ReaDySpeech for people with dysarthria after stroke:

a feasibility study

A thesis submitted to the University of Manchester for the degree of

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Word count: 44,840

List of Abbreviations

BI: Barthel Index

COAST: communication after stroke scale

CT: computer tomography

CTU: clinical trials unit

DIP: Dysarthria impact profile

Dysarthria TOMs: Therapy Outcome Measure specific to dysarthria

EQ-5D-5L: Euroquol quality of life scale

FDA II: Frenchay dysarthria assessment 2nd edition

ISRCTN: numerical identification of randomised controlled trials

ITT: intention-to-treat

MRI: magnetic resonance imaging

MRS: modified Rankin scale

NHS: UK National Health Service

NIHSS: National Institutes of Health Stroke Scale

PPI or PCPI: Patient, public or patient, carer, public involvement

RCT: randomised controlled trial

SLT: speech and language therapist

WHO: World Health Organisation

Thesis Abstract

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ReaDySpeech for people with dysarthria after stroke: a feasibility study

Dysarthria describes the impaired speech intelligibility caused by weakness of muscles involved in speech following stroke. This is a common consequence of stroke and can have a detrimental impact on self-confidence leading to social isolation for many. There is limited evidence for dysarthria intervention but we know that research into speech difficulties after stroke is a priority for stroke survivors. An online speech rehabilitation programme was developed, ReaDySpeech, with the potential to offer improved quality of independent practice, increased intensity of practice and the ability to record interaction. The research presented in this thesis aimed to systematically examine the existing evidence base, to carry out some preliminary acceptability work on ReaDySpeech, and implement a feasibility trial.

The initial study was a Cochrane systematic review of the effectiveness of interventions for people with non-progressive dysarthria after stroke or other adult-acquired brain injury. This found insufficient evidence to know whether dysarthria intervention is effective or not. This led to a study of early acceptability work for ReaDySpeech and whether there were any technical barriers to use. This found no significant technical barriers other than lack of Wi-Fi and it was acceptable to participants and therapists. This enabled a progression to a feasibility trial following amendments and improvements to the protocol and ReaDySpeech itself. The feasibility trial found recruitment, retention and the intervention were all feasible to carry out during a trial. Further in-depth consideration of the findings indicates more work is needed to widen recruitment and to develop the intervention, comparator and methodology of a future trial for this to be a success with valid clinical implications. This thesis reports this body of work and discusses potential future directions for dysarthria research.

Declaration

No portion of the work referred to in the thesis has been submitted in support of an application for another degree or qualification of this or any other university or other institute of learning.

Copyright Statement

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The Author

Claire Mitchell qualified with a BSc (Hons) degree in Speech Pathology and Therapy in 1995, from Manchester Metropolitan University. She has worked clinically as a speech and language therapist at Salford Royal Hospitals NHS Trust and Manchester Royal Infirmary ever since. Claire was awarded her MPhil in 2000 and went on to combine clinical work with working as a senior clinical lecturer (teaching focus) at The University of Manchester in 2003. In 2014 she was awarded a National Institute for Health Research Doctoral Research Fellowship.

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Thesis format

This thesis starts with a broad introduction to dysarthria and background to the study. This is then followed by five chapters, chapters two, three, four, five and six, which are written in a journal format. These have either been published, submitted or are ready for submission. The main themes of the thesis and potential future research directions are discussed in chapter seven, the discussion.

The inclusion of publication style chapters can lead to duplication with other sections of the thesis and this is acknowledged within the guidelines for this thesis format. The chapters are in the most logical order for the thesis but were not necessarily published in this order. Chapter three was published in 2016, so it does not refer to the Cochrane review, chapter two, which was published in 2017.

Chapter 1 Introduction

1.1 Overview of thesis structure

This introductory chapter will outline the background to the thesis. This will start with describing dysarthria after stroke, what it is and what is known about it. This will lead on to what interventions we currently offer to people with dysarthria after stroke and what evidence supports this. We will then discuss what we know about motor learning more widely in stroke rehabilitation and what lessons we can use from this literature to develop new approaches to intervention. The potential use of technology in rehabilitation will also be reviewed, specifically the evidence we have for this in similar stroke populations such as those with aphasia.

This broad scoping of the literature then leads into chapter 2 for the recently published Cochrane systematic review of interventions for dysarthria due to stroke and other adult-acquired, non-progressive brain injury. Chapter 3 explains the background to the development of the online programme ReaDySpeech. This first study examined the acceptability of ReaDySpeech and any technical barriers. The protocol for the feasibility randomised controlled trial for ReaDySpeech is outlined in chapter 4. The main empirical chapter reporting the findings from this trial is found in chapter 5. A more in-depth analysis of the interventions carried out as part of this trial are presented in chapter 6. The Discussion chapter in chapter 7 draws together the main research findings and themes which have emerged throughout the thesis, and provides a synthesis of these together with a consideration of ways forward for future research.

1.2 Dysarthria after stroke

Speech problems are widely recognised as a consequence of stroke and there are two main (but not exclusive) forms of clinical presentation affecting communication. Aphasia can be defined as a language disorder typically marked by impaired understanding or production of language, with symptoms such as word retrieval problems and/or deficits in sentence production very evident. Dysarthria in contrast refers to impairments in the neuromuscular control for speech which affects the precision of clarity and intelligibility of speech production (Darley et al., 1969).

Of the 150,000 individuals in the UK to survive a stroke each year (Stroke Association, 2014), approximately 20-30% (Lubart et al., 2005; Lawrence et al., 2001; Warlow, 2001), will experience dysarthria. We know that stroke survivors rate the need for communication research highly and the importance of communication for stroke survivors was illustrated, when the James Lind Alliance (Pollock et al., 2012) identified speech problems as one of their top ten priorities for stroke research. Interestingly the much more widely researched topic of aphasia after stroke has a similar incidence (Lubart et al., 2005; Tsouli et al., 2009; Ali et al., 2015). Although this study relates specifically to dysarthria, dysarthria co-occurs with aphasia in around 10% of strokes (Trapl et al., 2004). These figures suggest that up to 30,000-45,000 people in the UK have stroke related dysarthria although more detailed data of incidence and prevalence as well as the natural history of dysarthria is not available (Brady et al., 2011b). Despite the significant number of people who have dysarthria following stroke, there is limited research into the topic and the impact it has on activity and participation levels of functioning (WHO, 2007).

Dysarthria can be defined most comprehensively as a neurological motor speech impairment caused by slow, weak, imprecise, and/or poorly coordinated movements of the speech musculature. This can involve breathing, voice production, resonance and/or oral articulation (Yorkston, 1996). Dysarthric speech typically sounds less intelligible because of poor oral control of articulator muscles, particularly the tongue. It can also be quiet and/or underpowered and lack expressiveness because of weak use of the voice. Dysarthria includes a wide severity range after stroke with some patients having no useful speech, unintelligible to the listener, while at the milder end there may be lapses in speech accuracy, but speech is generally intelligible. There is research into progressive dysarthria that suggests intelligibility of speech does not necessarily predict conversation competence (Bloch and Tuomainen, 2017) and it may be that this is the case in stroke.

There is a paucity of research considering the impact dysarthria has on activity and participation (Brady et al., 2011b). There is significant evidence that stroke can have a devastating effect on an individual's self-identity and social interaction which directly leads to a reduced quality of life post stroke (Clarke and Black, 2005; Hommel et al., 2009). Ultimately, dysarthria is a communication disability and this inevitably directly impacts on an individual's ability to convey a sense of self and maintain social interaction at previous levels (Dalemans et al., 2008). This can have a detrimental impact on well-being (Haslam et al., 2008), cognition (Glymour et al., 2008) and functional outcomes (Kuelzer et al., 2008). Aphasia is more widely researched than dysarthria as shown in the number of studies found in the respective Cochrane reviews, 57 in aphasia (Brady et al., 2016) and five in the dysarthria review (Mitchell et al., 2017a). Much of the evidence around the impact of communication impairment

following stroke comes from the aphasia literature. These restrictions to social participation (Pallesen, 2014) resulting in increased isolation are likely to be similar for dysarthria.

It is clear that dysarthria, specifically, can have an impact on patients that goes beyond the communication impairments. It negatively affects their sense of identity, self-image, social participation, psychological well-being and level of outcome (Tilling et al., 2001; Brady et al., 2011b; Dickson et al., 2008). This may relate to the relatively frank nature of dysarthria, where speech tends to be consistently and often obviously affected, as well as the possible overlap with related symptoms such as facial asymmetry. It is important to note that there is some evidence to suggest that the impact of dysarthria on patients is not dependant on severity of dysarthria (Brady et al., 2011b). This work by Brady (2011b) looking at the impact of stroke-related dysarthria on social participation found that the impact on individuals could be just as severe for those with a mild dysarthria to try to return to work or pick up social roles with high communication demands as for those with a more severe degree of dysarthria. Whether an individual is able to return to their previous roles and activities is a significant factor in quality of life following stroke and may support the need to further investigate the impact of dysarthria on an individual as part of routine clinical assessment (Clarke and Black, 2005). In summary, there is still a lot to be learnt about dysarthria after stroke, particularly in relation to incidence and prevalence, the natural history of recovery and the wider impact of social participation.

1.3 Dysarthria intervention

Although speech and language therapy (SLT) input is highly valued by people with dysarthria after stroke (Brady et al., 2011b; Mackenzie et al., 2013), the evidence base for the treatment of dysarthria after stroke is limited by a lack of adequately powered well controlled trials (Sellars et al., 2005). This review was recently updated, see chapter 3 (Mitchell et al., 2017a), when five trials were selected but these were all small studies, not powered to show effectiveness so did not change clinical practice or guidelines.

Dysarthria guidelines (RCSLT, 2009 and NICE, 2013) recommend that speech and language therapy intervention should address all dimensions of the International Classification of Functioning, Disability, and Health Framework (World Health Organization 2001). The management of dysarthria is guided by three distinct approaches that address impairment, activity and participation:

i) physiological or impairment level intervention which aims to directly work on the strength, speed and/or function of the impaired musculature to change specific aspects of the function of respiration, resonance, phonation, articulation and prosody;

ii) compensatory approaches to promote activity and participation by minimising disability and promoting intelligibility by working on rate or volume control and other environmental modifications;

iii) augmentative approaches are used when speech does not meet communication needs and may range from mobile computer text-to-speech aids to an alphabet chart working at activity and participation level.

Dysarthria intervention in clinical practice usually draws on these approaches outlined above which are based on the best available evidence there is. This includes clinical case series studies and expert opinion to guide clinical best practice. This is usually a combination of impairment level exercises such as non-speech oro-motor or breathing exercises and activity level intervention such as advice on slowing the rate of speech or increasing volume. Intervention can also address participation level activities to support people psychologically and to return to everyday activity. The most recent National Clinical Guidelines for Stroke (ICSWP, 2016) reflect the lack of evidence for dysarthria intervention with greater emphasis on the importance of delivering activity and participation level interventions. Interestingly this advice is at odds with what speech and language therapists deliver in UK clinical practice. A recent UK-wide survey of dysarthria intervention for stroke (Miller and Bloch, 2017) indicated that UK speech and language therapists focus on impairment based intervention with limited participation level work. Furthermore many of the existing studies on dysarthria focus on physiological (impairment level) intervention such as oro-motor exercises and volume control using narrow speech specific outcome measures (Mackenzie, 2011; Palmer and Enderby, 2007; Nemec and Cohen, 1984; Tamplin, 2008; Kim and Jo, 2013; Ray, 2002).

There are many unanswered questions relating to dysarthria intervention. The key question is whether dysarthria intervention is effective. In order to answer this, the components of this complex intervention need to be understood. It may be that dysarthria intervention should include impairment, activity and participation foci or perhaps it is only elements of this range of foci which are beneficial. The other aspect of the intervention relates to the recovery process and whether different aspects of

intervention may be more beneficial at different time points for each individual. Intensity of intervention is a critically important variable in conducting effectiveness research which has had little attention to date.

1.3.1 Motor learning in stroke rehabilitation

The lack of research in dysarthria, a motor speech impairment, could however benefit from insights provided by the wider literature around motor impairment more generally after stroke. The most common physical symptom following stroke is a hemiparesis, which results in weakness on one side of the body typically reducing function and movement of the face, arm and/or leg (Warlow, 2001). The weakness is contralateral to the side of the brain affected by the stroke and will vary in severity of paresis. There is a relationship between the degree of weakness and the extent of the dysfunction which is why rehabilitation commonly attempts to strengthen movement with the intended outcome improving functional recovery (Jorgensen et al., 1995). A systematic review of rehabilitation studies made it clear that they considered studies of recovery and compensation to be the same process of recovery as it is not possible or necessary to distinguish between them (Langhorne et al., 2009). This review looked at the evidence from intervention trials and found that high-intensity, repetitive task-specific practice with feedback were key elements for motor learning, and these features reduced the degree of motor impairment and improved function after stroke (Langhorne et al., 2009). This approach to stroke rehabilitation has been reflected in recent stroke clinical guidelines (ICSWP, 2016), but the review concluded that more work is needed to identify particular treatment types to meet individuals' specific needs (Langhorne et al., 2009).

There is a significant body of neuroscience research that demonstrates the ability of the brain and the central nervous system to reorganise itself after injury such as stroke, which is known as neuroplasticity (Rossini et al., 2003), and within the cortex, cortical plasticity (Jain, 2002). The mechanics underlying this reorganisation post stroke are unclear (Albert and Kesselring, 2012) but there is consensus that repetition, specific learning conditions and a stimulating learning environment (specific to the individual) are most likely to induce and promote neural plasticity (Albert and Kesselring, 2012; Langhorne et al., 2009; Krakauer, 2006). There is also some evidence that, as part of the natural history of stroke recovery, there is some spontaneous neuroplasticity regardless of intervention (Kwakkel et al., 2004) but also that early, intensive intervention post stroke results in the most gain from rehabilitation (Kwakkel et al., 2002).

A fundamental principle of motor learning is that more frequent practice results in greater improvement of the motor activity (Schmidt and Lee, 1988). In essence, regular repetition of the same movement will result in an improved ability to carry out that movement. However this approach does not necessarily result in sustained change over time (Giuffrida et al., 2002) which is essential for functional recovery. It seems that for practice to be of sustained benefit, it is not as simple as pure repetition. Practice can be carried out in a variety of ways, some of which are more effective than others (Schmidt and Lee, 1988) in leading to sustained improvement and generalisation of the motor learning to other functions beyond that practiced. There have been numerous research studies into motor learning principles in healthy adults, including detailed investigation of practice schedules, task presentation and feedback (Schmidt and Lee, 1988; Shea and Kohl, 1991). There have been fewer clinical studies

with individuals with brain injury and even fewer on individuals with communication impairment as a result of brain injury. The studies that appear most pertinent to stroke and rehabilitation approaches to motor impairment are discussed below.

Different schedules of learning have been described; intensive or non-intensive practice schedules relates to the number of repetitions of the same movement before having a break (Hinckley and Carr, 2005; Mackay et al., 2002). 'Massed' practice relates to the practice of one movement/task while 'distributed' practice relates to the combination of different movements or tasks (Mackay et al., 2002).There is evidence to suggest that having rest breaks between repetitions results in better performance and retention over time (Mackay et al., 2002). In post stroke recovery, the benefits of different practice schedules can depend on the task and what stage the patient is at for that particular activity. Massed practice, with no breaks, may be of benefit to patients at an early stage of motor learning but later may be of less benefit due to mental or physical fatigue which is termed reactive impedance (Donovan and Radosevich, 1999). Hence, reactive impedance can be reduced by maintaining attention and engagement by changing the task and demands so it is not just repetition without learning as attempted with 'distributed practice' (Donovan and Radosevich, 1999). Longer rest periods have been found to improve learning (Verdaasdonk et al., 2007) although if this rest period becomes too long it can have a detrimental effect on learning (Savion-Lemieux and Penhune, 2005). However, both 'longer rest breaks' and 'too long' are never specified in terms of actual time. Some evidence indicates that the optimal length of time for a rest break is dependent on the complexity of the motor skill being carried out (Donovan and Radosevich, 1999).

Distributed practice is generally more effective than intensive learning and several theories have been proposed to explain this. It may be that changing tasks allows biochemical changes to occur (Verdaasdonk et al., 2007; BrashersKrug et al., 1996) or that learning is consolidated during sleep (Dail and Christina, 2004; Verdaasdonk et al., 2007). It has also been suggested that intensive practice simply involves more time practising which highlights the importance of the more practice the better as well as increasing the individuals awareness of their abilities (Moulton et al., 2006; Mackay et al., 2002). It seems that predictable practice conditions with errorless learning can be more effective in the early stages of learning and more challenging, distributed practice that is error-full can be more effective in later recovery. Being able to generalise learning is of significance in rehabilitation as the aim of motor learning is to impact on functional skills in everyday life (Rendell et al., 2011; Hanlon, 1996).

Another key part of motor learning is the importance of feedback to enhance and improve skill learning (Thorndike, 1927). This is a vital part of stroke rehabilitation where people need to re-learn skills and feedback has been found to play a crucial role where this must be specific to be effective. Feedback can be defined as intrinsic, so the feedback received by the individual from muscle movement, and extrinsic, where this can be the therapist or via equipment such as bio-feedback. The most effective type of feedback for motor learning has been found to vary according to stage and severity of recovery (Cirstea et al., 2006). Feedback can be given either through auditory or visual means and there is no evidence to suggest one is better than the other (Van Vliet and Wulf, 2006). Feedback studies in motor learning are mostly related to physiotherapy and little is known about what sort of feedback is delivered in speech rehabilitation

sessions. The importance of feedback during speech rehabilitation is a topic that needs further investigation considering the quantity, frequency, how it is given and by whom.

As dysarthria is caused by neuromuscular impairment (Darley et al., 1975), it would seem likely that motor learning principles can be applied to dysarthria rehabilitation. It is acknowledged that intact and impaired motor systems may respond differently to motor learning protocols but these principles are effective for physical limb rehabilitation in people with neurologically impaired systems (Hanlon, 1996; Krakauer, 2006) and it is possible they would have a similar effect if applied to muscles of speech as well the rest of the body. There are however no studies specifically examining the use of motor learning principles in dysarthria following stroke. A systematic review of the benefits of motor learning interventions for dysarthria or apraxia carried out by Bislick et al. (2012) found 7 relevant article. Two studies related to healthy adults, one to people with speech problems from Parkinson's disease and four studies relating to speech apraxia. None related to dysarthria following stroke and although the conclusion of the review found the results promising, there is clearly a need for further investigation of the potential of motor learning principles for motor speech disorders following stroke.

1.3.2 Technology to support rehabilitation

Rehabilitation can improve quality of life following stroke (Teasell et al., 2008) but access to rehabilitation at the right time and intensity for individuals may not be possible due to resource and time limitations. Quantity and quality of rehabilitation may also be affected by whether evidence based care is being delivered (Jutai and Teasell, 2003). A recent analysis in UK stroke units found most were operating below the recommended staffing level guidelines and therefore struggling to deliver the

recommended amount of therapy for optimum recovery (McHugh and Swain, 2014). Therapy time is therefore limited and dominated by assessment, liaison between health care professionals, and documentation (Putman et al., 2006; Foley et al., 2012; Clarke et al., 2015). This may be compounded by the fact that speech and language therapy was rarely available at weekends although this is now starting to change (Teasell et al., 2008).

The use of technology, including robotics, mobile devices and computers (Kwakkel et al., 2008) is a possible way to increase access to and the intensity of stroke therapy as an inexpensive adjunct or alternative to traditional face to face treatment with a therapist. This may enable patients to engage more in their rehabilitation, undertake greater repetition of tasks and the exercise during therapy sessions and/or enable patients to engage in therapy outside of therapy sessions. The uptake of technologies in rehabilitation has been limited to date due to challenges related to design and content of the technologies and acceptance by professionals, patients and services as well as the strength of the evidence base. It is acknowledged that technology implementation in healthcare is a complex area with many external and intrinsic factors affecting whether innovations are adopted or not (Greenhalgh et al., 2004).

Rehabilitation technology is, in theory, ideally placed to deliver therapy which is consistent with motor re- learning principles including intensity and task specificity (Krakauer, 2006; Kalra and Ratan, 2007; Langhorne et al., 2009). There has also been a contextual change within health care in the UK (ICSWP, 2016) with the aim of delivering rehabilitation in the community. This places additional demands on health

care professionals to deliver high quality therapy with the same intensity as would be delivered in hospital (ICSWP, 2012). There is a clear need to consider solutions to enable stroke patients to be less reliant on therapists, encouraging greater self-management and based in the home/community (Parker et al., 2014). Technology such as computer-based therapy may provide this as an alternative or an adjunct to 'traditional' face-to-face intervention (Ballinger et al., 1999) and thereby reduce costs (Siekierka et al., 2007). Going forward in terms of wider rehabilitation there will need to be exploration of whether there is an optimal delivery 'mix' between therapist and technology intervention that will achieve the optimal rehabilitation outcomes with regard to motor learning and self-management (Parker et al., 2014).

A Cochrane review into the benefits of one type of rehabilitation technology, virtual reality and gaming on stroke rehabilitation compared to another intervention or no intervention (Laver et al., 2011), found limited evidence that this approach improved outcome in arm movements and activities of daily living. There were mixed views between health care professionals and patients towards the use of technology in rehabilitation (Dijkers et al., 1991). There appear to be three key themes relating to the acceptance of new technology and whether it is adopted: knowledge; control; and, barriers to change (Chen and Bode, 2011). Clinicians are more likely to support successful implementation of a technology if they have information about cost benefits, the time and effort required to learn about the new technology and a perceived control over the decision to use the technology (Pare et al., 2006). Resistance to change or adoption can emerge from any of the stakeholders involved including the patient, therapist and the health care delivery system (Wallace et al., 2001). Generally all professionals considered patients' needs, practical implications

(such as availability on discharge), cost and clinical implications (such as impact on progress) to be critical to success (Chen and Bode, 2011).

There is a substantial body of research about computerised treatment of aphasia compared to dysarthria and there are promising results emerging (Stark and Warburton, 2016; Palmer et al., 2012). A recent systematic review of technology to treat anomia found improvements to naming but no clear evidence of carry-over to everyday speech (Lavoie et al., 2017). Several computer programmes are in current clinical use for people with aphasia to enable them to self-manage their aphasia rehabilitation. Potential barriers to accessing technology are reading and comprehension problems, fine motor control, motivation, cognition (e.g., deficits in sustained attention), expectations and family support (Brandenburg et al., 2013; Chen and Bode, 2011). Palmer et al., (2013) found support was important for patients with aphasia and their carers to carry out computer practice independently. An advantage of using computer-based technology is that the frequency of engagement with technology can be recorded and therefore adherence to therapy or practice can be monitored. Furthermore the programme can be modified thereby enabling individualised therapy as the individual progresses (Palmer et al., 2013). A recent pilot study reported that home-based computer therapy for aphasia was feasible (Palmer et al., 2012), while an earlier small trial (n=7) indicated that independent computer-based aphasia therapy can improve patients' perceived language, autonomy, communication activity, participation and confidence (Mortley et al., 2004; Palmer et al., 2012).

The use of computer based therapy for aphasia is commonly used in mainstream clinical practice but, in contrast, no commercially available computer-based therapy software exists for dysarthria. One small study compared traditional therapy with

computerized therapy in individuals with dysarthria from a variety of aetiologies with some promising results showing improvements to speech as effective as traditional therapy and potentially cost effective (Palmer et al., 2007). Development of technology based dysarthria interventions could help patients to overcome many problems with dysarthria intervention: lack of provision, lack of individualised input and lack of intensity of delivery.

In the next chapter, chapter 2, we have carried out a systematic review to ensure we have reliably searched for, with as little bias as possible and found, the best available evidence. The technology intervention, ReaDySpeech had been developed based on current clinical practice and existing best practice guidelines and would have been influenced by the Cochrane findings. The lack of definitive trials however, meant the acceptability work for ReaDySpeech could continue as described in chapter 3. This led to the development of the feasibility trial, protocol chapter 4, and the results of this are reported in chapters 5 and 6. The aim of this thesis is to report the development and evaluation of an online technology intervention for dysarthria after stroke.

1.4 Research questions posed within this thesis

1. What evidence is there for effectiveness of dysarthria interventions?
2. Is online therapy acceptable to people with post-stroke dysarthria, their therapists and accessible in an NHS clinical context?
3. Can we design a feasibility randomised controlled trial for an online therapy?
4. Is it feasible to carry out a randomised controlled trial of online therapy ReaDySpeech for people with dysarthria after stroke?
5. What was the ReaDySpeech and usual care intervention delivered during a feasibility randomised controlled trial?

Chapter 2 Interventions for dysarthria due to stroke and other adult-acquired, non-progressive brain injury (Cochrane Review)

This Cochrane Review has been published and is presented here in a format suitable for this thesis. This paper responds to the research question: What evidence is there for effectiveness of dysarthria interventions? A systematic review was considered appropriate methodology to ensure all recent relevant research had been sought and systematically evaluated in an attempt to minimise bias as outlined in The Principles of The Cochrane Collaboration (Higgins, 2013).

Mitchell C, Bowen A, Tyson S, Butterfint Z, Conroy P.

Interventions for dysarthria due to stroke and other adult-acquired, non-progressive brain injury.

Cochrane Database of Systematic Reviews 2017, Issue 1. Art. No.: CD002088.

DOI: 10.1002/14651858.CD002088.pub3.

2.1 Abstract

Background

Dysarthria is an acquired speech disorder following neurological injury that reduces intelligibility of speech due to weak, imprecise, slow and/or unco-ordinated muscle control. The impact of dysarthria goes beyond communication and affects psychosocial functioning. This is an update of a review previously published in 2005. The scope has been broadened to include additional interventions, and the title amended accordingly.

Objectives

To assess the effects of interventions to improve dysarthric speech following stroke and other non-progressive adult-acquired brain injury such as trauma, infection, tumour and surgery.

Search methods

We searched the Cochrane Stroke Group Trials Register (May 2016), CENTRAL (Cochrane Library 2016, Issue 4), MEDLINE, Embase, and CINAHL on 6 May 2016. We also searched Linguistics and Language Behavioral Abstracts (LLBA) (1976 to November 2016) and PsycINFO (1800 to September 2016). To identify further published, unpublished and ongoing trials, we searched major trials registers: WHO ICTRP, the ISRCTN registry, and ClinicalTrials.gov. We also handsearched the reference lists of relevant articles and contacted academic institutions and other researchers regarding other published, unpublished or ongoing trials. We did not impose any language restrictions.

Selection criteria

We selected randomised controlled trials (RCTs) comparing dysarthria interventions with 1) no intervention, 2) another intervention for dysarthria (this intervention may differ in methodology, timing of delivery, duration, frequency or theory), or 3) an attention control.

Data collection and analysis

Three review authors selected trials for inclusion, extracted data, and assessed risk of bias. We attempted to contact study authors for clarification and missing data as required. We calculated standardised mean difference (SMD) and 95% confidence interval (CI), using a random-effects model, and performed sensitivity analyses to assess the influence of methodological quality. We planned to conduct subgroup analyses for underlying clinical conditions.

Main results

We included five small trials that randomised a total of 234 participants. Two studies were assessed as low risk of bias; none of the included studies were adequately powered. Two studies used an attention control and three studies compared to an alternative intervention, which in all cases was one intervention versus usual care intervention. The searches we carried out did not find any trials comparing an intervention with no intervention. The searches did not find any trials of an intervention that compared variations in timing, dose, or intensity of treatment using the same intervention. Four studies included only people with stroke; one included mostly people with stroke, but also those with brain injury. Three studies delivered

interventions in the first few months after stroke; two recruited people with chronic dysarthria. Three studies evaluated behavioural interventions, one investigated acupuncture and another transcranial magnetic stimulation. One study included people with dysarthria within a broader trial of people with impaired communication.

Our primary analysis of a persisting (three to nine months post-intervention) effect at the activity level of measurement found no evidence in favour of dysarthria intervention compared with any control (SMD 0.18, 95% CI -0.18 to 0.55; 3 trials, 116 participants, GRADE: low quality, $I^2 = 0\%$). Findings from sensitivity analysis of studies at low risk of bias were similar, with a slightly wider confidence interval and low heterogeneity (SMD 0.21, 95% CI -0.30 to 0.73, $I^2 = 32\%$; 2 trials, 92 participants, GRADE: low quality). Subgroup analysis results for stroke were similar to the primary analysis because few non-stroke participants had been recruited to trials (SMD 0.16, 95% CI -0.23 to 0.54, $I^2 = 0\%$; 3 trials, 106 participants, GRADE: low quality).

Similar results emerged from most of the secondary analyses. There was no evidence of a persisting effect at the impairment (SMD 0.07, 95% CI -0.91 to 1.06, $I^2 = 70\%$; 2 trials, 56 participants, GRADE: very low quality) or participation level (SMD -0.11, 95% CI -0.56 to 0.33, $I^2 = 0\%$; 2 trials, 79 participants, GRADE: low quality) but substantial heterogeneity on the former. Analyses of immediate post-intervention outcomes provided no evidence of any short-term benefit on activity (SMD 0.29, 95% CI -0.07 to 0.66, $I^2 = 0\%$; 3 trials, 117 participants, GRADE: very low quality); or participation (SMD -0.24, 95% CI -0.94 to 0.45; 1 study, 32 participants) levels of measurement.

There was a statistically significant effect favouring intervention at the immediate, impairment level of measurement (SMD 0.47, 95% CI 0.02 to 0.92, P = 0.04, $I^2 = 0\%$; 4

trials, 99 participants, GRADE: very low quality) but only one of these four trials had a low risk of bias.

Authors' conclusions

We found no definitive, adequately powered RCTs of interventions for people with dysarthria. We found limited evidence to suggest there may be an immediate beneficial effect on impairment level measures; more, higher quality research is needed to confirm this finding.

Although we evaluated five studies, the benefits and risks of interventions remain unknown and the emerging evidence justifies the need for adequately powered clinical trials into this condition.

People with dysarthria after stroke or brain injury should continue to receive rehabilitation according to clinical guidelines.

2.1.1 Plain language summary

Interventions for speech problems (dysarthria) after stroke or other non-progressive brain injury

Review question

Does any type of treatment help people who have difficulty speaking clearly after a stroke or other types of brain injury acquired during adulthood?

Background

Brain damage caused by stroke, injury or other non-progressive disease can make speech unclear and difficult for listeners to understand. This condition is known as dysarthria and it occurs when face, tongue, and throat muscles are weak, slow, and unco-ordinated. Dysarthria can cause people who are affected to lose confidence when talking and become socially isolated, even if others see symptoms as mild. People with dysarthria do not have difficulties thinking, remembering, or retrieving words.

Treatment is usually provided by a speech and language therapist or speech pathologist and involves advice and education plus strategies and exercises to increase clarity of speech and to cope with social interaction. Other types of treatment used include acupuncture or brain stimulation.

We wanted to find out if any treatments work, if the effects are long lasting, and if so, which works best, when treatment should start, how frequent treatment should be, and for how long. To find out we searched for, evaluated, and summarised the quality of the existing research on this topic.

Search date

We searched the literature up to May 2016.

Study characteristics

We included five small trials that randomised only 234 people, almost all with stroke. Two trials investigated dysarthria treatment versus an attention control and three compared one treatment with usual care. There were no trials that compared one treatment to no treatment.

Key results

We found few randomised controlled trials of dysarthria treatment, and those that have been conducted involved small numbers of participants, or were not adequately designed or had serious reporting flaws.

We compared many different measures at various time points after treatment, so caution is recommended when interpreting results. We found no evidence of effectiveness on most measures, including long-lasting improvement in every day communication abilities. A positive finding was short-term improvement in muscle movement, such as tongue and lip control. However, this result is not reliable because it was based on small numbers of people, and we found concerns about the conduct and reporting of some trials. This finding needs to be investigated in a bigger, better designed trial.

We found insufficient evidence to tell us whether any one treatment is better than any other or whether treatment is better than general support, or no treatment. We found

no studies that examined timing, duration, or intensity of treatment. This is a clinically important question and should be considered in future trials.

Quality of the evidence

The included trials varied in quality but all included small numbers of participants. Overall, studies were rated as low to very low quality evidence.

Table 1 Summary of findings for the main comparison

Dysarthria intervention compared with another intervention, attention control, placebo or no intervention for people with dysarthria after stroke or other adult-acquired, non-progressive brain injury				
Patient or population: adults with dysarthria following stroke or other adult-acquired, non-progressive brain injury **Settings:** any **Intervention:** dysarthria intervention **Comparison:** another intervention, attention control, placebo or no intervention				
Outcomes	Standardised mean difference (95% CI)	No of participants (studies)	Quality of the evidence (GRADE)	Comments
Dysarthria intervention versus any control: persisting effects, activity level	0.18 (-0.18, 0.55)	116 participants 3 RCTs	⊕⊕⊖⊖ **low**	Very small numbers and none of the studies are adequately powered. Only two of the three studies considered low risk of bias
Dysarthria intervention versus any control: persisting effects, impairment level	0.07 (-0.91, 1.06)	56 participants 2 RCTs	⊕⊖⊖⊖ very low	Very small numbers, none of the studies are adequately powered. Only one of the two studies considered low risk of bias
Dysarthria intervention versus any control: persisting effects, participation level	-0.11 (-0.56, 0.33)	79 participants 2 RCTs	⊕⊕⊖⊖ **low**	Both studies considered low risk of bias but very small numbers and neither study adequately powered.
Dysarthria intervention versus any control for stroke subgroup: persisting effects, activity level	0.16 (-0.23, 0.54)	106 participants 3 RCTs	⊕⊕⊖⊖ **low**	Very small numbers and none of the studies are adequately powered. Only two of the three studies considered low risk of bias

Dysarthria intervention versus any control: immediate effects, activity level	0.29 (-0.07, 0.66)	117 participants 3 RCTs	⊕⊖⊖⊖ very low	Very small participant numbers, not adequately powered. Only one of the three studies considered to be low risk of bias
Dysarthria intervention versus any control: immediate effects, impairment level	0.47 (0.02, 0.92)	99 participants 4 RCTs	⊕⊖⊖⊖ very low	Very small participant numbers, not adequately powered. Only one of the four studies considered to be low risk of bias. This comparison shows a significant effect
GRADE Working Group grades of evidence **High quality:** Further research is very unlikely to change our confidence in the estimate of effect. **Moderate quality:** Further research is likely to have an important impact on our confidence in the estimate of effect and may change the estimate. **Low quality:** Further research is very likely to have an important impact on our confidence in the estimate of effect and is likely to change the estimate. **Very low quality:** We are very uncertain about the estimate.				

2.2 Background

Description of the condition

Dysarthria is a speech disorder affecting intelligibility due to disturbances in neuromuscular control. Dysarthria affects approximately 20% to 30% of stroke survivors and 10% to 60% of those who survive traumatic brain injury (Lawrence et al., 2001; Lubart et al., 2005; Wenke et al., 2008). It can occur in adults as an outcome of meningitis, encephalitis, post-surgical meningioma, and acoustic neuroma (Sellars et al., 2005).

Dysarthria is defined as a neurologic motor speech impairment causing the speech musculature to be slow, weak and/or imprecise (Duffy, 2013). This causes poor co-ordination of movements involving breathing, voice production, resonance, and oral articulation (Yorkston, 1996). People with dysarthric speech typically sound less intelligible or slurred because of poor oral control of articulators, particularly the tongue. Speech can also be quiet, underpowered, and lacking expressiveness because of respiratory control or impaired vocal cord function. Dysarthria includes a wide severity range; some people may be mostly unintelligible to the listener; people at the milder end of the range may experience lapses in speech accuracy, or fatigue, but speech is generally intelligible.

Dysarthria impacts beyond impaired communication. It can negatively affect psychological wellbeing, social participation, and rehabilitation (Brady et al., 2011a; Dickson et al., 2008; Tilling et al., 2001). Brady (Brady et al., 2011a) found that the psychological impact can be influenced by pre-morbid levels of communication demands. An individual with mild dysarthria, but high levels of communication before

their illness, may experience psychological impairment as severe as someone with more severe dysarthria.

Description of the intervention

Behavioural interventions by a speech and language therapist or speech language pathologist are the mainstay of dysarthria treatment. The primary aim is to maximise the patient's ability to communicate with others. UK treatment guidelines for dysarthria (Taylor-Goh, 2005) recommend that behavioural interventions address all dimensions of the International Classification of Functioning, Disability and Health (ICF) Framework; impairment, activity and participation (WHO, 2001). Impairment level exercises to improve the strength, speed, or function of the impaired musculature may be used. These are usually non-speech and oro-motor movements of affected muscles or muscle groups. This may include external stimulation of the muscles such as applying ice packs, brushing the skin, acupuncture (traditional and electrical), or transcranial magnetic stimulation of the brain. At the activity level, compensatory strategies to increase intelligibility through purposeful speech production such as over-articulation or slowing rate of speech may be used. In addition alternative ways to communicate, or support speech, may be used such as an alphabet chart or computers with artificial voice software. Participation level approaches may use facilitated group work, education, and feedback to support the psychological health of people living with dysarthria or advice to a communication partner may be implemented.

How the intervention might work

The interventions at the impairment level in the ‘Description of the intervention’ are likely to be focused on the recovery of impaired movement through exercises to

increase strength, range, precision and speed of movement required for speech. Treatment can utilise non-speech or more typically speech-focused movement tasks. Intervention for limb rehabilitation indicates some association between muscle strength and function of movement (Langhorne et al., 2009) but it is not known whether this is the case for muscles involved in speech. Interventions may examine intensity of intervention and may compare quantity, duration and frequency of input. We know from post-stroke research more generally that increased intensity of treatment may be a key element in recovery but the optimum frequency, duration and quantity of intervention is not known (ICSWP, 2016).

