Mechanisms of change underlying increased intelligibility for children with cerebral palsy following speech and language therapy focusing on breath support and speech rate

SHORT TRIAL TITLE

Speech change following dysarthria therapy

This protocol has regard for the HRA guidance and order of content

RESEARCH REFERENCE NUMBERS: 18/NW/0752; IRAS 249670

This protocol covers research undertaken as part of the study described in 18/NW/0752; IRAS 249670

This protocol focuses on changes to children's speech that account for gains in intelligibility following therapy targeting breath control, speech effort and rate.

18/NW/0752; IRAS 249670 also includes investigation of linguistic and acoustic-phonetic influences on children's speech intelligibility prior to therapy.

PROTOCOL VERSION NUMBER AND DATE: V3.0 14.12.2020

OTHER RESEARCH REFERENCE NUMBERS

SPONSOR: Newcastle University

FULL/LONG TITLE OF THE TRIAL

Mechanisms of change underlying increased intelligibility for children with cerebral palsy following speech and language therapy focusing on breath support and speech rate

SHORT TRIAL TITLE / ACRONYM

Speech change following dysarthria therapy

PROTOCOL VERSION NUMBER AND DATE

- V3.0 14.12.2020

KEY CONTACTS

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Committees	Study Oversight Committee Dr Leah Li UCL

i. LIST of CONTENTS

iii. STUDY SUMMARY

Trial Title	Mechanisms of change underlying increased intelligibility for children with cerebral palsy following speech and language therapy focusing on breath support and speech rate	
Internal ref. no. (or short title)	Speech change following dysarthria therapy	
Clinical Phase		
Trial Design	Observational Study: Secondary analysis of previously collected data.	
Trial Participants	42	
Planned Sample Size	42	
Treatment duration	6 weeks	
Follow up duration	1 week	
	Objectives	Outcome Measures
Primary	Is increased intelligibility associated with listener perception of sounds that demand the greatest intra-oral pressure and tightest constriction of the vocal tract (plosive sounds e.g. t, g; fricatives sh, z; affricates ch, j); sounds at the ends of words, because intra-oral pressure is sustained across the word; and sounds within clusters of consonants (e.g. str) from increased articulatory precision from maintaining a steady rate	Percentages of the following sounds perceived correctly: sounds at the start and end of words; individual sounds within consonant clusters; individual single consonants
		Measurements of features of children's speech waveforms known to differentiate individual speech sounds (e.g. bursts of acoustic energy in plosive sounds e.g. p).
Intervention	A speech and language therapy programme that aims to help children to speak more clearly by controlling their breath supply to create a stronger speech signal and by talking at a	

	slower rate to give them time to produce speech movements precisely.
Formulation, Dose, Route of Administration	1-1 therapy, delivered by a research speech and language therapist. 3 x 45 minute sessions per week for 6 weeks.

iv. FUNDING AND SUPPORT IN KIND

FUNDER(S)	**FINANCIAL AND NON FINANCIALSUPPORT GIVEN**
NIHR Efficacy and Mechanism Evaluation programme	Funding of study NIHR130967 £116,716

v. ROLE OF TRIAL SPONSOR AND FUNDER

Newcastle University has overall responsibility for the initiation and management of the study.

The research team from Newcastle University (L Pennington, G Khattab, V Ryan) and University of Sheffield (S Cunningham, S Sehgal) are responsible for study design, conduct, data analysis and interpretation, manuscript writing, and dissemination of results.

The Efficacy and Mechanism Evaluation programme is funded by the MRC and NIHR, with contributions from the CSO in Scotland, NISCHR in Wales and the HSC R&D, Public Health Agency in Northern Ireland. This report is managed by the NIHR Evaluation, Trials and Studies Coordinating Centre (NETSCC) (Efficacy and Mechanism Evaluation (EME), NIHR130967 - Mechanisms of change underlying increased intelligibility for children with cerebral palsy following speech and language therapy focusing on breath support and speech rate). The views expressed in this publication are those of the author(s) and not necessarily those of the MRC, NHS, the National Institute for Health Research or the Department of Health and Social Care.

vi. ROLES AND RESPONSIBILITIES OF TRIAL MANAGEMENT COMMITEES/GROUPS & INDIVIDUALS

Study Steering Committee, led by Dr Leah Li (UCL) will:

- Provide advice to the Project Funder (NIHR EME), the Project Sponsor (Newcastle University), the Chief Investigator (L Pennington on all appropriate aspects of the project
- Focus on progress of the study, adherence to the protocol, data security, and the consideration of new information of relevance to the research question
- Consider the rights, safety and well-being of the participants as the most important, prevailing over the interests of science and society

- Ensure appropriate ethical and other approvals are obtained in line with the project plan
- Agree proposals for substantial protocol amendments and provide advice to the sponsor and funder regarding approvals of such amendments
- Provide advice to the investigators on all aspects of the trial/project. The SSC will have a majority independent representation, including the Chair. It will meet regularly and send reports to the sponsor.

