orthogonal contrasts were performed at the data points within the follow-up phase, with the participants' data pooled. Contrasts were conducted of average performance across participants at (1) 1 day post-treatment with later points (i.e., 1 week, 1 month and 4 months post-treatment combined), (2) 1 week post-treatment with later points (i.e., 1 and 4 months post-treatment combined), and (3) 1 month post-treatment with 4 months post-treatment. Effect sizes were calculated using the protocol described by Beeson and Robey (2006): d_2 = (mean score in follow-up phase – mean score in baseline phase)/pooled standard deviation.

Table 2. Reliability information

	Probe items[a]			
	Pseudo-words	Real words	Control sounds	Treatment items[a]
Judgements of correctness				
Intra-rater	92	91.9	93.5	91
Inter-rater	89	87.3	81.5	88
Broad phonemic transcription				
Inter-rater	89.4	82.5	92.8	95
Inter-rater	84.9	78.5	80.5	94

Note: [a]Percentage agreement.

Reliability

Inter- and intra-rater reliability was calculated for phonemic transcription and the scoring of articulation accuracy, stress pattern and fluency of syllable transitions. Given the indications in the literature that perception of some sounds via video conferencing can be unsatisfactory (see the Introduction for details), reliability was calculated separately for pseudo-words items, real-word items and control items (table 2).

Treatment fidelity

The first author examined a randomly selected 10 min of each session for treatment fidelity. Assessment was made of the accuracy of the clinician's model, the number of trials given feedback, the accuracy of the feedback, the type of feedback (i.e., KP in pre-practice and KR in practice), and the timing of feedback. Average fidelity for treatment sessions was 95% (SD = 6.1, range = 75–100). Fidelity was lowest in the first two sessions, involving clinicians giving feedback without sufficient delay, and giving KP rather than KR feedback in the practice phase.

Results

Effects of treatment

Oliver's per cent accuracy with the to-be-treated items during baseline was 0–10% (figure 2, panel A). His per cent accuracy steadily improved during the treatment phase to 70%, and the difference between the baseline phase and the later phases was significant. The results of all significance testing can be found in table 3.

Table 3. Planned contrasts and effect sizes

		Effect size	BL versus later (i.e., T and FU combined)		T versus FU		
	Word set	d_2 =	t=	p=	t=	p=	Change
Oliver	Treated pseudo-words	9.64	6.595	0.001**	0.835	0.437	–
	Treated pseudo-words in phrases	2.04	2.418	0.065	5.156	0.68	–
	Untreated pseudo-words	1.79	3.397	0.015*	1.343	0.229	–
	Untreated real words	5.73	6.711	0.001**	0.731	0.497	–
	Control /r/ articulation	1.63	1.277	0.251	0.583	0.583	–
Jack	Treated pseudo-words	3.59	8.333	< 0.001**	0.177	0.111	–
	Treated pseudo-words in phrases	2.30	3.878	0.004**	0.282	0.784	–
	Untreated pseudo-words	3.59	8.333	< 0.001**	0.176	0.111	–
	Untreated real words	6.34	9.929	< 0.001**	1.136	0.286	–
	Control /s/ articulation	0.00	1.065	0.316	1.523	0.168	–
Emily	Treated pseudo-words	4.65	10.121	< 0.001**	2.705	0.003**	↓
	Treated pseudo-words in phrases	2.00	3.16	0.013*	0.321	0.757	–
	Untreated pseudo-words	3.48	5.172	0.001**	4.84	0.525	–
	Untreated real words						
	Control /r/ articulation	0.70	0.75	0.473	0.357	0.731	–
Luke	Treated pseudo-words	21.24	16.588	< 0.001**	1.452	0.193	–
	Untreated three-syllable pseudo-words	3.20	2.273	0.060	1.179	0.634	–
	Untreated pseudo-words	13.16	17.358	< 0.001**	0.515	0.275	–
	Untreated real words	3.12	4.37	0.003**	1.029	0.341	–
	Control /l/ clusters	0.64	5.952	0.001**	11.111	< 0.001**	↓
Lachlan	Treated pseudo-words	6.79	3.791	0.009**	1.329	0.232	–
	Treated pseudo-words in phrases						
	Untreated pseudo-words						
	Untreated real words	3.87	4.6	0.004**	2.741	0.034*	↑
	Control /s/ clusters	0.07	0.532	0.612	0.931	0.390	–

Note: Effect size = Cohen's d_2 using pooled standard deviations (Beeson and Robey 2006); BL, baseline phase; T, treatment phase; FU, follow-up phase; **significant at 0.01; *significant at 0.05; –, No difference between treatment and maintenance phase; ↓, decrease in the follow-up phase; ↑, increase in the follow-up phase. Contrasts for Emily's untreated real words and Lachlan's pseudo-words in phrases and untreated pseudo-words were not calculated because the data showed autocorrelation.

Jack's per cent accuracy during the baseline phase with the to-be-treated items ranged from 30% to 35% (figure 3, panel A). During the treatment phase, his per cent accuracy increased to 85–95%, resulting in a significant difference between baseline performance and later phase performance. Jack reached the a priori criterion of 80% accuracy on treated behaviours over two consecutive treatment sessions in the fifth treatment session. His therapy target was therefore changed from single pseudo-words to pseudo-words in carrier phrases from session 6. During baseline, Jack's per cent accuracy on treated pseudo-words in carrier phrases ranged between 0% and 10% (figure 3, panel B). In probe 7, following the introduction of treatment on single pseudo-words, his performance on pseudo-words in carrier phrases improved to 90% accuracy, suggesting generalization of treatment effects (see below). His accuracy with carrier phrases in probes 8 and 9 was similar to probe 7.

Emily's performance with the to-be-treated items during the baseline phase ranged from 45% to 60% accuracy (figure 4, panel A). During the treatment phase, her treated pseudo-word accuracy ranged from 84% to 90%, and planned contrasts confirmed Emily had significantly better performance in later phases than in baseline. Emily reached the a priori criterion of 80% accuracy on treated single pseudo-words over two consecutive treatment sessions in the 10th treatment session. Her treatment goal changed to the production of pseudo-words in carrier phrases from session 11. Figure 4, panel B, shows that during baseline Emily's per cent accuracy on treated pseudo-words in carrier phrases ranged between 5% and 15%. Her accuracy with pseudo-words in carrier phrases improved when she started treatment on single pseudo-words, suggesting generalization of treatment effects to more complex stimuli (see below).

Luke's per cent accuracy with the to-be-treated items during the baseline phase ranged from 5% to

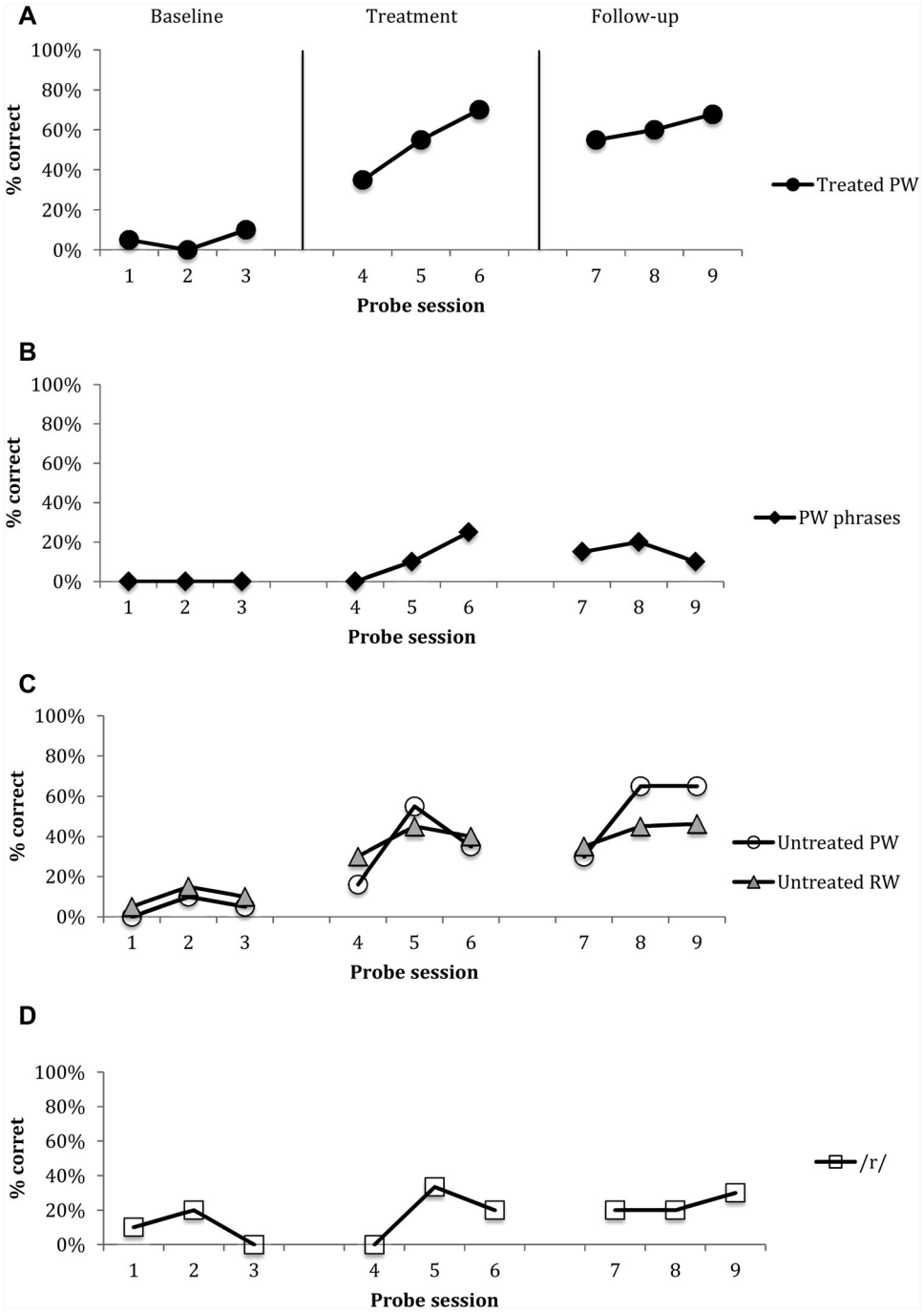

Figure 2. Oliver's results. PW, pseudo words; RW, real words.

10% (figure 5, panel A). During the treatment phase his accuracy with treated pseudo-words was 50–65% and planned contrasts confirmed that the improvement from baseline to the later phases (treatment and follow-up) was significant.

Lachlan's per cent accuracy with the to-be-treated items during baseline was 0–11% (figure 6, panel A). Within the treatment phase his per cent accuracy steadily improved to 75%, resulting in a significant difference between the baseline phase and the later phases.

Generalization of treatment effects

Oliver showed significant generalization to untreated pseudo-words and untreated real words (figure 2,

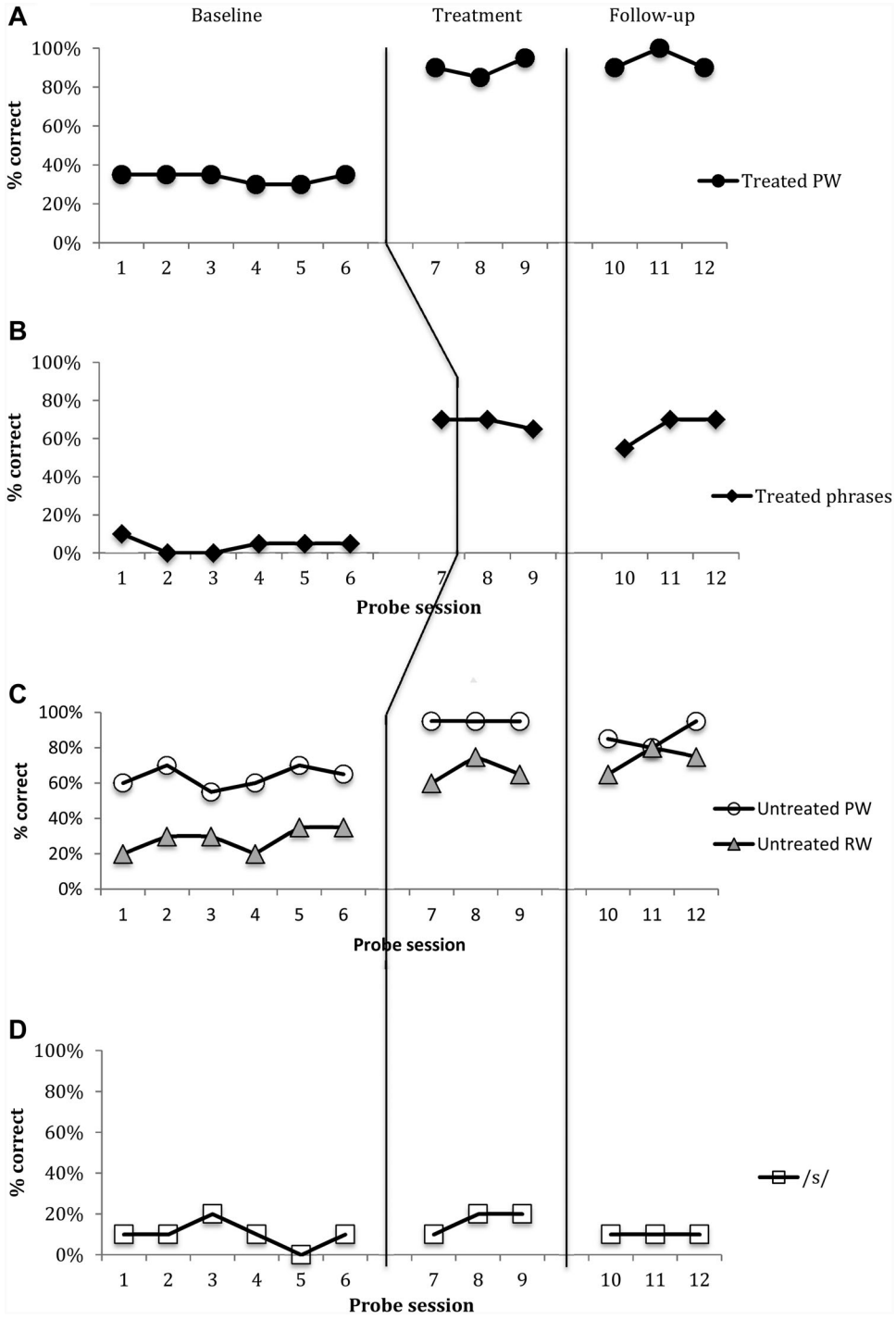

Figure 3. Jack's results. PW, pseudo words; RW, real words.

panel C). During baseline, his per cent accuracy with untreated pseudo-words and untreated real words was 5–15% and 0–10% respectively. During the treatment phase, his accuracy for these items improved significantly to 16–55% and 30–45% respectively. Visual inspection of Oliver's accuracy with pseudo-words in carrier phrases (figure 2, panel B) indicates a small improvement in these items during the treatment phase, which was not statistically significant.

Jack generalized his skill to similar, but untreated, pseudo-words and untreated real words, as shown in figure 3, panel C. In baseline, his accuracy was 60–70% for untreated pseudo-words and 20–35% for untreated real words. During the treatment phase, his performance

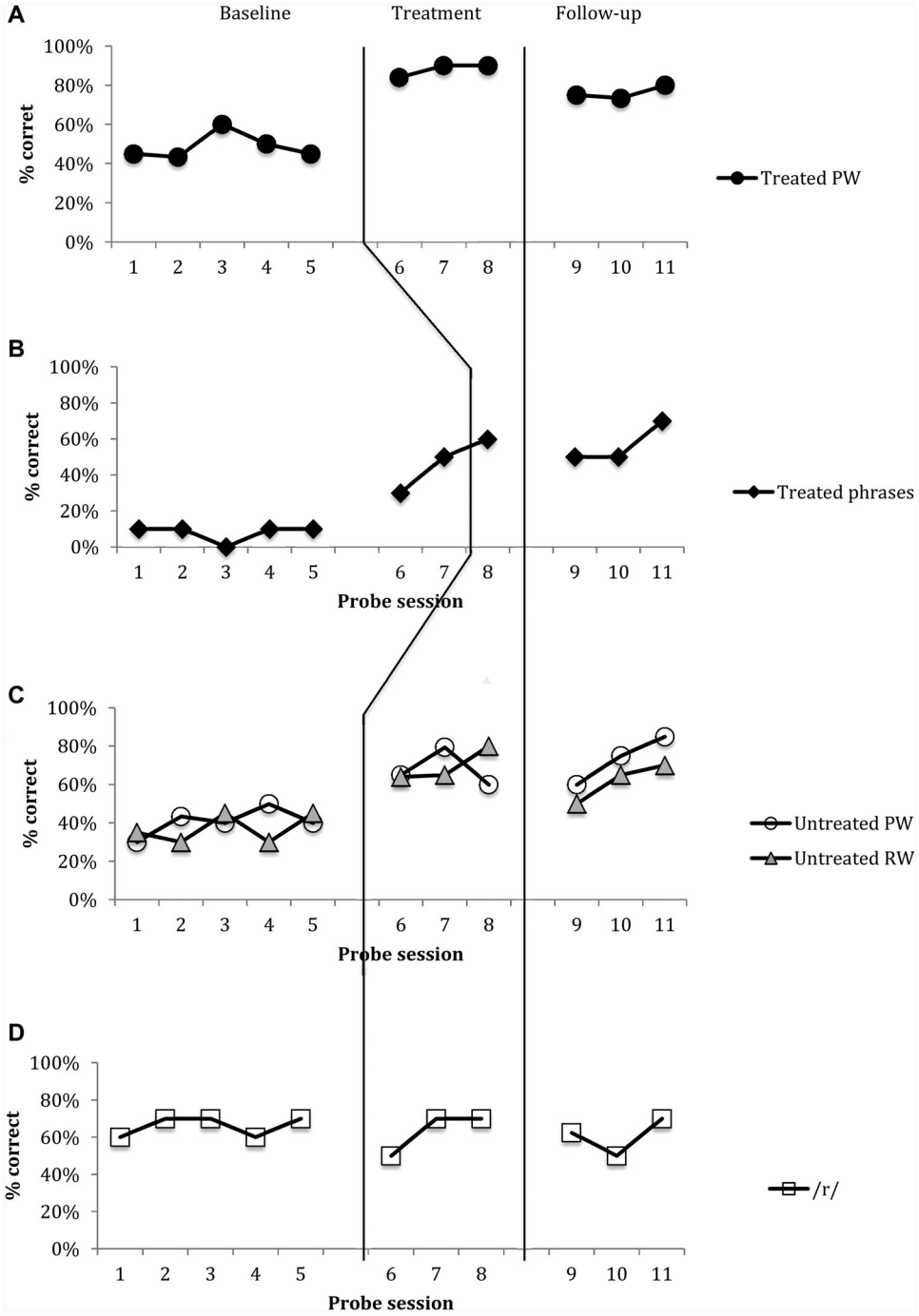

Figure 4. Emily's results. PW, pseudo words; RW, real words.

improved to 95% accuracy for untreated pseudo-words and 60–75% for untreated real words. Planned contrasts confirmed these differences were significant. As treatment shifted to treated pseudo-words in carrier phrases after probe 7, Jack's performance with treated words in carrier phrases in probe 7 was compared with his performance in the other baseline probes (figure 3, panel B). Jack's accuracy with pseudo-words in carrier phrases increased from < 10% in probes 1–6 to 70% in probe 7 following the introduction of treatment on single pseudo-words. Visual inspection indicated that there was no difference between performance on treated pseudo-words in carrier phrases between probe session 7 and later probe sessions, suggesting generalization to treated carrier phrases occurred once treatment began on the pseudo-words (figure 3, panel B).

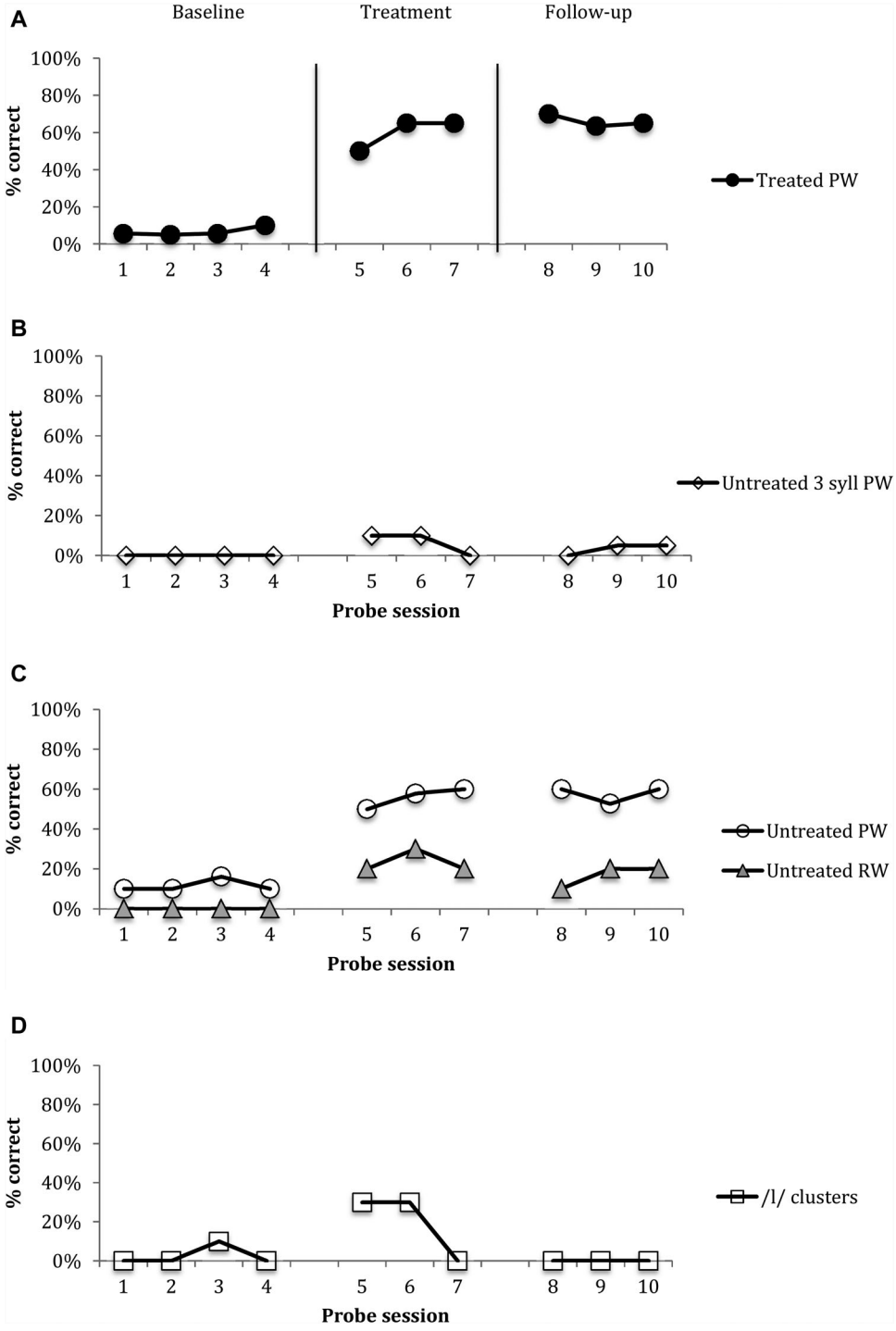

Figure 5. Luke's results. PW, pseudo words; RW, real words; syll., syllable.

Emily showed generalization to untreated pseudo-words and untreated real words (figure 4, panel C). During baseline, her per cent accuracy with untreated pseudo-words and untreated real words was 35–50% and 35–45% respectively. During the treatment phase, her accuracy for these items improved to 60–79% and 65–80% respectively, and the difference between performance in the baseline phase and later phases was significant. As discussed previously, Emily also showed generalization to carrier phrases with pseudo-words prior to treatment at the carrier phrase level. We compared her accuracy with carrier phrases in probe 7 (the last probe prior to treatment on pseudo-words in phrases) to probes 1–5 (prior to treatment on single

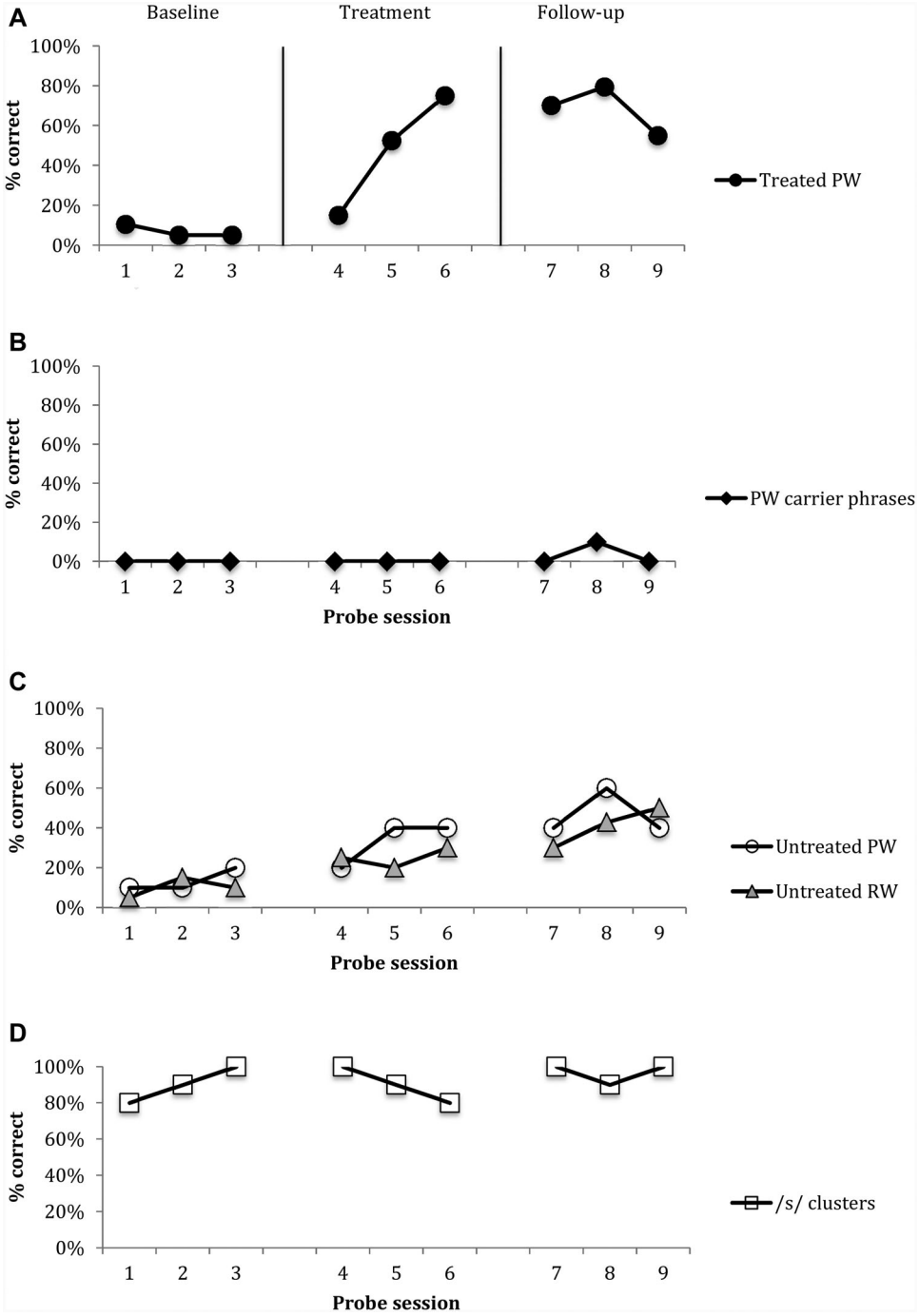

Figure 6. Lachlan's results. PW, pseudo words; RW, real words.

pseudo-words). In the first five probe sessions Emily achieved < 10% accuracy on treated pseudo-words in carrier phrases. Her accuracy with this behaviour improved steadily once treatment began on single pseudo-words, resulting in 50% accuracy in probe 7. This suggests generalization to carrier phrases with pseudo-words once treatment began on single pseudo-words (figure 4, panel B).

Luke generalized his skills to similar, but untreated, pseudo-words and untreated real words, as shown in figure 5, panel C. During baseline, his accuracy with untreated pseudo-words and untreated real words was 10–16% and 0% respectively. His accuracy improved on these untreated items during the treatment phase resulting in accuracy levels of 50–60% for untreated pseudo-words and 20–30% for untreated real words.

Planned contrasts confirmed that these improvements were significant. Although visual inspection indicates a small, temporary improvement with more complex items (three-syllable pseudo-words; figure 5, panel B), the change in these items between the baseline phase and later phases (i.e., treatment and follow-up) was not significant.

Lachlan showed significant generalization to untreated pseudo-words and untreated real words (figure 6, panel C). During baseline, his per cent accuracy with untreated pseudo-words and untreated real words was 10–20% and 5–15% respectively, both with slightly rising baselines. During the treatment phase, his accuracy with these items improved to 20–40% and 20–30% respectively with the slope greater than predicted by the rising baseline. His untreated pseudo-words showed autocorrelation of the residuals at lag 1 prohibiting statistical analyses. Planned contrasts indicated a significant difference between baseline and later phase performance on untreated real words. Visual inspection indicates Lachlan did not show generalization to pseudo-words in carrier phrases (figure 6, panel B); statistical analysis was not conducted on this data set due to autocorrelation of the residuals.

Maintenance of treatment and generalization effects

Most of the participants' treatment and generalization gains were maintained for 4 months post-treatment. Oliver, Jack, Lachlan and Luke maintained all treatment and generalization effects throughout the follow-up period. They had higher per cent accuracy at all follow-up points than baseline levels, for each of treated pseudo-words, similar but untreated words, and untreated real words. Planned contrasts revealed no significant difference between treatment phase and follow-up phase accuracy for any of these items for Oliver, Jack and Luke, supporting maintenance of effects to 4 months post-treatment. Lachlan had significantly higher accuracy in the follow-up phase than the treatment phase for untreated real words, indicating improving performance following the withdrawal of treatment. Jack also maintained his skill with treated pseudo-words in phrases. His per cent accuracy at two of the follow-up points was at the same level as probe 7 (the final probe prior to treatment on carrier phrases), and at all follow-up points was higher than baseline levels. No significant difference was found between his treatment phase and follow-up phase performance on pseudo-words in carrier phrases, supporting maintenance of skill.

Emily maintained some of her treatment gains and all of her generalization gains. With regard to maintenance of treatment gain, Emily lost some of her gain with treated pseudo-words. She had significantly lower accuracy in the follow-up phase for these items than the treatment phase, even though all follow-up points had higher accuracy than baseline levels. She did however maintain her treatment gain with treated pseudo-words in phrases. For these items, two follow-up points had the same per cent accuracy as probe 7 (the final probe prior to treatment on those items), and one had higher accuracy. Her performance was above baseline levels at all follow-up points for untreated pseudo-words, and untreated real words, and there was no significant difference between the treatment and follow-up phase accuracy these items, indicating maintenance of generalization effects.

In order to monitor the participants' progress at different time points *within* the follow-up phase, the data for the four participants were grouped and Helmert planned orthogonal contrasts were performed. There was no significant difference between the participants' performance at any of the time points, indicating stable, rather than improving or deteriorating performance in the follow-up phase'.

Control behaviour

Oliver, Jack and Emily did not show significant change in the behaviours we selected to monitor for maturational control (/r/, /s/, /r/ respectively) between the baseline and later phases. Luke's accuracy with the behaviour we selected to monitor to control for maturation effects (/l/ clusters), significantly improved during the treatment phase, and then significantly decreased in the follow-up phase. With regard to Lachlan, the behaviour we selected to monitor to control for maturation effects (/s/ clusters), demonstrated a ceiling effect (80–100% correct) in the baseline phase, prohibiting adequate evaluation of change during the treatment phase. However Lachlan's performance on a stimulus generalization measure (production of treated pseudo-words in carrier phrases) showed no significant change during the entire research period.

Adequacy of technology

Although 61% of the sessions were rated by the treating clinician as having technology difficulties, only one of the 113 sessions (< 1%) was cancelled due to a technical issue, namely the family had exceeded their service provider's monthly data allowance. One additional session was conducted partly by telephone, due to issues with sound transmission during the video conference. At the time of the final follow-up appointment, we assessed the speed of connection for all participants and clinicians. The download speed was above 50 Mbps for Jack, Emily and Lachlan, and below 4 Mbps for Oliver and Luke. Oliver and Luke had lower clinician ratings of technological adequacy than the other participants, with

average ratings of 5.45 and 6.73 out of 10 respectively, compared with an average rating for the other children across all sessions of 8.40. The most frequent technical difficulties experienced were difficulty establishing audio connection, web-camera freezing, and latency in the audio signal. At technology adequacy ratings of less than four (9% of sessions), clinicians reported feeling frustrated, annoyed, stressed, and disappointed with the technology. At technology adequacy rating levels above four (91% of sessions), clinicians reported feeling 'fine', 'comfortable', 'OK' and 'great'.

Satisfaction with video conferencing

The parents were very satisfied with the video conferencing treatment (average score = 9.5, range 7.5–10), and they reported their children were motivated to participate in video conferencing sessions (average score = 8, range = 6.5–10) and they found the home-based treatment very convenient (average score = 9.7, range = 8.5–10). The treating clinicians reported high levels of satisfaction (average score = 8.75, range 7.5–10) and convenience (average score = 9.25, range = 8.5–10) with the telehealth treatment.

Discussion

This study aimed to evaluate the efficacy of ReST treatment for children with CAS when provided by video conferencing. We hypothesized that treatment via video conferencing would result in (1) significant improvement in imitated pseudo-words, (2) significant generalization to related but untreated imitated speech behaviours, and (3) maintenance of treatment and generalization effects. The hypotheses were supported with all five children showing positive gains, and four of the five children maintaining their gains to 4 months post-treatment.

Experimental control was indicated by the establishment of stable baselines prior to the introduction of treatment, and the demonstration of improved performance on the dependent variable when treatment commenced for all five children. Additionally, control for maturation was demonstrated for all five children. Three children (Oliver, Jack and Emily) made no significant change with the behaviour we selected as a maturational control. For Luke, the behaviour we selected for this purpose, (/l/ clusters), co-varied with the treatment. His return to baseline levels following the withdrawal of treatment argues against a maturation effect. For Lachlan, the behaviour we selected to monitor for signs of maturation (/s/ clusters), demonstrated a ceiling effect in the baseline phase. Although a behaviour with lower levels of baseline performance would have ideally been selected, Lachlan did not have another speech behaviour appropriate for this purpose. His performance on a stimulus generalization task (production of pseudo-words in carrier phrases) was stable throughout the research period. Although unrelated behaviours are usually selected to monitor for maturational change, an alternative way is to monitor for stimulus generalization. Lachlan's lack of change with a stimulus generalization task argues against maturational change, and supports internal validity.

Video conferencing ReST treatment had similar effects to face-to-face treatment (Ballard *et al.* 2010, Murray *et al.* 2015, Thomas *et al.* 2014). Both service delivery methods resulted in significant acquisition of pseudo-words, with large effect sizes. Significant generalization to untreated but related behaviours, and maintenance of treatment and generalization gains to 4 months post-treatment was shown in both the face-to-face and telehealth modality.

Two of the participants not only generalized to untreated items at the same level as treatment, but also to more complex behaviours. Emily and Jack, who generalized to the more complex behaviour of pseudo-words in carrier phrases, had milder speech difficulties initially than the other participants, were older, had accuracy levels above 80% during treatment and some minimal knowledge of the more complex behaviour in baseline. Greater generalization in ReST treatment has been previously demonstrated for children with milder speech difficulties (Ballard *et al.* 2010, Thomas *et al.* 2014) and ReST treatment is generally more effective for older children with milder speech difficulties (Murray *et al.* 2013). Given that generalization to more complex behaviours occurred prior to treatment at that level, it raises the question of whether the children required treatment on the more complex behaviour. Further investigation of generalization to more complex behaviours during ReST treatment is warranted.

With the exception of Emily's accuracy with treated pseudo-words, all children maintained their gains to 4 months post-treatment. Emily's loss of some treatment gain with single pseudo-words is difficult to explain, particularly as she had high levels of treatment accuracy and strong generalization. Perhaps she did not maintain sufficient focus on the single pseudo-words after her treatment moved to phrases. Like the other participants, on all other behaviours, Emily had stable performance in the follow-up phase. This stable performance in the follow-up phase was also shown in face-to-face ReST treatment delivered twice weekly (Thomas *et al.* 2014), while face-to-face ReST treatment provided four times weekly resulted in significant ongoing improvement during the follow-up phase (Murray *et al.* 2015). This present study was different to that of Murray and colleagues in two significant ways: children with receptive language impairments were included and the mode of treatment was video conferencing rather than face to

face. Either of these factors, or a combination of the two, may account for the superior performance in the maintenance phase for children receiving face-to-face treatment versus video conferencing treatment of the same intensity.

Three of our participants had receptive language impairments, and four had expressive language impairments. The treatment effect for children with language impairments, particularly receptive impairments, may potentially be reduced. However, given that all participants demonstrated significant acquisition of the targeted pseudo-words and generalization effects, any limitation associated with the inclusion of participants with language impairments is minimal.

The stable performance during maintenance in this study was a positive finding, given the relatively low levels of treatment accuracy shown by Lachlan, Luke and Oliver. Previous studies with ReST and other motor speech disorders have indicated that high levels of treatment accuracy, around 70%, for approximately five treatment sessions are generally required for maintenance of treatment gains (Ballard *et al.* 2010, Wambaugh *et al.* 2013). ReST treatment, with its use of motor learning principles to facilitate generalization and maintenance, has previously demonstrated maintenance of treatment gains, even with treatment accuracy levels below 70% (Staples *et al.* 2008, Thomas *et al.* 2014). These findings suggest that clinicians may be able to use a lower criterion for treatment accuracy than is currently recommended for ReST treatment.

With regard to the technology used in the sessions, although the majority of the sessions had some technical difficulty, fewer than 1% of sessions were cancelled, indicating that the technical issues were tolerable for the families. Audio latency was the most troubling technical issues because it affected the interaction between clinician and client, as well as the ability to provide timely feedback and no solution was available for sessions with audio latency. Although most of the other technical issues could be resolved, in some cases problem solving took up to 10 min, which was more than 15% of the session. Parents reported that the two familiarization sessions were valuable for improving their technical skill and confidence. The time required for solving technical problems and familiarizing families with video conferencing systems needs to be factored in when considering using telehealth treatments.

Despite the technical challenges, ReST treatment was efficacious in this format. It may be that the nature of a high-production trial treatment with minimal need for physical prompts such as ReST is well suited to video conferencing. CAS treatments requiring more hands-on cueing such as Dynamic Temporal and Tactile Cueing (DTTC) (Strand *et al.* 2006) may be less suitable for video conferencing.

Parents and clinicians found the system convenient, motivating for the child, and were satisfied with their experience of therapy via video conferencing. The high levels of satisfaction and convenience may be related to the interactive games played using Adobe Connect's 'draw' function during session breaks and the reduction in travel time with home-based video conferencing. This high satisfaction is in keeping with previous telehealth studies (e.g., Constantinescu 2012). The children attended all of their treatment and probe sessions. It is possible that the benefits in terms of convenience helped outweigh technical difficulties experienced.

The reliability of phonemic transcription was similar in this study to face-to-face ReST treatment (cf. Thomas *et al.* 2014). Based on previous research indicating difficulty perceiving high frequency sounds, clusters, and phonemes without visible articulation (Eriks-Brophy *et al.* 2008, Waite *et al.* 2006) we would not have been surprised to find poor reliability for the control items (/s/, /l/ clusters, /s/ clusters, and /r/), however the average intra- and inter-rater reliability for the control items was acceptable at 93.5% and 81.5% respectively.