The interventions at the activity and participation level as outlined in the 'Description of the intervention' are likely to focus on strategies or patient specific goals to improve speech intelligibility that relate to a meaningful communication activity for that person. Stroke guidance suggests that goal setting should be used as a rehabilitation tool (ICSWP, 2016). This may include reducing rate of speech when talking on the phone, employing purposeful use of speech intonation to distinguish statements from questions in conversation, or advice to the key communication partner. Group or individual work to target confidence in use of communication is another treatment approach, which may incorporate principles of psychological interventions such as motivational interviewing. Environmental modification and education can also be utilised to optimise communication ease and success in a given context such as a family, hospital or nursing home setting.

Why it is important to do this review

The previous version of this review found no studies that met inclusion criteria (Sellars et al., 2005). Further trials have since been published, and this update broadened the scope of the search strategy applied by Sellars (Sellars et al., 2005) to include all interventions carried out by any health professional, people with dysarthria, a trained individual, or any other new approaches to treatment.

2.3 Objectives

To assess the effects of interventions to improve dysarthric speech following stroke and other non-progressive adult-acquired brain injury such as trauma, infection, tumour and surgery.

2.4 Methods

Criteria for considering studies for this review

Types of studies

We included randomised controlled trials (RCTs) of interventions to improve non-progressive dysarthric speech in adults with acquired brain injuries, including comparisons with no intervention, another intervention (which may be the same intervention approach but alternative method, theory, timing, duration or frequency), attention control, or placebo. We included data only from the first phase of cross-over trials to avoid contamination.

Types of participants

Adults (aged over 18 years) diagnosed with non-progressive dysarthria following acquired brain injury, principally stroke and traumatic brain injury, at any time since stroke onset or trauma event.

Types of interventions

We considered any type of intervention for acquired dysarthria including behavioural or psychological approaches, use of devices and medication, excluding surgical interventions. Interventions could be carried out by any healthcare professional, healthcare staff, trained volunteer, family member or carer, or the person with dysarthria.

Interventions addressed any level of the International Classification of Functioning Disability and Health (ICF)(WHO, 2001) including the following.

- Impairment level: interventions specifically targeting the impairment of function, e.g. non-speech and oro-motor exercises to improve speed, range, strength, accuracy of speech/respiratory musculature, external stimulation of the muscles such as applying ice packs, brushing the skin, transcranial magnetic stimulation of the brain, acupuncture (traditional and electrical).
- Activity level: interventions to increase intelligibility by modifying existing speech (e.g. modifying rate of speech) or the use of augmentative or alternative communication devices e.g. light tech aids (non-technical materials such as an alphabet chart) and high tech aids (such as text-to-talk computer devices).

- Participation level: interventions aimed at support or education for the individual with dysarthria or programmes for people with dysarthria and their conversational partners or conversational training as well as any psychological approaches to treatment that focus on increasing social participation.

We did not place any restrictions on frequency, intensity, or duration of the interventions.

2.4.1 Types of outcome measures

Primary outcomes

The primary outcome measure for this review was the long-term effectiveness of the dysarthria intervention on everyday speech (activity level, persisting effect) compared with any control (another intervention, attention control or placebo, or no intervention). Attempts to objectively measure everyday speech are usually based on listener perception grading scales such as dysarthria therapy outcome measures (Enderby et al., 2013) or the communication effectiveness measure (Mackenzie and Lowit, 2007). We defined evidence of a persistent beneficial effect as around six months post-intervention extracted as measures taken between three and nine months post-intervention.

When trials used more than one outcome measure at the activity level, we took the primary outcome as specified by the trial investigators. If a trial had not specified a primary outcome measure, we checked if a measure of functional communication had been used at the specified time points.

Secondary outcomes

Secondary outcomes included exploring effects:

- at other measurement levels (e.g. impairment, participation);
- at other time points (e.g. immediate post-intervention);
- compared with specific control groups (e.g. another intervention, attention control or placebo, or no intervention);
- for clinical subgroups (e.g. stroke, brain injury);
- for studies assessed at low risk of bias.

Secondary outcome measures were as follows.

- Communication at impairment level (immediate and persisting): speech impairment measure e.g. Frenchay Dysarthria Assessment edition I or II (Enderby, 1983; Enderby and Palmer, 2008), Iowa Oral Performance Instrument (IOPI)(Northwest, 2005), measures of intelligibility (e.g. Assessment of intelligibility of Dysarthric Speech)(Yorkston and Beuklman, 1981), acoustic and perceptual measures of voice and speech (e.g. vocal profile analysis, pitch, loudness, air flow, sound spectography).
- Communication at activity level (immediate): activity measure (e.g. Dysarthria Therapy Outcome Measure)(Enderby et al., 2013), listener acceptability measures.
- Communication-related quality of life (immediate and persisting participation level): patient perception of impact (e.g. Dysarthria Impact Profile)(Walshe et al., 2009); Communication Outcomes after Stroke Scale (Long et al., 2008).

- Generic quality of life measures: mood scales (e.g. Hospital Anxiety and Depression Scale) (Zigmond and Snaith, 1983); subjective health scales (e.g. EuroQol, SF-36)(Herdman et al., 2011).

Search methods for identification of studies

See the 'Specialized register' section in the Cochrane Stroke Group module. We did not impose any language restrictions and we sought translations for non-English language studies.

Electronic searches

We searched the Cochrane Stroke Group Trials Register (last searched by the Managing Editor to May 2016), the Cochrane Central Register of Controlled Trials (CENTRAL, Cochrane Library 2016, Issue 4; Appendix 1), MEDLINE (1946 to May 2016; Appendix 2), Embase (1974 to May 2016; Appendix 3), CINAHL (1937 to May 2016; Appendix 4), PsycINFO (1800 to September 2016; Appendix 5) and LLBA (1976 to November 2016; Appendix 6) using comprehensive search strategies.

We searched major trials registers for ongoing trials including the World Health Organization International Clinical Trials Registry Platform (who.int/ictrp/search/en/), the ISRCTN registry (isrctn.com/), ClinicalTrials.gov (clinicaltrials.gov/) and the Stroke Trials Registry (strokecenter.org/trials/).

Searching other resources

In an effort to identify other published, unpublished, and ongoing trials we hand searched the reference lists of relevant articles and contacted academic institutions and other researchers.

Data collection and analysis

Selection of studies

Our selection criteria were as follows.

- Research participants with dysarthria following stroke or other adult-acquired, non-progressive brain injury.
- Interventions designed to reduce the dysarthria or its impact on living with dysarthria.
- RCTs.

One author (CM) excluded any obviously irrelevant reports from the titles and abstracts retrieved in the search. Three authors (CM, AB, PC) independently examined the remaining abstracts and then the full-text to determine eligibility and exclude irrelevant reports. We resolved disagreements through discussion. No review author examined their own study. We pursued finding conference proceedings and dissertations that were difficult to retrieve using email contacts, university alumni societies, and conference committees. We arranged for reports published in languages other than English to be translated where required. Where possible, we contacted authors of studies for clarification to inform discussions around eligibility. All authors agreed final decisions on included studies and proceeded to data collection. The

studies we judged as ineligible for inclusion are listed with reasons for exclusion in Characteristics of excluded studies.

Data extraction and management

Three authors (CM, AB, PC) independently carried out data extraction from trial reports in pairs (avoiding authors' own trials), and extracted the following data.

- Methods: study design, study duration, sequence generation, allocation sequence concealment, blinding.
- Participants: total number, attrition, setting, diagnostic criteria, age, gender, country of research.
- Interventions: total number of intervention groups, specific intervention and details.
- Outcomes: outcomes and time points, outcome definition and measurement.
- Results: number of participants allocated to each intervention, sample size, missing participants, summary data.

We attempted to contact trial authors for further information where risk of bias was unclear or data were missing. We reconciled the independent data extraction between pairs of review authors and would have resolved any disagreements by discussion or with reference to an independent arbitrator (ST) if required.

2.4.2 Assessment of risk of bias in included studies

Three authors (CM, AB, PC) independently carried out the assessment of risk of bias and methodological quality within the pairs assigned for data extraction. The authors used Cochrane's 'Risk of bias' tool (Higgins, 2013). We examined the studies for the

following quality criteria: random sequence generation, allocation concealment, blinding of outcome assessors, incomplete outcome data, and selective reporting.

For random sequence generation (selection bias), we considered trials to be low risk if the random component was clearly described, at high risk of bias where randomisation was influenced by the availability of the intervention, or an unclear risk where there was insufficient information to decide. For allocation concealment (selection bias), we considered trials adequately concealed if the process made clear that participants and investigators could not possibly predict allocation. We considered a study to be at high risk if there was a possibility that allocation could be predicted (e.g. open random allocation schedule, open computer systems potentially accessible to the investigator), or where concealment was unclear and the study author was unable to provide sufficient information or did not respond.

It was accepted that the participants and the therapists delivering the intervention could not be blinded to the intervention. Thus, we considered blinding in terms of outcome assessment (performance bias and detection bias) and we considered studies to be at a low risk of bias if the outcome assessor was clearly blinded to the intervention; we considered studies to be at a high risk of bias if this was not the case, the blinding could be broken and an unclear risk of bias if there was insufficient information provided.

We considered incomplete outcome data (attrition bias) a low risk if there were:

- no missing outcome data;
- missing outcome data that were unlikely to be related to true outcome;

- missing outcome data that were balanced in numbers across intervention groups;
- similar reasons for missing data across groups; and
- missing data that had been imputed using appropriate methods that did not affect outcome and were reported as such.

We considered studies to be at a high risk of bias if they did not address:

- incomplete outcome data adequately;
- missing outcome data likely to be related to the true outcome;
- imbalance of numbers or reasons for missing data across the intervention groups;
- effect size among missing outcomes to induce clinically relevant bias;
- an intention-to-treat analysis done with substantial differences of the intervention received.

We considered selective reporting (reporting bias) within studies included in the review. We considered whether studies had reported all outcome data compared with their planned protocols (published or unpublished) where possible. Where this was not possible, we asked study authors for additional information on planned outcome reporting prior to the study. We considered study authors who did not respond to this request an unclear risk.

Measures of treatment effect

We treated the measures of functional speech as a continuous measure. We abstracted, calculated or requested means and standard deviations. We calculated

standardised mean differences (SMDs) and confidence intervals (CIs), using a random-effects model for the primary outcome and for any secondary outcomes measures included.

Unit of analysis issues

For continuous data we requested or calculated the mean and standard deviation (SD) data. We analysed outcomes as SMD and 95% CI. We used inverse variance and random-effects models. We entered data so that a higher score represented a favourable outcome.

We used RevMan 5 for all analyses (RevMan, 2014).

Dealing with missing data

We requested missing data from study authors as needed; this is reported in Characteristics of included studies.

Assessment of heterogeneity

We assessed heterogeneity between trials with the selected comparisons and outcomes comparing measures, time points, trial design and clinical subgroups. We determined statistical heterogeneity based on the statistic with Chi^2 distribution. We quantified heterogeneity using the I^2 statistic, which describes the proportion of total variance across trials. We considered heterogeneity of 40% or more as considerable and 70% or more as substantial (Higgins et al., 2003; Deeks et al., 2008). Heterogeneity below 40% was considered low.

Assessment of reporting biases

We planned to explore reporting bias if 10 or more trials were included in the review as outlined in The *Cochrane Handbook for Systematic Reviews of Interventions* (Higgins, 2013).

Data synthesis

The primary analysis pooled all trials in the meta-analysis, using a random-effects model, including the dysarthria intervention versus any control (another intervention, attention control, placebo or no intervention). We considered primary outcome data measures and secondary outcome measures at various time points (immediate and persistent) and various levels of functioning.

GRADE and 'Summary of findings' table

We created Summary of findings Table 1 for the main comparison and included the following outcomes:

1. dysarthria intervention versus any control: persisting effects, activity level;
2. dysarthria intervention versus any control: persisting effects, impairment level;
3. dysarthria intervention versus any control: persisting effects, participation level;
4. dysarthria intervention versus any control for stroke subgroup: persisting effects, activity level;
5. dysarthria intervention versus any control: immediate effects, activity level; and

6. dysarthria intervention versus any control: immediate effects, impairment level.

We used the five GRADE considerations (study limitations, consistency of effect, imprecision, indirectness and publication bias) to assess the quality of the body of evidence as it related to the included studies (Atkins et al., 2004). We used methods and recommendations described in Section 8.5 and Chapter 12 of the *Cochrane Handbook for Systematic Reviews of Interventions (Higgins, 2013)* using GRADEproGDT software (GRADEproGDT, 2015). We justified all decisions to down- or upgrade the quality of studies in footnotes, and provided comments to aid readers' understanding where necessary.

Subgroup analysis and investigation of heterogeneity

We carried out subgroup analysis to explore the effect of comparison with all controls (another intervention, attention control, placebo or no intervention). We carried out clinical subgroup analysis of stroke or brain injury and a subgroup sensitivity analysis where studies had low risk of bias.

Sensitivity analysis

We carried out sensitivity analysis to explore methodological heterogeneity including studies with adequate allocation concealment and adequate blinding, these were the studies we considered to be at low risk of bias.

2.5 Results

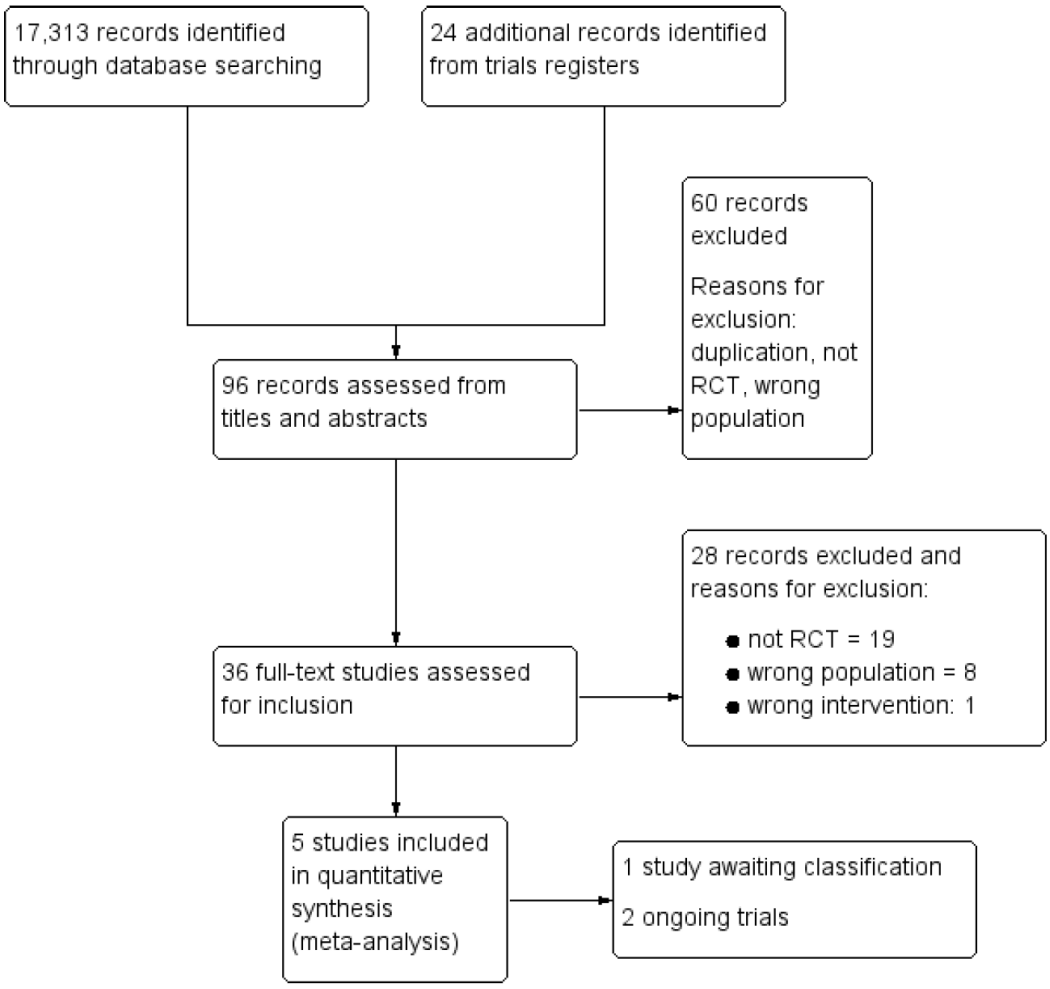
Description of studies

See: Characteristics of included studies; Characteristics of excluded studies; Characteristics of ongoing studies; and Characteristics of studies awaiting classification.

Results of the search

Our searches identified 17,313 records; the screening process is shown in the PRISMA flow diagram (Figure 1). Five papers met our inclusion criteria Bowen (Bowen et al., 2012a), Kwon (Kwon et al., 2015), Mackenzie (Mackenzie et al., 2014), Wenke (Wenke et al., 2010), Xu (Xu et al., 2010) and are described in Characteristics of included studies. We also identified two ongoing studies Peng (Peng et al., 2015) and ReaDySpeech (Mitchell et al., 2017b) see Characteristics of ongoing studies. Both ReaDySpeech, (Mitchell et al., 2017b) and Peng (Peng et al., 2015) presented insufficient detail to inform assessment, and will be assessed for inclusion in a future review update. The study authors of Peng (Peng et al., 2015) have been contacted for further information; we will monitor for publication of the study. You (You et al., 2010) included an English language abstract, but presents insufficient information to make a decision regarding inclusion; this study is presented in Characteristics of studies awaiting classification.

Figure 1 Study flow diagram



Included studies

The included trials randomised a total of 234 participants, ranging from 25 in Kwon (Kwon et al., 2015) to 66 in Bowen (Bowen et al., 2012a). The five trials are detailed in the Characteristics of included studies table (Table 2) and we have included the comparison data below. All included studies were RCTs and each contributed to more than one comparison. We present data that compared one dysarthria intervention with another dysarthria intervention and a dysarthria intervention with an attention control. We found no studies that compared dysarthria intervention with nothing or the same dysarthria interventions with variations in timing, duration, or frequency of delivery. Further information on intervention characteristics and the main comparisons are presented in Characteristics of included studies (Table 2) and Summary of findings (Table 1).

The previous version of this review did not include any studies (Sellars et al., 2005).

Participant characteristics

All five included trials recruited men and women; the proportion of men ranged from 56% in Bowen (Bowen et al., 2012a) to 85% in Kwon (Kwon et al., 2015). The average age ranged from 49 years in Wenke (Wenke et al., 2010) to 70 years in Bowen (Bowen et al., 2012a). Four studies included only people with stroke; Bowen (Bowen et al., 2012a); Kwon (Kwon et al., 2015); Mackenzie (Mackenzie et al., 2014); Xu (Xu et al., 2010); one study included people with stroke and a small number with traumatic brain injury Wenke (Wenke et al., 2010). Two studies tested interventions that were provided in the first four months, Bowen (Bowen et al., 2012a) and two months following stroke Kwon (Kwon et al., 2015). Two studies involved participants who were

in the chronic stage of recovery Mackenzie (Mackenzie et al., 2014); Wenke (Wenke et al., 2010), Xu (Xu et al., 2010) included people between one and 12 months after stroke.

Participants were recruited from hospital Bowen (Bowen et al., 2012a); Xu (Xu et al., 2010), the community, Mackenzie (Mackenzie et al., 2014) or the source of recruitment location was not specified Wenke (Wenke et al., 2010) or not clear Kwon (Kwon et al., 2015). Three studies reported dysarthria severity assessed and reported as part of study characteristics Bowen (Bowen et al., 2012a); Mackenzie (Mackenzie et al., 2014); Wenke (Wenke et al., 2010). People with severe dysarthria were excluded in Xu (Xu et al., 2010) and severity was not reported in Kwon (Kwon et al., 2015). Co-occurring communication impairment or cognitive problems were excluded by two studies Kwon (Kwon et al., 2015); Xu (Xu et al., 2010). Co-occurring aphasias were described in Bowen (Bowen et al., 2012a) and Mackenzie (Mackenzie et al., 2014) but not mentioned in Wenke (Wenke et al., 2010); however, Wenke (Wenke et al., 2010) identified co-existing cognitive impairment. Bowen (Bowen et al., 2012a) recruited people with communication difficulties after stroke including aphasia, dysarthria, or both. People with dysarthria were a planned subgroup within the study by Bowen (Bowen et al., 2012a) and we extracted dysarthria data from the trial data.

Intervention and control interventions

None of the included studies compared dysarthria interventions with no intervention. Two trials compared an intervention with an attention control (Bowen (Bowen et al., 2012a); Kwon (Kwon et al., 2015)). Bowen (Bowen et al., 2012a) investigated enhanced best practice speech and language therapy delivered by speech and language

therapists supported by assistants compared with an attention control (employees offering an equivalent amount of time and social contact but no therapy or therapist input). Kwon (Kwon et al., 2015) investigated repetitive transcranial magnetic stimulation versus sham repetitive transcranial magnetic stimulation; both groups received the same speech therapy intervention.

Three trials compared dysarthria interventions with usual dysarthria care (Mackenzie (Mackenzie et al., 2014); Wenke (Wenke et al., 2010); Xu (Xu et al., 2010)). Mackenzie (Mackenzie et al., 2014) examined oro-motor exercises compared with usual care. Wenke (Wenke et al., 2010) investigated Lee Silverman Voice Treatment (LSVT), an approach that focusses on increased volume of speech, with usual care. Xu (Xu et al., 2010) compared acupuncture with usual care. Usual care was described as behavioural strategies that address impairment and activity levels of functioning (Mackenzie et al., 2014); (Wenke et al., 2010); (Xu et al., 2010). Wenke (Wenke et al., 2010) and Mackenzie (Mackenzie et al., 2014) reported that usual care was based on existing literature and best practice guidelines; Wenke (Wenke et al., 2010) also included consensus agreement. Components of usual care were not reported in Xu (Xu et al., 2010).

There were no comparisons of one intervention versus the same intervention with variations in timing, intensity, or duration of treatment.

We referred to the template for intervention description and replication checklist (TiDier) when extracting the information on the interventions for each study (Hoffmann et al., 2014b).

Intervention compared with attention control

Two studies assessed dysarthria interventions compared with attention controls (Bowen et al., 2012a; Kwon et al., 2015); 86 participants). Bowen (Bowen et al., 2012a) investigated enhanced, flexible, best practice behavioural speech therapy, and Kwon (Kwon et al., 2015) examined repetitive transcranial magnetic stimulation. The enhanced, best practice intervention in Bowen (Bowen et al., 2012a) was described in sufficient detail to enable replication from the manual provided and was agreed by consensus of speech and language therapists to address impairment, activity, and participation levels of functioning. Kwon (Kwon et al., 2015) described the repetitive transcranial magnetic stimulation intervention, equipment used, and how motor-evoked potentials were calculated and established for each participant. The intervention was to be led by an experienced speech and language therapist in Bowen (Bowen et al., 2012a), and in Kwon (Kwon et al., 2015), the intervention was carried out by a physiatrist (physicians specialising in physical medicine and rehabilitation). The attention control applied in Bowen (Bowen et al., 2012a) was structured social contact, carried out by employed, part-time, visitors; five of nine visitors had high levels of educational attainment. In Kwon (Kwon et al., 2015) the attention control was sham repetitive transcranial magnetic stimulation, carried out by the same physiatrist using the same methods as the intervention, but holding the coil perpendicular to the skull rather than tangential to the skull surface.

The population in both studies was people with stroke, both interventions and attention control were delivered at the same time, soon after stroke, within the first two months Kwon (Kwon et al., 2015) and within the first four months Bowen (Bowen et al., 2012a).

Repetitive transcranial magnetic stimulation treatment duration was five days per week for two weeks (Kwon et al., 2015). Enhanced speech therapy was conducted for a maximum of 16 weeks, with duration and frequency as clinically indicated up to a maximum of three times per week (Bowen et al., 2012a). Bowen (Bowen et al., 2012a) mentioned homework, which was given as appropriate to people in the intervention arm, but not to the attention control arm participants. The unpublished intervention manual provided by the Bowen (Bowen et al., 2012a) study authors, includes a sheet to encourage documentation of homework by participants, but there is no further description of whether homework was carried out or completed. Participants in the intervention arm discussed homework and its impact during interviews conducted as part of the qualitative aspect of this study. Kwon (Kwon et al., 2015) describes that both groups had the same speech therapy intervention carried out for 30 minutes, five days per week for the two weeks of rTMS treatment. The content of the speech therapy intervention was not described, although it was carried out by a skilled speech therapist. There was no mention of homework in Kwon (Kwon et al., 2015). Participants in the study by Kwon (Kwon et al., 2015) were not aware of the intervention type they were randomised to receive either the active repetitive transcranial magnetic stimulation or the attention control sham therapy.

The outcome measure for Kwon (Kwon et al., 2015) was a blinded assessment of impairment level immediately post intervention. Participants in Bowen (Bowen et al., 2012a) were aware of the intervention type they were randomised to receive; the primary outcome was a blinded assessment of activity level functioning at six months post-entry to the study.

Intervention A compared with intervention B

Three trials, involving a total of 117 randomised participants, compared one intervention with another intervention Mackenzie (Mackenzie et al., 2014); Wenke (Wenke et al., 2010); Xu (Xu et al., 2010). All three studies compared usual care versus an alternative intervention Mackenzie (Mackenzie et al., 2014); Wenke (Wenke et al., 2010); Xu (Xu et al., 2010). There were no trials that compared one intervention with the same intervention but with variations in timing, duration, or intensity of delivery.

Intervention A in Wenke (Wenke et al., 2010) was Lee Silverman Voice Treatment (LSVT) which aims to increase vocal loudness. In Xu (Xu et al., 2010), intervention A was acupuncture; and in Mackenzie (Mackenzie et al., 2014) 10 minutes of non-speech oro-motor exercises (tongue and lip movements) replaced 10 minutes word and sentence practice.

Intervention A was delivered by the same speech pathologist trained in LSVT in Wenke (Wenke et al., 2010); traditional Chinese medical specialists carried out acupuncture in Xu (Xu et al., 2010); and the same experienced speech and language therapist provided treatment in Mackenzie (Mackenzie et al., 2014).

Intervention B in all three studies was usual care. Wenke (Wenke et al., 2010) and Mackenzie (Mackenzie et al., 2014) described intervention B as behavioural therapy, addressing impairment and activity levels of functioning. Both studies provided sufficient information to enable replication of the therapy. Xu (Xu et al., 2010), did not describe intervention B in sufficient detail to enable replication; there was no information around the content of the therapy, level of impairment, or how therapy was delivered.

Intervention B was delivered by an experienced speech pathologist in Wenke (Wenke et al., 2010); the same hearing and speech specialist delivered the usual care to participants in both arms in Xu (Xu et al., 2010) and the same experienced speech and language therapist delivered both intervention A and B in Mackenzie (Mackenzie et al., 2014).

Treatment timing was for people in the chronic phase of recovery following stroke or brain injury of more than six months or more than three months in Wenke (Wenke et al., 2010) and Mackenzie (Mackenzie et al., 2014) respectively. In Xu (Xu et al., 2010) timing ranged for people with acute to chronic dysarthria of between one and 12 months post stroke.

Treatment duration ranged from four weeks Wenke (Wenke et al., 2010), to eight weeks Mackenzie (Mackenzie et al., 2014) and nine weeks Xu (Xu et al., 2010).

Treatment frequency for interventions A and B was the same for Wenke (Wenke et al., 2010), at one hour per day, four days a week, and the same for Mackenzie (Mackenzie et al., 2014) at 40 minutes once a week. Xu (Xu et al., 2010) differed, with both arms receiving speech therapy for 30 minutes, five times per week but intervention A was delivered for four weeks, with a week-long break followed by four weeks of intervention A.

Independent practice of homework was described in Wenke (Wenke et al., 2010) and Mackenzie (Mackenzie et al., 2014) but was not used in Xu (Xu et al., 2010). In Wenke (Wenke et al., 2010), independent, daily homework was suggested between sessions for intervention B group participants only, but whether this was carried out and recorded was not described. In Mackenzie (Mackenzie et al., 2014), participants in

both intervention A and B were encouraged to carry out independent practice of their allocated intervention of around 30 minutes, five days a week during the seven between session practice weeks for a total of 1050 minutes. This was documented by participants in a diary and the results reported and analysed.

All participants in the three studies were aware of which intervention they were randomised to, none of the three studies had a primary outcome measure.

All three studies carried out an activity level measure, with this being considered to show persistent change for Wenke (Wenke et al., 2010) at six months post treatment, and Mackenzie (Mackenzie et al., 2014) at two months post intervention in a chronic population, but was only carried out immediately post intervention in Xu (Xu et al., 2010).

Outcomes

All five studies used different outcome measures and at various time points. The primary outcome for this review was to examine the persisting effect of the intervention at the activity level of functioning.

Four studies carried out activity level measures Bowen (Bowen et al., 2012a); Mackenzie (Mackenzie et al., 2014); Wenke (Wenke et al., 2010); Xu (Xu et al., 2010); Kwon (Kwon et al., 2015) did not carry out a measure of activity level of functioning.

Wenke (Wenke et al., 2010) and Xu (Xu et al., 2010) used a measure of perceived intelligibility by a speech and language therapist. Bowen (Bowen et al., 2012a) used the dysarthria therapy outcome measures (Enderby et al., 2013), and Mackenzie (Mackenzie et al., 2014) used the communication effectiveness measure and the

Speech Intelligibility Test (Yorkston, 1996). The only study that specified the primary outcome measure was Bowen (Bowen et al., 2012a).

For our analyses of persisting outcome, we took data from measures carried out at three to nine months post intervention; this included Wenke (Wenke et al., 2010) (six months post treatment) and Bowen (Bowen et al., 2012a) (measured at six months post randomisation). Mackenzie (Mackenzie et al., 2014) carried out the final outcome measure at two months (eight weeks) post intervention. The review authors discussed if these data should be included, because this was a chronic population with proximity to the proposed minimum time point of three months (12 weeks). We decided that the proposed time criterion (three months to nine months) in the review protocol was too tight, and agreed to relax timings to include the study data as a persisting effect. This change is reported in 'Differences between protocol and review'. The latest time point for the primary outcome measure, taken by Xu (Xu et al., 2010), was immediately post intervention, which did not meet our requirement of three to nine months post intervention to examine persistent change.

The secondary outcomes were other measures at various time points. This meant we examined data from the activity level measures at immediate time point post-intervention, and this had been carried out by Wenke (Wenke et al., 2010), Xu (Xu et al., 2010) and Mackenzie (Mackenzie et al., 2014). We considered 'immediate' measure to have been carried out at the end of the treatment period or the time period nearest to the end of treatment.

Communication impairment measures were used in four studies Kwon (Kwon et al., 2015); Mackenzie (Mackenzie et al., 2014); Wenke (Wenke et al., 2010); Xu (Xu et al., 2010). These were articulatory precision Wenke (Wenke et al., 2010), maximum

phonation time Xu (Xu et al., 2010), lip and tongue movements from the Frenchay dysarthria assessment (FDA-2) Mackenzie (Mackenzie et al., 2014), and an articulation test Kwon (Kwon et al., 2015). These impairment measures were carried out to show persistent effect between the three month and nine month time points by Wenke (Wenke et al., 2010) and Mackenzie (Mackenzie et al., 2014), but not Xu (Xu et al., 2010) or Kwon (Kwon et al., 2015). These measures were carried out immediately post-intervention by all four studies Kwon (Kwon et al., 2015); Mackenzie (Mackenzie et al., 2014); Wenke (Wenke et al., 2010); Xu (Xu et al., 2010). Measures at the participation level were used by Bowen (Bowen et al., 2012a), which used the Communication Outcomes after Stroke Scale (COAST; Long 2008), and Mackenzie (Mackenzie et al., 2014), which used the Communicative Effectiveness Survey (CES; (Donovan et al., 2007)). Both studies applied this participation level measure as a persistent measure of change between three month and nine months, but only Mackenzie (Mackenzie et al., 2014) applied this immediately post treatment.

Excluded studies

See: Characteristics of excluded studies (Table 3)

We excluded 28 studies primarily because they were not RCTs; Fitzgeralds-DeJean (Fitzgerald-DeJean, 2008); Fukusako (Fukusako et al., 1989); Garcia (Garcia and Dagenais, 1998); Huffman (Huffman, 1978); Huh (Huh et al., 2014); Hustad (Hustad et al., 2003); Ince (Ince and Rosenberg, 1973); Jones (Jones, 1972); Katić (Katic, 1973); Li (Li et al., 2013); Markov (Markov, 1973); Nagasawa (Nagasawa and Kamiyama, 1970); Palmer (Palmer and Enderby, 2004); Palmer (Palmer et al., 2007); Robertson (Robertson, 2001); Rosenbek (Rosenbek et al., 2006); Sakharov (Sakharov and Isanova,

2013); Togher (Togher et al., 2014); Varma (Varma, 2004). In several studies, participants were not dysarthric Behn (Behn, 2011); Behn (Behn et al., 2012); Braverman (Braverman et al., 1999); Sze (Sze et al., 2002); Togher (Togher et al., 2004), or had mixed aetiologies including progressive and congenital conditions; Cohen (Cohen and Masse, 1993); Kelly (Kelly et al., 2000); Main (Main, 1998), or a surgical intervention was investigated Qinglan (Qinglan et al., 2002)

Risk of bias in included studies

Overall risk of bias for the five included studies is depicted in Figure 2 and Figure 3.

Figure 2 Risk of bias graph

Review authors' judgements about each risk of bias item presented as percentages across all included studies

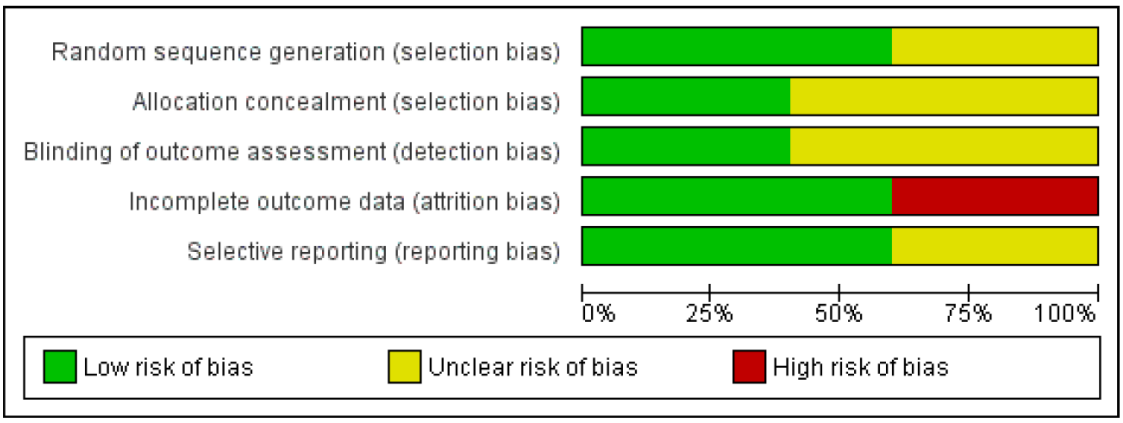
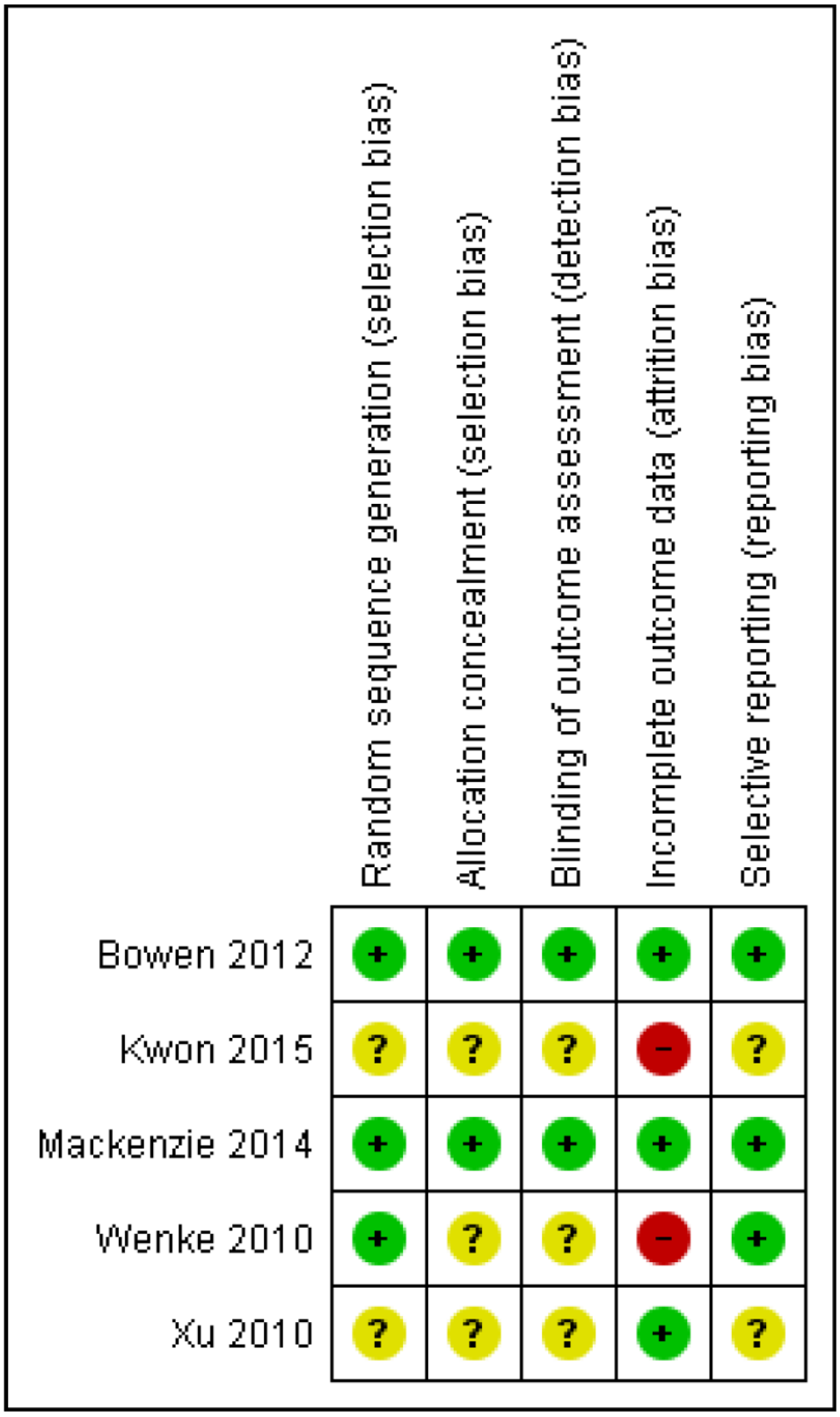


Figure 3 Risk of bias summary

Review authors' judgements about each risk of bias item for each included study



Three review authors independently assessed the included studies for methodological quality (avoiding their own studies) and discussed any discrepancies. We intended to carry out sensitivity analysis according to studies at low risk of bias for each domain. We considered that two studies were at low risk of bias overall, and these were included in the sensitivity analysis (Bowen et al., 2012a; Mackenzie et al., 2014). All five included studies reported inclusion and exclusion criteria.

