

Study Protocol:

Development and trial of a physical activity program for Aboriginal families with Machado Joseph Disease (MJD) living in the Top End of Australia

Principal Investigator: Associate Professor Ruth Barker

2nd Investigator: Associate Professor Anne Lowell, Charles Darwin University

3rd Investigator: Professor Alan Clough, James Cook University

4th Investigator: Gayangwa Lalara, MJD Foundation

5th Investigator: Joyce Lalara, MJD Foundation

6th Investigator: Bronwyn Daniels, MJD Foundation

7th Investigator: Libby Massey, MJD Foundation

8th Investigator: Nick Kenny, MJD Foundation

Student Investigator (PhD Candidate): Jennifer Carr, James Cook University

Version 1.2

Date: 01/11/2018

Contents

| | |
|----------------------------------------------------------------------------------------------------------------------------------------------------------------|-----------|
| STUDY PROTOCOL: | 1 |
| DEVELOPMENT AND TRIAL OF A PHYSICAL ACTIVITY PROGRAM FOR ABORIGINAL FAMILIES WITH MACHADO JOSEPH DISEASE (MJD) LIVING IN THE TOP END OF AUSTRALIA | 1 |
| SUMMARY | 4 |
| 2 BACKGROUND AND RATIONALE | 5 |
| 3 AIM | 6 |
| 4 RESEARCH QUESTIONS | 6 |
| 5 STUDY SETTING | 7 |
| 6 STUDY DESIGN | 7 |
| 7 PERMISSIONS AND INVITATIONS | 8 |
| 8 PHASE 1: PROGRAM DEVELOPMENT – EXPERIENCED BASED CO-DESIGN | 9 |
| 8.1 METHODS | 9 |
| 8.1.1 <i>Purpose</i> | 9 |
| 8.1.2 <i>Participants</i> | 9 |
| 8.1.3 <i>Sample size and selection</i> | 10 |
| 8.1.4 <i>Preparation for Co-design Workshops</i> | 10 |
| 8.1.5 <i>Forming the MJD Expert Panel</i> | 10 |
| 8.1.6 <i>Workshops</i> | 11 |
| 8.1.7 <i>Between Workshops</i> | 11 |
| 8.1.8 <i>Recording Workshops</i> | 12 |
| 8.1.9 <i>Team debriefing sessions</i> | 12 |
| 8.2 DATA HANDLING AND DATA ENTRY | 12 |
| 8.2.1 <i>File Names</i> | 12 |
| 8.2.2 <i>Data Handling</i> | 13 |
| 8.2.3 <i>Transcriptions and Translations</i> | 13 |
| 8.3 DATA ANALYSIS..... | 13 |
| 8.3.1 <i>Quantitative Data</i> | 13 |
| 8.3.2 <i>Qualitative Data</i> | 14 |
| 8.4 OUTCOMES AND DISSEMINATION OF FINDINGS | 14 |
| 9 PHASE 2: PROGRAM TRIAL - MIXED METHOD MULTIPLE-CASE STUDIES | 14 |
| 9.1 METHODS | 14 |
| 9.1.1 <i>Purpose</i> | 14 |
| 9.1.2 <i>Participants</i> | 14 |
| 9.1.3 <i>Sample size and selection</i> | 15 |
| 9.1.4 <i>Introduction and Consent</i> | 15 |
| 9.1.5 <i>Before Starting the Program</i> | 16 |
| 9.1.6 <i>Trialling the Program</i> | 17 |
| 9.1.7 <i>After the Trial</i> | 20 |
| 9.1.8 <i>Team debriefing sessions</i> | 21 |
| 10 DATA MANAGEMENT AND ANALYSIS | 21 |

| | | |
|-----------|------------------------------------------------------|-----------|
| 10.1 | DATA HANDLING AND DATA ENTRY | 21 |
| 10.1.1 | <i>File Names</i> | 21 |
| 10.1.2 | <i>Data Handling</i> | 21 |
| 10.1.3 | <i>Transcription and Translation</i> | 22 |
| 10.2 | DATA ANALYSIS | 23 |
| 10.2.1 | <i>Quantitative data</i> | 23 |
| 10.2.2 | <i>Qualitative Data</i> | 23 |
| 10.3 | MODIFYING THE PROGRAM FROM CASE STUDY FINDINGS..... | 24 |
| 11 | ENSURING ETHICS AND QUALITY IN PRACTICE | 24 |
| 11.1 | ETHICS..... | 24 |
| 11.2 | QUALITY ASSURANCE | 24 |
| 12 | TEAM ROLES..... | 25 |
| 13 | TIMELINES..... | 26 |
| 14 | REFERENCES | 28 |
| 15 | APPENDICES | 31 |

Summary

Aim

Develop (Phase 1) and trial (Phase 2) a meaningful, evidence-informed, feasible physical activity program for individuals and families with Machado Joseph Disease (MJD) in the Top End of Australia.

Significance

Worldwide, no research has been conducted to investigate the impact of physical activity on mobility for individuals with MJD, especially in the cultural and geographical context of Aboriginal communities in the Top End. MJD, is 100 times more prevalent in affected Aboriginal communities in the Top End than anywhere else in the world. James Cook University (JCU) researchers were invited by Aboriginal families with MJD from the Top End and the MJD Foundation (MJDF) to work together to reduce the impact of MJD on mobility. Hence this project has been designed to address a pressing need as expressed by individuals and families with MJD in the Top End of Australia.

Methodology

Grounded in Indigenous, constructivist and participatory methodologies, this study will develop a physical activity program that is derived from the voices of Anindilyakwa families with MJD and the best available western scientific literature. The study will be conducted in two phases that will overlap and inform each another.

Phase 1: Development of the physical activity program using Experience Based Co-Design (EBCD), with voices of families with MJD at the forefront.

Phase 2: Trial of the physical activity program using a mixed method multiple-case study design.

Expected Outcomes

- A program will be developed that will reduce the impact of MJD on ‘walking and moving around’ and keep families ‘stronger for longer’ and will be transferrable to other MJD affected Aboriginal communities.
- The research will build research capacity of community research partners (CRPs), and promote continued knowledge exchange between families with MJD, CRPs, and Non-Aboriginal researchers.
- Resources will be developed which will inform policy makers and health services to improve care for families with MJD in Australia.

2 Background and Rationale

Aboriginal families of Groote Eylandt, Ngukurr and related Aboriginal communities across the Top End of Australia have been severely affected by MJD (1). This rare hereditary neurodegenerative disease is also known as spinocerebellar ataxia 3 (SCA3) (2). Although found in almost every continent of the world, the prevalence of this neurodegenerative disease is estimated to be 100 times greater in affected Aboriginal communities of the Top End than anywhere else in the world (1).

A characteristic feature of MJD is ataxia leading to a progressive decline in mobility over 5 – 10 years. Most people become wheelchair bound or dependent upon others to complete activities of daily living within 10-15 years of symptoms emerging. With a mean life expectancy of 20 years from diagnosis, people suffer a high degree of disability burden for an extended period of time (3). This has a significant impact on the lifestyle of the individual with MJD, their families and their communities. Many individuals are forced to move to Darwin for care, away from their families, communities and homelands. Currently, there is no known cure available for MJD.

This research has been driven by Aboriginal families in the Top End who have experienced the devastating impact of MJD. In 2008, the MJDF was established on Groote Eylandt to help individuals and families living with MJD to ‘stay stronger for longer’. James Cook University (JCU) researchers were invited by Aboriginal families with MJD and the MJD Foundation (MJDF) to work together to reduce the impact of MJD on mobility. A PhD student was recommended by JCU researchers and selected and approved to undertake this current project by cultural leaders of families with MJD and MJDF executive. Hence this project has been designed to address a pressing need as expressed by individuals and families with MJD in the Top End of Australia.

Worldwide, there has been no reported research on the importance of mobility or the impact of physical activity on mobility for individuals with MJD. Research in Europe among older people with general functional decline found that mobility was considered pivotal to an individuals’ QOL and well-being as it provides the means for freedom, choice and independence (4). Studies conducted in Asia (5) and Europe (6) found walking and physical activity provided opportunities for social enjoyment, emotional support, and promoted skill development, a positive self-image and a sense of well-being. Within the specific cultural and geographical context of the Aboriginal communities of Groote Eylandt affected by MJD, the value placed on mobility has not been explored.

Accumulating evidence suggests that there are benefits from physical activity for individuals with a range of neurodegenerative conditions. The benefits include improvements in general health, disease specific benefits and disease-modifying effects (7). For example, people with Parkinson’s and Huntington’s Disease have demonstrated positive changes in gait, balance and function following regular exercise and physical activity (8, 9). For individuals with hereditary degenerative ataxias, recent evidence has demonstrated positive gains in mobility, balance, ataxia and participation in activities of daily living in response to interventions that involve walking, fitness training, balance activities, and functional tasks (10, 11). Hence, the benefits of meaningful physical activity on mobility and QOL for individuals and families with MJD in the Top End need to be investigated. To do so, a meaningful,

evidence-informed, feasible physical activity program needs to be developed, derived from the perspectives of families with MJD combined with the available western scientific literature. To ensure the program is meaningful and feasible, such a program needs to be tested and evaluated by families with MJD in the Top End.

The purpose of this current study is to develop and trial a physical activity program for individuals with MJD from the Top End of Australia. To that end, it is important to acknowledge that programs that target physical activity for Indigenous people risk failure if they do not take into account Indigenous views and concepts of physical activity and lifestyle, with respect to cultural and traditional practices (12). Hence, a meaningful, evidence-informed and feasible, physical activity program, derived from the perspectives of families with MJD as well as the available western scientific literature, is to be developed and trialled.

In preparation for this current study, two earlier studies have been undertaken. The first explored, from the perspective of families with MJD, ‘what is important’ and ‘what works best to keep people with MJD ‘walking and moving around’ (HREC 2016-2672). Participants emphasised the importance of staying strong on the outside (physically), and the inside (mentally, emotionally, spiritually) by ‘exercising your body’, ‘keeping yourself happy’, and ‘having something important to do’, ‘going on country’, ‘using traditional medicine’ and ‘families helping each other’. The second study, which is underway, is a review of the best available evidence from the western scientific literature to identify ways to keep people with hereditary neurodegenerative diseases ‘walking and moving about’.

This two–phase study, which is the subject of this application, will firstly involve development of the physical activity program (Phase 1) by combining the perspectives of families with MJD and western scientific evidence and secondly, trialling of the newly developed program to determine if it is a feasible physical activity program for families with MJD (Phase 2).

3 Aim

- Develop (Phase 1) and trial (Phase 2) a meaningful, evidence-informed and feasible, physical activity program for individuals and families with MJD in the Top End of Australia.

4 Research Questions

- What are the key principles/guidelines for a meaningful, evidence-informed and feasible physical activity program for families with MJD living in the Top End?
- Is the newly developed physical activity program feasible for individuals and families from Groote Eylandt and Ngukurr?