Trial Management Group

- The Trial Management Group will comprise Pennington, Khattab, Ryan and Cunningham.
- The TMC will meet fortnightly to ensure all practical details of the trial are progressing well and working well and everyone within the trial understands them.

vii. KEY WORDS: Secondary analysis, mechanisms of change, speech intelligibility, children, dysarthria, speech therapy

1 BACKGROUND

Cerebral palsy (CP) is thought to be the most common cause of physical disability in children, with an estimated 2-2.5 children being born with CP per 1000 live births (Sellier et al, 2016). Research by Parkes, Hill, Platt, & Donnelly (2010) showed that the speech of approximately one quarter of these children will be affected by their cerebral palsy, causing the motor speech disorder dysarthria.

One of the most prominent characteristics of dysarthria is poor speech intelligibility. Several studies have suggested acoustic-phonetic variables that may contribute to speech intelligibility. Ansel and Kent, 1992) found that listeners were unable to perceive contrasts between pairs of consonants that varied in voicing (vocal folds vibrating / open) and the manner in which they were made (full or partial constriction of the vocal tract). Weismer (2008) found a strong correlation between the size of an acoustic vowel space and speech intelligibility at single word level. Hustad and colleagues observed an association between intelligibility, intensity (perceived as loudness), vocal characteristics (relating to vocal fold vibration) and intelligibility (Lee et al, 2014; Allison and Hustad, 2018). The production of stress (measured by intensity) has been found to increase vowel intelligibility in words produced by speakers with mild dysarthria and CP (Connaghan & Patel, 2017).

We developed a speech and language therapy programme that aims to help children with cerebral palsy and dysarthria to speak more clearly by controlling their breath supply to create a stronger speech signal and by talking at a slower rate to give them time to produce speech movements precisely. Following the therapy, listeners have understood children better. In this study we will examine if increased intra-oral pressure generated by greater breath supply and precise, coordinated movement of the articulators, resulting from a steady speech rate, allow children to differentiate the production of individual speech sounds, thereby making them easier for listeners to distinguish. We predict that after intervention speech sounds appearing at the end of words will be produced more clearly, given that the children will maintain the loudness and precision across full words and across utterances. We also predict that sounds that demand the firmest contact between lips, tongue and palate will show the greatest change in production. These changes will be investigated at a perceptual level (the intelligibility of words pre- and post-therapy) and at an acoustic-phonetic level (using speech waveforms).

2 RATIONALE

The study will investigate the impacts of intervention that teaches children with dysarthria to control their breath supply and speech rate, which should help them to speak louder and more precisely. This investigation will allow us to ascertain which factors have the greatest influence on therapy success and to then tailor intervention to these characteristics. It is possible that factors influencing intelligibility will differ across children. The research will show which factors are most influential for children with particular pre-therapy speech characteristics. Results will be used to refine the therapy programme, to include measurement of the factors that are most influential in this study and the focus on the factors that have the greatest influence on intelligibility for individual children (e.g. being loud at the end of words for some children; being slow and clear in words containing particular speech sounds for others).

3 OBJECTIVES AND OUTCOME MEASURES/ENDPOINTS

The aim of this study is to investigate if increases in intelligibility following therapy focussing on breath control and speech rate are associated with changes to the production of individual sounds (phonemes) within words.

3.1 Primary objective

Therapy focusing on breath supply and speech rate will facilitate differentiation of the articulation of individual phonemes, enabling listeners to better identify individual phonemes in words spoken in isolation and in connected speech. Consequently:

1. Word-final consonants will be perceived more frequently following intervention, due to breath support being sustained across the word.
2. Word-initial consonant clusters (e.g. str) will be perceived correctly more frequently after intervention, as a steady rate and stronger speech signal will allow speakers to differentiate each phoneme in the cluster.
3. The manners of articulation that will increase in intelligibility most will be those requiring tightest constriction and intraoral pressure – plosives (e.g. p, d), affricates (e.g. ch) and fricatives (e.g. f, z), rather than nasals, approximants (r, l, w, y) and vowels. Switching from voiced to voiceless is likely to be related to the extent of children's motor disorder maybe outside their volitional control.
4. Acoustic change will be associated with phonetic change.