Allocation

We assessed two RCTs at low risk of bias for both random sequence generation and allocation concealment (Bowen et al., 2012a; Mackenzie et al., 2014). One study, while demonstrating random sequence generation, provided insufficient details to determine adequacy of allocation concealment (Wenke et al., 2010). Two studies provided insufficient details around random sequence generation and allocation concealment and we considered them to have unclear risk of bias without further clarification (Kwon et al., 2015; Xu et al., 2010). All included studies demonstrated adequate matching between randomised groups at baseline with no obvious concerns around risk in this area.

Blinding

Blinded outcome assessment on all measures was clearly described by Bowen (Bowen et al., 2012a) and Mackenzie (Mackenzie et al., 2014). It is not clear in Wenke (Wenke et al., 2010); Xu (Xu et al., 2010) or Kwon (Kwon et al., 2015) whether those involved in the outcome assessments were blind to the intervention. Although it was implied that those carrying out the outcome measures were not involved in the study, reporting

was not sufficiently clear for this to be assessed as low risk without further information and evidence that the blinding process was not easy to break.

Incomplete outcome data

Not all studies described completion of intervention, those that did reported a total of 14 (from 112 randomised participants) withdrawals, with no differences between intervention and control group participants (Bowen et al., 2012a; Kwon et al., 2015).

All five studies reported the number of participants lost to some or all of the follow-up assessments and across all five studies 33 out of the 234 randomised had either no follow up assessment or incomplete follow up assessment. We considered Xu (Xu et al., 2010) to be at low risk of bias; there was no attrition from recruitment to follow-up. Bowen (Bowen et al., 2012a) was assessed as low risk of bias for incomplete data; detailed explanations were provided in the study's data analysis. Missing data from Mackenzie (Mackenzie et al., 2014) was discussed with the study authors, who provided additional information about their analysis using imputed results and multiple imputations had made no difference to the findings; we rated this study as low risk of bias. Wenke (Wenke et al., 2010) reported treating missing data in a standard statistical way; however, implications were not fully addressed and without further information, this study was assessed at high risk of bias. Reporting in Kwon (Kwon et al., 2015) raised significant concerns about incomplete outcome data: five participants were randomised to both treatment arms, but three withdrew from the active treatment arm and two from the sham treatment. Data for these participants were withdrawn from the study; no intention-to-treat analysis was carried out or

discussion included around the implications of these withdrawn data on conclusions. We assessed Kwon (Kwon et al., 2015) at high risk of bias for this domain.

Adherence to intervention and dropout rates by included study are described in Characteristics of included studies.

Selective reporting

Bowen (Bowen et al., 2012a), Mackenzie (Mackenzie et al., 2014) and Wenke (Wenke et al., 2010) reported studies in full with specified outcome measures at specified time points. Bowen (Bowen et al., 2012a) also published a protocol and analyses.

Possible presence of selective reporting was harder to ascertain for Xu (Xu et al., 2010) and Kwon (Kwon et al., 2015). Both studies were assessed at unclear risk of bias for selective reporting. This assessment will be revised following confirmation of methods applied and clarification from the study authors.

Effects of interventions

See: Summary of findings (Table 1)

The results of this review are presented below to show the evidence for the objectives of the review. The main objective was to find whether there was an effect on dysarthric speech of any intervention and this is presented below under the three comparisons. In summary there was no evidence of a long-term effect of the dysarthria intervention on everyday speech compared to any control.

Results are described for comparisons in each outcome.

- Dysarthria intervention compared with another intervention, attention control, placebo or no intervention: persisting effects.
- Dysarthria Intervention compared with another intervention, attention control, placebo or no intervention: immediate effects.
- Dysarthria intervention A versus dysarthria intervention B (whether this is two different interventions or the same intervention with varying timing, duration, and frequency of delivery): persisting and immediate effects.

We included five studies that involved a total of 234 randomised participants. Comparisons were analysed according to our primary outcome of persisting effects of communication at activity level (three RCTs, 116 participants). Comparisons were further analysed for measurement of impairment and participation at immediate and persistent time points. Data were also considered for one subgroup of people with stroke because there were insufficient data for any other clinical subgroups.

We calculated standardised mean difference (SMD) and 95% confidence intervals (CI) because different measures were used of the same underlying construct. We used a random-effects model.

Comparison 1: dysarthria intervention versus any control: persisting effects (three to nine months post intervention), activity level

We found no evidence of an effect for persisting effects at communication activity level for any control Bowen (Bowen et al., 2012a), Mackenzie (Mackenzie et al., 2014), Wenke (Wenke et al., 2010) (116 participants): SMD 0.18, (95% CI -0.18 to 0.55, Tau^2 = 0.00; Chi^2 = 1.47, df = 2, P = 0.48; I^2 = 0%; GRADE: low quality). Findings were very similar for each study, with narrow CIs, but very small numbers of participants. None

of the studies were adequately powered to find an effect (Analysis 1.1). We considered two of the three studies to be at low risk of bias.

Secondary outcomes of dysarthria intervention versus any control: persisting effects (three to nine months), impairment or participation level

We found no evidence of a persisting effect on impairment level measures in favour of any treatment Mackenzie (Mackenzie et al., 2014), Wenke (Wenke et al., 2010) 56 participants, SMD 0.07, 95% CI -0.91 to 1.06; $Tau^2 = 0.35$; $Chi^2 = 3.32$, df = 1 (P = 0.07); $I^2 = 70\%$; GRADE: very low quality). There was substantial heterogeneity between the trials (Analysis 1.2). Both studies had small numbers of participants, and neither study was adequately powered. We considered one study at low risk of bias.

These two RCTs (79 participants) found no evidence of a persisting effect at the participation level (Bowen et al., 2012a; Mackenzie et al., 2014): SMD -0.11 (95% CI -0.56 to 0.33) and Heterogeneity: $Tau^2 = 0.00$; $Chi^2 = 0.16$, df = 1 (P = 0.69); $I^2 = 0\%$; GRADE: low quality (Analysis 1.3). These two studies have small numbers, they are not adequately powered, and only one has a low risk of bias.

Sensitivity analysis of dysarthria intervention versus any control (persisting effects, activity level) included two studies with adequate allocation concealment/adequate blinding (Bowen et al., 2012a; Mackenzie et al., 2014). The data from the sensitivity analysis of these two studies with 92 participants showed no effect and slight heterogeneity (SMD 0.21, 95% CI -0.30 to 0.73, heterogeneity: $Tau^2 = 0.05$; $Chi^2 = 1.47$, df = 1 (P = 0.23); $I^2 = 32\%$; GRADE: low quality) (Analysis 1.4).

Only one of the studies had a comparison of dysarthria intervention versus attention control with a measure of persisting effects at the activity level. This one study with 60

participants (SMD 0.00, 95% CI -0.51 to 0.51), indicated no evidence of an effect when comparing the intervention with an attention control (Bowen et al., 2012a) (Analysis 1.5).

The stroke subgroup for comparison 1 included three studies (Bowen et al., 2012a; Mackenzie et al., 2014; Wenke et al., 2010) 106 participants and showed no evidence of effect (SMD 0.16, 95% CI -0.23 to 0.54, $Chi^2 = 1.61$, df = 2, P = 0.45; $I^2 = 0\%$; GRADE: low quality; Analysis 1.6).

Comparison 2: dysarthria intervention compared with another intervention, attention control, placebo or no intervention: immediate effects at activity, impairment and participation level

Three included studies, with 117 participants, had measures of activity level immediately post intervention but found no evidence of an effect: (SMD 0.29, 95% CI -0.07 to 0.66) (Mackenzie et al., 2014) (Wenke et al., 2010) (Xu et al., 2010). The heterogeneity among studies was low but included very small numbers ($Chi^2 = 0.64$, df = 2 (P = 0.73); $I^2 = 0\%$) GRADE: very low quality) (Analysis 2.1).

Four studies measured impairment level immediately post intervention (Kwon et al., 2015; Mackenzie et al., 2014); (Wenke et al., 2010; Xu et al., 2010). These studies had a total of 99 participants, so each included small numbers of participants but there was a statistically significant effect favouring intervention (P value = 0.04), SMD 0.47 (95% CI 0.02 to 0.92) with low heterogeneity ($Chi^2 = 0.73$, df = 2 (P = 0.69); $I^2 = 0\%$). Only one study was low risk of bias, GRADE: very low quality (Analysis 2.2).

One study measured participation level immediately post intervention (Mackenzie et al., 2014). This single study had 32 participants: SMD -0.24 (95% CI -0.94 to 0.45) indicating no effect of the intervention (Analysis 2.3).

Comparison 3: dysarthria intervention A versus dysarthria intervention B: persisting and immediate effects at activity, impairment and participation level

Due to the small number of studies in this review there are only two comparisons in this section that have not already been carried out in the earlier analysis. It may be possible to populate this section more fully in the future as more trials are carried out.

Analysis 3.1 included two studies of 56 participants comparing intervention A versus B, with a measure of persisting effects at the activity level: SMD 0.38 (95% CI -0.15 to 0.91) indicating no effect of intervention (Mackenzie et al., 2014; Wenke et al., 2010). These studies have low heterogeneity (Heterogeneity: $Tau^2 = 0.00$; $Chi^2 = 0.43$, df = 1 (P = 0.51); $I^2 = 0\%$; GRADE: very low quality).

The second analysis of intervention A versus intervention B that has a measure of persisting effect at the participation level included one study: (Mackenzie et al., 2014). This study has 32 participants: SMD -0.22 (95% CI -0.92 to 0.47) and indicates no effect of the intervention (Analysis 3.2).

We would also have carried out analysis on intervention A versus intervention B, persisting effects at the impairment level but this has been carried out in Analysis 1.2.

We would have looked at intervention A versus intervention B, immediate effects; activity level (Analysis 2.1), impairment level (Analysis 2.2), participation level (Analysis 2.3) but these have already been carried out in the earlier comparisons.

2.6 Discussion

We examined the effectiveness of dysarthria interventions for people with speech problems due to stroke and other adult-acquired, non-progressive brain injury. We have built on the work of Sellars (Sellars et al., 2005) presented in the previous version of this review, by amending and updating objectives and review outcomes to reflect a more global perspective, and to consider new evidence. We considered whether dysarthria interventions were effective when compared with any control, whether the dysarthria intervention was more effective than an attention control, whether one type of dysarthria intervention was more effective than another, or whether one type of dysarthria intervention was more effective than the same intervention when delivered in a different way. We included five studies and presented data from 234 randomised participants.

Summary of main results

See: Summary of findings (Table 1). Meta-analyses demonstrated no evidence of a statistically significant persisting effect of dysarthria intervention compared with any control when communication was measured at either the activity (three studies, 116 participants), impairment (two studies, 56 participants), or participation level (two studies, 79 participants). This lack of effect did not change in the sensitivity analyses of only the studies with a low risk of bias (two studies, 92 participants), when the analysis was restricted to those with an attention control/placebo (one study, 60 participants), or to the subgroup of those with an underlying condition of stroke (three trials, 106 participants). Similarly, there was no evidence for the immediate effect of dysarthria intervention at the activity level (three studies, 117 participants) or participation level (one study, 32 participants). The one significant finding at the impairment level

immediately post-intervention, (four trials, 99 participants) means that clinically there may be some improvement of tongue and lip movement for example but there is no evidence that these persist long-term and the very small numbers and very low quality of the evidence make this an uncertain estimate.

2.6.1 Key findings from this review

- Despite one positive finding, there was insufficient evidence to enable firm conclusions to be drawn due to quality of the evidence.
- Evidence quality was graded as low or very low.
- There was low risk of bias in only two studies.
- There was no consensus on outcome measures or time points for measurement.

2.6.2 Overall completeness and applicability of evidence

We only identified five, small trials which indicates the evidence base is limited. In addition to the limited number of trials there were only small numbers of participants within the trials and there were also issues around quality and risk of bias. There is clearly much more that needs to be done before the objectives of the review can be fully addressed. The wide variety of outcome measures, where none of the five trials used any of the same outcome measures, indicates a need for consensus amongst researchers, people with dysarthria and therapists to identify which measures should be used in future research. However the included studies were all relevant to the review question in that they were all RCTs of dysarthria intervention for stroke and brain injury. The review set out to establish the evidence for all clinical groups who may have dysarthria but we found no RCTs for other types of non-progressive brain injury that may cause dysarthria. One of the studies excluded people with severe

dysarthria and one did not report severity so generalisation to the wider dysarthric population could be affected.

There were variable amounts of information relating to intervention and control description and replicability according to the TIDieR checklist that we used when evaluating the studies (Hoffmann et al., 2014a). In two of the studies this was clearly described in sufficient detail for replication Bowen (Bowen et al., 2012a); Mackenzie (Mackenzie et al., 2014). There was less detail in Wenke (Wenke et al., 2010), although the LSVT intervention used in this study cannot be described as the treatment is trademarked and not available publicly. Xu (Xu et al., 2010) gave minimal information about the usual care interventions in both arms, and this could not be replicated from the information given but they provided much more detail about the acupuncture delivery. Kwon (Kwon et al., 2015) gave detail around the transcranial magnetic stimulation intervention and how the sham/attention control was carried out. There was no detail around the speech therapy that was given to both groups to ensure they had the same treatment alongside the transcranial magnetic stimulation intervention and sham. There was variation in reporting whether the intervention was provided as intended by the protocol and this is detailed in Characteristics of included studies . Fidelity of the intervention and how this was monitored was not described in Wenke (Wenke et al., 2010), Xu (Xu et al., 2010) or Kwon (Kwon et al., 2015) which is important when considering applicability of the evidence. Fidelity to the interventions and attention control was described in detail, including information about how this was monitored, who carried this out, when and how, in Bowen (Bowen et al., 2012a) and Mackenzie (Mackenzie et al., 2014). Whether participants completed the intervention in the arm to which they were allocated was described in Bowen (Bowen

et al., 2012a) and Mackenzie (Mackenzie et al., 2014). Current practice in the UK around rehabilitation continues to focus on early intervention and the review included three studies of early intervention whereas the other two considered intervention with a chronic population.

Quality of the evidence

This review shows that we do not have a robust enough body of evidence to draw firm conclusions about the objectives of this review. It is a measure of progress that there were recent studies that could be included in the meta-analyses however we rated evidence quality for the key outcomes as low or very low (Summary of findings, Table 1). The primary objective of this review was reported by only three of the studies (116 participants; Analysis 1.1). However, none of the three studies were adequately powered to enable comparisons of the interventions because of the small numbers of participants. Bowen (Bowen et al., 2012a), while adequately powered to look at early communication intervention in aphasia and dysarthria, was not adequately powered to evaluate dysarthria intervention only. All secondary outcomes were downgraded due to small participant numbers and imprecision.

Only Bowen (Bowen et al., 2012a) and Mackenzie (Mackenzie et al., 2014) had low risk of bias; the other three studies all had areas of unclear risk or high risk. We carried out sensitivity analyses to remove any studies with high or unclear risk of bias but this did not alter the direction or the significance of the results (Analysis 1.4). The one significant finding was from four studies where we considered the overall quality of the evidence to be very low, which raises concerns around how confident we can feel about this estimate of effect (Analysis 2.2). The main message about the quality of the

evidence found in this review is that, in addition to being adequately powered, the reporting of RCTs must adhere to the CONSORT guidelines (Schulz et al., 2010) and follow the template for intervention description and replication (TIDieR; (Hoffmann et al., 2014a).

Potential biases in the review process

The search strategy was broadened for this review to include trials that may have been carried out by a range of professionals or non-professionals and we felt confident that we used search terms to reflect this broad scope. However, not knowing what potential professional or non-professional groups may be carrying out research may introduce the possibility of bias particularly where unpublished literature or ongoing trials were sought, as only those who have worked or are working in the field of dysarthria were approached.

The search strategy was in line with this broad approach and we documented reasons for study exclusions. We carried out searches with no time restrictions: the searches were all carried out in English language databases, and although we imposed no language restrictions, and had a paper published in Chinese (Xu et al., 2010) translated, this may have restricted our search method. It is highly probable that papers published in other languages were not identified, and this review may be biased toward English-speaking research studies. Xu (Xu et al., 2010) was published in Chinese and data extraction was carried out by two independent Chinese-speaking individuals, but neither were involved in the review team; discrepancies with data extraction may have occurred. There was some need for interpretation of information, which may not be entirely as intended by the author. Where clarification could not be obtained from

study authors, it is possible that information may have been interpreted incorrectly, and that the review is biased until information can be clarified.

Data collection was carried out by individual review authors and then compared in an attempt to reduce any bias around particular methodologies or intervention approaches. To ensure risk of bias judgements were carried out fairly this was considered independently and then compared and discussed by the review team.

The review team was conscious that a review author (AB) was also the lead author of an included study. We considered how to approach this before starting the review, should the study be eligible for inclusion. The review was structured to ensure the study author was not involved in assessing or making judgements about her own study. However, AB provided additional information and data when requested, and contributed her opinion to wider discussions where this was relevant. We were very conscious of the potential for bias in this particular situation and took steps to reduce bias as much as possible.

Agreements and disagreements with other studies or reviews

A previous Cochrane review of dysarthria intervention found no suitable studies for inclusion at that time (Sellars et al., 2005). There are no other systematic reviews of non-progressive dysarthria.

2.6.3 Authors' conclusions

Implications for practice

Research evidence is not yet sufficiently robust to guide clinical practice. It is therefore important for clinicians to continue to offer rehabilitation for people with dysarthria in line with current clinical guidelines.

Implications for research

Further research will need to be appropriately designed to avoid risk of bias, and evaluate persisting effects on activity level measures.

The absence of evidence for dysarthria interventions highlights the paucity of research for this distressing condition, and need for adequately-powered, methodologically-sound and well-reported studies.

Although inclusion of five studies (from none 10 years ago) is to be celebrated, much more needs to be done. Dysarthria research activity is in striking contrast to aphasia research, which has now amassed 57 trials of speech and language therapy interventions for aphasia following stroke (Brady et al., 2016).

Future dysarthria trials should clearly report methods governing randomisation, allocation concealment, clarity around attrition, and include evidence of full reporting of all outcomes. Where possible, blinding of outcome assessment is desirable, but is not always possible to achieve in rehabilitation research. When considering methodological approaches, researchers may want to consider a range of control groups such as comparing interventions with no treatment, or alternative treatment, or an attention control. These control arms answer different but important questions.

It is important to consider follow-up and intention-to-treat analysis: these are important factors in minimising bias.

Rehabilitation trialists will find it helpful to adhere to the CONSORT guidelines for all future studies. Future definitive trials must have adequate statistical power to detect clinically meaningful differences and this may be informed by feasibility and pilot trials.

It would be helpful if researchers could agree core outcome sets and timing of measurements. Interventions should be clearly described and replicable, and researchers would benefit from adherence to the TIDieR checklist.

Future studies should include patients' and carers' views on the available interventions and the most meaningful way of measuring treatment effects. Patients' and carers' views on acceptability of available interventions and acceptability measures (adherence or satisfaction scales) should be considered in future studies. The involvement of patients and carers in commissioning and designing research would greatly increase the quality of the research discussion especially related to potential interventions and possible outcome measures. We found no studies considering timing, intensity, and duration of interventions, which are concepts of clinical importance that need to be considered in future research.

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Contributions of authors

Claire Mitchell initiated and designed the review, conducted the searches, screened and retrieved references, contacted relevant authors, obtained translations for non-English publications, requested ongoing and unpublished study information, extracted data from included trials, evaluated methodological quality, entered and analysed the data, interpreted the findings, and wrote the review. Audrey Bowen designed the review, screened references for inclusion, extracted data from included trials, evaluated methodological quality, analysed the data, interpreted the findings and contributed to the writing of the review. Sarah Tyson supported decision-making for inclusion, contributed to the writing of the review, and commented on review drafts. Zoe Butterfint commented on the final versions of the updated review. Paul Conroy designed the review, screened references for inclusion, extracted data from included trials, evaluated methodological quality, analysed the data, interpreted the findings and contributed to the writing of the review.

Declarations of interest

Claire Mitchell is a speech and language therapist and is funded by a National Institute for Health Research Doctoral Research Fellowship (DRF-2014-07-043) and is registered with the Health and Care Professions Council, UK.

Audrey Bowen's salary is part funded by Stroke Association and partly by the National Institute for Health Research Collaboration for Leadership in Applied Health Research and Care (NIHR CLAHRC) Greater Manchester. Audrey Bowen has been involved in a study included in this review (Bowen et al., 2012a). She did not contribute to the assessment or interpretation of this study.

Sarah Tyson: none known.

Zoe Butterfint: none known.

Paul Conroy is a speech and language therapist, member of the Royal College of Speech and Language Therapists, and is registered with the Health and Care Professions Council, UK.

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2.6.5 Differences between protocol and review

The title of this review was changed from "Speech and language therapy for dysarthria due to non-progressive brain damage" to reflect the broader scope of the search, which is intended to have a more global reach. The search terms for this review now include interventions carried out by any health professional, people with dysarthria, or a trained individual (whether voluntary, employed, or family member) or any other possible approaches to delivery. This review has considered any type of intervention for acquired dysarthria including behavioural or psychological approaches, use of devices and medication, with the exception of surgical intervention. This review was

also designed to reflect the international levels of functioning including impairment, activity, and participation level effects (WHO, 2001). We included an examination of risk of bias in this review in accordance with current Cochrane methodology (Higgins and Green, 2011). This review now has a summary of findings table which includes the five GRADE considerations to assess the quality of the body of evidence of the studies included in the meta-analysis using GRADEproGDT software (GRADEproGDT, 2015). The primary outcome in the protocol was to examine long-term, persistent effectiveness between three and nine months post-intervention, but during the review process, we found this time criterion was too restrictive. Following discussion among review authors the timings were relaxed to include Mackenzie (Mackenzie et al., 2014), which was felt to be the most appropriate way forward, but this was a change from the original protocol.

Table 2 Characteristics of included studies

Bowen (Bowen et al., 2012a)

Methods	Study design: RCT **Study duration**: December 2006 to end of follow up July 2010 Pragmatic, parallel, superiority RCT with blinded outcome assessment This was a larger trial of all communication impairments following stroke and the dysarthria population was a planned subgroup from this larger trial. We were able to extract the data for the dysarthria population

Participants	Inclusion criteria
	• Setting: hospital, multicentre, North West England
	• Country: England
	• Health status: people with stroke and communication impaired due to aphasia or dysarthria; considered, by the speech and language therapist, able to engage in therapy; considered, by the speech and language therapist, likely to benefit from communication therapy; informed consent or proxy consent provided by carers
	• Number: 66 participants with dysarthria randomised (from the larger trial of participants with aphasia and/or dysarthria = 170); treatment (n = 34); control (n = 32)
	• Age: (mean, SD)
	  ○ treatment: 70 ± 11.4
	  ○ control: 67 ± 11.8
	• Sex (M/F): treatment (n = 27/7); control (n = 20/12)
	• Time post stroke/brain injury: this was a trial of early intervention so participants were within the first four months post stroke: both groups median time from stroke to randomisation: 12 days
	• Severity of dysarthria: 53% severe dysarthria, both groups
	• Other communication impairment: intervention: 25/34 had aphasia; attention control: 24/32 had aphasia
	
	Exclusion criteria
	• subarachnoid haemorrhage; dementia; pre-existing learning disabilities likely to prevent benefit from therapy; unable to communicate in the English language; other serious concomitant medication conditions; patient unable to complete eligibility screening after 3 attempts over 2-week period; family or carer objections; case when a speech and language therapist was asked to contribute to an urgent assessment of a person's mental capacity to consent to an NHS treatment, before the therapist had time to complete screening to determine eligibility for the trial

Interventions	Treatment group • Intervention: intervention was multifaceted and tailored to individual needs, but consisted of 6 core components

	○ assessment & information gathering, using standardised methods ○ information provision regarding communication difficulties, intervention goals, progress, etc. ○ communication materials to record interventions & activities, plus provision of AAC devices as appropriate ○ information and training for carers ○ indirect contact with MDT colleagues regarding patient needs ○ one-to-one contact involving intervention for speech and language impairment, psychosocial impacts, activities, etc., as appropriate to the individual dysarthria ○ intervention delivered was classified according to impairment type including: impairment (97%), activity (61%), participation (61%) • Start of treatment: intervention started approximately 2 weeks after admission to hospital and before 16 weeks • Duration: lasted a maximum of 16 weeks with three contacts per week - but this was variable • Frequency: participants were seen up to 3 times per week for a maximum of 4 months, as required mean 15 hours, 20 contacts • Administration: participants were seen by a highly qualified speech and language therapist intervention was designed, implemented and monitored by qualified SLT, employed by NHS trusts. SLTs delivered most of the one-to-one contacts but some were delivered by supervised assistants. 43% contacts experienced therapist, 54% contacts less experienced therapist • Fidelity: direct monitoring of therapy sessions, case notes, goal setting audit by experienced therapist involved in study • Location: intervention took place in a number of settings as appropriate to the participant's care pathway • Adherence: 33/34 completed • Homework: advised to carry this out as frequently as possible no data on this Attention Control group • Intervention: intervention started approximately 2 weeks after admission to hospital. Sessions consisted of 3 stages: ○ building rapport and getting to know each other, finding common ground ○ regular contact sessions including general conversation and activities ○ winding down sessions • Duration: lasted a maximum of 16 weeks with 3 contacts per week - but this was variable • Frequency: sessions were 60 minutes maximum duration and tailored to individual needs, with activities being participant-led. 15 hours, 19 contacts

	• Administration: employed, part-time visitors employed to carry out structured social contact with high level educational attainment, planned and implemented by part time staff employed for the study, with no prior experience or specific training in stroke rehabilitation • Fidelity: monitor-trained visitors, supervised and monitored sessions according to protocol • Location: intervention took place in a number of settings as appropriate to the participant's care pathway • Adherence: attention control: 27/32 completed • Homework: none for control group
Outcomes	Outcomes used in this review: • Primary outcome measure: Therapy Outcome Measure (TOM) activity sub scale Secondary outcomes: • participants' perception on the Communication Outcomes After Stroke scale (COAST) • carer's perceptions of participants from part of the Care COAST • carer wellbeing on Carers of Older People in Europe Index • quality of life items from Carer COAST • serious adverse events • economic evaluation • participants' utility (European Quality of LIfe-5 Dimensions, EQ-5D) Methods to measure outcomes: Primary outcome: blinded, functional communicative ability assessed on the TOM activity sub scale. A conversation with an unfamiliar conversation partner was rated using the TOM by an expert independent expert speech and language therapist Outcomes were evaluated at baseline and 6 months post randomisation, with 2-month gap between completion of intervention and final assessment Numbers lost to follow up: intervention lost 4/34; attention control lost 8/32
Notes	Funding source: this project was funded by the NIHR Health Technology Assessment programme. The Stroke Association funded part of the excess treatment costs Contact with study authors for additional information: primary outcome reported for subgroups of diagnosis (i.e. aphasia, dysarthria); secondary outcomes not reported separately Contacted the statistician involved in this paper for the dysarthria specific data of all outcomes; this was provided in full Other: we have ensured AB, author of this trial and involved in this Cochrane review, has had no involvement in the review of this study but she contributed her opinion and provided additional information when requested

Risk of bias table (Bowen)

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Low risk	Randomisation by an external, independent, web-based randomisation service using a computer-generated string of random permuted blocks. Participants were randomised using a 1:1 allocation ratio in blocks of 2, 4, and 6 with different combinations depending on site and stratified according to severity and study centre.
Allocation concealment (selection bias)	Low risk	External, independent, web-based
Blinding of outcome assessment (detection bias)	Low risk	Outcome assessment carried out by an independent speech and language therapist, blinded to treatment allocation and not involved in treating study participants
Incomplete outcome data (attrition bias)	Low risk	ITT used and dropouts specified in report
Selective reporting (reporting bias)	Low risk	Study protocol available and all statistical data included in the report

Kwon (Kwon et al., 2015)

Methods	**Study design:** RCT: single centre, prospective, randomised, double-blind, sham stimulation-controlled trial. **Study duration:** June 2013 to April 2015

Participants	Inclusion criteria
	• Setting: Department of Rehabilitation Medicine
	• Country: Korea
	• Health status: first-ever unilateral middle cerebral artery infarction
	• Number: 25 post-stroke patients were therefore recruited and randomised into the 2 study groups. A final total of 20 of these participants completed the study
	• Age (mean, SD): intervention: 69.4 ± 11.8; attention control: 68.8 ± 9.8
	• Sex (M/F): intervention: 10/0; attention control: 7/3
	• Time post stroke/brain injury: duration from stroke onset ranged from 1 week to 2 months but all had experienced their first-ever stroke
	  ○ intervention in days: 26.4 ± 15.0
	  ○ attention control in days: 26.5 ± 12.7
	• Severity of dysarthria: not reported
	• Other communication impairment: excluded from study if any other impairment communication or cognition
	
	Exclusion criteria
	• A total of 42 participants were initially enrolled in this study, but 17 were excluded after being assessed for eligibility. Among the excluded patients, 11 did not meet the inclusion criteria and 6 refused to participate
	• Cognitive and speech function and those who had aphasia, apraxia of speech, cognitive impairment (Mini Mental State Examination < 20), poor mental status, vocal cord palsy, history of epilepsy, or bilateral infarction were excluded

Interventions	Treatment group Intervention: • this procedure was carried out as part of the intervention to establish motor-evoked potentials. To determine the resting motor threshold and stimulation area, motor-evoked potentials were recorded from the orbicularis oris muscles on each participant's non-affected side using transcranial magnetic stimulation. Focal transcranial magnetic stimulation was applied using a Magstim Rapid magnetic stimulator (Magstim Company Ltd, Dyfed, UK). Briefly, a Magstim circular coil (external diameter, 90 mm) was placed onto each participant's contralateral motor cortex to identify the hotspot, defined as the area that produced the largest amplitude of motor-evoked potentials. The resting motor

	threshold was defined as the stimulus intensity required to produce motor-evoked potentials > 100 kV at a peak-to-peak amplitude during 3 of 5 consecutive trials on the orbicularis-oris. • the experimental intervention was LF stimulation, which involved being seated in a comfortable chair with foam ear plugs, each participant was treated with 10 consecutive sessions (5 times per week for 2 weeks) of repetitive transcranial magnetic stimulation, performed by a physiatrist who used a 70 mm, air cooled, figure-of-eight Y-shaped coil. Repetitive transcranial magnetic stimulation was performed at a low frequency (1 Hz), at 90% amplitude of evoked motor threshold, and with 1,500 stimulations/day on the hotspot • this group also received speech therapy for 30 minutes, 5 days per week from a skilled speech therapist who was blind to the nature of the study during the 2-week intervention period • Start of treatment: between 1 week and 2 months • Duration: 2 weeks • Frequency: 30 minutes, 5 days/week • administration: physiatrist • Fidelity: not described • Location: Department of Rehabilitation Medicine • Adherence: 3 participants were unable to complete the study in the repetitive transcranial magnetic stimulation (10 completed) • Homework: none Control group • Intervention: • this procedure was carried out as part of the intervention to establish motor-evoked potentials. To determine the resting motor threshold and stimulation area, motor-evoked potentials were recorded from the orbicularis-oris muscles on each participant's non-affected side using transcranial magnetic stimulation. Focal transcranial magnetic stimulation was applied using a Magstim Rapid magnetic stimulator (Magstim Company Ltd, Dyfed, UK). Briefly, a Magstim circular coil (external diameter, 90 mm) was placed onto each participant's contralateral motor cortex to identify the hotspot, defined as the area that produced the largest amplitude of motor-evoked potentials. The resting motor threshold was defined as the stimulus intensity required to produce motor-evoked potentials > 100 kV at a peak-to-peak amplitude during 3 of 5 consecutive trials on the orbicularis-oris • the sham stimulation occurred using the same protocol as that for the LF stimulation, except that the angle of the coil was perpendicular to the skull rather that tangential to it. Thus, the magnetic field could not penetrate the brain, although the

	participants could hear the sound that was produced • this group also received speech therapy for 30 minutes, 5 days per week from a skilled speech therapist who was blind to the nature of the study during the 2-week intervention period • Start of intervention: between 1 week and 2 months • Duration: 2 weeks • Frequency: 30 minutes, 5 days/week • Administration: physiatrist • Fidelity: not described • Location: Department of Rehabilitation Medicine • Adherence: 3 participants were unable to complete the study in the intervention group (10 completed) • Adherence: 2 participants were unable to complete the study in the sham stimulation group (10 completed) • Homework: none
Outcomes	Outcomes used in this review. No primary outcome identified • Urimal Test of Articulation and phonology (U-TAP) • Alternative motion rates (AMR) • Sequential motion rates (SMR) • Maximal phonation time (MPT) Dysarthria was evaluated by a single skilled speech therapist who was blind to the study protocol before and after the rTMS sessions These 4 measures were carried out prior to and immediately at the end of the 2-week treatment period
Notes	Funding source: not known We were unsuccessful in contacting the first author of the study for further information

Risk of bias table (Kwon)

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Unclear risk	Randomisation using a random numbers table; odd numbers went to the repetitive transcranial magnetic stimulation group and even numbers went to the sham stimulation group although it does not specify if this was equal randomisation. Insufficient information available
Allocation concealment (selection bias)	Unclear risk	No description of what method was used to ensure allocation concealment so

		this indicates a potential risk in the absence of further information
Blinding of outcome assessment (detection bias)	Unclear risk	Study reports the outcome assessor was blinded to protocol but insufficient detail as to how this was ensured; it may have been easy to break this blinding process
Incomplete outcome data (attrition bias)	High risk	5 participants were randomised to treatment groups but then failed to complete the treatment. These participants and their data were withdrawn from all the analysis and no consideration evident as to how this missing data was dealt with
Selective reporting (reporting bias)	Unclear risk	In the absence of a protocol this remains unclear

Mackenzie (Mackenzie et al., 2014)

Methods	**Study design: a** feasibility RCT **Study duration:** enrolment within 1 year
Participants	Inclusion criteria • Setting: community • Country: Scotland • Health status: stroke: minimum of 3 months since the last stroke, and dysarthria diagnosed by a referring speech and language therapist • Number: 39 recruited and randomised; Group A had 20 participants and group B had 19 participants • Age (mean, SD): intervention A: 62.80 ± 12.52; intervention B : 67.95 ± 12.10 • Sex (M/F): intervention A: 12/7; intervention B: 14/6 • Time post stroke/brain injury: o intervention A in months: 10.84 ± 7.09 o intervention B in months 9.3 ± 5.12 • Severity of dysarthria: intervention A: mild 12/severe 7; intervention B: mild 9/severe 11 • Other communication impairment: intervention A: 6/19 had aphasia; intervention B: 6/20 had aphasia Exclusion criteria • co-existing neurological condition; Mini Mental State

	Examination score < 24; Boston Diagnostic Aphasia Examination aphasia severity rating of 4 to 5; English not first language; vision and hearing not adequate despite required augmentation

Interventions	Intervention A group
	• Intervention:
	  ○ group A had the following intervention of behavioural, activity level practice of individually relevant speech sounds in words, sentences and conversation. Strategies for optimising speech, slowed rate, emphasis of key syllables, deliberate articulation were also used as required
	  ○ group A carried out 20 minutes of word and sentence practice as part of the 40-minute session
	• Start of treatment: more than 3 months post stroke
	• Duration: 8 weeks
	• Frequency: 40 minutes once/week
	• Administration: single experienced speech and language therapist
	• Fidelity: monitored by research team and Health Boards at 2 sessions.
	• Location: participants' homes
	• Adherence: intervention A:17/19 completed
	• Homework: 10 to 15 minutes, 5 days/week (1050 minutes), recorded in diary, 85% practised 1050 minutes
	Intervention B group
	• Intervention:
	  ○ group B had the following intervention of behavioural, activity level practice of individually relevant speech sounds in words, sentences and conversation. Strategies for optimising speech, slowed rate, emphasis of key syllables, deliberate articulation were also used as required
	  ○ group B also had non-speech oro-motor exercises (impairment level) and carried out 10 minutes of word and sentence practice and 10 minutes of oro-motor exercises as part of the 40-minute session
	• Duration: 8 weeks
	• Frequency: 40 minutes once/week
	• Administration: single experienced speech and language therapist
	• Fidelity: monitored by research team and Health Boards at 2 sessions.
	• Location: participants' homes
	• Adherence: intervention B:19/20 completed
	• Homework: 10 to 15 minutes, 5 days/week (1050 minutes), recorded in diary, 85% practised 1050 minutes

Outcomes	Outcomes used in this review. No primary outcome measure identified

	• speech intelligibility at sentence level with Speech Intelligibility Test (SIT) • communication effectiveness in conversation with Communication Effectiveness Measure (CEM) • lip and tongue movement tasks from Frenchay Dysarthria Assessment-2 (FDA-2) • communicative Effectiveness Survey - self-rating of communication effectiveness Intervention A lost 4/19 to follow-up Intervention B lost 4/20 to follow-up
Notes	Funding source: Dunhill Medical Trust We requested further information, which was provided, as well as a telephone consultation We were able to classify incomplete outcome data as low risk following discussion with the study author. They clarified that they had statistically analysed their findings appropriately and this had not affected the results: "Group A versus Group B difference was not indicated on any of the four measures, based on data for 32 completing participants: SIT $F(1, 30)=1.46$, $p=0.24$; CEM $F(1, 30) = 2.39$, $p = 0.13$, CES $F(1, 30) = 0.58$, $p = 0.45$; FDA-2 $F(1, 30) = 2.61$, $p = 0.12$. There was no significant interaction between group allocation and assessment point on any of the four measures for these participants: SIT $F(3, 90) = 0.88$, $p = 0.97$; CEM $F(3, 90) = 0.34$, $p = 0.80$; CES $F(3, 90) = 0.16$, $p = 0.92$; FDA $F(3, 90) = 0.12$, $p = 0.95$. In view of the scale nature of the CEM measure, non-parametric analysis was also undertaken and provided similar results. Imputation of results for seven additional cases with incomplete intervention and/or post-intervention assessments, by last observation carried forward and multiple imputation provided similar results for all measures."