5 Study Setting

Study Sites – Phase 1 and Phase 2

- Groote Eylandt Archipelago (Angurugu, Umbakumba and Milyakburra)
- Ngukurr
- Darwin (individuals from Groote Eylandt or Ngukurr who have relocated to Darwin)

The Aboriginal people who occupy the Groote Archipelago are an amalgamation of two cultures, the Anindilyakwa, who have occupied Groote Eylandt for around eight thousand years, and the Nunggubuyu from the mainland (13). Anindilyakwa is the main language spoken on Groote Eylandt and people are referred to as the Anindilyakwa people (13). The Anindilyakwa are made up of 14 clans who embrace their salt-water culture and ceremonial traditions (13). Although there has been rapid cultural change due to Non-Aboriginal influence since the mid 1900s, the Anindilyakwa people continue to uphold their strong cultural and traditional practices as an integral part of everyday life today (14). On the mainland close by, the Aboriginal people who have occupied Ngukurr in the Roper Gulf region for 40,000 years are seven clans living together and are referred to as the Yugul Mangi (15). The people of Ngukurr speak Kriol, English and other Aboriginal languages (15). Aboriginal families in this region have been severely affected by MJD. The MJDF supports families with MJD to improve QOL for Aboriginal Australians and their families living with MJD (16). This research has been approved by the Anindilyakwa Land Council (ALC) (peak representative body for Traditional Owners of the Groote Archipelago) and the Yugul Mangi Development Aboriginal Corporation (YMDAC) (main community reference group that represents the seven clans of South East Arnhem land).

6 Study design

Through a pragmatic approach, grounded in Indigenous and Participatory methodologies, a physical activity program will be developed and trialled by families living with MJD in the Top End. Anindilyakwa CRPs and families with MJD will guide and inform each stage of the research. The program will be derived from the voices of families with MJD and the best available western scientific evidence. This study will be described in two phases.

Phase 1: Development of a Physical Activity Program – Experience-Based Co-Design

The research team will combine views of families with MJD with western scientific evidence (see in Section 2). Background and Rationale) to identify key program principles. An Experience-based Co-design (EBCD) approach will be used to then refine the principles towards development of program guidelines, with voices of families with MJD at the forefront as experts with MJD (17). A series of workshops with an ‘MJD Expert Panel’ will be conducted to develop the program guidelines until agreement is reached by the panel.

EBCD is a participatory research approach that involves active collaboration of key stakeholders (i.e. experts, consumers, carers and clinicians) to inform the design of health care programs or interventions (17, 18). The co-design process will provide a method for the Student Investigator (SI) and CRPs to work collaboratively and flexibly with families with MJD and MJDF service providers to inform the program from planning to evaluation (17, 18). The process will ensure strong program principles and a strong framework for the program, and provide a basis for decision making and further guidance (17).

Phase 2: Trial of the Physical Activity Program - Mixed Method Multiple-Case Studies

Using a mixed method multiple-case study design, individuals with MJD will trial the program and feasibility, from the perspective of the participant, will be explored (19, 20). Within the qualitative component, in-depth interviews will be conducted to explore feasibility of the program from the participant perspective. Within the quantitative component, program feasibility (attendance, adherence, decline/did not complete) and reliability and validity of standardised measures of mobility and QOL will be explored (21). The case studies will provide information to modify and further develop the program in line with priorities of families with MJD.

A multiple-case study approach will provide a flexible and holistic investigation of participant's interactions with the newly developed program (22). Case studies will allow families with MJD to test what they have developed in collaboration with the SI, CRPs and MJDF service providers. Testing the program after development will ensure relevance of the program and will continue to acknowledge families as 'experts by experience' with MJD (17, 18). Mixed methods is supported in Indigenous research to inform useful and relevant intervention design and implementation (23).

7 Permissions and Invitations

- Permission to conduct this project has been granted by
 - Anindilyakwa Land Council (ALC) (*See Appendix A – Letter of Support: ALC*).
 - Yugul Mangi Development Aboriginal Corporation (YMDAC) (*See Appendix B - Letter of Support: YMDAC*).
 - MJDF Research Committee and MJDF on behalf of individuals and families with MJD living on Groote Eylandt (*See Appendix C – Letter of Support: MJDF*).
- After ethics approval from Menzies School of Health Research Ethics Committee (HREC) is gained, ethics approval will be sought from JCU HREC.

8 Phase 1: Program Development – Experienced Based Co-Design

8.1 Methods

8.1.1 Purpose

- Guidelines for a meaningful, evidence-informed and feasible physical activity program for individuals and families with MJD will be developed using an Experience-based Co-design (EBCD) approach. Use of this approach will allow collaboration of individuals with knowledge, experience and/or expertise regarding MJD in the Top End of Australia in the process of program guideline development.

8.1.2 Participants

- Participants ('MJD Expert Panel' members) will review program principles that have been developed and work towards development of the physical activity program guidelines.
- Individuals invited to trial the program will have the opportunity to communicate either verbally in their preferred language, using Aboriginal sign language or by using augmentative communication strategies, throughout the research and their participation in the program.
- The 'MJD Expert Panel' will be made up of three expert groups (see Inclusion Criteria):

Inclusion criteria

- Individuals with MJD and family representatives without MJD from Groote Eylandt or Ngukurr (**Group 1**)
- MJDF service providers employed by MJDF (**Group 2**)
- Health professional or researcher with expertise in MJD across the Top End and/or expertise in MJD and physical activity (**Group 3**).
- Able to provide consent
- Aged 18 years and over

Exclusion criteria

- Individuals with moderate to severe cognitive, intellectual or mental disability.

8.1.3 Sample size and selection

The MJD expert panel will consist of approximately 10 people, based on the number of people need to gain consensus as reported in previous studies (24). Panel members will be required to participate in approximately 3 workshops to collaborate on the development of the program (17).

The expert panel will be purposively sampled as follows:

- **Group 1** will be selected by the CRP and Cultural Advisor
- **Group 2** will be selected by the SI, CRP and Cultural Advisor
- **Group 3** and 4 will be recommended by the SI and confirmed by the CRP and Cultural Advisor

8.1.4 Preparation for Co-design Workshops

- The research team (SI, PI, CRPs) will assemble the guiding program principles for the program which will be compiled into a book or electronic resource for discussion at the workshops.
- Program principles and corresponding statements will be derived from findings of earlier studies on 'what matters most' and what 'works best' from the: 1) perspectives of individuals and families with MJD and 2) review of the best available western scientific evidence (see in Section 2. Background and Rationale).
- Statements will be written in controlled English (free from culturally specific metaphors, using terms, narrative and sentence structure that can be easily translated) (25) to ensure all participants can engage appropriately in the workshops.

8.1.5 Forming the MJD Expert Panel

The MJD Expert Panel will be formed in preparation for the consensus rounds. Suitable panel members will be identified collaboratively by the research team and the Cultural Advisor of MJDF. Panel members will be purposively selected to ensure a wide variation of opinions and views and a high level of expertise in MJD, physical activity and families with MJD living in the Top End.

- Panel members will be invited to participate by the research team either:
 - In-person
 - Via email
 - Via telephone.
- The manner in which they wish to participate in the consensus rounds will be identified at this time
 - i.e. in-person, via telephone or videoconference

Consent Process

Upon ethics approval, the SI will visit Groote Eylandt and Ngukurr to collaborate with CRPs and MJDF staff to seek potential panel members and gain informed consent.

Potential panel members will be:

- Provided with an information sheet and consent form (*See Study Protocol Appendices D and E*)
 - Forms will be read and explained by CRPs in Anindilyakwa or Kriol as required.
 - At a convenient location for the participant
- Asked to sign the consent form (*See Appendix E – Phase 1: Consent Form*). Alternatively:
 - Oral consent can be provided and audio recorded
 - Electronic certified signatures can be used
- Paid for their time and expertise by the Project Grant (i.e. individuals and families with MJD) or by their employing organisations (i.e. MJDF, JCU, Top End Health) and explained in the information sheet.
- Entered into the Recruitment Log and Enrolment Form (*See Appendices F and G*)

8.1.6 Workshops

- After recruitment of the Expert Panel, a series of workshops will take place to clarify and refine the program principles and develop the physical activity program. MJD expert panel members will collaborate on the program principles, parameters, procedures and logistics. Workshops will be flexible to suit the needs of panel members. Each panel member will participate in workshop discussions via their preferred method of engagement (i.e. face to face, phone, skype/zoom).
- Program principles will be displayed in either an A3 size worksheets or provided as an electronic resource when required.
- CRPs with the SI will facilitate the workshops, present and discuss each statement to the panel members in their preferred language, using their preferred method of communication.
- Discussions will be audio recorded with the participant's consent and translated/transcribed verbatim by CRPs and SI.
- Responses will be collated, amendments to the program will be made accordingly, and will be provided again to the panel for review until agreement is reached.

8.1.7 Between Workshops

After each workshop, notes and recordings from panel member discussions will be collated and analysed and the program will be further developed or amended accordingly. Changes will be fed back to the panel at the next workshop and reviewed again for further development until agreement is reached. The workshops will be repeated as many times as required until agreement is reached. This procedure is in line with the Australia's NHMRC Road Map Action Areas with regards to capacity exchange and improving the participation of Aboriginal and Torres Strait Islander people in research (20).

8.1.8 Recording Workshops

All responses will be audio recorded and transcribed.

Notes will be taken by the SI.

Information recorded that will also be described in the data analysis will include:

- Number of workshops
- Panel member participants in each workshop
- Panel member drop outs
- New principles generated from comments and suggestions of panel members
- Principles removed by panel members
- Panel member input will be presented in table form and structured according to themes related to the physical activity program. A similar table will also be provided for removed statements

8.1.9 Team debriefing sessions

Debriefing sessions will occur between research team members after each workshop (*See Appendix H - Phase 1: Team Debriefing Minutes Form*). The purpose of the debriefing meetings is:

- To provide a daily/weekly record of proceedings.
- For the SI to provide a summary of the results of each round to the research team in table form, discuss the findings, and discuss what principles will be removed, included or modified.
- For the research team to:
 - Update each other on progress with workshops
 - Discuss how findings will feed into program design
 - Discuss any problems/changes with the program principles/guidelines

8.2 Data handling and data entry

8.2.1 File Names

Panel members may choose how they wish to be identified in this phase. If the participant does not have a preference, a unique identification number will be given. The identifier will be used on all study documentation. The format for naming files will follow Table 1 – Labelling Files, e.g. *ID1M_Consensus Round#1*.