Limitations and future directions

This was a small, phase 1 study. It would be beneficial to investigate the use of video conferencing for ReST treatment in a larger group study, and to investigate the factors affecting treatment outcomes for children. It would also be beneficial to know if the results would be replicated within a community clinical setting, as our study was conducted within a university treatment research clinic.

Related to design, we had three to six data points in the baseline phase, and three in the treatment and follow-up phases. More data points in each phase, with a minimum of five in the treatment phase would be preferable. We demonstrated control for maturational effects on the selected behaviour for four of the five participants. We only demonstrated control for maturational effects for Lachlan on a stimulus control behaviour. Further studies should explore options for behaviours appropriate to monitor for maturational change, and explore stimulus generalization tasks and more complex behaviours as control measures for this purpose.

In this study, two participants demonstrated generalization to more complex speech behaviours. Further investigation of the factors associated with generalization in ReST treatment is required, and more data collection points within each phase may clarify the results. The participants in this study imitated the treatment and probe items, which may lead to limited generalization to spontaneous speech. Further investigation of the spontaneous speech production following ReST treatment is warranted.

The dependent variable in this study was the percentage of words produced with correct sounds, lexical stress and smooth connection between the syllables. It would be beneficial to investigate the change within each construct, within participant, over the course of treatment in order to provide information about precisely what changes for each child, when the change occurs, and any pattern of change. The participants in this study used headphones, which would have attenuated their auditory feedback from their own speech. This may have potentially reduced the treatment effect, but given the large treatment effects demonstrated may not be of significance on this occasion. Finally, future speech treatment studies using video conferencing should include routine testing of the bandwidth at the start of each session in order to provide information about the minimum bandwidth for effective treatment.

Conclusions

This study evaluated the efficacy of ReST treatment provided by telehealth to five children with CAS. Results showed significant acquisition of the imitated targeted pseudo-words, and generalization of the treatment effect to untreated imitated pseudo-words and real words. These results suggest that video conferencing as a service delivery method for ReST treatment may be beneficial for children with CAS. These results warrant larger scale studies.

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Notes

1. For children ≥ 11 years, the stimuli of the DEAP (Dodd *et al.* 2006) were not considered sufficiently challenging to assess inconsistency. For these children an inconsistency measure was calculated for 25 words from the Polysyllabic Word Test (Gozzard *et al.* 2006). As there are no guidelines for the severity of inconsistency with these stimuli, we assumed children of 11 years would show < 30% inconsistency. For children < 11 years we used the 40% criteria for inconsistency, as reported by Dodd *et al.* (2006).
2. Due to logistical constraints, the first author, experienced in ReST treatment via telehealth, conducted one treatment session of both Jack and Emily's 12 treatment sessions.

References

American Speech–Language–Hearing Association, 2007, *Childhood Apraxia of Speech*. Technical Report (available at: http://www.asha.org/policy/PS2007-00277.htm) (accessed on 22 of August 2015).

Ballard, K. J., Robin, D. A., Mccabe, P. and Mcdonald, J., 2010, A treatment for dysprosody in childhood apraxia of speech. *Journal of Speech Language and Hearing Research*, **53**, 1227–1245.

Beeson, P. M. and Robey, R. R., 2006, Evaluating single-subject treatment research: lessons learned from the aphasia literature. *Neuropsychology Review*, **16**, 161–169.

Carey, B., O'brian, S., Lowe, R. and Onslow, M., 2014, Webcam delivery of the Camperdown Program for adolescents who stutter: a phase II trial. *Language, Speech, and Hearing Services in Schools*, **45**, 314–324.

Ciccia, A. H., Whitford, B., Krumm, M. and Mcneal, K., 2011, Improving the access of young urban children to speech, language and hearing screening via telehealth. *Journal of Telemedicine and Telecare*, **17**, 240–244.

Constantinescu, G., 2012, Satisfaction with telemedicine for teaching listening and spoken language to children with hearing loss. *Journal of Telemedicine and Telecare*, **18**, 267–272.

Constantinescu, G., Theodoros, D., Russell, T., Ward, E., Wilson, S. and Wootton, R., 2011, Treating disordered speech and voice in Parkinson's disease online: a randomized controlled non-inferiority trial. *International Journal of Language and Communication Disorders*, **46**, 1–16.

Dodd, B., Zhu, H., Crosbie, S., Holm, A. and Ozanne, A., 2006, *Diagnostic Evaluation of Articulation and Phonology (DEAP)* (London: Psychology Corporation).

Dunn, L. M. and Dunn, D. M., 2007, *Peabody Picture Vocabulary Test (PPVT-4)* (Minneapolis, MN: Pearson Assessments).

Eriks-Brophy, A., Quittenbaum, J., Anderson, D. and Nelson, T., 2008, Part of the problem or part of the solution? Communication assessments of Aboriginal children residing in remote communities using video conferencing. *Clinical Linguistics and Phonetics*, **22**, 589–609.

Goldman, R. and Fristoe, M., 2000, *Goldman–Fristoe Test of Articulation 2 (GFTA-2)* (Minneapolis, MN: Pearson Assessments).

Gozzard, H., Baker, E. and Mccabe, P., 2006, Children's productions of polysyllables. *ACQuiring Knowledge in Speech, Language and Hearing*, **8**, 113–116.

Grogan-Johnson, S., Schmidt, A. M., Schenker, J., Alvares, R., Rowan, L. E. and Taylor, J., 2013, A comparison of speech sound intervention delivered by telepractice and side-by-side service delivery models. *Communication Disorders Quarterly*, **34**, 210–220.

Hill, A. J., Theodoros, D. G., Russell, T. G., Cahill, L. M., Ward, E. C. and Clark, K. M., 2006, An Internet-based telerehabilitation system for the assessment of motor speech disorders: a pilot study. *American Journal of Speech–Language Pathology*, **15**, 45–56.

Kazdin, A. E., 2011, *Single-Case Research Designs: Methods for Clinical and Applied Settings* (New York: Oxford University Press).

Keck, C. S. and Doarn, C. R., 2014, Telehealth technology applications in speech–language pathology. *Telemedicine and e-Health*, **20**, 653–659.

Lewis, B. A., Freebairn, L. A., Hansen, A. J., Iyengar, S. K. and Taylor, H. G., 2004, School-age follow-up of children with childhood apraxia of speech. *Language, Speech, and Hearing Services in Schools*, **35**, 122.

Maas, E., Gildersleeve-NEUMANN, C., Jakielski, K. and Stoeckel, R., 2014, Motor-based intervention protocols in

treatment of childhood apraxia of speech (CAS). *Current Developmental Disorders Reports*, **1**, 197–206.

Mashima, P. A. and Doarn, C. R., 2008, Overview of telehealth activities in speech–language pathology. *Telemedicine and e-Health*, **14**, 1101–1117.

Murray, E., Mccabe, P. and Ballard, K. J., 2012, A comparison of two treatments for childhood apraxia of speech: methods and treatment protocol for a parallel group randomised control trial. *BMC Pediatrics*, **12**, 122.

Murray, E., Mccabe, P. and Ballard, K. J., 2013, *Exploring factors that determined treatment success: data from a randomized control trial for childhood apraxia of speech*. Paper presented at the ASHA Convention. Chicago, IL, USA.

Murray, E., Mccabe, P. and Ballard, K. J., 2014, A systematic review of treatment outcomes for children with childhood apraxia of speech. *American Journal of Speech–Language Pathology*, **23**, 486–504.

Murray, E., Mccabe, P. and Ballard, K. J., 2015, A randomized control trial for children with childhood apraxia of speech comparing rapid syllable transition treatment and the Nuffield Dyspraxia Programme—Third Edition. *Journal of Speech, Language and Hearing Research*, **58**, 669–686.

Namasivayam, A. K., Pukonen, M., Goshulak, D., Hard, J., Rudzicz, F., Rietveld, T., Maassen, B., Kroll, R. and Lieshout, P., 2015, Treatment intensity and childhood apraxia of speech. *International Journal of Language and Communication Disorders*, **50**, 529–546. doi: 10.1111/1460-6984.12154

O'Brian, S., Smith, K. and Onslow, M., 2014, Webcam delivery of the Lidcombe Program for early stuttering: a phase I clinical trial. *Journal of Speech, Language, and Hearing Research*, **57**, 825–830.

O'Callaghan, A. M., Mcallister, L. and Wilson, L., 2005, Barriers to accessing rural paediatric speech pathology services: health care consumers' perspectives. *Australian Journal of Rural Health*, **13**, 162–171.

Reynolds, A. L., Vick, J. L. and Haak, N. J., 2009, Telehealth applications in speech–language pathology: a modified narrative review. *Journal of Telemedicine and Telecare*, **15**, 310–316.

Robbins, J. and Klee, T., 1987, Clinical assessment of oropharyngeal motor development in young children. *Journal of Speech and Hearing Disorders*, **52**, 271–277.

Ruggero, L., Mccabe, P., Ballard, K. J. and Munro, N., 2012, Paediatric speech–language pathology service delivery: an exploratory survey of Australian parents. *International Journal of Speech–Language Pathology*, **14**, 338–350.

Semel, E., Wiig, E. and Secord, W., 2006, *Clinical Evaluation of Language Fundamentals Fourth Edition, Australian Standardised Edition (CELF-4 Australian)* (Sydney, NSW: Pearson).

Skinder-Meredith, A., 2001, Differential diagnosis: Developmental apraxia of speech and phonologic delay. *Augmentative Communication News*, **14**, 5–8.

Speech Pathology Australia, n.d., *Code of Ethics* (available at: http://www.speechpathologyaustralia.org.au/library/Ethics/CodeofEthics.pdf\051 (accessed on 9 February 2015).

Staples, T., Mccabe, P., Ballard, K. J. and Robin, D. A., 2008, *Childhood apraxia of speech: treatment outcomes at 6 months of an intervention incorporating principles of motor learning*. Paper presented at the Joint New Zealand Speech–Language Therapy Association/Speech Pathology Australia Conference, Auckland, New Zealand.

Strand, E. A., Stoeckel, R. and Baas, B., 2006, Treatment of severe childhood apraxia of speech: a treatment efficacy study. *Journal of Medical Speech–Language Pathology*, **14**, 297–307.

Theodoros, D., 2011, Telepractice in speech–language pathology: the evidence, the challenges, and the future. *Perspectives on Telepractice*, **1**, 10–21.

Theodoros, D., 2013, Speech–language pathology and telerehabilitation. In S. Kumar and E. R. Cohn (eds), *Telerehabilitation* (London: Springer), 311–323.

Thomas, D. C., Mccabe, P. and Ballard, K. J., 2014, Rapid Syllable Transitions (ReST) treatment for childhood apraxia of speech: the effect of lower dose-frequency. *Journal of Communication Disorders*, **51**, 29–42.

Waite, M. C., Cahill, L. M., Theodoros, D. G., Busuttin, S. and Russell, T. G., 2006, A pilot study of online assessment of childhood speech disorders. *Journal of Telemedicine and Telecare*, **12**, 92–94.

Waite, M. C., Theodoros, D. G., Russell, T. G. and Cahill, L. M., 2012, Assessing children's speech intelligibility and oral structures, and functions via an Internet-based telehealth system. *Journal of Telemedicine and Telecare*, **18**, 198–203.

Wambaugh, J. L., Nessler, C., Cameron, R. and Mauszycki, S. C., 2013, Treatment for acquired apraxia of speech: examination of treatment intensity and practice schedule. *American Journal of Speech–Language Pathology*, **22**, 84–102.

Wiig, E. H., Secord, W. and Semel, E. M., 2004, *CELF Preschool 2: Clinical Evaluation of Language Fundamentals Preschool—Second Edition Australian Standardised Edition* (Sydney, NSW: Harcourt Assessment).

Supporting Information

Additional Supporting Information may be found in the online version of this article at the publisher's website:

Appendix 1 Probe stimuli for the children

Chapter 5: Delivery-Agent Modification–Combined Clinician–Parent Delivered Rapid Syllable Transition Treatment

Author attribution statement

Chapter 5 of this thesis has been published as Thomas, D. C., McCabe, P., & Ballard, K. J. (2017). Combined clinician–parent delivery of rapid syllable transition (ReST) treatment for childhood apraxia of speech. *International Journal of Speech-Language Pathology*, *April 26,* 1–16. Advance online publication.

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I declare that I made the following contribution to the study:

- Conception of the research question in collaboration with the other authors
- Design of the study with the other authors
- Collection of data
- Data entry and analysis
- Writing of the first draft of the manuscript
- Journal revisions and resubmission

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Date: 27.6.17

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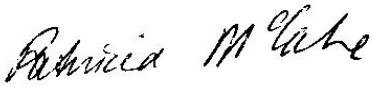
Sign: 

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Combined clinician-parent delivery of rapid syllable transition (ReST) treatment for childhood apraxia of speech

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Abstract

Purpose: Although speech-language pathologists use parent-delivered home-practice, little is known about the quality of this practice and its relation to treatment efficacy. This study reports both treatment outcomes and fidelity following combined clinician-parent delivery of Rapid Syllable Transition (ReST) treatment.
Method: Five children aged 5:1–11:7 with childhood apraxia of speech received 12 treatment sessions; six clinic-based and six at home, using multiple baselines across participants design. We investigated the children's acquisition of treated pseudo words, generalisation to untreated pseudo and real words, and maintenance of gains. We also assessed parent and clinician treatment fidelity and reliability of perceptual judgements.
Result: Two children improved on all treated behaviours; two showed treatment effect on one of their two treated behaviours, and one child had no treatment effect. Only two children generalised to the majority of untreated items. Variable treatment fidelity was found across parents and aspects of treatment. Child outcome was likely influenced by multiple factors, including treatment fidelity, reliability of perceptual judgements and child factors.
Conclusion: Combined clinician-parent delivery of ReST was less efficacious than previously reported clinician-only delivered ReST. Further investigation of the factors affecting outcome is recommended prior to clinical application of the combined model of service delivery.

Keywords: *therapy*; *caregiver*; *service-delivery*; *dyspraxia*; *treatment fidelity*

Introduction

Children with childhood apraxia of speech (CAS) are reported to require more frequent treatment sessions, across a longer time period than children with other speech impairments (Hall, Jordan, & Robin, 1993, Skinder-Meredith, 2001). A recent systematic review of treatments for CAS identified that effective treatment requires a minimum of 2–3 sessions per week, with at least 60 production trials per session (Murray, McCabe, & Ballard, 2014). This is significantly more than the one session per week, which is the most common Speech-Language Pathology (SLP) service delivery in Australia, England and Canada (Ruggero, McCabe, Ballard, & Munro, 2012). The limited service available leads to frustration for families (Bercow, 2008; Ruggero et al., 2012) and clinicians (Lim, McCabe, & Purcell, in press).

One common strategy to increase treatment intensity is to train parents to work on speech activities with children at home (Lim et al., in press; McLeod & Baker, 2014). Although parents expect to be involved in their children's therapy, they prefer regular therapy from a Speech-Language Pathologist (Glogowska & Campbell, 2000) rather than treatment via a home programme or parent training (Ruggero et al., 2012).

Involving carers in treatment delivery

Clinic sessions supplemented with regular home practice are effective in the treatment of many communication impairments including some language difficulties (Girolametto, 1988) and childhood stuttering (Jones et al., 2005). This combined model of clinic-based sessions with home-practice is a feature of several speech treatment programmes, such as Dynamic Temporal and Tactile Cueing (DTTC, Strand, Stoeckel, & Baas, 2006), Parents and Children Together (PACT, Bowen, 2010), as well as eclectic speech-sound treatment combining several different input and output tasks (Lancaster, Keusch, Levin, Pring, & Martin, 2010).

Exclusively parent-delivered treatment for speech production impairments is also associated with

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positive outcomes. A meta-analysis by Lawler, Taylor, and Shields (2013) indicated moderate evidence that treatment provided exclusively by parents produced statistically similar improvement in speech skills to treatment provided exclusively by clinicians. The meta-analysis included four studies with a total of 88 participants diagnosed with speech impairments. Stronger improvement was shown for treatment provided by parents in three studies, with stronger improvement following treatment provided by SLP clinicians in one study. None of the children included in the meta-analysis were diagnosed with CAS. It is not yet known whether parents are as effective with treating CAS, which is a more complex speech impairment.

Despite the promising indications of effectiveness for parent-delivered treatment in speech sound disorders, clinicians infrequently ask parents to be the primary intervention provider when delivering speech-sound therapy (Sugden, Baker, Munro, & Williams, 2016). The relative reluctance for clinicians to ask parents to be the primary provider of speech-sound therapy may be due to concerns about the perceived complexity of the disorder or the treatment (Sugden et al., 2016). Delivering treatment in the way it was initially described (i.e. ensuring high treatment fidelity) is essential for positive treatment outcomes and is a key feature of delivering Evidence-Based Practice (EBP) (Kaderavek & Justice, 2010). It remains to be seen whether parents can deliver treatment for CAS with high levels of treatment fidelity and whether such a treatment is effective.

Childhood apraxia of speech. Home-based speech practice is recommended for treatment of CAS (Maas, Gildersleeve-Neumann, Jakielski, & Stoeckel, 2014) and a recent systematic review found that 40% of CAS treatments prescribe some level of home-based practice (Murray et al., 2014). Given the reported need for high frequency treatment sessions, and the common employment of home-based practice for increasing treatment intensity for other communication impairments, it is important to evaluate the fidelity and the outcomes of parent-delivered treatment for CAS.

Purpose

In this study, we investigated the efficacy of combined clinician-parent delivered intervention for CAS. We selected the Rapid Syllable Transition (ReST) treatment programme as it has been shown efficacious for CAS with intensive clinic-based delivery (Murray, McCabe, & Ballard, 2015; Thomas, McCabe, & Ballard, 2014). It uses pseudo words with varied lexical stress patterns to improve children's speech sounds and prosody. When ReST is provided by a clinician in the clinic, for four sessions per week across three consecutive weeks, children's speech sound accuracy and prosodic accuracy typically improve for production of treated pseudo words, as well as untreated pseudo and real words (Murray et al., 2015). All previous studies applying ReST treatment have reported on the outcomes following exclusively clinician-delivered treatment without home-practice.

To facilitate comparison with previously published ReST studies, the participants in this study had the same number, frequency and duration of sessions as well as number of production trials. Our hypotheses were as follows:

(a) The combined model of clinician-parent delivered ReST treatment would be efficacious, in terms of:
   - acquisition of treated pseudo words,
   - generalisation to untreated pseudo words and real words,
   - maintenance of acquisition and generalisation gains to four-month post-treatment.

(b) Parent treatment fidelity would be $\geq$85% when parents are provided with individual training during their child's clinic-based sessions.

(c) The effect size for improvement in treated items would be within one standard deviation of that obtained by children receiving 12 sessions of clinician-delivered ReST treatment.

Method

The research project was approved by The University of Sydney's Human Ethics Committee – approval number 2012/2824.

Participants

Five parent-child dyads participated in the study. There were separate inclusion criteria for children and adults, and both halves of the dyad were required to meet the relevant criteria. The inclusion criteria for the children were: (a) a consensus diagnosis of CAS, as described below, (b) pure tone audiogram thresholds at least 20dB at 500Hz, 1, 2 and 4KHz bilaterally, (c) no evidence of oral structural impairments (Robbins & Klee, 1987), (d) no other existing neurological diagnoses, and (e) proficient English speaker. All child and parent participants were monolingual Australian-English speakers, except for Ben's parent Sam, who was a bilingual Turkish-English speaker. Although Ben was exposed to Turkish, parental report was that he did not speak Turkish.

In order to provide more detail about the children's speech and language skills relative to age-matched peers, we used three additional tests: (a) the Clinical Evaluation of Language Fundamentals – Preschool Second Edition (CELF-P2; Wiig, Secord, & Semel, 2004) or the Clinical Evaluation of Language Fundamentals – 4th Edition Australian version (CELF–4; Semel, Wiig, &

Secord, 2006), depending on age, (b) the Goldman Fristoe Test of Articulation 2 (Goldman & Fristoe, 2000), and (c) the Peabody Picture Vocabulary Test (Dunn & Dunn, 2007).

Inclusion criteria for the parents were: (a) normal receptive vocabulary (Dunn & Dunn, 2007), (b) pure tone audiogram thresholds as above, (c) non-word repetition skills within the normal range for young adults (Torgesen, Wagner, & Rashotte, 1999), and (d) proficient English speaker.

The diagnosis of CAS was made based on perceptual judgment of presence of the three core features of CAS (American Speech-Language-Hearing Association [ASHA], 2007) during a battery of speech production tests. We operationally defined these core features as: (a) when a child with a Percentage Consonants Correct (PCC) ≥ 90 showed >30% inconsistency over three separate administrations of 25 words from the Test of Polysyllables (Gozzard, Baker, & McCabe, 2006), or a child with PCC <90 showed >40% inconsistency during the Inconsistency subtest of the Diagnostic Evaluation of Articulation and Phonology (Dodd, Zhu, Crosbie, Holm, & Ozanne, 2006), (b) when a minimum of 10 polysyllabic words were perceived as having syllable segregation during the Test of Polysyllables (Gozzard et al., 2006), indicating difficulty transitioning between syllables, and (c) when $\geq$15% words on the Test of Polysyllables were produced with incorrect stress pattern (Gozzard et al., 2006). These tools have previously been reported for determining diagnosis in ReST treatment studies (e.g. Murray et al., 2015; Thomas et al., 2016). All three features were needed, from two independent raters, to assign the diagnosis of CAS. Parents, children and clinicians were assigned pseudonyms. The children's speech and language skills are reported in Table I.

Probe stimuli

A probe list was developed to allow for analysis of (a) acquisition and maintenance of treated pseudo words, and (b) generalisation of treatment effects to untreated pseudo words and untreated real words. Pseudo words were constructed with the vowels /i/, /a/, /ɔ/, and /ə/ and consonants /f/, /d/, /b/ and /k/. Syllable strings were concatenated CV syllables (e.g. /ˈbakə/, /ˈdəfəbi/). The stress patterns of strong-weak (SW or trochaic; e.g. /ˈbakəfi/) and weak-strong (WS or iambic; e.g. /fəˈkibə/) across the first two syllables were equally represented in each stimulus subset. The complete probe list is presented in Supplementary material A.

For Ben, Stacey and Eric, treatment targeted 3-syllable pseudo words. The 160-item probe list comprised 40 3-syllable (CVCVCV) pseudo words. Half of these (10 SW and 10 WS across the first two syllables) were selected for treatment and the other half were kept in baseline to assess generalisation to untreated related items. The probe set also included 20 carrier phrases (e.g. I want a _____) with the 3-syllable strings (10 treated and 10 untreated), and 20 each of 2- and 4-syllable pseudo words, and 2-, 3- and 4-syllable real words.

Matt commenced treatment on 3-syllable pseudo words in carrier phrases. His probe list contained 180 items; the 160 items outlined above, and 20 carrier phrases containing two pseudo words (e.g. "I found a /bəˈdikə/ and a /ˈfadəbi/") to allow analysis of his performance on more complex items.

Julian commenced treatment on 2-syllable pseudo words. His probe list included 160 items, 40 2-syllable pseudo words (20 treated and 20 untreated), 20 carrier phrases with the 2-syllable pseudo words, 20 2-syllable strings with the same consonant repeated (e.g. /ˈfarfə/), 20 3-syllable pseudo words, and 20 each of 2-, 3-, and 4- syllable real words.

Design

The study used a multiple baselines across participants design (Kazdin, 2011). Baseline sessions were twice-weekly and participants had 3-, 4- or 5-baseline probes prior to the commencement of therapy. The number of baseline sessions was randomly allocated. The children each had 12 treatment sessions across 3 weeks, with performance probed three times during the treatment phase and three times in the follow-up phase (see Figure 1).

During the period from the start of baseline testing until one-month post-treatment none of the children had any other speech therapy. The children were permitted to return to their community SLP after the one-month follow-up point. Stacey and Ben did so, with Stacey receiving therapy for language skills and Ben receiving further ReST treatment.

Baseline and probe sessions. Identical procedures were used for the baseline and probe sessions. The probe stimuli were presented in one of three randomised orders. The participants viewed a PowerPoint slide show, with one stimulus item per slide. All items were imitated; a pre-recorded sound file by an Australian-English female speaker was used for each non-word item and the examiner provided a live model for each real word item.

Procedure

The first author, a qualified SLP, carried out the eligibility assessments and baseline probes. Six of the 12 treatment sessions were delivered by a trained SLP student in the clinic under the supervision of the first and second authors, one clinician per child. All SLP student clinicians and clinical educators were Australian-English speakers. One parent was designated to attend all of the child's clinic-based sessions and deliver the six home-based sessions. The participants had more clinic-based sessions in

Table I. Participants' initial assessment results.

Test	Stacey (5 yrs:1 mth)	Eric (7yrs:7mth)	Ben (5yrs:10mth)	Matt (11yrs:7mth)	Julian (10yrs:6mth)
Clinical Evaluation of Language Fundamentals – Preschool Second Edition (CELF-P2) or Fourth Edition (CELF-4)					
Receptive Language Index					
Standard Score (%ile)	103 (58)	88 (21)	73 (4)	100 (50)	73 (4)
Interpretation	WNL	WNL	mod.	WNL	mod.
Expressive Language Index					
Standard Score (%ile)	113 (81)	95 (37)	62 (1)	99 (47)	60 (0.4)
Interpretation	WNL	WNL	sev.	WNL	sev.
Peabody Picture Vocabulary Test 4					
Standard Score (%ile)	129 (97)	103 (58)	84 (27)	101 (53)	76 (5)
Interpretation	>NL	WNL	WNL	WNL	mod.
Goldman Fristoe Test of Articulation					
Standard Score (%ile)	92 (17)	99 (27)	48 (<1)	59 (<1)	<40 (<1)
Interpretation	WNL	WNL.	sev.	sev.	sev.
Test of Auditory Perception – Third Edition, Word discrimination subtest					
Scaled Score (%ile)	11 (63)	16 (7)	7 (16)	9 (37)	–
Interpretation	WNL	WNL	WNL	WNL	–
Inconsistency Assessment (> 30 = inconsistent)					
% Inconsistency	60	40	76	32	40
Interpretation	Inconsist.	Inconsist.	Inconsist.	Inconsist.	Inconsist.
Test of Polysyllables					
% Consonants Correct	80	94	25	93	54
% Vowels Correct	79	91	62	94	79
% Phonemes Correct	80	93	41	94	64
% Stress Pattern Errors	50	19	71	19	40
No. Syllable Segregations[a]	24	13	14	13	29
Oral and Motor Speech Protocol					
Structure					
Raw Score	22	22	24	34	35
Interpretation	WNL	^	WNL	^	^
Function					
Raw Score	96	102	84	89	68
Interpretation	<NL	^	<NL	^	^
Other observations	Intermittent hypernasality			Intermittent hypernasality. Limited range of intonation	Intermittent hypernasality. Loud speaking volume

WNL: within normal limits; NL: normal limits; mod.: moderate; sev.: severe; –: missing data.
[a]Perceptually identified absence of smooth joining of the syllables within a word. ^= outside of age range for normative scores.

the first week and more home-based sessions in the final week of therapy (see Figure 1).

Treatment

The Rapid Syllable Transitions Treatment (ReST) was used (Murray et al., 2015), with modifications for parent training and delivery as detailed herein. Each session began with ∼10 minutes of pre-practice to explain the task to the child and for the child to have supported production of the targets. Following pre-practice, the participants moved to the practice phase, each completing 100 trials: five trials of each of the 20 treated items, in random order. Knowledge of results (KR, i.e. right/wrong) feedback was provided on ∼50% of the items, after a delay of 3–5 seconds. After every 20 trial items, a 2-minute rest break was provided.

Once a participant achieved ≥80% correct on two consecutive practice sessions, the client began treatment on the next, more complex, treatment level. Eric and Matt both met the progression criterion; Eric moved to the production of carrier phrases with a single pseudo word (e.g. "I found the /bəfɔkə/") in session 7, and Matt moved to the production of carrier phrases with two pseudo words (e.g. "She held a /kɔbədə/ not a /fidəkə/") in session 9. If a participant achieved ≤5% correct in the practice phase of two consecutive sessions, the client began treatment at a less complex level. Ben met this criterion in session 2 and from session 3 his treatment goal moved to 2-syllable pseudo words.

Parent training. Each parent was provided with a parent treatment manual in the child's first treatment session. The manual included information about ReST treatment, what constitutes a correct response, cueing techniques for the pre-practice phase, a session flow chart, and details of the type, timing, and frequency of feedback for each phase of the treatment session.

In the first treatment session, the clinician demonstrated ReST treatment with the child while the parent observed. During the second treatment session, the clinician trained the parent to make perceptual judgements about the accuracy of the child's speech. Parents were instructed to attend to the accuracy of the sounds and the prosody. Within prosody, they were asked to attend to both lexical stress (termed "beats") and the smooth connection between the syllables (termed "smoothness"). The parents were instructed to judge whether a word was correct in its entirety (i.e. sounds, beats, and smoothness). If any aspect of the word was incorrect, the parents were to

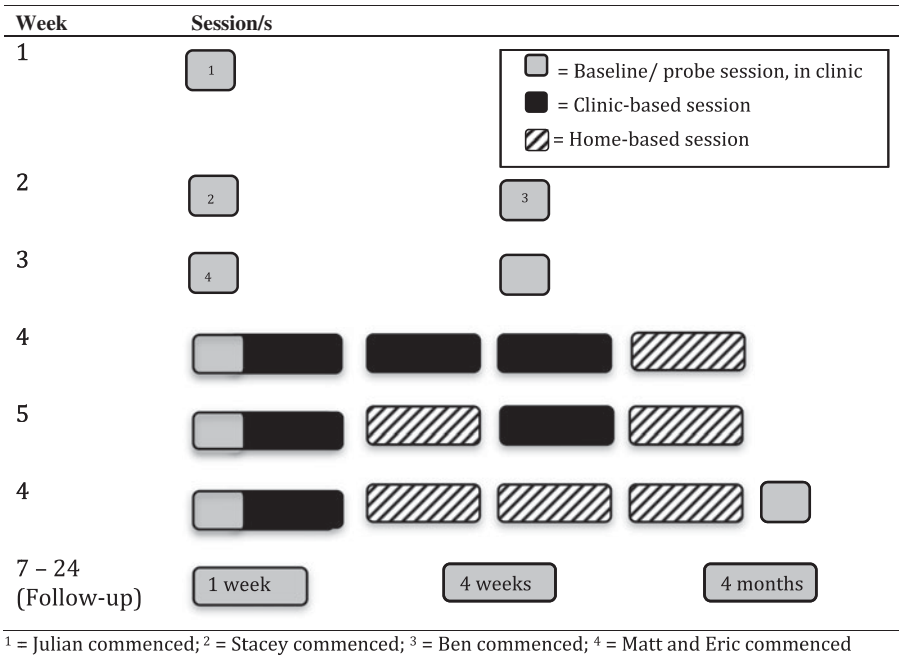

Figure 1. Schedule of baseline, experimental probe and treatment sessions for the five participants.

score a production as incorrect. After this explanation, the parent and the clinician individually made judgements about the child's speech production during the practice. During the scheduled breaks between blocks of production of pseudo words, the clinician calculated the point-by-point agreement between the parent's judgements and their own, discussed any differences, and provided strategies to improve the parent's perceptual judgements. If the parent demonstrated 80% agreement with the clinician across 40 production trials (2 blocks), the parent gave feedback to the child for the next 20 practice items (1 block), while the clinician continued to compare perceptual judgements with the parent in the rest breaks. Parents were provided with data sheets indicating with an asterisk which trials required feedback. Provided that 80% agreement was maintained, the parent and the clinician alternated delivery of feedback to the child every 20 practice items, during sessions two and three.

During the third session, the clinician trained the parent to conduct the pre-practice ("training") phase of treatment. The clinician discussed and modelled strategies to help the child approximate more correct productions. Parents were encouraged to conduct some pre-practice trials and the clinician provided feedback and encouragement. Parents conducted the fourth treatment session with their child at home. In order to maintain consistency between participants regarding dose and scheduling of clinician versus parent-delivered training, all parents provided a parent-delivered home-based treatment session following three clinician-delivered clinic-based sessions, even if they had not demonstrated 80% agreement with the clinician for perceptual judgements.

From therapy session five, each clinic-based session began with a discussion about the home-based therapy followed by a review of the audio recording of a section of home-based pre-practice and practice phases. The clinician and parent discussed the recording, and the clinician provided feedback for the parent. The clinician conducted approximately two-thirds of each remaining clinic-based sessions, and the parent conducted one-third. The clinician provided ongoing support, modelling and training for the parent during the clinic-based sessions.

Equipment

All home-based sessions were audio-recorded with an Olympus VN-711PC digital voice recorder with the child wearing an unbranded lapel microphone. Clinic-based sessions were video recorded using a Bosch NBN – 832V-P camera, and an Electrovoice RE90HW microphone connected to a Bosch DIVAR IP 7000 2U DVD. The initial assessments were audio-recorded with an AKG C520 headset microphone at 5cm mouth-to-microphone distance and Roland Quad Capture UA-55, and video recorded using the Bosch system listed above.

Dependent measures and data analysis

Probe items were phonemically transcribed, and the phonemes, lexical stress and fluency of syllable transitions scored as correct or incorrect. As in the treatment, a response was only scored "overall correct" when all aspects (i.e. phonemes, lexical

stress and fluency of syllable transitions) were judged as accurately matching the model. During the clinic-based treatment sessions, the treating clinician phonemically transcribed and scored each response as for probe sessions. The child received feedback in their treatment sessions based on the perceptual judgements of the person conducting the trial. In the first clinic-based session, this was exclusively the clinician, in all home-based sessions this was the parent, and in the remaining sessions it was approximately two-thirds the clinician and one-third the parent.

Data for each participant were graphed for visual analysis of the trend, variability, level, overlap, and immediacy of effect. Where possible, visual analyses were supported with a version of analysis of variance (ANOVA), Helmert planned orthogonal contrasts. We tested the assumptions of ANOVA, specifically independence of data and equal variance across phases. Where variances were unequal, the ANOVA was calculated not assuming equal variances. Where the assumption of independence of data was violated only visual analyses were performed. When ANOVA was appropriate, Helmert planned orthogonal contrasts were performed for each participant, testing differences across phases (baseline, treatment, follow-up) within behaviours. The Helmert contrast compared (a) average performance in the baseline phase versus pooled treatment and follow-up phases and (b) average performance in the treatment phase versus follow-up phase. Significance at both 0.05 and 0.01 levels are indicated in Table II.

Effect size. In order to develop population-specific estimations of magnitude for ES (Beeson & Robey, 2006), we calculated the average effect size and quartiles across the 18 participants in the four single-case ReST studies using similar dependent measures (current study; McCabe, Macdonald-D'Silva, van Rees, Ballard, & Arciuli, 2014; Thomas et al., 2014; Thomas et al., 2016). We calculated effect sizes (ES) per treated behaviour per child using the standard mean difference procedure (Busk & Serlin, 1992). We subtracted the mean performance in the baseline phase from the mean performance in the follow-up phase and divided this by the standard deviation (SD) in the baseline phase. In order to manage the potential 0% variance in the baseline phase, we pooled the SD for treated and similar untreated pseudo words across participants as recommended by Glass (1977) and shown below.

d_2 = (mean score follow-up phase – mean score baseline phase)/pooled SD across participants in the baseline phase.

The pooled SD across participants in all four studies was 0.061 (see Supplementary material B for ESs of individual published ReST studies using this method). An approximation of ES required for small-, medium-, and large magnitudes of change across the four studies is 3.99, 5.46 and 8.18, respectively. These values are used to interpret effect sizes in the current study. As expected, these figures are substantially higher than estimations for group studies (Cohen, 1988). However they are similar to the benchmarks obtained for single-case experimental studies of treatment for childhood phonological impairments (ES of 1.40, 3.61 and 10.12; Gierut, Morrisette, & Dickinson, 2015) and for adult apraxia of speech (ES of 5.9, 7.12 and 10.19; Bailey, Eatchel, & Wambaugh, 2015).

Reliability

Twenty percent of responses from each probe and each clinic-based treatment session was analysed for intra- and inter-rater (i.e. clinician: first author) reliability of phonemic transcription and scoring of articulation accuracy, stress pattern and fluency. Intra-rater and inter-rater point-to-point agreement for phonemic transcription of experimental probe responses was 97% (SD = 3.3%) and 91% (SD = 5.3%), respectively, and 96% (SD= 3%) and 89% (SD = 4.6%), respectively, for average judgments of phoneme accuracy, lexical stress and syllable fluency.

The reliability of the parents' judgements during the home-based sessions was calculated by comparing 100% of the parent's correct/incorrect judgements with the treating clinician's "overall correct" judgement. The parent scored the child's productions online, and the treating clinician transcribed and scored from the audio recording. Inter clinician reliability was also calculated, by comparing the treating clinician's transcription and scoring with that of the first author for each home-based session. The point-by-point agreement on "overall correct" was 89% (SD = 6.5) between the first author and the treating clinician and 76% (SD = 9.1) between the parent and the treating clinician.

Result

Results for all children are shown in Figures 2–6 and the results of all significance testing can be found in Table II.

Treatment efficacy

Acquisition of treated items. Four children showed a treatment effect on one or more of their treated behaviours. Stacey showed a treatment effect on her treated 3-syllable pseudo words (see Figure 2). Eric showed a treatment effect for 3-syllable pseudo words and also generalisation of the treatment effect to the to-be-treated carrier phrase level (see Figure 3). Eric achieved 20–30% accuracy with these phrases in baseline probes 1–3; his accuracy rose to 50% after initiation of practice on the

Table II. Planned contrasts, and effect sizes for baseline (BL) vs. follow-up phases.