We will investigate if patterns of change are common across children who share characteristics, e.g. motor disorder, pre-therapy level of speech intelligibility.

3.2 Outcome measures/endpoints

Perceptual data

Single words

We will carry forward the mean percentage of words understood in single word speech pre- and post-therapy from our previous studies.

We will generate the following mean percentage of listeners' correct perception of (across words):

- single consonants at the start of words
- single consonants at the end of words
- consonant clusters at the start of words
- consonant clusters at the end of words
- monosyllabic words perceived as monosyllables
- bisyllabic words perceived as bisyllabic

For single consonants we also examine speakers' ability to mark voice, place and manner correctly at the beginning and end of words pre- and post-therapy. In each word position, we will calculate the mean percentage of listeners' correct perception of:

- Voiced consonants; voiceless consonants
- Bilabial; labiodental; alveolar; velar consonants
- Plosives; fricatives; affricates; nasal consonants

For clusters at the beginning and end of words, we will calculate the proportion of clusters that contain two consonants (e.g. <u>sw</u>ing) and three consonants (e.g. <u>str</u>ing) perceived correctly at pre and post therapy.

Connected speech

The same proportions for will be generated for words in connected speech as those in single words. We will also examine:

- The percentage of content words and function words perceived correctly pre- and post-therapy.
- The percentage of stressed words perceived correctly pre- and post-therapy.

Acoustic data

The mean and SD of each measure will be taken for each child for words in the single words condition and the connected speech condition at pre- and post-therapy.

4 STUDY DESIGN

Observational study: Secondary analysis of data collected in three phase II studies.

5 STUDY SETTING

Single centre study.

6 PARTICIPANT ELIGIBILITY CRITERIA

The data from 42 children with cerebral palsy and dysarthria who have taken part in previous studies and whose speech samples were analysed in RfPB PB-PG-0712-28077 (comprising data from three studies) will be used in this project.

Fifty-three children with CP and dysarthria were recruited by local speech and language therapists (SLTs) within the north of England. They were all diagnosed with moderate to severe dysarthria, as assessed by local their SLT. All classifications of CP were included; spastic, dyskinetic, ataxic, mixed type and Worster Drought syndrome (Clark, Carr, Reilly & Neville, 2000). The group comprised 30 boys, 23 girls, age range 6-18 years (Mean = 10.6 years, SD = 3.69). Twenty-nine children had spastic type CP, fifteen had dyskinetic type, three had ataxic, two had Worster Drought. Participants were excluded from the studies if they had one or more of the following; bilateral hearing impairments (>50dBHL), visual impairments (not correctable with glasses) or difficulties following simple instructions.

Forty-two of the children received individual speech and language therapy focussed on breath control and speech rate in 18 sessions, provided thee times per week for six weeks. Children were recorded on two separate days at: six weeks and one week pre-therapy and one week, six weeks and twelve weeks after therapy completion.

Adult listeners, who had no experience of conversing with children or adults with speech disorders, also took part in the study as raters of the speech (three listeners per recording).

7 PROCEDURES

Database development

This project will conduct further analyses of the children's single word and connected speech data and will produce four linked databases:

1. Single word speech database showing the consonants perceived (in)correctly in word initial and word final positions pre- and post-therapy, from orthographic transcriptions (both as singletons and in consonant clusters)

2. Connected speech database showing the words perceived (in)correctly in each sentence position and phonemes (vowels and consonants) perceived correctly within each word position pre- and post-therapy, from orthographic transcriptions
3. Phonetic-acoustic database of measures showing segmental characteristics of children's single word speech that differentiate the voicing and manner of consonants, taken from spectrographic analysis. Scripts will be developed for PRAAT software for phonetic-acoustic analysis of single words. Further computer scripts will be developed to automate the uploading of acoustic measurements to the database.
4. Phonetic-acoustic database of segmental measures of children's connected speech, showing the characteristics that differentiate individual vowels and the voicing and manner of consonants taken from spectrographic analysis speech for each word produced in a phrase, plus the length of the phrase in words, the position of the word in the phrase, and the type of word (see below). PRAAT scripts will be developed for phonetic-acoustic analysis of individual words in connected speech. Further scripts will be developed to automatically upload the measurements to the database.

1. Single word speech database showing the phonemes perceived (in)correctly in word initial and word final position pre- and post-therapy

The database will show which consonants are perceived correctly in single words before and after therapy for each child.