Table 1 Labelling files

| <i>Participant Identifier</i> | <i>Gender of participant_</i> | <i>Type of data collection</i> |
|-----------------------------------|-------------------------------|----------------------------------------------------|
| [skin name, pseudonym, ID number] | M or F | Consensus Round#(insert number of consensus round) |

8.2.2 Data Handling

- All study data will be kept on the person of the SI at all times or in a locked vehicle, lockable room of the SI or SI password protected computer, backed up regularly and only shared within the research team.
- Panel members will not be allowed to view study data at any time and any discussions will not be revealed to anyone else.
- Names of panel members will not be used at any stage
- Audio recordings will not start until the consent has been given

8.2.3 Transcriptions and Translations

- Recordings of responses in the workshops will be interpreted by the CRP and transcribed into a word document by the SI in English.
- English interpretations by the CRP will be audio recorded to verify the written transcript.
- English transcriptions will be reviewed by CRPs by listening to recordings and cross-checking the transcription. The SI will make corrections and cross-checking will continue until a clear transcription/translation is reached, confirmed by CRPs.
- English transcriptions will be saved as new Word files, and labelled with the panel members ID followed by the language the transcript was translated into (i.e. English) e.g. 'ID1M_English' and exported into NVivo (QSR International, Cambridge, MA) qualitative data management software for coding and analysis.

8.3 Data Analysis

8.3.1 Quantitative Data

- Statistical analysis of consensus scoring will be conducted using Excel. Descriptive statistics will be used to describe: response rate, number of workshops, number of drop outs

8.3.2 Qualitative Data

- Thematic analysis with qualitative description will be used to analyse comments for all workshops with NVivo software (27).

8.4 Outcomes and Dissemination of Findings

- Outcomes of Phase 1 will be prepared as a resource to guide the trial of the program (Phase 2).
- Outcomes of Phase 1 and implications for Phase 2 will be disseminated to individuals and families with MJD from Groote Eylandt and Ngukurr including those families who have relocated to Darwin. The research team will meet with individuals and their families either one on one or in a group setting according to their preference snacks and refreshment will be provided at these meetings.

9 Phase 2: Program Trial - Mixed Method Multiple-Case Studies

9.1 Methods

9.1.1 Purpose

Feasibility of the newly developed physical activity program, based on the experiences of families with MJD (19, 20), will be determined in Phase 2. A trial of the program following Phase 1 will action what families with MJD have developed and provide practical feedback in preparation for a larger trial to follow.

- A mixed method multiple-case study design will be used to explore program feasibility (19, 20).
- The program will be determined feasible if it is acceptable and useful from the perspective of the participants (consumers) using the program.
- Within the qualitative component, in-depth interviews will be conducted to explore feasibility of the program from participant experiences.
- Within the quantitative component, program feasibility will be measured through program attendance/adherence and appropriateness of outcome measures to quantify mobility and QOL (21).

9.1.2 Participants

Participants will be required to participate in

- Pre- and post-program measurement of function and QOL

- Physical activity program as per the newly developed program guidelines
- Post-program interviews

Individuals invited to trial the program will have the opportunity to communicate either verbally in their preferred language, using Aboriginal sign language or by using augmentative communication strategies, throughout the research and their participation in the program.

Inclusion criteria

- Clinical or genetic diagnosis of MJD
- Aged 18 years and over
- Identified as belonging to either the Groote Eylandt or Ngukurr families with MJD
- Physically able to participate in functional activities and a likely minimum score of 2 for question 1 of the Scale for the Assessment and Rating of Ataxia (SARA) [2 = Gait clearly abnormal, tandem walking > 10 steps not possible] (to be confirmed following completion of Phase 1)
- Able to provide consent

Exclusion criteria

- Individuals with an additional health condition that may impact on mobility e.g. a neurological condition such as stroke or spinal cord injury; a vascular condition e.g. amputation.
- Individuals with moderate to severe cognitive, intellectual or mental disability
- Deemed medically unable to participate by their doctor

9.1.3 Sample size and selection

- Four (4) individuals with MJD from either Groote Eylandt or Ngukurr will be purposively selected to participate in the program to achieve maximum variation in terms of age, functional level, gender and family role.
- Recruitment will continue until four participants complete the program for the full duration (or four case studies are completed) in line with similar studies (28, 29).

9.1.4 Introduction and Consent

- The research team (PI, SI, CRP) will collaborate with and MJD Family Leaders to identify potential participants for the trial.
- Participants will be
 - Provided with an information sheet and consent form (See Appendices I and J)

- Forms will be read and explained by CRPs in Anindilyakwa or Kriol
- At a convenient location for the participant (MJD office, residence).
- Asked to sign the consent form. Alternatively,
 - Oral consent can be provided and audio recorded
- Asked to confirm their age, age of onset of MJD, the story of their health, their family and who lives at home with them, whether they are working or not, and information about their level of physical ability and physical activity throughout each week.
- Entered into the Recruitment Log and Enrolment Form (See Appendices F and G)

9.1.5 Before Starting the Program

- A comprehensive assessment will be conducted with each individual with MJD (accompanied by any of their nominated family members or caregivers) in accordance with the framework of the World Health Organisation International Classification of Functioning, Disability and Health (ICF) by CRPs and SI. The assessment will include:
 - Identification of individuals' specific goals and priorities for walking and moving around
 - Identification of personal and environmental factors
 - Identification of impairments, activity limitations and participation restrictions
 - Identification of supportive factors to keep 'strong on the inside and on the outside' (see Figure 1), as identified in earlier studies (*see Section 2. Background and Rationale*)
 - Outcome measures, based on mobility, function, and QOL completed by an independent assessor of the individual's specific goals, priorities and aspirations (*see in Section 8.1.7 – Measurement*).
- A tailored program plan for each participant will be developed based on the participant goals and priorities, assessment findings and according to program guidelines (See Appendix M – Phase 2: Participant Program Plan). Copies of the completed assessment and program plan will be provided to the participant.
- The plan will be reviewed each week to establish what other activities need to be included/removed.
- Changes made will be recorded (See Appendix N – Phase 2: Participant Progress Notes).
- The physiotherapist facilitating program delivery (SI) will complete progress notes and report adverse events.



Figure 1 - What is important and work works best to keep families with MJD Strong on the inside and the outside to keep 'walking and moving around'

9.1.6 Trialling the Program

- The physical activity program will be delivered by the SI who is a registered and experienced physiotherapist in the area of neurological rehabilitation
- CRPs and Cultural Advisors will provide guidance throughout delivery of the program.

| | |
|------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Delivery | <ul style="list-style-type: none"> • Program delivered according to Participant Program Plan by SI and CRPs <ul style="list-style-type: none"> ○ Family members can be involved at the preference of the participant • 6 weeks, 3 times a week, distributed according to wishes of participant <ul style="list-style-type: none"> ○ To be confirmed as part of Phase 1 |
| Location | <ul style="list-style-type: none"> • Selected according to activities relevant to that participant. May include <ul style="list-style-type: none"> ○ MJDF Office ○ By the beach or the river ○ The bush ○ In the community |
| Equipment | <ul style="list-style-type: none"> • Provided by MJDF Exercise bikes, tricycles, walking machine, fishing gear, and various sporting equipment items, stationary. • Waterproof, wearable accelerometers (<i>See also Section 8.1.7 – Measurement</i>) • Snacks and refreshments provided |

| | |
|----------------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Transport | <ul style="list-style-type: none"> • Provided by MJDF according to standard practice <ul style="list-style-type: none"> ○ Four-wheel drive, wheelchair accessible vehicles, and modified vehicles for those with mobility difficulties available |
| Documentation | <ul style="list-style-type: none"> • Progress notes will be completed at the end of each day and will include <ul style="list-style-type: none"> ○ Subjective / objective information and observations ○ Tasks completed, duration and intensity ○ Location of activities ○ Incidents /adverse events |

Measurement

| | |
|-----------------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Demographic Data | <ul style="list-style-type: none"> • Documented on Participant Enrolment Form <i>(See Appendix L - Participant Enrolment Form)</i>. • Collected on enrolment by SI and CRP |
| Feasibility Measures | <ul style="list-style-type: none"> • Number of sessions completed • Number of participants who <ul style="list-style-type: none"> ○ Decline participation/recruitment ○ Commence but do not complete program ○ Complete the program ○ Would wish to continue to the program • Reasons for drop out • Complications • Semi structured in-depth interviews to gain perceptions of individuals with MJD on the program |
| Outcome Measures | <ul style="list-style-type: none"> • Waterproof, wearable accelerometers will likely be used to measure physical activity (30). • Standardised, valid and reliable outcome measures for mobility and QOL for individuals with MJD/SCA to quantify changes in mobility perceived as ‘important’. <ul style="list-style-type: none"> ○ To be confirmed as part of Phase 1 ○ Details will be provided to the ethics committee once Phase 1 has been completed • Independent assessor - Physiotherapist <ul style="list-style-type: none"> ○ Baseline: Upon recruitment of the participant onto the program ○ Pre-Program: Four weeks after baseline on commencement of the program ○ During program: Fortnightly during the 6-week program period ○ Post program: On completion of the program (within one week) ○ Follow up: Four weeks post completion of the program |

Safety Considerations

- The physical activity program will be delivered by the SI who is a registered and experienced physiotherapist in the area of neurological rehabilitation.
- Investigators will act within existing MJDF policies and procedures in relation to MJD Clients, Travel, Vehicles, Workplace Health and Safety, and general Professional Conduct.
- Investigators will adhere to JCU policies on responsible conduct of research, the National Statement on Ethical Conduct in Human Research, NHMRC Road Map II and the NHMRC/ARC/UA Australian Code for the Responsible Conduct of Research (2007).
- Participants will be able to stop participating at any time without giving a reason.
- Investigators will act within MJDF policies and procedures in relation to MJD Clients, Travel, Vehicles, Workplace Health and Safety, and general Professional Conduct.
- Below is an outline of potential risks and risk mitigation strategies relevant to this study.

| Potential Risk | Management Strategy |
|------------------------------------------------------------------------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Failing to respect cultural and ethical principles in Aboriginal communities | <ul style="list-style-type: none"> ▪ Work under the guidance of CRPs and MJD family leaders ▪ Adhere to conditions of ethical approval ▪ Secure land and work permits prior to visiting each community and liaise with CRPs regarding cultural protocols. ▪ Respect community protocols e.g. reschedule visits if sorry business arises or follow community protocols if sorry business occurs during field work. |
| Failure to engage participants | <ul style="list-style-type: none"> • Ensure MJD family leaders and CRPs take a lead role in selection of MJD Expert Panel members • Provide early invitations and regular engagement with panel members |
| Risks associated with physical activity | <ul style="list-style-type: none"> • Participants in the physical activity program will require medical clearance to participate |
| Risks associated with remote work | <ul style="list-style-type: none"> • Complete risk assessments for field work • Log field trips using Riskware • Work within JCU and MJDF policies and procedures for remote community work • Complete work permits annually for approval to visit and work on Groote Eylandt and in Ngukurr • When going on field trips, provide trip details and contact details to at least two key MJDF staff situated at MJDF offices. • Inform local MJDF staff if going 'on country' with families with MJD and advise of planned leave and return times • Take the MJDF satellite phone on all trips 'on country' where there is limited phone reception |

9.1.7 After the Trial

Interviews

- Will be led by CRPs in collaboration with SI, in each participant's preferred language, in a culturally safe way.
 - Time and location will be organised to suit the participant, to ensure participants are comfortable to share their experiences.
 - Transport and light refreshments will be provided.
 - Audio recording equipment will be available.
- Prior to interview commencement, confidentiality and anonymity will be re-explained and each participant will be asked if he/she consents to be interviewed and audio recorded.
- Audio recording will allow note taking and transcriptions to occur after completion of the interview. Field notes will record relevant participant responses, non-verbal behaviour, and the setting and atmosphere of the interview.
- The topic guide will be followed by CRPs and SI but the pace will be set by the participant to tell their own story about the program (*See Appendix O – Phase 2: Post-Trial Interview Topic Guide*).
- Feedback provided by the participant during the program will be collected from Participant Progress Notes, collated prior to the interviews, and provided to the participant for comment and verification.
- On completion, the participant will be thanked for their participation and reminded that their information will be kept confidential.
- Short summaries of fieldwork observations will also be translated from Anindilyakwa or Kriol into English by CRPs.
- Interviews that occurred in Anindilyakwa or Kriol will be interpreted and transcribed by CRPs, in collaboration with SI, into English.
- All transcriptions and field notes will be typed into Word in English and uploaded into NVivo for analysis at the end of each day or the next day.