Child	Probe stimuli	Effect size d_2 =	BL v (Tx and Follow-up) t =	BL v (Tx and Follow-up) p =	Tx v Follow up t =	Tx v Follow up p =	Change
Stacey	Treated 3 Syll. pseudo words	6.40	11.301	<0.001**	3.713	0.08	–
	Untreated 3 Syll. pseudo words		3.178	0.016*	1.716	0.13	–
	Untreated 2 Syll. pseudo words		Visual analysis only				
	Untreated 4 Syll. pseudo words		3.896	0.006**	4.928	0.002**	↑
	Phrases: 3 Syll. pseudo words (× 1)		3.669	0.008**	2.467	0.043*	↑
	Untreated 2 Syll. real words		Visual analysis only				
	Untreated 3 Syll. real words		2.535	0.039*	2.138	0.07	–
	Untreated 4 Syll. real words		4.430^	0.034*	2.541	0.126	–
Eric	Treated 3 Syll. pseudo words	7.98					
	Treated phrases: 3 Syll. pseudo word (× 1)	6.05					
	Untreated 3 Syll. pseudo words		Visual analysis only				
	Untreated phrases: 3 Syll. pseudo word (× 1)						
	Untreated 2 Syll. pseudo words		7.348	<0.001**	4.243	0.005**	↑
	Untreated 4 Syll. pseudo words		3.457	0.014*	0.272	0.795	–
	Untreated 2 Syll. real words		1.136	0.88	0.236	0.822	–
	Untreated 3 Syll. real words		Visual analysis only				
	Untreated 4 Syll. real words		2.63	0.037*	1.852	0.114	–
Ben	Treated 2 Syll. pseudo words	8.52	Visual analysis only				
	Treated 3 Syll. pseudo words	2.76	1.395^	0.295	0.930	0.448	–
	Untreated 3 Syll. pseudo words		3.565^	0.33	0.324	0.765**	–
	Phrases: 3 Syll. pseudo words (× 1)		Visual analysis only				
	Untreated 4 Syll. pseudo words		0.953	0.368	1.414	0.195	–
	Untreated 2 Syll. real words		−0.136	0.896	0.236	0.822	–
	Untreated 3 Syll. real words		Visual analysis only				
	Untreated 4 Syll. real words		2.181^	0.12	0.530	0.646	–
Matt	Treated phrases: 3 Syll. pseudo word (× 1)	4.4	2.631	0.046*			
	Treated phrases: 3 Syll. pseudo word (× 2)	2.75	1.94	0.11			
	Untreated phrases: 3 Syll. pseudo word (× 1)		1.94	0.11			
	Untreated phrases: 3 Syll. pseudo word (× 2)		0.337	0.75			
	Untreated 2 Syll. pseudo words		0.473	0.656	1.372	0.228	–
	Untreated 3 Syll. pseudo words		Visual analysis only				
	Untreated 4 Syll. pseudo words		3.078	0.028*	0.888	0.415	–
	Untreated 2 Syll. real words						
	Untreated 3 Syll. real words		Visual analysis only				
	Untreated 4 Syll. real words		2.344	0.066	0.000	1.000	–
Julian	Treated 2 Syll. pseudo words	2.48	3.007	0.250	0.850	0.417	–
	Untreated 2 Syll. pseudo words		0.598^	0.576	0.128	0.904	–
	Untreated 2 Syll. pseudo words (same cons.)		0.033	0.055	0.005	0.389	–
	Untreated 3 Syll. pseudo words		Visual analysis only				
	Untreated 1 Syll. real words		1.958^	0.107	1.00	0.423	–
	Untreated 2 Syll. real words		0.543	0.600	1.152	0.279	–
	Untreated 3 Syll. real words		1.258^	0.253	0.378	0.742	–

Effect size 3.99 = small; 5.5 = medium; 8.18 = large; BL: baseline phase; Tx: treatment phase; **Significant at 0.01; *Significant at 0.05; – = no significant change from end of treatment to follow-up; ↑ Increase from end of treatment to follow-up; ^ = contrast tests did not assume equal variance due to significantly different variances across phases; shading = contrasts were not performed due to violation of independence of data; hash = contrasts were not performed due to insufficient data points within the treatment phase, due to the audio data from Matt's probe session 4 not being on the recording.

isolated words, indicating generalisation across level (see Figure 3, panel A). No clear improvement was noted with direct treatment on the phrases. Ben commenced treatment on 3-syllable pseudo words, but was downgraded to 2-syllable stimuli when he achieved <2% correct in the first two sessions. He showed a clear treatment effect for 2-syllable pseudo words (see Figure 4). Matt began treatment on phrases with one 3-syllable pseudo word. He achieved >80% correct on these phrases in two consecutive treatment sessions in session 8, and from session 9, began treatment on phrases with two pseudo words. He showed a significant treatment effect for carrier phrases containing one pseudo word, but not for carrier phrases with two pseudo words (see Figure 5). Julian was treated on 2-syllable pseudo words. Although he showed some improvement during the sessions, there was no significant treatment effect in the probes (see Figure 6).

The average effect size for treated behaviours in this present study was 5.55 (SD = 2.41, range = 2.48–8.52) (see Table II). The previously published single-case clinician-delivered ReST treatment studies reporting similar dependent measures (McCabe et al., 2014; Thomas et al., 2014; Thomas et al., 2016) had an average ES of 6.46 (SD = 2.70, range 3.3–11.21; see Supplementary material B). The behaviours demonstrating a treatment effect in the present study had ES ≥1SD above the average ES in previous studies.

Generalisation to untreated items. Although four children showed some degree of generalisation to

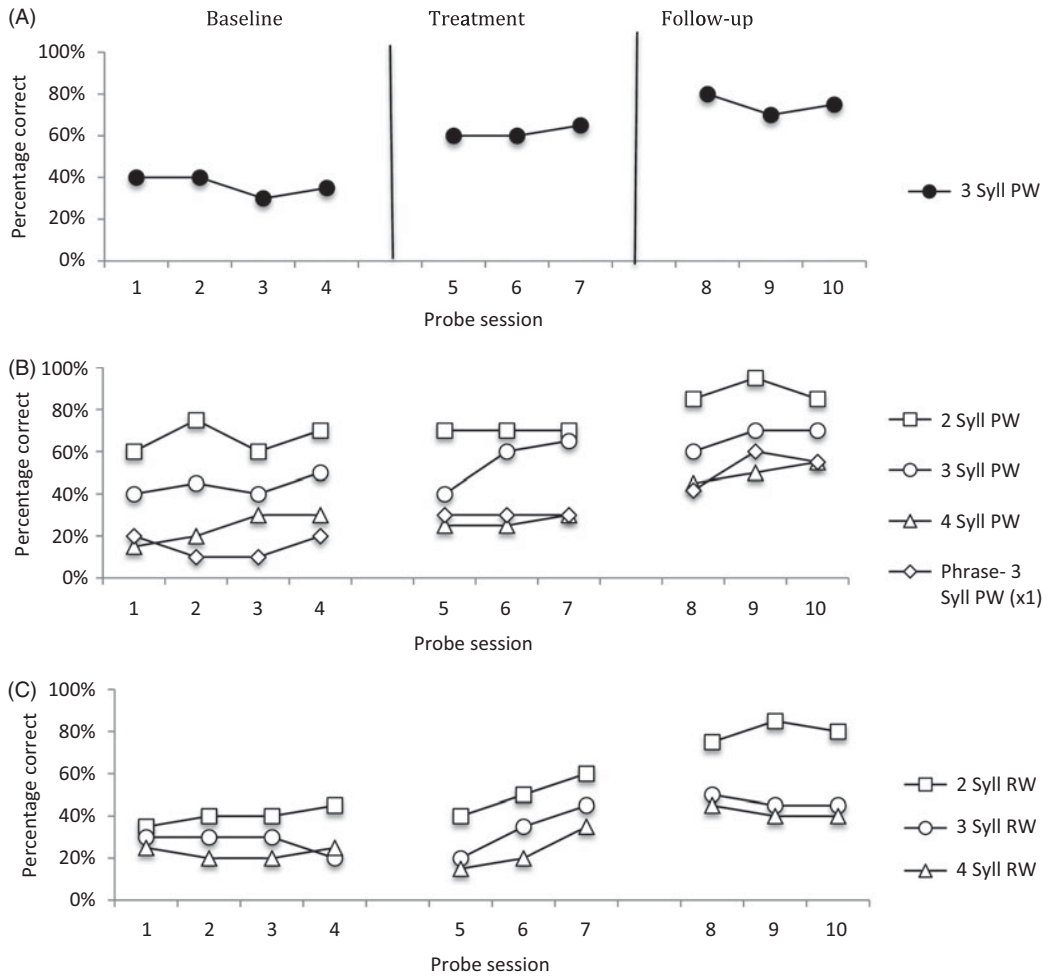

Figure 2. Percent correct for Stacey on treated and untreated items in baseline, treatment and follow-up phases of the study. Panel A shows performance for the treated 3-syllable pseudo words (PW), panel B shows untreated pseudo words as well as 3-syllable pseudo words in carrier phrases, and panel C shows untreated real words (RW).

untreated behaviours, only Stacey and Eric generalised to the majority of untreated pseudo and real words. Stacey had significant generalisation to similar-but-untreated 3-syllable pseudo words, as well as untreated 2- and 4-syllable pseudo words, and phrases containing one 3-syllable pseudo word. She also improved her production of untreated real words with 2-, 3- and 4-syllables. Eric showed a generalisation effect for both of his similar-but-untreated behaviours – 3-syllable pseudo words and phrases containing one 3-syllable pseudo word. Eric demonstrated significant generalisation of the treatment effect to untreated 2- and 4-syllable pseudo words and untreated 4-syllable real words, but not to untreated 2- or 3- syllable real words. Ben showed significant generalisation to only one untreated behaviour – 3-syllable real words. He did not show generalisation to 2-syllable real words. Due to the initial intention to treat 3-syllable pseudo words, Ben's probe list did not include similar-but-untreated 2-syllable pseudo words, 2-syllable pseudo words in carrier phrases, or less-complex pseudo words and real words. Matt only showed generalisation to one untreated behaviour: 4-syllable pseudo words. Julian had no generalisation effect to any of the untreated behaviours.

Maintenance of treatment and generalisation effects. All four children who demonstrated a treatment effect maintained these gains to four months' post-treatment. Stacey had higher accuracy at all follow-up points than baseline levels for her treated behaviour. Eric maintained his treatment gains for 3-syllable pseudo words. He had higher accuracy at two of the three follow-up points than baseline levels for phrases with one pseudo word, and at all follow-up points his accuracy was above the level obtained prior to the commencement of any treatment. Ben had higher accuracy in the follow-up period than all baseline points for 2-syllable pseudo words. Matt maintained his treatment gains for phrases with one pseudo word.

As for generalisation effects, all gains that children had made at the end of treatment were maintained to four months' post-treatment. Additionally, Stacey demonstrated ongoing improvement with her generalisation to 2- and 4-syllable pseudo words, phrases with a pseudo word, and 2-syllable real words. Eric also demonstrated significant ongoing improvement

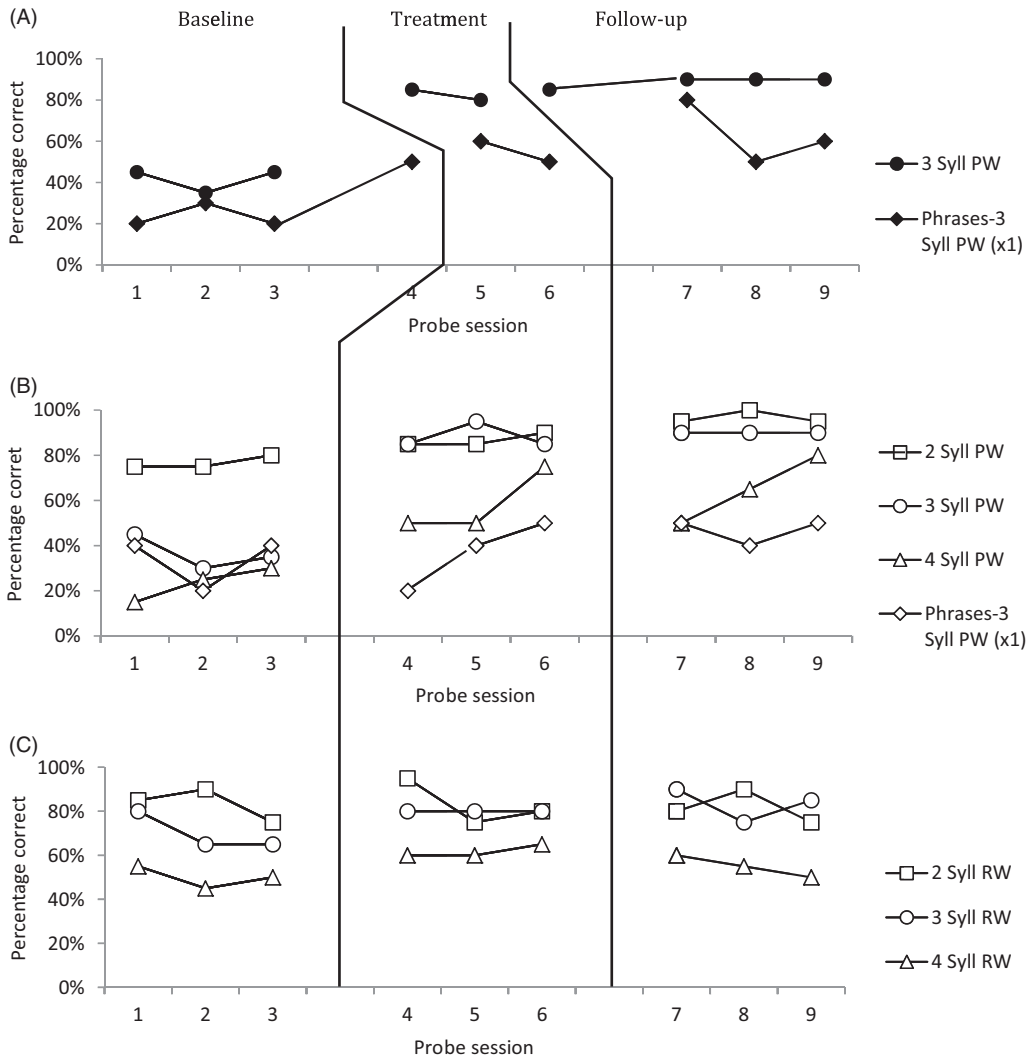

Figure 3. Percent correct for Eric on treated and untreated items in baseline, treatment and follow-up phases of the study. Panel A shows performance for the treated 3-syllable pseudo words (PW), and phrases with one 3-syllable pseudo word, panel B shows untreated syllable pseudo words as well as carrier phrases with a single untreated 3-syllable pseudo word, and panel C shows untreated real words (RW).

within the follow-up phase for untreated 2-syllable pseudo words.

Relationship between effect size and child variables

Scatterplots were created to compare effect size with age, receptive language index, expressive language index, PCC, and Peabody Picture Vocabulary Test (PPVT) score. The highest correlations were between ES and parental perceptual judgements ($r^2 = 0.29$), and ES and age ($r^2 = 0.31$) indicating a small positive correlation between ES and parental perceptual judgements, and a small negative correlation between ES and age. Neither of these correlations were statistically significant. Furthermore, no other correlations were evident, indicating no relationship between effect size and receptive or expressive language ability, or initial speech severity.

Treatment fidelity

The first author calculated treatment fidelity on a randomly selected 10 minutes of the practice phase of each clinician-delivered session, and 100% of each parent-delivered home-based session. Items included in fidelity analysis were: adult model of the stimulus items had correct sounds and prosody, feedback was accurate, KR-style feedback, feedback provided on the trials scheduled for feedback, feedback followed a 3–7 second delay. Matt's parent did not record the final two sessions, despite conducting the sessions and completing the paper record form. All fidelity calculations for the final two sessions used the remaining four parents only.

Average fidelity was 93% (SD = 5.6%) for clinician-delivered practice sessions and 77% (SD = 26.7%) for parent–delivered practice sessions. The fidelity of the parent-delivered sessions varied over the course of the treatment, across parents and across aspects of treatment. The parents improved their fidelity across the sessions, from an average fidelity of 59% (SD = 27, range 11–81) in the first home-session to an average fidelity of 93% (SD = 3, range 89–97) in the final two home-sessions. With

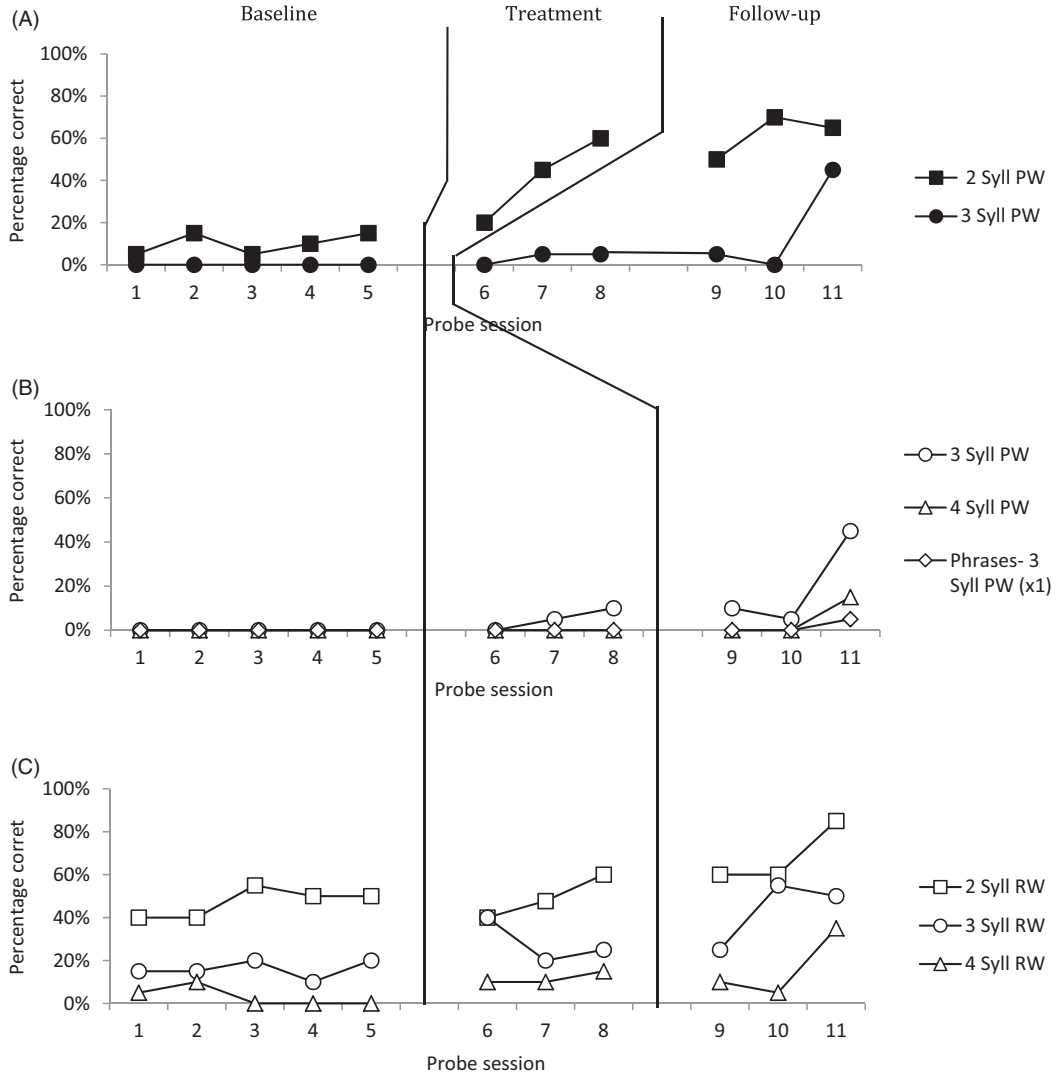

Figure 4. Percent correct for Ben on treated and untreated items in baseline, treatment and follow-up phases of the study. Panel A shows performance for the treated 2 syllable pseudo words (PW) and 3-syllable pseudo words, panel B shows untreated pseudo words as well as single 3-syllable pseud words in carrier phrases, and panel C shows untreated real words (RW).

regard to aspects of treatment, the parents had highest fidelity for providing Knowledge of Results (KR) feedback (90%, SD = 11, range 75–100), providing a correct model (88%, SD = 13, range 68–100), and providing feedback on only the trials scheduled for feedback (88%, SD = 12, range 69–97). The parents had the lowest fidelity for providing feedback within 3–7 seconds (66%, SD = 33, range = 21–91) and giving accurate feedback about whether the child's production was correct (78%, SD = 3, range 75–81). The parents improved with their provision of delayed feedback, using the required delay over 85% of the time by home-session 4. They did not however, improve with their provision of accurate feedback, a skill that depends on the ability to make reliable perceptual judgements. One parent, Chris, had lower fidelity scores than the other parents (average fidelity rating = 41%). Chris did not provide any feedback at all during the first two sessions, infrequently used the sound file of the stimulus items, gave feedback without the required delay, and did not record the final two home sessions.

Discussion

This study aimed to evaluate the outcomes and treatment fidelity of combined clinician-parent delivered ReST treatment for children with CAS. We hypothesised that: (a) the combined model of clinician-parent delivered treatment would be effective for acquiring treated pseudo words and generalising to untreated items, with maintenance of gains for 4 months' post-treatment, (b) parent treatment fidelity would be above 85%, and (c) the effect size would be within one standard deviation of that obtained in clinician-delivered treatment. Our first hypothesis was partially confirmed as not all of the children demonstrated change in their treated behaviours and generalisation items, even

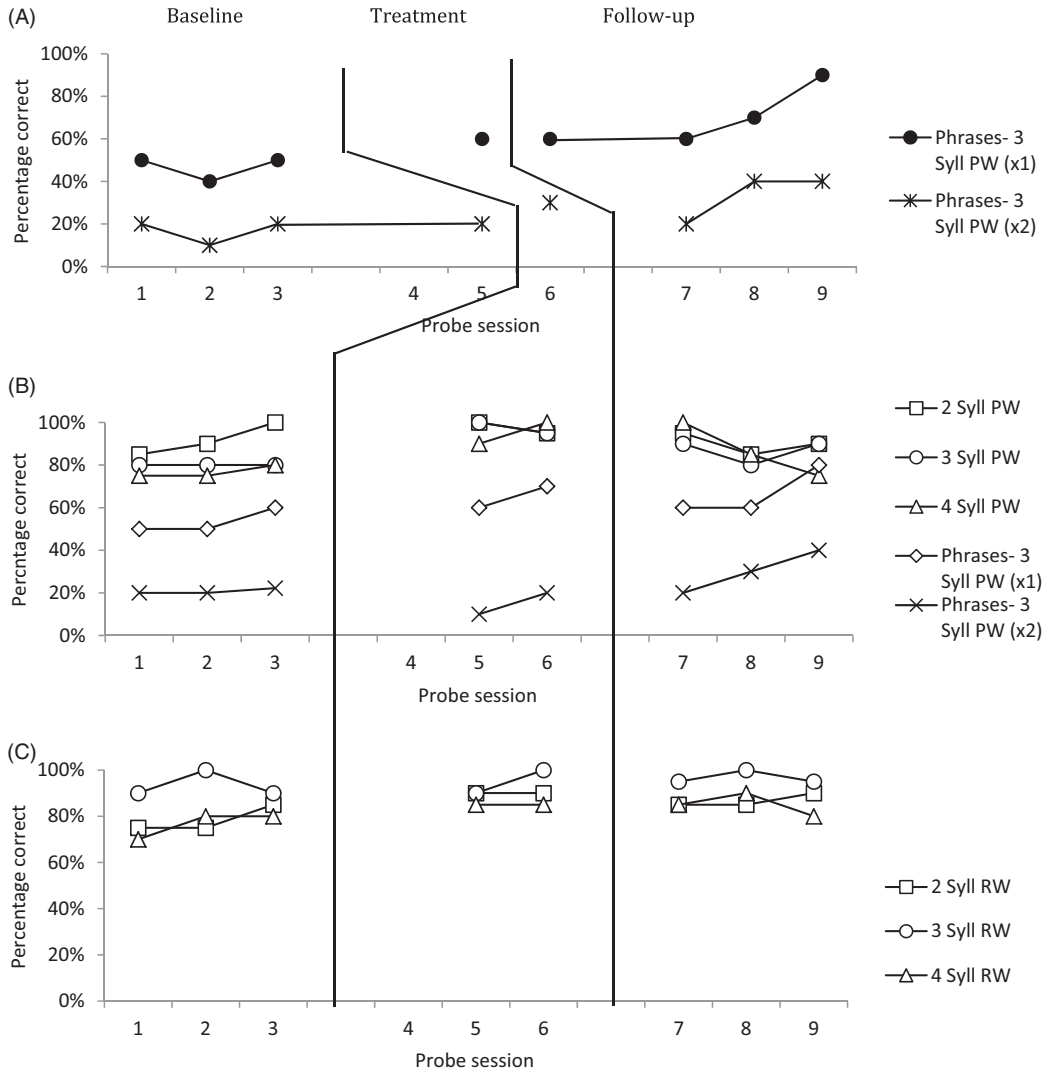

Figure 5. Percent correct for Matt on treated and untreated items in baseline, treatment and follow-up phases of the study. Panel A shows performance for the treated carrier phrases with one 3-syllable pseudo word and two 3-syllable pseudo words (PW), panel B shows untreated pseudo words as well as carrier phrases containing a single untreated 3-syllable pseudo word and two untreated 3-syllable pseudo words, and panel C shows untreated real words (RW). There are no data for Matt's probe 4 because the audio data from that session were not on the recording.

though they maintained all gains to 4 months' post-treatment. Our second hypothesis was not confirmed; parents had variable treatment fidelity levels and the average treatment fidelity was below 85%. Our final hypothesis was confirmed; all behaviours demonstrating a treatment effect had a similar effect size to previously reported clinician delivered treatment.

Combined clinician-parent delivered ReST treatment was efficacious for fewer children than treatment delivered only by clinicians. Previous single-case experimental studies of ReST using only clinician-delivery have reported significant improvement for all participants for all treated behaviours (Ballard, Robin, McCabe, & McDonald, 2010; McCabe et al., 2014; Thomas et al., 2014; Thomas et al., 2016). Despite the combined model of treatment being efficacious for fewer children than the same number of clinician-delivered sessions, in cases where the treatment was effective, the effect size was as large as obtained in clinician-delivered treatment.

Not only did the combined model produce a treatment effect for fewer children than clinician-delivered treatment, it also resulted in more limited generalisation. In comparison to clinician-delivered ReST treatment, fewer children generalised to untreated pseudo words or untreated real words, at any level of complexity. Generalisation to untreated exemplars of the treated behaviour is common in many speech disorder treatments, including ReST (Ballard et al., 2010; McCabe et al., 2014; Murray et al., 2015; Thomas et al., 2016). Matt's lack of generalisation to untrained exemplars of the treated behaviour is difficult to explain. It may be that, despite being statistically significant, Matt's 10% improvement on his treated behaviour (phrases with a single pseudo word) was insufficient to generate system-wide change necessary for production of novel items at the same level. Ben did not generalise

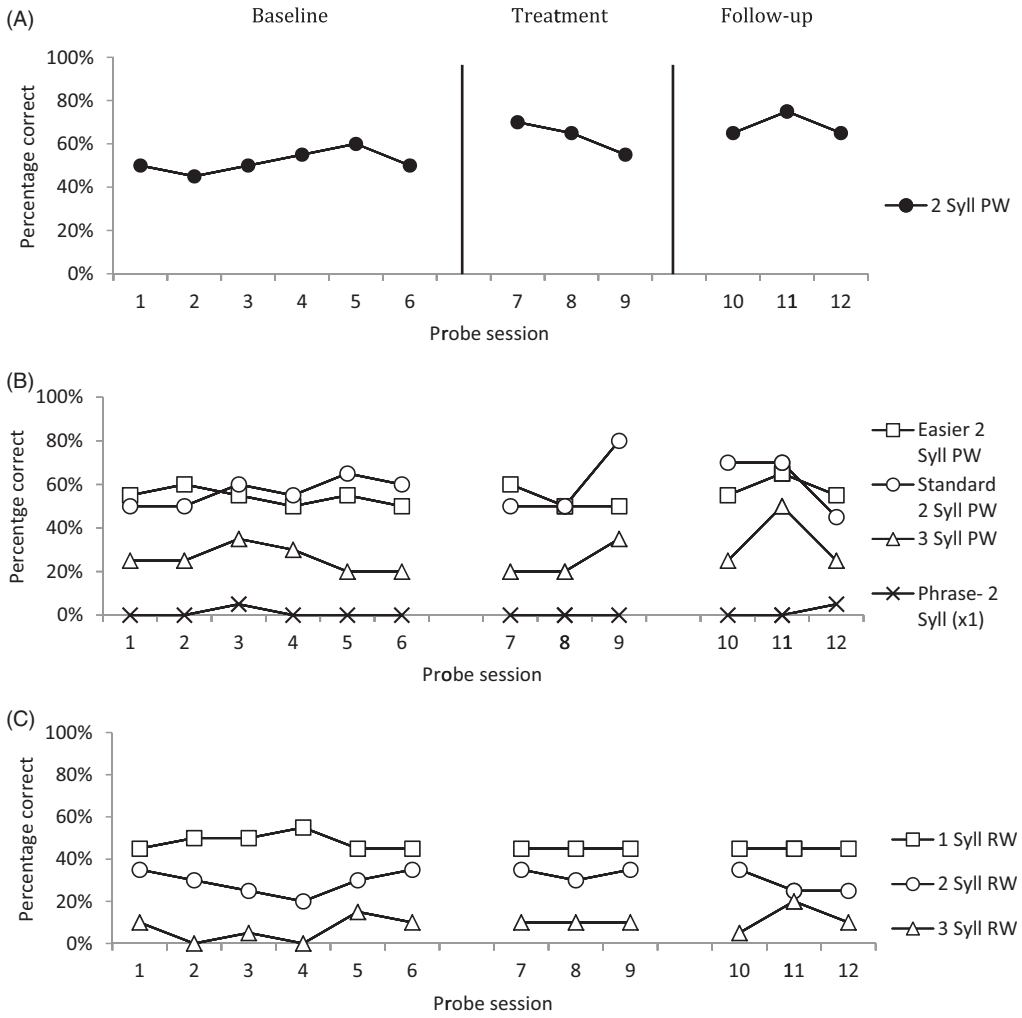

Figure 6. Percent correct for Julian on treated and untreated items in baseline, treatment and follow-up phases of the study. Panel A shows performance for the treated 2-syllable pseudo words (PW), panel B shows untreated less-complex pseudo words with the same consonant repeated (i.e. $C_1V_1C_1V_2$) as well as untreated 2-syllable and 3-syllable pseudo words, and phrases with 2-syllable pseudo words, and panel C shows untreated real words (RW).

to real words at the same level as treatment (two syllables). The real words probed here had more variability than the treated pseudo words; some had differing syllable structures, some had sound classes not treated, while others had more phonemes. It is important for a non-word treatment such as ReST to probe real word productions in order to measure ecological validity. Despite the greater variability of real words compared to the trained items, generalisation to real words is frequently reported following clinician-delivered ReST treatment (Murray et al., 2015; Thomas et al., 2014; Thomas et al., 2016), presumably due to generalised improvement in planning and programming speech motor movements with resultant improvement in prosody and the targeted speech classes (i.e. voiced and voiceless plosives and fricatives).

Generalisation effects are thought to be related to stimulus characteristics, principles of motor learning, and patient characteristics (Ballard, 2001). There was no change in stimulus characteristics between this study and previously reported clinician-delivered ReST treatment, and the same principles of motor learning were employed (i.e. random presentation of items, high number of trials with low frequency, delayed knowledge-of-results feedback). The fidelity of the parent-delivered sessions was lower than typically achieved in clinician-delivered treatment, indicating that some of the principles of motor learning were not applied as faithfully as the manualised treatment and this may have had some effect on generalisation. Additionally, the combined clinician-parent delivery model had less variability in practice conditions than clinician-only treatment. This occurred because sound files were used to present the stimulus item during parent-delivered practice trials. This meant there was no variation in dynamic level or prosodic contour between models used during the parent-delivered sessions, unlike the live models used in clinician-delivered treatment. The lack of variability in the auditory models or lack of visual modelling may have contributed to the lack of generalisation (Maas et al., 2008).

The efficacy of combined clinician-parent delivered treatment appears to be influenced by multiple factors, including treatment fidelity, inter-judge reliability of perceptual judgements, child factors, and parent factors. A breakdown of these factors can be found in Supplementary Material C. Although there were small correlations between treatment outcome and reliability of parental perceptual judgements and younger age of the child, these were not statistically significant. Although no overall correlations were shown between ES and either PPVT-IV results (Dunn & Dunn, 2007) or language ability, it is interesting to note that the child the poorest response to treatment (Julian) had below average PPVT-IV score, and the two children who had incomplete response to treatment had both receptive and expressive language delay. PPVT-IV score has been shown to correlate with intelligence measures (e.g. Vance, West, & Kutsick, 1989), and it would not be unexpected for general cognitive ability to affect treatment outcome. Given that children with language impairments have previously shown significant treatment and generalisation effects following ReST treatment (Thomas et al., 2014; Thomas et al., 2016) it may be that there is an interaction between language skills and service delivery mode with respect to treatment outcome. Further larger-scale studies would be required to further investigate the relationship between these variables and treatment outcome.

For comparison purposes, we maintained the treatment dose at 12 hours as reported in all other ReST treatment studies (6 clinic sessions, 6 home sessions). This meant that the parent training was provided within the 6 clinic visits. Six clinic sessions is not uncommon in public funded services (Glogowska, Roulstone, Enderby, & Peters, 2000; Ruggero et al., 2012). However, when parents are trained to deliver home-based treatment they are typically given more hours of training (e.g. Carr Swift et al., 2011; Sugden et al., 2016).

We found that although the average parental treatment fidelity was below our benchmark of 85%, there was variability across parents. This variability in fidelity across different parents, has been shown in other parent-delivered treatments (Carr Swift et al., 2011). Three parents had treatment fidelity levels above the 85% benchmark, one was just below, and the other parent, Chris, achieved only 41% treatment fidelity. Our pre-treatment assessment battery did not identify any differences between Chris and the other parents that would foreshadow the difficulties Chris had conducting the sessions. Chris held a Bachelor degree and worked in a professional role. Although Chris mentioned having trouble reading b's and d's throughout life, Chris had not received any formal assessment, treatment or diagnoses of speech, language or reading abilities. Two other parents, Andy and Morgan, also had a history of undiagnosed and untreated literacy difficulties, but had higher levels of treatment fidelity indicating that literacy alone is unlikely to explain Chris's difficulty implementing the treatment. The three parents with a history of literacy difficulty had the lowest inter-rater reliability for perceptual judgements about their child's speech. It is important for clinicians and researchers to consider parental speech, language and literacy skills for children with CAS before replacing clinician delivered treatment with parent delivered treatment.

One of the areas with lowest parental treatment fidelity was the provision of accurate feedback to the child on speech accuracy. Treatment fidelity is the degree to which delivery of a treatment is in accordance with the "gold standard" or manualised treatment (Kaderavek & Justice, 2010). We can divide the elements of fidelity into perceptual and procedural components. The perceptual component of fidelity is evaluated through measurement of reliability of perceptual judgements of the child's speech. Although the parent's verbal feedback to the child matched their recorded perceptual judgement (i.e. correct/incorrect) there was relatively low agreement between the parents' feedback on speech accuracy and the clinician's judgment of accuracy. The reliability of the parent's perceptual judgements had a small positive correlation with ES ($r^2 = 0.29$), indicating the potential relevance of this aspect for treatment outcome. The parents' inter-rater reliability on speech accuracy did not improve over the course of treatment. In contrast, fidelity for *procedural* aspects of the treatment, such as giving feedback after a 3–7 second delay, showed early and rapid improvement, reaching >80% by session 3.

It is not surprising that the parents' perceptual judgement skill did not show the same improvement as their procedural treatment accuracy. The perceptual task was complex, and, unlike the clinicians, the parents had no phonetic training. We had no reason to suspect the parents would be different from the general population with regard to their perceptual abilities, as they passed hearing screening, and had normal non-word repetition skills. Untrained listeners are strongly influenced by prosodic features in speech (Cutler, 2012). However, it is uncommon for people to be asked to judge the prosodic features of lexical stress and syllable segmentation. Not only did we ask parents to judge something that is not commonly judged explicitly; we believe their years of attending previous therapy focussed on sound accuracy rather than prosody may have biased their perception. Anecdotally, parents told us they were unfamiliar with the concept of prosody and were more confident judging speech sounds than prosody. Of the treatments Murray et al. (2014) report as having sufficient evidence to warrant clinical application, few explicitly train prosody, despite prosodic impairment comprising two of the three features in the ASHA consensus criteria (ASHA, 2007). It may be that a perceptual training programme, specifying

minimum inter-rater reliability thresholds is required before a clinician, student or parent is asked to deliver ReST.

Limitations and future directions

This study was a small-scale investigation and findings may not generalise to a larger sample. The study design did not permit evaluation of whether the parent-delivered sessions enhanced the children's speech outcomes. This would require a comparison group who received six clinician-delivered sessions only. Also, the study included a multiple baseline across participants' component but did not include a maturational control.

During the parent-delivered sessions, pre-recorded clinician models of target stimuli were delivered via PowerPoint slideshow. While this was done to ensure fidelity, it may have operated to reduce small variations in stimulus characteristics from trial to trial and diminished generalisation effects.

Across the literature in CAS and other speech and language treatments, there are limited investigations of treatment fidelity in parent-delivered interventions. Modifications to the parent training methodology used here, such as training over a longer period and/or beginning parent training prior to the child's treatment could also be explored. We also recommend including specific teaching about prosody with inter-rater reliability thresholds for perceptual judgements to be achieved prior to commencing treatment. Given that weak syllables are marked in English by both a weak vowel *and* a specific lexical stress pattern (Cutler, 2012) and parents have greater confidence perceiving speech sounds than prosody, it may be preferable to train parents to identify weak vowels rather than lexical stress patterns. Further investigations should investigate the parent factors associated with perceptual reliability and treatment fidelity as well as the child factors associated with treatment outcome and the interaction between these factors.

Conclusion

This study evaluated the outcomes and treatment fidelity of a combined model of clinician-parent delivered ReST treatment, reporting both the efficacy and treatment fidelity for five children with CAS. Not all children showed an improvement in their treated items, and only two children showed generalisation to the majority of the untreated pseudo and real words. Although there is a growing body of literature supporting the use of ReST treatment for CAS, all previous research has been conducted via clinician-delivery of the treatment. The strong treatment and generalisation effects previously shown with clinician-delivered ReST were not evident when the treatment was delivered in a combined model. In general, the fidelity of parent-delivered sessions was lower than the clinician-delivered sessions, with variability within and across parents. Parents did not improve over time with their perceptual judgements about the child's speech. Due to the limited treatment and generalisation effect shown in this study, clinicians need to be cautious about substituting clinician-delivered sessions with parent-delivered sessions until we have a better understanding of the parent and child factors affecting magnitude of treatment response.

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Supplementary material

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References

American Speech-Language-Hearing Association. (2007). *Childhood apraxia of speech* [Technical Report]. 12th of May, 2016. Retrieved from http://www.asha.org/policy/TR2007-00278/

Bailey, D.J., Eatchel, K., & Wambaugh, J. (2015). Sound production treatment: Synthesis and quantification of outcomes. *American Journal of Speech-Language Pathology*, *24*, S798–S814. doi: 10.1044/2015_ajslp-14-0127

Ballard, K.J. (2001). Response generalization in apraxia of speech treatments: Taking another look. *Journal of Communication Disorders*, *34*, 3–20. http://dx.doi.org/10.1016/S0021-9924(00)00038-1

Ballard, K.J., Robin, D.A., McCabe, P., & McDonald, J. (2010). A treatment for dysprosody in childhood apraxia of speech. *Journal of Speech Language & Hearing Research*, *53*, 1227–1245. http://dx.doi.org/10.1044/1092-4388(2010/09-0130)

Beeson, P.M., & Robey, R.R. (2006). Evaluating single-subject treatment research: lessons learned from the aphasia literature. *Neuropsychology Review*, *16*, 161–169. doi: 10.1007/s11065-006-9013-7

Bercow, J. (2008). The Bercow Report: A review of services for children and young people (0–19) with speech, language and communication needs. 12th of May, 2016. Retrieved from http://dera.ioe.ac.uk/8405/7/7771-dcsf-bercow_Redacted.pdf.

Bowen, C. (2010). Parents and children together (PACT) intervention. In Williams A. L., McLoeod, S., & McCauley, R. J (Eds.), *Interventions for Speech Sound Disorders in Children*. (pp. 407–426). Baltimore Maryland, USA: Paul Brookes.

Busk, P.L., & Serlin, R.C. (1992). Meta-analysis for single-case research. In T. R. Kratochwill & J. R. Levin (Eds.), *Single-case research design and analysis: New directions for psychology and education*. Hillsdale, NJ: Lawrence Earlbaum Associates.

Carr Swift, M., O'brian, S., Hewat, S., Onslow, M., Packman, A., & Menzies, R. (2011). Investigating parent delivery of the Lidcombe Program. *International Journal of Speech-Language Pathology*, *13*, 308–316. http://dx.doi.org/10.3109/17549507.2011.550692

Cohen, J. (1988). *Statistical power analysis for the behavioral sciences:* Vol. 2. Hillsdale, NJ: Lawrence Earlbaum Associates.

Cutler, A. (2012). *Native listening: Language experience and the recognition of spoken words*. London, England: The MIT Press.

Dodd, B., Zhu, H., Crosbie, S., Holm, A., & Ozanne, A. (2006). *Diagnostic evaluation of articulation and phonology (DEAP)*. London, England: Psychology Corporation.

Dunn, L.M., & Dunn, D.M. (2007). *Peabody picture vocabulary test, (PPVT-4)*. *Minneapolis*, *MN: Pearson Assessments*.