Because words vary in their constituent number of syllables and phonemes, and the therapy may affect children's speech in different ways, the database will show whether whole words were perceived correctly and whether constituent phonemes were perceived correctly.

We will develop computer scripts to automatically expand the existing database, which currently shows each of the 100 target words produced pre-therapy and the 100 target words produced post-therapy, plus the words perceived by each listener. The scripts will generate the following data for each word:

- The target word:
    - The consonant/consonant cluster in word initial and word final position
    - The number of consonants in target cluster
    - The number of syllables in the word
      For each consonant in within the word:
        - target consonant place of articulation (bilabial, labiodental, alveolar, velar)
        - target consonant voicing (voiced/voiceless)
        - target consonant manner (plosive, fricative, affricate, approximant, nasal)
- The word perceived by each listener:
    - The consonant / consonant cluster perceived in word initial and word final position
    - The number of consonants perceived
    - The number of syllables perceived
      For each individual consonant perceived:
        - consonant place of articulation
        - consonant voicing
        - consonant manner

2. Connected speech database showing the words perceived correctly in each sentence position and phonemes (vowels and consonants) perceived correctly within each word position pre and post therapy from orthographic transcriptions

We will expand the database that currently contains the phrases each child spoke, the phrases listeners perceived the children to say and the percentage of words perceived correctly by each listener in the pre-therapy and post-therapy recordings. We will add: the number of words in each target phrase; the position of the target word within the phrase; and the linguistic class of target and perceived words: content words (nouns, verbs); modifiers (adjectives, adverbs); function words (articles, prepositions, conjunctions, pronouns); 'social/other' (e.g. 'yeah' and 'shh'). These factors may act as moderators in intelligibility. The influence of phrase length may be bidirectional. For speakers with less impaired speech, longer phrases may be more intelligible than shorter ones, as listeners will have greater access to top-down processing using world and linguistic knowledge if they can detect initial words. For speakers with very severe dysarthria, the stress to speech motor control engendered by longer utterances may further reduce the articulatory differentiation of individual sounds, making them less easy to perceive and providing little access to top-down processing. Content words and modifiers are open class, meaning they contain a larger number of words (for example, 'sunflower', 'cup', 'pretty') and so are more difficult to process using top-down knowledge. However, they usually spoken at higher intensity than function words, and the greater control on breath supply following intervention should aid their perception. Function words are often monosyllabic, and hence easier to produce. They are closed class (e.g. it, in, with) and appear frequently in British English making them predictable to listeners, aiding their decoding in top down processing.

For each word, we will follow the process described for the single word orthographic data in section 1. above, to generate complementary data derived in 1.1 - 1.3 for individual words produced in connected speech.

3. Phonetic-acoustic database of measures showing segmental characteristics of children's single word speech

We will measure acoustic features that differentiate the manner of consonant production and their voicing using PRAAT acoustic analysis software. While place of articulation is less well reliably determined in acoustic analysis, we will also use known measures in stop bursts and the onset/offset of surrounding vowels in order to examine changes in place of plosives. We will analyse word initial and final consonants from monosyllabic words. We hypothesise that the production of word final consonants may change due to increased breath support and steady speech rate. Production of phonemes is affected by the sounds that immediately precede and follow them (coarticulation), so separate characteristics will be measured for word initial and final consonants. Our analysis will be two-fold: for each category of sound, we will carry out an acoustic profiling which looks for the presence of the main characteristics of these sounds pre- and post-therapy; this will be followed by measures of the magnitude and duration of sound/sound features in order to examine the strength of the articulation.

- Plosives: Plosives are signalled by a closure, followed by a release burst, and in the case of voiceless stops, aspiration. For initial stops we will measure burst intensity, voice onset time, and f0 (mean fundamental frequency) and formant frequencies in the following vowel. For final stops we will measure the duration of the preceding vowel along with formants at offset, closure duration and burst intensity.