Interviewer debriefing sessions

- SI and CRP interviewer will meet to debrief and discuss findings after each interview
- The contact summary form will be completed and typed into a computer file, labelled with the unique participant identifier, stored with the audio file (*See Appendix P – Interview Contact Summary Form*).
- The SI will also record field notes in a journal throughout the data collection process.

9.1.8 Team debriefing sessions

Weekly debriefing sessions will occur between the research team. Progress Notes will be used for reference and the Debrief Meeting Form will be completed (*See Appendix Q – Investigator Debrief Meeting Form*). The aims of the meetings are:

- For all investigators and independent assessors to update each other on progress with data collection
- For investigators to discuss key findings from data collection, including differences and similarities
- For investigators to discuss how preliminary findings will feed into the design of the program
- For investigators to discuss problems/changes with the program
- To provide a record of proceedings during the trial of the program.

10 Data Management and Analysis

10.1 Data handling and data entry

10.1.1 File Names

Participants may choose how they wish to be identified in this phase. If the participant does not have a preference, a unique identification number will be given. The identifier will be entered on any notes, audio files or transcript documents. The format for naming will follow Table 2 - Labelling of files. E.g. ID1M_Trial_Progress Notes

Table 2 Labelling files

| <i>Participant Identifier</i> | <i>Gender of participant_</i> | <i>Study Phase_</i> | <i>Type of data collection</i> |
|--------------------------------|-------------------------------|---------------------|--------------------------------------------------------|
| [skin name, pseudonym, number] | M or F | Trial | Post-Trial Interview Progress Notes Program Plan |

10.1.2 Data Handling

Study data will be kept on the person of the SI at all times or in a locked vehicle, lockable room of the SI or password protected computer. Participants will not be allowed to view notes at any time. Discussions relating to the program trial will not be revealed to anyone else.

- Names of participants will not be used at any stage
- Any audio recordings will not start until the consent has been given

- First names may be used during post-program interviews to refer to participants. However, notes taken will use participant identifiers described above and recorded/written records will only display participant identifiers, not individual names.
- Field notes will be in English unless discussions had been held in other languages, in which case translation will occur as detailed for the interviews.

10.1.3 Transcription and Translation

- Translation will take a meaning-based approach from the original language into English. The translator will convey the meaning of the source language within the natural grammar of the target language – English. Clarifications will be made in brackets, to capture and interpret for the reader meaningful elements of the source material, and the way the elements combine to form meaning of the text as a whole. Clarifications will repackage the original narratives and utterances into words, grammar and idioms specific to the target language (English) (31).
- Quality criteria for the translations will be comprehensibility (especially relating to culture-specific concepts), appropriateness (in content and approach) and accuracy (faithful to the source text and key facts) (31). For difficult terms, phrases and corrections, the CRPs and SI will sit together and discuss, such that final and most appropriate translations are agreed upon.
- Transcription files will be labelled with the interview ID followed by the language that the transcript was translated into (English) e.g. 'IDIM01 English' and exported into NVivo (QSR International, Cambridge, MA) for analysis. A standardised layout will be applied to transcripts to facilitate the comparison of data at the analysis stage (32).
- Transcription will reflect a descriptive qualitative approach, describing experiences and perspectives of participants. Hesitations, pauses, utterances, cross-talking and incomplete sentences will be recorded. Major interruptions by other people or telephones will be recorded to contextualise any breaks in speech or repetitions. Minor interruptions will not be recorded to ensure the flow of the transcript supports interpretation and analysis (32).
- At the top of the transcript should be a table detailing the unique participant identifier, interviewer and note-taker names, date of the interview, language of the interview, location, date transcript completed and recording details. See the example below:

| | |
|-----------------------------------------|-------------------|
| Participant Identifier | _ _ _ _ _ _ _ _ _ |
| Interviewer name: | |
| Note taker name: | |
| Date of Interview (dd/mm/yyyy) | |
| Language of Interview: | |
| Location of Interview: | |
| Date completed transcript: (dd/mm/yyyy) | |
| Start time of recording | |
| End time of recording | |
| Length of recording | |

Cross-checking

Each transcript will be reviewed by the CRPs by listening to or watching sections of the recordings and cross-checking the transcription. If errors are identified, the SI will make corrections and cross-checking will continue to occur until a clear transcription/translation is reached as confirmed by CRPs.

10.2 Data analysis

10.2.1 Quantitative data

SPSS and Simulation Modelling Analysis (SMA) software will be used (33).

- Demographics and program data will be analysed using descriptive statistics in SPSS.
- SMA, which is used to analyse small sets of single subject data over time, will be used to reflect the rate of change in outcome measure scores for each participant (28, 34). The mean change in scores between the baseline and trial phase will be identified (28, 33).

10.2.2 Qualitative Data

- A descriptive content analysis will be undertaken with assistance from NVIVO software.
- Interview data will be analysed collaboratively between CRPs and SI, to draw out participant perceptions of the program, positives and negatives of the program and recommendations for optimisation.

- Coding will be conducted by reading the data line by line, identifying underlying meaning or concepts behind statements (35). Each line, or few lines, will be labelled according to the idea(s) in the transcript, using a short title, and used to create a new tree node in NVivo. When the idea appears again, it will be coded to the same node. Nodes will be grouped according to the question asked in relation to the program. As themes emerge, nodes will be arranged in groups under a parent node labelled as the theme.
- Oral feedback and verification sessions will also occur between CRPs and SI to share, confirm and discuss emerging findings (36).
- A narrative will be developed bridging the original research concerns with the participants' subjective experiences and ideas about the program related back to the study aims and research questions.
- The findings will be presented to participants for verification, and to optimise the program.

10.3 Modifying the Program from Case Study Findings

- The program will be modified and strengthened after the program trial is complete in accordance with feedback and findings gathered from participants.
- Participants who trialled the program will then be approached by CRPs and SI to verify the findings, modify the program accordingly and disseminate the final findings.
- The final program will be put together into a book for participants and communities to keep. The findings of this study will be published in an international journal

11 Ensuring Ethics and Quality in Practice

11.1 Ethics

All investigators have been briefed and provided resources on the National Statement on Ethical Conduct in Human Research and will place these priorities of protecting study participants, anticipating harms, avoiding undue intrusion, rights to confidentiality and anonymity, intellectual property rights and involvement in the study ahead of other requirements, whether for their own gain or that of the project. Non-Aboriginal researchers will be principally familiar with the National Statement on Ethical Conduct in Human Research, particularly in regard to research with Aboriginal and Torres Strait Islander Peoples (Chapter 4.7), the Australian Code for Responsible Conduct of Research, NHMRC Guidelines for Ethical Conduct in Aboriginal and Torres Strait Islander Health Research and the Commonwealth Privacy Act (Section 95 and 95A).

11.2 Quality assurance

The protocol will be used by the research team to guide conduct of the study and will provide a method to assess quality assurance. At least one assessment of quality assurance will be conducted during the data collection phase by a moderator who will be external to the field team, preferably a collaborator

from a similar study from JCU. Assessors will use the protocol as guidance for the evaluation. If issues or concerns arise as a result of this, the moderator will take action as appropriate.

12 Team Roles

Student Investigator:

- Work under the guidance of CRPs, families with MJD and all other investigators
- Collaborate with all investigators for study planning
- Assemble expert panel
- Consult with families with MJD
- Coordinate consensus rounds and case studies
- Responsible for data collection, analysis, dissemination of results, and write up

Principal Investigator:

- Responsible for designing and supervising the study, responsible for monitoring progress of data collection, data analysis and write up.

2nd and 3rd Investigator:

- Provide guidance on study design and data collection, supervise the study, monitor progress of data collection, data analysis and write up.

4th, 5th, 6th Investigators – Aboriginal Community Research Partners

- Provide cultural guidance to investigators and oversee study planning and study conduct
- Collaborate with all investigators to assemble MJD expert panel and develop program principles
- Lead consultation and communication with families with MJD
- Monitor conduct of case studies
- Collaborate with SI to translate all discussions and interviews with participants in Anindilyakwa/Kriol and translate the recordings into English for transcription.
- Collaborate with SI throughout data collection, data analysis, write up and dissemination of results

7th, 8th Investigators and MJDF Staff

- Assist with study planning and preparation on Groote Eylandt and in Ngukurr
- Provide ongoing communication to families with MJD regarding preparation and commencement of the study
- Collaborate with CRPs, SI and PI to provide logistic support required for coordination and conduct of the study