Gierut, J.A., Morrisette, M.L., & Dickinson, S.L. (2015). Effect size for single-subject design in phonological treatment. *Journal of Speech, Language, and Hearing Research*, *58*, 1464–1481. http://dx.doi.org/10.1044/2015_JSLHR-S-14-0299

Girolametto, L.E. (1988). Improving the social-conversational skills of developmentally delayed children: An intervention study. *Journal of Speech and Hearing Disorders*, *53*, 156–167. http://dx.doi.org/10.1044/jshd.5302.156

Glass, G.V. (1977). Integrating findings: The meta-analysis of research. *Review of Research in Education*, *5*, 351–379. http://dx.doi.org/10.2307/1167179

Glogowska, M., Roulstone, S., Enderby, P., & Peters, T.J. (2000). Randomised controlled trial of community based speech and language therapy in preschool children. *British Medical Journal*, *321*, 923. http://dx.doi.org/10.1136/bmj.321.7266.923

Glogowska, R., & Campbell, M. (2000). Investigating parental views of involvement in pre-school speech and language therapy. *International Journal of Language & Communication Disorders*, *35*, 391–405. doi: 10.1080/136828200410645

Goldman, R., & Fristoe, M. (2000). *Goldman-Fristoe Test of Articulation 2 (GFTA -2)*. Minneapolis, MN: Pearson Assessments.

Gozzard, H., Baker, E., & McCabe, P. (2006). Children's productions of polysyllables. *ACQuiring Knowledge in Speech, Language and Hearing*, *8*, 113–116. doi: 10.1177/0265659008096292

Hall, P.K., Jordan, L.S., & Robin, D.A. (1993). *Developmental apraxia of speech: Theory and clinical practice*. Austin, TX: Pro-ed.

Jones, M., Onslow, M., Packman, A., Williams, S., Ormond, T., Schwarz, I., & Gebski, V. (2005). Randomised controlled trial of the Lidcombe programme of early stuttering intervention. *British Medical Journal*, *331*, 659. http://dx.doi.org/10.1136/bmj.38520.451840.E0

Kaderavek, J.N., & Justice, L.M. (2010). Fidelity: An essential component of evidence-based practice in speech-language pathology. *American Journal of Speech-Language Pathology*, *19*, 369–379. http://dx.doi.org/10.1044/1058-0360(2010/09-0097)

Kazdin, A.E. (2011). *Single-case research designs: Methods for clinical and applied settings*. New York, NY: Oxford University Press.

Lancaster, G., Keusch, S., Levin, A., Pring, T., & Martin, S. (2010). Treating children with phonological problems: Does an eclectic approach to therapy work?. *International Journal of Language & Communication Disorders*, *45*, 174–181. doi: 10.3109/13682820902818888

Lawler, K., Taylor, N.F., & Shields, N. (2013). Outcomes after caregiver-provided speech and language or other allied health therapy: A systematic review. *Archives of Physical Medicine and Rehabilitation*, *94*, 1139–1160. doi: http://dx.doi.org/10.1016/j.apmr.2012.11.022.

Lim, J.M., McCabe, P., & Purcell, A. (in press) Challenges and solutions in speech-language pathology service delivery across Australia and Canada. *European Journal for Person Centered Healthcare*.

Maas, E., Gildersleeve-Neumann, C., Jakielski, K., & Stoeckel, R. (2014). Motor-based intervention protocols in treatment of childhood apraxia of speech (CAS). *Current Developmental Disorders Reports*, *1*, 197–206. doi: 10.1007/s40474-014-0016-4

Maas, E., Robin, D.A., Austermann Hula, S.N., Freedman, S.E., Wulf, G., Ballard, K.J., & Schmidt, R.A. (2008). Principles of motor learning in treatment of motor speech disorders. *American Journal of Speech-Language Pathology*, *17*, 277–298. http://dx.doi.org/10.1044/1058-0360(2008/025)

McCabe, P., Macdonald-D'silva, A.G., van Rees, L.J., Ballard, K.J., & Arciuli, J. (2014). Orthographically sensitive treatment for dysprosody in children with Childhood Apraxia of Speech using ReST intervention. *Developmental Neurorehabilitation*, *17*, 137–145. http://dx.doi.org/10.3109/17518423.2014.906002

McLeod, S., & Baker, E. (2014). Speech-language pathologists' practices regarding assessment, analysis, target selection, intervention, and service delivery for children with speech sound disorders. *Clinical Linguistics & Phonetics*, *28*, 508–531. doi: 10.3109/02699206.2014.926994

Murray, E., McCabe, P., & Ballard, K.J. (2014). A systematic review of treatment outcomes for children with childhood apraxia of speech. *American Journal of Speech-Language Pathology*, *23*, 486–504. doi: 10.1044/2014_AJSLP-13-0035

Murray, E., McCabe, P., & Ballard, K.J. (2015). A randomized control trial for children with Childhood Apraxia of Speech comparing Rapid Syllable Transition treatment and the Nuffield Dyspraxia Programme – third edition. *Journal of Speech, Language and Hearing Research*, *58*, 669–686. doi: 10.1044/2015_JSHLR-S-13-0179

Robbins, J., & Klee, T. (1987). Clinical assessment of oropharyngeal motor development in young children. *Journal of Speech and Hearing Disorders*, *52*, 271–277. http://dx.doi.org/10.1044/jshd.5203.271

Ruggero, L., McCabe, P., Ballard, K.J., & Munro, N. (2012). Paediatric speech-language pathology service delivery: An exploratory survey of Australian parents. *International Journal of Speech-Language Pathology*, *14*, 338–350. http://dx.doi.org/10.3109/17549507.2011.650213

Semel, E., Wiig, E., & Secord, W. (2006). *Clinical evaluation of language fundamentals fourth edition, Australian standardised edition (CELF-4 Australian)*. Sydney, Australia: Pearson Inc.

Skinder-Meredith, A. (2001). Differential diagnosis: Developmental apraxia of speech and phonologic delay. *Augmentative Communication News*, *14*, 5–8.

Strand, E.A., Stoeckel, R., & Baas, B. (2006). Treatment of severe childhood apraxia of speech: A treatment efficacy study. *Journal of Medical Speech-Language Pathology*, *14*, 297–307.

Sugden, E., Baker, E., Munro, N., & Williams, A.L. (2016). Involvement of parents in intervetnion for childhood speech disorders: A review of the evidence. *International Journal of Language & Communication Disorders*, *51*, 597–625. doi: 10.1111/1460-6984.12247.

Thomas, D.C., McCabe, P., & Ballard, K.J. (2014). Rapid syllable transitions (ReST) treatment for childhood apraxia of speech: The effect of lower dose-frequency. *Journal of Communication Disorders*, *51*, 29–42. doi: 10.1016/j.jcomdis.2014.06.004.

Thomas, D.C., McCabe, P., & Ballard, K.J. (2016). Telehealth delivery of rapid syllable transition treatment for childhood apraxia of speech. *International Journal of Language & Communication Disorders*, *51*, 654–671. doi: 10.1111/1460-6984.12238.

Torgesen, J., Wagner, R., & Rashotte, C. (1999). Comprehensive test of phonological processing. Austin, TX: Pro-Ed.

Vance, B., West, R., & Kutsick, K. (1989). Prediction of Wechsler Preschool and Primary Scale of Intelligence IQ scores for preschool children using the Peabody Picture Vocabulary Test-R and the Expressive One Word Picture Vocabulary Test. *Journal of Clinical Psychology*, *45*, 642–644. http://dx.doi.org/10.1002/1097-4679(198907)45:4<642::AID-JCLP2270450421>3.0.CO;2-Q

Wiig, E.H., Secord, W., & Semel, E.M. (2004). *Clinical evaluation of language fundamentals preschool* (2nd ed). Australian standardised edition. Sydney, Australia: Harcourt Assessment.

Chapter 6: Comparing Rapid Syllable Transition Treatment Efficacy Across Service-Delivery Approaches

So far, this thesis has reported the results of individual children receiving 12 sessions of ReST treatment in one of three service-delivery contexts. Chapter 3 reported on twice-weekly clinician-delivered face-to-face delivery; Chapter 4 reported on telehealth delivery four times per week with a clinician; and Chapter 5 reported on combined clinician–parent delivery four times per week, face-to-face. Each child participated in only one of the studies and therefore received only one service-delivery approach.

This chapter has three purposes. Firstly, it will compare the efficacy of the service-delivery approaches, by considering the presence of treatment and generalisation effects for individual children; effect sizes (ESs); and change scores, using both visual and statistical analyses. Secondly, it will explore the factors associated with treatment response across the service-delivery approaches. Thirdly, it will revisit the EBP framework that was presented in Chapter 2 and thus provide context for Chapter 7, which will report on parents' experiences of telehealth-delivered ReST treatment and combined clinician–parent delivered treatment.

6.1 Effects for individual children within each service-delivery approach

6.1.1 Twice-weekly delivery

In the twice-weekly delivery model, all four participants had significant improvement on all treated behaviours. All four participants had significant improvement on untreated real words, and two of the four participants also had significant improvement on untreated pseudo words. All participants maintained their treatment and generalisation gains for 4 months post-treatment for the majority of behaviours. However, there was no evidence of widespread ongoing improvement in the maintenance phase, as reported following four times per week clinician-delivered face-to-face ReST treatment (Murray et al., 2015).

6.1.2 Telehealth delivery

In the telehealth delivery model, all five participants demonstrated significant improvement on their treated behaviours, and significant generalisation to untreated pseudo words and untreated real words. All children maintained the treatment and generalisation gains for the majority of their behaviours. As with the twice-weekly service delivery, there was no widespread demonstration of ongoing improvement in the maintenance phase as demonstrated by Murray et al. (2015).

6.1.3 Combined clinician–parent delivery

The combined clinician–parent delivery model described in Chapter 5 provided a more mixed pattern of treatment and generalisation effects across participants. Of the five participants, two demonstrated significant improvement with all treated behaviours; two significantly improved with one of their two treated behaviours; and one showed no improvement with his treated behaviour.

A similar pattern of mixed response across participants was shown for generalisation. The two children who responded well to treatment generalised to the majority of untreated items. The two children who had partial treatment response generalised to some of the generalisation behaviours, and, as might be expected, the child with no treatment response showed no generalisation.

As for maintenance of treatment effect, all children who made gains maintained their treatment and generalisation gains for 4 months post-treatment. Of the 11 behaviours for which significant treatment or generalisation gains were demonstrated, eight had statistically similar performance in the maintenance phase. The remaining three behaviours had statistically stronger performance in the maintenance phase than in the treatment phase, indicating improved performance once treatment had ceased, which is comparable to improvements in the maintenance phase in Murray et al. (2015), where children received treatment from a clinician four times per week.

6.2 Effect size

An alternative way of measuring improvement in treatment studies is to calculate ES. ESs provide a means of quantifying the improvement (Beeson & Robey, 2006), and as such, are frequently used in single-case investigations (e.g. Gierut, Morrisette, & Dickinson, 2015; Skelton & Hagopian, 2014). ES is computed using the performance of a given individual on a given behaviour pre- and post-treatment. One of the most common means of calculating an ES is to subtract the mean baseline performance from the mean post-treatment performance, and to divide the difference by the standard deviation of the baseline phase (Busk & Serlin, 1992).

One consideration in using ES measures in single-case research is that standard deviations are a summary of variation, and variation may be less if the measures are non-independent. However, there was little evidence of autocorrelation within the data examined in this thesis (see Chapters 3, 4 and 5), and as such, ES calculations are valid in this instance.

The magnitude of ES is typically larger in single-case methodology studies than in group studies because the variance used for the denominator is frequently smaller in single participants than across a group (Beeson & Robey, 2006). As such, it is not appropriate to compare the ES from this study with the ES from the group study—a randomised controlled trial—reported by Murray et al. (2015).

Different strategies for ES calculation are required when an individual has zero variance in the baseline phase. Zero variance in baseline means a standard deviation of zero, and it is not possible to calculate an ES when the denominator of the equation is zero (Beeson & Robey, 2006). Several different strategies have been proposed to estimate the variance for the denominator to avoid this problem. Glass (1977) proposed using an average of the standard deviation across participants with the same impairment, whereas Beeson and Robey (2006) suggested either using the pooled standard deviation of baseline and follow-up phases within behaviour and participant or using the

average baseline standard deviation across behaviours within participant (Beeson & Robey, 2006).

In this chapter, standard deviation was pooled across participants with the same impairment, as advocated by Glass (1977). ES was calculated per participant, per treated behaviour, by subtracting the mean pre-treatment performance from the mean post-treatment performance and dividing the difference by the mean baseline standard deviation on the production of pseudo words across participants in single-case ReST treatment studies with similar dependent measures (McCabe et al., 2014; Thomas, McCabe, & Ballard, 2014; Thomas, McCabe, & Ballard, 2017; Thomas, McCabe, Ballard & Lincoln, 2016). Ballard et al. (2010) was not included as they did not report similar dependent measures. The equation used for ES calculation is as follows:

d_2 = (mean score follow-up phase − mean score baseline phase/pooled *SD* across participants in the baseline phase)

Calculations were made of the ES for the papers presented in this thesis. In cases where a participant had more than one treated behaviour, an ES was calculated for each treated behaviour, and an average ES was calculated across all their treated behaviours. An average ES per service-delivery model (twice-weekly treatment, telehealth mode, combined clinician-parent delivery) was also calculated by using one ES per participant. In cases where participants had more than one treated behaviour, the average ES across behaviours was used. All ESs are reported in Table 6.1.

Table 6.1: Effect sizes for treated behaviour(s) across service-delivery models

Service-delivery method	Participant	Treated behaviour	Effect size
Twice-weekly, face-to-face clinician-delivered	M1	3 syllable pseudo words	8.18
		Phrases with 1x3 syllable pseudo word	4.12
		Average ES for M1	*6.15*[#]
	F1	3 syllable pseudo words	11.21
	M2	3 syllable pseudo words	3.44
	F2	2 syllable pseudo words	4.95
	Mean (*SD*) across participants		**6.44 (3.36)**
Telehealth, four times per week, clinician-delivered	Oliver	3 syllable pseudo words	9.23
	Luke	2 syllable pseudo words	9.82
	Jack	3 syllable pseudo words	3.85
		Phrases with 1x3 syllable pseudo word	7.76
		Average ES for Jack	*5.81*[#]
	Emily	3 syllable pseudo words	4.65
		Phrases with 1x3 syllable pseudo word	5.98
		Average ES for Emily	*5.31*[#]
	Lachlan	3 syllable pseudo words	10.09
	Mean (*SD*) across participants		**8.05 (1.94)**
Combined clinician–parent delivery, four times per week, face-to-face	Stacey	3 syllable pseudo words	6.40
	Eric	3 syllable pseudo words	7.98
		Phrases with 1x3 syllable pseudo word	6.05
		Average ES for Eric	*7.01*[#]
	Ben	3 syllable pseudo words	2.76
		2 syllable pseudo words	8.52
		Average ES for Ben	*5.64*[#]
	Matt	Phrases with 1x3 syllable pseudo word	4.40
		Phrases with 2x3 syllable pseudo words	2.75
		Average ES for Matt	*3.58*[#]
	Julian	2 syllable pw	2.48
	Mean (*SD*) across participants		**5.02 (1.92)**

Note. ES = effect size; # = score is an average of the two treated behaviours, and used in the calculations of mean ES across participants for the service-delivery model.

6.2.1 Effect size across service-delivery models

There was variability of ES for treated behaviours, both within and across service-delivery groups. The ESs for individual participants on individual treated behaviours ranged between 2.48 and 11.21. The average ES for twice-weekly therapy, telehealth and combined clinician–parent delivery was 6.44 (range = 3.44–11.21), 8.05 (range = 3.85–10.09) and 5.02 (range = 2.48–8.52) respectively. See 'Interpreting effect sizes' below for interpretation.

6.2.2 Effect size magnitude calculations

As discussed in Chapter 5, it is possible to estimate ES magnitudes for specific clinical populations (Gierut et al., 2015). Estimations of ES magnitudes enable researchers and clinicans to determine whether the performance of a given individual or group is similar to the performance of other people who have received the treatment. As detailed in Chapter 5, ES magnitude was estimated following 12 sessions of ReST treatment, across the 18 participants in the ReST studies reported in this thesis and other studies with similar dependent measures and methodology (McCabe et al., 2014; Thomas et al., 2016; Thomas, McCabe, & Ballard, 2014, 2017). An approximation of the ES required for small, medium, and large magnitudes of change (Beeson & Robey, 2006) across the four studies was 3.99, 5.46 and 8.18 respectively. As expected, these figures are substantially higher than estimations for group studies using Cohen's *d* (0.20, 0.50 and 0.80; Cohen, 1988). However, they are similar to the benchmarks obtained for single-case experimental studies of treatment for childhood phonological impairments (1.40, 3.61 and 10.12; Gierut et al., 2015) and for acquired apraxia of speech in adults (5.9, 7.12 and 10.19; Bailey, Eatchel, & Wambaugh, 2015).

6.2.3 Interpreting effect sizes

The largest mean ES was obtained from participants receiving telehealth treatment and the lowest from participants receiving combined clinician–parent delivered treatment; however, there was considerable overlap between the ES of differing service-delivery modes. Across all single-case

methodology studies for 12 sessions of ReST treatment, including traditional four times per week service delivery and the newer approaches of twice-weekly, telehealth and combined clinician–parent service delivery, an ES of 3.99 was required for a small magnitude of change. Within the service-delivery approaches investigated in this thesis (twice-weekly delivery, telehealth mode and combined clinician-parent delivery), four of the 20 treated behaviours had ESs below the level indicating a small magnitude of change. Of these four, three were from participants in the combined clinician–parent delivery and were associated with statistically non-significant change following treatment. The final one was from a participant in the twice-weekly model and was associated with marginal treatment efficacy ($p = 0.048$). These ES calculations indicate that, on average, there is a trend for the largest treatment gain to be from telehealth treatment, and the smallest treatment gain may be from combined clinician–parent delivered treatment. Further investigations with larger numbers of children would be required to determine whether these trends reflect significant differences.

6.3 Change scores

A change score is a measure of difference, calculated by subtracting one measure from another (Peter, Churchill, & Brown, 1993). Change scores have intuitive appeal and are frequently used in small-scale speech pathology treatment research. Change scores enable a comparison of performance across participants with differing baseline performance. Change scores were used by Murray et al. (2015) to compare the difference in performance of children following either 12 sessions of ReST treatment or 12 sessions of the NDP-3 (Williams & Stephens, 2004).

Change scores are not without their limitations, as they do not take into account the likelihood of change based on differing baseline levels of skill. For example, a change score of 20% from a baseline accuracy of 10% is a 200% improvement. The same 20% change score from a baseline accuracy of 40% is only a 50% improvement. However, both of these improvements are considered a 20% change score.

6.3.1 Method of calculating change scores

Change scores were calculated within participant and within behaviour for each study. Change scores were calculated for all post-treatment data points (i.e., 1 week, 1 month and 4 months post-treatment) and were calculated by subtracting the highest baseline % accuracy from the post-treatment % accuracy at each data point. To conservatively estimate the efficacy of the treatment, the highest baseline measure was used, employing the following calculation:

Change score calculation: Post-treatment % accuracy − highest baseline % accuracy

In order to calculate a single change score for each participant's treated items, an average of the change scores for treated behaviours was calculated in cases where a participant had more than one treated behaviour. To calculate the score for untreated pseudo words, the participant's change scores for untreated pseudo words at the same level(s) of treatment was used. As for treated items, in cases where participants had more than one level of treatment, an average of the change scores for their two similar but untreated items was calculated. For untreated real words, the change scores were calculated on real words with the same number of syllables as the treated pseudo words.

The change scores were then averaged across participants, within behaviour (treated items, untreated pseudo words, untreated real words) and service-delivery type (twice-weekly, telehealth, combined clinician–parent delivery). The change scores for treated items, untreated pseudo words, and untreated real words were graphed, alongside the change scores for four times per week face-to-face clinician-delivered ReST treatment (Murray et al., 2015) see Figures 6.1, 6.2 and 6.3 respectively).

6.3.2 Visual analysis of change scores

6.3.2.1 Treated items

As shown in Figure 6.1, there was considerable variability in the performance of participants within each service-delivery group, as indicated by the standard error bars. Notwithstanding the variability, some trends in the means of the service-delivery groups are worth noting. Similar levels of change for treated items were noted in participants who received twice-weekly face-to-face treatment and those reported by Murray et al. (2015) following four times per week clinician-delivered treatment. This indicates broadly similar levels of improvement with treated items for twice-weekly and four times per week clinician-delivered treatment. On average, participants who received telehealth treatment had larger change scores than participants in all other service-delivery methods, indicating higher levels of improvement with treated items. On average, participants who received combined clinician–parent delivered treatment had lower change scores for treated items than participants in the other service-delivery methods, indicating lower levels of improvement with treated items.

Maintenance of treatment effect

Children in the telehealth treatment group showed a stable pattern of maintenance, neither improving nor deteriorating in performance up to 4 months post-treatment. Children in the twice-weekly treatment group had deteriorating performance between one month and four months post-treatment. The children who received combined clinician–parent delivered treatment improved their accuracy on treated items during the maintenance period, as did those studied by Murray et al. (2015).

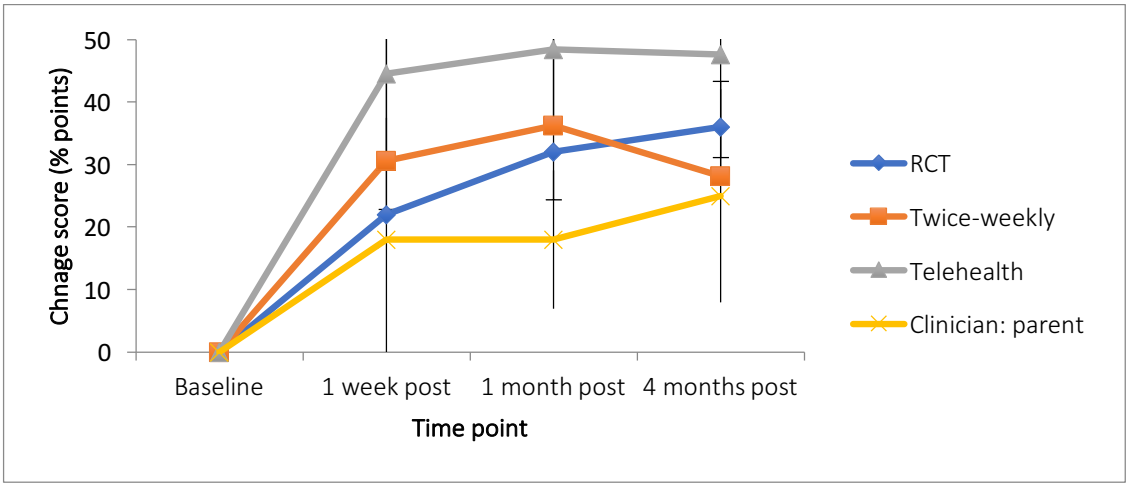

Figure 6.1: Change scores for treated items in each of the service-delivery approaches. These are compared with those reported by Murray et al. (2015) from their RCT (randomised control trial). Error bars indicate standard deviation at each time point for each service-delivery approach.

6.3.2.2 Untreated pseudo word items

As shown by the error bars in Figure 6.2, there was significant variability within members of each service-delivery group. The average change scores for untreated pseudo words of children receiving telehealth treatment were similar to those reported by Murray et al. (2015), indicating a similar degree of generalisation. The change scores for untreated pseudo words are lower for twice-weekly face-to-face treatment and combined clinician–parent treatment delivered four times per week than for the other service-delivery models.

Maintenance of pseudo word generalisation effect

As shown in Figure 6.2, all groups showed ongoing improvement from 1 week post-treatment to 4 months post-treatment.

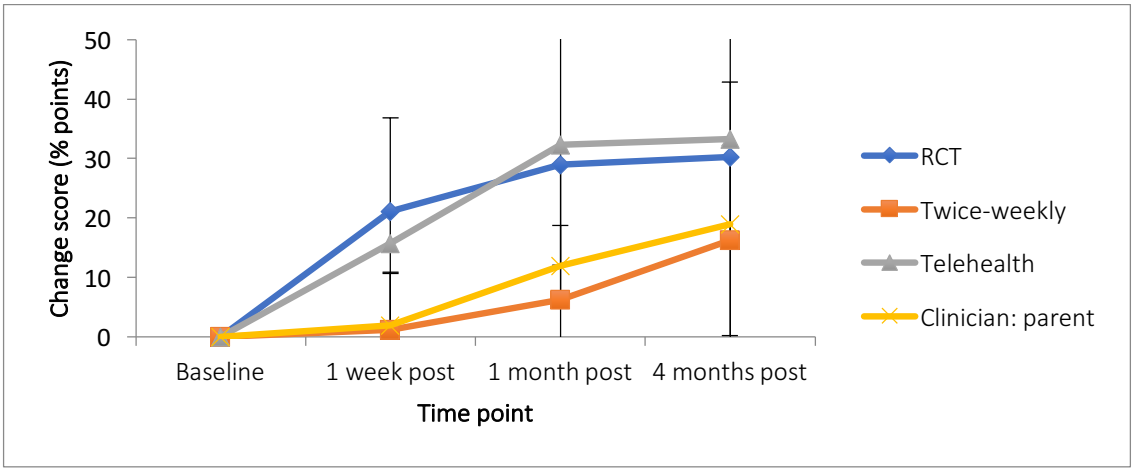

Figure 6.2: Change scores for untreated pseudo-word items in each of the service-delivery approaches. These are compared with those reported by Murray et al. (2015) from their RCT (randomised control trial). Error bars indicate standard deviation at each time point for each service-delivery approach.

6.3.2.3 Untreated real word items

The variability of change scores within each service-delivery group was lower for untreated real words than for treated and untreated pseudo words, as shown by the error bars in Figure 6.3. The change scores for untreated real words were similar for participants receiving telehealth, twice-weekly face-to-face and four times per week face-to-face ReST treatment (Murray et al., 2015), indicating similar levels of generalisation to real words. Participants receiving combined clinician–parent delivered treatment had lower change scores for untreated real words than participants receiving any other service-delivery model, indicating poorer generalisation to real words.

Maintenance of real word generalisation effect

The telehealth, combined clinician–parent, and twice-weekly treatment groups maintained their generalisation gain with a stable rather than improving or deteriorating profile in the maintenance period. This contrasts with the significant ongoing improvement within the maintenance phase for untreated real words shown by Murray et al. (2015) in their group study of children receiving treatment four times per week.

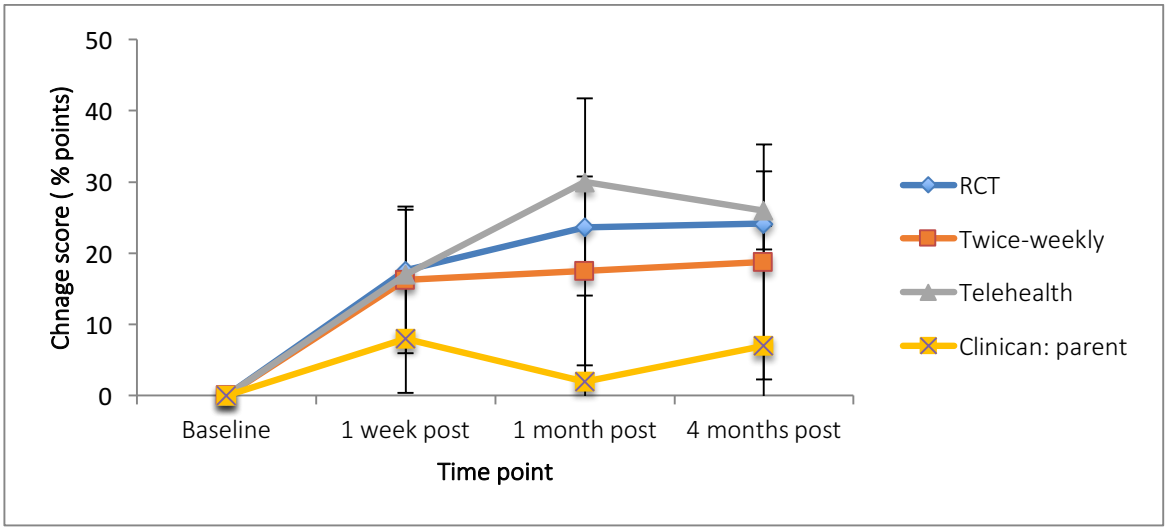

Figure 6.3: Change scores for untreated real words in each of the service-delivery approaches. These are compared with those reported by Murray et al. (2015) from their RCT (randomised control trial). Error bars indicate standard deviation at each time point for each service-delivery approach.

6.3.3 Statistical analyses of change scores

Although this research was designed as a series of single-case studies, it is possible to perform exploratory statistical investigations of the differences between the change scores of the service-delivery approaches. Such analysis may demonstrate whether any of the service-delivery approaches produced outcomes significantly different from the other approaches, and may indicate whether one approach was inferior or superior. It is important to note, however, that these investigations are only preliminary and are limited by the number of participants in each group (n = 4, 5, 5).

6.3.3.1 Method and results

Statistical analyses of the difference between change scores across service-delivery approaches were performed via independent pairwise comparisons between service-delivery groups within behaviour (treated items, untreated pseudo words, untreated real words) using Statistical Package for Social Sciences (IBM Corp, 2011). See Table 6.2 for the results.

There were no significant differences between service-delivery groups for treated items or untreated pseudo words. There was, however a marginally significant difference (p = 0.05) between the change scores for the telehealth group and the combined clinician–parent group on untreated real words. This result is subject to Type 1 error; however, it indicates a potentially stronger change on real words for participants receiving telehealth than for participants receiving combined clinician–parent delivered treatment.

Table 6.2: Pairwise comparisons of mean change scores averaged across 1 week, 1 month and 4 months post-treatment

		Treated items		Untreated pseudo words		Untreated real words	
Service delivery (a)	Service delivery (b)	Mean difference (a − b)	Sig.	Mean difference (a − b)	Sig.	Mean difference (a − b)	Sig.
Twice-weekly	Telehealth	−13.87	0.757	−19.22	0.228	−6.83	1.00
Twice-weekly	Clinician–parent	11.33	1.000	−3.08	1.000	11.83	0.362
Telehealth	Clinician–parent	25.20	0.120	16.13	0.327	18.67	0.050*

Note: Significance is based on estimated marginal means, with Bonferroni adjustment for multiple comparisons. Sig. = significance. * = marginally significant difference between groups.

6.3.4 Interpreting the change scores

These change scores indicate that children have a similar change in accuracy of treated items regardless of the service-delivery approach—that is, whether they have treatment twice-weekly face-to-face, via telehealth four times per week, or via a combined clinician–parent delivery model four times per week. The average change in accuracy of treated items for each of these models is similar to that reported following four times per week face-to-face clinician-delivered treatment (Murray et al., 2015). The small numbers and high variance within each group mean the possibility of Type 2 errors cannot be excluded.

Although visual analysis suggested a trend for poorer generalisation to untreated pseudo words following twice-weekly or combined clinician–parent delivered treatment than telehealth delivery, children had statistically similar generalisation in all service-delivery approaches. Significantly stronger generalisation to untreated real words was shown following telehealth treatment than following combined clinician–parent delivered treatment. For a treatment such as ReST that uses pseudo-word stimuli, the most ecologically valid measure of improvement is the generalisation to untreated real words. This finding of significantly stronger generalisation to real words following telehealth treatment than following combined clinician–parent treatment is therefore not only significantly significant, but also clinically important.

6.4 Putting all the aspects of efficacy together

Across all analyses of the data reported thus far in this thesis, telehealth treatment has been shown a trend towards higher efficacy than the other trialled service delivery models. Telehealth delivery resulted in the most number of children having a statistically significant change in the accuracy of their treated and untreated pseudo-word items. It also resulted in the highest average ES for treated items of the service-delivery approaches. The children in the telehealth group also had significantly stronger ESs for generalisation to real words than the combined clinician–parent group.

The other trend within the data is that combined clinician–parent delivered treatment has lower efficacy than the other service-delivery models. This combined model produced statistically significant treatment and generalisation effects for only some of the children treated. It had the lowest average ES of the three service-delivery methods, and three of the treated behaviours in this approach had ESs below what is considered a small treatment effect. It resulted in the lowest change scores for treated and untreated pseudo words, and on the most important measure of ecological validity—the change to untreated real words—it resulted in significantly poorer generalisation than did telehealth treatment.

6.5 Determining the factors associated with treatment effect

In Chapter 5, the factors that correlated with treatment outcome were explored within the combined clinician–parent delivery approach alone, to determine if any factors were associated with the differential performance across children. The correlation between ES and child factors (e.g., age, initial speech severity, and language ability), parent factors (history of speech/language/literacy difficulties, parental score on the Peabody Picture Vocabulary Test—fourth edition [PPVT-4], and reliability of perceptual judgements of child's speech) and treatment fidelity factors was investigated. In this chapter, this chapter now considers whether there are relationships between ES and other variables across all service-delivery approaches. Given that telehealth and twice-weekly delivery approaches do not have parent data to be considered, this chapter explores the correlations between ES and child variables and treatment fidelity variables only.

6.5.1 Method

ESs were calculated for all 20 treated behaviours across the 14 participants in the three service-delivery approaches, and scatterplots and correlation coefficients were calculated. To establish the relationship between ES and child variables, scatterplots and correlation coefficients of ES against each of the following child variables were created:

- age, in months;
- PPVT-4 (Dunn & Dunn, 2007) standard score from the pre-treatment assessment;
- percentage of phonemes correct on the Test of Polysyllables (Gozzard, Baker, & McCabe, 2006) from the pre-treatment assessment;
- Expressive Language Index standard score from the Clinical Evaluation of Language Fundamentals—Preschool-2 (CELF-P2) and/or Clinical Evaluation of Language Fundamentals—fourth edition (CELF-4; (Semel, Wiig, & Secord, 2006; Wiig, Secord, & Semel, 2004) from the pre-treatment assessment; and

- Receptive Language Index standard score from the CELF-P2 and/or CELF-4 (Semel et al., 2006; Wiig et al., 2004) from the pre-treatment assessment.

Readers may recall that for children who received treatment for more than one behaviour, each behaviour was treated for fewer than 12 sessions. In order to investigate the association between the number of sessions of treatment per behaviour and ES, a scatterplot of these two variables was created. The correlation coefficient (R^2) between the two factors was calculated, and the significance of the correlation using two-tailed Pearson correlation coefficients was determined.

6.5.2 Results

The results of the correlation analyses are shown in Table 6.3. With the exception of ES versus age in months and ES versus number of sessions treating the target behaviour, no apparent relationships were indicated, as all remaining correlation coefficients (R^2) were < 0.03. ES versus age in months showed a small negative correlation (R^2 = 0.218) that was statistically significant (p = 0.038; see Figure 6.4). ES versus number of sessions treating the target behaviour showed a small positive correlation (R^2 = 0.178) that was not statistically significant (see Figure 6.5).

Table 6.3: Correlation coefficient (R2) and two-tailed significance (p) for Pearson correlations between effect size and various variables

ES versus variable	RLI	ELI	PPVT-4	PCC	No. sessions treating goal	Age
R^2	0.00447	0.00008	0.0363	0.00223	0.17829	0.21857
p	0.7794	0.9702	0.8433	0.8432	0.0636	0.03766*

Note. Variables were as follows: receptive language, expressive language, receptive vocabulary, initial speech severity, number of sessions treating the target, and age in months; RLI = Receptive Language Index Standard Score from Clinical Evaluation of Language Fundamentals—fourth edition (CELF-4; Semel et al., 2006) or Clinical Evaluation of Language Fundamentals—Preschool-2 (CELF-P2; Wiig et al., 2004); ELI = Expressive Language Index Standard Score on CELF-4 or CELF-P2 (Semel et al., 2006; Wiig et al., 2004); PPVT-4 = Peabody Picture Vocabulary Test—fourth edition (Dunn & Dunn, 2007); PCC = percentage consonants correct on the Test of Polysyllables (Gozzard et al., 2006) at initial assessment; age = age in months when treatment commenced.

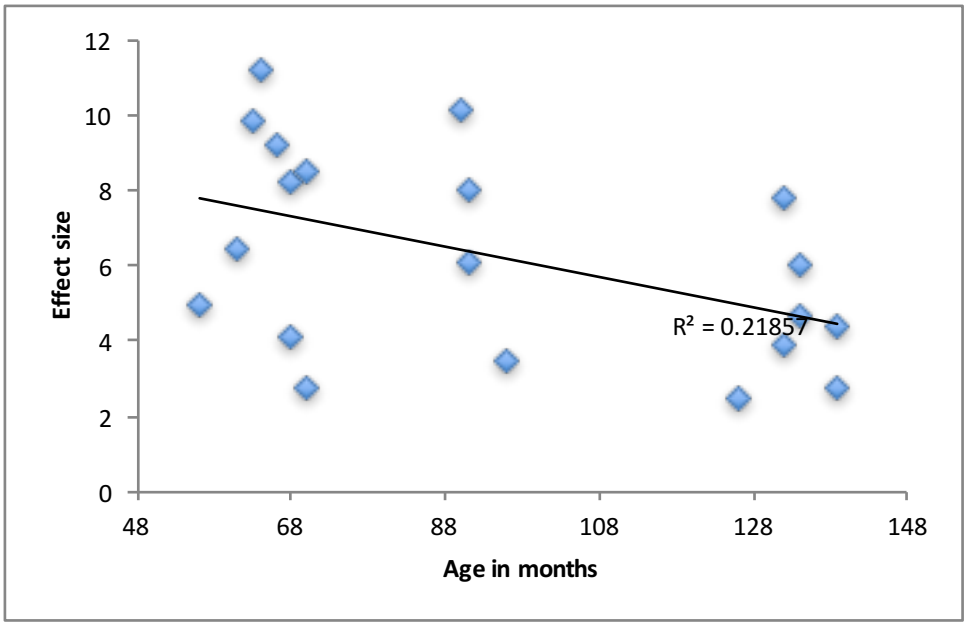

Figure 6.4: Effect size versus age in months for the 20 treated behaviours across the 14 participants in the three service-delivery approaches. This figure shows the trend line and R^2 value.

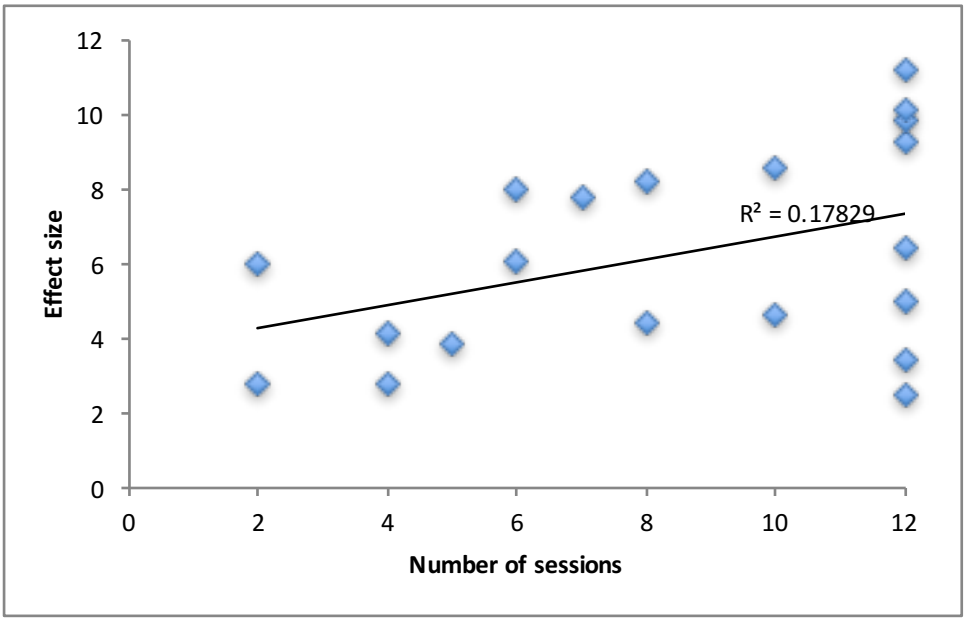

Figure 6.5: Effect size versus number of sessions treating target behaviour for the 20 treated behaviours across the 14 participants in the three service-delivery approaches. This figure shows the trend line and R^2 value.