- Fricatives: Fricatives are recognised by the presence and duration of frication. Fricatives are differentiated from affricates by their duration and their rate of onset of the frication (rise time). Fricatives have longer rise time than affricates. We will measure the total duration of frication and the rise time, as the point of frication to its maximum. We will also measure spectral moments to explore place of articulation.
- Affricates: Affricates involve a stop-like burst followed by frication followed. Using the contrast information for fricatives, we will measure burst intensity and duration, the duration of frication, rise time, and spectral moments.
- Nasals: Nasals have low amplitude formant-like structure and antiformants due to the extra nasal cavity. We will measure duration, formants and a range of amplitude and harmonic measures (*A*1-*P*1, *A*1-*P*0*; B*1*; H*1-*A*1) in the nasal and preceding/following vowel in order to examine voice quality effects of nasal production and potential excess breathiness or creak.
- Voicing: Voiced and voiceless stops are distinguished by voice onset time (VOT), f0 and the duration of the preceding/following vowel. Voicing in fricatives is distinguished by their overall duration, energy and their influence on f0, formants and duration of following vowels. Cues for voicing will be measured in the consonants themselves as well as their following vowels.
- Consonant clusters: acoustic profiling will look for evidence of occurrence of two separate consonants. In the absence of one we will still look for cues of partial acquisition of clusters in terms of duration, voicing patterns, and intensity of the consonant that is produced and the surrounding vowels.

NB. Although we hypothesise that the sounds requiring the tightest constriction and greatest precision will change most, we need to measure all consonants to ascertain why consonants are misperceived; for example a plosive (e.g. *p)* may be perceived as a nasal *(*e.g. *m)* because there is insufficient burst.

4. ***Phonetic-acoustic database of segmental measurements of children's connected speech***

We will create time-aligned transcriptions for utterances produced by the children in PRAAT, with separate tiers for utterance-, phrase-, word- and segment-level realisation. These 'text grids' show the portions of the waveforms that relate to individual words and phonemes. We currently have time-aligned transcriptions for 11 children. We will carry out the same type of acoustic profiling and measurements of target sounds as detailed above, with added tagging for position of the segment in relation to word and phrase-level stress, and sentence position.

Across the connected speech samples, we will also look at vowel realisation as a function of prosodic position, examining the effect of syllable/word/phrase stress of vowel realisation. In sentence medial position, we will examine the potential effect of co-articulation arising from speech connected processes on the realisation of initial and final consonants. We will also measure the effect of connected speech on consonant realisation and potential compression/reduction. We will use methods developed at the University of Sheffield to quantify the amount of deviation in the phase of the speech signal. Previous work has shown this measure can be used to quantify the coordination of articulations, and be useful for predicting intelligibility(.

Computer scripts will transfer the resulting acoustic measurements to databases 3 and 4.

7.2 Consent

The analysis proposed in this study has been given a favourable opinion by North West - Preston Research Ethics Committee (18/NW/0752). We have permission to analyse anonymised perceptual data (transcriptions) from all 42 participants and nonanonymised data from the 24/42 participants who gave permission for their data to be further analysed in further research. Nonanonymised data are sound files, which researchers need to listen to in order to time-align text grids for acoustic analysis in PRAAT. We have time-aligned text grids for acoustic data from a further 11 participants from our first study, which could allow for anonymous analysis of their acoustic data (giving a total of 35 participants in acoustic analysis). A substantial amendment to (18/NW/0752) permitted analysis of these data (Amendment 1, date of favourable opinion 26/10/2020).

7.3 Storage and analysis of clinical samples

Data to be analysed in this study are stored on a password-protected Newcastle University server.

8 STUDY TREATMENTS

No treatment will be provided as part of this study.

9 STATISTICS AND DATA ANALYSIS

For each child the change in each outcome measure from pre- to post-therapy will be described using numerical and graphical summaries (e.g. spider web plots), with the aim being to identify patterns of change across the children by visual inspection. Exploratory multivariate graphical and numerical descriptive techniques, will also be utilised with the aim of potentially reducing the dimensionality of the dataset, identifying groups of outcome measures that are more closely associated (principal component analysis) or uncovering groups of children with common intelligibility features (cluster analysis).

Based on the data from the three studies included in this secondary analysis, the standard deviation of the change in percent of single words heard from pre intervention to 1 week post intervention was approximately 10% - if similar variability is observed for the change from baseline for other outcome measures (e.g. % change in articulation measures) then, based on a sample size of 42, a mean change from baseline would be estimated with a 95% CI of ±3.1%. Further, with n=42 a 95% CI for an observed correlation of 0.5 would be 0.23 to 0.70. Considering the sample size requirements for principal components analysis and cluster analysis, a recent simulation study concluded that sample sizes of 40+ are sufficient for exploratory multivariate analyses. Previous studies exploring the relationships between patterns of speech and intelligibility in patients with dysarthria have involved comparable numbers of patients.

