ISRCTN17619184 https://doi.org/10.1186/ISRCTN17619184

Effectiveness of speech therapy for people with Friedreich's ataxia

Submission date
05/11/2018

Recruitment status
No longer recruiting

Registration date
16/11/2018

Overall study status
Completed

Last Edited
03/09/2020

Condition category
Nervous System Diseases

[_] Prospectively registered
[_] Protocol
[_] Statistical analysis plan
[X] Results
[_] Individual participant data

Plain English summary of protocol
Ataxia is characterised by uncoordinated movements, which can affect walking, hand movements as well as speech. The resulting speech disorder is called ataxic dysarthria. People with ataxic dysarthria experience imprecise articulation, changes to voice quality, slow speech and running out of breath while speaking. This results in reduced speech intelligibility and a breakdown in communication, which is reported to be one of the most upsetting symptoms of their disease by people who are diagnosed with a degenerative disease. Whilst researchers have found out lot about the nature of the communication problems experienced by speakers with ataxic dysarthria, there are very few intervention studies. One treatment approach that has potential to increase communication efficiency is Lee Silverman Voice Treatment (LSVT). This treatment approach focuses on increasing the level of loudness in a person's speech. The method has been shown to improve breath support for speech, and improve voice quality and articulation, whilst at the same being simple enough for the patient to implement in everyday communication. There are many reports showing the benefits of LSVT for people with Parkinson' s Disease (PD), and it is the only treatment for dysarthria that has been investigated according to gold standard clinical trial guidelines with a randomised controlled study. Although LSVT was initially designed to help people with PD, there have also been reports of its effectiveness in other types of dysarthria, such as cerebral palsy, traumatic brain injury and stroke. In addition, a case study on one patient with ataxic dysarthria found improvements in speech intelligibility following a course of LSVT. We are performing a feasibility study to investigate whether this result can be replicated with a larger group of patients, whether there might be any negative side effects and what the best measures are to test whether the treatment has worked.

We will focus on people with hereditary ataxia, specifically Friedreich's ataxia. We will measure their speech difficulties before and after therapy, and In addition, explore the acceptability of the treatment to patients by asking them about their opinions and experiences.

Together, these data will allow us to design a future large scale randomised control study into finding an effective treatment for communication problems in this population.

Background and study aims
Ataxia is characterised by uncoordinated movements, which can affect walking and hand movements, as well as speech. The resulting communication disorder is called ataxic dysarthria.

People with ataxic dysarthria experience problems with pronouncing words correctly, speaking loud and fast enough, and having enough breath to talk. This means other people have trouble understanding them. The resulting breakdown in communication is often reported to be amongst the most upsetting symptoms of their disease. Researchers have found out a lot about the communication problems experienced by speakers with ataxic dysarthria and why they are happening, but don't know yet how to help these people improve their speech.
This study aims to test whether a treatment approach called Lee Silverman Voice Treatment (LSVT), which was successfully developed for Parkinson's Disease, can also help people with ataxia. We want to find out whether this therapy can improve communication, whether there might be any side effects and what the best measures are to test whether the treatment has worked. These results will allow us to design a future larger study for finding an effective treatment for communication problems in people with ataxia.

Who can participate?
We will recruit people with hereditary ataxia, mainly Friedreich's ataxia.

What does the study involve?
Participants will receive 16 one-hour sessions of LSVT treatment over a period of 8 weeks from an accredited speech and language therapist, i.e. 2 sessions per week. In addition, they will be asked to complete homework practice. The treatment will be delivered remotely via Skype.
We will measure participants' speech and how it impacts on their lives twice before and twice after therapy. We will also ask them about their opinions and experiences of the therapy programme and Skype delivery after treatment is complete.

What are the possible benefits and risks of participating?
Participants will potentially gain direct benefit from the therapy if it helps them with their communication. There is a small risk that their voice could be negatively affected, in which case we will immediately stop the treatment. We will also monitor fatigue very carefully to ensure the treatment does not impact on this.

Where is the study run from?
Strathclyde University

When is the study starting and how long is it expected to run for?
March 2017 to July 2019

Who is funding the study?
Ataxia UK

Who is the main contact?
Professor Anja Lowit, a.lowit@strath.ac.uk

Contact information

Type(s)
Scientific

Contact name
Prof Anja Lowit

ORCID ID

http://orcid.org/0000-0003-0842-584X

Contact details
School of Psychological Sciences and Health
Strathclyde University
40 George St
Glasgow
United Kingdom
G1 1QE
+44 (0)141 5483058
a.lowit@strath.ac.uk

Additional identifiers

EudraCT/CTIS number

IRAS number
224092

ClinicalTrials.gov number

Secondary identifying numbers
161378, UEC/25, IRAS 224092

Study information

Scientific Title
Effectiveness of Lee Silverman Voice Treatment in improving communication in people with Friedreich’s ataxia

Study objectives
Lee Silverman Voice Treatment will result in improved communication of people with Friedreich's ataxia.

Ethics approval required
Old ethics approval format

Ethics approval(s)
South Central - Hampshire B Research Ethics Committee, 10/04/2017, REC ref: 17/SC/0161. Protocol number: UEC/25; IRAS project ID: 224092

Study design
Non-randomised feasibility study

Primary study design
Interventional

Secondary study design
Non randomised study

Study setting(s)
Home

Study type(s)
Treatment

Participant information sheet
Not available in web format, please use contact details to request a participation information sheet

Health condition(s) or problem(s) studied
Friedreich's ataxia, other hereditary ataxias

Interventions
A group of 20 patients with Friedreich's Ataxia and communication problems will be recruited. There are no restrictions regarding the severity of the ataxia or the speech problem. All participants will receive the same treatment and no control group will be recruited. Participants will be recruited internationally through charity channels, as well one UK health board.