Risk of bias table (Mackenzie)

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Low risk	Randomisation was computer generated and the block system was employed to facilitate the logistics of recruitment and intervention. This would not affect sequence generation. Participants were referred in batches of 8 and then randomised within each block so 4 to group A and 4 to group B
Allocation concealment (selection bias)	Low risk	This was provided in opaque envelopes after the initial assessment by the 'assessor' and just before the intervention treatment started by the 'intervention' researcher
Blinding of outcome assessment (detection bias)	Low risk	Single blinded experienced speech and language therapy research assessor collected the outcome measurements. These were rated or transcribed by groups of blinded graduating speech and language therapy students
Incomplete outcome data (attrition bias)	Low risk	Missing outcome not likely to clinically impact, discussed with study author and confirmed all data included and adjusted where appropriate
Selective reporting (reporting bias)	Low risk	Feasibility study but all data and outcomes reported

Wenke (Wenke et al., 2010)

Methods	**Study design:** RCT; an experimental research design was used to investigate the effects of 2 treatments at multiple follow-up time points Study duration: not known
Participants	Inclusion criteria • Setting: not known • Country: Australia • Health status: 6 months post onset of stroke or brain injury • Number: 26 13 in the TRAD (traditional dysarthria therapy) intervention group and 13 in the LSVT (Lee Silverman Voice Treatment) intervention group • Age (mean, SD): total for study: 48.6 ± 21.3 • Sex (M/F): intervention A (TRAD): 7/6; intervention B (LSVT): 9/4(usual care) • Time post stroke/brain injury: o total study in years: 3.4 ± 4.75 (range: 5 to 21 years) • Severity of dysarthria: intervention A: mild/moderate 7, moderate/severe 6; intervention B: mild/moderate 7, moderate/severe 6 • Other communication impairment: intervention A cognitive impairment: 11/13; intervention B: cognitive impairment: 10/13 Exclusion criteria • Co-existing significant aphasia, hearing loss, dementia, apraxia of speech, post traumatic amnesia, or pre-existing laryngeal pathology and/or dysfunction as identified during a video laryngoscopic examination, people with a significant respiratory dysfunction unrelated to the neurological disorder; unable to speak or understand English, unable to increase/alter habitual vocal volume or quality during the pre-treatment assessment
Interventions	Treatment group A • Intervention: o TRAD used behavioural techniques at impairment and activity level. This involved phonation and/or oro-motor exercises, strategies to improve articulation, respiratory/phonatory therapy, resonance and prosody exercises. Daily 5 to 10 minutes of homework exercises. Maintenance task of exercises 5 to 10 minutes per day, 3 to 5 days a week, for 6 months were given at the end of treatment • Duration: 4 weeks • Frequency: intervention A: 1 hour/day, 4 days/week for 4 weeks • Administration: speech pathologist certified in intervention; intervention A: delivered by 1 speech pathologist • Fidelity: not described

	• Location: not known • Adherence: intervention A: all completed • Homework: intervention B: asked to practice 5 to 10 minutes daily homework during treatment. Intervention A: on completion of 4 week treatment asked to practice daily, 5 to 10 minutes, 3 to 5 days/week for 6 months. No description of whether practice was recorded and this was not reported Treatment group B • Intervention: ○ LSVT treatment was delivered in strict accordance with the manual by a therapist trained in LSVT, which employs increased vocal loudness and maximum physiological effort. Maintenance exercises were given following treatment to be carried out for 5 to 10 minutes per day, 3 to 5 days a week, for 6 months • Duration: 4 weeks • Frequency: intervention B: 1 hour/day, 4 days/week for 4 weeks • Administration: speech pathologist certified in intervention; intervention B: delivered by 1 speech pathologist • Fidelity: not described • Location: not known • Adherence: intervention B: all completed • Homework: intervention B: asked to practice 5 to 10 minutes daily homework during treatment. Intervention B: on completion of 4 week treatment asked to practice daily, 5 to 10 minutes, 3 to 5 days/week for 6 months. No description of whether practice was recorded and this was not reported
Outcomes	26 randomised Intervention A lost 4/13 to some follow-up assessments Intervention B lost 4/13 to some follow-up assessments No primary outcome measure specified. • Perceptual measure of articulatory precision and intelligibility using direct magnitude estimation • Acoustic analysis of vowels • Acoustic analysis of consonants We used intelligibility measure as the primary outcome measure at activity level and articulatory precision as the secondary impairment level measure The data presented in the paper analysed the vowels and consonants separately, which meant data extraction was not possible without further information from the authors
Notes	Funding source: not known Contact with study authors: the study authors responded to 1 email answering questions relating to randomisation. We were unable to pursue a telephone consultation with the authors to discuss further

Risk of bias table (Wenke)

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Low risk	Stratified randomisation according to severity levels was carried out and allocation based on the results of this clinical judgement. Computer generated randomisation confirmed by author
Allocation concealment (selection bias)	Unclear risk	Further information suggested a pre-generated list was used and stored on a computer in an Excel file, but it was not clear who had access to this list and how easily accessible this list was
Blinding of outcome assessment (detection bias)	Unclear risk	2 certified speech-language pathologists served as independent listeners. This implies they are not involved in the study but does not specify whether they were blind or not to the intervention
Incomplete outcome data (attrition bias)	High risk	Unable to find out more from study author; missing outcome data showing imbalance across the 2 groups
Selective reporting (reporting bias)	Low risk	All outcome measures reported at all time points

Xu (Xu et al., 2010)

Methods	**Study design:** RCT to observe the effect of acupuncture combined with speech therapy for dysarthria versus speech therapy only Study duration: not known

Participants	Inclusion criteria
	• Setting: hospital
	• Country: China
	• Health status: people diagnosed with stroke by CT and/or MRI; people diagnosed as dysarthric by the hearing and speech specialist
	• Number: 61; 30 in the intervention group (speech therapy and acupuncture); 31 in the control group (speech therapy only)
	• Age (mean, SD): intervention A: 52.6 ± 12.7; control group: 52.2 ± 12.3
	• Sex (M/F): intervention A: 23/7; control group: 26/5
	• Time post stroke/brain injury:
	  ○ Intervention A in months: 2.80 ± 2.13
	  ○ Control group in months: 2.48 ± 1.69
	• Severity of dysarthria: severe dysarthria excluded
	• Other communication impairment: excluded from study
	
	Exclusion criteria
	• Mother tongue not Mandarin; severe dysarthria or dysarthria with aphasia and apraxia of speech; cognitive impairment; could not tolerate speech therapy; Parkinsons Disease or other cerebellar lesion; myocardial infarction or renal dysfunction, severe infection or severe diabetes; unable to tolerate acupuncture, or having syncope

Interventions	Treatment group (acupuncture)
	• Intervention:
	  ○ Speech therapy intervention for both groups is impairment and activity level intervention. Breathing training, articulation work, nasality work, tone and intonation
	  ○ Acupuncture at Lianquan (CV 23), Jinjin (EX-HN 12), Yuye (EX-HN 13), Fengchi (GB 20), Yifeng (TE 17) and Wangu (GB 12) as major acupoints
	  ○ Acupuncture needles were inserted at the acupoints in different ways. The needles were pulled out when the skin sites of the major acupoints Jinjin and Yuye began to bleed. The needles inserted into the other major acupoints and additional points except these two points were left for 30 minutes at a time
	• Start of treatment: between 1 to 12 months post stroke/brain injury
	• Duration: 9 weeks with 1 week of no treatment at week 5 and speech therapy

	• Frequency: acupuncture for 30 minutes, 5 times/week and speech therapy for 30 minute sessions, 5 times per week for 9 weeks • Administration: traditional Chinese medicine specialist delivered acupuncture and speech therapy delivered by a speech therapist • Fidelity: not described • Location: hospital • Adherence: intervention A: all completed • Homework: none Control group (usual care) • Intervention: ○ Speech therapy intervention for both groups is impairment and activity level intervention. Breathing training, articulation work, nasality work, tone and intonation • Duration: 9 weeks • Frequency: 30 minutes, 5 times/week • Administration: speech therapist • Fidelity: not described • Location: hospital • Adherence: all completed • Homework: none
Outcomes	No primary outcome measure identified Outcome measures used were: • perceptual evaluation of articulation intelligibility using the Chinese Rehabilitation Research Centre Dysarthria Examination method • the maximum phonation time measuring air flow Outcome measures carried out immediately post treatment when the 9-week treatment period ended. The outcome measures were carried out before and immediately after the trial by the hearing and speech specialists who did not know the details of the trial No participants were lost to follow up from either group
Notes	Funding source: not known We were unsuccessful in contacting the first author of the study for further information

Risk of bias table (Xu)

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Unclear risk	Refers to a random number table but limited information make this judgment difficult
Allocation concealment (selection bias)	Unclear risk	There is no information about allocation concealment without further discussion with the author of the study
Blinding of outcome assessment (detection bias)	Unclear risk	The participants were tested before and after the treatment by the same hearing and speech therapist who did not know the detail of the trial. This implies they were blinded to the intervention but no further information
Incomplete outcome data (attrition bias)	Low risk	Appears to have no missing data with all participants recruited remaining in the trial to follow-up
Selective reporting (reporting bias)	Unclear risk	We were unable to verify selective reporting after an unsuccessful attempt to contact the authors

Table 3 Characteristics of excluded studies

Study	Reason for Exclusion
Behn 2011	Excluded people with dysarthria
Behn 2012	Intervention for carers not people with dysarthria
Braverman 1999	RCT; included people with communication problems other than dysarthria Intervention for cognition not dysarthria
Cohen 1993	Mixed aetiology of progressive and non-progressive adult-acquired and congenital brain injury
Fitzgerald-DeJean 2008	Not an RCT; wrong intervention (language)
Fukusako 1989	Not an RCT
Garcia 1998	Not an RCT
Huffman 1978	Not an RCT
Huh 2014	Not an RCT
Hustad 2003	Not an RCT
Ince 1973	Not an RCT
Jones 1972	Not an RCT
Katić 1973	Not an RCT
Kelly 2000	Mixed aetiology of participants, progressive and non-progressive
Li 2013	Not an RCT
Main 1998	Mixed aetiology of participants, progressive and non-progressive
Markov 1973	Not an RCT
Nagasawa 1970	Not an RCT
Palmer 2004	Not an RCT
Palmer 2007	Not an RCT
Qinglan 2002	Wrong intervention (surgical)
Robertson 2001	Not an RCT
Rosenbek 2006	Not an RCT
Sakharov 2013	Not an RCT
Sze 2002	Intervention not for people with dysarthria
Togher 2004	Intervention not for people with dysarthria
Togher 2014	Not an RCT
Varma 2004	Not an RCT

Table 4 Characteristics of studies awaiting classification

You 2010

Methods	The effects of transcranial direct stimulation (tDCS) on dysarthria in stroke patients In a prospective, double blinded, randomised case control study performed between January 2007 and December 2008, 6 people were randomised to anodal tDCS application and conventional speech therapy, and 6 participants were randomised to the sham group, which received only conventional speech therapy. tDCS was delivered for 30 minutes at 2 milliampere (mA) with 25 cm^2, five times/week, for a total of 2 weeks. The effects were assessed in maximal phonation time (MPT), alternative motion rates (AMR)-Pa, AMR-Ta, AMR-Ka, and sequential motion rates (SMR)-PaTaKa using the Multi-Media Dimension Voice Program
Participants	12 participants who developed dysarthria after acute middle cerebral artery infarction were included in this study
Interventions	Experimental intervention: anodal tDCS application and conventional speech therapy Usual care intervention: conventional speech therapy only
Outcomes	Pre-treatment patient evaluation showed no significant difference between the 2 groups for all parameters. The MPT, AMR-Pa, AMR-Ta, AMR-Ka, and SMR-PaTaKa were improved pre- and post-treatment in the stimulation group, while MPT, SMR-PaTaKa were improved in the sham group ($P < 0.05$). The AMR-Pa significantly improved in the stimulation group compared with the sham group ($P < 0.05$)
Notes	This study is in Korean and needs to be translated and data extracted before it can be considered for inclusion in the review. We were unsuccessful in contacting the first author for further information

Table 5 Characteristics of ongoing studies

Peng 2015

Study name	Modified VitalStim electroacupuncture improves the speech function in people with spastic dysarthria after stroke
Methods	32 people with spastic dysarthria after stroke within 1 month were randomly divided into VitalStim group (n = 16) and control group (n = 16). Basic medical therapy, physical therapy, occupational therapy, and speech therapy were used in both groups. Additionally, modified VitalStim electroacupuncture at acupoints of Yiming (EXHN14), Fengchi (GB20), Dazhui (BU14), Lianquan (RN23), Baihui (DU20), and lateral Jinjinyuye was performed in Vitalstim group. Participants in VitalStim group received extra 30-minute VitalStim therapy once a day, for a total of 28 days. The outcomes were evaluated by using modified Barthel index (MBI) and Frenchay Dysarthria Assessment (FDA), and the practical significance of VitalStim electroacupuncture were statistical analysed
Participants	32 participants with spastic dysarthria after stroke within 1 month
Interventions	Basic medical therapy, physical therapy, occupational therapy, and speech therapy were used in both groups. Additionally, modified VitalStim electroacupuncture at acupoints of Yiming (EXHN14), Fengchi (GB20), Dazhui (BU14), Lianquan (RN23), Baihui (DU20) and lateral Jinjinyuye was performed in Vitalstim group. Participants in the VitalStim group received extra 30-minute VitalStim therapy once a day, for a total of 28 days
Outcomes	The outcomes were evaluated by using modified Barthel index (MBI) and Frenchay Dysarthria Assessment (FDA). MBI increased significantly after treatment in both groups ($P < 0.01$). Compared with both groups, MBI increased more significantly in VitalStim group ($P < 0.05$). Significant improvements were found in VitalStim group in relation to 20 FDA items, such as lips spread, tongue at rest and palate maintenance ($P < 0.05$). The performance of the patients in VitalStim group on the rest of FDA items also showed an improvement trend compared with that of control ($P > 0.05$) except for the two items in relation to tongue alternate and jaw in speech.
Starting date	Not known
Contact information	YN Peng, Y Yin, BT Tan, W Jiang, B Zheng, YY Deng, LH Yu The Second Affiliated Hospital of Chongqing Medical University, Rehabilitation Medicine, Chongqing, China Chongqing Medical University, Rehabilitation Therapy, Chongqing, China
Notes	This study is available as an abstract only and no full report can be found. We unsuccessfully attempted to contact the authors to obtain further information about this study, including if the full study has been published.

	WCPT Congress 2015/Physiotherapy 2015; 101 (Suppl 1): eS833–eS1237 eS1189 Ethics approval: Ethical approval obtained from the Ethics Committee of the Second Affiliated Hospital of Chongqing Medical University. http://dx.doi.org/10.1016/j.physio.2015.03.2113 Research Report Poster Presentation

ReaDySpeech 2017

Study name	ReaDySpeech for people with dysarthria after stroke: protocol for a feasibility RCT
Methods	A feasibility RCT will recruit 36 people with post-stroke dysarthria who are more than 1 week post-stroke. Participants will be externally randomised in a 2:1 ratio to receive either ReaDySpeech and usual care (24 participants) or usual care only (12 participants). This study is single blind with the researcher carrying out the baseline and outcome measures blinded to treatment allocation. The primary objective is to assess the feasibility of conducting a definitive trial. Secondary objectives include recruitment rate, and determining: numbers of eligible patients recruited and reasons for non-recruitment; loss of participants to follow-up and reasons; acceptability of randomisation and the intervention; adherence to the intervention; acceptability of outcome measures; defining 'usual' care; and the implications of the intervention for the patient/family/carer
Participants	The study population includes adults (aged ≥18 years) with dysarthria as a result of stroke
Interventions	ReaDySpeech is an online programme which delivers exercises to improve breathing; intonation; facial expression; rate of speech; and oro-motor control (including range of movement, strength and speed). ReaDySpeech is set up and amended by the treating therapist according to the participant's progress. The participant accesses these exercises online, via any Wi-Fi enabled device (smart phone, tablet computer, laptop computer or personal computer). It can be used in a variety of ways: as part of face-to-face therapy during a session with a speech and language therapist or a therapy assistant, or the participant can use it independently outside of the therapy sessions, with or without the support of family or carers. The therapists select clinically relevant exercises and negotiate agreed intensity and duration of use with the participant, adherence to which is monitored by the software programme which will record the exercises selected by the therapist. Therapists will have an instruction booklet with screen shots to support their use of ReaDySpeech. The proof of concept work has shown that ReaDySpeech can be delivered by any qualified speech and language therapist of any level of experience. In this trial, participating therapists will use ReaDySpeech with participants who meet the inclusion criteria alongside 'usual' care for a maximum of 10 weeks. No specifications

	about the intensity of ReaDySpeech care will be made and this will be decided according to the therapist's clinical judgement in consultation with the participant
Outcomes	Primary outcome: Dysarthria Therapy Outcome Measure (Therapist-reported activity level measure) Secondary outcomes: COAST (communication outcome after stroke scale), Dysarthria Impact Profile (patient-reported outcome measure, activity and participation level), Frenchay Dysarthria Assessment 2nd edition (therapist-reported impairment level measure); Euroquol 5D-5L (patient-reported generic health outcome measure)
Starting date	September 2015
Contact information	claire.mitchell@manchester.ac.uk
Notes	ISRCTN84996500

2.7 Data and analyses

Table 6 Comparison 1
Dysarthria intervention compared with another intervention, attention control, placebo or no intervention: persisting effects

Outcome or Subgroup	Studies	Participants		Statistical Method	Effect Estimate
1.1 Primary outcome of dysarthria intervention versus any control: persisting effects, activity level		3	116	Std. Mean Difference (IV, Random, 95% CI)	0.18 (-0.18, 0.55)
1.2 Secondary outcome of dysarthria intervention versus any control: persisting effects, impairment level		2	56	Std. Mean Difference (IV, Random, 95% CI)	0.07 (-0.91, 1.06)
1.3 Secondary outcome of dysarthria intervention versus any control: persisting effects, participation level		2	79	Std. Mean Difference (IV, Random, 95% CI)	-0.11 (-0.56, 0.33)
1.4 Primary outcome of dysarthria intervention versus any control: persisting effects, activity level: adequate allocation concealment/adequate blinding		2	92	Std. Mean Difference (IV, Random, 95% CI)	0.21 (-0.30, 0.73)
1.5 Secondary outcome of dysarthria intervention versus attention control, placebo or no intervention: persisting effects, activity level		1	60	Std. Mean Difference (IV, Random, 95% CI)	0.00 (-0.51, 0.51)
1.6 Secondary outcome of dysarthria intervention versus any control for stroke subgroup: persisting effects, activity level		3	106	Std. Mean Difference (IV, Random, 95% CI)	0.16 (-0.23, 0.54)

Table 7 Comparison 2
Dysarthria Intervention compared with another intervention, attention control, placebo or no intervention: immediate effects

Outcome or Subgroup	Studies	Participants	Statistical Method	Effect Estimate
2.1 Secondary outcome of dysarthria intervention versus any control: immediate effects, activity level	3	117	Std. Mean Difference (IV, Random, 95% CI)	0.29 (-0.07, 0.66)
2.2 Secondary outcome of dysarthria intervention versus any control: immediate effects, impairment level	4	99	Std. Mean Difference (IV, Random, 95% CI)	0.47 (0.02, 0.92)
2.3 Secondary outcome of dysarthria intervention versus any control: immediate effects, participation level	1	32	Std. Mean Difference (IV, Random, 95% CI)	-0.24 (-0.94, 0.45)

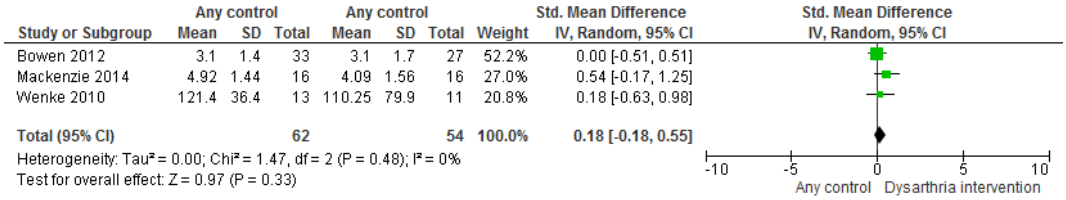
Table 8 Comparison 3
Dysarthria intervention A versus dysarthria intervention B: persisting and immediate effects

Outcome or Subgroup	Studies	Participants	Statistical Method	Effect Estimate
3.1 Secondary outcome of dysarthria intervention A versus dysarthria intervention B: persisting effects, activity level	2	56	Std. Mean Difference (IV, Random, 95% CI)	0.38 (-0.15, 0.91)
3.2 Secondary outcome of dysarthria intervention A versus dysarthria intervention B: persisting effects, participation level	1	32	Std. Mean Difference (IV, Random, 95% CI)	-0.22 (-0.92, 0.47)

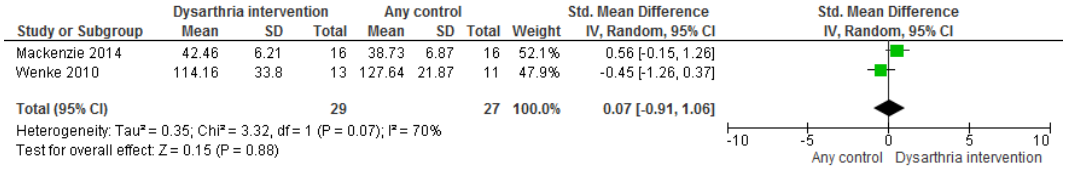
2.7.1 Comparison 1

Dysarthria intervention compared with another intervention, attention control, placebo or no intervention: persisting effects

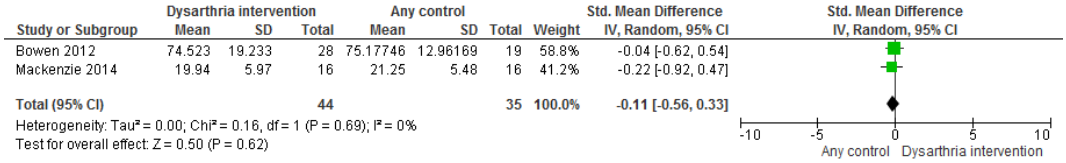
Analysis 1.1 Primary outcome of dysarthria intervention versus any control: persisting effects, activity level



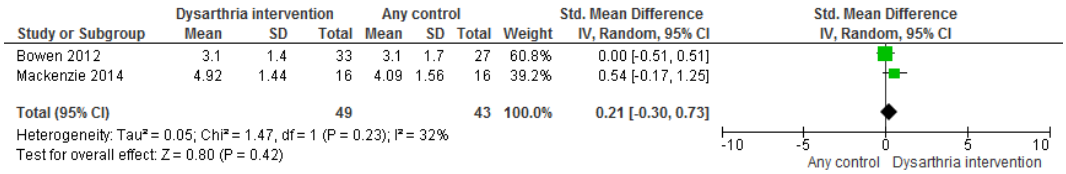
Analysis 1.2 Secondary outcome of dysarthria intervention versus any control: persisting effects, impairment level



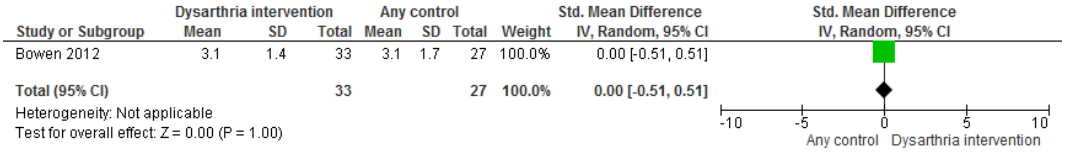
Analysis 1.3 Secondary outcome of dysarthria intervention versus any control: persisting effects, participation level



Analysis 1.4 Primary outcome of dysarthria intervention versus any control: persisting effects, activity level: adequate allocation concealment/adequate blinding



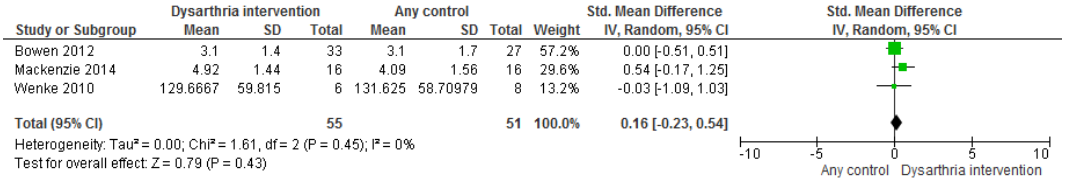
Analysis 1.5 Secondary outcome of dysarthria intervention versus attention control, placebo or no intervention: persisting effects, activity level



Study or Subgroup	Dysarthria intervention Mean	SD	Total	Any control Mean	SD	Total	Weight	Std. Mean Difference IV, Random, 95% CI
Bowen 2012	3.1	1.4	33	3.1	1.7	27	100.0%	0.00 [-0.51, 0.51]
Total (95% CI)			**33**			**27**	**100.0%**	**0.00 [-0.51, 0.51]**

Heterogeneity: Not applicable
Test for overall effect: Z = 0.00 (P = 1.00)

Analysis 1.6 Secondary outcome of dysarthria intervention versus any control for stroke subgroup: persisting effects, activity level



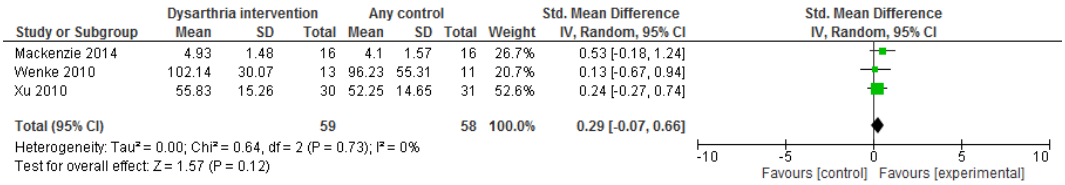
Study or Subgroup	Dysarthria intervention Mean	SD	Total	Any control Mean	SD	Total	Weight	Std. Mean Difference IV, Random, 95% CI
Bowen 2012	3.1	1.4	33	3.1	1.7	27	57.2%	0.00 [-0.51, 0.51]
Mackenzie 2014	4.92	1.44	16	4.09	1.56	16	29.6%	0.54 [-0.17, 1.25]
Wenke 2010	129.6667	59.815	6	131.625	58.70979	8	13.2%	-0.03 [-1.09, 1.03]
Total (95% CI)			**55**			**51**	**100.0%**	**0.16 [-0.23, 0.54]**

Heterogeneity: Tau² = 0.00; Chi² = 1.61, df = 2 (P = 0.45); I² = 0%
Test for overall effect: Z = 0.79 (P = 0.43)

2.7.2 Comparison 2

Dysarthria Intervention compared with another intervention, attention control, placebo or no intervention: immediate effects

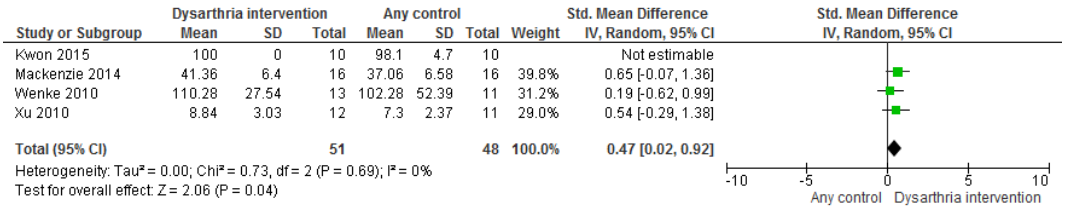
Analysis 2.1 Secondary outcome of dysarthria intervention versus any control: immediate effects, activity level



Study or Subgroup	Dysarthria intervention Mean	SD	Total	Any control Mean	SD	Total	Weight	Std. Mean Difference IV, Random, 95% CI
Mackenzie 2014	4.93	1.48	16	4.1	1.57	16	26.7%	0.53 [-0.18, 1.24]
Wenke 2010	102.14	30.07	13	96.23	55.31	11	20.7%	0.13 [-0.67, 0.94]
Xu 2010	55.83	15.26	30	52.25	14.65	31	52.6%	0.24 [-0.27, 0.74]
Total (95% CI)			**59**			**58**	**100.0%**	**0.29 [-0.07, 0.66]**

Heterogeneity: Tau² = 0.00; Chi² = 0.64, df = 2 (P = 0.73); I² = 0%
Test for overall effect: Z = 1.57 (P = 0.12)

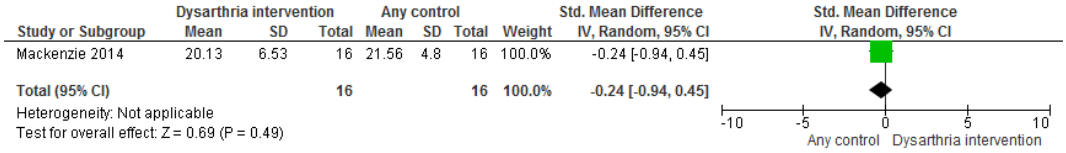
Analysis 2.2 Secondary outcome of dysarthria intervention versus any control: immediate effects, impairment level



Study or Subgroup	Dysarthria intervention Mean	SD	Total	Any control Mean	SD	Total	Weight	Std. Mean Difference IV, Random, 95% CI
Kwon 2015	100	0	10	98.1	4.7	10		Not estimable
Mackenzie 2014	41.36	6.4	16	37.06	6.58	16	39.8%	0.65 [-0.07, 1.36]
Wenke 2010	110.28	27.54	13	102.28	52.39	11	31.2%	0.19 [-0.62, 0.99]
Xu 2010	8.84	3.03	12	7.3	2.37	11	29.0%	0.54 [-0.29, 1.38]
Total (95% CI)			**51**			**48**	**100.0%**	**0.47 [0.02, 0.92]**

Heterogeneity: Tau² = 0.00; Chi² = 0.73, df = 2 (P = 0.69); I² = 0%
Test for overall effect: Z = 2.06 (P = 0.04)

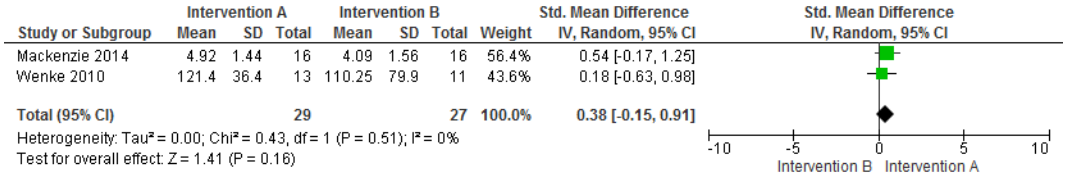
Analysis 2.3 Secondary outcome of dysarthria intervention versus any control: immediate effects, participation level



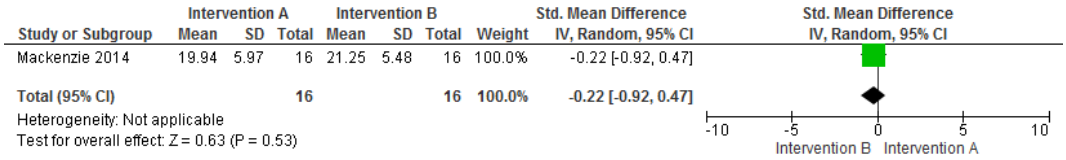
2.7.3 Comparison 3

Dysarthria intervention A versus dysarthria intervention B: persisting and immediate effects

Analysis 3.1 Secondary outcome of dysarthria intervention A versus dysarthria intervention B: persisting effects, activity level



Analysis 3.2 Secondary outcome of dysarthria intervention A versus dysarthria intervention B: persisting effects, participation level



Chapter 3 If we build it, will they use it?

Phase I observational evaluation of ReaDySpeech, an online therapy programme for people with dysarthria after stroke

This article has been published and is presented here in publication format. This chapter aims to answer the research question: Is online therapy acceptable to people with post-stroke dysarthria, their therapists and accessible in an NHS clinical context? ReaDySpeech had been developed based on current clinical NHS speech and language therapy usual care based on best practice profession specific guidelines and what is typically delivered (Taylor-Goh, 2005). ReaDySpeech was developed through working with therapists and patients using small focus groups and interviews. The intention was for ReaDySpeech to build on the motor learning principles described in the introductory chapter particularly around considering increasing intensity.

Mitchell C, Bowen A, Tyson S, Conroy P: **If we build it, will they use it? Phase I observational evaluation of ReaDySpeech, an online therapy programme for people with dysarthria after stroke.** *Cogent Medicine* 2016:1257410.

https://www.cogentoa.com/article/10.1080/2331205X.2016.1257410

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3.1 Abstract

Purpose: To explore the acceptability of using ReaDySpeech, an online speech therapy programme for people with dysarthria after stroke, within usual clinical practice. This early clinical testing underpins future research evaluation of ReaDySpeech.

Methods: A prospective, observational design involving interviews with speech and language therapists with experience of using ReaDySpeech. This included the usability of ReaDySpeech, therapists' training/support needs, ease of recruitment of therapist and patient participants, ReaDySpeech technical issues and therapy content. Therapists also provided feedback from the patient participants.

Results: Six therapists working in hospital and community-based settings used ReaDySpeech with five patients (12-28 weeks post-stroke, four female, mean age 71 years). Therapists found it was easy to use, training/support was sufficient and they reported positive feedback from participants. Areas to address involved patients' access to Wi-Fi, ease of navigation, content improvements and difficulties recruiting people more than 12 weeks post-stroke as most patients had already been discharged.

Conclusions: ReaDySpeech was acceptable and generally feasible to use in clinical practice. This early phase research testing has been essential to improve navigation within the therapy software and content. ReaDySpeech can now be further evaluated with a phase two feasibility trial with earlier recruitment following stroke.

3.2 Introduction

Dysarthria following stroke results in impaired intelligibility of speech from the neurologic damage that causes speech musculature to be slow, weak, imprecise and/or poorly coordinated (Yorkston, 1996). This can negatively affect an individual's sense of identity, self-image, social participation and psychological well-being (Tilling et al., 2001; Dickson et al., 2008; Brady et al., 2011a). Research into treatment of post-stroke dysarthria is limited; no high quality trials were identified in a Cochrane review (Sellars et al., 2005). Research into motor learning after stroke indicates that although increased intensity and duration of treatment may improve recovery the increase in therapist time would be costly for health service providers (Langhorne et al., 2009). The use of electronic technology (or e-rehabilitation) may be a way to increase the dose of dysarthria treatment without increasing the therapist demand and is being explored for wider stroke rehabilitation (Mawson et al., 2014; Lindqvist and Borell, 2012). Recent studies have suggested that using technology increased the amount of therapy received by patients and may benefit patients (Palmer et al., 2007; Palmer et al., 2012; Lemoncello et al., 2011). It is also important to ensure patients have the opportunity to access high quality independent practice so they can make that choice if they wish, to support patient centred intervention.

This report details early work to assess the acceptability of using a novel computerised rehabilitation programme for people with dysarthria following stroke. The original idea for the technology (called ReaDySpeech) was suggested to the author (CM) by patients with post stroke dysarthria. They commented that generic, paper exercises which are part of standard care in the UK were not particularly easy or motivating to use and asked if these could be computer based. This suggestion coupled with the need for

greater intensity, repetition and functional activities led to the development of ReaDySpeech which has the potential to use technology to improve the quantity and quality of therapy and ultimately, the outcomes for stroke survivors. Extensive searches indicated that there were no complete commercial computer-based programmes specifically for dysarthria. Thus 'ReaDySpeech', an online programme that could be tailored for individuals by a speech and language therapist was developed by the first author (2014) in collaboration with speech and language therapists and stroke survivors with dysarthria. The content was based on best practice guidelines 12 including exercises for facial and oral muscles and strategies for increasing intelligibility. We now report this initial proof-of-concept work to explore acceptability of ReaDySpeech and whether it should progress to further evaluation of efficacy as outlined by the MRC Framework for the Development and Evaluation of RCTs for Complex Interventions to Improve Health (Campbell et al., 2000; Anderson, 2008). If we found ReaDySpeech acceptable for clinical use we would then proceed to a feasibility randomised controlled trial, which if feasible would allow us to proceed to a larger trial of efficacy in the future.

Aims: The main objective is to find out if ReaDySpeech is acceptable to use during every-day clinical practice. The other objectives are to: establish if it is possible to recruit NHS therapists to carry out the testing, identify and recruit patients with dysarthria more than 12 weeks post stroke.