13 Timelines

| Dates | Study Phases | Activities | Who |
|------------|---------------------------------------------------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|---------------------------------------------------------------------------------------------------------------|
| Completed | Planning | <ul style="list-style-type: none"> • Draft interview and consent forms • Gain approvals from Anindilyakwa Land Council (ALC) to conduct the proposed research • Draft interview and consent forms • Develop protocol • Collaborate and discuss study to be conducted to families with MJD, CRPs and MJD staff • Engage with community and families with MJD • Engage with MJDF staff regarding study protocol, study planning and data collection | <ul style="list-style-type: none"> • All investigators MJDFS, CRPs, families with MJD |
| April 2018 | Scoping, preparation and community engagement completed | <ul style="list-style-type: none"> • Two studies completed that have been conducted in earlier phases of this study to inform the development of the program. <ul style="list-style-type: none"> ○ Study 1: Lived experiences of families with MJD living in the Top End of Australia: what matters most and what works best to keep 'walking and moving around'. ○ Study 2: Interventions that promote 'walking and moving around' for individuals with SCA: A scoping review) • Engage with community and families with MJD about study commencement • Collaborate with CRPs employed by MJDF for further study planning • Identify suitable dates for the study to commence • Ethics approval gained <p><i>Please note, through previous studies conducted, all investigators are well known to families with MJD on Groote Eylandt and Ngukurr and extensive community engagement has taken place between 2016 and 2018. MJDF staff involved this research have been known to families with MJD since 2008.</i></p> | <ul style="list-style-type: none"> • All investigators MJDFS, CRPs, families with MJD |
| May 2018 | Guidelines for program developed | <ul style="list-style-type: none"> • Guiding principles developed • Meetings conducted with all investigators to refine the program and identify MJD expert panel. • Principles prepared for workshops for collaboration and comment by MJD expert panel. | <ul style="list-style-type: none"> • All investigators MJDFS, CRPs, MJD family representatives |

| Dates | Study Phases | Activities | Who |
|----------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| June 2018 | 'MJD Expert Panel' recruited | <ul style="list-style-type: none"> • Suitable panel identified by investigators • Meetings face to face, via telephone or zoom to invite panel members to participate • Information sheets and consent forms provided to panel members • Completed consent forms finalised | <ul style="list-style-type: none"> • All investigators MJDFS, CRPs, MJD family representatives • MJD expert panel |
| July 2018 | 1 st consensus round commences | <ul style="list-style-type: none"> • Program principles finalised • First consensus round distributed | <ul style="list-style-type: none"> • All investigators MJDFS, CRPs • MJD expert panel |
| October 2018 | <ul style="list-style-type: none"> • Consensus rounds completed • Meeting with investigators and dissemination of findings • Preparation for case studies | <ul style="list-style-type: none"> • Table produced which outlines accepted and removed statements • Program guidelines confirmed • Meetings in Darwin, Groote Eylandt and Ngukurr with all investigators, MJD family leaders and families with MJD to discuss study findings • MJD family leaders and MJDF staff identify participants to trial the program • Meetings face to face, via telephone or zoom to invite panel members to participate • Information sheets and consent forms provided to panel members • Completed consent forms finalised | <ul style="list-style-type: none"> • All investigators MJDFS, CRPs • MJD expert panel • Families with MJD on Groote Eylandt and Ngukurr |
| November 2018 | Case studies underway | <ul style="list-style-type: none"> • SI and CRPs facilitate participants to trial the program according to principles developed <ul style="list-style-type: none"> ○ Initial assessment completed ○ Measures commenced ○ Trial underway | <ul style="list-style-type: none"> • All investigators MJDFS, CRPs, MJD family representatives |
| February 2019 – April 2019 | Interviews with case study participants | <ul style="list-style-type: none"> • SI and CRPs will meet interview participants to gain feedback on the positives, negatives and areas for improvement • Translation and transcription of interviews • Optimise the program with feedback provided by participants during the program and from participant perspectives gained from the interviews. • Verify the findings with participants and MJD family leaders | <ul style="list-style-type: none"> • All investigators MJDFS, CRPs • Families with MJD on Groote Eylandt and Ngukurr |
| April 2019 – June 2019 | Program refined and study findings finalised | <ul style="list-style-type: none"> • Findings disseminated to all communities / panel members • Manuscript prepared for publication | <ul style="list-style-type: none"> • All investigators MJDFS, CRPs • Families with MJD on Groote Eylandt and Ngukurr • MJD expert panel |

14 References

1. Massey L, Jane A, Lindop N, Christian E. Disability Audit – NE Arnhem Land/NT Gulf – A Snapshot of Indigenous Australian Disability in the Very Remote Communities of the Groote Eylandt Archipelago (Angurugu, Umbakumba, Milyakburra), Elcho Island (Galiwin'ku), and Ngukurr (including Urapunga). *Australian Indigenous Health Bulletin*. 2013;13(4).
2. Martins S, Soong B-W, Wong VCN, Giunti P, Stevanin G, Ranum LPW, et al. Mutational origin of Machado-Joseph Disease in the Australian Aboriginal communities of Groote Eylandt and Yirrkala. *Archives of Neurology*. 2012;69(9):746-51.
3. Massey L, Jane A, Lindop N, Christian E. Disability Audit – NE Arnhem Land/NT Gulf – A Snapshot of Indigenous Australian Disability in the Very Remote Communities of the Groote Eylandt Archipelago (Angurugu, Umbakumba, Milyakburra), Elcho Island (Galiwin'ku), and Ngukurr (including Urapunga).". In: Foundation MJD, editor. 2013.
4. Bourret EM, Bernick LG, Cott CA, Kontos PC. The meaning of mobility for residents and staff in long-term care facilities. *J Adv Nurs*. 2002;37(4):338-45.
5. Jepson R, Harris FM, Bowes A, Robertson R, Avan G, Sheikh A. Physical activity in South Asians: an in-depth qualitative study to explore motivations and facilitators. *PLoS One*. 2012;7(10):e45333.
6. Ferrand C, Perrin C, Nasarre S. Motives for regular physical activity in women and men: a qualitative study in French adults with type 2 diabetes, belonging to a patients' association. *Health & Social Care in the Community*. 2008;16(5):511-20.
7. Speelman AD, van de Warrenburg BP, van Nimwegen M, Petzinger GM, Munneke M, Bloem BR. How might physical activity benefit patients with Parkinson disease? *Nat Rev Neurol*. 2011;7(9):528-34.
8. Khalil H, Quinn L, van Deursen R, Dawes H, Playle R, Rosser A, et al. What effect does a structured home-based exercise programme have on people with Huntington's disease? A randomized, controlled pilot study. *Clin Rehabil*. 2013;27(7):646-58.
9. Goodwin VA, Richards SH, Taylor RS, Taylor AH, Campbell JL. The effectiveness of exercise interventions for people with Parkinson's disease: A systematic review and meta-analysis. *Movement Disorders*. 2008;23(5):631-40.
10. Ilg W, Synofzik M, Brotz D, Burkard S, Giese MA, Schols L. Intensive coordinative training improves motor performance in degenerative cerebellar disease. *Nat Rev Neurol*. 2009;73(22):1823-30.
11. Milne SC, Corben LA, Georgiou-Karistianis N, Delatycki MB, Yiu EM. Rehabilitation for Individuals With Genetic Degenerative Ataxia: A Systematic Review. *Neurorehabil Neural Repair*. 2017;31(7):609-22.
12. Gray C, Macniven R, Thomson N. Review of physical activity among Indigenous people. *Australian Indigenous Health/InfoNet*. 2013;13:1-17.
13. Anindilyakwa Land Council. Culture 2015 [Available from: <http://anindilyakwa.com.au/culture>].
14. Remote Area Health Corps. East Arnhem Region Community Profile: Groote Eylandt. Australian Government; 2009.
15. Department of Local Government and Community Services. Ngukurr 2012 [Available from: http://www.dlgcs.nt.gov.au/about_us/remote_service_delivery/major_remote_towns/ngukurr/profile].

16. MJD Foundation. MJD Foundation: quality of life for MJD sufferers and their families. Alyangula, Northern Territory: MJD Foundation; 2009.
17. Donetto S, Pierri P, Tsianakas V, Robert G. Experience-based Co-design and Healthcare Improvement: Realizing Participatory Design in the Public Sector. *The Design Journal*. 2015;18(2):227-48.
18. Dawda P, Knight A. Experience Based Co-design: A toolkit for Australia: Australian Healthcare and Hospitals Association and Consumers Health Forum of Australia; 2017 [cited 2018 6 October]. Available from: <http://ahha.asn.au/experience-based-co-design-toolkit>.
19. Yin RK. Applications of case study research: Sage; 2011.
20. Powell BJ, Proctor EK, Glisson CA, Kohl PL, Raghavan R, Brownson RC, et al. A mixed methods multiple case study of implementation as usual in children's social service organizations: study protocol. *Implementation Science*. 2013;8(1):92.
21. Thabane L, Ma J, Chu R, Cheng J, Ismaila A, Rios LP, et al. A tutorial on pilot studies: the what, why and how. *BMC Med Res Methodol*. 2010;10:1.
22. Yin RK. Case study research: Design and methods: Sage publications; 2013.
23. Dawson A, Toombs E, Mushquash C. Indigenous Research Methods: A Systematic Review. *The International Indigenous Policy Journal*. 2017;8(2).
24. Akins RB, Tolson H, Cole BR. Stability of response characteristics of a Delphi panel: application of bootstrap data expansion. *BMC Medical Research Methodology*. 2005;5(1):37.
25. Eades D. Aboriginal ways of using English: Aboriginal Studies Press; 2013.
26. Hasson F, Keeney S, McKenna H. Research guidelines for the Delphi survey technique. *J Adv Nurs*. 2000;32(4):1008-15.
27. Sandelowski M. Whatever happened to qualitative description? *Res Nurs Health*. 2000;23(4):334-40.
28. Harrison AM, McCracken LM, Jones K, Norton S, Moss-Morris R. Using mixed methods case-series evaluation in the development of a guided self-management hybrid CBT and ACT intervention for multiple sclerosis pain. *Disability and Rehabilitation*. 2017;39(18):1785-98.
29. Abu-Zidan FM, Abbas AK, Hefny AF. Clinical "case series": a concept analysis. *Afr Health Sci*. 2012;12(4):557-62.
30. Darvall JN, Parker A, Story DA. Feasibility and acceptability of remotely monitored pedometer-guided physical activity. *Anaesth Intensive Care*. 2016;44(4):501-6.
31. Downing B, Bogoslaw L. Effective patient-provider communication across language barriers: a focus on methods of translation. The Robert Wood Johnson Foundation - *Hablamos Juntos*; 2003.
32. McLellan-Lemal E. Transcribing data for team-based research. In: Guest G, MacQueen K, editors. *Handbook for Team-Based Qualitative Research*. Plymouth, UK: Altamira Press; 2008.
33. Borckardt JJ, Nash MR. Simulation modelling analysis for small sets of single-subject data collected over time. *Neuropsychological rehabilitation*. 2014;24(3-4):492-506.
34. Yin RK. Analysing Case Study Evidence. *Case Study Research: Design and Methods*. Thousand oaks: SAGE publications; 2009. p. 14.

35. Strauss A, Corbin J. Basics of qualitative research: Grounded theory, procedures and techniques. Newbury Park, CA: Sage; 1998.
36. Lowell A, Maypilama E, Yikaniwuy S, Rrapa E, Williams R, Dunn S. "Hiding the story": indigenous consumer concerns about communication related to chronic disease in one remote region of Australia. *Int J Speech Lang Pathol.* 2012;14(3):200-8.

15 Appendices

- Appendix A** - Letter of Support: ALC
- Appendix B** - Letter of Support: YMDAC
- Appendix C** - Letter of Support: MJDF
- Appendix D** - Phase 1: Information sheet
- Appendix E** - Phase 1: Consent Form
- Appendix F** - Phase 1: MJD Expert Panel Recruitment Log
- Appendix G** - Phase 1: MJD Expert Panel Enrolment Form
- Appendix H** - Phase 1: Team Debriefing Minutes Form
- Appendix I** - Phase 2: Information Sheet
- Appendix J** - Phase 2: Consent Form
- Appendix K** - Phase 2: Participant Recruitment Log
- Appendix L** - Phase 2: Participant Enrolment Form
- Appendix M** - Phase 2: Participant Program Plan
- Appendix N** - Phase 2: Participant Progress Notes
- Appendix O** - Phase 2: Post-Trial Interview Topic Guide
- Appendix P** - Phase 2: Interview Contact Summary Form
- Appendix Q** - Phase 2: Investigator Debrief Meeting Form



**Anindilyakwa
Land
Council**

Ph 08 8987 4006
Fax 08 8987 4099
admin@alcnt.com.au

30 Bougainvillea Drive
PO Box 172
Alyangula NT 0885

To Whom It May Concern

The Anindilyakwa Land Council (ALC) is the custodian of traditional information, land, and people of the Grootte Eylandt Archipelago and work for the benefit of Grootte Eylandt people, on all matters that affect the land, rehabilitation and the people.