6.5.3 Interpreting the correlation results

The correlations indicate that younger children had higher ES than older children. Age has previously been identified as a factor in treatment outcomes for children with CAS (Murray,

McCabe, & Ballard, 2013). Murray et al. (2013) reported that younger children had stronger gains following treatment with the NDP-3 (Williams & Stephens, 2004) and older children had stronger gains following ReST treatment. In this study, the influence of age on treatment outcome was the opposite to that reported by Murray et al. (2015).

There was also a small correlation between ES and the number of sessions treating the target behaviour. Although this correlation was not statistically significant, it may be worth further exploration in larger scale studies. A correlation between ES and the number of sessions training a target would not be unexpected, as the number of sessions is one of the key factors that drives motor (re)learning (Kleim, 2013). No correlations were found between ES and the other child variables, such as initial speech severity, language skill and PPVT-4 score.

6.6 Evidence-based practice framework

As discussed in Chapter 2, EBP provides a framework for health professionals to make clinical decisions regarding treatment. The E^3BP model (Dollaghan, 2007) incorporates three sources of evidence: (a) external evidence from systematic research, (b) internal evidence regarding clinical practice, and (c) the preferences of an informed client or carer. Only the first of those sources of evidence—external evidence from systematic research—has been explored thus far in this thesis in the quest to understand the effect of service-delivery variations in ReST treatment for CAS.

In speech pathology, there is comparatively more external evidence than the other E^3BP elements (Kovarsky, 2008; Kovarsky & Curran, 2007). For example, to date, there is no information about families' experiences with ReST treatment, and there is only limited information about families' experiences with parent-delivered treatment for children with SSD (e.g. Watts Pappas, McAllister, & McLeod, 2016). Such limited literature makes it difficult for clinicians to provide information to prospective families to assist them to make informed choices about treatment preferences and therefore fully implement the E^3BP model.

Chapter 7 presents a study addressing the paucity of information about parents' experiences of ReST treatment and alternative service-delivery models. Using qualitative methodology, Chapter 7 explores the parents' experiences, and compares and contrasts the experiences of parents when their child received ReST treatment via telehealth or a combined clinician–parent delivery model.

Chapter 7: Parent Experiences with Variations in Service Delivery for Rapid Syllable Transition Treatment

Author Attribution Statement

Chapter 7 of this thesis has been published as Thomas, D. C., McCabe, P., Ballard, K. J., & Bricker-Katz, G. (2017). Parent experiences of variations in service delivery of Rapid Syllable Transition (ReST) treatment for childhood apraxia of speech. *Developmental Neurorehabilitation, May 23,* 1–11.

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I declare that I made the following contribution to the study:

- Conception of the research question in collaboration with the other authors
- Design of the study with the other authors
- Data analysis, in conjunction with Geraldine Bricker-Katz
- Writing of the first draft of the manuscript
- Journal revisions and resubmission

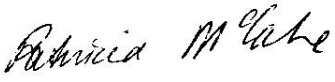
Name: Donna Thomas

Sign: 

Date: 27.6.17

As a co-author of the above paper and primary supervisor for the candidate upon which this thesis is based, I can confirm that the authorship attribution statements above are correct.

Name: Tricia McCabe

Sign: 

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ORIGINAL ARTICLE

Parent experiences of variations in service delivery of Rapid Syllable Transition (ReST) treatment for childhood apraxia of speech

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ABSTRACT

Purpose: To understand parents' perceptions of Rapid Syllable Transition (ReST) treatment and their experience of either telehealth or combined parent-clinician delivery of speech–language pathology. *Method*: Thematic analyses of semi-structured interviews were conducted with 10 parents (5 telehealth, 5 parent-clinician) after their child completed 12 sessions of ReST treatment. *Results*: Three themes were unique to telehealth: "telehealth was a million times easier," "technical problems weren't deal breakers," and "telehealth therapy has different boundaries." Three themes were unique to parent-clinician delivery: "therapy is something to get over and done with," "I wasn't very good at doing therapy," and "my child doesn't like me as his therapist." Both groups had themes related to the significance of childhood apraxia of speech, the importance of specialist treatment, and ReST being a "different way forward." *Conclusions*: Speech–language pathologists should carefully consider the suitability of caregiver-provided ReST treatment, and increase telehealth delivery of ReST treatment.

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Caregiver; dyspraxia; intervention; qualitative; therapy; telepractice

Introduction

Children with childhood apraxia of speech (CAS) require a greater number of treatment sessions, more frequently, and for a longer period than children with most other speech impairments.[1–3] An expert consensus paper recommended children with CAS receive individual treatment 3–5 times per week,[1] and a systematic review of CAS treatments reported effective treatments utilize 2–3 sessions per week.[4]

There are two main barriers to the implementation of such frequent, ongoing treatment for children with CAS. First, there is the time and cost involved in traveling to services, which has a disproportionate effect on families living in rural and remote locations.[5–7] Second, there are limits to the services available, and restrictions on the frequency of sessions when therapy is provided by publicly funded organizations.[8] The discrepancy between the requirements for effective treatment and available services can lead to frustration for families[9–11] and for clinicians.[12]

Alternative service delivery models provide potential solutions to the barriers of distance and limited services.[13,14] Telehealth applications such as videoconferencing enable real-time connection between client and clinician via the Internet. Telehealth is an efficacious service delivery for a range of speech and language impairments[15] including CAS.[16] Caregiver-provided therapy has the potential to overcome the barrier of limited speech–language pathology service, as it enables children to receive treatment at home between clinic visits. Caregiver-provided treatment is widely used by speech–language pathologists (SLPs)[17] with positive outcomes for several speech and language impairments.[18] Although home-practice is recommended for children with CAS,[4] the only investigation of caregiver-provided treatment for this population showed limited efficacy, with only 2 of 5 children demonstrating significant treatment effect and generalization.[19]

Effective implementation of Evidence Based Practice (E^{3}BP) requires consideration of three elements: the best available information regarding treatment efficacy, individual clinical expertise and consideration of the patient (or family's) preference.[20] However, compared to efficacy research, there is relatively limited information about patients' and families' preferences within the field of speech–language pathology.[21] Within the area of CAS, most reports of parents' experiences[22,23] relate to the parents' adjustment and well-being rather than their experience of specific treatments or service delivery approaches. In order to understand client and family perspectives, researchers frequently rely on qualitative methodologies.[20] These methods enable SLPs to access the more subjective and complex perspectives of those who experience a specific phenomenon such as a less familiar service delivery model and novel treatment. Understanding these perspectives enables SLPs to facilitate the implementation of E^{3}BP.

Rapid Syllable Transition (ReST) treatment is a relatively new treatment for CAS that has been trialed in alternative service delivery models. The treatment simultaneously improves the prosody and speech sound impairment associated with CAS. ReST is efficacious when delivered face to face four times a week,[24–26] as well as via telehealth four times a week.[16] The treatment has lower efficacy when delivered in a combined parent-clinician delivery model[19] compared with exclusively clinician-delivered treatment. ReST has some features that differ

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from traditional speech sound treatments.[27] It uses pseudo word stimuli, combined with treatment principles derived from the motor learning literature.[28] In ReST treatment, the aim is for the child to produce pseudo words with simultaneously correct sounds, beats (lexical stress pattern), and smoothness (absence of segregation between the syllables).[27] Each session begins with a "teaching" phase (termed "pre-practice" in the motor learning literature) where the child learns how to produce the pseudo word items correctly. During this phase the child is given immediate, specific feedback about each production, along with cues about how to improve any incorrect production (i.e., "knowledge of performance" or "KP" feedback). The majority of each session is spent in a "practice" phase, where the child attempts 100 productions of the pseudo words, presented in random order, and receives low-frequency right/wrong (i.e., "knowledge of results" or "KR" feedback) following a 3 second delay. Understanding families' experience of ReST can potentially guide future developments of the treatment, improve treatment implementation and professional training[29] as well as provide information to future families seeking to make an informed choice about treatment.

Investigation of parents' experience with stuttering treatment revealed that parents find aspects of home-based treatment challenging. Parents lack confidence in their ability to implement the Lidcombe Program for stuttering treatment.[30] They find it difficult to fit in home-based treatment,[29,31] and experience emotions such as guilt when their child has limited progress.[29,31] Regarding service delivery for childhood speech and language impairments, parents are willing to be involved at home,[32] provided they are supported and not left "to get on with it."[33] Rating scale studies indicate that parents generally find telehealth treatment convenient[34]; however, detailed and "rich" data about the caregiver's experience is not available using questionnaires and rating scales. To gather a more in-depth perspective of caregiver experiences of delivering treatment, it is prudent to use a qualitative methodology where participants can reveal their experiences and the richer data can be appropriately analyzed to derive meaning.[35]

We therefore used thematic analyses[35] to investigate the experiences of parents whose child received ReST treatment delivered by a clinician via telehealth, and parents whose child received ReST delivered in a combined parent-clinician model face to face. Our research question was as follows:

What is the parent experience when a child receives ReST treatment? Specifically:

a. What is the parent's experience of the specific service delivery model their child received–either telehealth delivery or combined parent-clinician delivery?
b. What is the parent's experience of ReST treatment?

Method

Author reflexivity

Reflexivity is the process by which qualitative researchers become conscious of the experiences, values, and biases they bring to the research, in order to understand their motivations and the lens through which they understand the findings.[35] All authors of this study are SLPs, university academics, and researchers. This article is one of a series of studies resulting from DT's PhD research, investigating service delivery options for children with CAS using the ReST treatment program. PM and KB are DT's PhD supervisors, and they have been authors on all ReST treatment papers published to date. This article arose from DT's interest in applying research efficacy findings to real-world contexts, and her desire to understand the acceptability of ReST treatment and the specific service delivery models employed. GBK was involved in the project as an independent qualitative researcher; she participated in data collection and data analysis.

Participants

Ten parents participated in the study; all had a child with CAS who received 12 sessions of ReST treatment across 3 weeks. The participants' children participated in one of two treatment efficacy studies, one investigating clinician-delivered treatment via telehealth,[16] and the other investigating combined parent-clinician delivered treatment with 6 clinician-delivered and 6 parent-delivered face-to-face sessions.[19] The efficacy studies were conducted sequentially, with the combined parent-clinician delivered treatment study completed prior to the clinician-delivered telehealth treatment study. Children were recruited to whichever study was running at the given time. Two of five families enrolled in the telehealth study were from rural areas, and the remaining three were from the metropolitan area; all five families enrolled in the combined parent-clinician delivery study were from the metropolitan area. The parent involved in the child's treatment was invited to participate in an individual semi-structured interview, 4 weeks after their child's final treatment session. Nine parents were female and one was male. Parents were assigned gender-neutral pseudonyms in order to protect their identity. The pseudonyms used for the children in the quantitative analysis of their treatment outcome[16,19] are also used here. Demographic information is available in Table 1. Ethical approval for the study was provided by the relevant Human Ethics Committee–approval numbers 2012/2824 and 2014/080.

Design

A qualitative approach was central to the research design and the data were treated by way of reduction and the extraction of themes to develop an understanding of how the treatment and the service delivery were experienced by the participants.[35] Parents were invited to participate in a semi-structured interview. The semi-structured nature of the interviews allowed the interviewer to explore issues raised by the parents and ask follow-up questions as required.[35] The analysis used a data-driven, inductive approach.[38,39]

Table 1. Demographic information.

Parent name	Age bracket	Highest educational level	Currently employed	Employment category (status)	Child name (age)	Child PCC	Service delivery
Sam	35–44	4 years high school	Yes	Clerical support (PT)	Ben (5:10)	25	Combined clinician: parent
Alex	35–44	Bachelor's degree	Yes	Professional (PT)	Eric (7:7)	94	Combined clinician: parent
Morgan	35–44	Bachelor's degree	No	-	Julian (10:6)	54	Combined clinician: parent
Chris	55+	Diploma	Yes	Professional (PT)	Matt (11:7)	93	Combined clinician: parent
Andy	35–44	6 years high school	No	-	Stacey (5:1)	80	Combined clinician: parent
Kim	35–44	Trade certificate	Yes	Elementary (PT)	Luke (5:3)	36	Telehealth
Mel	35–44	Postgraduate degree	Yes	Professional (PT)	Oliver (5:6)	76	Telehealth
Tracy	35–44	4 years high school	No	–	Lachlan (7:6)	67	Telehealth
Kerry	45–54	Bachelor's degree	Yes	Professional (FT)	Jack (11:0)	84	Telehealth
Shannon	35–44	Diploma	Yes	Service and sales (PT)	Emily (11:2)	85	Telehealth

Age brackets = <25, 25–34, 35–44, 45–54, 55+ years; Employment status categories determined using the International Standard Classification of Occupations-08;[36] PCC = percentage of consonants correct on the Test of Polysyllables;[37] PT = part time, <35 hr per week; FT = full time ≥35 hr per week; child age is (years:months).

Procedure

The semi-structured interviews were conducted either face to face (3) or via telephone (7); depending on the parent's preference. Interviews lasted between 24 and 53 min. An SLP experienced in qualitative research, but not associated with the treatment phase of the project, conducted the interviews. The interviews were audio recorded, transcribed verbatim, and sent to the parents for checking approximately 2 weeks after the interview. Parents were invited to review the transcripts and, if required, to make notes on the transcripts and clarify their intended meaning. Two of the 10 parents returned the transcripts with minor notes to enhance clarity.

Data analysis

As advocated by Boyatzis,[38] analysis was conducted initially within each service-delivery group (telehealth and combined parent-clinician delivery), then finally *across* the two groups. Authors DT and GBK independently analyzed the data by reading the transcripts several times in their entirety, and making notes on the transcript. The data were analyzed within each parent by summarizing each utterance and noting the key idea/s communicated by the parent. Multiple ideas emerged from each parent's data, and these ideas were analyzed for areas of convergence, which were coded as themes.[35] Following the independent identification of themes, authors DT and GBK compared their themes, and discussed any differences until they achieved consensus. The data were then analyzed across participants within each service delivery group, independently by the two authors, prior to discussion of any differences. The sections of text relating to each theme were copied from the original transcript into a separate document to enable comparison with other sections of text within the same theme. During this process, the themes were combined and divided as required to best represent the ideas communicated by the participants.[35] During each stage of analysis the researchers re-read the complete transcripts to check for context. Finally, the data were analyzed across service delivery models by DT and GBK independently, prior to discussion of differences until consensus was achieved.

Results

There were similarities and differences in the themes within parents across the different service delivery models. Three themes were common to both the telehealth and the parent-clinician service delivery groups—the significance of CAS, the importance of a specialist CAS service, and "ReST is a different way forward." An additional three themes were specific to parent-clinician delivery: Therapy is something to "get over and done with," "I wasn't very good at doing therapy," and "my child doesn't like me as his therapist." Finally, three different themes were unique to telehealth: "telehealth was better than I expected", "technical problems weren't deal-breakers," and "telehealth therapy has different boundaries." The themes and subthemes common to both service delivery groups and unique to each group are presented in Figure 1.

Themes common to both groups of parents

The themes and subthemes common to both groups are presented in Table 2, those unique to combined parent-clinician treatment in Table 3, and those unique to telehealth treatment in Table 4.

The significance of CAS

The parents felt the need to explain the context of having a child with CAS, even though the interview questions did not specifically explore this. Parents provided a context for their experiences and discussed the challenges of having a child with CAS. The parents reported that they encountered many obstacles in obtaining the correct diagnosis, sufficient treatment, and recognition of their child's impairment. They reported visiting many health professionals and up to seven SLPs before their child received a definitive diagnosis. For example, Kim had difficulty obtaining a diagnosis for Luke's speech difficulties, and the process involved numerous challenges and hurdles:

> When we found out, about the apraxia…, it was just like, "Oh here's another hurdle to go through" because, we just kept hitting brick walls. (Kim)

Parents reported that they needed to advocate for their child in order to get the recognition and services that they required. They felt there was a lack of recognition about CAS among SLPs, other health professionals, schools, and the

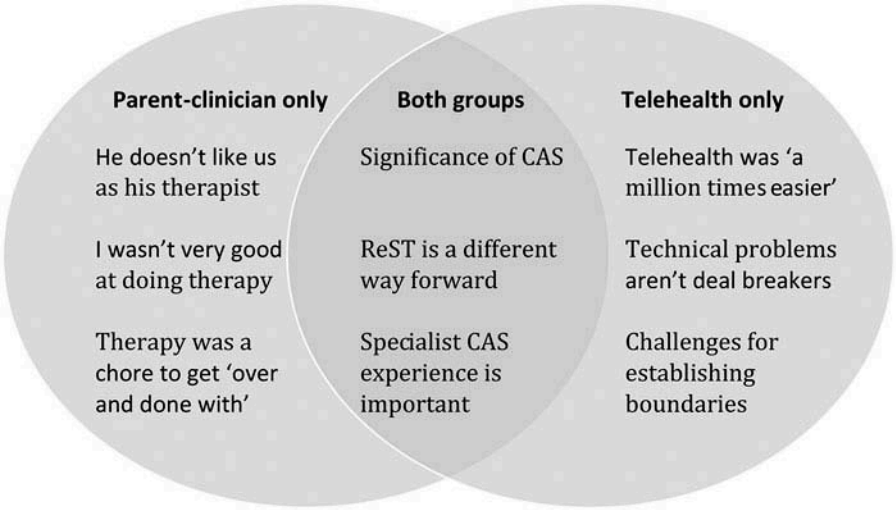

Figure 1. Themes unique to parent-clinician delivery, unique to telehealth delivery, and themes common to both groups. Themes common to both groups are shown at the intersection of the two circles.

Table 2. Themes and subthemes common to both groups.

Theme	Subtheme
Significance of CAS	Hard to get diagnosis
	Need long-term treatment
	Hard to get appropriate treatment
	Desperate for treatment
ReST is a different way forward	New, exciting, different
	Suits the needs of apraxia
	Generalizes to real words
	Why wasn't there more teaching?
Specialist CAS experience is important	Seeking specialist SLP
	Specialist treatment → hope
	Feeling of sadness when treatment ends

Table 3. Themes and subthemes unique to parent-clinician delivery.

Theme	Subtheme
He doesn't like us as his therapist	"Therapist" is a new role for me
	He is less compliant for us
	I didn't want to say "that's wrong"
I wasn't very good at doing therapy	I wanted to do a good job
	I mucked it up
	It was hard to remember how he said it
	I couldn't judge beats and smoothness
Therapy was a chore "to get over with"	It was intense
	It required reorganization of the family

Table 4. Themes and subthemes unique to telehealth.

Theme	Subtheme
Telehealth was "a million times easier"	Time efficient
	My child liked it
	Fitted better with home-life
Technical problems aren't deal breakers	I had technical problems
	It was OK
	My confidence increased
Challenges for establishing boundaries	No traditional markers of therapy
	Created a "dedicated space"
	Parent/clinician role less clear than face to face

broader community. In the example below, Andy describes the frustration associated with the limited support for children with CAS and the need to advocate for Stacey:

> I was feeling frustrated because there's no recognition for CAS kids. There's no support in schools, there's no support in speech therapists … I couldn't get recognition for Stacey having, if you want to call it a disability, a disability. Basically where Stacey's at today is because I have pushed, I have worked with her. (Andy)

The parents described their association with speech–language pathology as a "long journey" (Morgan). Their children had been attending therapy for a long time, and many parents felt there was no end in sight. Speech therapy, while being viewed as very important, brought additional burdens to parents in terms of locating an appropriate therapist, traveling to appointments, paying for private therapy, and practicing at home. This was "really tricky" (Kerry) and "quite stressful" (Kerry) for some parents. Shannon describes the financial struggle associated with private speech–language pathology:

> It's always a financial struggle, but it's more that with apraxia of speech they need huge amounts of speech therapy, so it's worse. (Shannon)

In some cases the challenges associated with accessing therapy became overwhelming. For Tracy, the barrier of distance and associated travel time resulted in Lachlan withdrawing from his community therapy:

> It was an hour one-way to get to therapy. And to only be there for sort of half an hour or 45 min, it was too much. So we put a halt to it there. (Tracy)

Despite the challenges, the parents were desperate to access treatment for their children. Many parents told of joining the ReST treatment trial even if they were unsure what the benefit would be because "any treatment is better than nothing." (Kim)

The importance of specialist CAS knowledge and experience

Most parents expressed a desire for their child to receive therapy from an SLP with specialist knowledge in CAS. Several parents made a distinction between generalist SLPs ("regular speechies") and those with CAS experience ("specialists"). Shannon described this distinction as follows:

> Now that I understand the condition better I also understand that neither of her two speech therapists she's had over the years really understands her condition, nor how to help it … If the regular speechies, and I put both of them in that category, don't know about her condition, how can they communicate that to me. (Shannon)

Although the parents appreciated the relationship they had with their general SLP, they expressed a level of disappointment with the assessment and treatment their child received. Some parents felt that generalist therapists treat CAS like any other speech impairment, and questioned whether the treatment was appropriate. Alex described Alex's concerns as follows:

> Personally, I've had lots of problems trying to find people who are actually trained and have an understanding of verbal dyspraxia. Often they use the same methods as they would for an articulation difficulty, or a different speech disorder. (Alex)

Several parents expressed excitement about being part of treatment research, attending a University clinic, and receiving treatment from "the newest scientists" (Morgan). They felt that participating in the research enabled their child to receive therapy from a team experienced with CAS. Alex expressed this as follows:

> I was confident in the fact that the people who are facilitating it, XXX Uni, had in-depth knowledge about verbal dyspraxia. (Alex)

Andy was relieved to find someone who understood Stacey's condition:

> When this came along it was like a sigh of relief that there was somebody out there that understood where I was coming from and offer[ing] a bit of relief … for Stacey, and I didn't have to do it all on my own. (Andy)

However, for Andy, the feeling of relief was replaced by a feeling of sorrow when the research treatment phase came to an end. Andy felt that the end of therapy signaled a return to the challenges of finding an appropriate therapist and accessing affordable treatment for Stacey.

> When the home sessions finished I went home and I cried because it was like "my job's done. I have nothing left to do, where do I go from here?" (Andy)

ReST is a different way forward

The parents were intrigued by ReST as it was novel and different to other treatments their child had received. They were excited to try something different and described it as "a different way forward." (Morgan). They felt "it was really refreshing to be doing something completely different" (Mel). Although they were excited, they were skeptical about the non-word stimulus items.

> Uh, it's very different to what I imagined. I mean non-words! I can't imagine how you learn to speak if you're saying things that no one is ever going to say again in your presence. (Chris)

Many parents felt that ReST treatment's explicit training of prosody met their child's needs. This alignment of the treatment goals and the parents' concerns gave the parents reason to feel "very hopeful" (Morgan) of a positive outcome for their child.

> I was just very excited to find something that kind of really fitted where he was at, like with his prosody. It just really seemed to fit what his needs were. (Alex)

Several parents told of improvements in their child's speech, including generalization to untrained real words. Given their initial skepticism about the non-words, the parents were surprised by this improvement. Kerry expressed her surprise as follows:

> Once I saw Jack starting to do [the therapy] … all of a sudden words that I knew he couldn't say properly he started saying properly … So I am just blown away. I mean I understand what they're trying to do, but I really don't understand how you can get them saying, you know, "darfebee" and for that to affect other words. (Kerry)

On the other hand, there were aspects of ReST treatment that the parents felt weren't well suited to their child. These were the motor learning features designed to enhance retention and generalization.[28,40] Specifically, the parents felt the children did not have sufficient time to learn the words in the pre-practice phase before they commenced the practice phase within each session. Mel expressed this as follows:

> I wondered why more time wasn't spent teaching him how to say the words. I know that is how the therapy was designed … but I was surprised that it was only a short bit. (Mel)

The parents also felt that the use of low frequency right/wrong (i.e. Knowledge of Results, KR) feedback in the practice phase of the session was not well suited to their child. Alex expressed concerns as follows:

> I think initially while he's still learning how to use that prosody, he could have done with more feedback… he also needs to know what exactly he said wrong. (Alex)

Themes specific to combined parent-clinician treatment

He doesn't like us as his therapist

Although the parents felt their child generally enjoyed spending time with them, this didn't extend to doing ReST therapy. The parents felt their child was less cooperative for them, more "whiney" (Kim) and that they generally worked better in the clinic than at home. Several parents mentioned that their child had less emotional regulation at home with them than in the clinic with the therapist. At home the child expressed more frustration, anger, and annoyance than in the clinic.

> He got more frustrated with me than he would have been with the therapist … I know that he's less tolerant with me because I'm mum. (Morgan)

The parents said that when doing therapy they needed to adopt a new role, that of "therapist" (Chris). There were aspects of that role that caused discomfort for both them and their child. Parents wanted to give feedback related to the child's level of effort rather than the accuracy of their production. Their normal role as "parent" was to give positive feedback and encourage their child, and when delivering ReST treatment they needed to provide a higher proportion of negative feedback. The parents reported that they felt uncomfortable giving their child negative feedback and worried about upsetting the child and damaging their morale. The potential impact on their child's emotions weighed heavily on them.

> Matt is very set that his teachers teach him and we don't…. He doesn't he doesn't view us as his therapists. He doesn't like us as his therapists. (Chris)

Chris reflected on the emotional toll of delivering therapy and explained concerns about giving feedback on accuracy rather than effort.

> At the time I didn't mind [giving negative feedback] because we had a task to perform, but later you feel bad putting an x next to your son [when it's] a hard attempt by your child to achieve something that they're struggling with. They're struggling, they're trying hard and you're having to say "no, no sorry." That's quite hard. (Chris)

Similarly, Sam felt bad about giving negative feedback because of the effect on Ben's morale. When discussing giving negative feedback Sam said:

> But for me, myself saying it, maybe I don't like to say it. I'd like to have said "oh I'm happy." By me saying "no that's not right," I did feel bad, of course. I didn't want to say "no," that he was doing it wrong…. I didn't like saying it was incorrect because I could see his little morale type of thing go down every time I said "incorrect." (Sam)

Andy's concerns about the emotional impact of the treatment on both Andy and Stacey were so significant that Andy considered withdrawing from the treatment, as the following example explains:

> At one point I thought of giving up, and I said "it's not the fact that I want to give up, it's the fact that I had to be the one to hurt her" so and I didn't like that feeling. (Andy)

I wasn't very good at doing therapy

The parents wanted to "do a good job" (Morgan). Although most were initially confident about their ability to do the task, it was harder than they expected. They didn't want to let down their child or the research team. The following example shows Chris explaining the desire to do a good job and not let people down:

> I was worried about me not hearing it correctly. I lay awake in bed at two am worrying about it. Because I wanted to do well, and I wanted Matt to do well and I wanted the research to go smoothly rather than it be like "ah we've got this parent on the research that's mucked it up." (Chris)

The "teaching" (i.e., pre-practice) phase was harder for the parents than the practice phase, because they had to select the word to teach, determine which elements were correct/in error, and provide cues. The parents were particularly concerned about their ability to produce the words correctly, and many attempted to read the words rather than use the sound file provided for the items. This difficulty with producing the words was magnified for Chris who had difficulty with reading and writing "b" and "d" in the non-word stimuli, as explained below:

> I've had so many mistakes. I had forgotten because I don't write things down anymore, but I had trouble with b and d. I don't know which side of the stick to put the ball. … I had a lot of trouble. I mucked Matt up by mispronouncing. (Chris)

The parents reported difficulty perceiving whether their child's productions were correct. Although several felt they improved across the six parent-delivered sessions, most felt their judgments were still unreliable at the end of the program. They had difficulty recalling exactly how their child produced the word or sentence and this made it difficult for them to judge whether the "sounds," "beats," and "smoothness" were correct.

> I really wasn't happy with the way I responded. I thought I was inconsistent. Even though I heard it, when I'd think about it again I couldn't replicate exactly what Matt had just said with the beat and sort of accent on words. I could just hear him saying the sentence, but exactly. I couldn't repeat it over and over in my head to check his beat and smoothness. It was like my recollection was an idealized version not exactly what I had just heard. (Chris)

The parents had more difficulty making prosodic judgments than judgments about sound accuracy. They felt their difficulty with prosody was related to the relative lack of emphasis on this construct in previous therapy. They had difficulty calibrating their prosodic judgments at the start of each home-based session. Morgan explained this in the following examples:

> With sounds I had [some experience] but not with beats, not with thinking about the smoothness of the word. I hadn't really thought about those things before I guess. For so long, we were just so concerned with getting Julian to produce the sounds, that had been our focus. (Morgan)

Therapy was a chore to "get over with"

Parents in the combined clinician-parent treatment group considered the home-based therapy sessions important, but not always lots of fun, a little like taking medicine. To both them and their child the treatment sessions were something to "get over with" (Chris), as Morgan explains:

> Julian just knew this was something that I had to do to help him, and that it wasn't always going to be great fun, and we'd have to persevere sometimes. (Morgan)

Although the intense nature of the treatment put pressure on the family, the parents wanted to honor the commitment they had made. All parents considered the intense treatment to be sustainable for the three-week period, even though many would have preferred shorter, less-frequent home-based sessions.

> I'm busy all the time, so I was sort of trying to get it in, so I was glad it was over. But if it was less time at home, maybe not as many days, just say once a week, doing that [would be better]. (Sam)

For Andy, however, the hour-long sessions were symbolic of the commitment and "full on" nature of parenting a child with CAS:

> The treatment was full on, it was going out to the university during the holidays … but we are used to committing to things and doing them for an intensive period of time. With the time I've had to put in with her, over the last few years especially, the hour sessions were nothing. (Andy)

Implementing home-based sessions prohibited parents from doing some of their regular home duties. Andy's partner needed to do the cooking, and Alex's partner needed to mind the other children. This put additional pressure on the parents.

Themes specific to telehealth

"It was a million times easier"

The parents whose children received exclusively telehealth-delivered treatment were surprised by the convenience, time-efficiency, and relative ease of the telehealth delivery. Not having to travel to appointments saved them time, effort, and money. Several parents also commented that they were better able to care for their other children when their child received telehealth treatment than clinic-based face-to-face treatment. Mel explains the convenience and time-saving in the example below:

> It takes less out of our day. We have other children and we both work, and we're pretty busy so to be able to say "we're off to speech therapy now" and it's just one hour out of your day rather than three hours, was great. (Mel)

The parents found telehealth treatment easy. As Kim noted "telehealth treatment is the same as normal treatment, just easier." They found their role within the sessions straightforward – they logged onto the web conferencing system, brought the child to the computer, and redirected the child's attention as required. Their child's willingness to engage in the sessions made their job easier than with previous clinic-based face-to-face therapy. When their child was eager and "enthusiastic" (Tracy) about therapy, the parent avoided conflict with their child about therapy. Many parents felt the novelty of the telehealth modality increased their child's enjoyment of the treatment.

> Lachlan was so enthusiastic, willing to do it, really. I'm not quite sure why, but he just seemed more enthusiastic and more willing to do it over the Internet. It was so convenient and good and it wasn't the hassle to get him to do it. (Tracy)

The parents felt that telehealth opened up the treatment options for their child that would otherwise be unavailable. They reported that telehealth could potentially enable their child to see an SLP with CAS experience, have more frequent sessions, and receive therapy when illness or injury would otherwise have prevented clinic-based visits. Several parents expressed a desire to seek out telehealth treatment in the future.

> I would jump at the chance to do video conferencing again. (Kerry)

Technical problems weren't deal breakers

All parents experienced technical difficulty during the study. The difficulties mostly related to audio and/or visual connection between the child and the therapist, transmission delay, and "freezing" of the videoconference. The parents had one or two teleconferencing familiarization sessions before treatment commenced, and they reported feeling confident following these sessions, and able to attempt to solve technical problems. Some of the technical issues could be quickly resolved, but others could not. Some parents had more technical issues than other parents, and there was variation within and across sessions. Even though the parents felt frustrated when they experienced unresolved technical issues, on balance, they still felt that telehealth was a viable mode of treatment for their child.

> We had a few rough days there that third week where the connection wasn't fantastic, it kept pausing. But apart from that, the rest of the time, it was fabulous I thought. (Tracy)

> Other than the delay, I don't think there was much problem, and as I said, the delay wasn't always bad. (Mel)

The parents had differing opinions about the effect that the telehealth modality had on the rapport between the child and the therapist. Mel felt that Oliver "might have been more engaged face-to-face," but Kerry felt that "rapport was just amazing … I thought maybe it would feel like we're talking over the computer, but that wasn't the case at all."

Telehealth presents new challenges for establishing boundaries

Most parents attempted to create a specific space for the telehealth treatment that was separated in some way from normal family life. They felt that home-based telehealth treatment didn't have the traditional markers of therapy such as a therapist's office, and special desks and chairs. Even though they were not specifically asked to, all parents created a specific space for telehealth sessions such as the child's bedroom or study, which limited noise and other distractions, and enabled the child to focus. Most parents reported that despite their efforts to limit distractions, there were still many distractions at home.

Even though the parents strived to ensure the sanctity of the therapy time, they were unable to communicate this to extended family and friends. The parents had not expected this to be a problem, and found it difficult to anticipate some of the interruptions to therapy. Shannon explains this as follows:

> My parents dropped around in the middle of one of the sessions and I was like "We can't talk now, we're in the middle of a session," but they just lacked that understanding of what was going on and the fact that I needed to be there with Emily … If you were with a speech therapist they would understand that. (Shannon)

Discussion

This study aimed to understand parents' perceptions of ReST treatment and their experience of either telehealth or combined parent-clinician delivery. This provided insight into

how families view different service delivery models as well as their perceptions of ReST treatment, and having a child with CAS. What emerged will have application for clinicians working with other families with children with CAS.

The parents whose children received combined parent-clinician delivered treatment had a less positive experience than those who received clinician-delivered telehealth. There were two main issues raised by the parents in the combined parent-clinician group, firstly, delivering therapy caused them to adopt the role of a "therapist," which was uncomfortable, and secondly, they lacked confidence and competence when delivering therapy. Parents are sometimes resistant to implementing home-based treatment,[32,41,42] and can experience negative emotions associated with home-based treatment[29]; however, the emotions experienced by the parents in the present study were specifically related to the tasks required when administering the treatment. Given that E^{3}BP requires knowledge of the preferences of clients and families,[20,21] SLPs can use the experiences of these parents to ensure that future parents are provided with information about how they may feel when implementing therapy, and to ensure that sufficient emotional support is provided during the course of home-based treatment.

The parents' experience of conducting therapy was characterized by doubt about their competence, particularly in regard to making perceptual judgments about the prosodic aspects (the "beats" and "smoothness") of their child's speech. Many parents reported having no prior awareness of the construct of prosody, in part because their child's treatment to date had focused exclusively on sounds. Prosody has taken a more prominent role in the diagnosis of CAS in the last decade[1]; however, based on the experiences of these parents, treatment for prosody is not yet commonplace in community clinics. Although some treatments (e.g., Dynamic and Temporal Cueing, DTTC,[43] ReST[27]) focus explicitly on the prosodic aspects of children's speech from the outset, other treatments (e.g., Nuffield Dyspraxia Program[44]) address prosody only in the later stages of treatment. Given the central nature of prosodic difficulties within CAS, it may be that a focus on prosody is warranted even in early stages of treatment. Treatment that addresses prosody may indirectly improve parents' perceptual awareness and judgments of this construct.

Clinicians need to carefully consider the type of therapy appropriate for parents to do at home. Although ReST treatment has demonstrated efficacy in clinician-delivered modalities,[24–26,45] it may be that the complexity of the treatment is not well suited to a parent-delivered modality. Complex impairments and treatments are less frequently provided via caregiver-delivered treatment.[17] ReST treatment is complex, requiring real-time evaluation of articulation accuracy, lexical stress, and concatenation of the syllables, as well as the provision of feedback with specific styles, delays, and schedules in different parts of the session. It may be that the complexity of the treatment, combined with parents' unfamiliarity with prosody, contributed to their perception of being "no good at doing therapy."

There is a need to explore the type of training, and level of competence required before parents deliver therapy at home. The amount of training received by the parents in the parent-clinician-delivered ReST group was low (cf. Sugden[17]), but similar to what would be received in a community clinic.[46] Greater amounts of training may improve parents' confidence and competence, but the time required to provide the training would need to be balanced against the finite resource of clinician time. Even if longer training produced increased levels of competence and confidence for parents, the time spent in training the parents may mean that, on balance, caregiver-provided treatment is not particularly time efficient.

Telehealth treatment is generally found to be convenient and motivating,[34,47] and the parents in this study agreed with these ideas. There is however a perception that telehealth treatment is for clients living in rural and remote areas.[48] This study shows that families living in metropolitan areas obtain benefits from treatment via a telehealth modality. SLPs should consider telehealth services for metropolitan clients as well as rural clients. The parents here confirmed the need for clinicians to explain to families the likely telehealth technical difficulties, and strategies to manage the technical problems. In order to improve efficacy, clinicians might engage parents in a discussion about ways to delineate telehealth sessions from regular family life and to minimize distractions within the home environment. Further research could investigate whether the children's willingness to engage in telehealth continues even when this modality is no longer novel.

The parents in both service delivery groups felt ReST addressed their child's speech needs; however, they were unsure about the suitability of some of the motor learning principles and the non-word stimuli. Although they understood at a theoretical level the purpose and processes of the treatment, at an emotional level they felt uncertain of whether the treatment provided sufficient support for their child. The principles that caused the most concern were those designed to facilitate retention and generalization,[28] (i.e., the provision of delayed, low-frequency, KR feedback). The parents may have been more familiar with principles designed to facilitate short-term acquisition (i.e., immediate, KP feedback on each trial), as these principles are exclusively used in many other speech treatments.[44,49] The experience of these parents highlights the need for clinicians to understand that parents feel uncomfortable about some motor learning principles, and that they may need additional support and discussion regarding the purpose of these principles within their child's treatment. Further research is warranted regarding the optimal use of motor learning principles in speech treatment for children, such as an understanding of the point at which treatment should move from focusing on principles to facilitate acquisition to facilitating retention and generalization.

All parents felt that their child's CAS presented challenges, and that obtaining a definitive diagnosis and appropriate treatment was challenging. Accessing sufficient speech–language therapy is well known to be difficult,[5,8,9,50,51] and acutely so when the child has a condition requiring ongoing intensive treatment.[1] The diagnosis of CAS has historically been fraught with difficulty, due to overdiagnosis[52] and the myriad symptoms considered to be associated with the impairment.[53] The increasing clarity regarding the diagnostic indicators,[1] combined with information about the

prelinguistic behaviors of children with CAS,[54–56] means that diagnosis should be less challenging now. These parents revealed the considerable pressure faced when seeking a diagnosis. It may be that dynamic assessment[57] is appropriate for children about whom there is some query regarding their diagnosis. In this way a child's treatment is not delayed pending the statement of diagnosis, and the child's progress during the dynamic assessment assists with the confirmation (or otherwise) of the diagnosis. Parents find it helpful to speak with other parents[22] and it may be that clinicians are in a position to link parents with existing parent support networks, particularly at times of concern such as when they are seeking a diagnosis.

Finally, the parents in this study expressed a desire for their child to see a therapist with specialist skills and experience in CAS. Although some professional organizations permit SLPs to receive specialist training and promote their specialist skills, this is not possible in all countries. There are a growing number of resources available to support clinicians working with children with CAS.[1,4,58,59] While SLPs can use the available resources to increase their skill level in the area of CAS, they have an ethical imperative to recognize the limitations of their experience and knowledge, and to make referrals to colleagues with more specialized knowledge and experience when necessary.[60]

Limitations

This was a small group study of Australian parents, and as such the generalization to the wider population of parents of children with CAS is limited. Due to the nature of the sampling, where participants were invited based on their child's participation in a treatment research study, we were not able to continue with sampling until saturation of themes was achieved. The themes identified in this study are potentially not exhaustive. The timing of the study, where all participants in a service delivery group completed their treatment at the same time, prohibited later participants from confirming or rejecting the themes of earlier participants. Other than inviting participants to check their transcripts, no further member checking was conducted, prohibiting participants from confirming that the identified themes resonated with their experience.