The Intervention

ReaDySpeech is a dysarthria programme that aims to rehabilitate speech at impairment and activity level. It includes exercises to improve articulation; breathing; intonation; facial expression; rate of speech; and range of movement, strength and

speed of the oro-motor musculature. It is intended that it will be suitable for people in the acute and chronic stages of recovery and can be delivered alongside therapy to increase participation and other aspects of speech and language therapy. It is anticipated that ReaDySpeech can be used in a variety of ways: as part of face to face therapy with a speech and language therapist or a therapy assistant practitioner, or the person with dysarthria can use it independently outside of the therapy sessions, with or without the support of family or carers. ReaDySpeech is set up and amended by the treating therapist according to the patients' level of difficulty and rate of progress. The therapist selects clinically relevant exercises and negotiates agreed intensity and duration of use with the patient, adherence to which is monitored by the software programme. The patient is then able to access these exercises online, on any Wi-Fi enabled device (smart phone, tablet computer, lap top computer, personal computer). In this study, participating therapists used ReaDySpeech with people who met the inclusion criteria (details below) alongside 'usual' care for up to 10 weeks. The 'usual' care intervention was not specified and was provided by the treating therapist as they deemed appropriate. No specifications about the intensity of ReaDySpeech care were made and this was decided according to the therapists' clinical judgement.

3.3 Methods

Ethical approval

Ethics approval for the study was obtained from the UK NHS research ethics committee (REC reference number: 14/SC/1320) and research permissions gained from NHS Trusts.

Two groups of participants

1. **Group 1: Qualified speech and language therapists** (of any grade) who worked with people with stroke, in acute care, rehabilitation or community settings in the four participating stroke services were eligible to participate. We aimed to recruit a minimum of four therapists, across the sites so one per site, with each expected to recruit one or two patients over a five month recruitment period.
2. **Group 2: Patients with post-stroke dysarthria**, who were known to participating speech therapy services, more than 12 weeks post-stroke (no upper time limit), willing and able to undertake and benefit from dysarthria therapy (in therapists' opinion), medically stable and able to give informed consent were recruited. The exclusion criteria for people with dysarthria were any co-existing neurological condition, needing a translator to participate in therapy, significant hearing, physical, cognitive, language or visual problems that would prevent using ReaDySpeech. Therapists kept a log of patients who were ineligible and why, and those who were eligible but declined to participate and why.

Procedure

A prospective, observational design was used to interview the participating speech and language therapists. Therapists who consented to participate were given training and support about the study and in how to deliver therapy using ReaDySpeech. An instruction booklet for ReaDySpeech and face to face training were provided. Therapists then used ReaDySpeech with recruited patients, as described above, as part of usual care. They would set up each patient with access to ReaDySpeech, selecting the specific exercises that were needed and either go through these with them as part

of their therapy session or ask them to go through the programme independently. Therapists were able to borrow a tablet computer if needed or would use the patient's own device.

On completion of the intervention period the first author interviewed therapists face to face, using a semi-structured questionnaire containing open and closed questions, and recorded the responses in writing. The open questions asked for comments e.g. 'please comment on ease of use' and the closed questions offered a rating scale: very easy to use, easy to use, not sure either way, not particularly easy to use, not at all easy to use. The questions covered four key areas: i) ease of use of ReaDySpeech and therapists' training and support needs; ii) patients' views as communicated to therapists; iii) technical issues related to use in various settings; iv) the content of ReaDySpeech, strengths and/or weaknesses of the programme and how it could be improved.

The data analysis plan was to summarise the quantitative and qualitative data, drawing out practical suggestions for developments to the ReaDySpeech programme and to finalise the design of a subsequent phase 2 feasibility randomised controlled trial.

3.4 Results

Eight speech and language therapists were recruited, six of whom identified and recruited suitable patients from three sites. Two therapists from a fourth site were unable to recruit patients, due to a lack of Wi-Fi at a rehabilitation centre, and could therefore not continue further with the project. The six included therapists were female with a mean of 11.5 years clinical experience.

Ten eligible patients with dysarthria were identified, five of whom participated. Three declined (two because they disliked computers and one because they feared it might

delay discharge from in-patient care) and two could not participate because they did not have access to Wi-Fi in a rehabilitation facility. Four of the five participating patients were male, with a mean age of 71 years (61-76 years) and mean time post stroke of 16 weeks (12 to 28 weeks). All had dysarthria in the absence of aphasia (language impairment). All five participants had suffered an ischaemic stroke; one was lacunar; one was in the posterior circulation; two were in the anterior circulation and one location was unknown. Two participants were recruited from an-inpatient rehabilitation unit from one site; two were recruited in their own homes from the community-based service of a second site and the fifth was recruited from the in-patient rehabilitation unit of the third site.

The training to use ReaDySpeech was rated as 'thorough' by five therapists, with one rating of 'unsure'. They highlighted that training needed to be flexible to be appropriate for a range of therapists' ability to use technology. All six therapists reported that they accessed support via email or phone call and found this sufficient. They rated ReaDySpeech as 'easy' or 'very easy' to use in terms of selecting the relevant exercises and setting up individual programmes for patients and their five patients were able to use ReaDySpeech with no reported difficulties. All the patient participants completed the intervention, with no drop-outs. Three of the five patients' provided feedback to therapists including that it was "*fine"*, another patient "*liked it"* and reported it was better than "*the other speech therapy"* meaning the paper-based work used prior to involvement in the study. One patient commented that it was "*easy to use"* and liked the fact it was "*more environmentally friendly"* than the paper-based exercises. Therapists were also positive about using ReaDySpeech reporting that they felt it was "*more motivating for patients*" and "*more interactive for patients*" and "was *more professional*" than using paper exercises, as well as "*easier than photocopying*".

Therapy was delivered equally in community and in-patient settings. The therapists also discussed how they had used the programme in different ways. At one site ReaDySpeech was used with a therapy assistant; at another, therapists used it during their therapy session; in the third site, ReaDySpeech was used by one patient independently and with another as part of their therapy session. Several suggestions about potential improvements were made, mostly regarding changes to the functionality of ReaDySpeech to improve navigation around the programme (n=15 comments) or to enhance the content (n=11). Technical difficulties were raised eight times and related to limited Wi-Fi access in clinical settings.

Therapists reported that identifying patients with dysarthria at more than 12 weeks post stroke was difficult as most patients had been discharged from speech and language therapy by this point. They felt the intervention could have been used with people earlier post-stroke. An additional four people with dysarthria were ineligible for this initial study because they were less than 12 weeks post-stroke.

3.5 Discussion

Ease of recruitment and a willingness of therapists to engage in testing out new technology indicates a future study is possible. This study found that while therapists were easily recruited, patients more than twelve weeks post stroke were harder to identify. Our subsequent studies will recruit people earlier after stroke to more accurately reflect clinical practice and the population receiving dysarthria rehabilitation. The findings showed that ReaDySpeech was easy to use and the support and training sufficient according to the therapists. All patient participants were able to use ReaDySpeech with no difficulties reported to the therapists. This early clinical testing has provided further feedback and suggestions to improve the functionality and

content of ReaDySpeech. This study did not specify how ReaDySpeech should be used, so the predominant use by the therapist or therapy assistants as part of face to face therapy provides some useful indicators for further evaluation.

This study wanted to establish, not just recruitment rate of patients, but willingness to participate in technology clinical testing. As most stroke patients are older and presumed to be less familiar with technology concerns had been raised in the initial development phases that this population may not wish to engage with technology testing. Two of the 10 eligible patients approached declined participation due to a dislike of computers. Although a very small sample this suggests that concerns about the willingness of patients following stroke to engage with technology testing may be exaggerated and do not mitigate against progressing development of ReaDySpeech.

The ease of use reported by therapists and patients indicated that this programme could go forward to further evaluation without the need for significant changes. However, suggested improvements have been incorporated for future evaluation. This included enhancing the 'user manuals' with ongoing phone support and demonstration videos.

3.5.1 Study Limitations

The small numbers of participants mean that generalisability is limited and one cannot assume that the same results would be found if used in other rehabilitation units, more varied clinical context or with a wider range of participants. Furthermore the lack of a control group and a direct evaluation of patients' views adds potential bias to the results. They do however give us the initial proof of concept that support our plans to progress to a larger phase II feasibility trial.

3.5.2 Interpretation

This study found that it was feasible to use ReaDySpeech in clinical practice and it was acceptable to patients and therapists. We were able to recruit therapists and eligible patients were willing to participate in testing rehabilitation regardless of age but the inclusion criteria for time post stroke would need to be earlier. Amendments have been made to ReaDySpeech to improve content and functioning, with training and support remaining flexible according to need. ReaDySpeech will continue to be used in whatever way is most suited to the patient according to clinical need.

3.5.3 Future directions

This study has shown the importance of early development work around a novel intervention and the methodology most suited to examining ReaDySpeech in more depth. It has provided the information needed to ensure a larger, more costly phase II feasibility trial is well designed. This testing phase now means that a phase II feasibility, randomised controlled trial comparing 'usual care' with ReaDySpeech is warranted and this is currently underway.

Chapter 4 ReaDySpeech for people with dysarthria after stroke: protocol for a feasibility randomised controlled trial

This paper has been published in Pilot and Feasibility Studies

Claire Mitchell[1,3], Audrey Bowen[1], Sarah Tyson[2] & Paul Conroy[1]

This paper is addressing the research question: Can we design a feasibility randomised controlled trial for an online therapy?

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4.1 Abstract

Background: Dysarthria, disordered speech production resulting from neuro-muscular impairment, is a common symptom after stroke. It causes significant problems for patients' speech intelligibility, communication, psychological well-being, social engagement and stroke recovery. Rehabilitation for dysarthria is variable in quality, intensity and duration, which may be, in part, due to the lack of good quality evidence. An online therapy programme, ReaDySpeech, has the potential to improve quality, intensity and duration of speech rehabilitation and was considered in a proof-of-concept study to be acceptable to speech and language therapists and patients which warranted further evaluation. The present study aims to examine the feasibility of running a trial using the ReaDySpeech intervention.

Methods/Design: A feasibility, randomised controlled trial, will recruit a minimum of 36 people with post-stroke dysarthria who are more than one week post stroke. Participants will be externally randomised in a 2:1 ratio to receive either ReaDySpeech and usual care (24 participants) or usual care only (12 participants). This study is single blind with the researcher carrying out the baseline and outcome measures while blinded to treatment allocation. The primary objective is to assess the feasibility of conducting a larger phase III trial. Specific objectives are to determine: recruitment rate and reasons for non-recruitment; loss of participants to follow up; acceptability of randomisation; adherence to the intervention; delivery of ReaDySpeech and content; acceptability of outcome measures; success of blinding strategies; defining 'usual' care; and, the implications of the intervention for the patient/family/carer.

Discussion: This study will involve a regional, multi-centre, randomised controlled feasibility trial of a complex intervention in order to evaluate whether a phase 3 randomised controlled trial is feasible.

Trial registration: Current Controlled Trials ISRCTN84996500

Keywords: Dysarthria, stroke, Speech/language therapy (SLT), Feasibility, Randomised controlled trial

4.2 Background

Stroke is the second leading cause of death worldwide (Feigin et al., 2014) and approximately 20-30% of stroke survivors (Warlow, 2001) will experience dysarthria. Dysarthria following stroke has been found to have a negative impact on functional recovery, psychological well-being, social engagement and participation (Tilling et al., 2001; Brady et al., 2011b; Dickson et al., 2008). Dysarthric speech is less intelligible than that of healthy individuals due to poor control of oral articulator muscles, particularly the tongue and lips and poor respiratory control. Dysarthria affects people in many different ways depending on which muscle groups are impaired such as unclear articulation of words, nasal speech or a quiet voice with no expression for example. This variation in presentation also includes a wide severity range with some patients having no useful speech while at the milder end speech is generally intelligible but there may be lapses in speech accuracy or fatigue. The extent of disability also varies according to an individual's communication demands, such as work and social situations, where mild dysarthria can be hugely disabling.

The evidence base regarding treatment for dysarthria after stroke is limited by a lack of adequately powered, well controlled trials. A Cochrane review found no trials (Sellars

et al., 2005) and a more recent update found that while five trials could be included these were considered low to very low in quality (Mitchell et al., 2017a). These more recent randomised controlled trials are inconclusive about which intervention for dysarthria rehabilitation is most effective (Mackenzie et al., 2014; Bowen et al., 2012a; Xu et al., 2010; Wenke et al., 2010; Kwon et al., 2015). Thus further high quality trials are needed to benefit people with dysarthria given the potential for depression, social exclusion and worse quality of life (Brady et al., 2011b; Dickson et al., 2008; Mackenzie, 2011). Researchers and therapists continue to evaluate and seek guidance about which intervention works best for dysarthria post stroke and what frequency and duration of intervention will give the best outcomes (Mackenzie, 2011). There is growing interest in the use of computer technology to help patients access therapy for dysarthria after stroke and in doing so, enhance the individualisation and intensity of treatment delivery as well as ensuring choices are available so patient centred care is accessible. Using technology to support dysarthria intervention could be cost effective and enhance clinical and patient-reported outcomes from rehabilitation after stroke.

This paper summarises a protocol for our feasibility trial of the online programme 'ReaDySpeech' for people with dysarthria, accessed via any Wi-Fi enabled device. In terms of the ICF (International Classification of functioning, disability and health (WHO, 2007)) the ReaDySpeech programme addresses dysarthria impairment (improving speech musculature) as well as activity (compensatory strategies) levels. This programme was initially developed with exploratory interviews with speech and language therapists and patients (Mitchell et al., 2016), and then further enhanced by input from a research advisory group 'Ever-ready', made up of patients who have experienced speech problems after stroke. ReaDySpeech can be used in the acute and

chronic stages of recovery and can be delivered alongside work to address participation. Proof-of-concept testing found ReaDySpeech to be acceptable to therapists and patients when used in a clinical context (Mitchell et al., 2016). This protocol is the next step in evaluating ReaDySpeech. Here we describe a Phase II feasibility trial which will enable us to determine the feasibility of carrying out this research on a larger scale (Campbell et al., 2000; Anderson, 2008). This protocol has followed the CONSORT (CONsolidated Standards of Reporting Trials) guidelines (Schulz et al., 2010) and the SPIRIT (Standard Protocol Items: Recommendations for Interventional Trials) statement (Chan et al., 2013) as well as the TIDieR (Template for Intervention Description and Replication) checklist and guide (Hoffmann et al., 2014b).
Aim: The primary objective is to assess the feasibility of conducting a Phase III randomised controlled trial comparing ReaDySpeech with usual care versus usual care only.

4.2.1 Study objectives:

To determine:

1) number of participants eligible for the study, recruitment rates and reasons for declining, retention rates and reasons for loss of patients for future trial sample size
2) delivery of ReaDySpeech and content selection
3) adherence to the technology in the intervention arm
4) content of 'usual' care: activities, intensity, duration
5) clinical utility and acceptability of outcome measures to patients and effectiveness of blinding
6) impact of intervention on patient, family/carer/partner

4.3 Methods/design

Study design: This is a feasibility, single blind, individually randomised controlled trial of ReaDySpeech with usual care versus usual care.

Setting

This is a multi-centre study recruiting from four NHS sites in North West England over 14 months. Recruitment and treatment will take place in hospital and community locations including patients' homes.

Population: The study population includes adults (aged ≥18 years), with dysarthria as a result of stroke.

Entry criteria for participation in the trial

Inclusion criteria are as follows:

1. Diagnosis of dysarthria caused by stroke.
2. More than one week post stroke, no upper time limit.
3. Medically stable, as judged by the clinical team.
4. Considered, by their speech and language therapist, to be likely to benefit from speech rehabilitation.
5. Sufficient English to participate in therapy without a translator.

Exclusion criteria are:

1. Co-existing progressive neurological conditions.

2. Co-existing communication, cognitive, hearing or visual problems significant enough to prevent use ReaDySpeech.

Identification and recruitment of trial participants

Potential participants will be identified by NHS speech and language therapists in the participating stroke services from their patient caseloads including hospital and community settings. The ReaDySpeech study screening log will be completed by the speech and language therapists to record the number of dysarthric stroke patients who are not eligible for the study and the reasons why, and the number of those who are eligible for the study but decline participation including their reasons if provided. The speech and language therapists will identify patients meeting the inclusion criteria, give out the patient information sheet to those willing to find out more, and inform the researcher (with the potential participant's agreement). The researcher will contact the patient once they have had 24 hours to read the study information and answer any questions. If the participant meets the inclusion criteria and is willing to participate, the researcher will obtain signed, fully informed consent in line with Research Ethics Committee guidance and Good Clinical Practice Standards. Baseline assessments will be carried out at this point prior to randomisation and intervention will start immediately after allocation.

Randomisation

Participants will be randomised using a 2:1 allocation ratio to ReaDySpeech and usual care (intervention arm) or usual care only (control arm) respectively. The primary researcher will enter minimal anonymised patient details onto the external web based randomisation programme held by an independent clinical trials unit to ensure

allocation concealment. The programme will generate an un-blinded email to the treating therapist, who will inform the patient of their treatment allocation when intervention starts. Minimisation will be stratified by the four sites and by acute stroke (≤12 weeks post stroke) or chronic stroke (≥12 weeks post stroke).

Blinding

Given that rehabilitation involves active participation it is not possible for the participants and treating therapist to remain blinded to the treatment allocation. The primary researcher will be blinded to the intervention allocation and ask the speech and language therapists delivering the intervention and the patients or family to not to reveal the treatment allocation. They will be reminded of this at the start of every interaction. The primary researcher will remain blind until after the outcome measures have been carried out at 8/10 weeks post randomisation. At that point the researcher will record which group they thought the patient was in to look at the effectiveness of the blinding process as well as documenting any reasons where blinding was affected.

Sample size justification

No formal sample size calculation was carried out. Thus the sample size for this feasibility trial was governed pragmatically by the resources available. Based on our experience of undertaking communication related trials we decided to recruit for 14 months, from four NHS sites in the North West of England, estimating this would provide around 36 participants. This will give us an indication of the expected variability of service delivery, resource availability and produce more generalizable recruitment and retention rate. Recruitment will be reviewed 4 months into the study to whether the recruitment strategy needs to change. The 2:1 allocation provides a

larger group with whom to explore intervention delivery and fidelity. It means the smaller group would have a minimum sample of 12 which is considered acceptable for a feasibility study (Julious, 2005).

Description of the Intervention

ReaDySpeech

ReaDySpeech is an online programme to deliver dysarthria therapy at impairment and activity levels of functioning for people following stroke. The intervention is described in detail in Appendix 7 following the TIDieR checklist (Hoffmann et al., 2014b). ReaDySpeech will record what exercises were prescribed by the therapist and which were completed by the patient.

Usual speech and language therapy care

Usual speech and language therapy will be accessed by those randomised to the control group as well as the ReaDySpeech arm. This would be expected to follow existing best practice guidelines which address impairment, activity and participation levels of functioning (Taylor-Goh, 2005) and is described in detail in Appendix 7 following the TIDieR checklist (Hoffmann et al., 2014b). The frequency, duration and content of the sessions will be extracted retrospectively from the clinical speech and language therapy notes by the primary researcher in partnership with the therapist.

Assessment of objectives

Feasibility will be determined by the recruitment and retention rates found at the 4 sites over the 14 month trial. This will enable us to look at the sample size needed and the number of sites required to recruit to a phase III trial from a formal power

calculation. Data on reasons for exclusion and eligible participants declining involvement in the study will also help to assess whether this study is feasible for a larger population and whether recruitment should be amended in any way.

Fidelity to look at how ReaDySpeech was delivered will cover delivery, access and support. We will record who delivered ReaDySpeech, whether this was independent use, therapist, assistant or family led. Whether computers were loaned to participants and whether participants were supported and trained to use the programme. Adherence data will also be examined to assess participants' use of ReaDySpeech. The programme software will record exercises selected by the therapist and which of these exercises are recorded online as having been completed by the participant. The usual care provision adherence data will be taken from the clinical case notes and will allow us to describe current provision of 'usual' speech and language therapy in the four participating sites with a potential to formulate what this could look like in a future trial.

Following completion of the outcome measures, patients will be asked structured and open questions by the primary researcher in a face to face interview, and answers will be written down. The questions will explore four key areas: i) what the participants thought about the research study, whether they understood the study and what was going to happen, their views on randomisation, whether the time taken for the outcome measures was acceptable and whether they felt the outcome measures reflected their views of their speech; ii) what was delivered in 'usual' care and/or ReaDySpeech from their perspective including who delivered it and when; iii) the impact of 'usual' care or ReaDySpeech on themselves, family, partner, carer; iv) any

other comments on the study and/or the interventions for the research team to consider.

This will enable the research team to consider the implications of participating in this research and participant's views of the study itself as well as the acceptability of the outcome measures.

Baseline measures: Demographic data (age and gender), stroke information (time since stroke, type of stroke - haemorrhagic or infarction, and stroke classification), levels of pre-morbid and current functioning (modified Rankin Scale (Vanswieten et al., 1988)) and current activities of daily living (Barthel Index, 10 item scale with 5 point increments, based on Mahoney and Barthel's tool (Mahoney and Barthel, 1965; Collin et al., 1988; Quinn et al., 2011)) and the co-existence of other language impairments such as aphasia (severity, how it was diagnosed) will be extracted from notes and documented prior to randomisation. Measures completed at baseline, prior to randomisation, will be: therapist reported speech at activity level (Dysarthria Therapy Outcome Measure, TOMS Activity score (Enderby et al., 2013)); patient reported communication at activity and participation level (Communication after Stroke Scale, COAST (Long et al., 2008)); patient reported communication at activity and participation level (Dysarthria Impact Profile, DIP (Walshe et al., 2009)); therapist reported speech at impairment level (Frenchay Dysarthria Assessment 2nd edition, FDA II (Enderby and Palmer, 2008)); and, a patient reported health outcome measure (EQ-5D-5L (Herdman et al., 2011)) that may provide useful feasibility data for a future cost effectiveness study.

Outcome measures: The measures carried out at baseline will also be carried out at the end of intervention (8/10 weeks post randomisation) by the primary researcher

either in hospital or the participants home (if they have been discharged). Change from baseline scores will be examined to determine the sensitivity of the measures.

Assessment of blinding

To assess the effectiveness of the blinding, the primary researcher will guess group allocation for each participant once the outcome measures have been completed. They will then check this guessed allocation against the randomised allocation record to examine effectiveness of blinding when the database is locked and the code un-blinded.

Data management

Personal data, case report forms and participant questionnaires will be treated as confidential documents and held securely in accordance with the NHS research ethics committee regulations as outlined in the ethics approval process. Each consenting participant will have a unique identifier that will be used for randomisation and identification. The externally held randomisation programme at the clinical trials unit will have no identifying patient information and all data entered on the programme will be stored securely in accordance with the standard operating procedures at the clinical trials unit. There will be no dates of birth or NHS numbers recorded during this study, only age. Time since stroke will be recorded, not the date of the stroke. A data monitoring committee was not deemed necessary for this small feasibility trial given the intervention (online speech rehabilitation) is so low risk and not expected to lead to serious related adverse events.

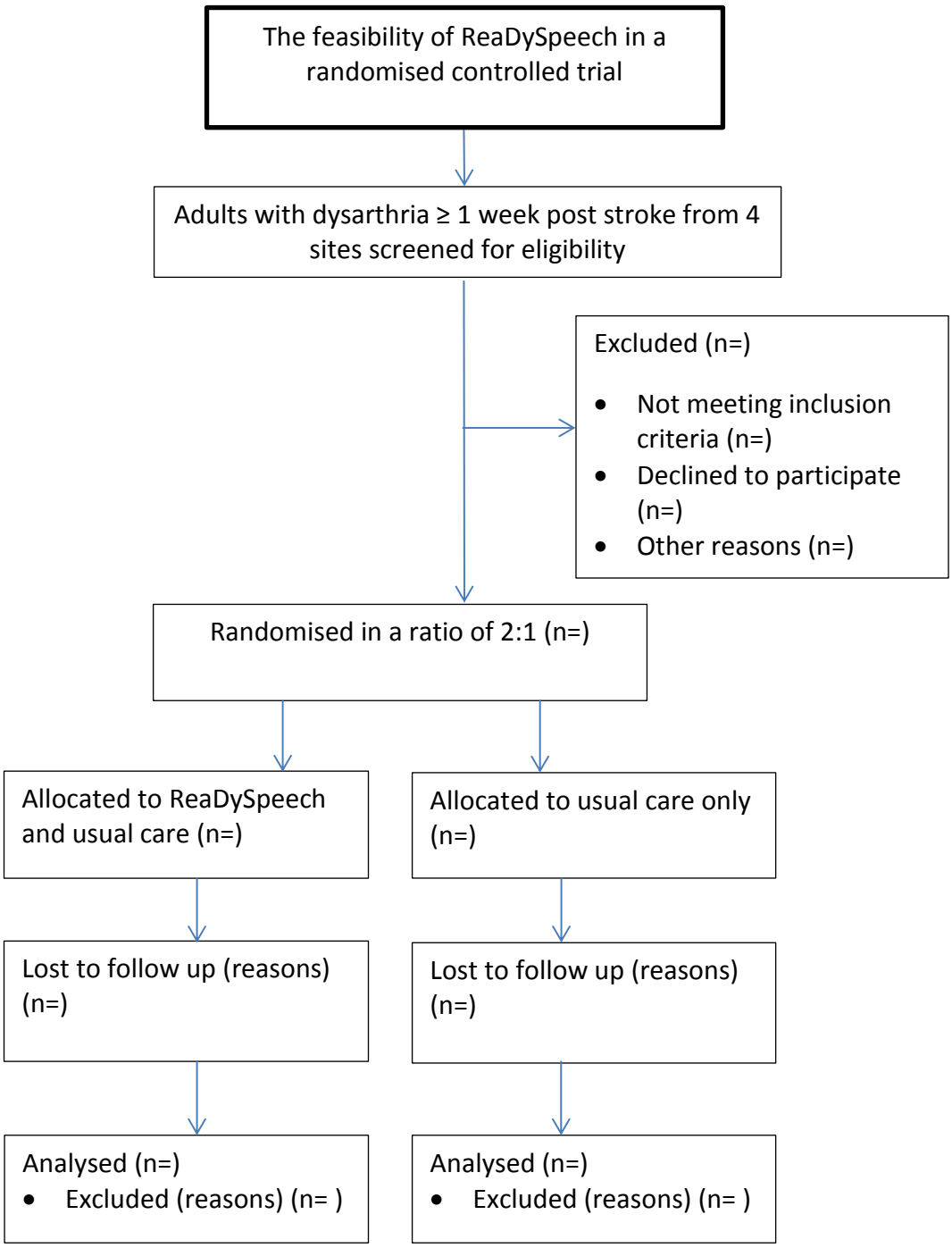
Data analysis

Analysis for this single blind, multi-centre, feasibility randomised controlled trial will be descriptive as the study is not designed to look at the effect of the intervention and would not have sufficient statistical power.

Descriptive summary statistics will consider the numbers of patients who were eligible, recruitment to the study and attrition rates according to site and intervention arm. A CONSORT (CONsolidated Standards of Reporting Trials) and SPIRIT (Standard Protocol Items: Recommendations for Interventional Trials) flow chart will present the overall recruitment to the study (Chan et al., 2013) (Figure 4).

Demographic data will be examined to identify if there are any patterns of recruitment related to age, gender, stroke type, severity of disability or health, whether the randomised groups are balanced at baseline and whether this recruited sample is reflective of the stroke population. We will consider change between baseline and follow up measures to consider possible effect sizes of the potential primary outcome measure for the future trial. The recruitment and retention rate data will support the sample size calculation for a future larger trial and the likely number of sites needed to achieve this sample. No analysis of outcome measures will be undertaken until all follow up assessments have been completed. Both the ReaDySpeech and usual care therapy provided will be described including fidelity and adherence data and interview data (Hoffmann et al., 2014b) (Appendix 7).

Figure 4 ReaDySpeech participant flowchart through trial



Safety monitoring and adverse events

A risk assessment on using the technology as part of this trial indicates that risk of harm is low. The proof of concept work also showed it was safe (Mitchell et al., 2016). Any previously unidentified risks of the experimental intervention will be documented and reviewed with the study sponsor. The study can be audited at any point by the funders, sponsor or REC and documentation for this purpose will be maintained in a study master file.

4.4 Discussion

This study will provide evidence for the feasibility of a randomised controlled trial into the effectiveness of ReaDySpeech for people with dysarthria after stroke. The qualitative and quantitative data produced will inform the decision about the potential for a subsequent trial. The paucity of existing randomised controlled trials of interventions for dysarthria after stroke mean the findings will also be of interest to other researchers working in this area or wanting to examine recruitment for other technology studies in similar populations. The likely variations in usual care will be an additional finding from this research that will be of interest to both researchers and therapists.

This feasibility trial's findings will be presented at national and international stroke and rehabilitation conferences and submitted for publication in peer-reviewed journals. They will also be disseminated to stroke survivors and study participants in a user-friendly format which will be produced in partnership with our research advisory group 'Ever-ready'. Finally the results will also be disseminated through social media.

Trial status

Participant recruitment started in September 2015 and is due to finish recruiting by the end of October 2016. The trial is registered with ISRCTN84996500

List of abbreviations

CTU: clinical trials unit; ISRCTN: numerical identification of randomised controlled trials; NHS: UK National Health Service; ReaDySpeech: online programme for the rehabilitation of dysarthric speech; RCT: randomised controlled trial; SLT: speech and language therapy or therapists; Dysarthria TOMs: Therapy Outcome Measure specific to dysarthria.

Declarations

Ethics approval and consent to participate

Ethics approval for this study was granted by the UK National Research Ethics Service Committee Northwest (15/NW/0371) and local research and development management approval was granted by the four NHS trust sites involved in the study. The trial will be conducted in compliance with the Declaration of Helsinki. Informed written consent will be obtained from all participants. Any significant protocol amendments will be discussed with the funder, sponsor and REC and updated protocols will be submitted.

This study is sponsored by the University of Manchester, Faculty of Biology, Medicine and Health, Oxford Road, Manchester M13 9PT.

Consent for publication

Not applicable

Availability of data and material

The final anonymised trial dataset as a result of this study will be available to other researchers on request from the corresponding author.

Competing interests

The authors declare that they have no competing interests.

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Authors' contributions

CM, chief investigator on the ReaDySpeech study, developed the initial project idea and the ReaDySpeech online programme. CM, AB, ST & PC developed the protocol, the study methods and contributed to the ongoing support and development of this study. CM, AB, ST & PC have all been involved in monitoring the trial progress and have developed and reviewed the drafts of this manuscript. All authors read and approved the final manuscript.

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Chapter 5 A feasibility randomised controlled trial of ReaDySpeech for people with dysarthria after stroke

This paper has been submitted for publication in Clinical Rehabilitation and is under review. This paper is addressing the research question: Is it feasible to carry out a randomised controlled trial of online therapy ReaDySpeech for people with dysarthria after stroke?

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5.1 Abstract

Objective: To evaluate the feasibility of a multi-centre randomised controlled trial of ReaDySpeech, an online speech therapy programme for people with dysarthria.

Design: Feasibility randomised controlled trial, 2:1 minimisation procedure.

Setting: Four UK NHS services across hospital and community.

Participants: Forty participants with dysarthria at least one week post-stroke.

Interventions/comparator: ReaDySpeech with usual care (n=26) versus usual care only (n=14).

Main outcomes: We assessed the feasibility of the trial and intervention by: recruitment and retention rate, time taken to carry out assessments, success of outcome assessor blinding as well as fidelity and adherence. Participant outcome measures collected immediately after 8-10 weeks of intervention were the Frenchay Dysarthria Assessment II, Therapy Outcome Measure, Communication Outcomes After Stroke Scale, EQ-5D-5L, Dysarthria Impact Profile.

Results: The 40 participants recruited represented 54% of those eligible, 1-13 weeks post stroke, mean age 69 years (37-99). Retention was very high (92%). Assessor-blinding was not achieved with intervention allocation correctly guessed for 70% of participants (26/37). Time to carry out assessments was acceptable and we identified promising outcome measures. ReaDySpeech was delivered to 16/26 allocated participants, who completed 55% of prescribed activities, but ReaDySpeech and usual care were both delivered at low intensity (mean 6.6 face to face sessions of 40 minutes duration).

Conclusions: This randomised controlled trial of computerised therapy for dysarthria is feasible. However, further work is needed to widen recruitment and generalisability; address staffing and increase intervention delivery, intensity, adherence and independent use; achieve assessor-blinding by video-recording outcome assessments and to determine sample size.

Keywords: Dysarthria, stroke, computer therapy, feasibility, randomised controlled trial

5.2 Introduction

Dysarthria describes the impaired speech intelligibility caused by weak or uncoordinated muscles in the speech tract and is thought to affect 20-30% of stroke survivors (Warlow, 2001). This can be hugely disabling for individuals affected leading to social isolation and poor health outcomes (Brady et al., 2011b). This disorder occurs when any of the respiratory, laryngeal and/or oral articulator muscles; tongue, lips, cheeks, palate are affected (Darley et al., 1975). Severity of symptoms may range from completely unintelligible to slow speech or articulation difficulties. Intervention for dysarthria typically involves motor recovery exercises to strengthen the weak muscles, strategies to improve intelligibility and psychological support. However, research is extremely limited and there is no robust evidence indicating what intervention works best, when treatment should start, nor optimal duration or intensity of treatment. The lack of adequately powered, well-controlled trials in dysarthria was illustrated in a recent Cochrane review of five small trials and the overall body of evidence was considered low quality (Mitchell et al., 2017a). This is in marked contrast to the wealth of research on aphasia, the other main communication disorder experienced after stroke (Brady et al., 2016; Palmer et al., 2012).

Evidence from other aspects of stroke rehabilitation indicates that high intensity, repetitive task specific practice may be the most effective way to promote motor recovery after stroke (Langhorne et al., 2009). Patients in clinical practice reported paper-based dysarthria exercises were not particularly clear or motivating and this led the first author to develop an online programme to promote these principles in dysarthria rehabilitation. ReaDySpeech was developed to provide a tailored programme of exercises in a more user-friendly, accessible and engaging way with the

expectation this might increase uptake and, crucially, treatment intensity. In line with guidance on developing complex interventions, preliminary work found ReaDySpeech to be acceptable to speech and language therapists and patients (MRC, 2008; Mitchell et al., 2016). The aim of this study was to evaluate the feasibility of conducting a Phase III trial comparing ReaDySpeech with usual care for people with dysarthria after stroke.

5.3 Method

We registered this study with the International Standard Randomised Controlled Trials Number register (ISRCTN84996500) and obtained ethics approval from the UK National Research Ethics Service Committee Northwest (15/NW/0371). The study benefitted from Patient, Carer and Public Involvement. Four patient advisors formed the 'Ever Ready' group, advising on the design, conduct and dissemination of the trial.

We recruited participants from four NHS sites with both hospital and community stroke provision, in England over 14 months. The site speech and language therapist identified patients from their caseload and recorded reasons for exclusion or declining participation. Those meeting the inclusion criteria who were interested in hearing more about the study met the chief investigator, who recorded informed written consent or non-identifiable reasons for declining study participation.

Eligible for inclusion were new or current patients with post-stroke dysarthria and who were: more than one week post stroke (no upper limit), medically stable, and likely to benefit from speech and language therapy with sufficient English language skills to participate in therapy without a translator. Patients with a co-occurring aphasia were eligible and would only be excluded if severity precluded the use of ReaDySpeech. Exclusion criteria were: co-existing progressive neurological conditions or co-existing

cognitive, hearing or visual problems at a level that would prevent use of ReaDySpeech.

At baseline, prior to random allocation we recorded: demographic data (age and gender), stroke information (time since stroke, type of stroke - haemorrhagic or infarction, and stroke classification), levels of pre-morbid and current functioning (modified Rankin scale (Vanswieten et al., 1988)), current activities of daily living (Barthel Index (Mahoney and Barthel, 1965)) and the co-existence of aphasia (severity, how it was diagnosed).

Completed at baseline and follow up:

- Dysarthria impairment level: Frenchay dysarthria assessment (2nd edition FDA II)(Enderby and Palmer, 2008).
- Dysarthria activity level: Dysarthria therapy outcome measures activity (TOM A)(Enderby et al., 2013).
- Dysarthria participation level:
    - Communication after stroke scale (COAST) (Long et al., 2008).
    - Dysarthria impact profile (DIP) (Walshe et al., 2009).
- General health quality of life: EQ-5D-5L and visual analogue scale (Herdman et al., 2011).

Following baseline measurements, participants were randomly allocated to ReaDySpeech with usual care or usual care only. To ensure allocation concealment, a third party system used minimisation by the recruiting site and by time since stroke, acute (≤12 weeks post stroke) or chronic (≥12 weeks post stroke). Blinded outcome assessment by the chief investigator using the measures listed above was attempted

immediately after the 8-10 weeks intervention period. These data were collected by the assessor face-to-face and scored without the use of video-recording.

5.3.1 Interventions

In the control group participants received usual care only which would vary by site, from no intervention to best practice guidelines (WHO, 2007; Taylor-Goh, 2005). This could include: specific exercises for speech muscles, articulation work (impairment level); strategies such as slowing speech, education or awareness training (activity level); psychological support or advice and/or strategies to communication partners (participation level). Intervention details were recorded in speech therapy notes and retrieved following completion of follow up.

In the intervention group participants received usual care (as described in control group above) as well as access to ReaDySpeech, an online computer programme, delivered in any way considered clinically appropriate by the treating therapist. ReaDySpeech, was accessible using any Wi-Fi enabled device (smart phone, tablet, computer). ReaDySpeech addresses impairment and activity level functioning including articulation, breathing, rate of speech, volume, facial expression, intonation, oro-motor exercises (WHO, 2007). These included words and phrases appearing on screen, strategies to reduce speech rate, and video clips of oro-motor exercises to copy. This could be used during face to face therapy sessions with a therapist initially and thereafter with an assistant, supported by family or independent practice. We wanted to explore how often the intervention was delivered and used although we expected duration to be up to 8/10 weeks, we did not specify intensity or duration. Similarly, participants were able to practise independently if they wanted to but were not specifically required to do so. The programme was intended to be easier to understand

than the paper exercise sheets often used in clinical practice and it was hoped would prove more engaging and motivating. Exercises selected by the therapist and completion rate were recorded automatically by the programme data. Both usual care and ReaDySpeech interventions are outlined in greater detail in the protocol (Mitchell et al.).