Correlations (with 95% confidence intervals (CIs)) between the change in the percentage of words understood by listeners and the change in articulation measures will be calculated. For each outcome measure, the mean change from pre to post therapy (with 95% CIs) will be estimated using a linear mixed model with child fitted as random to account for multiple measures on the same child at each of two time points. Both model fit summary statistics and graphical examination of model residuals will be used to assess the adequacy of the specified model. Sensitivity analyses will be performed around any possible misspecification identified in the model checking. If data do not approximately follow a Normal distribution, appropriate techniques will be employed where possible, such as the use of a simple transformation or the use of an alternate error structure. If necessary, a summary measures approach will

be used to derive a single measure at each time point for each child and fixed effect analyses will be performed. Correlations (with 95% CIs) between the change in the percentage of words understood by listeners and the change in articulation measures will be calculated. There will be no adjustment to the CIs for multiple comparisons, but the total number of outcome measures analysed will be reported.

10 DATA MANAGEMENT

10.1 Data Management Plan

The following data management plan has been developed using the Medical Research Council guidance and template for data management in research

0. Proposal name
Mechanisms of change underlying increased intelligibility for children with cerebral palsy following speech and language therapy focusing on breath support and speech rate
1. Description of the data
1.1 Type of study Observational study: secondary analysis of previously collected data **1.2 Types of data** A. Orthographic transcriptions of words spoken by children with dysarthria and cerebral palsy and words perceived by naive listeners. B. Sound files (wav format) of words spoke by children, all identifiable data removed. **1.3 Format and scale of the data** A: Words are stored in two MS Excel spreadsheets, one containing repeated single words and one containing phrases produced by children when describing pictures and answering questions. Each child repeated 100 single words pre-therapy and 100 words post-therapy and produced up to 12 phrases (containing up to 11 words) pre- and post-therapy. Single word and phrases were heard by three listeners. B: One wav file per single word and phrase spoken by each child. Estimated files size 6300 MB - 12600 MB
2. Data collection / generation
A: Databases of single words and phrases spoken and perceived were created for previous research. We will add: the number of words in each target phrase; the position of the target word within the phrase; and the linguistic class of target and perceived words: content words (nouns, verbs); modifiers (adjectives, adverbs); function words (articles, prepositions, conjunctions, pronouns); 'social/other' (e.g. 'yeah' and 'shh'). B: We will create time-aligned transcriptions for utterances produced by the children in PRAAT (acoustic-phonetic analysis software), with separate tiers for utterance-, phrase-, word- and

segment-level realisation. These 'text grids' show the portions of the waveforms that relate to individual words and phonemes.

3. Data management, documentation and curation

The confidentiality, integrity and availability of the data will be managed through adherence to the 10 Data Guardian Standards.

3.1 Managing, storing and curating data.

During the project data will be stored on the Newcastle University UK based file store which hosts the university's Microsoft account and data. The file store is hosted across two data centres, equipped with fire detection, suppression equipment and secure audited access procedures. The University's file store system operates 'Shadow Copies' , which are taken four times daily. An incremental copy to backup tape is taken nightly, and a full copy monthly. Backups are kept for ninety days. Inactive tapes are stored in on-campus fireproof safes.

Data will be managed and stored in accordance with University of Newcastle processes: https://research.ncl.ac.uk/rdm/duringaproject/datastorage/

Collaborators at the University of Sheffield will analyse data on a remotely-accessed PC holding a copy of the data (a 'virtual machine'), with any means of downloading a copy of the dataset disabled.

1.2 Metadata standards and data documentation

Metadata will include title, description, keywords, creators, funders and access conditions. Supporting documentation will describe the data and the context of the data including: software used, methodology and dates of collection. Metadata and documentation will exist in the university and the funding agency. University guidance on metadata and documentation will be followed:
https://research.ncl.ac.uk/rdm/duringaproject/organise/documentation/#d.en.484401

1.3 Data preservation strategy and standards

The Newcastle University policy on data preservation is for a 10-year storage period. Data to be stored will include the data in spreadsheets, analysed data, and meetings that underpin publications and/or with long-term value.

4. Data security and confidentiality of potentially disclosive information

Qualitative data collected by interviews and the Community of Practice will be anonymised at transcription. Video recorded data will only be accessible to the research team and anonymised during analysis.

4.1 Formal information/data security standards

National Data Guardian standards and principles will be adhered to and guidance for data security and standards for internet-mediated research[16] will be used. Newcastle University IT has a dedicated security team, which includes a member who is trained in ISO/IEC 27001:2005 and ISO/IEC 27001:2013 auditing and is also a certified PCI-SSC Internal Security Assessor. An internal information security risk assessment is completed every three months. The findings of this risk assessment are subject to review by the University's IT Information Security Forum and form the basis of a risk treatment plan. This risk treatment plan is a key part of an on-going quality assurance process to ensure that information security risks are mitigated through improvement of the managed information security controls.