The ALC is responsible for land visitation by non-Indigenous people, illegal entry to lands, issuing of permits for visitation rights, ranger inspections and other daily management issues.

In 2008 and 2009, the ALC gifted the Machado Joseph Disease Foundation (MJDF) substantial grants in recognition of the needs of a growing number of people in the community who suffer with and care for family members with Machado Joseph Disease.

A strong relationship has since developed between the ALC and the MJDF, with a permanent position on the MJDF board held by an executive of the ALC. In this capacity we have been able to closely scrutinize and guide the activities of the MJDF ensuring that they continue to operate in accordance with the traditional and cultural requirements of the Anindilyakwa people.

The proposal by James Cook University in collaboration with the MJD Foundation to conduct an investigation into the physical activity needs of the MJD families in this region is another important step toward understanding the needs of these people and providing them with the best quality care available.

The ALC has considered the proposal and is supportive of the research. The researchers have met with representatives of the ALC to seek advice about the proper cultural process for the project. They have provided the ALC with documentation of the proposed research methodology as well as information regarding the storage and security of documents and data. These conditions for conduct of the research are acceptable to the ALC.

Yours sincerely

A handwritten signature in black ink, appearing to read 'Mark Hewitt', written over a horizontal line.

Mark Hewitt
Chief Executive Officer

18/07/16



anindilyakwa.com.au

Attn: Chairperson
Human Research Ethics Committee of the
Northern Territory Department of Health
and Menzies School of Health Research
PO Box 41096, Casuarina NT 0811
15th January 2018



To whom it may concern,

Re: Yugul Mangi Development Aboriginal Corporation support for the research project: *'Developing a physical activity program for Aboriginal families with Machado Joseph Disease (MJD) living in the Top End of Australia'*.

The Yugul Mangi Development Aboriginal Corporation has supported the MJD Foundation since early 2010, who provide support for families with MJD in Ngukurr. Yugul Mangi Development Aboriginal Corporation has provided assistance with cultural support, accommodation, airport transfers, car hire, storage of equipment.

This research is about finding the best way to help families with MJD keep 'walking and moving around' and 'staying stronger for longer'. As part of this research, we understand that researchers from James Cook University and MJD Foundation staff will be working alongside Aboriginal co-researchers from Ngukurr. The researchers will be consulting with families with MJD in Ngukurr to inform the development of a physical activity program, based on what matters and what works best to families with MJD. The program will be built on the priorities of families with MJD, as experts with the disease, combined with western science and research to support their ideas. Families with MJD in Ngukurr will be supported in this research to test the program they have worked to develop, to make sure it works and keeps families 'stronger for longer'.

Yugul Mangi Development Aboriginal Corporation supports this research, part of which will be conducted in the community of Ngukurr.

Yours sincerely,

Bill Blackley
CEO Yugul Mangi Development Aboriginal Corporation
Ngukurr, South East Arnhem Land
NT 0852

Yugul Mangi Development Ab.Corp.
CMB 6 Ngukurr 0852 ABN 40719772364

Menzies School of Health Research
Aboriginal Ethics Sub-Committee AND
Human Research Ethics Committee

Via email: ethics@menzies.edu.au

Date: 17/01/2018



Re: Support for the research project titled: Developing a physical activity program for Aboriginal families with MJD

To Whom It May Concern,

The MJD Foundation seeks to provide a better quality of life for Indigenous Australians and their families living with Machado Joseph Disease (MJD) in Arnhem Land and beyond. The MJD Foundation provides advocacy and practical support for individuals and families with MJD, information about MJD best practice and research coordination according to the priorities of individuals and families with MJD from the Top End of Australia.

The MJD Foundation has been extensively involved in the planning and preparation of this research project in collaboration with James Cook University researchers. On behalf of the MJD families of Groote Eylandt, the MJD Foundation Research Advisory Committee and the MJD Foundation Board have approved this research project.

The MJD Foundation will provide in-kind support for accommodation, transport and equipment as required for all research visits to Groote Eylandt and Ngukurr, in conjunction with funding from the Lowitja Institute. Staff of the MJD Foundation will assist with this research project and have been heavily involved in the development of this research. In particular, Gayangwa Lalara (MJD Foundation Vice-Chairperson and Senior Cultural Advisor) and Joyce Lalara (MJD Foundation Community Researcher) will be Associate Investigators for the study and local community research partners. Community Research Partners will play a significant role in participant recruitment, informed consent, conduct of the consensus rounds and the physical activity and lifestyle program, data collection, translation, transcription, data analysis, write up and dissemination of results. Additionally, these local investigators will provide cultural guidance and support for the study, coordinate community access for the research and communicate closely with MJD clients and families throughout the research project.

This research project is a priority for individuals and families living with MJD on Groote Eylandt and Ngukurr. Families with MJD will be able to share their knowledge on the best way to keep families with MJD 'walking and moving around' and living a good life. Families feel strongly that exercise helps keep people with MJD stronger for longer, and this research will help families demonstrate this to other families with MJD, health professionals and researchers around the world. This will help families with MJD across the Top End. The MJD Foundation has considered this research proposal carefully and provides full support for conduct of the research.

Yours sincerely,

A handwritten signature in black ink, appearing to read 'Libby Massey', written over a decorative graphic of a path of colored dots.

Libby Massey
Director, Research & Community Services
Mob: 0417 279 120
Email: libby.massey@mjd.org.au

MJD Foundation Limited
ABN: 65 159 208 867
PO Box 414,
Alyangula NT 0885
www.mjd.org.au
info@mjd.org.au
1300 584 122



THIS IS FOR YOU TO KEEP



Developing a physical activity program for Aboriginal families with MJD

We would like you to be part of our study as an expert in MJD in families with MJD in the Top End of Australia. We would like to show you an exercise and lifestyle program that we have made for families with MJD, from listening to their stories and looking at the best science from in Australia and overseas. You will be part of a group of **experts** and asked what you think about the program. We would like to know if the program is strong and true. We would like you to tell us how this program could be the best it can be. It is important that we make sure all **experts** agree that the program is a strong one, so it is right for families to test it.

If you would like to join in the research as an MJD Expert, there are some things you'll be asked to do:

- Sign a form
 - Saying you are happy to be part of the MJD Expert Group
- Sit together with other MJD experts and the researchers
 - Look at the main stories about what keeps people with MJD walking and moving around
 - Talk about what should be in the program to keep people with MJD strong
 - Tell us how to make it better
 - Give your ideas about the program so it can be strong for families with MJD
 - We can read and explain the program to you in your preferred language

**If you agree, we would like to record your answers,
and record your voice if you want to tell us what you think in person**

If you have any worries about the study, you can talk to someone from the MJD Foundation or someone from ALC or the clinic.

**You do not have to do this research. It is your choice.
If you don't want to be in the MJD Expert Group, you can just stop.**

A lady called Jen is a physiotherapist, who has been working with Gayangwa Lalara and Joyce Lalara (researchers with the MJD Foundation), will be talking to you about the program. The other people who will be helping her Ruth Barker from James Cook University, Anne Lowell from Charles Darwin University and Alan Clough from James Cook University. Other staff of the MJD Foundation will help too.

What you tell us will make the program better and will be written in a paper and read by other people who want to know about MJD. We won't use your name or your family's name. If you are from Groote Eylandt or Ngukurr, the MJD Foundation will provide transport for you to be part of the research, and have a computer or iPad available when you need to look at research work. All of your information will be kept safe so that only the researchers for this study will be able to see it. If you have any questions, please ask the staff of the MJD Foundation. If you have any concerns or complaints regarding the ethical conduct of the study, you are invited to contact Ethics Administration, Human Research Ethics Committee of the NT Department of Health and Menzies School of Health Research on 8946 8687 or 89468692 or email ethics@menzies.edu.au

Are there any benefits in taking part in the study?

You will be paid for being involved as an expert in this research (\$75 per workshop). Your expert ideas will help explain to health professionals and other people with MJD the best way to help people with MJD to keep 'walking and moving around' and 'stronger for longer'.

Are there any risks in taking part in the study?

It may be difficult to talk about MJD at times, especially if you or someone in your family has MJD. You can stop at any time if you feel upset or don't want to keep going. Your personal information will be kept private so that only the researchers can see it and kept in a locked office or in a locked computer.

Ethics

This study has been approved by the ALC, YMDAC, the MJD Research the Human Research Ethics Committee of the NT Department of Health and Menzies School of Health Research and James Cook University Human Research Ethics Committee.

Research Team

| | |
|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| <p>Jennifer Carr Student Investigator PhD student James Cook University Ph: 0407968512 Email: jennifer.carr2@my.jcu.edu.au</p> <p>Associate Professor Ruth Barker Principal Investigator Associate Professor – Rehabilitation College of Healthcare Sciences James Cook University Ph: 07 42321614 Email: ruth.barker@jcu.edu.au</p> <p>Associate Professor Anne Lowell 2nd Investigator School of Health Charles Darwin University Casuarina NT 0909 T: 08 8946 6297 E: anne.lowell@cdu.edu.au</p> <p>Professor Alan Clough 3rd Investigator College of Healthcare Sciences James Cook University Ph: 07 4232 1604 Email: alan.clough@jcu.edu.au</p> <p>Gayangwa Lalara 4th Investigator Community Research Partner Vice-Chairperson Senior Cultural Advisor Machado Joseph Disease Foundation PO Box 414, Alyangula NT 0885 Ph: 1300-584-122 gayangwa.lalara@mjd.org.au</p> | <p>Joyce Lalara 5th Investigator Community Research Partner Aboriginal Health Worker Machado Joseph Disease Foundation PO Box 414, Alyangula NT 0885 Phone: 1300-584-122 Email: c/ libby.massey@mjd.org.au</p> <p>Bronwyn Daniels 6th Investigator Community Research Partner Aboriginal Health Worker Machado Joseph Disease Foundation PO Box 414, Alyangula NT 0885 Phone: 1300-584-122 Email: c/ libby.massey@mjd.org.au</p> <p>Libby Massey 7th Investigator Community Research Partner Director of Research and Community Services Machado Joseph Disease Foundation Phone: 0417279120 Email: libby.massey@mjd.org.au</p> <p>Nick Kenny 8th Investigator Physiotherapist Manager of Community Services, Ngukurr/Groote Eylandt Machado Joseph Disease Foundation Phone: 0417279120 Email: nick.kenny@mjd.org.au</p> |
|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|



**Developing a physical activity program for Aboriginal families with MJD
(THIS MEANS YOU CAN SAY NO)**

| | | |
|-----------------------|--------------------------------------------------------------------------------------------------------------|----------------------------------------------------------------------------------|
| INVESTIGATORS: | Jennifer Carr Associate Professor Ruth Barker Associate Professor Anne Lowell Professor Alan Clough | Gayangwa Lalara Joyce Lalara Bronwyn Daniels Libby Massey Nick Kenny |
|-----------------------|--------------------------------------------------------------------------------------------------------------|----------------------------------------------------------------------------------|

| | |
|---------------------|--------------------------------------------------------------------------------|
| STUDY TITLE: | Developing a physical activity program for Aboriginal families living with MJD |
|---------------------|--------------------------------------------------------------------------------|

I know that the purpose of this research is to be part of a group of experts telling the researchers whether the program they have come up will be the best way to keep families walking and moving around and having a good life.