5.3.2 Data handling and analysis

The study was not designed to be statistically powered to test for a between group difference in outcomes. Instead, we aimed to recruit 24 people to the ReaDySpeech intervention group and a minimum of 12 participants to the control group as recommended for a feasibility study (Julious, 2005).

We explored the feasibility of the trial processes by monitoring recruitment and retention rates including reasons for declining participation and for withdrawal, time taken to carry out outcome assessments and the success or otherwise of outcome assessor blinding. Recruitment and retention rates were analysed using descriptive summary statistics looking at patterns and reasons for non-participation.

Data on patients' ability and willingness to participate in the intervention were extracted from therapists' records and directly from usage data captured by the ReaDySpeech software. Participants willing to proceed to follow up were also interviewed face to face by the chief investigator about the impact of the study set-up, assessments and interventions on their involvement using yes/no questions, open questions and 5-point rating scales about the trial and the intervention.

Outcome data were reported on all participants with an intention-to-treat approach (White et al., 2012) from baseline to outcome using descriptive summary statistics:

mean, standard deviation and 95% confidence intervals (95% CI). We determined effectiveness of outcome assessor-blinding by comparing guessed with actual allocation, presented as a percentage and analysed using the kappa statistic. We used exploratory analysis using summary statistics to report the feasibility of delivering the intervention by fidelity and adherence. Responses to interview questions were reported as similar themes but no formal qualitative analyses were used.

5.4 Results

The results are reported in order of study objectives starting with the feasibility of the trial processes (recruitment, retention and measurement) and then the feasibility of delivering the online intervention (fidelity and adherence), highlighting findings that would inform the design of a definitive randomised controlled trial or further feasibility work to get to that point.

5.4.1 Recruitment and retention of participants

We achieved a consent rate of 54% (40 of the 74 eligible participants identified) at the four UK NHS sites between September 2015 and October 2016 (average rate 2.9/month). Full details of recruitment and retention are shown as a CONSORT flow diagram (Schulz et al., 2010) in Figure 5 and study participant baseline characteristics are described in Table 9.

For the majority of people who were deemed ineligible, this was because of therapists' perceptions that cognition was too impaired for therapy or because service limitations precluded participation from those without sufficient English language skills. A dislike of computers or low prioritisation of dysarthria by this predominantly acute stroke population, were the main reasons for declining involvement. However, those declining were not obviously older than those consenting and participants up to the

age of 99 were recruited to this study. Co-existing aphasia was not an exclusion criterion and was rarely reported by therapists as a reason for exclusion. However, as shown in Table 9, fewer than expected participants had aphasia. Despite our open-ended eligibility criteria, we recruited a predominantly acute population post stroke because there was nobody on the current NHS caseload beyond 13 weeks of stroke.

Figure 5 CONSORT Flow diagram for participants

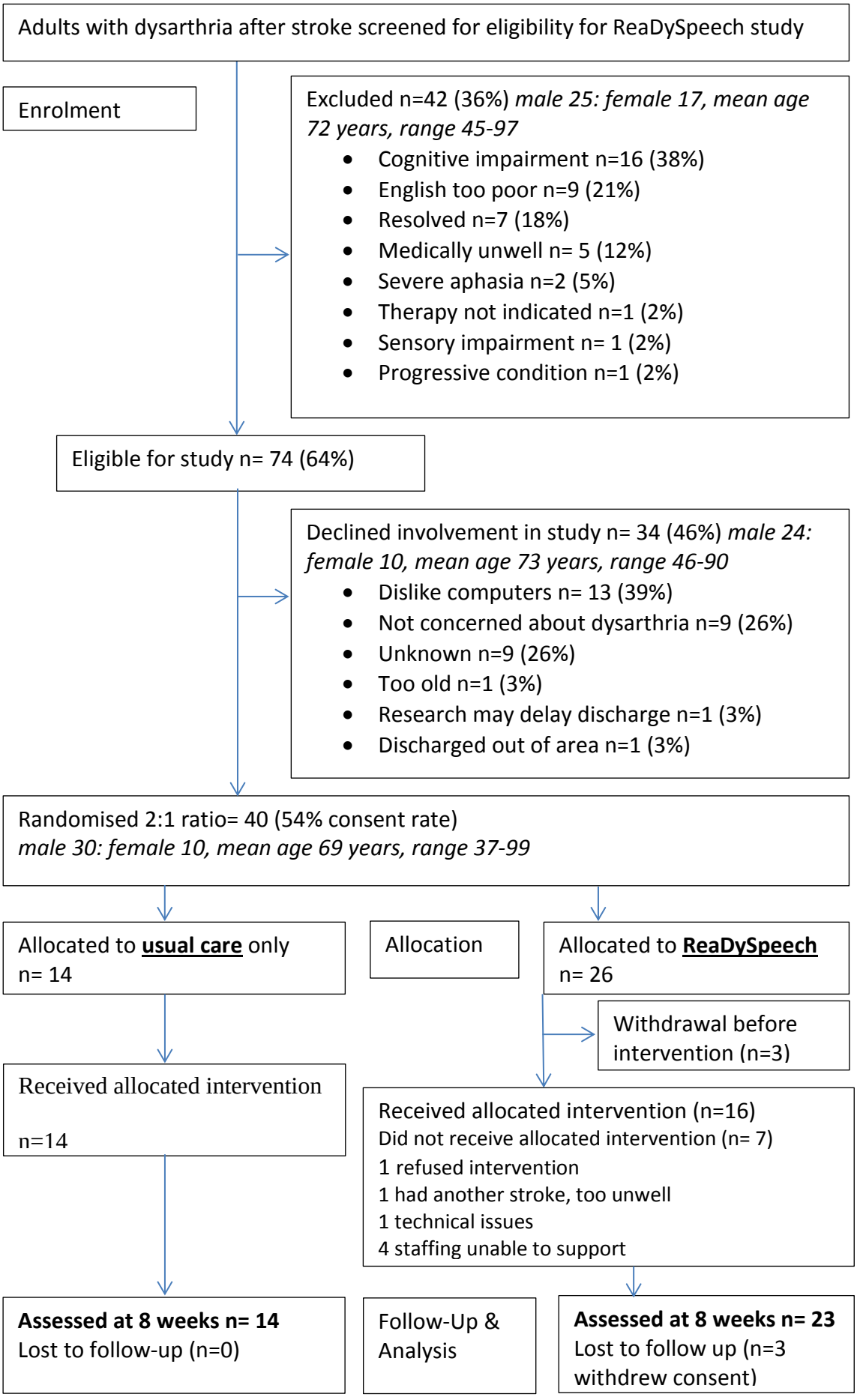

Table 9 Baseline characteristics of participants by treatment allocation

Characteristic	ReaDySpeech (n=26)	Usual care (n=14)
Mean age, years (min-max)	70 (37-99)	67 (55-85)
Male/Female	18/8	12/2
Recruitment location		
Hospital	16	5
Community	10	9
Days post-stroke mean (min-max)	24 (8-67)	27 (8-90)
Aphasia present (severity)	2 (mild aphasia)	2 (mild aphasia)
Stroke severity, mean (standard deviation)		
Baseline Barthel Index, 0 dependent, 100 independent	56 mean (42.5)	83 mean (29.6)
Baseline Modified Rankin Scale, 0 no symptoms, 6 death	3 mean (1.4)	2 mean (1.5)
Stroke lesion location: Lacunar, Partial anterior circulation, Total anterior circulation, Posterior circulation	Lacunar=11 Partial anterior circulation=6 Total anterior circulation=3 Posterior circulation=4 Not known=2	Lacunar=7 Partial anterior circulation=4 Total anterior circulation=1 Posterior circulation=1 Not known=1

The study retention rate was high. We followed up 37 of the 40 recruits (8% attrition) to outcome assessment and analysis at around eight weeks (range 5 to 16 weeks). It was observed that almost everyone understood the need for randomisation and no-one withdrew because of their group allocation. The three participants lost to follow up were from the ReaDySpeech intervention group (Figure 5). They withdrew consent for the study before intervention and having reviewed their reasons we consider these three withdrawals as missing completely at random and have not imputed outcome data.

Feasibility of measurements

The 2:1 minimisation procedure meant the groups (ReaDySpeech n=26, Usual care n=14) were well matched at baseline for occurrence of aphasia, lesion location and days post stroke (see Table 9). The groups were less well matched for levels of everyday functioning and dependence, with the ReaDySpeech group more impaired and more likely to be recruited in hospital. The ReaDySpeech group were also found to have more impaired communication as measured at the impairment, activity and participation level. The ReaDySpeech group also reported lower baseline quality of life scores and more problems with mobility, self-care, usual activity, pain/discomfort, anxiety/depression.

We found it was feasible to carry out the Frenchay Dysarthria Assessment II (Enderby and Palmer, 2008), dysarthria activity level from the Therapy Outcome Measure (Enderby et al., 2013), Communication Outcome After Stroke Scale (Long et al., 2008) and EQ-5D-5L (Herdman et al., 2011) on this primarily acute stroke population. It was not feasible to use the Dysarthria Impact Profile (Walshe et al., 2009). This is designed

to be used with people adjusting to long term dysarthric symptoms and was not suitable in this trial due to the predominantly acute population recruited. It took 60-90 minutes to complete all assessments at follow up which was considered appropriate by 97% (36/37) of participants. Most participants reported during the interviews that they felt these assessments reflected their speech and health difficulties.

The success of assessor-blinding was judged by the assessor's ability to guess allocation group and whether blinding was achieved. The assessor was un-blinded either explicitly or inadvertently, in 11 cases (10 ReaDySpeech and 1 usual care). The assessor guessed treatment allocation correctly for 10 out of 14 in the usual care group and 16 out of 23 in the ReaDySpeech group. Observed agreement is 70% compared with 51% expected by chance. Kappa is 0.39 (p=0.008) giving evidence of agreement (i.e. correct prediction) beyond chance.

Results of outcome measures from baseline to follow up are shown in Table 10. For the group as a whole, irrespective of allocation, outcomes improved over time. On all measures, confidence intervals of the mean difference from baseline to outcome excluded zero. The standard deviation of the Frenchay Dysarthria Assessment II at follow-up was considerably higher than that of its change score from baseline to follow-up. The same was true for dysarthria Therapy Outcome Measure, activity score. If used in a definitive trial, this finding suggests that it would be beneficial in terms of statistical power to assess these scales at baseline to allow analysis of covariance. Conversely, there was no suggestion of a similar reduction in standard deviation when comparing 'follow-up' with 'change from baseline' for the Communication Outcome After Stroke Scale. This suggests there was no statistical benefit from baseline assessment of this scale.

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Table 10 Outcome measures from baseline to follow up

Outcome measure	Baseline, mean (standard deviation) All n=40, UC n=14, RS n=26	Outcome, mean (standard deviation) All n=37, UC n=14, RS n=23	Whole group, mean difference (SD, 95%CIs)
Impairment: FDA II	*All: 159 (37.5)* UC: 170 (20.2), RS: 153 (43.3)	*All: 179 (32.7)* UC: 184 (20.4), RS 177: (38.6)	*21 (21.2, 27.8 to 14)*
Activity: Dysarthria TOMs	*All 3 (1.0)* UC: 3.5 (0.8), RS: 3.2 (1.1)	*All: 3.7 (0.9)* UC: 3.9 (0.6), RS: 3.6 (1.1)	*0.5 (0.7, 0.7 to 0.2)*
Participation: COAST	*All 59 (16.3)* UC 63.1(15.0), RS: 56 (16.7)	*All 67 (16.1)* UC: 70.8 (15.3), RS: 65.3(16.6)	*8.5 (16.6, 14 to 3)*
Participant reported health quality of life states: EQ-5D-5L			
Visual analogue scale median (IQR)	UC n=14, Median = 63 (25^{th}=50, 75^{th}=84) RS n=26, Median = 50 (25^{th}=25, 75^{th}=64)	UC n=14, Median = 76.5 (25^{th}=55, 75^{th}=86) RS n=23, Median = 65 (25^{th}=50, 75^{th}=80)	
EQ-5D-5L	**Baseline % problems**	**Outcome % problems**	
Mobility	UC 9 (64%) : RS 24 (92%)	UC 8 (57%) : RS 16 (70%)	
Self-care	UC 7 (50%) : RS 21 (81%)	UC 3 (21%) : RS 15 (65%)	
Usual activity	UC 8 (57%) : RS 24 (92%)	UC 7 (50%) : RS 24 (92%)	
Pain/ discomfort	UC 6 (43%) : RS 17 (65%)	UC 7 (50%) : RS 14 (61%)	
Anxiety/depression	UC 3 (21%) : RS 16 (62%)	UC 4 (29%) : RS 15 (65%)	

UC = usual care : RS = ReaDySpeech
FDA II = Frenchay dysarthria assessment II
Dysarthria TOMs = Dysarthria therapy outcome measure, activity score
COAST = Communication after stroke scale

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5.4.2 Feasibility of the Intervention

In terms of delivery, we found that 16 of the 26 participants randomised to the ReaDySpeech group did access the ReaDySpeech intervention. Of the 10 who did not, three had already withdrawn and refused follow up. One had refused intervention and one had another stroke but these two both agreed to follow up. Five participants did not have access to ReaDySpeech due to lack of staffing (three from a single hospital).

For those receiving ReaDySpeech and usual care, face to face sessions were of similar low intensity of 6.6 sessions per participant, (min 1 – max 24), with a mean session time of 43 minutes (SD 28: min 10 – max 120 minutes). Different models of therapy provision were observed with assistants carrying out 81 of the 151 (54%) sessions in the ReaDySpeech group compared to 20 of the 95 (21%) sessions in usual care, the rest were by qualified speech and language therapists. In the ReaDySpeech group all of the face to face sessions used ReaDySpeech, with exercise selection including impairment and activity level exercises and in addition two participants had psychological support.

When considering adherence for the 16 participants set up with access to ReaDySpeech, completion rate of the exercises by the participants was 55% across all sites (therapy data around exercise selection and completion will be reported separately). Of these 16 participants, nine used it independently outside of face to face sessions (56%), mostly in the community with their own computers. The majority found the programme straightforward and easy to use. They commented specifically on the videos, as well as being able to practise when it was convenient to them and reported an improved confidence in their speech. For this reason, all agreed they would recommend the treatment to someone in their position.

5.5 Discussion

This study of ReaDySpeech for people with dysarthria post-stroke found that it is feasible to undertake a randomised controlled trial within the context of the NHS, provided staffing resources are in place. A lack of NHS therapy provision for people with chronic dysarthria meant that we only recruited acute stroke participants. A broader sample would be more reflective of the general stroke population and this would include chronic stroke, those with aphasia and those with no computer skills. We found that random allocation using minimisation did not result in balance across groups on key variables that may be important for outcomes. We also need to ensure a future trial achieves assessor blinding and to determine the sample size needed for adequate statistical power with this blinded measure. ReaDySpeech delivery was difficult to achieve at sites with low therapy staffing but the unexpectedly high rate of successful delivery by assistants at several sites may be a way to improve access. We observed relatively low intensity for both interventions, including independent use, which needs to be addressed in a future trial.

Recruitment was carried out through initial identification by speech and language therapists. Recruitment to randomised controlled trials can be difficult, particularly for vulnerable participants early post-stroke (Treweek et al., 2013; Horne et al., 2015) so we considered a recruitment rate of 54% to be reasonable. This study shows we can identify and potentially recruit those early post-stroke within the first three to four months only. We know nine of the eligible patients (26%) declined participation as they were not concerned about their speech but this may alter over time as priorities change as people return to everyday life. To widen recruitment to include chronic

stroke, we will also want to examine the feasibility of recruitment through charity, independent sector and using online stroke forums.

Interestingly, we found that there were very few patients identified who also had a co-occurring aphasia and we recruited a lower than expected proportion of participants with aphasia, (n=4, 10%), compared to other studies with 29-31% (Flowers et al., 2013; Bowen et al., 2012a). We may need to screen all communication impaired patients to avoid subconsciously excluding those with a co-occurring aphasia. There is potential that a future trial could use clinical research nurses to identify all stroke admissions with a communication impairment, which could also reduce the therapist work-load (Treweek et al., 2013).

The most likely reason for eligible participants to decline was due to a dislike of computers for 13 eligible patients (38%) despite the treatment making minimal technical demands on users. Although technology is becoming ever more common, useful lessons have been learnt about how to describe the research and intervention more carefully (Donovan et al., 2009). Describing tablets and smart phones instead of computers may be less daunting. Screening of a broader range of patients, carefully considered wording and training around equipoise to identify potential participants will be introduced in a future study (Treweek et al., 2013).

Retention rates were high in this trial with 37 out of 40 (92%), being followed up to outcome measure. This was a relatively short follow up of around eight weeks. A future trial may need to consider longer time to follow up to evaluate sustained improvement which may affect retention. Retention may be a factor if other changes are made to a future trial such as broadening the stroke population, using video at

outcome as well as any changes to intensity of delivery. These would all need to be considered when calculating a future sample size.

Following recruitment and randomised allocation by minimisation we found that the groups were not balanced at baseline. The ReaDySpeech group had worse physical health and more severely impaired speech. To achieve balance in a future trial we would consider minimising by severity of speech at the activity level using the Dysarthria Therapy Outcome Measure activity score and this would be monitored. The potential to use this measure as the primary outcome for a future trial will be discussed with the patient user group.

A key limitation of the study was the unsuccessful assessor-blinding of outcome measures as participants were often keen to discuss their intervention. This was particularly the case for those who had been allocated to ReaDySpeech, despite being asked not to disclose this. Video assessment, including of the Therapy Outcome Measure, has been used in other trials of communication impairment after stroke which successfully blinded the outcome assessor (Bowen et al., 2012b; Palmer et al., 2015). Further feasibility work will explore whether the benefit of the use of videos outweigh any adverse impact on recruitment and retention.

Frequency and intensity of ReaDySpeech and usual care delivery was low, particularly in acute hospital settings due to staffing levels. At two of the sites, therapists reported actively seeking out assistants to deliver the ReaDySpeech intervention to reduce their time and this warrants future health economic investigation. Just over half of the ReaDySpeech group used the online therapy programme for independent practice, and this study has raised awareness of some of the barriers to independent practice in acute settings. A future trial will need to be more prescriptive about how, when and

how often to deliver it but this could impact on adherence to treatment and retention, which will need to be taken into account. It will also be important to emphasise the philosophy of guided self-management underpinning ReaDySpeech through which intensity of engagement with the intervention could be achieved through flexible, self-administration by patients. The application of ReaDySpeech in the feasibility trial suggested it became an alternative format for intervention delivery but the flexible self-administration approach was not supported sufficiently by clinical settings.

In summary, to ensure the success of a phase III trial we plan to carry out a further feasibility trial around widening recruitment, improving fidelity, adherence and intensity of ReaDySpeech achieving assessor-blinding by video-recording outcome assessments and determining sample size.

5.5.1 Clinical messages

- People with dysarthria early after stroke are willing to engage in research using ReaDySpeech, an online programme to support speech rehabilitation.
- Innovative service delivery models are required to increase the amount of therapy for people with dysarthria as usual care is currently provided in low doses and for a short duration.

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Author contributions

Claire Mitchell, Audrey Bowen, Sarah Tyson and Paul Conroy were all involved in the conception and design of the study. Audrey Bowen, Sarah Tyson and Paul Conroy were the steering group for the trial. All the authors were involved in data analysis and interpretation of data. All of the authors were involved in the drafting and revision of this article.

Competing interests

None declared.

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Chapter 6 Detailed exploration of online therapy (ReaDySpeech) and Usual Care intervention as delivered during a feasibility randomised controlled trial.

This paper will be submitted to the International Journal of Communication Disorders for the themed call: The use of technology in speech and language therapy

This paper is attempting to address the research question: What was the ReaDySpeech and usual care intervention delivered during a feasibility randomised controlled trial?

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6.1 Abstract

Background: Dysarthria after stroke can have a profound psychological impact on those affected yet there is limited evidence about the efficacy of interventions. Online therapy is a promising way to deliver speech rehabilitation. Here we detail the online therapy (ReaDySpeech) and usual care delivered during a feasibility randomised controlled trial focusing on describing fidelity and adherence rather than trial outcomes which have been previously reported.

Aims: To investigate the type, dose and delivery of exercises prescribed by therapists and carried out by patients, and to explore patients' views of ReaDySpeech and usual care.

Methods & procedures: Forty participants with dysarthria, at least one week post-stroke, were randomised (2:1 minimisation procedure) in a feasibility trial: ReaDySpeech with usual care (n=26) versus usual care only (n=14). Intervention period of 8/10 weeks with immediate follow up. Information about what and how ReaDySpeech was delivered was extracted from the online programme history at follow up. Details of the usual care delivered were extracted from participants' clinical notes at follow up. Participants were interviewed about the intervention they received at follow up.

Outcomes and results: Trial participants included 30 men, 10 women, mean age 69 years (range 37-99), all within 13 weeks of stroke. 37 of the 40 were followed up at 8/10 weeks. When using ReaDySpeech, therapists selected impairment and activity level exercises for every participant. The most frequently selected were activity level articulation tasks (selected 40 times/96 total exercises for all participants) and most likely to be completed by participants were impairment level oro-motor tasks (25

completed/31 times selected, 80% completion). Usual care also included mainly impairment and activity level exercises. Both groups received limited participation level intervention. ReaDySpeech completion rate by participants was higher with assistant support (17/29, 59%) compared to no assistant support (3/9, 33%). Participants reported positive experiences from both intervention approaches.

Conclusions and implications: Impairment and activity level exercises are the intervention of choice whether delivered online or in person. The findings add to our understanding of how speech and language therapy for dysarthria is delivered in the NHS and its uptake by patients. In future research, trialists will need to consider carefully what aspects of experimental and usual care interventions are sufficiently different to detect a difference, and how to ensure fidelity to the interventions under investigation.

What this paper adds?

What is already known on the subject?

We know that the psychological impact of dysarthria can be devastating for people particularly those returning to work and/or with a busy, active social life. We also know that there is a distinct lack of research into dysarthria after stroke particularly when compared to aphasia research. Clinical guidance is limited but generally suggests speech and language therapists address impairment, activity and participation levels of activity. There is no evidence to suggest whether clinical intervention is effective or not for dysarthria rehabilitation, or which type of intervention may be most effective.

What this paper adds to existing knowledge?

This paper adds to our understanding of how speech and language therapy for dysarthria is delivered in every day clinical practice in the UK. It also furthers our knowledge of the interventions that can feasibly be delivered during a trial, that are acceptable to patients and therapists and may improve adherence.

What are the potential or actual clinical implications of this work?

Understanding what and how interventions are delivered informs the feasible development of both future clinical trials and clinical practice and is particularly important for service planning. It will help guide research questions, the development and definition of trial and control interventions as well as facilitate priority setting work with patients and therapists.

6.2 Introduction

Dysarthria is a commonly occurring communication disorder post stroke with variable clinical presentation from minimal impairment, through to severely unintelligible depending on the weakness of the speech muscles involved (Darley et al., 1975; Duffy, 2013; Mackenzie, 2011). The psychological impact of altered speech is not merely dependent on severity but also on the communication demands of an individual's social and working life prior to their stroke (Brady et al., 2011b; Tilling et al., 2001; Dickson et al., 2008). Clinical guidelines indicate that intervention should be delivered according to the individual's speech presentation, but little is known about dysarthria and how best to treat it. A recent update of the adult acquired non-progressive dysarthria intervention Cochrane review identified only five trials suitable for inclusion, which were graded as low to very low quality due to subgroups or small numbers and therefore lack of statistical power (Mitchell et al., 2017a). Thus, there is limited

evidence to guide therapists' intervention choices, including what should be delivered, when it should be delivered, at what intensity and for how long.

Best practice guidance for dysarthria intervention after stroke traditionally follows the World Health Organisation's International Classification of Functioning and Health framework (WHO, 2001). Intervention may therefore consist of therapeutic exercises to improve impairments (such as oro-motor exercises, breathing exercises and articulation of sounds) and/or activity (by providing strategies like slowing the rate of speech, purposeful production of prosody and or articulation) while intervention to improve participation aims to address psychological needs, communicative confidence, supporting conversation and integration in everyday life (Taylor-Goh, 2005; World Health Organisation, 2001). Regardless of the treatment focus, intervention is traditionally delivered in face to face sessions by a speech and language therapist demonstrating exercises, giving feedback and offering generic, paper-based exercises for patients to use for practice. The most recent UK National Clinical Guideline for Stroke indicated that anyone with dysarthria should be assessed and intervention offered that includes activity and participation level strategies but doesn't specifically mention impairment level exercises (ICSWP, 2016).

However, a recent UK wide survey of 146 speech and language therapists working in stroke, highlighted that impairment based exercises are a common feature of current clinical practice (Miller and Bloch, 2017). This survey reported time spent on dysarthria treatment was limited and variable between clinical settings. Time is spent on education and explanation with informal assessment more likely than formal assessment. Dysarthria intervention continues to be carried out as part of clinical practice because of the striking clinical need, and current intervention approaches are

based on motor learning theories, drawn from post stroke limb rehabilitation. This suggests recovery is most effective when task specific activities are carried out at high intensity (Langhorne et al., 2009). More research around whether dysarthria intervention is effective and at what intensity, for people after stroke is a key topic that needs to be addressed.

We know therapists have limited time to support dysarthria exercises, as reported in the Miller, 2017 survey, and people with dysarthria are given paper-based worksheets for independent work in response to this. Current clinical guidelines suggest 45 minutes of active rehabilitation a day and the Miller, 2017 survey suggests this is not being met for dysarthria (NICE guidelines 2013 long term rehabilitation for stroke).It may be that technology could support the need to increase time spent on speech rehabilitation and increase independent practice. The idea for ReaDySpeech, an online speech therapy programme, was devised by the first author in consultation with patients and therapists following guidance around developing complex interventions to start the development process (MRC, 2008; Mitchell et al., 2016). Following acceptability work, a feasibility randomised controlled trial evaluated the delivery of ReaDySpeech and usual care and of undertaking a subsequent Phase III randomised controlled trial (Mitchell et al., 2017b); (Mitchell et al., 2017c, Submitted). The trial was found to be feasible in terms of the numbers of participants recruited, 40 out of 74 eligible, and the success of retention to 8 week follow up (37 out of 40 participants).

The delivery of ReaDySpeech was also considered feasible, although challenging in acute settings where staffing levels were low. However, the study concluded that further feasibility research was needed to widen recruitment strategies and criteria, avoid bias by blinding outcome assessment as well as evaluating the impact of longer

follow up and increased intensity of intervention. A feasibility study also affords the opportunity to report crucially important clinical aspects of the trial, which have been thus far unreported. Specifically, the current paper provides a detailed description of ReaDySpeech, how it was delivered as an example of a technology-based treatment approach to dysarthria, and how acceptable it was to the patients. Furthermore, data from the trial's control group contributes to the discussion around what usual care for dysarthria after stroke currently looks like, at least in the context of UK clinical NHS care to guide future research (Langhorne et al., 2011).

6.3 Method

Design

We registered this study with the International Standard Randomised Controlled Trials Number register (ISRCTN84996500) and obtained ethics approval from the UK National Research Ethics Service Committee Northwest (15/NW/0371). Four patient advisors formed the 'Ever Ready' consultation group who advised on the setting up, progress and dissemination of the trial.

The Method for the trial, including participants, randomisation and outcome measures are described in detail in the protocol (Mitchell et al., 2017b) and the trial report (Mitchell et al., 2017c, Submitted). In summary, we recruited participants over a 14-month period, from four English NHS sites with both hospital and community stroke provision. The main inclusion criterion was post-stroke dysarthria, more than one-week post stroke (no upper limit) with no co-existing progressive neurological condition. Baseline assessments were carried out prior to the minimisation procedure and outcome measures were completed following the 8/10 week intervention period. Participants were randomly allocated, using a minimisation procedure, in a 2:1 ratio to

ReaDySpeech with usual care or usual care only. The intended ReaDySpeech intervention and usual care are outlined in greater detail following the TIDieR (Template for Intervention Description and Replication) checklist and guide (Hoffmann et al., 2014a), in the published protocol (Mitchell et al., 2017b). Figure 6 shows the ReaDySpeech programme menu, which includes impairment and activity level exercises that can be selected by the therapist. When using the programme words and phrases will appear on screen and there are video clips for all oro-motor exercises.

Figure 6 Screen shot of ReaDySpeech exercise selection overview

New Exercise Session

Exercise Session name

Week 10

Expected Attempts per Day

3

Session Duration (Days)

Week 10

Exercises

Articulation
Bilabials
Labiodentals
Dental
Alveolar
Post alveolar
Glottal
Velar
Blends
Single syllable words
Two syllable words
Three syllable words
Questions to answer
Short phrases
Word final complex consonant clusters
Word final Dental
Word final Bilabial
Word final Velar
Word final post alveolar

Breathing
Breathing and counting to 8/5/10
Breathing and increasing volume counting 1 – 5
Breathing and decreasing volume counting 1 – 5
Breathing on /s/ various
Breathing and words
Breathing and talking

Slowing speech
Alphabet chart and talking exercises
Pacing Board and talking exercises

Volume
Volume work word level
Volume work and talking

Facial expression
Facial expression exercises

Intonation
Intonation exercises

Oro-motor exercises:
Range
Strength
Speed

Data collection

To describe the intervention that was delivered and examine fidelity by therapists (Persch and Page, 2013) and adherence by patients the following were recorded. For ReaDySpeech this information was extracted from the programme usage. For usual care, it was extracted from the patient notes.

- The actual exercises selected by the therapists
- Adherence: the exercises carried out by the participants
- Who delivered the intervention (therapist, assistant or self-directed)
- How therapy was delivered (face to face, telephone or any other means)

Participants' views of both interventions were documented by the researcher during face to face interviews via yes/no questions, open questions and 5-point rating scales and comments (Appendix 8).

Analysis

Exploratory analysis using summary statistics reported the intervention selection, completion, delivery and participants' views.

6.4 Results

Content selection and delivery

A total of 151 ReaDySpeech sessions were carried out for those allocated to the ReaDySpeech arm (n=23, 3 withdrew before intervention). The number of sessions per participant varied but on average this was 6.6 sessions over 8/10 weeks of intervention. A total of 96 exercises were selected from the ReaDySpeech programme for all participants across all sites. Exercise programmes were altered throughout the

intervention and the programme history recorded all that were selected. Exercise selection by therapists using ReaDySpeech followed similar patterns across all four participating sites including both impairment based exercises and activity level strategies. Articulation exercises, were the most common exercises selected (n=40), followed by oro-motor exercises (n=31), then breathing exercises (n=15) with rate of speech, volume, facial expression and intonation only being selected a few times (Table 11). The majority of sessions used the ReaDySpeech programme only, but there were four sessions (out of the 151 ReaDySpeech sessions) that also included participation level activities. In the ReaDySpeech group assistant practitioners (both generic and speech/language therapy assistants) carried out 81 sessions with participants compared to 70 sessions delivered by a speech and language therapist. Nine participants used the programme independently outside of their therapy sessions, predominantly those at home with their own computers and Wi-Fi. One participant reported involving family in independent practise.

A total of 95 sessions of usual care only sessions were carried out for those allocated to that arm (n=14) which was an average number of sessions per participant of 6.8 during the 8/10 week intervention period. The majority of usual care sessions involved impairment level oro-motor exercises and articulation work, as well as activity level articulation work including strategies for rate of speech and volume, in the same manner as ReaDySpeech. In the usual care only group, mirror work was included in several sessions for impairment level oro-motor exercises and reading from a book or newspaper for activity level tasks was also carried out. In four of the sessions, participants also received psychological support and two more had telephone support to improve confidence. In the usual care group, 75 sessions were delivered by a speech

and language therapist and only 20 were carried out by an assistant practitioner. In response to the question about practise outside of therapy 10 out of the 14 in the usual care group said they had practised. One participant reported they sought family support for this.

Table 11 Exercise selection (by therapist) and completion (by patient) for ReaDySpeech only

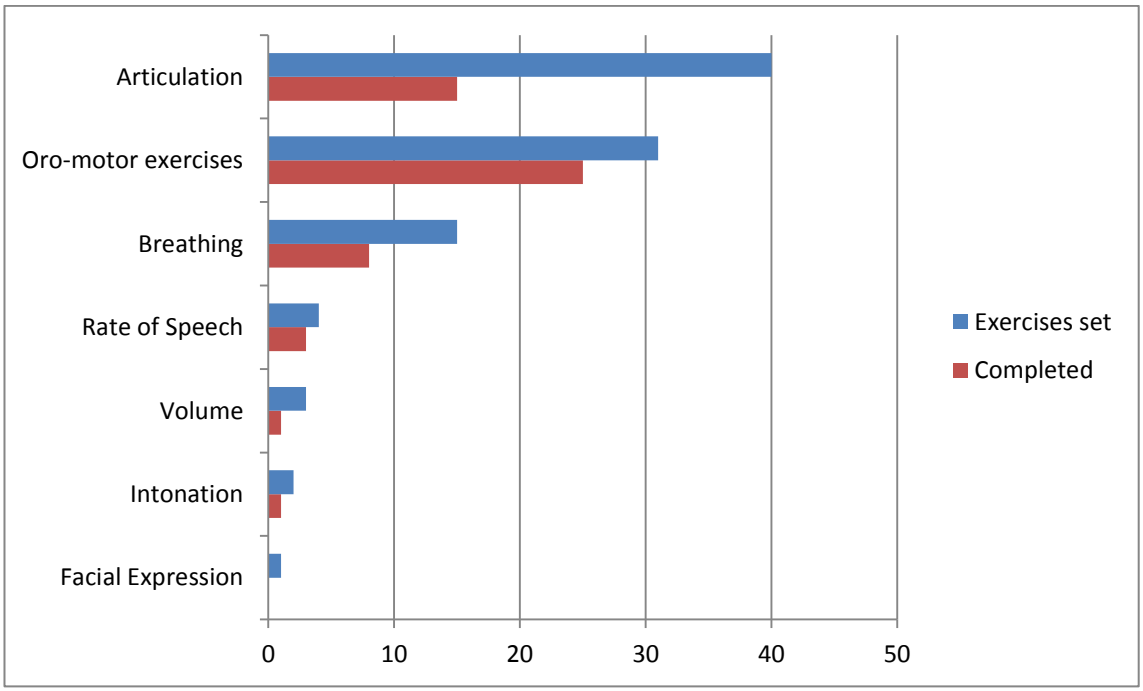
		ReaDySpeech exercises selected by speech/language therapist	Completion by participant	Total exercises set (completed)
Articulation	Word initial	14	5	40 (15)
	Single syllable words	4	0	
	2 syllable words	2	1	
	3 syllable words	5	0	
	Questions/phrases	5	3	
	Word final	10	6	
Breathing	Counting	5	2	15 (8)
	Increasing/decreasing volume	5	3	
	Breathing and words	2	1	
	Breathing and talking	3	2	
Rate of speech	Alphabet chart			4 (3)
	Pacing board and talking	4	3	
Volume	words	1	0	3 (1)
	talking	2	1	
Facial expression		1	0	1 (0)
Intonation		2	1	2 (1)
Oro-motor	Range	13	9	31 (25)
	Strength	9	7	
	speed	9	9	

Exercise completion

The ReaDySpeech exercises most frequently completed by participants were the impairment level oro-motor exercises (25 completed out of 31 selected, 80%), followed by activity level breathing exercises (8 completed out of 15 selected, 53%), followed by activity level articulation tasks (15 completed out of 40 selected, 38%) (Figure 7). This includes completion during both face to face intervention and independent practice. Completion rates of ReaDySpeech varied by site and assistant support led to a higher rate of completion. Where assistant support was used there was an average of 59% completion, with 17 exercises completed out of 29. Without assistant support participants completed 3 exercises out of the 9 selected (33% completion).

The only data for completion of usual care exercises is from clinical notes, where all participants were considered to have been fully involved in participating in the sessions. There are no data for independent practice.

Figure 7 Exercises set and completion for ReaDySpeech programme



Participant reported experience of ReaDySpeech

The participants in the ReaDySpeech arm found the training and support to use the programme sufficient, and the majority found it straightforward and easy to use. The most likely response when asked about what the intervention was trying to achieve related to the use of video clips and that participants knew what was expected of them. Participants reported they liked the videos with comments such as "*its visual you can see it, using the video with mouth movements"* and this made it easy to understand and follow. Participants frequently commented that using a computer was convenient and could be used independently, for example "*good to use a computer so you could do it when you feel like it"*. The majority of comments were positive, participants reported that it improved their confidence and they would recommend it to others in their position. Three participants reported that they would have liked greater feedback, and one participant reported motivation would be better if the programme was more like a computer game.

Participant reported experience of usual care

Eight out of the 14 usual care participants were clear about what the intervention was trying to do, with six not being clear and one of them described it as *"pieces of paper"*. Participants receiving usual care felt this approach gave them an understanding of their problem and suggestions about what they could do about it. Most found, the intervention was "*very easy*" or "*fairly easy*" to carry out. For example, *"told where the problem is and told exercises that help that".* They valued this and found it helpful, and on this basis, would recommend it to someone in their position. On the whole comments were positive about usual care intervention in terms of understanding, ability to carry out and perceived helpfulness.