Participants will be offered Lee Silverman Voice Treatment (LSVT) in its extended form, i.e. two 1-hour sessions per week over a period of 8 weeks. Sessions will be conducted via Skype in the participant's own home.

Intervention Type
Behavioural

Primary outcome measure
Participants will be assessed before and after treatment. Multiple baseline assessments will be taken (assessments 1 & 2, approximately 2 weeks apart). Performance will then be re-assessed immediately following (assessment 3) and 6-8 weeks after the end of treatment (assessment 4).
1. Maximum loudness on prolonged /a/ and mean loudness of a reading passage measured acoustically in decibel at assessments 1, 2, 3 & 4
2. Voice quality of prolonged /a/ evaluated acoustically (shimmer, jitter, harmonics to noise ratio) at assessments 1, 2, 3 & 4
3. Articulation and speech rate of a reading passage assessed acoustically in syllables per minute at assessments 1, 2, 3 & 4
4. Intelligibility of a reading passage measured perceptually on a 9-point scale according to Dobinson (2007) at assessments 1, 2, 3 & 4

Secondary outcome measures
Participants will be assessed before and after treatment. Multiple baseline assessments will be taken (assessments 1 & 2, approximately 2 weeks apart). Performance will then be re-assessed immediately following (assessment 3) and 6-8 weeks after the end of treatment (assessment 4).
1. Communication participation assessed with the Communication Participation Item Bank short form (CPIB, Baylor et al. 2012) at assessments 1 & 3
2. Impact of the voice problem assessed with the Voice Handicap Index (VHI, Jacobson et al. 1997) at assessments 1 & 3
3. Levels of communication impact, confidence and engagement evaluated with a self-constructed, unpublished questionnaire on a Visual Analogue Scale, and baseline and exit semi-structured interviews at assessments 1 & 3
4. Participants’ evaluation of therapy programme and Skype delivery established during exit

interview at assessment 3
5. Fatigue measured with the Fatigue Severity Scale (FSS, Krupp et al, 1989) at assessments 1 & 3

Overall study start date
08/03/2017

Completion date
30/07/2019

Eligibility

Key inclusion criteria
1. Confirmed diagnosis of hereditary ataxia, primarily Friedreich's ataxia
2. Presence of a speech impairment
3. Ability to perform assessment and treatment tasks
4. No uncorrected hearing loss
5. Availability of appropriate technology to record voice and operate Skype

Participant type(s)
Patient

Age group
Adult

Sex
Both

Target number of participants
The target number of participants is 20

Total final enrolment
20

Key exclusion criteria
1. Ataxia due to non-hereditary reasons
2. Lack of speech impairment
3. Inability to perform assessment or treatment tasks
4. Significant hearing loss that would impact on speech performance
5. Unable to participate in Skype conversations

Date of first enrolment
01/07/2017

Date of final enrolment
31/10/2018

Locations

Countries of recruitment
England

Switzerland

United Kingdom

Study participating centre
Ataxia UK
12 Broadbent Close
London
United Kingdom
N6 5JW

Study participating centre
ACHAF
La Chenaletta
St.Aubin
Switzerland
1566

Study participating centre
Sheffield Teaching Hospitals NHS Foundation Trust
Northern General Hospital
Herries Road
Sheffield
United Kingdom
S5 7AU

Sponsor information

Organisation
Strathclyde University, Research and Knowledge Exchange Services

Sponsor details
50 George St
Glasgow
Scotland
United Kingdom
G1 1QE

Sponsor type
University/education

Website
https://www.strath.ac.uk/

ROR
https://ror.org/00n3w3b69

Funder(s)

Funder type
Charity

Funder Name
Ataxia UK

Alternative Name(s)
Ataxia

Funding Body Type
Private sector organisation

Funding Body Subtype
Other non-profit organizations

Location
United Kingdom

Funder Name
Association Suisse de l’ Ataxie de Friedreich (ACHAF) - Swiss Association of Friedreich's Ataxia

Results and Publications

Publication and dissemination plan
We will publish our results in scientific journals targeted at speech and language therapists, other allied health professionals and neurologists. We will also disseminate the findings to people with ataxia through ataxia charities.

2019 results presented at the International Ataxia Research Conference (IARC) 2019
2020 Poster session presented at Motor Disorders Society Virtual Congress 2020 (added 03/09/2020)
2020 Poster session presented at Twentieth Biennial Conference on Motor Speech: Motor Speech Disorders and Speech Motor Control, United States (added 03/09/2020)

Intention to publish date

01/08/2019

Individual participant data (IPD) sharing plan

Anonymised speech data and participant demographics will be made available indefinitely in an open access database based at Strathclyde University for those participants who consented to this once all data have been collected and edited. The chief investigator, Prof Lowit, can be contacted to gain access to these data for academic purposes once the database has been made available.
Anonymised copies of the questionnaires and speech data analysis results will furthermore be stored in the Strathclyde University data repository and can be shared on request to the chief investigator.

IPD sharing plan summary

Stored in repository

Study outputs

Output type	Details	Date created	Date added	Peer reviewed?	Patient-facing?
Results article	results	01/10/2020	03/09/2020	Yes	No
HRA research summary			26/07/2023	No	No