I am happy to be in the MJD Expert Group, and have been told what will happen, and I have been given an information sheet to keep.

I am happy for the researchers to use the expert information I give in papers for other people to read.

I know that:

- All the things I have to do have been explained to me
- Taking part in this study is my choice. I can stop taking part at any time without reason.
- Only the researchers and MJD staff will hear my story and use it in papers. My name won't be written in the papers.

Please circle applicable: Written consent / Oral consent (Please tick to indicate consent)*

| | | | | |
|---------------------------------------------------------------------------------------|--|-----|--|----|
| I am happy to be on the MJD Expert group | | Yes | | No |
| I am happy to talk about this program and to tell researchers what I think about it | | Yes | | No |
| I am happy for my comments to be recorded, either in writing or with a voice recorder | | Yes | | No |

Name: *(please print)* **Please refer to recorded oral consent if identified above and witness signature below*

| | | |
|-------------------------------------------------------|--------------------------------------|--------------|
| Signature: | Date: | |
| Name of Parent/Guardian: <i>(please print)</i> | Signature of Parent/Guardian: | Date: |
| Name of Witness: <i>(please print)</i> | Signature of Witness: | Date: |

For questions, please contact: Associate Professor Ruth Barker (Principal Investigator), James Cook University.
Ph: 07 42321614 - Email: ruth.barker@jcu.edu.au (See information sheet for more study information)

Appendix F Phase 1: MJD Expert Panel Recruitment Log

| ID | First name | Age | Gender (M/F/NA) | Expert with MJD: (Y/N - If so, please state age of onset) | Profession / Expertise in MJD (Please describe) | Mode of workshop participation (Face to face, Phone, Skype/Zoom) | Audio recorded (Yes/No) | Best Contact Details |
|----|------------|-----|--------------------|--------------------------------------------------------------|----------------------------------------------------|---------------------------------------------------------------------|----------------------------|----------------------|
| 1 | | | | | | | | |
| 2 | | | | | | | | |
| 3 | | | | | | | | |
| 4 | | | | | | | | |
| 5 | | | | | | | | |
| 6 | | | | | | | | |
| 7 | | | | | | | | |
| 8 | | | | | | | | |
| 9 | | | | | | | | |
| 10 | | | | | | | | |

Appendix G Phase 1: MJD Expert Panel Enrolment Form

| Participant Name, contact number and email | Date Approached | Gender | Background of expertise in MJD <i>(Please describe)</i> | Diagnosis of MJD <i>(Y or N)</i> | Location | Preferred mode of workshop participation <i>(face to face, videoconference, over the phone)</i> | Interested? <i>(Y or N)</i> | Consent? <i>(Y or N)</i> | Special Requirements or requests for participation? <i>(List any i.e. transport, require carer present))</i> |
|--------------------------------------------------|--------------------|--------|--------------------------------------------------------------------------|--------------------------------------------|----------|----------------------------------------------------------------------------------------------------------------------|------------------------------------|---------------------------------|-----------------------------------------------------------------------------------------------------------------------------------|
| 1 | | | | | | | | | |
| 2 | | | | | | | | | |
| 3 | | | | | | | | | |
| 4 | | | | | | | | | |
| 5 | | | | | | | | | |
| 6 | | | | | | | | | |
| 7 | | | | | | | | | |
| 8 | | | | | | | | | |
| 9 | | | | | | | | | |

Team debriefing session minutes – Workshops

Date: |_|_|/|_|_|/|_|_|

List those present:

Workshop number:

- Was the workshop conducted as planned? If not, what were the reasons?

- What parts of the program need to be changed/removed after this workshop?

- Was there any new information gathered from the workshop that needs to be included in the guidelines or following workshops?

- Discuss any problems with the workshop



THIS IS FOR YOU TO KEEP



Developing a physical activity program for Aboriginal families with MJD Information Sheet

We would like you to be part of our research to try out an exercise program for families with MJD, designed to help keep people with MJD walking and moving around and living a good life.

We've worked with families with MJD, experts in MJD and the MJD Foundation to bring ideas together about what is important for this program. We want to invite you to try the program for yourself. A lady physiotherapist named Jen, will be working together with local researchers and MJD Foundation staff and helping you to try the program. These people will sit with you to work out how this program can be made so that is just right for you, doing the things you want to do. Jen will help you do the program, and your family can join in too. We would like to help you try the program for about 6 weeks.

If you would like to join in the research and try the program, there are some things you'll be asked to do:

- Sign a form to say you are happy to be part of the research to try the program
- Talk with the researchers about your MJD and what you'd like to work on with walking and moving around.
- Try out an exercise and lifestyle program designed for you for 6 weeks
- Do some walking, strength and balance tests before, during and after the program
- Answer some questions about your life with MJD before and after the program
- Tell them what you thought of the program, your ideas, and how it can be better

**If you agree, we would like to record your answers,
and record your voice if you want to tell us what you think in person**

If you have any worries about the study, you can talk to someone from the MJD Foundation or someone from ALC or the clinic.

The other people who will be helping her Ruth Barker from James Cook University, Anne Lowell from Charles Darwin University and Alan Clough from James Cook University. Other staff of the MJD Foundation will help too.

What you tell us will and what the tests tell us will be written in a paper and read by other people who want to know about MJD. We won't use your name or your family's name. The MJD Foundation will provide transport for you to be part of the research, and there will be a computer or iPad available when you need to look at research work. All of your information will be kept safe so that only the researchers for this study will be able to see it.

**You do not have to do this research. It is your choice.
If you decide you don't want to do the program, you can just stop**

If you have any questions, please ask the staff of the MJD Foundation. If you have any concerns or complaints regarding the ethical conduct of the study, you are invited to contact Ethics Administration, Human Research Ethics Committee of the NT Department of Health and Menzies School of Health Research on 8946 8687 or 89468692 or email ethics@menzies.edu.au

Are there any benefits in taking part in the study?

There will be no direct benefits to you from participating in the study. However, your stories will help explain to health professionals and other people with MJD ways to help families with MJD to stay strong and keep 'walking and moving around.' There will be no payment provided to you to be in the study.

Are there any risks in taking part in the study?

Exercising might make you feel tired and make your muscles sore. It may be difficult to talk about MJD at times. However, you can stop at any time if you feel upset or don't want to do the exercise anymore. Your personal information will be kept private so that only the researchers can see it and kept in a locked office or in a locked computer.

Ethics

This study has been approved by the ALC, YMDAC, the MJD Research the Human Research Ethics Committee of the NT Department of Health and Menzies School of Health Research and James Cook University Human Research Ethics Committee.

Research Team

| | |
|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| <p>Jennifer Carr Student Investigator PhD student James Cook University Ph: 0407968512 Email: jennifer.carr2@my.jcu.edu.au</p> <p>Associate Professor Ruth Barker Principal Investigator Associate Professor – Rehabilitation College of Healthcare Sciences James Cook University Ph: 07 42321614 Email: ruth.barker@jcu.edu.au</p> <p>Associate Professor Anne Lowell 2nd Investigator School of Health Charles Darwin University Casuarina NT 0909 T: 08 8946 6297 E: anne.lowell@cdu.edu.au</p> <p>Professor Alan Clough 3rd Investigator College of Healthcare Sciences James Cook University Ph: 07 4232 1604 Email: alan.clough@jcu.edu.au</p> <p>Gayangwa Lalara 4th Investigator Community Research Partner Vice-Chairperson Senior Cultural Advisor Machado Joseph Disease Foundation PO Box 414, Alyangula NT 0885 Ph: 1300-584-122 gayangwa.lalara@mjd.org.au</p> | <p>Joyce Lalara 5th Investigator Community Research Partner Aboriginal Health Worker Machado Joseph Disease Foundation PO Box 414, Alyangula NT 0885 Phone: 1300-584-122 Email: c/ libby.massey@mjd.org.au</p> <p>Bronwyn Daniels 6th Investigator Community Research Partner Aboriginal Health Worker Machado Joseph Disease Foundation PO Box 414, Alyangula NT 0885 Phone: 1300-584-122 Email: c/ libby.massey@mjd.org.au</p> <p>Libby Massey 7th Investigator Community Research Partner Director of Research and Community Services Machado Joseph Disease Foundation Phone: 0417279120 Email: libby.massey@mjd.org.au</p> <p>Nick Kenny 8th Investigator Physiotherapist Manager of Community Services, Ngukurr/Groote Eylandt Machado Joseph Disease Foundation Phone: 0417279120 Email: nick.kenny@mjd.org.au</p> |
|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|