Conclusion

This study evaluated the experience of parents whose children received ReST treatment either from a clinician via telehealth or in a combined parent-clinician face-to-face model. The parents whose children received clinician-delivered telehealth treatment had more positive experiences than those who received combined parent-clinician face-to-face delivered treatment. When parents delivered the treatment they lacked confidence, particularly regarding their perceptual judgments about the child's speech, and they felt uncomfortable providing negative feedback to their child. When children received telehealth treatment, parents found it convenient, time efficient, and motivating for their child. The parents felt that ReST treatment was well suited to their child, but some queried the use of non-word stimuli and most expressed discomfort with the application of some motor-learning principles. Both groups of parents felt that having a child with CAS presented unique challenges in terms of obtaining a definitive diagnosis, and accessing sufficient and appropriate therapy.

Declaration of interest

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the article.

References

1. American Speech-Language-Hearing Association. Childhood apraxia of speech (Technical Report); 2007. http://www.asha.org/policy/TR2007-00278/%3E
2. Skinder-Meredith A. Differential diagnosis: developmental apraxia of speech and phonologic delay. Augmentative Communication News 2001;14:5–8.
3. Namasivayam AK, Pukonen M, Goshulak D, Hard J, Rudzicz F, Rietveld T, et al. Treatment intensity and childhood apraxia of speech. International Journal of Language & Communication Disorders 2015;50:529–546.
4. Murray E, McCabe P, Ballard KJ. A systematic review of treatment outcomes for children with childhood apraxia of speech. American Journal of Speech-Language Pathology 2014;23(3):486–504.
5. Verdon S, Wilson L, Smith-Tamaray M, McAllister L. An investigation of equity of rural speech-language pathology services for children: a geographic perspective. International Journal of Speech-Language Pathology 2011;13(3):239–250.
6. Verdon S, McLeod S, McDonald S. A geographical analysis of speech-language pathology services to support multilingual children. International Journal of Speech-Language Pathology 2014;16(3):304–316.
7. Wilson L, Lincoln M, Onslow M. Availability access, and quality of care: inequities in rural speech pathology services for children and a model for redress. International Journal of Speech-Language Pathology 2002;4(1):9–22
8. Baker E. The experience of discharging children from phonological intervention. International Journal of Language & Communication Disorders 2010;12(4):325–328.
9. Bercow J. The Bercow Report: a review of services for children and young people (0–19) with speech, language and communication needs; 2008. Available from http://dera.ioe.ac.uk/8405/7/7771-dcsf-bercow_Redacted.pdf%3E
10. Ruggero L, McCabe P, Ballard KJ, Munro N. Paediatric speech-language pathology service delivery: an exploratory survey of Australian parents. International Journal of Speech-Language Pathology 2012;14(4):338–350.
11. McAllister L, McCormack J, McLeod S, Harrison LJ. Expectations and experiences of accessing and participating in services for childhood speech impairment. International Journal of Speech-Language Pathology 2011;13(3):251–267.
12. Lim JM, McCabe P, Purcell A. Challenges and solutions in speech-language pathology service delivery across Australia and Canada. European Journal for Person Centered Healthcare (in press).
13. Keck CS, Doarn CR. Telehealth technology applications in speech-language pathology. Telemedicine and e-Health 2014;20(7):653–659.
14. Theodoros D. Speech-language pathology TD. In Kumar S, Cohn ER, (Eds), Telerehabilitation (pp. 311–323). London: Springer-Verlag, 2013.
15. Speech-Pathology-Australia. Telepractice in speech pathology; 2014. Available from http://www.speechpathologyaustralia.org.

au/spaweb/Document_Management/Public/Position_Statements.aspx%3E.
16. Thomas DC, McCabe P, Ballard KJ, Lincoln M. Telehealth delivery of Rapid Syllable Transitions (ReST) treatment for childhood apraxia of speech. International Journal of Language & Communication Disorders 2016;51(6):654–671.
17. Sugden E, Baker E, Munro N, Williams AL. Involvement of parents in intervention for childhood speech disorders: a review of the evidence. International Journal of Language & Communication Disorders 2016;51(6):597–625.
18. Lawler K, Taylor NF, Shields N. Outcomes after caregiver-provided speech and language or other allied health therapy: a systematic review. Archives of Physical Medicine and Rehabilitation 2013;94(6):1139–1160.
19. Thomas DC, McCabe P, Ballard KJ. Combined clinician-parent delivery of Rapid Syllable Transition (ReST) treatment for childhood apraxia of speech. International Journal of Speech-Language Pathology 2017.
20. Dollaghan CA. The handbook for evidence-based practice in communication disorders. Baltimore, MD: Paul H. Brookes, 2007.
21. Kovarsky D. Representing voices from the life-world in evidence-based practice. International Journal of Language & Communication Disorders 2008;43(sup1):47–57.
22. Allen LF, Babin EA. Associations between caregiving, social support, and well-being among parents of children with childhood apraxia of speech. Health Communication 2013;28(6):568–576.
23. Miron C. The parent experience: when a child is diagnosed with childhood apraxia of speech. Communication Disorders Quarterly 2012;33(2):96–110.
24. Murray E, McCabe P, Ballard KJ. A randomized control trial for children with childhood apraxia of speech comparing rapid syllable transition treatment and the Nuffield dyspraxia programm3rd edition. Journal of Speech, Language and Hearing Research 2015;58:669–686.
25. Ballard KJ, Robin DA, McCabe P, McDonald J. A treatment for dysprosody in childhood apraxia of speech. Journal of Speech Language & Hearing Research 2010;53(5):1227–1245.
26. McCabe P, Macdonald-D'Silva AG, Van Rees LJ, Ballard KJ, Arciuli J. Orthographically sensitive treatment for dysprosody in children with Childhood Apraxia of Speech using ReST intervention. Developmental Neurorehabilitation 2014;17(2):137–145.
27. Murray E, McCabe P, Ballard KJ. A comparison of two treatments for childhood apraxia of speech: methods and treatment protocol for a parallel group randomised control trial. BMC Pediatrics 2012;12(1):112.
28. Schmidt RA, Lee T. Motor control and learning: a behavioral emphasis, 5th. Champaign, IL: Human Kinetics; 2011.
29. Goodhue R, Onslow M, Quine S, O'Brian S, Hearne A. The Lidcombe program of early stuttering intervention: mothers' experiences. Journal of Fluency Disorders 2010;35(1):70–84.
30. Onslow M, Attanasio J, Harrison E. Parents talk about the Lidcombe program. In Onslow M, Packman A, Harrison E, (Eds). The Lidcombe program of early stuttering intervention: a clinician's guide (2nd edn, pp. 193–206). Austin, TX: Pro-Ed, 2003.
31. Hayhow R. Parents' experiences of the Lidcombe Program of early stuttering intervention. International Journal of Speech-Language Pathology 2009;11(1):20–25.
32. Watts Pappas N, McAllister L, McLeod S. Parental beliefs and experiences regarding involvement in intervention for their child with speech sound disorder. Child Language Teaching and Therapy 2016;32(2):223–239.
33. Glogowska R, Campbell M. Investigating parental views of involvement in pre-school speech and language therapy. International Journal of Language & Communication Disorders 2000;35(3):391–405.
34. Constantinescu G. Satisfaction with telemedicine for teaching listening and spoken language to children with hearing loss. Journal of Telemedicine and Telecare 2012;18(5):267–272.
35. Creswell JW. Qualitative inquiry and research design: choosing among five approaches. Thousand Oaks, CA: Sage Publications, 2013.
36. International_Labour_Orgainization. 2016 International Standard Classification of Occupations (ISC)-88). Available from http://www.ilo.org/public/english/bureau/stat/isco/isco08/index.htm%3E.
37. Gozzard H, Baker E, McCabe P. Children's productions of polysyllables. ACQuiring Knowledge in Speech, Language and Hearing 2006;8:113–116.
38. Boyatzis RE. Transforming qualitative information. Thematic analysis and code development. Thousand Oaks, CA: Sage Publications, 1998.
39. Braun V, Clarke V, Terry G. Thematic Analysis. In Rohleder P, Ac L, (Eds). Qualitative research in clinical and health psychology (pp. 95–113). Palgrave Macmillan, 2015.
40. Maas E, Robin DA, Austermann Hula SN, Freedman SE, Wulf G, Ballard KJ, et al. Principles of motor learning in treatment of motor speech disorders. American Journal of Speech-Language Pathology 2008;17(3):277–298.
41. Carroll C. "It's not everyday that parents get a chance to talk like this": Exploring parents' perceptions and expectations of speech-language pathology services for children with intellectual disability. International Journal of Speech-Language Pathology 2010;12(4):352–361.
42. Campbell M, Glogowska R. Investigating parental views of involvement in pre-school speech and language therapy. International Journal of Language & Communication Disorders 2000;35(3):391–405.
43. Strand E, Skinder A. Treatment of developmental apraxia of speech: integral stimulation methods. In Caruso AJ, Strand EA, (Eds). Clinical management of motor speech disorders in children (pp. 109–148). New York: Thieme, 1999
44. Williams P, Stephens H. Nuffield centre dyspraxia programme, 3rd edn. Windsor, England: The Miracle Factory, 2004.
45. Thomas DC, McCabe P, Ballard KJ. Rapid Syllable Transitions (ReST) treatment for childhood apraxia of speech: the effect of lower dose-frequency. Journal of Communication Disorders 2014;51:29–42.
46. Roulstone S, Wren Y, Bakopoulou I, Lindsay G. Interventions for children with speech, language and communication needs: an exploration of current practice. Child Language Teaching and Therapy 2012;28(3):325–341.
47. Mashima PA, Doarn CR. Overview of telehealth activities in speech-language pathology. Telemedicine and e-Health 2008;14(10):1101–1117.
48. O'Callaghan AM, McCallister L, Wilson L. Consumers' proposed solutions to barriers to access of rural and remote speech pathology services. International Journal of Speech-Language Pathology 2005;7(2):58–64.
49. Secord W. The traditional approach to treatment. In Creaghead N, Newman P, Secord W, (Eds). Assessment and remediation of articulatory and phonological disorders, (2nd edn, pp. 127–157). Columbus, OH: Merrill, 1989.
50. McCormack J, McAllister L, McLeod S, Harrison L. Knowing, having, doing: the battles of childhood speech impairment. Child Language Teaching and Therapy 2012;28(2):141–157.
51. Paradice R, Adewusi A. "It's a continuous fight isn't it?" parents' views of the educational provision for children with speech and language difficulties. Child Language Teaching & Therapy 2002;257-288:257–288.
52. Davis BL, Jakielski KJ, Marquardt TP. Developmental apraxia of speech: determiners of differential diagnosis. Clinical Linguistics & Phonetics 1998;12(1):25–45.
53. Forrest K. Diagnostic criteria of developmental apraxia of speech used by clinical speech-language pathologists. American Journal of Speech-Language Pathology 2003;12(3):376–380.
54. Overby M, Caspari SS. Volubility, consonant, and syllable characteristics in infants and toddlers later diagnosed with childhood apraxia of speech: a pilot study. Journal of Communication Disorders 2015;55:44–62.
55. Highman C, Hennessey NW, Leitão S, Piek JP. Early development in infants at risk of childhood apraxia of speech: a longitudinal investigation. Developmental Neuropsychology 2013;38(3):197–210.

56. Highman C, Leitao S, Hennessey N, Piek J. Prelinguistic communication development in children with childhood apraxia of speech: a retrospective analysis. International Journal of Speech-Language Pathology 2012;14(1):35–47.
57. Strand EA. Dynamic assessment of motor speech disorders in children. In Bowen C, (Ed). Children's speech sound disorders (pp. 353–356). Oxford: Wiley-Blackwell, 2015.
58. Maas E, Gildersleeve-Neumann C, Jakielski K, Stoeckel R. Motor-based intervention protocols in treatment of Childhood Apraxia of Speech (CAS). Current Developmental Disorders Reports 2014;1:197–206.
59. The Childhood Apraxia of Speech Association of North America (CASANA). The Apraxia Center on-demand webinars; n.d. Available from: http://apraxia-kids.org/webinars/%3E
60. Speech-Pathology-Australia. Code of Ethics; 2010. https://www.speechpathologyaustralia.org.au/spaweb/Document_Management/Public/Ethics.aspx%3E

Chapter 8: Discussion

At the start of this thesis, a problem was identified—that is, how to ensure that children with CAS are able to access sufficient intervention. The thesis then explored three service-delivery approaches as potential solutions to the problem, and it evaluated the efficacy of treatment for each approach and explored the parents' experiences with two of the three approaches. This chapter revisits the initial problem of barriers to service delivery, and critique each service-delivery approach in terms of its efficacy, acceptability and potential for overcoming access barriers. This chapter also considers the parents' perceptions of ReST treatment and service delivery, comparing and contrasting their perceptions with the available information about principles of practice for motor learning. Lastly, the chapter explains the limitations of the research and proposes directions for future research.

8.1 Access barriers and service-delivery approaches

As discussed in Chapter 1, despite evidence of treatment effectiveness, many children are unable to access treatment at the intensity shown to be effective (Ruggero et al., 2012). These difficulties with access are broadly attributable to structural, geographical and financial barriers (Verdon et al., 2011). The following section evaluates ReST treatment provided in each service-delivery approach in terms of efficacy, acceptability to parents, and potential to overcome service access barriers.

8.1.1 Dose-parameter modification—Low dose-frequency

8.1.1.1 Efficacy and acceptability

ReST treatment was efficacious with a lower dose-frequency of two sessions per week across a period of 6 weeks than the standard dose-frequency of four sessions per week for three weeks (see Chapter 3). Specifically, the lower dose-frequency produced significant acquisition of trained items and generalisation to most untreated behaviours for all participants. Although the participants maintained their treatment gain for 4 months post-treatment, they did not demonstrate the ongoing improvement shown in higher dose-frequency treatment (Murray et al., 2015). The

investigation reported in the published article reproduced as Chapter 3 of the thesis was the first investigation of dose-frequency modification in speech pathology treatment for children with CAS. Although higher dose-frequencies of three to five sessions per week are recommended for CAS (e.g. American Speech Language Hearing Association, 2007b), commonly used in CAS treatments (e.g. Strand et al., 2006), and supported theoretically from a motor learning (Maas et al., 2008) and neural plasticity perspective (Kleim, 2013), a lower dose-frequency of twice-weekly was efficacious.

It is important to note, however, there were some differences in the performance of the children receiving twice-weekly treatment and those receiving a higher dose-frequency (McCabe et al., 2014; Murray et al., 2015). The children receiving lower dose-frequency ReST treatment did not demonstrate ongoing improvement following the cessation of treatment (c.f. Murray et al., 2015), and only two of the four children demonstrated generalisation to untreated pseudo words. It may be that there is a threshold for dose-frequency in treatment for CAS and that dose-frequencies below this level have lower efficacy. In the speech and other motor-learning literature, there is evidence that more practice sessions per week produce greater treatment effect (e.g. Kleim, 2013; Maas et al., 2008; Schmidt & Lee, 2011). The small differences in generalisation and maintenance in the twice-weekly dose-frequency compared with four times per week may indicate that twice-weekly treatment nears this theoretical threshold. Further investigations of the efficacy of treatment at even lower dose-frequencies would be required to test this hypothesis. In other areas of paediatric speech and language therapy, there is evidence of a relationship between dose per session and frequency of sessions with regard to treatment outcome, such that low dose-frequency treatment is most effective with a high dose per session (e.g. Schmitt et al., 2017). It may be that the low dose-frequency investigated in this thesis was efficacious due to the high dose within the sessions. Although treatment of CAS is frequently underpinned by motor-learning principles that emphasise the need for large amounts of treatment (See Maas et al., 2014 for a review) rather than more-general learning theory that emphasises the need for cognitive consolidation between treatment sessions (e.g. Rohrer & Pashler, 2010), future studies in CAS should investigate

whether a similar relationship between dose, dose-frequency and treatment outcome exists within treatment for this population.

8.1.1.2 Impact on access barriers

Providing treatment at a lower dose-frequency of two sessions per week may help improve access to therapy by overcoming the structural barriers related to the number of sessions per week permitted within a speech pathology service. However, low dose-frequency alone is unlikely to be sufficient to overcome all structural barriers. Although two sessions per week is closer to routine service delivery than four sessions per week, it is still some way from the most common treatment frequency in Australia of one to two sessions per month (Ruggero et al., 2012) and in other countries of one session per week (Keilmann et al., 2004; Pascoe et al., 2010; Pring et al., 2012). It is important to remember that although a low dose-frequency was employed, the children discussed in Chapter 3 received 12 sessions of treatment, and many services have policies regarding the maximum number of sessions permitted (Baker, 2010; Ruggero et al., 2012). Such policies, which specify the maximum number of sessions allowed before a temporary or permanent discharge from the service, continue to present structural barriers, even within a lower dose-frequency service of two sessions per week.

It is not known whether ReST treatment would be efficacious at even lower dose-frequencies, such as one session per week, nor if ReST treatment would be efficacious at lower cumulative intervention intensities, such as six sessions. Although there are theoretical indications that a motor speech treatment such as ReST needs to be delivered at a minimum dose-frequency of twice-weekly (Murray et al., 2014) with high numbers of trials per session across an extended period (American Speech Language Hearing Association, 2007b), these constructs have not been confirmed with regard to ReST treatment specifically or CAS treatments more generally.

Providing treatment at a lower dose-frequency does not reduce the geographical barrier to access for families or the financial barrier. Although some families facing a geographical barrier may

find it more tenable to travel to a clinic for treatment twice per week rather than four times per week, twice-weekly treatment still required children to attend 12 clinic visits, with the same number of kilometres to be travelled, with the same associated travelling expenses and time costs. The investigations of distance decay (Eyles & Woods, 2014) indicate that families are willing to travel further for a less-frequent service (65 km for a fortnightly service [Wilson et al., 2002] cf. 50 km for a weekly service [Verdon et al., 2011). However, there has been no investigation of distance decay for children receiving treatment at a frequency of greater than one session per week. Lower dose-frequency treatment does not reduce the direct or indirect financial cost barriers known to affect the ability of families to access treatment (Dew et al., 2012; O'Callaghan, McAllister, et al., 2005).

8.1.1.3 Summary

Twice-weekly ReST treatment is efficacious and may help overcome some of the structural barriers to speech pathology access. Further efficacy research is required with dose-frequencies more similar to routine clinical practice and with reduced cumulative intervention intensities. Further investigations could also consider the parents' experience when their child receives clinic-based, clinician-delivered ReST treatment.

8.1.2 Mode modification—Telehealth

8.1.2.1 Efficacy and acceptability

ReST treatment delivered via telehealth modality four times per week by a clinician was efficacious (see Chapter 4), with larger treatment and generalisation gains than for the other service-delivery approaches investigated in the thesis. All children who received treatment via telehealth modality made significant treatment and generalisation gains, with three of the five participants demonstrating a large treatment effect, one a moderate treatment effect and one a small treatment effect (see section 6.2.2). Telehealth treatment also produced a larger change on treated items than any of the other treatment approaches investigated and than face-to-face

treatment of the same intensity (see section 6.3.3.1). Children who received treatment via telehealth modality showed significantly stronger generalisation to untreated real words than children who received the combined clinician–parent delivered treatment, again indicating the strong generalisation gains.

Treatment delivered via telehealth modality was well liked by parents and children. Parents considered telehealth to be more convenient than clinic-based sessions, motivating for their child, and helpful for them in managing their other home duties (see Chapter 7). These findings were unsurprising given that high satisfaction is frequently reported for telehealth treatments (e.g. Bridgman, Onslow, O'Brian, Jones, & Block, 2016; Molini-Avejonas et al., 2015). The parents in this study were concerned about the frequency of the sessions (four times per week); however, they reported that the telehealth modality was more convenient for intensive treatment than face-to-face clinic-based sessions. This implies that it was the length and frequency of the sessions of the treatment that was challenging for parents rather than the telehealth modality. Parents reported that their child was enthusiastic about sessions using telehealth and that this was because sessions using the computer via telehealth were novel for their child. Such factors, which improve a child's motivation and attention, can have positive effects on their performance in speech intervention (Kwiatkowski & Shriberg, 1998). Future research could investigate whether children remain enthusiastic and motivated about treatment via telehealth when it is no longer considered novel.

8.1.2.2 Impact on access barriers

Treatment delivered via the telehealth modality has long been advocated as a way of overcoming geographical barriers to accessing health services, as this modality eliminates the need for clients to travel to a clinic to receive services (e.g. Mashima & Doarn, 2008; Theodoros, 2013; Wilson et al., 2002). The parents' opinions in this study concurred with those of families in previous investigations of telehealth treatment, which have consistently reported that families find telehealth convenient because it eliminates the need to travel to appointments (Ciccia, Roizen, Garvey, Bielefeld, & Short, 2015; Constantinescu, 2012; Molini-Avejonas et al., 2015). The telehealth modality, however, does not overcome structural barriers related to policies regarding amount of service or direct financial barriers related to fees for services, and it may be most useful for clients whose primary concern is geographical barriers to access. Although there may be some potential reduction in indirect financial costs associated with travelling to appointments, there may be financial costs for some families related to the technology and Internet required to facilitate telehealth treatment in the home (Keck & Doarn, 2014).

8.1.2.3 Summary

Telehealth ReST treatment delivered four times per week by a clinician is efficacious and well accepted, indicating alignment of external evidence and parent preferences (Dollaghan, 2007). It may help alleviate the geographical barrier to access for families in locations distant from the clinician, but it has little impact on structural barriers and an equivocal effect on financial barriers. Although the financial costs associated with telehealth are not significant for most families, the costs associated with access to a computer with a webcam, microphone and Internet connection may be excessive for families with socioeconomic disadvantage. Future research could investigate clients' and parents' perceptions of telehealth treatment when this modality is no longer novel to the family. If clients are less motivated by telehealth when it is no longer novel, it would be important to consider whether this affects compliance and/or treatment efficacy.

8.1.3 Delivery-agent modification—Clinician–parent

8.1.3.1 Efficacy and acceptability

When a parent delivered half the treatment sessions, the treatment was less efficacious than when all sessions were delivered by a clinician (see Chapters 5 and 6). It was efficacious for two of the five children, had mixed efficacy for another two children and was not efficacious for the final child. This model had the lowest overall ES of all the service-delivery models investigated, with significantly lower generalisation to real words than telehealth treatment.

It was not immediately clear why the treatment was efficacious for some but not all the children. As discussed in Chapter 5, the child's treatment outcome appeared to be influenced by child, parent and treatment fidelity factors. Given that the performance of only five children was studied in the combined clinician–parent delivery approach, it was not possible to fully unpack the complex relationship between child, parent and fidelity variables. Age, pre-treatment speech severity and language ability have all previously been shown to influence treatment outcomes in clinician-delivered treatment for CAS in children without language impairments (Murray et al., 2013; Murray, McKechnie, & Williams, 2017). Further investigation of the factors that influence treatment outcome in a combined clinician–parent model is warranted to determine which children may benefit from a combined clinician-parent model.

One of the limiting factors in the implementation of a combined clinician–parent delivery model was the ability of the parents to make accurate perceptual judgements. Without accurate perceptual judgements, parents were unable to provide correct feedback to their child, and, unsurprisingly, reliability of parental judgement correlated with child's treatment outcome. All parents in this service-delivery approach provided treatment to their child after three clinic-based sessions, regardless of their accuracy of perceptual judgements. As discussed in Chapter 6, making explicit judgements about prosodic accuracy without reference to lexical information is a non-typical task (Cutler, 2012). The 3 hours of training that the parents received prior to implementing treatment and the 6 hours of training in total may have been insufficient for parents

to develop the cognitive constructs required for prosody judgements. Although designed around a parent-training model that has been successful elsewhere (Lidcombe Program, Onslow et al., 2003), this amount of training is substantially less than 15.8 hours—the average hours of training received across other studies of parent-delivered SSD treatment (Sugden et al., 2016). Future research iterations of combined clinician–parent delivered treatment should consider more-explicit training for parents regarding perceptual judgements of the child's speech, as well minimum accuracy thresholds for perceptual judgements prior to parents implementing treatment with their child.

Another factor limiting the clinical application of a combined clinician–parent service delivery was the variability of parents' treatment fidelity. Although most parents were able to implement ReST treatment with high levels of fidelity, one parent was not. As discussed in Chapter 6, the screening assessments for parents did not reveal any difference between this parent and the other parents that would foreshadow the very significant difficulties this parent had in implementing treatment. Further research investigation of parent factors associated with treatment fidelity is required before combined clinician–parent delivered ReST treatment could be recommended. Given the importance of treatment fidelity to treatment outcome (Kaderavek & Justice, 2010), it would be prudent for clinicians to establish minimum levels of fidelity for parent-delivered treatment and to assess the fidelity of parent-delivered treatment, both prior to and during treatment. If parent-delivered treatment does not meet these levels, further training and support could be provided or an alternative model provided.

Several parents in the combined clinician–parent delivery model had a history of speech, language and/or literacy difficulty. This finding should not be surprising, given the genetic influence in SSDs generally (Felsenfeld et al., 1994) and CAS specifically (Carrigg et al., 2016; B. Peter et al., 2016). Parents of children with CAS are more likely to have had a history of speech, language or literacy difficulties than parents of children without communication impairments (Felsenfeld et al., 1994; Lewis, Freebairn, Hansen, Gerry Taylor, et al., 2004) and more likely to have residual

speech and language difficulties (Felsenfeld et al., 1992). Even when the outward signs of a childhood speech or language difficulty are no longer present, there may be residual difficulties with phonological memory, phonological processing and literacy (Carrigg et al., 2016; Lewis, Freebairn, Hansen, Gerry Taylor, et al., 2004; Zaretsky, Velleman, & Curro, 2010). Therefore, parents of children with CAS are at higher risk of phonological memory and processing difficulties that could impair their ability to make accurate perceptual judgements about their child's speech (Carrigg et al., 2016). Further investigations of parents' phonological processing and phonological memory skills are warranted prior to providing parents with home-based speech-practice activities.

A combined intervention-agent model, where half the sessions were implemented by a parent, had lower levels of parent satisfaction than a fully clinician-delivered treatment in telehealth modality. As discussed in Chapter 7, the parents did not feel comfortable performing the role of clinician. They felt uncomfortable giving their child negative feedback, and they wanted to praise effort rather than accuracy. Furthermore, they felt their child was less compliant for them than for the clinician and that implementing treatment at home affected their ability to engage in other home duties. These findings are not unexpected, as parents have previously reported a preference for the clinician to take the primary role in implementing speech treatment (Glogowska & Campbell, 2000; Watts Pappas et al., 2016), and, despite a desire to be involved in treatment, parents have reported feeling overwhelmed when implementing treatment at home (Marshall & Goldbart, 2008). Many of the practical strategies for supporting parents when implementing home-based treatment (Sugden, Munro, Trivette, Baker, & Williams, 2017), such as provision of a treatment manual, treatment resources and a folder for treatment materials, were already in place for parents in the combined clinician–parent delivery model. It may be that the challenges for parents in delivering ReST treatment were related less to the procedural aspects and more to the perceptual aspects, the intensity of the treatment and its use of motor-learning principles.

As discussed in Chapter 7, some of the parents' concerns related specifically to the employment of motor-learning principles in ReST treatment. For example, parents reported relatively more difficulty administering the pre-practice phase of the sessions, as they needed to determine which of the three elements of the word (sounds, lexical stress and smoothness) were in error and provide their child with cues to improve the production. Although they found the practice phase easier, they felt unsure of their judgements about the prosody of the child's speech, which affected their confidence about the feedback they provided to their child. Given that prosodic elements are two of the three criteria judged in each ReST production trial, parent's limited confidence with prosodic constructs is a concern for parent-delivered ReST treatment.

Future research iterations of combined clinician–parent delivery agent for ReST treatment could explore avenues to work within the limitations expressed by parents. These may include investigation of clinician-delivered sessions consisting of exclusively pre-practice, with parent-delivered sessions consisting of exclusively practice. Alternatively, as voice recognition technology improves, it may be possible for home-based intervention sessions to be conducted using a computer application (Parnandi et al., 2015; Shahin et al., 2015), so that the parent's role becomes one of facilitating the child's engagement with the program, similar to the parent's role in telehealth delivered by a speech pathologist, rather than directly implementing therapy.

8.1.3.2 Impact on access barriers

Of all the service-delivery approaches investigated, modification of delivery agent has the most potential for overcoming access barriers. When parents deliver some of the treatment sessions, it reduces the need for clinician-delivered sessions, thereby overcoming structural barriers related to policies about the frequency of treatment sessions or number of treatment sessions. This model also reduces the geographical barrier to access because it negates the need for travelling to a clinic for all treatment sessions. As children receive fewer clinician-delivered sessions in this model, it also reduces the direct financial barriers. On balance, however, the indirect costs associated with a combined clinician–parent delivery model are likely to be equivocal. Although there are fewer

travel costs with this model, parents have indirect costs associated with time spent implementing the treatment and the subsequent inability to participate in work or other roles during this time.

8.1.3.3 Summary

Combined clinician–parent delivered ReST treatment has the potential for overcoming the most access barriers, however it is only efficacious for some children. At present, it is not clear which children will benefit from combined clinician–parent delivery. The two elements of E3BP investigated (Dollaghan, 2007)—that is, external evidence and the parents' preferences—align to indicate that in its present form, combined clinician-delivered treatment should not be used clinically. However, given the potential of this model to overcome access barriers and given the efficacy of parent-delivered treatment for other SSDs (e.g. Bowen & Cupples, 1999; Broen & Westman, 1990; Lawler et al., 2013), further investigations regarding how to improve the efficacy and acceptability are warranted. The broad avenues for such investigations include child and parent factors associated with treatment outcome, phonological processing skills associated with perceptual judgements of prosody, the optimal parent-training regime, the best configuration of clinic-based and home-based sessions, and computer applications that may assist with parent training and home-based implementation of treatment.

8.2 Reconciling parent preferences with empirical evidence

There was a discrepancy between the parents' desire for effective CAS treatment and their preferences regarding session length and frequency and the feedback provided to the child. Parents reported wanting a treatment that specifically targets the needs of children with CAS and being pleased that ReST treatment targeted prosody in addition to sounds. However, there were several aspects of ReST treatment that caused discomfort for parents. These were related to the amount of feedback their child received, the type of feedback, the length of the pre-practice phase, and the length and frequency of the sessions.

ReST treatment aims to treat the core deficit in CAS: speech motor planning and programming (American Speech Language Hearing Association, 2007b; McCabe et al., 2017). It uses principles from the motor-learning literature to guide target selection, presentation of stimuli and conditions of verbal feedback. In the motor-learning literature, a distinction is made between short-term performance and true learning (Maas et al., 2014; Maas et al., 2008; Schmidt & Lee, 2005). True learning is demonstrated when a skill is both retained and appropriately generalised to other contexts and behaviours. The factors that enhance short-term performance, such as accurately producing a target word in a clinic session, differ from the factors that enhance the long-term retention of the skill and its generalisation to other words with the same sounds. Although the majority of the literature on motor learning has been conducted with limb motor learning, there is growing evidence that these principles also apply to speech motor learning for typical speakers (Maas et al., 2014). The motor-learning literature is less clear when it comes to the application of these principles for people with speech impairments, particularly children (e.g., Maas et al., 2008; Maas et al., 2012; Maas et al., 2014).

A small number of investigations have examined the application of motor-learning principles to treatments for CAS, and they reveal mixed findings. Three studies have explicitly investigated the use of motor-learning principles for children with CAS (Edeal & Gildersleeve-Neumann, 2011; Maas et al., 2012; Maas & Farinella, 2012), and one has compared treatments that employ differing motor-learning principles (Murray et al., 2015). In accordance with the general trend for other motor-learning literature, a higher number of production trials produced better skill retention and generalisation when compared with a lower number of production trials (Edeal & Gildersleeve-Neumann, 2011). Similarly, treatment with ReST using delayed low-frequency knowledge-of-results feedback produced improved retention relative to treatment with the NDP-3 using immediate knowledge-of-performance feedback (Murray et al., 2015). However, mixed results were shown for the effect of blocked versus random presentation of stimuli (Maas & Farinella, 2012), and for high versus low frequency of verbal feedback (Maas et al., 2012) on

retention and generalisation of skill, possibly indicating an interaction between principles, child age and severity of CAS (Maas et al., 2014).

ReST treatment employs principles from the motor-learning literature that should theoretically facilitate retention and generalisation. It uses random presentation of stimuli, a high number of treatment trials, and a predominance of delayed low-frequency knowledge-of-results feedback. Parents, by contrast, expressed a desire for shorter, less-frequent sessions where their child had high levels of success in sessions and was provided with frequent, specific feedback. In short, they wanted treatment that did not use motor-learning principles associated with long-term retention and generalisation (Maas et al., 2008; Schmidt & Lee, 2011). It is possible that their preferences were due to their prior experience with other treatment approaches that employ principles that facilitate short-term performance or acquisition of speech skills (Maas et al., 2008; Schmidt & Lee, 2011), such as the NDP-3 (Williams & Stephens, 2004) which is commonly used for CAS treatment in Australia. In such treatments, the child's progress is monitored by in-session performance with targeted sounds, words and phrases (e.g. Van Riper, 1972; Williams & Stephens, 2004). Possibly, the parents did not understand the long-term beyond-clinic aim of ReST treatment and would therefore be helped by an explicit discussion of the principles being employed in the treatment. Parents may find it reassuring to be informed of their child's progress with generalisation tasks during treatment in addition to their progress with the treatment tasks (Sugden et al., 2017).

Such a discrepancy between the preferences of parents and the external evidence base (Dollaghan, 2007) creates conflict for clinicians (Kenny & Lincoln, 2012). Clinicians want to provide treatment that considers all three elements of E^3BP (Dollaghan, 2007), and may resolve the discrepancy between parent preference and empirical evidence by modifying the treatment (Lim et al., 2017; McCurtin & Roddam, 2012; Roulstone, 2015). Although Chapter 7 reported that parents would prefer shorter sessions with more-frequent specific feedback, there is no evidence to suggest that ReST treatment will continue to be effective if modified in accordance with their

wishes. Until such time as any modifications of the motor-learning principles in ReST treatment have been investigated, the treatment should be administered as manualised (McCabe et al., 2017) in clinical practice.

Future research, however, could consider the application of other motor-learning principles in ReST treatment. For example, it would be valuable to establish the ideal point to change from knowledge-of-performance feedback to knowledge-of-results feedback, the effect of feedback delay, the length of the pre-practice phase and the criterion for moving to the practice phase. Such investigations may improve the efficacy of ReST treatment and may have broader application to the understanding of motor-learning principles in treatment for children with speech impairments.

Potential discrepancies between parent preferences and the external evidence affecting the implementation of E^3BP may be resolved through discussion with parents and the development of parent information materials. In clinical practice, such discussions could cover the procedures and learning principles employed in various treatments and the rationale for these, as well as information about the experiences of previous parents (see Chapter 7). This information, combined with details of the efficacy of various treatments, may enable parents to make a truly informed choice about whether ReST treatment is an appropriate choice for their child.

8.3 Study strengths

These studies presented in this thesis were the first to empirically investigate the impact of service delivery on treatment outcomes in CAS. The findings support the clinical application of clinician-delivered ReST treatment either twice-weekly face-to-face or four times per week via telehealth. They do not support the application of parent-delivered ReST treatment.

The series of studies in this thesis added to the limited literature regarding treatment efficacy for children with CAS (Maas et al., 2014; Morgan & Vogel, 2009; Murray et al., 2014), adding further support for ReST treatment. Controlling the number of treatment trials and sessions not only enabled comparison across the studies in this thesis but also facilitated comparison with other

ReST investigations (McCabe et al., 2014; McCabe et al., 2010; Murray et al., 2015). The use of single-case methodology permitted detailed within-participant analysis, enabling the first evaluation of ReST treatment efficacy for children with receptive language impairment.

The studies were strengthened by both qualitative and quantitative investigations for two of the service-delivery models. This enabled evaluation of the rich and complex interplay between the empirical evidence base and the preferences of families.

8.4 Limitations

This thesis and the studies contained within it have several limitations. Firstly, the lack of qualitative information about parents' experiences with the low dose-frequency treatment limited the ability to evaluate the acceptability of the intensity-modification approach. The studies were conducted sequentially, and it was only after the low dose-frequency treatment had been completed that the decision was made to collect qualitative data. Future studies of service-delivery approaches with ReST treatment should collect qualitative data about parents' experiences. Future studies should also explore the application of internal evidence for speech pathologists when selecting intervention approaches (Dollaghan, 2007).

Some of the child participants in these studies had concomitant language impairments. Although concomitant language impairment is common in children with CAS (American Speech Language Hearing Association, 2007b; Lewis, Freebairn, Hansen, Iyengar, et al., 2004), the interpretation of their results was less clear-cut than in studies where such children are excluded.

There were limitations in the study design of the multiple baselines studies, discussed previously in Chapters 3, 4 and 5. At the time of study design, three points per phase was considered adequate (Kazdin, 2011); however, five data points per phase is now recommended (Kratochwill & Levin, 2014). Although the multiple baseline approach provides within-participant control, it is preferable to include a control for maturation in studies involving children, and no maturational control was included in the combined clinician–parent delivered treatment. Each study included

only a small number of children, either four or five. Small studies are common in low-incidence impairments such as CAS (Murray et al., 2014), but this may limit the generalisability of the results.

As discussed in Chapter 7, there were limitations related to design in the qualitative study. The nature of the study, with a fixed number of children receiving each service-delivery approach, prevented sampling until saturation of themes was reached. Potentially, the themes reported were not exhaustive. The study design, with all participants completing treatment simultaneously, did not permit parents to comment on the themes raised by previous participants. However, parents were invited to comment on the transcription of their interview as a means of member checking.

Although this thesis explored how varying service-delivery models may overcome barriers to accessing speech pathology, there is still some distance between the service-delivery approaches investigated and routine clinical practice (Ruggero et al., 2012). Further investigations should consider the efficacy and acceptability of ReST treatment in service-delivery approaches more similar to routine clinical practice. Following a series of Phase 2 trials, such as those contained within this thesis, a Phase 3 trial would typically be recommended (Robey, 2004). However, as ReST treatment has previously demonstrated efficacy in a randomised controlled trial (Murray et al., 2015), and CAS is a rare speech impairment, the next investigations of ReST treatment for CAS as outlined above, are more suited to further Phase 2 trials.

8.5 Future directions and conclusions

Although this thesis has provided evidence supporting the clinical application of two of the three service-delivery approaches investigated, it has also raised many questions. Firstly, it speaks of the need for further investigations of the efficacy and acceptability of service-delivery approaches with ReST treatment and with treatments for CAS more generally. Secondly, it exposes the complexities of parent-delivered treatment for children with CAS. Lastly, and arguably most

importantly, it argues for the need for the profession to question the seemingly inflexible structural barriers to accessing speech pathology services.

Although ReST treatment was efficacious in two of the assessed service-delivery models, there is a need to investigate other models. Given that speech pathology treatment in Australia is commonly delivered once per week (Ruggero et al., 2012), it is essential to investigate the efficacy and acceptability of weekly clinician-delivered treatment. It would also be valuable to confirm that telehealth treatment is efficacious in other dose-frequencies, such as twice-weekly, and that telehealth modality continues to be efficacious when it is no longer novel. Further investigations could consider the feasibility of training other delivery agents, such as allied health assistants, to deliver ReST treatment. Any such investigations should consider treatment efficacy; fidelity; and acceptability to clients, families and intervention agent. Future research should also explore clinicians' use of internal evidence in treatment for children with CAS.

Parent-delivered treatment has enormous potential for overcoming barriers to accessing services; however, not all children benefit from a combined clinician–parent delivery model. Further investigations are warranted to identify child and parent variables that may be associated with success in such a model. Additional assessment of parent phonological processing skills and phonological memory is recommended prior to any investigations of parent-delivered SSD treatment in order to explore the relationship between these parent variables and the fidelity of parent-delivered treatment.