4.2 Main risks to data security

Potential risks include poor quality data, data loss, breaches of anonymity and confidentiality, and invasions of privacy. For mitigating these, please see above. Additional security measures will include: up-to-date anti-virus software on data collection devices; use of private Wi-Fi networks; and use of apps licensed to the university. Access to data will be carefully controlled and restricted to the research team until made formally available. MRC and Data Guardian guidance on the confidentiality and data security will be followed, together with the University Good Research Practice polices, and NHS Research Ethics Governance requirements.

5. Data sharing and access

5.1 Suitability for sharing

Permission for sharing data was not obtained at their collection for the original studies and hence data analysed for this study are not suitable for sharing.

5.2 Discovery by potential users of the research data

A metadata record will be created in Newcastle University's externally facing data catalogue and within the funders' online resources. A link to the metadata record will be included in publications that use data from the project, and on a study web page.

5.3 Governance of access

A data access policy will be developed by the research team and the sponsor with support from Newcastle University's Research Data Service. Based on the policy, a data access statement will be produced to accompany the metadata.

5.4 The study team's exclusive use of the data

The study team will retain access to the data.

5.5 Restrictions or delays to sharing, with planned actions to limit such restrictions

The CI will discuss with the Newcastle University Research and Enterprise Service before any metadata is made public.

5.6 Regulation of responsibilities of users

No data will be shared externally.

6. Responsibilities
The CI will have the overall responsibility for quality of the data and data management with supervision and guidance from the wider research team. The Research Data Service team at Newcastle University https://www.ncl.ac.uk/research/researchgovernance/researchdata-openaccess/#researchdatamanagement will assist with data management, storage, documentation, and sharing. The University Information Security Officer can provide guidance and training on data protection. Newcastle University, as sponsor, will ensure the research is conducted in line with the standards and protocol outlined.
7. Relevant institutional, departmental or study policies on data sharing and data security

Policy	**URL or Reference**
Data Management Policy & Procedures	https://research.ncl.ac.uk/rdm/beforeaproject/expectations/
Data Security Policy	https://research.ncl.ac.uk/rdm/duringaproject/datastorage /
Data Sharing Policy	https://mrc.ukri.org/documents/pdf/mrc-data-sharing-policy/
Institutional Information Policy	https://www.ncl.ac.uk/itservice/policies/
Other: University Code of Good Research Practice UK policy Framework for Health and Social Care Research	https://www.ncl.ac.uk/research/researchgovernance/goodpractice/ https://www.hra.nhs.uk/planning-and-improving-research/policies-standards-legislation/uk-policy-framework-health-social-care-research/

8. Author of this Data Management Plan (Name) and, if different to that of the Principal Investigator, their **telephone & email contact details**
Lindsay Pennington Lindsay.pennington@ncl.ac.uk

10.2 Archiving

- No personal data will be used in this study.
- Original data (collected as part of previous studies) will be archived in accordance with the approvals obtained for the previous studies.
- Electronic databases, analyses and reports created for this study will be archived securely by Newcastle University as per the Data Management Plan.

11 ETHICAL AND REGULATORY CONSIDERATIONS

11.1 A favourable opinion of the study was granted by North West - Preston Research Ethics Committee, as an amendment to 18/NW/0752 Motor speech disorder in childhood: association between word and speaker characteristics on intelligibility (date of favourable opinion 26/10/2020).

11.2 Public and Patient Involvement

The study will involve three key groups of stakeholders - young people (aged 12 to 18 years), parents and speech and language therapists - as PPI advisors. Three advisory groups will comprise: 1) five young people with dysarthria and cerebral palsy; 2) five parents of children with cerebral palsy and dysarthria (not necessarily the parents of the young people's PPI group); 3) five speech and language therapists who work with children with dysarthria in mainstream and special and schools and colleges. There will be two meetings for each of the groups to orientate the groups to the aims of the study, its methods and their role as advisors; discuss the results in relation to advisors' experience of the impact of articulation on intelligibility (e.g. difficulties with certain speech sounds, sounding out the end of words) and the potential the optimisation of the therapy protocol. PPI advisors will also try out the computer scripts we develop to measure children's speech, to check that they can be used easily in therapy. PPI members will be invited to contribute to dissemination activities, such as social media, regional meetings and formal reports, and to be involved in the future application to test the individualisation of the refined therapy protocol to be submitted to NIHR HTA programme.