6.5 Discussion

This study reports the detail of the ReaDySpeech intervention and 'usual care' carried out in four typical NHS hospital and community settings during a randomised controlled trial to examine feasibility of the intervention for evaluation in a subsequent trial. Although ReaDySpeech is a different mode of delivering usual care, we had still anticipated greater differences in terms of what was actually delivered in usual care content, how it was delivered and intensity of delivery. Both interventions delivered similar numbers of face to face sessions of primarily impairment and activity level exercises, advice and strategies with limited participation level. Intervention delivery supported by assistants led to greater likelihood of participants completing the exercises selected but there was less independent use of ReaDySpeech due to access restrictions particularly for those in acute settings.

The similar content delivered for both treatment groups was an important finding from the feasibility trial. Therapists for both groups had delivered what was clinically indicated with no specific direction. It is important to take into account that the data retrieved from the ReaDySpeech programme was much more detailed than the usual care data but we were able to establish the main focus of the usual care sessions. There were more therapist delivered, face to face impairment level sessions in usual care than we expected mainly because of the lack of evidence around these exercises and the impact of limited therapist resources (Foley et al., 2012). It is possible that the amount and frequency of usual care could have influenced therapists input purely by involvement in a trial and this may not reflect usual care outside of a trial (McCambridge et al., 2014).

Recent guidance for stroke (ICSWP, 2016) includes activity and participation level approaches with less emphasis on impairment level interventions, such as non-verbal oral exercises. The use of non-speech oro-motor exercises continues to be a topic of much debate, with a growing number of researchers suggesting these exercises are so far removed from speaking they have no place in clinical practice (Mackenzie et al., 2010; Mackenzie et al., 2014; Ziegler and Ackermann, 2013). While the theoretical reasoning for this argument may be coherent, there are no definitive trials to back up this view and it continues to be the mainstay of clinical practice (Miller and Bloch, 2017). This study reports that the second most highly selected exercises by therapists and most frequently completed by participants were the impairment level oro-motor exercises. It is likely this debate will continue until we have clear evidence supporting or refuting their use.

Another interesting similarity between the ReaDySpeech (with usual care) intervention and usual care only, was the significant lack of participation level intervention. There were only eight sessions out of the total 246 for both interventions that involved this level of functioning, offering psychological support and opportunity to engage in everyday tasks. It may be that participation level work is considered more time-consuming which was found to be the case with aphasia rehabilitation (Laliberte et al., 2016) and may reflect clinical habit or tradition by the predominantly medical model of in-patient care. This may also reflect the acute nature of the recruited trial population with many in hospital settings. It has been found that hospital-based therapists may focus more on impairment due to the constraints of acute care, even when people have progressive dysarthria (Collis and Bloch, 2012). Further work looking at what

participation level intervention involves and what the barriers are could be useful further research.

One of the motivations behind the development of ReaDySpeech was the possibility it could increase intervention intensity by enabling participants to practise and exercise independently outside therapy sessions. Research is now showing that therapists in stroke units spend less than half of their time in face to face rehabilitation (Putman et al., 2006; Foley et al., 2012; Clarke et al., 2015) due to other administrative duties. However, ReaDySpeech, in this study, was predominantly used during face to face intervention with therapists. When participants did practise independently, this was usually only the case for those living at home with their own computers (tablets, smart phones, lap-tops). Although we offered tablets to therapists for participants to borrow while in hospital, these were not usually left with participants outside of therapy sessions because of therapists'/assistants' perceptions about support needs or fears they would be lost or damaged. We did, however find that participants using ReaDySpeech more frequently worked with an assistant, rather than a therapist, than those receiving usual care. As ReaDySpeech was considered self-explanatory, assistants needed little explanation or support to use the programme set and monitored by the therapist. The completion levels appear higher when using ReaDySpeech with an assistant so this may be a relatively acceptable way to increase the intensity while minimising resources. However further work is needed to decide what is the most effective level of intensity of therapy, and how to effectively implement independent practice.

Participants were overwhelmingly positive about both the interventions they received. It must be remembered though that a dislike of computers was the main reason for

declining involvement in the ReaDySpeech study, so the participants were already willing to use computers as part of rehabilitation (Mitchell et al., 2017c, Submitted). The usual care participants also expressed positive views and expressed gratitude for the intervention, which may influence comments and would likely benefit from more in-depth discussion. It was evident that those allocated to ReaDySpeech, even those who scarcely used the programme, considered themselves as having been lucky to be given the opportunity. This highlights the need to promote equipoise in the language used with those involved in identification of participants and patient information. It also indicates that we need to carefully consider whether another intervention, placebo or attention control is the best approach for a future trial of effectiveness. It will be key to ensure there is a sufficient difference between interventions, whether this is content, intensity or delivery, or indeed a placebo or attention control, to successfully examine effectiveness in future research.

Conclusions

This article describes the content of usual care and ReaDySpeech as used during a randomised controlled trial of feasibility. We found impairment and activity exercises are the most likely to be delivered in an online intervention and usual care. We found there was some independent use of the online programme with the potential to increase intensity but future research will need to evaluate the appropriate timing, frequency and intensity of intervention to improve outcomes. We know patients after stroke are willing to engage in technology to support speech rehabilitation but further work is needed to establish better understanding of whether interventions work for those with dysarthria and if they do, what point post stroke and at what intensity is acceptable.

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Chapter 7 Discussion

7.1 Background to the research

This body of work has originated from clinical concerns raised by patients with dysarthria after stroke reporting a lack of intervention options available to them. Therapists also reported the weak evidence base meant they had poor information on which to base clinical advice for patients. This started the process of developing a complex intervention of an online programme, ReaDySpeech, in an attempt to address some of these issues around increasing intensity and offering clearer information through video clips when carrying out exercises independently. This on-line development was based on existing best clinical practice and what is typically delivered in routine NHS care. This discussion chapter will present the findings and relate them to the non-linear stages described in the MRC framework (MRC, 2008).

The development stage is a process that we have described with the initial scoping of the literature in chapter 1. This development stage led to a more in-depth and robust identification of the evidence base with the Cochrane systematic review in chapter 2. We continued development stage work by considering potential technical barriers and clinical acceptability work in the study described in chapter 3. This enabled us to progress to the feasibility stage. This included writing the protocol for, and evaluating the feasibility of a randomised controlled trial as described in chapter 4 and 5 respectively. A more detailed exploration of the ReaDySpeech and usual care intervention were described in chapter 6 as part of the feasibility stage. The limitations of the work and how this can be addressed going forward to the evaluation stage of ReaDySpeech will be considered.

7.2 Development stage

The research questions relating to this stage were:

1. What evidence is there for effectiveness of dysarthria intervention?
2. Is online therapy acceptable to people with post-stroke dysarthria, their therapists and accessible in an NHS clinical context?

The Cochrane review, while finding more studies than in any previous Cochrane review on dysarthria (Sellars et al., 2005) still found that there is no, or insufficient evidence, that intervention for dysarthria is effective. This was the case for both stroke and brain injury. From a clinical perspective, this really demonstrates the need to continue to follow clinical guidelines. The Cochrane review led to a jointly written 'Evidently Cochrane' blog with a ReaDySpeech research advisor to explain the findings of the review in an accessible way for health professionals and patients (Appendix 10). The conclusions from this Cochrane review clearly state that we need research that is adequately powered, methodologically sound, clearly reported and free from bias.

The findings from the initial acceptability work confirmed recruitment of therapists and participants to try ReaDySpeech was possible, and that the ease of use, training and support were all considered acceptable to both therapists and participants. There were improvements made to the online programme during the trial in response to feedback around usability, navigation and content and some further amendments at the end of this initial testing. There were fewer technical access issues than expected. One particular stroke unit could not access Wi-Fi, which would need to be considered going forward. Although this was a small testing out phase, with a self-selecting group of existing technology users, it was considered sufficient support to take this intervention through to the next stage of feasibility testing.

7.3 Feasibility stage

The research questions for this stage were:

1. Can we design a feasibility randomised controlled trial for an online therapy?
2. Is it feasible to carry out a randomised controlled trial of online therapy ReaDySpeech for people with dysarthria after stroke?
3. What was the ReaDySpeech and usual care intervention delivered during a feasibility randomised controlled trial?

The feasibility of the randomised controlled trial was measured on recruitment and retention rates, time taken to carry out assessments, success of outcome assessor blinding as well as fidelity and adherence to the intervention. The findings from this trial did show it has the potential to be run as a large randomised controlled trial with reasonable recruitment and retention. However, prior to this, further feasibility work is indicated if we want to successfully reflect the wider stroke population to recruit participants longer than 13 weeks post stroke. We found the minimisation procedure we used did not result in balance across the two intervention groups. The groups, while well-matched for lesion location and days post-stroke, were not well matched for everyday functioning and dependence, with the ReaDySpeech group more impaired. We did not successfully blind the outcome assessor and would consider using videos for a future study. Low intensity of both ReaDySpeech and usual care intervention was found. ReaDySpeech was less likely to be delivered where sites had staffing problems although assistant delivery increased adherence. The findings from the in-depth description of the interventions, found that both ReaDySpeech and usual care intervention consisted of mostly impairment and activity level input. A future trial of effectiveness will need to consider how the intervention and control differ, whether

this is by: intervention content, timing, frequency, intensity, placebo or attention control.

7.4 Strengths of thesis

This work arose at the initial ideas stage from patient feedback and patient, carer public involvement (PCPI) is a strong feature of this research study. A lot of time at the development stage prior to the start of the research funding was spent with stroke groups in the community, specific research PCPI stroke groups and interviews or small groups with patients for their thoughts on technology solutions for rehabilitation. This progressed to patient involvement in designing and testing the ReaDySpeech programme and the research methodology. This has progressed to a working group of four then three patients who have experienced speech difficulties after stroke, called 'Ever-ready' to support the on-going research development, queries, opinions around the trial and ensuring the results can be easily understood by all participants (Appendix 9). This group have been an essential part of the research, shaping progress and future directions.

We have attempted throughout this research to provide clarity and openness about our research intentions, what we were trying to achieve and where we were unable to prevent bias. One way to do this was to publish our protocol so we could highlight differences from our planned protocol to the trial itself. This was a feasibility study and for this to be a realistic measure of whether a future trial is possible we attempted to reduce bias by working with an external clinical trials unit. They developed the minimisation programme for randomisation which was held externally, with anonymous emails to the lead researcher, who was attempting to carry out blinded outcome measures. They ensured at the end of the trial that all data had been saved and locked prior to releasing the minimisation code to avoid any bias around analysis.

7.5 Limitations of the thesis

This work demonstrated the development and feasibility testing of a novel computer-based dysarthria programme following the MRC framework for developing and evaluating complex interventions. The limitations of the methodology employed will be addressed in this section.

Both, during the development and feasibility stages of the process alternative approaches could have been selected. Although there will always be limitations to any approach chosen, on analysis there is always more information that would have been useful and we consider how this could be improved for future studies. There was potential to use more qualitative methodology in both the acceptability and feasibility study. This could have achieved more detailed and deeper insights using a more systematic interviewing and evaluation process.

In the development stage looking at acceptability, we carried out structured interviews with therapists only but did not interview the patients. The intention was for participants to give feedback to their therapists, who they already had a relationship with. We felt this 'light-touch' approach without introducing a research interviewer would be better for the participants so they could talk honestly and openly about the ReaDySpeech programme without the time taken to establish a rapport (DiCicco-Bloom and Crabtree, 2006). However, on reflection, in-depth interviews could have given much richer data than we achieved as we found that participants did not offer much information to the therapists other than being accepting of the intervention. Interviewing and analysis can be time consuming but the benefits of this approach

early in the development stage may have offered greater insight going forward to the next stage of feasibility.

The interviews with the therapists in the acceptability study and with the participants in the feasibility study involved structured and open questions. We found the therapists had a lot to say, but the participants could have been offered more support to encourage their views. Although comments were encouraged participants often needed prompting to elaborate and opportunity to do this (DiCicco-Bloom and Crabtree, 2006). The therapist interviews while providing rich data, were only broadly analysed into similar topics mainly due to time constraints. This data was used directly to improve the methodology and intervention for the feasibility study but there may have been useful information within this to offer a broader context. A more systematic approach to these data, potentially using thematic analysis which would have offered a flexible method of identifying and reporting the themes that were apparent in our interview data may have offered more insight into the therapist perspective (Braun and Clarke, 2006).

The feasibility work presented several limitations, some of which we had carefully planned for and thought we had avoided, others presented that were not expected. In terms of recruitment we had expected to get a higher proportion of acute referrals and to ameliorate this spent extra time with community colleagues prior to the start of the study in an effort to reduce this disparity. This approach did not address the lack of any referrals more than 13 weeks post stroke. We could potentially have pursued other recruitment options via charity and voluntary sectors but this would inevitably have had resource and time implications that would not have been sustainable for a feasibility trial. This mainly acute population recruited also affected the success of the

minimisation procedure we used as the groups were stratified by site and time post stroke. The resulting lack of balance between the randomised groups would need to be addressed in future work by stratifying by speech activity level or stroke severity and further feasibility work will test this out.

Staying with recruitment, we did not expect so few co-occurring aphasia and dysarthria referrals. We attempted to raise therapist awareness of the lack of these referrals through monthly newsletters and contact at recruitment, but for various possible reasons this referral rate did not change. Searching the evidence base for data indicated that little is known about the natural history of dysarthria, dysarthria with aphasia after stroke, as well as a lack of data around incidence and prevalence.

The benefits of process evaluation to carry out observation of and discussion with therapists following the feasibility trial would have enabled us to explore therapist views on identification and referral into the trial as well as intervention delivery (Moore et al., 2015). This feasibility trial carried out as part of a PhD did not plan to include process evaluation. It is possible however to conduct a process evaluation as part of a feasibility trial and this would need to be considered for future feasibility trials as well as a larger effectiveness trial. Some of the interesting issues raised in the research around the lack of participation intervention, lack of intensity, delivery by assistants and exercise selection versus adherence would be of interest to investigate in more depth. The use of process evaluation interviews and observation during the trial to monitor fidelity may have helped guide future research directions (Oakley et al., 2006). From a broader perspective, this type of analysis in a future trial may help us to go beyond the outcomes evaluation finding and look at whether wider

implementation is possible, what the intervention is doing and how this is influenced by the context of delivery (Carroll et al., 2007).

To avoid bias at outcome assessment, we attempted to implement several ways to avoid the outcome assessor finding out intervention allocation. The assessor was not involved in the intervention allocation as we used an external clinical trial unit for this and sent blinded emails so only the treating therapist knew allocation. Therapists and participants were asked not to reveal allocation but this proved surprisingly difficult, particularly for participants who were keen to discuss their treatment or inadvertently gave clues. Future studies will need to employ videos to ensure assessor blinding as our inability to blind the outcome assessor means the study will be considered biased and of reduced quality (Guyatt et al., 2008). There is evidence of clear risk of bias when the assessor is not blind (Hrobjartsson et al., 2013), although this has been reported transparently from the planned published protocol (Mitchell et al., 2017b) to the publication of the trial findings (Mitchell et al., 2017c, Submitted). This risk of bias was compounded by the participants allocated to the ReaDySpeech intervention, who reported being pleased to have been allocated to the experimental group. This highlights the importance of attempting to maintain equipoise throughout the trial in terms of our patient information language (Freedland et al., 2011; Pagoto et al., 2013). Inevitably, this high risk of bias would affect the level of evidence achieved (Burns et al., 2011) although we have been clear this study was not powered to compare groups (Ioannidis et al., 2017) and reported the inevitable bias of failure to blind outcome assessment.

This trial was designed to offer participants an opportunity to use a self-directed, online programme, so a different mode of delivery. As part of examining the feasibility

of this trial the content, timing and intensity of delivery of both interventions was not specified and it was not known how either would be implemented. Therapists were asked to carry out both interventions, according to their typical clinical practice. Both interventions in this trial were carried out at low intensity and the content of both were more similar than anticipated. We had expected that usual care would involve less face to face intervention and it could be possible that therapists involved in the trial offered a greater level of intervention than would usually be given outside of a trial situation due to the Hawthorne effect (McCambridge et al., 2014). This was a feasibility trial so although these are not necessarily limitations of this trial, they certainly indicate the limitations of our current knowledge. These findings have only increased the need to examine the detail of dysarthria intervention and whether differences in intensity, timing or a placebo or attention control should be examined before an effectiveness trial can be carried out.

7.6 Directions for further research

The limitations of the thesis outlined in the previous section will guide the direction of future research and improve the methodology and success as we progress towards the next stage of dysarthria research. Some of the limitations that need to be addressed relate to a more balanced use of qualitative and quantitative methods with a more systematic approach to analysis. Recruitment needs to be more reflective of the wider stroke population. We need to reduce bias around blinded outcome assessment, as well as bias around participant group allocation. The interventions or intervention and control need to be sufficiently different to measure change without compromising participant bias towards one or the other. We need to know more about the interventions, particularly intensity to be effective without a detrimental impact on adherence and for this to be clearly described following relevant guidelines (QUAlity and Transparency Of health Research (EQUATOR, 2014)).

The introduction of process evaluation to the next stage of feasibility research may enable us to improve our understanding of these limitations prior to a larger phase II trial of intervention effectiveness. The future work direction would be prioritised in the following work streams.

Workstream 1: The development work to establish the evidence base has confirmed that there is still variation being reported globally about the incidence and prevalence of dysarthria. This study reported fewer than expected numbers of patients with co-occurring dysarthria and aphasia but this remains an unknown (Flowers et al., 2013; Bowen et al., 2012a). Work would be carried out to analyse Sentinel Stroke National Audit Programme (SSNAP) data to establish the incidence and prevalence of dysarthria after stroke. This SSNAP data is based on the continuous reported data from all stroke

services in England. There is an acknowledgment that the diagnosis of dysarthria as part of the National Institutes of Health Stroke Scale (NIHSS) is not likely to be carried out by a speech and language therapist and may not be wholly accurate (Brott et al., 1989). However, concerns around accuracy of diagnosis must be considered alongside the advantages of the large sample size and inclusion of all stroke units in England, which demonstrates it is the best information we have despite possible limitations. Establishing the factual findings for dysarthria could potentially lead to quality improvement (Bray et al., 2016) and help to provide much needed information around early stroke motor and communication impairment (Dunn et al., 2016).

Workstream 2: From the development stage of this work the Cochrane review demonstrated the need for the development of a core outcome set as one of its recommendations (Mitchell et al., 2017a). The Cochrane review found there were eleven outcome measures used by the five trials included in the review. There are ethical concerns around the use of inadequate outcome measures used in research leading to wasted resources (Chalmers and Glasziou, 2009). Across medicine as a whole, there is real impetus to evaluate outcome measures (Hatemi et al., 2014) and a clear need, across other Cochrane groups, such as oral health, to establish core outcome sets (Taylor et al., 2014). It is clear that, as dysarthria research is in its infancy, establishing a set of standardised, valid, meaningful to patients and clinically relevant outcome measures should be a priority. This work stream would initially carry out a systematic review of available measures including the psychometric properties to establish which produce the most robust data. Following this an international working group of patients, speech and language therapists and other researchers would be set up and registered with COMET (Core Outcome Measures in Effectiveness Trials:

www.comet-initiative.org) to develop a consensus statement (Williamson et al., 2012). Again, there is much to be gleaned from the aphasia research data collaboration which has involved work to establish a core outcome set with patient, carer, therapist and research input to ensure they are both clinically relevant and meaningful to patients. International stroke organisations have also recognised the importance of data sharing (Saver et al., 2012) and developing a common set of outcome measures (Lees et al., 2012). The hope is that this will improve quality and relevance of research where a common goal is to examine effectiveness of intervention leading to improved communication in everyday activities and settings (Brady et al., 2014).

Workstream 3: To address several of the limitations reported in the development and feasibility stages of this research further feasibility work is indicated. Development of the feasibility trial protocol will be carried out with patient research advisors, speech and language therapists and statisticians to consider the comparator and whether we compare different interventions (whether this is content, timing or intensity), a placebo or attention control. The impact of this on recruitment and retention will need to be measured as part of this trial once this has been decided. This feasibility trial would aim to answer some of the following research questions potentially using an embedded process evaluation with a mixed methods approach:

- Can we recruit a more chronic stroke population i.e more than 12 weeks post stroke?
- Can we recruit and retain to a trial where the intervention may be self-delivered?
- Can we retain participants in a study where intensity is specified?

- Can we use videoed outcome measures without affecting recruitment and retention to the trial?
- Can we stratify by speech activity level impairment and achieve balance across allocation groups?

This would enable us to consider how or indeed whether to progress to the evaluation stage of ReaDySpeech. It seems essential that progress to a larger phase III trial of clinical effectiveness and cost-effectiveness of technology-based therapy for people with dysarthria after stroke needs to be given the best chance of producing a succesful trial. Whether effective or not, it is imperative that for those with dysarthria we produce good quality research findings that are clinically relevant to avoid the waste of research resources that occurs too often (Chalmers and Glasziou, 2009).

7.7 Final Conclusions

The aims of this thesis were to identify the existing evidence base with a systematic review, to carry out further development stage work testing out ReaDySpeech by finding out if it was acceptable with any technical barriers to use. If ReaDySpeech was acceptable to therapists and patients, the aim was then to carry out a feasibility randomised controlled trial. All of the intended aims were met, with the Cochrane systematic review carried out and completed, as well as the two studies one of early acceptability testing and the second a feasibility randomised controlled trial. As with all research there were some interesting and unexpected findings and there were many unanswered questions. Different methodological approaches may have enabled us to find out more information than we achieved but we were also constrained by the time limitations of a PhD thesis with one researcher to carry out the research. This reflection has enabled us to consider what could have been done differently and what should be done next.

The Cochrane review confirmed the lack of research studies into dysarthria, and the existing studies were all small and not adequately powered to report effectiveness of intervention. We found ReaDySpeech was acceptable for use in an NHS clinical context by therapists and selected patients, with no obvious technical barriers to use other than a lack of Wi-Fi in certain locations. The feasibility trial key finding was that we could recruit and retain participants so patients after stroke were willing to engage with technology as part of speech rehabilitation. We found that the time taken for the outcome measures were acceptable but assessor blinding was not successful. ReaDySpeech was less likely to be delivered with low staffing, but there was more

chance of participant adherence when assistants were involved in delivery. Intensity for both interventions was low, with similar content delivered.

Although we met the planned objectives and attempted to carry out transparent and unbiased research, it is clear there are always limitations to what can be achieved. There will always be wider philosophical considerations within a professional context about the use of randomised controlled trials with complex speech interventions (Lazar et al., 2008; Dunn et al., 2016) where little is still known about early stroke recovery. There are still differences between what is clinically carried out and what is considered best practice in published clinical guidance (Grol and Grimshaw, 2003). Research can still play an important role in offering clear evidence around clinically relevant unanswered questions to support implementation of evidence based findings.

This research has certainly highlighted the importance of the need to know more factual information about dysarthria after stroke and this work around incidence and prevalence in England should be a priority going forward. The lack of consensus around core outcome sets for dysarthria is another problem for moving forward and ensuring that dysarthria research is fit for purpose to examine effectiveness of intervention. Many questions remain about the intervention and how it works, or indeed whether it works at all. This complex intervention was found to mainly target the impairment and activity level with limited participation level intervention. Intensity was low and we are no further forward in finding out what the active ingredient are for dysarthria after stroke or whether intensity makes a difference to everyday speech outcome. The similarity of the interventions in this trial mean that a future trial of effectiveness will need to consider what the best comparator will be to ensure a meaningful outcome from the trial. Methodological improvements to stratify by speech or stroke severity

and ensuring outcome assessor blinding all need to be examined further in feasibility work.

Dysarthria after stroke is typically a devastating communication disability. The impact this can have on patients psychologically and in terms of participating in everyday life should not be underestimated. Having completed this body of research, there is greater need than ever to consider dysarthria research a priority, we need to gain momentum and drive forward the research agenda. This needs to involve collaborative working, data sharing and prioritisation agreement with people who have dysarthria, speech and language therapists and researchers. Clinical intervention must continue to follow the relevant guidelines working with patients, adhering to goal setting and using intervention options best suited to that individual. As therapists, we must influence research direction until we can produce better evidence to guide our intervention and influence best practice guidelines.

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Appendices

Appendix 1 CENTRAL

Cochrane Library databases (CDSR, DARE, CENTRAL, HTA) searched to May 2016
1. MeSH descriptor: [Cerebrovascular Disorders] this term only
2. MeSH descriptor: [Basal Ganglia Cerebrovascular Disease] explode all trees
3. MeSH descriptor: [Brain Ischemia] explode all trees
4. MeSH descriptor: [Carotid Artery Diseases] explode all trees
5. MeSH descriptor: [Cerebrovascular Trauma] explode all trees
6. MeSH descriptor: [Intracranial Arteriovenous Malformations] explode all trees
7. MeSH descriptor: [Intracranial Arterial Diseases] explode all trees
8. MeSH descriptor: [Intracranial Embolism and Thrombosis] explode all trees
9. MeSH descriptor: [Intracranial Hemorrhages] explode all trees
10. MeSH descriptor: [Stroke] this term only
11. MeSH descriptor: [Brain Infarction] explode all trees
12. MeSH descriptor: [Stroke, Lacunar] this term only
13. MeSH descriptor: [Vasospasm, Intracranial] this term only
14. MeSH descriptor: [Vertebral Artery Dissection] this term only
15. MeSH descriptor: [Hypoxia, Brain] explode all trees
16. stroke* or "post stroke" or poststroke or post-stroke or apoplex* or cerebrovasc* or CVA or SAH or "cerebral vasc*" (Word variations have been searched)
17. (brain or cerebr* or cerebell* or vertebrobasil* or hemispher* or intracran* or intracerebral or infratentorial or supratentorial or "middle cerebr*" or mca* or "anterior circulaion" or "basilar artery" or "vertebral artery") and (ischaemi* or ischemi* or thrombos* or thromboem* or emboli* or occlus* or hypoxi*) (Word variations have been searched)
18. (brain* or cerebr* or cerebell* or intracerebral or intracran* or parenchymal or intraparenchymal or intraventricular or infratentorial or supratentorial or "basal gangli*" or putaminal or putamen or "posterior fossal" or hemisphere* or subarachnoid) and (haemorrhag* or hemorrhag* or haematoma* or bleed*) (Word variations have been searched)
19. MeSH descriptor: [Hemiplegia] explode all trees
20. MeSH descriptor: [Paresis] explode all trees
21. MeSH descriptor: [Aphasia] explode all trees
22. MeSH descriptor: [Gait Disorders, Neurologic] explode all trees
23. (hemipar* or hemipleg* or paresis or paretic or aphasi* or dysphasi*) (Word variations have been searched)
24. MeSH descriptor: [Brain Damage, Chronic] explode all trees
25. MeSH descriptor: [Brain Injuries] this term only
26. MeSH descriptor: [Brain Concussion] explode all trees
27. MeSH descriptor: [Brain Hemorrhage, Traumatic] explode all trees
28. MeSH descriptor: [Brain Injury, Chronic] this term only
29. MeSH descriptor: [Diffuse Axonal Injury] this term only
30. MeSH descriptor: [Craniocerebral Trauma] this term only
31. MeSH descriptor: [Head Injuries, Closed] explode all trees
32. MeSH descriptor: [Intracranial Hemorrhage, Traumatic] explode all trees
33. MeSH descriptor: [Brain Abscess] explode all trees
34. MeSH descriptor: [Central Nervous System Infections] explode all trees
35. MeSH descriptor: [Encephalitis] explode all trees
36. MeSH descriptor: [Meningitis] explode all trees

37. (encephalitis or meningitis or "head injur*") (Word variations have been searched)
38. MeSH descriptor: [Brain Neoplasms] explode all trees
39. (brain or cerebr*) and (injur* or hypoxi* or damage* or concussion or trauma* or neoplasm* or lesion* or tumor* or tumour* or cancer* or infection) (Word variations have been searched)
40. {or #1-#39}
41. MeSH descriptor: [Dysarthria] this term only
42. MeSH descriptor: [Articulation Disorders] this term only
43. MeSH descriptor: [Speech Articulation Tests] this term only
44. MeSH descriptor: [Speech Disorders] this term only
45. MeSH descriptor: [Voice Disorders] this term only
46. MeSH descriptor: [Aphonia] this term only
47. MeSH descriptor: [Dysphonia] this term only
48. MeSH descriptor: [Communication Disorders] this term only
49. (dysarth* or dysphon* or anarth* or dyspros* or aphon* or dysfluen* or stutter* or stammer*) (Word variations have been searched)
50. (speech or articul* or disarticul* or phonat* or phonolog* or voice or vocal or prosod* or intonat* or respirat* or communicat* or fluen*) and (disorder* or impair* or problem* or difficult*) (Word variations have been searched)
51. speech and (slow* or weak* or imprecis* or intelligibil* or unintelligibil* or accuracy or fatigue) (Word variations have been searched)
52. {or #41-51}
53. MeSH descriptor: [Mouth] explode all trees
54. MeSH descriptor: [Larynx] explode all trees
55. MeSH descriptor: [Laryngeal Muscles] explode all trees
56. MeSH descriptor: [Pharynx] explode all trees
57. MeSH descriptor: [Pharyngeal Muscles] explode all trees
58. MeSH descriptor: [Facial Muscles] this term only
59. MeSH descriptor: [Palatal Muscles] this term only
60. (mouth or tongue or lingual or palat* or laryn* or pharyn* or orofacial or oro-facial or "face musc*" or facial musc*) (Word variations have been searched)
61. {or #53-#60}
62. MeSH descriptor: [Movement Disorders] this term only
63. MeSH descriptor: [Ataxia] this term only
64. MeSH descriptor: [Dystonia] this term only
65. MeSH descriptor: [Dystonic Disorders] this term only
66. MeSH descriptor: [Hyperkinesis] this term only
67. MeSH descriptor: [Hypokinesia] explode all trees
68. MeSH descriptor: [Muscle Hypertonia] this term only
69. MeSH descriptor: [Muscle Hypotonia] this term only
70. MeSH descriptor: [Muscle Weakness] this term only
71. MeSH descriptor: [Muscular Diseases] this term only
72. MeSH descriptor: [Muscle Spasticity] this term only
73.(atax* or dyston* or hyperkin* or hypokin* or hypoton* or hyperton* or flaccid* or spastic*) (Word variations have been searched)
74. {or #62-#73} 75. #61 and #74

Appendix 2 MEDLINE

MEDLINE (PubMed) from 1946 to May 2016
1. Search (("Cerebrovascular Disorders"[Mesh:noexp]) OR "Basal Ganglia Cerebrovascular Disease"[Mesh]) OR "Brain Ischemia"[Mesh]) OR "Carotid Artery Diseases"[Mesh]) OR "Cerebrovascular Trauma"[Mesh]) OR "Intracranial Arteriovenous Malformations"[Mesh]) OR "Intracranial Arterial Diseases"[Mesh]) OR "Intracranial Embolism and Thrombosis"[Mesh]) OR "Intracranial Hemorrhages"[Mesh]) OR "Stroke"[Mesh:noexp]) OR "Brain Infarction"[Mesh]) OR "Stroke, Lacunar"[Mesh:noexp]) OR "Vasospasm, Intracranial"[Mesh:noexp]) OR "Vertebral Artery Dissection"[Mesh:noexp]) OR "Hypoxia, Brain"[Mesh])
2. Search (stroke*[Text Word] OR "post stroke"[Text Word] OR poststroke[Text Word] OR post-stroke[Text Word] OR apoplex*[Text Word] OR cerebrovasc*[Text Word] OR CVA[Text Word] OR SAH[Text Word] OR cerebral vasc*[Text Word])
3. Search ((brain[Text Word] OR cerebr*[Text Word] OR cerebell*[Text Word] OR vertebrobasil*[Text Word] OR hemispher*[Text Word] OR intracran*[Text Word] OR intracerebral[Text Word] OR infratentorial[Text Word] OR supratentorial[Text Word] OR middle cerebr*[Text Word] OR mca*[Text Word] OR anterior circulation[Text Word] OR basilar artery[Text Word] OR vertebral artery[Text Word])) AND (Ischemi*[Text Word] OR infarct*[Text Word] OR thrombos*[Text Word] OR thromboem*[Text Word] OR emboli*[Text Word] OR occlus*[Text Word] OR hypoxi*[Text Word]))
4. Search (((Brain*[Text Word] OR cerebr*[Text Word] OR cerebell*[Text Word] OR intracerebral[Text Word] OR intracran*[Text Word] OR parenchymal[Text Word] OR intraparenchymal[Text Word] OR intraventricular[Text Word] OR infratentorial[Text Word] OR supratentorial[Text Word] OR basal gangli*[Text Word] OR putaminal[Text Word] OR putamen[Text Word] OR posterior fossa[Text Word] OR hemisphere*[Text Word] OR subarachnoid[Text Word])) AND (haemorrhag*[Text Word] OR hemorrhag*[Text Word] OR haematoma*[Text Word] OR hematoma*[Text Word] OR bleed*[Text Word]))
5. Search (("Hemiplegia"[Mesh]) OR "Paresis"[Mesh]) OR "Aphasia"[Mesh]) OR "Gait Disorders, Neurologic"[Mesh])
6. Search (Hemipar*[Text Word] OR hemipleg*[Text Word] OR paresis[Text Word] OR paretic[Text Word] OR aphasi*[Text Word] OR dysphasi*[Text Word])
7. Search (("Brain Damage, Chronic"[Mesh]) OR "Brain Injuries"[Mesh:noexp]) OR "Brain Concussion"[Mesh]) OR "Brain Hemorrhage, Traumatic"[Mesh]) OR "Brain Injury, Chronic"[Mesh:noexp]) OR "Diffuse Axonal Injury"[Mesh:noexp])
8. Search (("Craniocerebral Trauma"[Mesh:noexp]) OR "Head Injuries, Closed"[Mesh]) OR "Intracranial Hemorrhage, Traumatic"[Mesh])
9. Search (("Brain Abscess"[Mesh]) OR "Central Nervous System Infections"[Mesh]) OR "Encephalitis"[Mesh]) OR "Meningitis"[Mesh])
10. Search (encephalitis[Text Word] OR meningitis[Text Word] OR head injur*[Text Word])
11. Search "Brain Neoplasms"[Mesh]
12. Search (((brain[Text Word] OR cerebr*[Text Word])) AND (injur*[Text Word] OR hypoxi*[Text Word] OR damage*[Text Word] OR concussion[Text Word] OR trauma*[Text Word] OR neoplasm*[Text Word] OR lesion*[Text Word] OR tumor*[Text Word] OR tumour*[Text Word] OR cancer*[Text Word] OR infection[Text Word]))

13. Search (#1 OR #2 OR #3 OR #4 OR #5 OR #6 OR #7 OR #8 OR #9 OR #10 OR #11 OR #12)
14. Search (("Dysarthria"[Mesh:noexp]) OR "Articulation Disorders"[Mesh:noexp]) OR "Speech Articulation Tests"[Mesh:noexp])
15. Search ("Speech Disorders"[Mesh:noexp]) OR "Voice Disorders"[Mesh:noexp]) OR "Aphonia"[Mesh:noexp]) OR "Dysphonia"[Mesh:noexp]) OR "Communication Disorders"[Mesh:noexp])
16. Search (dysarth*[Text Word] OR dysphon*[Text Word] OR anarth*[Text Word] OR dyspros*[Text Word] OR aphon*[Text Word] OR dysfluen*[Text Word] OR stutter*[Text Word] OR stammer*[Text Word])
17. Search (((speech[Text Word] OR articul*[Text Word] OR disarticul*[Text Word] OR phonat*[Text Word] OR phonolog*[Text Word] OR voice[Text Word] OR vocal[Text Word] OR prosod*[Text Word] OR intonat*[Text Word] OR respirat*[Text Word] OR communicat*[Text Word] OR fluen*[Text Word])) AND (disorder*[Text Word] OR impair*[Text Word] OR problem*[Text Word] OR difficult*[Text Word]))
18. Search (speech[Text Word]) AND (slow*[Text Word] OR weak*[Text Word] OR imprecis*[Text Word] OR intelligibil*[Text Word] OR unintelligibil*[Text Word] OR accuracy[Text Word] OR fatigue[Text Word])
19. Search ("Mouth"[Mesh]) OR "Larynx"[Mesh]) OR "Laryngeal Muscles"[Mesh]) OR "Pharynx"[Mesh:noexp]) OR "Pharyngeal Muscles"[Mesh]) OR "Facial Muscles"[Mesh:noexp]) OR "Palatal Muscles"[Mesh:noexp])
20. Search (mouth[Text Word] OR tongue[Text Word] OR lingual[Text Word] OR palat*[Text Word] OR laryn*[Text Word] OR pharyn*[Text Word] OR orofacial[Text Word] OR oro-facial[Text Word] OR face musc*[Text Word] OR facial musc*[Text Word])
21. Search (#19 OR #20)
22. Search ("Movement Disorders"[Mesh:noexp]) OR "Ataxia"[Mesh:noexp]) OR "Dystonia"[Mesh:noexp]) OR "Dystonic Disorders"[Mesh:noexp]) OR "Hyperkinesis"[Mesh:noexp]) OR "Hypokinesia"[Mesh:noexp]) OR "Muscle Hypertonia"[Mesh:noexp]) OR "Muscle Hypertonia"[Mesh]) OR "Muscle Hypotonia"[Mesh:noexp]) OR "Muscle Weakness"[Mesh:noexp]) OR "Muscular Diseases"[Mesh:noexp]) OR "Muscle Spasticity"[Mesh:noexp])
23. Search (atax*[Text Word] OR dyston*[Text Word] OR hyperkin*[Text Word] OR hypokin*[Text Word] OR hypoton*[Text Word] OR hyperton*[Text Word] OR flaccid*[Text Word] OR spastic*[Text Word])
24. Search (#22 OR #23)
25. Search (#21 AND #24)
26. Search (#14 OR #15 OR #16 OR #17 OR #18 OR #25)
27. Search "Randomized Controlled Trials as Topic"[Mesh:noexp]
28. Search "Random Allocation"[Mesh:noexp]
29. Search "Controlled Clinical Trials as Topic"[Mesh:noexp]
30. Search "Control Groups"[Mesh:noexp]
31. Search ("Clinical Trials as Topic"[Mesh:noexp]) OR "Clinical Trials, Phase I as Topic"[Mesh:noexp]) OR "Clinical Trials, Phase II as Topic"[Mesh:noexp]) OR "Clinical Trials, Phase III as Topic"[Mesh:noexp]) OR "Clinical Trials, Phase IV as Topic"[Mesh:noexp])
32. Search "Double-Blind Method"[Mesh:noexp]
33. Search "Single-Blind Method"[Mesh:noexp]
34. Search "Placebos"[Mesh:noexp]