Given the specific difficulty that parents experienced in making perceptual judgements of their child's prosody, future investigations should consider ways to develop these perceptual abilities. One option may be to use a computer application such as BRIDGE (Madill, 2017) or Scale of Suprasegmentals (SASS, Murray, 2017) to develop parents' competence and confidence with making perceptual judgements. Such programs separate the training of perceptual judgements from the other tasks involved in delivering treatment, enabling dedicated cognitive attention to the task of developing perceptual judgement (Madill, 2017). These programs have an added

advantage in that learners can use the program independently on any computer with Internet access. Future investigations should explore the use of applications such as BRIDGE (Madill, 2017) or CAPTain (Madill et al., 2016) for training adults with and without a history of speech, language and/or literacy difficulties to make reliable judgements of prosody.

Further investigations of parent-training methodologies are warranted to determine whether greater ReST treatment fidelity can be achieved by changing the amount, structure or sequencing of training. Future research should determine minimum accuracy criteria for treatment fidelity and perceptual judgement reliability in ReST as well as in SSD treatments more broadly. Such research could inform new ways to enhance parent confidence and competence with home-based treatment. Modifications to the tasks that parents implement during parent-delivered treatment should also be explored. It may be that a model in which parents only implement practice may be more appropriate than one where they implement both pre-practice and practice. Such a model would overcome the concerns that parents reported about implementing pre-practice.

Lastly, this thesis argues for the need for clinicians to consider whether the structural barriers encountered by clients when attempting to access speech pathology can be eliminated. Within the profession, there can be a perception that policies and procedures are inflexible and that the best a professional can do for their clients is to assist the client to navigate these policies. In terms of children with CAS, this may mean that clinicians assist families to understand the amount of publicly funded service provision their child is entitled to; which government-subsidised programs, if any, the child is eligible for; and where to access private providers. Moving forward, speech pathologists need to consider the mandate from their peak professional body (e.g. Competency Based Occupational Standards, Speech Pathology Australia, 2011) to advocate for their clients and to use the growing body of evidence to challenge policies that create structural barriers to accessing much-needed services.

References

Akamai. (2016). *State of the internet, Q4 report*. Retrieved from https://content.akamai.com/ap-en-pg8285-q4-2016-soti-connectivity-report.html?utm_source=GoogleSearch&gclid=CN2J25mjptQCFZIKKgodZsEDSw

Allard, E. R., & Williams, D. F. (2008). Listeners' perceptions of speech and language disorders. *Journal of Communication Disorders, 41*(2), 108–123.

Allen, M. M. (2013). Intervention efficacy and intensity for children with speech sound disorder. *Journal of Speech, Language, and Hearing Research, 56*, 865–877. https://doi.org/10.1044/1092-4388(2012/11-0076)

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders (DSM-5®)*. Washington, D.C: American Psychiatric Pubishing.

American Speech Language Hearing Association. (2005). Speech-language pathologists providing clinical services via telepractice: Position statement. *ASHA Supplement, 25*.

American Speech Language Hearing Association. (2007a). Childhood Apraxia of Speech [Position statement]. Retrieved from http://www.asha.org/policy/PS2007-00277/

American Speech Language Hearing Association. (2007b). Childhood apraxia of speech [Technical Report]. Retrieved from http://www.asha.org/policy/TR2007-00278/

Anthony, J. L., Aghara, R. G., Dunkelberger, M. J., Anthony, T. I., Williams, J. M., & Zhang, Z. (2011). What factors place children with speech sound disorders at risk for reading problems?. American Journal of Speech-Language Pathology, 20(2), 146-160.

Australian Government Department of Health. (2012). National E-Health strategy. Retrieved from http://www.health.gov.au/internet/main/publishing.nsf/Content/National+Ehealth+Strategy

Australian Government Department of Health. (2014). Chronic Disease Management (formerly Enhanced Primary Care or EPC) – GP services. Retrieved from http://www.health.gov.au/internet/main/publishing.nsf/content/mbsprimarycare-chronicdiseasemanagement

Baas, B. S., Strand, E. A., Elmer, & Barbaresi, W. J. (2008). Treatment of severe childhood apraxia of speech in a 12-year-old male with CHARGE association. *Journal of Medical Speech-Language Pathology, 16*(4), 181–190.

Bailey, D. J., Eatchel, K., & Wambaugh, J. (2015). Sound Production Treatment: Synthesis and quantification of outcomes. *American Journal of Speech Language Pathology, 24*(4), S798–S814. https://doi.org/10.1044/2015_ajslp-14-0127

Baker, E. (2010). The experience of discharging children from phonological intervention. *International Journal of Language and Communication Disorders, 12*(4), 325–328. https://doi.org/10.3109/17549507.2010.488326

Baker, E. (2012). Optimal intervention intensity. *International Journal of Speech-Language Pathology, 14*(5), 401–409.

Baker, E., & McLeod, S. (2011). Evidence-based practice for children with speech sound disorders: Part 1 narrative review. *Language, Speech, and Hearing Services in Schools, 42*(2), 102.

Ballard, K. J., Robin, D. A., McCabe, P., & McDonald, J. (2010). A treatment for dysprosody in childhood apraxia of speech. *Journal of Speech Language & Hearing Research, 53*(5), 1227–1245.

Ballard, K. J., Wambaugh, J. L., Duffy, J. R., Layfield, C., Maas, E., Mauszycki, S., & McNeil, M. R. (2015). Treatment for acquired apraxia of speech: A systematic review of intervention research between 2004 and 2012. American Journal of Speech-Language Pathology, 24(2), 316-337.

Barnett, W. S., Escobar, C. M., & Ravsten, M. T. (1988). Parent and clinic early intervention for children with language handicaps: A cost-effectiveness analysis. *Journal of Early Intervention, 12*(4), 290–298.

Bashir, A. S., Grahamjones, F., & Bostwick, R. Y. (1984). A touch-cue method of therapy for developmental verbal apraxia. *Seminars in Speech and Language, 5*(2), 127–137.

Beeson, P. M., & Robey, R. R. (2006). Evaluating single-subject treatment research: Lessons learned from the aphasia literature. *Neuropsychology Review, 16*(4), 161–169. https://doi.org/10.1007/s11065-006-9013-7

Behl, D. D., & Kahn, G. (2015). Provider perspectives on telepractice for serving families of children who are deaf or hard of hearing. *International Journal of Telerehabilitation, 7*(1), 1–12.

Bercow, J. (2008). The Bercow Report: A review of services for children and young people (0–19) with speech, language and communication needs. Retrieved from http://dera.ioe.ac.uk/8405/7/7771-dcsf-bercow_Redacted.pdf

Beukelman, D., & Mirenda, P. (2005). *Augmentative and alternative communication*. Baltimore: Paul H. Brookes.

Binger, C., Kent-Walsh, J., Berens, J., Del Campo, S., & Rivera, D. (2008). Teaching Latino parents to support the multi-symbol message productions of their children who require AAC. *Augmentative and Alternative Communication, 24*(4), 323–338.

Binger, C., & Light, J. (2007). The effect of aided AAC modeling on the expression of multi-symbol messages by preschoolers who use AAC. *Augmentative and Alternative Communication, 23*(1), 30–43.

Binger, C., Maguire-Marshall, M., & Kent-Walsh, J. (2011). Using aided AAC models, recasts, and contrastive targets to teach grammatical morphemes to children who use AAC. *Journal of Speech, Language, and Hearing Research, 54*(1), 160–176.

Bornman, J., Alant, E., & Meiring, E. (2001). The use of a digital voice output device to facilitate language development in a child with developmental apraxia of speech: a case study. *Disability and Rehabilitation, 23*(14), 623–634.

Bowen, C. (2010). Parents and children together (PACT) intervention. In A. L. Williams, S. McLeod, & R. J. McCauley (Eds.), *Interventions for Speech Sound Disorders in Children* (pp. 407–426). Baltimore Maryland, USA: Paul Brookes.

Bowen, C., & Cupples, L. (1999). Parents and children together (PACT): a collaborative approach to phonological therapy. *International Journal of Language and Communication Disorders, 34*(1), 35–83.

Boyle, J., McCartney, E., Forbes, J., & O'Hare, A. (2007). A randomised controlled trial and economic evaluation of direct versus indirect and individual versus group modes of speech and language therapy for children with primary language impairment. *Health Technology Assessment, 11*(25), 1–139.

Brandel, J., & Frome Loeb, D. (2011). Program intensity and service delivery models in the schools: SLP survey results. *Language, Speech, and Hearing Services in Schools, 42*(4), 461–490.

Bridgman, K., Onslow, M., O'Brian, S., Jones, M., & Block, S. (2016). Lidcombe Program webcam treatment for early stuttering: A randomized controlled trial. *Journal of Speech, Language, and Hearing Research, 59*(5), 932–939.

Broen, P. A., & Westman, M. J. (1990). Project parent: A preschool speech program implemented through parents. *Journal of Speech and Hearing Disorders, 55*(3), 495–502.

Brumbaugh, K. M., & Smit, A. B. (2013). Treating children ages 3–6 who have speech sound disorder: A survey. *Language Speech and Hearing Services in Schools, 44*(3), 306–319. https://doi.org/10.1044/0161-1461(2013/12-0029)

Burgess, L., Holtel, M. R., Syms, M. J., Birkmire-Peters, D. P., Peters, L. J., & Mashima, P. A. (1999). Overview of telemedicine applications for otolaryngology. *The Laryngoscope, 109*(9), 1433–1437.

Busk, P. L., & Serlin, R. C. (1992). Meta-analysis for single-case research. In T. R. Kratochwill & J. R. Levin (Eds.), *Single-case research design and analysis: New directions for psychology and education*. Hillsdale, NJ: Lawrence Earlbaum Associates.

Canadian Association of Speech Language Pathologists and Audiologists. (2006). Position paper on the use of telepractice for CASAPLA Speech-Language Pathologists and Audiologists. Retrieved from http://sac-oac.ca/professional-resources/resource-library/sac-position-paper-use-telepractice-sac-s-lps-and

Carpenter, C. J. (2010). A meta-analysis of the effectiveness of health belief model variables in predicting behavior. *Health Communication, 25*(8), 661–669.

(2015). Persistent speech sound disorder in a 22-year-old male: communication, educational, socio-emotional, and vocational outcomes. *Perspectives on school-based issues, 16*, 37–49.

Carrigg, B., Parry, L., Baker, E., Shriberg, L. D., & Ballard, K. J. (2016). Cognitive, linguistic, and motor abilities in a multigenerational family with childhood apraxia of speech. *Archives of Clinical Neuropsychology, 31*(8), 1006–1025. https://doi.org/10.1093/arclin/acw077

Carrillo, J. E., Carrillo, V. A., Perez, H. R., Salas-Lopez, D., Natale-Pereira, A., & Byron, A. T. (2011). Defining and targeting health care access barriers. *Journal of Health Care for the Poor and Underserved, 22*(2), 562–575.

Carroll, C. (2010). 'It's not everyday that parents get a chance to talk like this': Exploring parents' perceptions and expectations of speech-language pathology services for children with intellectual disability. *International Journal of Speech-Language Pathology, 12*(4), 352–361.

Carter, P. P., & Edwards, S. S. (2004). EPG therapy for children with long-standing speech disorders: predictions and outcomes. *Clinical Linguistics & Phonetics, 18*(6–8), 359–372.

Ciccia, A. H., Roizen, N., Garvey, M., Bielefeld, R., & Short, E. J. (2015). Identification of neurodevelopmental disabilities in underserved children using telehealth (INvesT): Clinical trial study design. *Contemporary Clinical Trials, 45*, 226–232.

Cirrin, F. M., Schooling, T. L., Nelson, N. W., Diehl, S. F., F., F. P., Maureen, S., . . . Adamczyk. (2010). Evidence-based systematic review: Effects of different service delivery models on communication outcomes for elementary school–age children. *Language, Speech & Hearing Services in Schools, 41*, 223–264.

Cohen, J. (1988). *Statistical power analysis for the behavioral sciences* (Vol. 2). Hillsdale, NJ: Lawrence Earlbaum Associates.

Constantinescu, G. (2012). Satisfaction with telemedicine for teaching listening and spoken language to children with hearing loss. *Journal of Telemedicine and Telecare, 18*(5), 267–272.

Culp, D. (1989). Developmental apraxia and augmentative or alternative communication–a case example. *Augmentative and Alternative Communication, 5*(1), 27–34.

Cumley, G. D., & Swanson, S. (1999). Augmentative and Alternative Communication options for children with developmental apraxia of speech: Three case studies. *Augmentative and Alternative Communication, 15*(2), 110–125.

Cunningham, B. J., Washington, K. N., Binns, A., Rolfe, K., Robertson, B., & Rosenbaum, P. (2017). Current methods of evaluating speech-language outcomes for preschoolers with communication disorders: A scoping review using the ICF-CY. *Journal of Speech, Language and Hearing Research, 60*(2), 1–18. https://doi.org/10.1044/2016_JSLHR-L-15-0329

Cutler, A. (2012). *Native listening: Language experience and the recognition of spoken words*. London, England: The MIT Press.

Dale, P. S., & Hayden, D. A. (2013). Treating speech subsystems in childhood apraxia of speech with tactual input: the PROMPT approach. *American Journal of Speech-Language Pathology, 22*(4), 644–661.

Daniel, G. R., & McLeod, S. (2017). Children with speech sound disorders at school: Challenges for children, parents and teachers. *Australian Journal of Teacher Education, 42*(2), 6.

Davies, K. E., Marshall, J., Brown, L. J. E., & Goldbart, J. (2016). Co-working: Parents' conception of roles in supporting their children's speech and language development. *Child Language Teaching and Therapy, 33*(2), 0265659016671169. https://doi.org/10.1177/0265659016671169

Dew, A., Veitch, C., Lincoln, M., Brentnall, J., Bulkeley, K., Gallego, G., . . . Griffiths, S. (2012). The need for new models for delivery of therapy intervention to people with a disability in rural and remote areas of Australia. *Journal of Intellectual and Developmental Disability, 37*(1), 50–53.

Dodd, B. (2005). *Differential diagnosis and treatment of children with speech disorder, 2nd Edition*. Philadelphia: Whurr.

Dodd, B., Holm, A., Crosbie, S., & McIntosh, B. (2006). A core vocabulary approach for management of inconsistentspeech disorder. *International Journal of Speech-Language Pathology, 8*(3), 220–230. https://doi.org/10.1080/14417040600738177

Dollaghan, C. A. (2007). *The handbook for evidence-based practice in communication disorders*. Baltimore, MD: Paul H. Brookes.

Duffy, J. R. (2005). *Motor Speech Disorders: Substrates*. St Louis, MO: Elsevier Mosby.

Dunn, L. M., & Dunn, D. M. (2007). Peabody Picture Vocabulary Test, (PPVT-4). *Minneapolis, MN: Pearson Assessments*.

Eadie, P., Morgan, A., Ukoumunne, O. C., Ttofari Eecen, K., Wake, M., & Reilly, S. (2015). Speech sound disorder at 4 years: prevalence, comorbidities, and predictors in a

community cohort of children. *Developmental Medicine and Child Neurology, 57*(6), 578–584.

Edeal, D. M., & Gildersleeve-Neumann, C. E. (2011). The importance of production frequency in therapy for childhood apraxia of speech. *American Journal of Speech-Language Pathology, 20*(2), 95–110.

Eiserman, W. D., McCoun, M., & Escobar, C. M. (1990). A cost-effectiveness analysis of two alternative program models for serving speech-disordered preschoolers. *Journal of Early Intervention, 14*(4), 297–317. https://doi.org/10.1177/105381519001400402

Eyles, J., & Woods, K. J. (2014). *The Social Geography of Medicine and Health*. Florence: Taylor and Francis.

Felsenfeld, S., Broen, P. A., & McGue, M. (1992). A 28-year follow-up of adults with a history of moderate phonological disorder: Linguistic and personality results. *Journal of Speech, Language, and Hearing Research, 35*(5), 1114–1125.

Felsenfeld, S., Broen, P. A., & McGue, M. (1994). A 28-year follow-up of adults with a history of moderate phonological disorder: Educational and occupational results. *Journal of Speech, Language, and Hearing Research, 37*(6), 1341–1353.

Fey, M. E. (1986). *Language intervention with young children*. Boston: Allyn and Bacon.

Fish, M. (2015). *Here's how to treat childhood apraxia of speech*. San Diego: Plural Publishing.

Forrest, K. (2003). Diagnostic criteria of developmental apraxia of speech used by clinical speech-language pathologists. *American Journal of Speech-Language Pathology, 12*(3), 376–380.

Gierut, J. A., Morrisette, M. L., & Dickinson, S. L. (2015). Effect size for single-subject design in phonological treatment. *Journal of Speech, Language, and Hearing Research, 58*(5), 1464–1481.

Gillon, G. T., & Moriarty, B. C. (2007). Childhood apraxia of speech: children at risk for persistent reading and spelling disorder. *Seminars in Speech and Language, 28*(1), 48–57.

Glass, G. V. (1977). Integrating findings: The meta-analysis of research. *Review of Research in Education, 5*, 351–379.

Glogowska, M., & Campbell, R. (2000). Investigating parental views of involvement in pre-school speech and language therapy. *International Journal of Language and Communication Disorders, 35*(3), 391–405. https://doi.org/10.1080/136828200410645

Glogowska, M., Campbell, R., Peters, T. J., Roulstone, S., & Enderby, P. (2002). A multimethod approach to the evaluation of community preschool speech and language therapy provision. *Child: Care, Health and Development, 28*(6), 513–521.

Glogowska, M., Roulstone, S., Enderby, P., & Peters, T. J. (2000). Randomised controlled trial of community based speech and language therapy in preschool children. *British Medical Journal, 321*(7266), 923.

Gozzard, H., Baker, E., & McCabe, P. (2006). Children's productions of polysyllables. *ACQuiring Knowledge in Speech, Language and Hearing, 8*, 113–116.

Gruber, F. A. (1999). Probability estimates and paths to consonant normalization in children with speech delay. *Journal of Speech, Language, and Hearing Research, 42*(2), 448–459.

Hall, P. K., Jordan, L. S., & Robin, D. A. (1993). *Developmental apraxia of speech: Theory and clinical practice*: Pro-ed Austin, TX.

Hall, P. K., Jordan, L. S., & Robin, D. A. (2007). *Developmental apraxia of speech: Theory and clincal practice, second edition*. Austin, Texas: Pro-Ed.

Harris, L., Doyle, E. S., & Haaf, R. (1996). Language treatment approach for users of AAC: Experimental single-subject investigation. *Augmentative and Alternative Communication, 12*(4), 230–243.

Hayden, D. (2006). The PROMPT model: Use and application for children with mixed phonological-motor impairment. *International Journal of Speech-Language Pathology, 8*(3), 265–281. https://doi.org/10.1080/14417040600861094

Hayhow, R. (2009). Parents' experiences of the Lidcombe Program of early stuttering intervention. *International Journal of Speech-Language Pathology, 11*(1), 20–25.

Health Workforce Australia. (2014). Australia's health workforce series: Speech Pathologists in focus.

Helfrich-Miller, K. R. (1994). A clinical perspective: melodic intonation therapy for developmental apraxia. *Clinics in Communication Disorders, 4*(3), 175–182.

Hill, A. J., & Miller, L. E. (2012). A survey of the clinical use of telehealth in speech-language pathology across Australia. *Journal of Clinical Practice in Speech-Language Pathology, 14*(3), 110–117.

Hines, M., Lincoln, M., Ramsden, R., Martinovich, J., Fairweather, C. (2015). Speech pathologists' perspectives on transitioning to telepractice: What factors promote acceptance? Journal of Telemedicine and Telecare, 21(8), 469-473.

Hodson, B. W., & Paden, E. P. (1991). *Targeting intelligible speech: A phonological approach to remediation, 2nd ed.* Austin, TX: Pro-Ed.

Hussain, R., & Tait, K. (2015). Parental perceptions of information needs and service provision for children with developmental disabilities in rural Australia. *Disability and Rehabilitation, 37*(18), 1609–1616.

IBM Corp. (2011). IBM SPSS statistics for Windows, Version 22.0. Armonk, NY: IBM Corp.

Iuzzini, J., & Forrest, K. (2010). Evaluation of a combined treatment approach for childhood apraxia of speech. *Clinical Linguistics & Phonetics, 24*(4–5), 335–345.

Iuzzini-Seigel, J., Hogan, T. P., Guarino, A. J., & Green, J. R. (2015). Reliance on auditory feedback in children with childhood apraxia of speech. *Journal of Communication Disorders, 54*, 32–42.

Joffe, V., & Pring, T. (2008). Children with phonological problems: a survey of clinical practice. *International Journal of Language and Communication Disorders, 43*(2), 154–164. https://doi.org/10.1080/13682820701660259

Johnson, C. J., Beitchman, J. H., & Brownlie, E. B. (2010). Twenty-year follow-up of children with and without speech-language impairments: Family, educational, occupational, and quality of life outcomes. *American Journal of Speech-Language Pathology, 19*(1), 51–65.

Kaderavek, J. N., & Justice, L. M. (2010). Fidelity: An essential component of evidence-based practice in speech-language pathology. *American Journal of Speech-Language Pathology, 19*(4), 369–379.

Kaipa, R., & Peterson, A. M. (2016). A systematic review of treatment intensity in speech disorders. *International Journal of Speech-Language Pathology, 18*(6), 507–520.

Kazdin, A. E. (2011). *Single-case research designs: Methods for clinical and applied settings*. New York, NY: Oxford University Press.

Keck, C. S., & Doarn, C. R. (2014). Telehealth technology applications in speech-language pathology. *Telemedicine and e-Health, 20*(7), 653–659. https://doi.org/10.1089/tmj.2013.0295

Keilmann, A., Braun, L., & Napiontek, U. (2004). Emotional satisfaction of parents and speech-language therapists with outcome of training intervention in children with speech and language disorders. *Folia phoniatrica et logopaedica, 56*(1), 51–61.

Kenny, B., & Lincoln, M. (2012). Sport, scales, or war? Metaphors speech-language pathologists use to describe caseload management. *International Journal of Speech-Language Pathology, 14*(3), 247–259.

King, A. M., Hengst, J. A., & deThorne, L. S. (2013). Severe speech sound disorders: An Integrated Multimodal Intervention. *Language, Speech, and Hearing Services in Schools, 44*, 195–210. https://doi.org/10.1044/0161-1461(2012/12-0023)

Kleim, J. (2013). *Neural plasticity: foundations for neurorehabilitation*. Scottsdale, Arizona: TANAS Publishing.

Klick, S. L. (1994). Adapted cuing technique: facilitating sequential phoneme production. *Clinics in Communication Disorders, 4*(3), 183–189.

Kovarsky, D. (2008). Representing voices from the life-world in evidence-based practice. *International Journal of Language and Communication Disorders, 43*(sup1), 47–57.

Kovarsky, D., & Curran, M. (2007). A missing voice in the discourse of evidence-based practice. *Topics in Language Disorders, 27*(1), 50–61.

Kratochwill, T. R., & Levin, J. R. (2014). *Single-case intervention research: Methodological and statistical advances*: American Psychological Association.

Krauss, T., & Galloway, H. (1982). Melodic intonation therapy with language delayed apraxic children. *Journal of Music Therapy, 19*(2), 102–113.

Kwiatkowski, J., & Shriberg, L. D. (1998). The capability-focus treatment framework for child speech disorders. *American Journal of Speech-Language Pathology, 7*(3), 27.

Lambier, J., & Atherton, M. (2003). General membership survey: Speech Pathology Association of Australia 2003. *Melbourne, Australia: The Speech Pathology Association of Australia Limited*.

Larrivee, L. S., & Catts, H. W. (1999). Early reading achievement in children with expressive phonological disorders. *American Journal of Speech-Language Pathology, 8*(2), 118–128.

Law, J., & Conti-Ramsden, G. (2000). Treating children with speech and language impairments: Six hours of therapy is not enough. *BMJ: British Medical Journal, 321*(7266), 908.

Law, J., Dennis, J. A., & Charlton, J. J. (2017). Speech and language therapy interventions for children with primary speech and/or language disorders (Protocol). *The Cochrane Database of Systematic Reviews, 1*. https://doi.org/10.1002/14651858.CD012490

Law, J., Garrett, Z., & Nye, C. (2004). The efficacy of treatment for children with developmental speech and language delay/disorder: A meta-analysis. *Journal of Speech, Language and Hearing Research, 47*(4), 924.

Law, J., Zeng, B., Lindsay, G., & Beecham, J. (2012). Cost-effectiveness of interventions for children with speech, language and communication needs (SLCN): a review using the Drummond and Jefferson (1996) 'Referee's Checklist'. *International Journal of Language and Communication Disorders, 47*(1), 1–10. https://doi.org/10.1111/j.1460-6984.2011.00084.x

Lawler, K., Taylor, N. F., & Shields, N. (2013). Outcomes after caregiver-provided speech and language or other allied health therapy: A systematic review. *Archives of Physical Medicine and Rehabilitation, 94*(6), 1139–1160. https://doi.org/10.1016/j.apmr.2012.11.022

Lewis, B. A., Freebairn, L. A., Hansen, A., Gerry Taylor, H., Iyengar, S., & Shriberg, L. D. (2004). Family pedigrees of children with suspected childhood apraxia of speech. *Journal of Communication Disorders, 37*(2), 157–175.

Lewis, B. A., Freebairn, L. A., Hansen, A. J., Iyengar, S. K., & Taylor, H. G. (2004). School-age follow-up of children with childhood apraxia of speech. *Language, Speech, and Hearing Services in Schools, 35*(2), 122.

Lewis, B. A., Freebairn, L. A., Tag, J., Ciesla, A. A., Iyengar, S. K., Stein, C. M., & Taylor, H. G. (2015). Adolescent outcomes of children with early speech sound disorders with and without language impairment. *American Journal of Speech-Language Pathology, 24*(2), 150–163.

Purcell, A. (2017). Challenges and solutions in speech-language pathology service delivery across Australia and Canada. *European Journal for Person Centered Healthcare, 5*(1), 120–128. https://doi.org/10.5750/ejpch.v5i1.1244

Lincoln, M., Hines, M., Fairweather, C., Ramsden, R., & Martinovich, J. (2014). Multiple stakeholder perspectives on teletherapy delivery of speech pathology services in rural schools: A preliminary, qualitative investigation. *International Journal of Telerehabilitation, 6*(2), 65–74. https://doi.org/10.5195/IJT.2014.6155

Little, A., & Grasselli, M. (2013). *Shifting the wait: meeting the demands for paediatric speech pathology services*. Paper presented at the Strong Commitment, Bright Future 12th National Rural Health Conference, Adelaide Convention Centre, SA, 7–10th April.

Lowe, R., O'Brian, S., & Onslow, M. (2014). Review of telehealth stuttering management. *Folia Phoniatrica et Logopaedica, 65*(5), 53–68.

Lüke, C. (2016). Impact of speech-generating devices on the language development of a child with childhood apraxia of speech: a case study. *Disability and Rehabilitation: Assistive Technology, 11*(1), 80–88. https://doi.org/10.3109/17483107.2014.913715

Lundeborg, I., & McAllister, A. (2007). Treatment with a combination of intra-oral sensory stimulation and electropalatography in a child with severe developmental dyspraxia. *Logopedics Phoniatrics Vocology, 32*(2), 71–79.

Maas, E., Butalla, C. E., & Farinella, K. A. (2012). Feedback frequency in treatment for Childhood Apraxia of Speech. *American Journal of Speech-Language Pathology, 21*(3), 239–257.

Maas, E., & Farinella, K. A. (2012). Random versus blocked practice in treatment for childhood apraxia of speech. *Journal of Speech, Language, and Hearing Research, 55*(2), 561–578.

Maas, E., Gildersleeve-Neumann, C., Jakielski, K. J., & Stoeckel, R. (2014). Motor-based intervention protocols in treatment of Childhood Apraxia of Speech (CAS). *Current Developmental Disorders Reports*(1), 197–206. https://doi.org/10.1007/s40474-014-0016-4

Maas, E., Robin, D. A., Austermann Hula, S. N., Freedman, S. E., Wulf, G., Ballard, K. J., & Schmidt, R. A. (2008). Principles of motor learning in treatment of motor speech disorders. *American Journal of Speech-Language Pathology, 17*(3), 277–298.

Maassen, B. (2002). Issues contrasting adult acquired versus developmental apraxia of speech. *Seminars in Speech and Language, 23*(4), 257–266.

Mabie, H., & Shriberg, L. (2017). *Speech and motor speech measures and reference data for the Speech Disorders Classification System (SDCS)*. Retrieved from

Madill, C. (2017). *BRIDGE: A bridge between theory and practice*. Paper presented at the Asia-Pacific Educational Collaboration in Speech-Language Pathology, Sydney.

Madill, C., Purcell, A., Murray, E., McCabe, P., Lowe, R., & Onslow, M. (2016). CAPTain: Comprehensive (Auditory) Perceptual Training tool: DVC Education, The University of Sydney.

Marquardt, T. P., Jacks, A., & Davis, B. L. (2004). Token-to-token variability in developmental apraxia of speech: three longitudinal case studies. *Clinical Linguistics & Phonetics, 18*(2), 127–144.

Marshall, J., & Goldbart, J. (2008). 'Communication is everything I think'. Parenting a child who needs Augmentative and Alternative Communication (AAC). *International Journal of Language and Communication Disorders, 43*(1), 77–98.

Martikainen, A.-L., & Korpilahti, P. (2011). Intervention for childhood apraxia of speech: A single-case study. *Child Language Teaching and Therapy, 27*(1), 9–20. https://doi.org/10.1177/0265659010369985

Martin, M. K., Wright, L. E., Perry, S., Cornett, D., Schraeder, M., & Johnson, J. T. (2016). Children with Developmental Verbal Dyspraxia: Changes in articulation and perceived resilience with intensive multimodal intervention. *Child Language Teaching and Therapy, 32*(3), 261–275.

Mashima, P. A., & Doarn, C. R. (2008). Overview of telehealth activities in speech-language pathology. *Telemedicine and e-Health, 14*(10), 1101–1117.

May, J., & Erickson, S. (2014). Telehealth, Why not? *Journal of Clinical Practice in Speech-Language Pathology, 16*(3), 147–151.

McAllister, L., McCormack, J., McLeod, S., & Harrison, L. J. (2011). Expectations and experiences of accessing and participating in services for childhood speech impairment. *International Journal of Speech-Language Pathology, 13*(3), 251–267.

McCabe, P., Macdonald-D'Silva, A. G., van Rees, L. J., Ballard, K. J., & Arciuli, J. (2014). Orthographically sensitive treatment for dysprosody in children with childhood apraxia of speech using ReST intervention. *Developmental Neurorehabilitation, 17*(2), 137–146. https://doi.org/10.3109/17518423.2014.906002

McCabe, P., McDonald-D'Silva, A., van Rees, L. J., Arciuli, J., & Ballard, K. J. (2010). *Using orthographic cues to improve speech production in children with & without childhood apraxia of speech*. Paper presented at the Motor Speech Conference, Savannah, GA.

McCabe, P., Murray, E., Thomas, D. C., & Evans, P. (2017). Clinician manual for Rapid Syllable Transition Treatment (ReST). Retrieved from http://sydney.edu.au/health-sciences/rest-media/rest-clinician-manual.pdf

McCabe, P., Preston, J., Murray, E., Bricker, G. & Morgan A., (2017). What happens when they grow up? *Experiences of adults who were diagnosed with childhood apraxia of speech as chidlren.* Paper presented at the Speech Pathology Australia National Conference, Sydney, Australia

McCormack, J., McAllister, L., McLeod, S., & Harrison, L. (2012). Knowing, having, doing: The battles of childhood speech impairment. *Child Language Teaching and Therapy, 28*(2), 141–157.

McCormack, J., McLeod, S., Harrison, L. J., & McAllister, L. (2010). The impact of speech impairment in early childhood: Investigating parents' and speech-language pathologists' perspectives using the ICF-CY. *Journal of Communication Disorders, 43*(5), 378–396. https://doi.org/10.1016/j.jcomdis.2010.04.009

McCormack, J., McLeod, S., McAllister, L., & Harrison, L. J. (2010). My speech problem, your listening problem, and my frustration: The experience of living with childhood speech impairment. *Language, Speech, and Hearing Services in Schools, 41*(4), 379–392.

McCormack, J., & Verdon, S. (2015). Mapping speech pathology services to developmentally vulnerable and at-risk communities using the Australian Early Development Census. *International Journal of Speech-Language Pathology, 17*(3), 273–286. https://doi.org/10.3109/17549507.2015.1034175

McCurtin, A., & Roddam, H. (2012). Evidence-based practice: SLTs under siege or opportunity for growth? The use and nature of research evidence in the profession. *International Journal of Language and Communication Disorders, 47*(1), 11–26.

McLaughlin, E., Lincoln, M., & Adamson, B. (2008). Speech-language pathologists' views on attrition from the profession. *International Journal of Speech-Language Pathology, 10*(3), 156–168.

McLeod, S. (2004). Speech pathologists' application of the ICF to children with speech impairment. *Advances in Speech Language Pathology, 6*(1), 75–81. https://doi.org/10.1080/14417040410001669516

McLeod, S. (2006). An holistic view of a child with unintelligible speech: Insights from the ICF and ICF-CY. *Advances in Speech Language Pathology, 8*(3), 293–315. *Clinical Linguistics & Phonetics, 28*(7–8), 508–531. https://doi.org/10.3109/02699206.2014.926994

McLeod, S., & Baker, E. (2017). Children's speech: An evidence-based approach to assessment and intervention: Boston, MA: Pearson Education.

McLeod, S., Harrison, L. J., McAllister, L., & McCormack, J. (2013). Speech sound disorders in a community study of preschool children. *American Journal of Speech-Language Pathology, 22*(3), 503–522.

McNeill, B. C., Gillon, G. T., & Dodd, B. (2009a). Effectiveness of an integrated phonological awareness approach for children with childhood apraxia of speech (CAS). *Child Language Teaching and Therapy, 25*(3), 341–366.

McNeill, B. C., Gillon, G. T., & Dodd, B. (2009b). A longitudinal case study of the effects of an integrated phonological awareness program for identical twin boys with childhood apraxia of speech (CAS). *International Journal of Speech-Language Pathology, 11*(6), 482–495. https://doi.org/10.1080/17549500902842583

McNeill, B. C., Gillon, G. T., & Dodd, B. (2009c). Phonological awareness and early reading development in childhood apraxia of speech (CAS). *International Journal of Language and Communication Disorders, 44*(2), 175–192.

McNeill, B. C., Gillon, G. T., & Dodd, B. (2010). The longer term effects of an integrated phonological awareness intervention for children with childhood apraxia of speech. *Asia Pacific Journal of Speech, Language & Hearing, 13*(3), 145–161.

Miccio, A. W., & Elbert, M. (1996). Enhancing stimulability: A treatment program. *Journal of Communication Disorders, 29*(4), 335–351. https://doi.org/10.1016/0021-9924(96)00016-0

Molini-Avejonas, D. R., Rondon-Melo, S., Amato, C. A., & Samelli, A. G. (2015). A systematic review of the use of telehealth in speech, language and hearing sciences. *Journal of Telemedicine and Telecare, 21*(7), 367–376. https://doi.org/10.1177/1357633X15583215

Morgan, A. T., & Vogel, A. P. (2009). A Cochrane review of treatment for childhood apraxia of speech. *European Journal of Physical and Rehabilitation Medicine, 45*(1), 103–110.

Moriarty, B. C., & Gillon, G. T. (2006). Phonological awareness intervention for children with childhood apraxia of speech. *International Journal of Language and Communication Disorders, 41*(6), 713–734.

Mullen, R., & Schooling, T. (2010). The national outcomes measurement system for pediatric speech-language pathology. *Language, Speech, and Hearing Services in Schools, 41*(1), 44–60.

Murray, E. (2017). *Reliability of a new tool for rating of articulation and supra-segmental speech: Scale of Articulation and Suprasegmentals*. Paper presented at the Speech PAthology Australia National Conference, Sydney.

Murray, E., McCabe, P., & Ballard, K. J. (2012). A comparison of two treatments for childhood apraxia of speech: methods and treatment protocol for a parallel group randomised control trial. *BMC Pediatrics, 12*, 1–9. https://doi.org/10.1186/1471-2431-12-112

Murray, E., McCabe, P., & Ballard, K. J. (2013). *Exploring factors that determined treatment success: data from a Randomized Control Trial for Childhood Apraxia of Speech*. Paper presented at the ASHA Convention, Chicago, USA.

Murray, E., McCabe, P., & Ballard, K. J. (2014). A systematic review of treatment outcomes for children with childhood apraxia of speech. *American Journal of Speech-Language Pathology, 23*(3), 486–504. https://doi.org/10.1044/2014_AJSLP-13-0035

Murray, E., McCabe, P., & Ballard, K. J. (2015). A randomized control trial for children with Childhood Apraxia of Speech comparing Rapid Syllable Transition treatment and the Nuffield Dyspraxia Programme—third edition. *Journal of Speech, Language and Hearing Research 58*, 669–686. https://doi.org/10.1044/2015_JSHLR-S-13-0179

Murray, E., McKechnie, J., & Williams, P. (2017). *Exploring factors for treatment success in childhood apraxia of speech following intervention using the Nuffield Dyspraxia Programme: 3rd edition*. Paper presented at the Speech Pathology Australia National Conference, Sydney.

Namasivayam, A. K., Pukonen, M., Goshulak, D., Hard, J., Rudzicz, F., Rietveld, T., . . . Lieshout, P. (2015). Treatment intensity and childhood apraxia of speech. *International Journal of Language and Communication Disorders, 50*, 529–546. https://doi.org/10.1111/1460-6984.12154

NDIS. National Disability Insurance Scheme. Retrieved from https://www.ndis.gov.au/about-us.html

Nippold, M. A. (2012). Different service delivery models for different communication disorders. *Language, Speech & Hearing Services in Schools, 43*, 117–120. https://doi.org/10.1044/0161-1461(2012/ed-02)

Nordness, A. S., & Beukelman, D. R. (2010). Speech practice patterns of children with speech sound disorders: the impact of parental record keeping and computer-led practice. *Journal of Medical Speech-Language Pathology, 18*(4), 104–108.

O'Brian, S., Iverach, L., Jones, M., Onslow, M., Packman, A., & Menzies, R. (2013). Effectiveness of the Lidcombe Program for early stuttering in Australian community clinics. *International Journal of Speech-Language Pathology, 15*(6), 593–603.

O'Callaghan, A. M., McAllister, L., & Wilson, L. (2005a). Barriers to accessing rural paediatric speech pathology services: Health care consumers' perspectives. *Australian Journal of Rural Health, 13*(3), 162–171.

O'Callaghan, A. M., McAllister, L., & Wilson, L. (2005b). Consumers' proposed solutions to barriers to access of rural and remote speech pathology services. *International Journal of Speech-Language Pathology, 7*(2), 58–64.

O'Halloran, R., & Larkins, B. (2008). The ICF Activities and Participation related to speech-language pathology. *International Journal of Speech-Language Pathology, 10*(1–2), 18–26. https://doi.org/10.1080/14417040701772620

Onslow, M., Packman, A., & Harrison, E. (2003). *The Lidcombe Program of Early Stuttering Intervention: A Clinician's Guide*. Austin, TX: Pro-Ed.

Ozanne, A. (1995). Search for developmental verbal dyspraxia. In B. Dodd (Ed.), *Differential diagnosis and treatment for children with speech disorders* (pp. 91–109). London: Whurr.

Paradice, R., & Adewusi, A. (2002). 'It's a continuous fight isn't it?': Parents' views of the educational provision for children with speech and language difficulties. *Child Language Teaching and Therapy, 18*(3), 257–288.

Parnandi, A., Karappa, V., Lan, T., Shahin, M., McKechnie, J., Ballard, K., . . . Gutierrez-Osuna, R. (2015). Development of a remote therapy tool for childhood apraxia of speech. *ACM Transactions on Accessible Computing, 7*(3), 10.

Pascoe, M., Maphalala, Z., Ebrahim, A., Hime, D., Mdladla, B., Mohamed, N., & Skinner, M. (2010). Children with speech difficulties: A survey of clinical practice in the Western Cape. *South African Journal of Communication Disorders, 57*(1), 66.