11.3 Amendments

Amendments will be submitted to REC and favourable opinion provided prior to their implementation.

12 DISSEMINIATION POLICY

A project website will be developed at the start of the project. We will use email and social media for dissemination that can be distributed online or through Facebook or Twitter. In addition to our report to the funder, we will prepare at least two articles for submission to major journals in motor disorders (Movement Disorders) and clinical speech research (Journal of Speech, Language, and Hearing Research). Papers will be open access so that they are readily available to therapists as well as researchers. We will present the findings at the European Academy of Childhood Disability annual meeting, which is attended by parents of children with cerebral palsy, multidisciplinary researchers and clinicians from across Europe and internationally. We will also submit our results to the International Congress of Phonetic Sciences and Conference of the International Speech and Communication Association (with no costs incurred as members of the team already attend these meetings), to bring the testing and application of acoustic measurements in childhood dysarthria to the attention of theoretical phoneticians, linguists, speech scientists and computer scientists. We will discuss the study and its results, including the acoustic measurement scripts, with parents and clinicians at regional neurodisability meetings and speech and language Clinical Excellence Network to further encourage the provision of therapy focussing on speech for children with dysarthria. We have previously presented at these meetings and been invited to give regular updates on our intervention.

13 REFERENCES

Allison, K. M., & Hustad, K. C. (2014). Impact of sentence length and phonetic complexity on intelligibility of 5-year old children with cerebral palsy. International Journal of Speech-Language Pathology, 16(4), 396-407.

Ansel, B. M., & Kent, R. D. (1992). Acoustic-phonetic contrasts and intelligibility in the dysarthria associated with mixed cerebral palsy. Journal of Speech, Language and Hearing Research, 35, 296-308.

Clark, M., Carr, L., Reilly, S., & Neville, B. G. R. (2000). Worster-Drought Syndrome, a mild tetraplegic perisylvian cerebral palsy. Brian, 123, 2160-70.

Connaghan, K. P., & Patel, R. (2017). The Impact of Contrastive Stress on Vowel Acoustics and Intelligibility in Dysarthria. Journal of Speech, Language and Hearing Research, 60, 38-50.

Lee, J., Hustad, K. C., & Weismer, G. (2014). Predicting Speech Intelligibility with a Multiple Speech Subsystems Approach in Children with Cerebral Palsy. Journal of Speech, Language and Hearing Research, 57, 1666-1678.

Parkes, J., Hill, N., Platt, M. J., & Donnelly, C. (2010). Oromotor dysfunction and communication impairments in children with cerebral palsy: a register study. Developmental Medicine and Child Neurology. 52, 1113-1119.

Patel, R., Hustad, K. C., Connaghan, K. P., & Furr, W. (2012). Relationship Between Prosody and Intelligibility in Children with Dysarthria. Journal of Medical Speech Language Pathology, 20(4), 17.

Sellier E, Platt MJ, Andersen GL, Krägeloh-Mann I, De La Cruz J, Cans C, et al. Decreasing prevalence in cerebral palsy: A multi-site European population-based study, 1980 to 2003. Developmental Medicine and Child Neurology. 2016;58(1):85-92. 7.

Weismer, G. (2008). Speech intelligibility. In M. J. Ball, M. R. Perkins, N. Muller, & S. Howard (Eds), The handbook of clinical linguistics (pp. 568-582). Oxford, UK: Blackwell.

Protocol change log

Amendment No.	Protocol version no.	Date approved	Author(s) of changes	Details of changes made
1	2	REC favourable opinion 26/11/2020	Lindsay Pennington	Title: Motor speech disorder in childhood: association between word and speaker characteristics on intelligibility Added comparison of perceptual and acoustic features of single word and connected speech to original protocol that considered pre-therapy data only.
1	3.0		Lindsay Pennington	Title changed to 'Mechanisms of change underlying increased intelligibility for children with cerebral palsy following speech and language therapy focusing on breath support and speech rate' This version of the protocol considers comparison of perceptual and acoustic features of single word and connected speech pre- and post-therapy. This version of the protocol covers the work funded by 'EME Project:NIHR130967 - Mechanisms of change underlying increased intelligibility for children with cerebral palsy following speech and language therapy focusing on breath support and speech rate' only Format of the protocol changed in accordance with HRA guidance REC approval covered by Amendment 1 (dated 26/10/2020).