35. Search "Placebo Effect"[Mesh:noexp]
36. Search "Cross-Over Studies"[Mesh:noexp]
37. Search randomized controlled trial[Publication Type]
38. Search controlled clinical trial[Publication Type]
39. Search (clinical trial[Publication Type] OR clinical trial, phase i[Publication Type] OR clinical trial, phase ii[Publication Type] OR clinical trial, phase iii[Publication Type] OR clinical trial, phase iv[Publication Type])
40. Search (random*[Text Word] OR RCT[Text Word] OR RCTs[Text Word])
41. Search (controlled[Text Word]) AND (trial*[Text Word] OR stud*[Text Word])
42. Search (clinical*[Text Word] AND trial*[Text Word])
43. Search (control[Text Word] OR treatment[Text Word] OR experiment*[Text Word] OR intervention[Text Word])) AND (group*[Text Word] OR subject*[Text Word] OR patient*[Text Word])
44. Search (quasi-random*[Text Word] OR quasi random*[Text Word] OR pseudo-random*[Text Word] OR pseudo random*[Text Word])
45. Search (control[Text Word] OR experiment*[Text Word] OR conservative[Text Word])) AND (treatment[Text Word] OR therapy[Text Word] OR procedure[Text Word] OR manage*[Text Word])
46. Search (singl*[Text Word] OR doubl*[Text Word] OR tripl*[Text Word] OR trebl*[Text Word])) AND (blind*[Text Word] OR mask*[Text Word])
47. Search (cross-over[Text Word]) OR cross over[Text Word]) OR crossover[Text Word])
48. Search (placebo*[Text Word] OR sham[Text Word])
49. Search trial[Title]
50. Search (assign*[Text Word] OR allocat*[Text Word])
51. Search controls[Text Word]
52. Search (#27 OR #28 OR #29 OR #30 OR #31 OR #32 OR #33 OR #34 OR #35 OR #36 OR #37 OR #38 OR #39 OR #40 OR #41 OR #42 OR #43 OR #44 OR #45 OR #46 OR #47 OR #48 OR #49 OR #50 OR #51)
53. Search (#13 AND #26 AND #52)
54. Search ("Animals"[Mesh]) NOT "Humans"[Mesh:noexp])
55. Search (#53 NOT #54)

Appendix 3 Embase

Embase (Ovid) from 1974 to May 2016
1. CEREBROVASCULAR DISEASE/ or exp BASAL GANGLION DISEASE/ or exp BASAL GANGLION HEMORRHAGE/ or exp BRAIN ISCHEMIA/ or exp CAROTID ARTERY DISEASE/ or exp CEREBROVASCULAR ACCIDENT/ or exp CEREBRAL ARTERY DISEASE/ or exp BRAIN ARTERIOVENOUS MALFORMATION/ or exp BRAIN EMBOLISM/ or exp OCCLUSIVE CEREBROVASCULAR DISEASE/ or exp BRAIN HEMORRHAGE/ or exp BRAIN INFARCTION/ or LACUNAR STROKE/ or STROKE/ or BRAIN VASOSPASM/ or ARTERY DISSECTION/ or exp BRAIN HYPOXIA/
2. (stroke$ or post stroke or poststroke or post-stroke or apoplex$ or cerebral vasc$ or cerebrovasc$ or cva or SAH).ti,ab
3. ((brain or cerebr$ or cerebell$ or vertebrobasil$ or hemispher$ or intracran$ or intracerebral or infratentorial or supratentorial or middle cerebr$ or mca$ or anterior circulation or basilar artery or vertebral artery) adj5 (isch?emi$ or infarct$ or thrombo$ or emboli$ or occlus$ or hypoxi$)).ti,ab.
4. ((brain$ or cerebr$ or cerebell$ or intracerebral or intracran$ or parenchymal or intraparenchymal or intraventricular or infratentorial or supratentorial or basal gangli$ or putaminal or putamen or posterior fossa or hemispher$ or subarachnoid) adj5 (h?emorrhag$ or h?ematoma$ or bleed$)).ti,ab
5. exp HEMIPLEGIA/ or exp PARESIS/ or exp APHASIA/ or exp NEUROLOGIC GAIT DISORDER/
6. (hemipar$ or hemipleg$ or paresis or paretic or aphasi$ or dysphasi$).ti,ab
7. exp BRAIN DAMAGE, CHRONIC/ or BRAIN INJURY/ or exp BRAIN CONCUSSION/ or exp BRAIN HAEMORRHAGE, TRAUMATIC/ or BRAIN INJURY, CHRONIC/ or DIFFUSE AXONAL INJURY/
8. HEAD INJURY/ or exp HEAD INJURIES, CLOSED/ or exp INTRACRANIAL HEMORRHAGE, TRAUMATIC/
9. exp BRAIN ABSCESS/ or exp CENTRAL NERVOUS SYSTEM INFECTION/ or exp ENCEPHALITIS/ or exp MENINGITIS
10. (encephalitis or meningitis or head injur$).ti,ab.
11. exp BRAIN TUMOR/
12. ((brain or cerebr$) adj5 (injur$ or hypoxi$ or damage$ or concussion or trauma$ or neoplasm$ or lesion$ or tumor$ or tumour$ or cancer$ or infection$)).ti,ab.
13. 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12
14. DYSARTHRIA/ or SPEECH SOUND DISORDER/ or SPEECH ARTICULATION TESTS/
15. SPEECH DISORDER/ or VOICE DISORDER/ or APHONIA/ or DYSPHONIA/ or COMMUNICATION DISORDER/
16. (dysarth$ or dysphon$ or anarth$ or dyspros$ or aphon$ or dysfluen$ or stutter$ or stammer$).ti,ab
17. ((speech or articul$ or disarticul$ or phonat$ or phonolog$ or voice or vocal or prosod$ or intonat$ or respirat$ or communicat$ or fluen$) adj5 (disorder$ or impair$ or problem$ or difficult$)).ti,ab
18. (speech adj5 (slow$ or weak$ or imprecis$ or intelligibil$ or unintelligibil$ or accuracy or fatigue)).ti,ab
19. exp MOUTH/ or exp LARYNX/ or exp LARYNX MUSCLE/ or PHARYNX/ or exp PHARYNGEAL MUSCLE/ or FACE MUSCLE/ or PALATE/

20. (mouth or tongue or lingual or palat$ or laryn$ or pharyn$ or orofacial or oro-facial or face musc$ or facial musc$).ti,ab
21. 19 or 20
22. MOTOR DYSFUNCTION/ or ATAXIA/ or DYSTONIC DISORDER/ or HYPERKINESIA/ or HYPOKINESIA/ or MUSCLE HYPOTONIA/ or exp MUSCLE HYPOTONIA/ or MUSCLE WEAKNESS/ or MUSCLE DISEASE/ or SPASTICITY/
23. (atax$ or dyston$ or hyperkin$ or hypokin$ or hypoton$ or hyperton$ or flaccid$ or spastic$).ti,ab
24. 22 or 23
25. 21 and 24
26. 14 or 15 or 16 or 17 or 18 or 25
27. "RANDOMIZED CONTROLLED TRIAL (TOPIC)"/
28. RANDOMIZATION/
29. "CONTROLLED CLINICAL TRIAL (TOPIC)"/
30. CONTROL GROUP/
31. "CLINICAL TRIAL (TOPIC)"/ or "PHASE 1 CLINICAL TRIAL (TOPIC)"/ or "PHASE 2 CLINICAL TRIAL (TOPIC)"/ or "PHASE 3 CLINICAL TRIAL (TOPIC)"/ or "PHASE 4 CLINICAL TRIAL (TOPIC)"/
32. DOUBLE BLIND PROCEDURE/
33. SINGLE BLIND PROCEDURE/
34. PLACEBO/
35. PLACEBO EFFECT/
36. CROSSOVER PROCEDURE/
37. RANDOMIZED CONTROLLED TRIAL/
38. CLINICAL TRIAL/
39. PHASE 1 CLINICAL TRIAL/ or PHASE 2 CLINICAL TRIAL/ or PHASE 3 CLINICAL TRIAL/ or PHASE 4 CLINICAL TRIAL/
40. (random$ or RCT or RCTs).ti,ab
41. (controlled adj5 (trial$ or stud$)).ti,ab
42. (clinical$ adj5 trial$).ti,ab.
43. ((control or treatment or experiment$ or intervention) adj5 (group$ or subject$ or patient$)).ti,ab
44. (quasi-random$ or quasi random$ or pseudo-random$ or pseudo random$).ti,ab
45. ((control or experiment$ or conservative) adj5 (treatment or therapy or procedure or manage$)).ti,ab.
46. ((singl$ or doubl$ or tripl$ or trebl$) adj5 (blind$ or mask$)).ti,ab
47. (cross-over or cross over or crossover).ti,ab
48. (placebo$ or sham).ti,ab.
49. trial.ti
50. (assign$ or allocat$).ti,ab
51. controls.ti,ab.
52. 27 or 28 or 29 or 30 or 31 or 32 or 33 or 34 or 35 or 36 or 37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45 or 46 or 47 or 48 or 49 or 50 or 51
53. 13 and 26 and 52
54. exp ANIMALS/ not HUMANS/
55. 53 not 54

Appendix 4 CINAHL

CINAHL (NICE Evidence Services Portal HDAS) search strategy CINAHL (Ovid) from 1937 to May 2016

1. CEREBROVASCULAR DISORDERS/ OR exp BASAL GANGLIA CEREBROVASCULAR DISEASE/ OR exp HYPOXIA-BRAIN,ISCHEMIA/ OR exp CAROTID ARTERY DISEASES/ OR exp CEREBROVASCULAR CIRCULATION/ OR exp INTRACRANIAL ARTERIAL DISEASES/ OR exp ARTERIOVENOUS MALFORMATIONS/ OR exp INTRACRANIAL EMBOLISM AND THROMBOSIS/ OR exp INTRACRANIAL HEMORRHAGE/ OR STROKE/ OR STROKE,LACUNAR/ OR CEREBRAL VASOSPASM/ OR VERTEBRAL ARTERY DISSECTIONS/ OR exp HYPOXIA,BRAIN
2. (stroke* OR "post stroke" OR poststroke OR post-stroke OR apoplex* OR "cerebral vasc*" OR cerebrovasc* OR cva OR SAH OR "brain infarction" OR "cerebrovascular trauma").ti,ab
3. ((brain OR cerebr* OR cerebell* OR vertebrobasil* OR hemispher* OR intracran* OR intracerebral OR infratentorial OR supratentorial OR "middle cerebr*" OR mca* OR "anterior circulation" OR "basilar artery" OR "vertebral artery") adj5 (ischemi* OR ischaemi* OR infarct* OR thrombo* OR emboli* OR occlus* OR hypoxi*)).ti,ab;
4. ((brain* OR cerebr* OR cerebell* OR intracerebral OR intracran* OR parenchymal OR intraparenchymal OR intraventricular OR infratentorial OR supratentorial OR "basal gangli*" OR putaminal OR putamen OR "posterior fossa" OR hemispher* OR subarachnoid) adj5 (hemorrhag* OR haemorrhag* OR hematoma* OR haematoma* OR bleed*)).ti,ab;
5. exp HEMIPLEGIA/ OR exp PARALYSIS/ OR exp APHASIA/ OR exp GAIT DISORDERS,NEUROLOGIC/;
6. (hemipar* OR hemipleg* OR paresis OR paretic OR aphasi* OR dysphasi*).ti,ab;
7. exp BRAIN DAMAGE,CHRONIC/ OR BRAIN INJURIES/ OR exp BRAIN CONCUSSION/ OR exp INTRACRANIAL HEMORRHAGE/
8. ("chronic brain injury" OR "diffuse axonal injury" OR "craniocerebral trauma" OR "closed head injur*" OR "intracranial hemorrhag*").ti,ab
9. exp BRAIN ABSCESS/ OR exp CENTRAL NERVOUS SYSTEM INFECTIONS/ OR exp ENCEPHALITIS/ OR exp MENINGITIS/
10. (encephalitis OR meningitis OR "head injur*" OR "traumatic brain hemorrhag*" OR "chronic brain injury" OR "diffuse axonal injury" OR "craniocerebral trauma" OR "closed head injur*" OR "intracranial hemorrhag*").ti,ab
11. exp BRAIN NEOPLASMS/
12. ((brain OR cerebr*) adj5 (injur* OR hypoxi* OR damage* OR concussion OR trauma* OR neoplas* OR lesion* OR tumor* OR tumour* OR cancer* OR infection*)).ti,ab
13. 1 OR 2 OR 3 OR 4 OR 5 OR 6 OR 7 OR 8 OR 9 OR 10 OR 11 OR 12
14. DYSARTHRIA/ OR ARTICULATION DISORDERS/ OR SPEECH ARTICULATION TESTS/
15. SPEECH DISORDERS/ OR VOICE DISORDERS/ OR APHONIA/ OR DYSPHONIA,SPASMODIC/ OR DYSPHONIA,MUSCLE TENSION/ OR COMMUNICATIVE DISORDERS/
16. (dysarth* OR dysphon* OR anarth* OR dyspros* OR aphon* OR dysfluen* OR stutter* OR stammer*).ti,ab
17. ((speech OR articul* OR disarticul* OR phonat* OR phonolog* OR voice OR vocal OR prosod* OR intonat* OR respirat* OR communicat* OR fluen*) adj5 (disorder* OR impair* OR problem* OR difficult*))

18. (speech adj5 (slow* OR weak* OR imprecis* OR intelligibil* OR unintelligibil* OR accuracy OR fatigue)).ti,ab
19. exp MOUTH/ OR exp LARYNX/ OR exp LARYNGEAL MUSCLES/ OR PHARYNX/ OR exp PHARYNGEAL MUSCLES/ OR FACIAL MUSCLES/ OR PALATAL MUSCLES/
20. (mouth OR tongue OR lingual OR palat* OR laryn* OR pharyn* OR orofacial OR oro-facial OR "face musc*" OR "facial musc*").ti,ab
21. 19 OR 20
22. MOVEMENT DISORDERS/ OR ATAXIA/ OR DYSTONIA/ OR DYSTONIC DISORDERS/ OR HYPERKINESIS/ OR HYPOKINESIA/ OR MUSCLE HYPOTONIA/ OR exp MUSCLE HYPERTONIA/ OR MUSCLE WEAKNESS/ OR MUSCULAR DISEASES/ OR MUSCLE SPASTICITY/
23. (atax* OR dyston* OR hyperkin* OR hypokin* OR hypoton* OR hyperton* OR flaccid* OR spastic*).ti,ab
24. 22 OR 23 25. 21 AND 24
26. 14 OR 15 OR 16 OR 17 OR 18 OR 25
27. RANDOMIZED CONTROLLED TRIALS/
28. RANDOM ASSIGNMENT/
29. CLINICAL TRIALS/
30. CONTROL GROUP/
31. ("clinical trials" OR "clinical trials,phase i" OR "clinical trials,phase ii" OR "clinical trials,phase iii" OR "clinical trials,phase iv").ti,ab
32. DOUBLE-BLIND STUDIES/
33. SINGLE-BLIND STUDIES/
34. PLACEBOS/
35. PLACEBO EFFECT/
36. CROSSOVER DESIGN/
37. "randomized controlled trial".pt
38. "controlled clinical trial".pt
39. ("clinical trial" OR "clinical trial phase i" OR "clinical trial phase ii" OR "clinical trial phase iii" OR "clinical trial phase iv").pt
40. (random* OR RCT OR RCTs).ti,ab
41. (controlled adj5 (trial* OR stud*)).ti,ab
42. (clinical* adj5 trial*).ti,ab
43. ((control OR treatment OR experiment* OR intervention) adj5 (group* OR subject* OR patient*)).ti,ab
44. (quasi-random* OR "quasi random*" OR pseudo-random* OR "pseudo random*").ti,ab
45. ((control OR experiment* OR conservative) adj5 (treatment OR therapy OR procedure OR manage*)).ti,ab
46. ((singl* OR doubl* OR tripl* OR trebl*) adj5 (blind* OR mask*)).ti,ab
47. (cross-over OR "cross over" OR crossover).ti,ab
48. (placebo* OR sham).ti,ab
49. trial.ti
50. (assign* OR allocat*).ti,ab
51. controls.ti,ab
52. 27 OR 28 OR 29 OR 30 OR 31 OR 32 OR 33 OR 34 OR 35 OR 36 OR 37 OR 38 OR 39 OR 40 OR 41 OR 42 OR 43 OR 44 OR 45 OR 46 OR 47 OR 48 OR 49 OR 50 OR 51
53. 13 AND 26 AND 52
54. exp ANIMALS/ NOT HUMAN/ 55. 53 NOT 54

Appendix 5 PsycINFO

PsycINFO (Ovid) from 1800 to September 2016
1. cerebrovascular disorders/ or cerebral hemorrhage/ or exp cerebral ischemia/ or cerebral small vessel disease/ or cerebrovascular accidents/ or subarachnoid hemorrhage/
2. (stroke$ or poststroke or apoplex$ or cerebral vasc$ or brain vasc$ or cerebrovasc$ or cva$ or SAH).tw.
3. ((brain or cerebr$ or cerebell$ or vertebrobasil$ or hemispher$ or intracran$ or intracerebral or infratentorial or supratentorial or middle cerebral artery or MCA$ or anterior circulation or posterior circulation or basilar artery or vertebral artery or space-occupying) adj5 (isch?emi$ or infarct$ or thrombo$ or emboli$ or occlus$ or hypoxi$)).tw.
4. ((brain$ or cerebr$ or cerebell$ or intracerebral or intracran$ or parenchymal or intraparenchymal or intraventricular or infratentorial or supratentorial or basal gangli$ or putaminal or putamen or posterior fossa or hemispher$ or subarachnoid) adj5 (h?emorrhag$ or h?ematoma$ or bleed$)).tw.
5. hemiparesis/ or hemiplegia/
6. (hemipleg$ or hemipar$ or paresis or paretic).tw.
7. head injuries/ or exp brain concussion/ or brain damage/ or exp traumatic brain injury/
8. ((brain or cerebr$) adj5 (injur$ or hypoxi$ or damage$ or concussion or trauma$ or neoplasm$ or lesion$ or tumor$ or tumour$ or cancer$ or infection$)).tw.
9. 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8
10. dysarthria/ or articulation disorders/
11. dysphonia/ or speech disorders/
12. (dysarth$ or dyphon$ or anarth$ or dyspros$ or aphon$ or dysfluen$ or stutter$ or stammer$).tw.
13. ((speech or articul$ or disarticul$ or phonat$ or phonolog$ or voice or vocal or prosod$ or intonat$ or respirat$ or communicat$ or fluen$) adj5 (disorder$ or impair$ or problem$ or difficult$)).tw.
14. (speech adj5 (slow$ or weak$ or imprecis$ or intelligibil$ or unintelligibil$ or accuracy or fatigue)).tw.
15. "mouth (anatomy)"/ or exp tongue/ or larynx/ or pharynx/ or vocal cords/ or facial muscles/
16. (mouth or tongue or lingual or palat$ or laryn$ or pharyn$ or orofacial or oro-facial or face musc$ or facial musc$).tw.
17. 14 or 15
18. muscular disorders/ or movement disorders/ or ataxia/ or bradykinesia/ or dyskinesia/ or hyperkinesis/ or neuromuscular disorders/ or spasms/ or muscle spasms/
19. (atax$ or dyston$ or hyperkin$ or hypokin$ or hypoton$ or hyperton$ or flaccid$ or spastic$).tw.
20. 18 or 19
21. 17 and 20
22. 10 or 11 or 12 or 13 or 14 or 21
23. clinical trials/ or treatment effectiveness evaluation/ or placebo/
24. (random$ or RCT or RCTs).tw.

25. (controlled adj5 (trial$ or stud$)).tw.
26. (clinical$ adj5 trial$).tw.
27. ((control or treatment or experiment$ or intervention) adj5 (group$ or subject$ or patient$)).tw.
28. (quasi-random$ or quasi random$ or pseudo-random$ or pseudo random$).tw.
29. ((control or experiment$ or conservative) adj5 (treatment or therapy or procedure or manage$)).tw.
30. ((singl$ or doubl$ or tripl$ or trebl$) adj5 (blind$ or mask$)).tw.
31. (cross-over or cross over or crossover).tw.
32. (placebo$ or sham).tw.
33. trial.ti.
34. (assign$ or allocat$).tw.
35. controls.tw.
36. 23 or 24 or 25 or 26 or 27 or 28 or 29 or 30 or 31 or 32 or 33 or 34 or 35
37. 9 and 22 and 36

Appendix 6 LLBA

Linguistics and Language Behavior Abstracts (LLBA) search strategy
LLBA (ProQuest) 1976 to November 2016
(((dysarth* OR dysphon* OR anarth* OR dyspros* OR aphon* OR dyston*) OR ((speech OR articulat* OR voice OR vocal OR communicat*) AND (disorder* OR impair* OR problem* OR difficult*)) OR ((phonat* OR prosod* OR intonat* OR respirat*) AND (disorder* OR impair* OR problem* OR difficult*)) OR SU("Articulation Disorders" OR "Dysarthria"))) AND (SU("Brain Damage" OR "Stroke") OR (stroke* OR "post stroke" OR poststroke OR post-stroke OR apoplex* OR cerebrovasc* OR CVA OR SAH OR "cerebral vasc*"))

Appendix 7 Tidier from protocol

Template for Intervention Description and Replication for the ReaDySpeech intervention

- ReaDySpeech is an online programme which delivers exercises and strategies to improve intelligibility for people with dysarthria after stroke.
- This intervention is based on existing best practice guidelines which address impairment and activity levels of functioning.
- The exercises are selected by a speech and language therapist (slt) for each individual patient to be accessed via the ReaDySpeech online programme https://amie-test.herokuapp.com/. This can then be accessed with a username and password.
- The slt will select the intervention according to clinical need when considering the duration, intensity or dose of intervention as agreed with the patient.
- The programme will be adapted to the patients' progress.
  The activities used in the intervention include;
    - Practising articulation of
        - specific sounds in isolation
        - words of increasing syllable length
        - words with specific sounds in word initial position
        - words with specific sounds in word final position
        - complex clusters in word final position
        - short phrases and questions to repeat or answer
    - Breathing exercises
        - maintaining breath support
        - controlled breathing
        - breathing and speaking
    - Practising rate of speech and intelligibility strategies using an alphabet chart or a pacing board
    - Volume work
        - word level
        - speaking in short sentences
    - Facial expression
    - Intonation exercises
        - sentence level
    - Oro-motor exercises to improve range of movement, strength and speed of movement.
- All the activities appear on screen in written format and show progress, the oro-motor exercises include video clips and verbal instructions for each one.

- The intervention is used by the individual independently or with support from family, therapy assistant or speech and language therapist. There is no required level of training to support this intervention.
- The intervention is provided via the internet on any Wi-Fi enable device.
- The intervention could be accessed in any setting that has Wi-Fi access, for this study that could be in the acute or rehabilitation in-patient setting or the community in a patients home, residential or nursing home setting. If Wi-Fi access is not available this could be provided via a Wi-Fi enabled device using a paid for sim card.

Template for Intervention Description and Replication for 'USUAL CARE'

- Usual speech and language therapy for dysarthria is based on Royal College of Speech and Language Therapy Clinical Guidelines (Taylor-Goh, 2005) to deliver intervention at impairment, activity and participation levels of functioning. The guidelines are based on the best available evidence from research trials, case studies and expert opinion.
- Usual care is delivered by face to face sessions with a speech and language therapist or therapy assistant. Paper based materials describing exercises and including words/sentences to practice are used during the sessions and left with the patient for independent practice.
- Impairment level therapy focuses on exercises for function, strength, speed and precision of impaired musculature. This can also include: breathing exercises, work to improve resonance, phonation, articulation and prosody. Activity level therapy addresses compensatory approaches to speech such as reducing rate of speech, enhanced articulation, as well as environmental modifications and augmentative approaches such as an alphabet chart or text-to-speech aids. Participation level approaches involve education, psychological support, working with conversation partners and other person-centred approaches to support individuals in their work and life following stroke.
- Usual care is delivered by speech and language therapists, assistants or with family support according to the speech and language therapy departments' usual model of service delivery and responsive to patient need.
- Usual care is delivered in acute and rehabilitation in-patient settings and in any community setting.
- No specifications were given to therapists delivering usual care regarding the duration, intensity or dose of intervention other than to use it according to clinical need as agreed between therapist and patient. Usual care is expected to be responsive to patient need and variable between patients and departments according to service delivery.

Appendix 8 ReaDySpeech Feasibility Study: Participant interview questions

Section 1: What you thought about the treatment you received?

Usual care intervention:

Do you feel clear what 'usual' speech therapy care is? 5 yes completely clear what it is, 4 mostly clear about what it is, 3 not sure either way, 2 not much of an idea about what it is, 1 no idea at all about what it is

Can you describe what you think 'usual' speech therapy care is?

Do you think your speech therapy was helpful? 5 yes extremely helpful, 4 fairly helpful, 3 neither helpful nor unhelpful, 2 not particularly helpful, 1 not at all helpful

What 'effect' did the therapy have on you?

Do you think the therapy 'worked'?

What bit particularly worked and why?

Was the therapy easy to do? 5 yes very easy, 4 fairly easy, 3 not sure either way, 2 quite difficult, 1 very difficult

Would you recommend speech therapy care to others in your position? Yes/no

If you only had usual speech therapy care – did you do this work on your own as homework? Yes/no

How often did a therapist come to see you to carry out speech therapy?

Comments on your therapy input?

ReaDySpeech intervention:

Did you know how to use ReaDySpeech? Yes/no

Would you say your training was? 5 very thorough, 4 sufficient to use it, 3 Not sure either way, 2 Not particularly thorough, 1 Not at all thorough

How did you find using ReaDySpeech? 5 straightforward and easy to use, 4 fairly easy to use, 3 neither easy nor hard, 2 quite difficult to use, 1 really difficult to use

How often would you say you used ReaDySpeech? 5 every day, 4 every week, 3 when I remembered, 2 once a month or less, 1 rarely or never

What effect did ReaDySpeech have on you?

Do you think ReaDySpeech worked for you? If so why or why not?

Support needs – did you need help using ReaDySpeech? Yes or No

Who would help you with ReaDySpeech? Your therapist? Your family? Your partner?

Would you recommend ReaDySpeech to someone else in your position? Yes or No

Any thoughts on ReaDySpeech?

Section 2: What you thought the impact of the treatment was on you, your family and your wife/husband/partner/carer?

As part of your 'usual' care SLT treatment did any member of your family or friends take part in the treatment session? Yes or no

If the answer was yes can you say how often this was? 5 every single treatment session possible, 4 most of the treatment sessions, 3 attended now and again, 2 rarely attended, 1 never attended

If you had ReaDySpeech treatment as well as 'usual' SLT care how often did a family member or relative get involved in this work? Yes or no

If yes can you say how often? 5 Every time I used ReaDySpeech, 4 most times when I used ReaDySpeech, 3 Occasionally when I used ReaDySpeech, 2 rarely involved, 1 never involved

Did you feel 'usual' care SLT treatment affected any family members or friends in any way? Yes or no?

Did you feel ReaDySpeech affected family or friends in any way? Yes or no?

Section 3: Any other comments/questions/thoughts for the research team to consider?

ReaDySpeech Research Study

Thank you to everyone who took part

September 2015 to October 2016

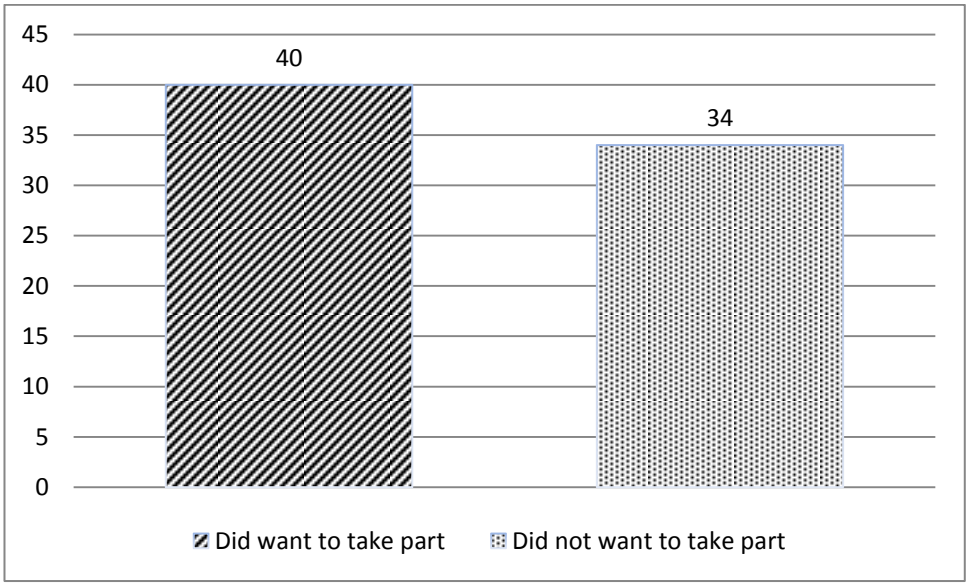
Claire.mitchell@manchester.ac.uk

Please email me for more detailed information

The research was looking at whether people would be interested in taking part in a study using an online programme for their speech exercises after stroke.

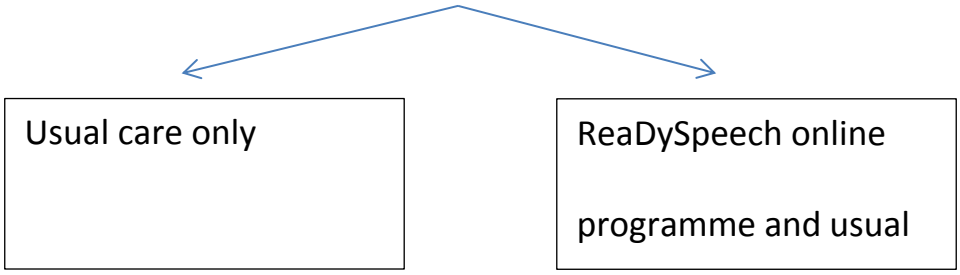
What we found?

- More than half of people (40 out of 74) who had speech problems after their stroke wanted to take part.



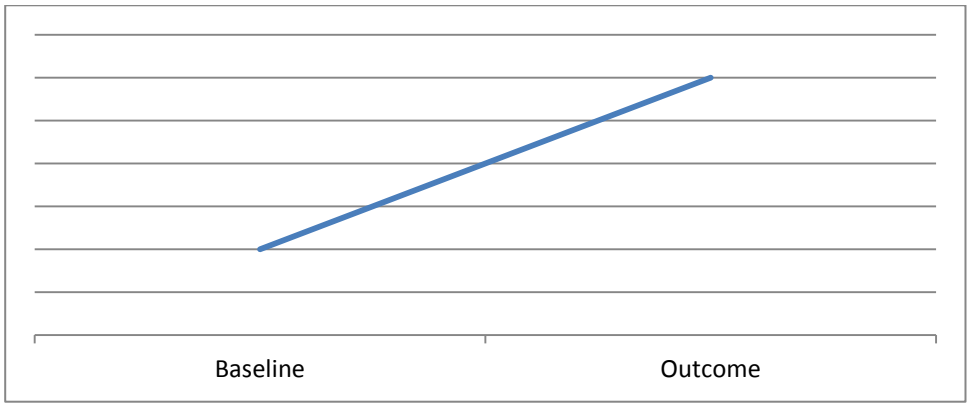
- The main reason for the 34 who didn't want to take part was a dislike of computers.
- ✓ Only 3 people dropped out.

The 40 who did take part were put (randomly) into 2 groups.



- Everyone understood the need to be put randomly in one group or another 
- Everyone thought the speech tests took an ok amount of time 
- Not everyone in the ReaDySpeech group got to use it, mostly due to low staff numbers
- Everyone improved their speech regardless of what treatment they got

The whole group showing improvements to speech



What's next?

- We want to recruit people who had their stroke longer than 3 months ago
- We want to know what happens if we ask people to practise more
  - Will it put them off?
  - Will they do better?
- We can then do a bigger research project
- With more people
- Does this really help people improve their speech after stroke?

Appendix 10 Evidently Cochrane blog

Sorry, what did you say? Living with dysarthria (unclear speech) after stroke

In this guest blog, Annette shares her story of living with dysarthria after stroke and Claire Mitchell (@ReaDySpeech), Speech & Language Therapist and author of a newly updated Cochrane review on dysarthria, explains the findings.

Annette's story

Two and a half year check-up today. I'm in good health. I should be feeling great. BUT, any mention of my dysarthria had me reaching for the tissues. I've finally got my head round my wobbly post-stroke body. I can't get my head around my wobbly speech.

A stroke 2 years ago left me with dysarthria (sounding as though I've drunk a few pints too many; reduced intelligibility; difficulty forming words; an inability to increase volume and pace). I am a 64 year old female. Pre-stroke, a bit of a social butterfly … and a talker! I am a changed person since developing dysarthria. I prefer being alone and in familiar places.

Dysarthria's effects are many, and profound. Why draw attention to my speech difficulties? If I stay home and keep quiet I can pretend that I'm 'normal'. I hate my new voice. It's not mine. Why put myself through the embarrassment of watching people straining to hear or understand me? Why put myself through the mental fatigue of constantly having to repeat myself?

But friends tell me I'm lucky to be affected so 'mildly'. Mildly??! Yes, my speech could be worse, even non-existent, but the emotional and psychological effects of dysarthria make it impossible to consider myself 'lucky'.

'Lucky' is a life without dysarthria.

I try to avoid the mental and physical effort required to form words – my mouth dries; my throat tightens; my tongue tires (weird). I want to go home. I want to be silent. That isn't me. Well, it isn't the 'me' of 2 years ago…. I force myself to socialise but any excuse to escape and I'm off. I even avoid engaging my nearest and dearest in conversation. Tiring, upsetting and frustrating, the reality of dysarthria smacks me in the mouth every time I attempt speech. I can be in bits with the effect dysarthria has had on my life. The reality of dysarthria smacks me in the mouth every time I attempt speech.

As a divorcee bringing up a child on my own I was used to standing on my own two feet. Now I feel vulnerable because of the restricted communication which my dysarthric speech places on me. I rely on other's patience to hear me out. In my experience people generally (wrongly) associate dysarthria with low or impaired intellect because it isn't 'normal'.

Adapting to the physical restrictions of stroke was painfully slow but the long term emotional effects of my speech problems seem to get worse, not better.

I seem unable to accept the change. I want further improvement but I don't know if that's possible or how to achieve it. I had my standard 6 weeks of speech therapy but I want more, I would be willing to try anything. I avoid interactions… misunderstandings affect me emotionally. I'm a tough old bird but I could cry when I have to repeatedly repeat myself. I just withdraw.

Before my stroke I occasionally worked as a TV extra. The agency said I could return whenever I felt ready. Physically, I've been ready for months but my confidence to speak has evaporated. Others may view my dysarthria as 'mild' but its effects have been (and still are) devastating and stifling. Dysarthria has a stranglehold on every aspect of my life. I've always worked and been financially independent but now feel this opportunity is no longer open to me.

Dysarthria is imprisoning, limiting my life to the people and places that I know. I would no more engage in a new friendship or relationship than fly. Dysarthria has robbed me of the confidence to try.

Why put my head above the parapet and risk being shot?

Stick with what I know.

It's safer.

It's also uncharacteristic (of me).

Dysarthria has created a psychological barrier which I am struggling to get over. Dysarthria has robbed me of confidence. I now prefer to avoid speaking. I do my best but it's exhausting. I just want to go home and be alone and quiet.

What's the evidence for dysarthria treatment after stroke or non-progressive brain injury?

Annette's story is a powerful reminder for clinicians and researchers of the impact dysarthria can have and highlights the importance of rehabilitation research.

In this updated Cochrane systematic review we wanted to find out if there is evidence that any dysarthria treatments work. We found surprisingly few trials (5) with small numbers of patients (234) and almost all participants were post-stroke.

None of the included studies had sufficient numbers to answer the question about whether dysarthria intervention works. We still don't know the benefits or risks of intervention and it is important that people with dysarthria continue to receive rehabilitation according to clinical guidelines. Our final conclusion for this review is that there have been no definitive trials focussed solely on dysarthria.

What more do we need to do?

Research into dysarthria has clearly been neglected over the years and this seems inadequate when considering the impact on people such as Annette. This review shows we urgently need better quality trials with bigger numbers of patients to find out more about dysarthria treatments and consider patients' views on interventions and how we measure change.

What does Annette think about the research?

We don't yet know what works best for people and we need to find out. "In a strange way it was heartening to discover that so little research has been done on dysarthria. It gave me hope that with further research people like me might move on with a useful and meaningful life. It is not saying nothing works it is saying we don't yet know what works best for people and we need to find out. I feel that dysarthria has severely restricted my recovery. Ongoing research is required into something which affects recovery so profoundly. Even slight improvements would improve the quality of my life drastically through increased confidence and self-belief."

The lack of evidence around dysarthria treatment after stroke prompted Claire to look at how treatment could be developed. She is Chief investigator of the ReaDySpeech feasibility study which is looking at whether a computer programme can be used in a randomised controlled, multi-centre trial for dysarthria treatment after stroke. Annette is currently acting as a research advisor for the ReaDySpeech study and was asked to comment on the Cochrane systematic review from a patient perspective.

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