Pertile, J., & Page, F. (2003). The Maroondah Approach to Clinical Service (MACS): an integrated service delivery system. *ACQuiring Knowledge in Speech, Langauge and Hearing, 5*(2), 55–58.

Peter, B., Wijsman, E. M., Nato Jr, A. Q., Matsushita, M. M., Chapman, K. L., Stanaway, I. B., . . . Raskind, W. H. (2016). Genetic candidate variants in two multigenerational families with Childhood Apraxia of Speech. *PloS One, 11*(4), 1–27. https://doi.org/10.1371/journal.pone.0153864

Peter, J. P., Churchill, G. A., & Brown, T. J. (1993). Caution in the use of difference scores in consumer research. *Journal of Consumer Research, 19*(4), 655–662. https://doi.org/10.1086/209329

Preston, J. L., Brick, N., & Landi, N. (2013). Ultrasound biofeedback treatment for persisting childhood apraxia of speech. *American Journal of Speech-Language Pathology, 22*(4), 627–643.

Preston, J. L., Leece, M. C., & Maas, E. (2016). Intensive treatment with ultrasound visual feedback for speech sound errors in childhood apraxia. *Frontiers in Human Neuroscience, 10*(440), 1–9. https://doi.org/ 10.3389/fnhum.2016.00440

Preston, J. L., Maas, E., Whittle, J., Leece, M. C., & McCabe, P. (2016). Limited acquisition and generalisation of rhotics with ultrasound visual feedback in childhood apraxia. *Clinical Linguistics & Phonetics, 30*(3–5), 363–381.

Pring, T., Flood, E., Dodd, B., & Joffe, V. (2012). The working practices and clinical experiences of paediatric speech and language therapists: a national UK survey. *International Journal of Language and Communication Disorders, 47*(6), 696–708. https://doi.org/10.1111/j.1460-6984.2012.00177.x

Roberts, M. Y., & Kaiser, A. P. (2011). The effectiveness of parent-implemented language interventions: A meta-analysis. *American Journal of Speech-Language Pathology (Online), 20*(3), 180–199.

Robey, R. R. (2004). A five-phase model for clinical-outcome research. *Journal of Communication Disorders, 37*(5), 401–411.

Rohrer, D., & Pashler, H. (2010). Recent research on human learning challenges conventional instructional strategies. *Educational Researcher, 39*(5), 406–412.

Rosenstock, I. M. (1990). The health belief model: Explaining health behavior through expectancies. In K. Glanz, F. M. Lewis, & B. K. Rimer (Eds.), *Health Behaviour and Helath Edication* (pp. 39–62). San Francisco: Josey-Baase.

Roulstone, S. (2015). Exploring the relationship between client perspectives, clinical expertise and research evidence. *International Journal of Speech-Language Pathology, 17*(3), 211–221. https://doi.org/10.3109/17549507.2015.1016112

Royal College of Speech & Language Therapists. (2011). *RCSLT policy statement Developmental Verbal Dyspraxia*. Retrieved from https://www.ndp3.org/documents/rcslt2011dvdPolicyStatement.pdf

Ruggero, L., McCabe, P., Ballard, K. J., & Munro, N. (2012). Paediatric speech-language pathology service delivery: An exploratory survey of Australian parents. *International Journal of Speech-Language Pathology, 14*(4), 338–350.

Rvachew, S., & Rafaat, S. (2014). Report on benchmark wait times for pediatric speech sound disorders. *Revue canadienne d'orthophonie et d'audiologie| Vol, 38*(1).

Sackett, D. L., Rosenberg, W. M., Gray, J. M., Haynes, R. B., & Richardson, W. S. (1996). Evidence based medicine: what it is and what it isn't: British Medical Journal Publishing Group.

Schmidt, R. A., & Lee, T. (2011). *Motor control and learning: A behavioral emphasis (5th ed.)*. Champaign, IL: Human Kinetics.

Schmidt, R. A., & Lee, T. D. (2005). *Motor Control and Learning-4th: A Behavioral Emphasis*. Champaign, IL: Human Kinetics Publishers.

Schmitt, M. B., Justice, L. M., & Logan, J. A. (2017). Intensity of language treatment: contribution to children's language outcomes. *International Journal of Language and Communication Disorders, 52*(2), 155–167.

Schooling, T., Venediktov, R., & Leech, H. (2010). Evidence-based systematic review: Effects of service delivery on the speech and language skills of children from birth to 5 years of age. Retrieved from http://www.asha.org/uploadedFiles/EBSR-Service-Delivery.pdf

Semel, E., Wiig, E., & Secord, W. (2006). Clinical evaluation of language fundamentals fourth edition, Australian standardised edition. Sydney, Australia: Pearson Inc.

Shahin, M., Ahmed, B., Parnandi, A., Karappa, V., McKechnie, J., Ballard, K. J., & Gutierrez-Osuna, R. (2015). Tabby Talks: An automated tool for the assessment of childhood apraxia of speech. *Speech Communication, 70*, 49–64.

Shriberg, L. D. (1994). Five subtypes of developmental phonological disorders. *Clinics in Communication Disorders, 4*(1), 38–53.

Shriberg, L. D., Fourakis, M., Hall, S. D., Karlsson, H. B., Lohmeier, H. L., McSweeny, J. L., . . . Wilson, D. L. (2010). Extensions to the Speech Disorders Classification System (SDCS). *Clinical Linguistics & Phonetics, 24*(10), 795–824.

Shriberg, L. D., Gruber, F. A., & Kwiatkowski, J. (1994). Developmental phonological disorders III. Long-term speech-sound normalization. *Journal of Speech, Language, and Hearing Research, 37*(5), 1151–1177.

Shriberg, L. D., Kwiatkowski, J., & Gruber, F. A. (1994). Developmental phonological disorders II. Short-term speech-sound normalization. *Journal of Speech, Language, and Hearing Research, 37*(5), 1127–1150.

Shriberg, L. D., Lohmeier, H. L., Strand, E. A., & Jakielski, K. J. (2012). Encoding, memory, and transcoding deficits in Childhood Apraxia of Speech. *Clinical Linguistics & Phonetics, 26*(5), 445–482.

Silverman, F. H., & Falk, S. M. (1992). Attitudes of teenagers toward peers who have a single articulation error. *Language, Speech, and Hearing Services in Schools, 23*(2), 187–187.

Silverman, F. H., & Paulus, P. G. (1989). Peer reactions to teenagers who substitute /w/ for /r/. *Language, Speech, and Hearing Services in Schools, 20*(2), 219–221.

Singh, P., & Trivedi, S. (2016). Therapy outcome of combined treatment approach in Childhood Apraxia of Speech (CAS). *Asia Pacific Journal of Research, 1*(46), 34–45.

Skeat, J., Wake, M., Ukoumunne, O. C., Eadie, P., Bretherton, L., & Reilly, S. (2014). Who gets help for pre-school communication problems? Data from a prospective community study. *Child Care Health Development, 40*(2), 215–222. https://doi.org/10.1111/cch.12032

Skelton, S. L., & Hagopian, A. L. (2014). Using randomized variable practice in the treatment of childhood apraxia of speech. *American Journal of Speech-Language Pathology, 23*, 599–611.

Skinder-Meredith, A. (2001). Differential diagnosis: Developmental apraxia of speech and phonologic delay. *Augmentative Communication News, 14*, 5–8.

Smith-Lock, K., Leitão, S., Lambert, L., Prior, P., Dunn, A., Cronje, J., . . . Nickels, L. (2013). Daily or weekly? The role of treatment frequency in the effectiveness of grammar treatment for children with specific language impairment. *International Journal of Speech-Language Pathology, 15*(3), 255–267.

Snowling, M., & Stackhouse, J. (1983). Spelling performances of children with developmental verbal dyspraxia. *Developmental Medicine and Child Neurology, 25*(4), 430–437. https://doi.org/10.1111/j.1469-8749.1983.tb13787.x

Speech Pathology Australia. (2011). Competency-based Occupational Standards for Speech Pathologists. Retrieved from https://www.speechpathologyaustralia.org.au/spaweb/Document_Management/Public/CBOS.aspx

Speech Pathology Australia. (2014a). An inquiry into the prevalence of different types of speech, language and communication disorders and speech pathology services in Australia. *Submission to the Community Affairs References Committee of the Senate*.

Speech Pathology Australia. (2014b). Telepractice in Speech Pathology. Retrieved from http://www.speechpathologyaustralia.org.au/spaweb/Document_Management/Public/Position_Statements.aspx

Strand, E. A., & Debertine, P. (2000). The efficacy of integral stimulation intervention with Developmental Apraxia of Speech. *Journal of Medical Speech-Language Pathology, 8*(4), 295–300.

Strand, E. A., & McCauley, R. J. (2008). Differential diagnosis of severe speech impairment in young children. *The ASHA Leader, 13*(10), 10–13.

Strand, E. A., & Skinder, A. (1999). Treatment of Developmental Apraxia of Speech: Integral Stimulation Methods. In A. J. Caruso & E. A. Strand (Eds.), *Clinical Managment of Motor Speech Disorders in Children* (pp. 109–148). New York: Thieme.

Strand, E. A., Stoeckel, R., & Baas, B. (2006). Treatment of severe childhood apraxia of speech: A treatment efficacy study. *Journal of Medical Speech-Language Pathology, 14*(4), 297–307.

Sugden, E., Baker, E., Munro, N., & Williams, A. L. (2016). Involvement of parents in intervention for childhood speech disorders: a review of the evidence. *International Journal of Language and Communication Disorders, 51*(6), 597–625. https://doi.org/10.1111/1460-6984.12247

Sugden, E., Munro, N., Trivette, C., Baker, E., & Williams, A. L. (2017). *The value of home practice for speech sound disorders: What do parents think?* Paper presented at the Speech Pathology Australia National Conference, Sydney.

The PROMPT Instutute. (n.d.). *Prompt training*. Retrieved from http://www.promptinstitute.com/?page=PROMPTTraining

Theodoros, D. (2008). Telerehabilitation for service delivery in speech-language pathology. *Journal of Telemedicine and Telecare, 14*(5), 221–224. https://doi.org/10.1258/jtt.2007.007044

Theodoros, D. (2011). Telepractice in speech-language pathology: The evidence, the challenges, and the future. *Perspectives on Telepractice, 1*(1), 10–21.

Theodoros, D. (2013). Speech-language pathology and telerehabilitation. In S. Kumar & E. R. Cohn (Eds.), *Telerehabilitation* (pp. 311–323). London: Springer-Verlag.

Thomas, D. C., McCabe, P., & Ballard, K. J. (2014). Rapid Syllable Transitions (ReST) treatment for Childhood Apraxia of Speech: The effect of lower dose-frequency. *Journal of Communication Disorders, 51*, 29–42. https://doi.org/10.1016/j.jcomdis.2014.06.004

Thomas, D. C., McCabe, P., & Ballard, K. J. (2017). Combined clinician–parent delivery of rapid syllable transition (ReST) treatment for childhood apraxia of speech. *International Journal of Speech-Language Pathology*. http://www.tandfonline.com/action/showCitFormats?doi=10.1080/17549507.2017.1316423

Thomas, D. C., McCabe, P., Ballard, K. J., & Lincoln, M. (2016). Telehealth delivery of Rapid Syllable Transitions (ReST) treatment for childhood apraxia of speech. *International Journal of Language and Communication Disorders, 51*(6), 654–671. https://doi.org/10.1111/1460-6984.12238

Tierney, C. D., Pitterle, K., Kurtz, M., Nakhla, M., & Todorow, C. (2016). Bridging the gap between speech and language: using multimodal treatment in a child with apraxia. *Pediatrics, 138*(3), e2–e8.

Tindall, L. R. (2013). Implementation and management of a successful telerehabilitation program in speech language pathology. In S. Kumar & E. R. Cohen (Eds.), *Telerehabilitation* (pp. 91–99). London: Springer-Verlag.

Togher, L., Power, E., Rietdijk, R., McDonald, S., & Tate, R. (2012). An exploration of participant experience of a communication training program for people with traumatic brain injury and their communication partners. *Disability and Rehabilitation, 34*(18), 1562–1574.

Vallino-Napoli, L. D., & Reilly, S. (2004). Evidence-based health care: A survey of speech pathology practice. *International Journal of Speech-Language Pathology, 6*(2), 107–112.

Van Riper, C. (1972). *Speech correction*. Englewood Cliffs, NJ: Prentice-Hall.

Velleman, S., Strand, K., Bernthal, J., & Bankson, N. (1994). Developmental verbal dyspraxia. *Child phonology: Characteristics, assessment, and intervention with special populations*, 110–139.

Verdon, S., Wilson, L., Smith-Tamaray, M., & McAllister, L. (2011). An investigation of equity of rural speech-language pathology services for children: A geographic perspective. *International Journal of Speech-Language Pathology, 13*(3), 239–250.

Wade, V. A., & Eliott, J. (2012). The role of the champion in telehealth service development: a qualitative analysis. *Journal of Telemedicine and Telecare, 18*(8), 490–492.

Wambaugh, J. L., Nessler, C., Cameron, R., & Mauszycki, S. C. (2013). Treatment for acquired apraxia of speech: examination of treatment intensity and practice schedule. *American Journal of Speech-Language Pathology, 22*(1), 84–102. https://doi.org/10.1044/1058-0360(2012/12-0025)

Warren, S. F., Fey, M. E., & Yoder, P. J. (2007). Differential treatment intensity research: a missing link to creating optimally effective communication interventions. *Mental Retardation & Developmental Disabilities Research Reviews, 13*(1), 70–77.

Watson, M. M., & Leahy, J. (1995). Multimodal therapy for a child with Development Apraxia of Speech: A case study. *Child Language Teaching and Therapy, 11*(3), 264–272.

Watts Pappas, N., McAllister, L., & McLeod, S. (2016). Parental beliefs and experiences regarding involvement in intervention for their child with speech sound disorder. *Child Language Teaching and Therapy, 32*(2), 223–239. https://doi.org/10.1177/0265659015615925

Watts Pappas, N., McLeod, S., McAllister, L., & McKinnon, D. H. (2008). Parental involvement in speech intervention: a national survey. *Clinical Linguistics & Phonetics, 22*(4–5), 335–344.

Wiig, E. H., Secord, W., & Semel, E. M. (2004). *Clinical Evaluation of Language Fundamentals Preschool, 2nd edition, Australian standardised edition*. Sydney, Australia: Harcourt Assessment.

Williams, P., & Stephens, H. (2004). *Nuffield Centre Dyspraxia Programme: Third Edition*. Windsor, England: The Miracle Factory.

Wilson, L., Lincoln, M., & Onslow, M. (2002). Availability, access, and quality of care: Inequities in rural speech pathology services for children and a model for redress. *International Journal of Speech-Language Pathology, 4*(1), 9–22.

World Health Organization. (2007). *International Classification of Functioning, Disability, and Health: Children & Youth Version: ICF-CY*: World Health Organization.

Zaretsky, E., Velleman, S. L., & Curro, K. (2010). Through the magnifying glass: Underlying literacy deficits and remediation potential in childhood apraxia of speech. International Journal of Speech-Language Pathology, 12(1), 58–68.

Appendices

Appendix 1: Supplementary material from Chapter 4—Probe stimuli

Probe item type	Probe item		Included in probe list for				
	Orthography	Phonemic script	Oliver	Jack	Emily	Luke	Lachlan
2 syllable pseudo word: strong - weak (SW) stress	deeka	/dikə/				Y*	
	farbi	/fabi/				Y*	
	karby	/kabi/				Y*	
	deefa	/difə/				Y*	
	korfa	/kɔfɔ/				Y*	
	korba	/kɔbə/				Y*	
	darfa	/dafə/				Y*	
	bardi	/badi/				Y*	
	kordi	/kɔdi/				Y*	
	deeba	/dibə/				Y*	
	barka	/bakə/				Y	
	beeda	/bidə/				Y	
	farba	/fabə/				Y	
	korda	/kɔdə/				Y	
	feedy	/fidi/				Y	
	keeda	/kidə/				Y	
	darfy	/dafi/				Y	
	dorfa	/dɔfə/				Y	
	fardi	/fadi/				Y	
	fordy	/fɔdi/				Y	
2 syllable pseudo word: weak - strong (WS) stress	kebar	/kəba/				Y*	
	bedee	/bədi/				Y*	
	fedor	/fədɔ/				Y*	
	kefee	/kəfi/				Y*	
	febee	/fəbi/				Y*	
	debor	/dəbɔ/				Y*	
	fekar	/fəka/				Y*	
	fedar	/fəda/				Y*	
	kedee	/kədi/				Y*	
	fekee	/fəki/				Y*	
	befar	/bəfa/				Y	
	bekor	/bəkɔ/				Y	
	kedor	/kədɔ/				Y	
	defee	/dəfi/				Y	
	bekar	/bəka/				Y	
	dekar	/dəka/				Y	
	febar	/fəba/				Y	
	bedar	/bəda/				Y	
	kefor	/kəfɔ/				Y	
	fekor	/fəkɔ/				Y	
	farbeda	/fabədə/	Y*	Y	Y*		Y*
	karbefi	/kabəfi/	Y*	Y	Y	Y	Y*

3 syllable	barkefi	/bakəfi/	Y	Y*	Y*	Y	Y
pseudo word:	deefeba	/difəbə/	Y*	Y	Y*	Y	Y*
SW stress	korfedi	/kɔfədi/	Y	Y	Y*		Y*
	beedafa	/bidəfə/	Y*	Y*	Y		Y
	korbefa	/kɔbəfə/	Y	Y	Y		Y*
	farbekee	/fabəki/	Y*	Y*	Y*	Y	Y
	korbida	/kɔbədə/	Y	Y*	Y		Y
	darfebee	/dafəbi/	Y*	Y	Y*		Y*
	feedika	/fidəkə/	Y	Y*	Y	Y	Y
	fordabee	/fɔdəbi/	Y	Y*	Y*		Y
	keedefi	/kidəfi/	Y*	Y*	Y		Y
	bardefa	/badəfə/	Y*	Y	Y*	Y	Y*
	farbeka	/fabəkə/	Y*	Y*	Y*	Y	Y
	kordefi	/kɔdəfi/	Y	Y	Y	Y	Y*
	deekeba	/dikəbə/	Y	Y	Y		Y*
	deekefa	/dikəfa/	Y	Y	Y*	Y	Y*
	dorfebi	/dɔfəbi/	Y*	Y*	Y		Y
	fardekee	/fadəki/	Y	Y*	Y	Y	Y
3 syllable	bedeeka	/bədikə/	Y*	Y	Y	Y	Y*
pseudo word:	febeeda	/fəbidə/	Y	Y	Y	Y	Y*
WS stress	beforka	/bəfɔkə/	Y*	Y*	Y*	Y	Y
	deborfee	/dəbɔfi/	Y	Y	Y	Y	Y*
	kafordi	/kəfɔdi/	Y*	Y*	Y*	Y	Y
	bekorda	/bəkɔdə/	Y	Y*	Y*	Y	Y
	kedorfi	/kədɔfi/	Y*	Y*	Y*		Y
	defeeka	/dəfikə/	Y*	Y*	Y*		Y
	fekardi	/fəkadi/	Y	Y	Y		Y*
	bekarfi	/bəkafi/	Y*	Y*	Y*		Y
	fedarbi	/fədabi/	Y	Y	Y		Y*
	fekorba	/fəkɔbə/	Y*	Y*	Y*	Y	Y
	fedorki	/fədɔki/	Y	Y	Y	Y	Y*
	dekarba	/dəkabə/	Y*	Y*	Y*		Y
	befardi	/bəfadi/	Y*	Y*	Y*		Y
	kedeefa	/kədifə/	Y	Y	Y		Y*
	debarfi	/dəbafi/	Y	Y*	Y*	Y	Y
	fekeeba	/fəkibə/	Y	Y	Y		Y*
	febarki	/fəbaki/	Y*	Y	Y	Y	Y*
	kefeeda	/kəfidə/	Y	Y	Y		Y*
Phrase	I found a _____		Y	Y	Y		Y
containing	I want a _____		Y	Y	Y		Y
treated SW	I went to the _____		Y	Y	Y		Y
pseudo word	Who took the _____?		Y	Y	Y		Y
	He bought a _____		Y	Y	Y		Y
Phrase containing	Where did you put my _____?		Y	Y	Y		Y
	I dropped the _____		Y	Y	Y		Y

treated WS	Look at the _____	Y	Y	Y		Y
pseudo word	She took a big _____	Y	Y	Y		Y
	Can I have a _____?	Y	Y	Y		Y
Phrase containing	Where did you put my _____?	Y	Y	Y		Y
untreated SW	I dropped the _____	Y	Y	Y		Y
pseudo word	Look at the _____	Y	Y	Y		Y
	She took a big _____	Y	Y	Y		Y
	Can I have a _____?	Y	Y	Y		Y
Phrase	I found a _____	Y	Y	Y		Y
containing	I want a _____	Y	Y	Y		Y
untreated WS	I went to the _____	Y	Y	Y		Y
pseudo word	Who took the _____	Y	Y	Y		Y
	He bought a _____	Y	Y	Y		Y
Real word	tiger				Y	
with 2	garden				Y	
syllables:	teacher				Y	
SW stress	sugar				Y	
	table				Y	
Real word	between				Y	
with 2	forget				Y	
syllables:	begin				Y	
WS stress	today				Y	
	correct				Y	
Real word	barbecue	Y	Y	Y		Y
with 3	butterfly	Y	Y	Y		Y
syllables:	carpenter	Y	Y	Y	Y	Y
SW stress	hibernate	Y	Y	Y		Y
	kangaroo	Y	Y	Y	Y	Y
	photograph	Y	Y	Y		Y
	broccoli	Y	Y	Y		Y
	cavity	Y	Y	Y		Y
	cardigan	Y	Y	Y	Y	Y
	fisherman	Y	Y	Y	Y	Y
Real word	detergent	Y	Y	Y	Y	Y
with 3	potato	Y	Y	Y	Y	Y
syllables:	tomato	Y	Y	Y		Y
WS stress	mechanic	Y	Y	Y	Y	Y
	recorder	Y	Y	Y		Y
	banana	Y	Y	Y		Y
	toboggan	Y	Y	Y		Y
	tomorrow	Y	Y	Y	Y	Y
	magician	Y	Y	Y		Y
	karate	Y	Y	Y	Y	Y
/r/ control	rice	Y		Y		
word	ribbon	Y		Y		

	radio	Y		Y		
	red	Y		Y		
	ring	Y		Y		
	dragon	Y		Y		
	earring	Y		Y		
	Fairy	Y		Y		
	pirate	Y		Y		
	giraffe	Y		Y		
/s/ control	soap		Y			
word	sock		Y			
	salt		Y			
	stamp		Y			
	spoon		Y			
	school		Y			
	bus		Y			
	face		Y			
	juice		Y			
	horse		y			
/l/ cluster	plane				Y	
control word	plum				Y	
	flood				Y	
	fly				Y	
	clap				Y	
	clock				Y	
	glue				Y	
	glide				Y	
	black				Y	
	blue				Y	
/s/ cluster	skate					Y
word	school					Y
	spear					Y
	spider					Y
	spot					Y
	sleigh					Y
	slow					Y
	stamp					Y
	stick					Y
	stop					Y

Appendix 2: Supplementary material from Chapter 5—Probe stimuli

Probe item type	Probe item		Included in probe list for				
	Orthography	Phonemic script	Stacey	Ben	Eric	Matthew	Julian
2 Syllable	farfy	fafi					Y
pseudo word:	keeka	kikə					Y
same	borbi	bɔbi					Y
consonant SW	deedy	didə					Y
stress	karka	kakə					Y
	barba	badə					Y
	forfa	fɔfə					Y
	korky	kɔki					Y
	dardy	dadi					Y
	feefa	fifə					Y
2 Syllable	bebor	bəbɔ					Y
pseudo word:	kekar	kəka					Y
same	dedee	dədi					Y
consonant WS	fefor	fəfɔ					Y
stress	bebar	bəba					Y
	fefee	fəfi					Y
	dedar	dəda					Y
	kekor	kəkɔ					Y
	fefar	fəfɔ					Y
	kekee	kəki					Y
2 syllable	fardi	fadi					Y*
pseudo word:	karba	kabə					Y*
strong - weak	barky	baki					Y*
(SW) stress	deeba	dibə					Y*
	korfi	kɔfi					Y*
	beeda	bidə					Y*
	korba	kɔbə	Y	Y^	Y	Y	Y*
	farbi	fabi					Y*
	kardi	kadi					Y*
	darfa	dafə	Y	Y^	Y	Y	Y*
	feeda	fidə	Y	Y^	Y	Y	Y
	dorky	dɔki	Y	Y^	Y	Y	Y
	keeda	kidə	Y	Y^	Y	Y	Y
	bardi	badi	Y	Y^	Y	Y	Y
	farba	fabə	Y	Y^	Y	Y	Y
	kordi	kɔdi	Y	Y^	Y	Y	Y
	deefa	difə	Y	Y^	Y	Y	Y
	darky	daki	Y	Y^	Y	Y	Y
	dorfi	dɔfi					Y
	farda	fadə					Y
2 syllable	bedee	bədi					Y*
pseudo word:	febee	fəbi					Y*

weak - strong	befar	bəfa					Y*
(WS) stress	debor	dəbɔ					Y*
	kefar	kəfa					Y*
	bekor	bəkɔ	Y	Y^	Y	Y	Y*
	kefor	kəfɔ	Y	Y^	Y	Y	Y*
	defee	dəfi	Y	Y^	Y	Y	Y*
	fekar	fəka	Y	Y^	Y	Y	Y*
	bekar	bəka	Y	Y^	Y	Y	Y*
	fedar	fəda	Y	Y^	Y	Y	Y
	fekor	fəkɔ	Y	Y^	Y	Y	Y
	fedor	fədɔ	Y	Y^	Y	Y	Y
	dekar	dəka	Y	Y^	Y	Y	Y
	befar	bəfa	Y	Y	Y	Y	Y
	kedee	kədi					Y
	debar	dəba					Y
	fekee	fəki					Y
	febar	fəba					Y
	kefee	kəfi					Y
3 syllable	farbeda	/fabədə/	Y	Y*	Y*	Y*	
pseudo word:	karbefi	/kabəfi/	Y	Y*	Y*	Y*	
SW stress	barkefi	/bakəfi/	Y*	Y	Y	Y*	
	deefeba	/difəbə/	Y	Y	Y*	Y	
	korfedi	/kɔfədi/	Y	Y*	Y*	Y*	
	beedafa	/bidəfə/	Y*	Y*	Y*	Y	
	korbefa	/kɔbəfə/	Y	Y	Y	Y*	
	farbekee	/fabəki/	Y*	Y*	Y*	Y	
	korbida	/kɔbədə/	Y*	Y	Y	Y*	
	darfebee	/dafəbi/	Y	Y*	Y*	Y	Y
	feedika	/fidəkə/	Y*	Y	Y	Y*	Y
	fordabee	/fɔdəbi/	Y*	Y	Y	Y	Y
	keedefi	/kidəfi/	Y*	Y*	Y*	Y	Y
	bardefa	/badəfə/	Y	Y*	Y*	Y	Y
	farbeka	/fabəkə/	Y*	Y*	Y*	Y	Y
	kordefi	/kɔdəfi/	Y	Y	Y	Y*	Y
	deekeba	/dikəbə/	Y	Y	Y	Y	Y
	deekefa	/dikəfa/	Y	Y	Y	Y	Y
	dorfebi	/dɔfəbi/	Y*	Y*	Y*	Y	Y
	fardekee	/fadəki/	Y*	Y*	Y	Y*	
3 syllable	bedeeka	/bədikə/	Y	Y	Y	Y*	
pseudo word:	febeeda	/fəbidə/	Y	Y	Y	Y*	
WS stress	beforka	/bəfɔkə/	Y*	Y*	Y*	Y	
	deborfee	/dəbɔfi/	Y	Y	Y	Y*	
	kafordi	/kəfɔdi/	Y*	Y*	Y*	Y	
	bekorda	/bəkɔdə/	Y*	Y*	Y*	Y	
	kedorfi	/kədɔfi/	Y*	Y*	Y*	Y*	
	defeeka	/dəfikə/	Y*	Y*	Y*	Y*	

	fekardi	/fəkadi/	Y	Y	Y	Y*	
	bekarfi	/bəkafi/	Y*	Y*	Y*	Y	
	fedarbi	/fədabi/	Y	Y	Y	Y*	Y
	fekorba	/fəkɔbə/	Y*	Y*	Y*	Y	Y
	fedorki	/fədɔki/	Y	Y	Y	Y	Y
	dekarba	/dəkabə/	Y*	Y*	Y*	Y	Y
	befardi	/bəfadi/	Y*	Y*	Y*	Y	Y
	kedeefa	/kədifə/	Y	Y	Y	Y*	Y
	debarfi	/dəbafi/	Y*	Y*	Y*	Y	Y
	fekeeba	/fəkibə/	Y	Y	Y	Y*	Y
	febarki	/fəbaki/	Y	Y	Y	Y*	Y
	kefeeda	/kəfidə/	Y	Y	Y	Y	Y
4 syllable	fardekeeba	/fadəkibə/	Y	Y	Y	Y	
pseudo words:	keedaforby	/kidəfɔbi/	Y	Y	Y	Y	
SW stress	beekadarba	/bikədabə/	Y	Y	Y	Y	
	forbedarky	/fɔbədaki/	Y	Y	Y	Y	
	korfedeeka	/kɔfədikə/	Y	Y	Y	Y	
	farbekardee	/fabəkədi/	Y	Y	Y	Y	
	kordefeeba	/kɔdəfibə/	Y	Y	Y	Y	
	deekebarfi	/dikəbafi/	Y	Y	Y	Y	
	deekeforba	/dikəfɔbə/	Y	Y	Y	Y	
	dorfebarka	/dɔfəbakə/	Y	Y	Y	Y	
4 syllable	bekeefidor	/bəkifədɔ/	Y	Y	Y	Y	
pseudo words:	kedarfebee	/kədafəbi/	Y	Y	Y	Y	
WS stress	befordikar	/bəfɔdəka/	Y	Y	Y	Y	
	fedarkebee	/fədakəbi/	Y	Y	Y	Y	
	debarkifee	/dəbakəfi/	Y	Y	Y	Y	
	kedeefebor	/kədifəbɔ/	Y	Y	Y	Y	
	defarbekee	/dəbafəki/	Y	Y	Y	Y	
	fekeebador	/fəkibədɔ/	Y	Y	Y	Y	
	febarkidee	/fəbakədi/	Y	Y	Y	Y	
	kefeedabar	/kəfidəba/	Y	Y	Y	Y	
Phrase	I found a _____		Y	Y	Y	Y	Y
containing	I want a _____		Y	Y	Y	Y	Y
treated SW	I went to the _____		Y	Y	Y	Y	Y
pseudo word	Who took the _____?		Y	Y	Y	Y	Y
	He bought a _____		Y	Y	Y	Y	Y
Phrase	Where did you put my		Y	Y	Y	Y	Y
containing	_____?						
treated WS	I dropped the _____		Y	Y	Y	Y	Y
pseudo word	Look at the _____		Y	Y	Y	Y	Y
	She took a big _____		Y	Y	Y	Y	Y
	Can I have a _____?		Y	Y	Y	Y	Y
Phrase	Where did you put my		Y	Y	Y	Y	Y
containing	_____?						
	I dropped the _____		Y	Y	Y	Y	Y

untreated SW	Look at the _____	Y	Y	Y	Y	Y
pseudo word	She took a big _____	Y	Y	Y	Y	Y
	Can I have a _____?	Y	Y	Y	Y	Y
Phrase	I found a _____	Y	Y	Y	Y	Y
containing	I want a _____	Y	Y	Y	Y	Y
untreated WS	I went to the _____	Y	Y	Y	Y	Y
pseudo word	Who took the _____	Y	Y	Y	Y	Y
	He bought a _____	Y	Y	Y	Y	Y
Phrase	I found a _____ and a _____				Y	
containing	I want a _____ not a _____				Y	
two treated	She held a _____ and a _____				Y	
pseudo words	I went to the _____ not the _____				Y	
	Who took the _____ and the _____ ?				Y	
	I found a _____ not a _____				Y	
	I want a _____ and a _____				Y	
	She held a _____ a not a _____				Y	
	I went to the _____ not the _____				Y	
	Who took the _____ and the _____?				Y	
Phrase	I found a _____ not a _____				Y	
containing	I want a _____ and a _____				Y	
two untreated	She held a _____ and a _____				Y	
pseudo words	I went to the _____ not the _____				Y	
	Who took the _____ and the _____?				Y	
	I found a _____ not a _____				Y	
	I want a _____ not the _____				Y	
	She held a _____ not a _____				Y	
	I went to the _____ and the _____				Y	
	Who took the _____ and the _____?				Y	
Real word	feet					Y
with 1	fork					Y
syllable:	bed					Y
CVC structure	gate					Y
	take					Y
	feed					Y
	cave					Y

	dig					Y
	book					Y
	peak					Y
Real word	frog					Y
with 1	blood					Y
syllable:	track					Y
CCVC	flap					Y
structure	drum					Y
	drop					Y
	tram					Y
	plane					Y
	clap					Y
	flag					Y
Real word	tiger	Y	Y	Y	Y	Y
with 2	garden	Y	Y	Y	Y	Y
syllables:	teacher	Y	Y	Y	Y	Y
SW stress	perfect	Y	Y	Y	Y	Y
	rocket	Y	Y	Y	Y	Y
	package	Y	Y	Y	Y	Y
	sugar	Y	Y	Y	Y	Y
	table	Y	Y	Y	Y	Y
	digger	Y	Y	Y	Y	Y
	number	Y	Y	Y	Y	Y
Real word	giraffe	Y	Y	Y	Y	Y
with 2	between	Y	Y	Y	Y	Y
syllables:	Japan	Y	Y	Y	Y	Y
WS stress	forget	Y	Y	Y	Y	Y
	begin	Y	Y	Y	Y	Y
	behave	Y	Y	Y	Y	Y
	conduct	Y	Y	Y	Y	Y
	today	Y	Y	Y	Y	Y
	correct	Y	Y	Y	Y	Y
	forgive	Y	Y	Y	Y	Y
Real word	barbecue	Y	Y	Y	Y	Y
with 3	butterfly	Y	Y	Y	Y	Y
syllables:	carpenter	Y	Y	Y	Y	Y
SW stress	hibernate	Y	Y	Y	Y	Y
	kangaroo	Y	Y	Y	Y	Y
	photograph	Y	Y	Y	Y	Y
	broccoli	Y	Y	Y	Y	Y
	cavity	Y	Y	Y	Y	Y
	cardigan	Y	Y	Y	Y	Y
	fisherman	Y	Y	Y	Y	Y
Real word	detergent	Y	Y	Y	Y	Y
with 3	potato	Y	Y	Y	Y	Y
syllables:	tomato	Y	Y	Y	Y	Y

WS stress	mechanic	Y	Y	Y	Y	Y
	recorder	Y	Y	Y	Y	Y
	banana	Y	Y	Y	Y	Y
	toboggan	Y	Y	Y	Y	Y
	tomorrow	Y	Y	Y	Y	Y
	magician	Y	Y	Y	Y	Y
	karate	Y	Y	Y	Y	Y
Real word	caterpillar	Y	Y	Y	Y	
with 4	mathematics	Y	Y	Y	Y	
syllables:	calculator	Y	Y	Y	Y	
SW stress	kookaburra	Y	Y	Y	Y	
	combination	Y	Y	Y	Y	
	commentator	Y	Y	Y	Y	
	decorator	Y	Y	Y	Y	
	germination	Y	Y	Y	Y	
	macaroni	Y	Y	Y	Y	
	babycino	Y	Y	Y	Y	
Real word	Canadian	Y	Y	Y	Y	
with 4	majority	Y	Y	Y	Y	
syllables:	community	Y	Y	Y	Y	
WS stress	thermometer	Y	Y	Y	Y	
	pedometer	Y	Y	Y	Y	
	traditional	Y	Y	Y	Y	
	particular	Y	Y	Y	Y	
	divisible	Y	Y	Y	Y	
	photographer	Y	Y	Y	Y	
	perimeter	Y	Y	Y	Y	
Total items		160	160	160	180	160

Appendix 3: Supplementary material from Chapter 5—Effect sizes across ReST studies

Study	Participant	Treated behaviour	Effect size
McCabe et al., (2014) SLP student clinicians delivered treatment, 4 times per week for 3 weeks, in the clinic	P1	2 syllable pw	8.25
	P2	2 syllable pw	4.13
	P3	2 syllable pw	4.41
	P4	2 syllable pw	3.30
Thomas et al., (2014) SLP student clinicians delivered treatment, 2 times per week for 6 weeks, in the clinic	M1	3 syllable pw	8.18
		Phrases with 1 3 syllable pw	4.12
	F1	3 syllable pw	11.21
	M2	3 syllable pw	3.44
	F2	2 syllable pw	4.95
Thomas et al., (2016) SLPs and student clinicians delivered treatment, 4 times per week for 3 weeks, via video conference	Oliver	3 syllable pw	9.23
	Luke	2 syllable pw	9.82
	Jack	3 syllable pw	3.85
		Phrases with 1 3 syllable pw	7.76
	Emily	3 syllable pw	4.65
		Phrases with 1 3 syllable pw	5.98
	Lachlan	3 syllable pw	10.09
The present study: Combined SLP student clinician-and parent-delivery, 4 sessions a week for 3 weeks, 6 sessions in the clinic and 6 sessions at home	Stacey	3 syllable pw	6.40
	Eric	3 syllable pw	7.98
		Phrases with 1 3 syllable pw	6.05
	Ben	3 syllable pw	2.76
		2 syllable pw	8.52
	Matt	Phrases with 1 3 syll. pw	4.40
		Phrases with 2 3 syll. pw	2.75
	Julian	2 syllable pw	2.48

Appendix 4: Supplementary material from Chapter 5—Comparison of treatment fidelity, perceptual reliability, child, and parent factors, with treatment, generalisation, and maintenance effects

		Stacey	**Eric**	**Ben**	**Matt**	**Julian**
Person delivering treatment	Parent Clinician	Andy Claire	Alex Lisa	Sam Hannah	Chris Hannah	Morgan Claire
Child's treatment outcome	Treatment effect?	Yes (1 of 1)	Yes (2 of 2)	Partial (1 of 2)	Partial (1 of 2)	No (0 of 1)
	Generalisation effect?	Yes (7 of 7)	Partial (5 of 7)	Partial* (1 of 7)	Partial (1 of 7)	No (0 of 6)
	Maintenance of gains for 4 months?	Yes (↑ for 4 of 7)	Yes (↑ for 1 of 7)	Yes (↑ for 1 of 7)^	Yes	N/A
Child variables	Receptive language	WNL	WNL	<NL mod.	WNL	<NL mod.
	Expressive language	WNL	WNL	<NL sev.	WNL	<NL sev.
	PPVT score	>NL	WNL	WNL	WNL	<NL mod.
	PCC	80	94	25	93	54
	Hypernasality	Yes	No	No	Yes	Yes
Parent variables	Highest education level	Year 10	Bachelor degree	Year 12	Bachelor degree	Bachelor degree
	History of speech, language or literacy difficulties?	Yes - Spelling difficulty - childhood	No	No	Yes - Confusion of written b and d	Yes - Reading difficulty - high school
Treatment fidelity	Clinician-delivered sessions	91%	98%	92%	89%	92%
	Parent –delivered sessions	83%	86%	88%	41%	89%
Perceptual judgement reliability	Clinician-delivered sessions	87%	91%	94%	87%	83%
	Parent-delivered sessions	73%	83%	87%	65%	71%

*Ben's probe list was generated for a child being treated on 3 syllable pseudo words. Because he had low success with this behavior his treatment goal was changed to 2 syllable pseudo words from session 3. His probe list did not allow the evaluation of generalization to similar pseudo words and less complex items ^ Ben showed an improvement at the 4-month follow-up point. He received ReST therapy in the community following the one-month follow-up point.