**Developing a physical activity program for Aboriginal families with MJD living in the Top End of Australia: Consent Form
(THIS MEANS YOU CAN SAY NO)**



| | | | | | | | | | | | |
|---------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|--------------------------------------------------------------------------------------------------------------|----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----|--------------------------|----|-----|--------------------------|----|-----|--------------------------|----|
| INVESTIGATORS: | Jennifer Carr Associate Professor Ruth Barker Associate Professor Anne Lowell Professor Alan Clough | Gayangwa Lalara Joyce Lalara Bronwyn Daniels Libby Massey Nick Kenny | | | | | | | | | |
| STUDY TITLE: | Developing a physical activity program for Aboriginal families with MJD living in the Top End of Australia | | | | | | | | | | |
| <p>I know that the purpose of this research is to be part of a research project to try out a physical activity and lifestyle program for families with MJD, to help keep people with MJD walking and moving around and living a good life.</p> <p>I am happy to be part of the research, and try the program for myself. I have been told what will happen, and I have been given an information sheet to keep. I am happy for the researchers to use what I say about the program in papers for other people to read. I know that my real name or my families name won't be used in these papers.</p> <p>I know that:</p> <ul style="list-style-type: none"> • All the things I have to do have been explained to me • Taking part in this study is my choice. I can stop taking part at any time without reason. • Only the researchers and MJD staff will hear my story and use it in papers. My name won't be written in the papers. | | | | | | | | | | | |
| <i>Please circle applicable: Written consent / Oral consent*</i> | | <i>(Please tick to indicate consent)</i> | | | | | | | | | |
| I am happy to try the program out | <input type="checkbox"/> | <table border="1" style="width:100%; border-collapse: collapse;"> <tr> <td align="center" style="width:33%;">Yes</td> <td align="center" style="width:33%;"><input type="checkbox"/></td> <td align="center" style="width:33%;">No</td> </tr> <tr> <td align="center">Yes</td> <td align="center"><input type="checkbox"/></td> <td align="center">No</td> </tr> <tr> <td align="center">Yes</td> <td align="center"><input type="checkbox"/></td> <td align="center">No</td> </tr> </table> | Yes | <input type="checkbox"/> | No | Yes | <input type="checkbox"/> | No | Yes | <input type="checkbox"/> | No |
| Yes | <input type="checkbox"/> | No | | | | | | | | | |
| Yes | <input type="checkbox"/> | No | | | | | | | | | |
| Yes | <input type="checkbox"/> | No | | | | | | | | | |
| I am happy to do some walking, strength and balance tests and tell researchers what I think about the program | <input type="checkbox"/> | <table border="1" style="width:100%; border-collapse: collapse;"> <tr> <td align="center" style="width:33%;">Yes</td> <td align="center" style="width:33%;"><input type="checkbox"/></td> <td align="center" style="width:33%;">No</td> </tr> <tr> <td align="center">Yes</td> <td align="center"><input type="checkbox"/></td> <td align="center">No</td> </tr> <tr> <td align="center">Yes</td> <td align="center"><input type="checkbox"/></td> <td align="center">No</td> </tr> </table> | Yes | <input type="checkbox"/> | No | Yes | <input type="checkbox"/> | No | Yes | <input type="checkbox"/> | No |
| Yes | <input type="checkbox"/> | No | | | | | | | | | |
| Yes | <input type="checkbox"/> | No | | | | | | | | | |
| Yes | <input type="checkbox"/> | No | | | | | | | | | |
| I am happy for my comments to be recorded, either in writing or with a voice recorder | <input type="checkbox"/> | <table border="1" style="width:100%; border-collapse: collapse;"> <tr> <td align="center" style="width:33%;">Yes</td> <td align="center" style="width:33%;"><input type="checkbox"/></td> <td align="center" style="width:33%;">No</td> </tr> <tr> <td align="center">Yes</td> <td align="center"><input type="checkbox"/></td> <td align="center">No</td> </tr> <tr> <td align="center">Yes</td> <td align="center"><input type="checkbox"/></td> <td align="center">No</td> </tr> </table> | Yes | <input type="checkbox"/> | No | Yes | <input type="checkbox"/> | No | Yes | <input type="checkbox"/> | No |
| Yes | <input type="checkbox"/> | No | | | | | | | | | |
| Yes | <input type="checkbox"/> | No | | | | | | | | | |
| Yes | <input type="checkbox"/> | No | | | | | | | | | |
| Name: (please print) _____ <i>*Please refer to recorded oral consent if identified above and witness signature below</i> | | | | | | | | | | | |
| Signature: | Date: | | | | | | | | | | |
| Name of Parent/Guardian: (please print) | Signature of Parent/Guardian: | Date: | | | | | | | | | |
| Name of Witness: (please print) | Signature of Witness: | Date: | | | | | | | | | |

For questions, please contact: Associate Professor Ruth Barker (Principal Investigator), James Cook University
Ph: 07 42321614 - Email: ruth.barker@jcu.edu.au (See information sheet for more study information)

Appendix K Phase 2: Participant Recruitment Log

| ID | Participant Name | M/F | Date Approached | Successful contact? | Eligible to Participate (Y or N) | Interested? | Consent? | Initial assessment date arranged | Location | Transport Required |
|----|------------------|-----|-----------------|---------------------|----------------------------------|-------------|----------|----------------------------------|----------|--------------------|
| 1 | | | | | | | | | | |
| 2 | | | | | | | | | | |
| 3 | | | | | | | | | | |
| 4 | | | | | | | | | | |
| 5 | | | | | | | | | | |
| 6 | | | | | | | | | | |
| 7 | | | | | | | | | | |
| 8 | | | | | | | | | | |
| 9 | | | | | | | | | | |
| 10 | | | | | | | | | | |
| 11 | | | | | | | | | | |

Appendix L Phase 2: Participant Enrolment Form

| ID | First name | Age | Community | Age of onset of MJD | Times since onset of MJD | Mobility status Walking independently - with or without mobility aids (I), walking with assistance (A), wheelchair dependent (WC)) | Describe current level of physical activity (exercise or leisure activities) | Living situation (alone/with family/with carer) | Working? (Y/N – if yes, please enter work role) | Consent to be audio recorded (Yes/No) |
|----|------------|-----|-----------|---------------------|--------------------------|-------------------------------------------------------------------------------------------------------------------------------------------|-------------------------------------------------------------------------------------|--------------------------------------------------------|--------------------------------------------------------|----------------------------------------------|
| 1 | | | | | | | | | | |
| 2 | | | | | | | | | | |
| 3 | | | | | | | | | | |
| 4 | | | | | | | | | | |
| 5 | | | | | | | | | | |
| 6 | | | | | | | | | | |
| 7 | | | | | | | | | | |
| 8 | | | | | | | | | | |
| 9 | | | | | | | | | | |
| 10 | | | | | | | | | | |
| 11 | | | | | | | | | | |



My plan to keep myself walking and moving around

Participant ID:

Date:



| My Goal: What's important to me | Things I am doing to keep myself strong inside and outside | | | | | | | |
|-------------------------------------------------------------------------------|------------------------------------------------------------|------------------------------------------------------------------------------------------------------------|---------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-------------------------------------------------------------------------------------------------------------------------|---------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|--------|------------------------------------------------------------------------------------------------------|--------|
| | Week | Monday | Tuesday | Wednesday | Thursday | Friday | Saturday | Sunday |
| <i>Example:</i> Walking in the bush looking for Mangkarrkba with my family | 1 | <ul style="list-style-type: none"> Go walking out bush with Jen and Gayangwa when it's cool | <ul style="list-style-type: none"> Practice walking over uneven ground at the back of Aged Care with Jen and Joyce Ride the exercise bike in the aircon | <ul style="list-style-type: none"> On country with family and MJD Foundation – fishing, hunting, walking | <ul style="list-style-type: none"> Practice stepping over big things and soft things and making my legs strong Family night – big feed and watch movies | | <ul style="list-style-type: none"> Big bush trip: Fishing and looking for Mangkarrkba | |
| | 2 | | | | | | | |
| | 3 | | | | | | | |
| | 4 | | | | | | | |
| | 5 | | | | | | | |
| | 6 | | | | | | | |

Appendix N Phase 2: Participant Progress Notes

Progress Notes



ID NO: |_|_|_|_|

Audio file #: |_|_|_|_| | **Community:** |_____|

Today's date: |_|_|/|_|_|/|_|_|



Program Week: |__|

Subjective:

Objective:

Activities done today:

(how long, how many, where, who with?)

Issues/Adverse Events:

Comments from Participant:

What worked well today?

What didn't work so well?

Any issues with the program plan/forms?

Appendix O Phase 2: Post-Trial Interview Guide

Post-Trial – Interview Topic Guide



ID NO: |_|_|_|_|_|

Audio file #: |_|_|_|_|_| Community: |_____|

Today's date: |_|_|_|/|_|_|_|/|_|_|_|

Tell us your story about this program?

What was good about the program?

What wasn't good about the program?

What's missing from the program?

How could it be better?

What would you change?

What do you think about keeping going with the program?

What do you think about this program for other families with MJD?

Appendix P Phase 2: Interview Contact Summary Form

Contact Summary Form (for each post-trial interview)



ID NO: |_|_|_|_|_|

Audio file #: |_|_|_|_|_| Community: |_____|

Today's date: |_|_|_|/|_|_|_|/|_|_|_|

- How would you describe the atmosphere and context of the interview?
- What were the main points made by the participant during this interview?
- What new information did you gain through this interview?
- Was there anything surprising to you? Or that made you think differently?
- What messages did you take from this interview?
- Were there any problems with the topic guide (e.g. wording, order of topics, missing topics) used during the interview?

Appendix Q Phase 2: Investigator Debrief Meeting Form

Investigator Debrief Meeting Form

ID NO: |_|_|_|_|_|



Audio file #: |_|_|_|_|_| **Investigators Present:**

Today's date: |_|_|_|/|_|_|_|/|_|_|_|

Progress with data collection?

What have been the key findings so far?

What are the implications of these findings on the program?

Have there been any problems with the program?

Have any changes to the program been made?

18 January 2018

Miss Jennifer Carr
PhD Student,
James Cook University

Email: jennifer.carr2@my.jcu.edu.au



the
Lowitja
Institute

Australia's National Institute for Aboriginal and
Torres Strait Islander Health Research

Dear Miss Carr,

Re: Developing a physical activity program for Aboriginal families with Machado Joseph Disease (MJD) living in the Top End of Australia.

Thank you for applying to the 2017 Lowitja Institute research funding round.

We are pleased to advise that your application has been successful.

Research project applications were subject to rigorous evaluation by an Aboriginal and Torres Strait Islander majority assessment panel with an independent chairperson. Each application was assessed by at least three people with experience and expertise in Aboriginal and Torres Strait Islander health research, and/or policy development and practice.

We will contact you very shortly with more detail around project commencement, and in particular, contract development.

Should you have any queries in the meantime, please contact Mary Guthrie - mary.guthrie@lowitja.org.au or via phone on 03 8341 5504.

Yours sincerely,

Romlie Mokak
CEO

RESEARCH ACTIVITY FUNDING AGREEMENT

BETWEEN

**NATIONAL INSTITUTE FOR ABORIGINAL AND TORRES STRAIT ISLANDER HEALTH RESEARCH
LIMITED**

trading as

THE LOWITJA INSTITUTE

and administering

**THE LOWITJA INSTITUTE ABORIGINAL AND TORRES STRAIT ISLANDER HEALTH CRC
(‘the Company’)**

and

**James Cook University
(‘Administering Institution’)**

Regarding provision of funding for a Research Activity 017-SF-005

**The Lowitja Institute Aboriginal and Torres Strait Islander Health CRC
Research Activity Funding Agreement**

Appendix R Evidence of Lowitja Institute Funding Support - Cont

Schedule 3 – Project Cash and In-kind Contributions

Item 1 – Cash Contributions

| Entity | 2018 | 2019 | Total |
|---------------------------------|------|------|-------|
| Administering Institution | | | |
| Participating Institution 1 | | | |
| Participating Institution 2 | | | |
| Total Cash Contributions | Nil | Nil | Nil |

Item 2 – In-kind Contributions

| Staff In-Kind – Administering Institution | 2018 | 2019 | Total |
|--------------------------------------------------|---------|------|---------|
| <i>Position</i> | | | |
| Program Leader/Senior Manager | 1.2 | | 1.2 |
| Key Researcher/Manager/Project Leader | | | |
| Researcher/Professional | 0.2 | | 0.2 |
| Other (support staff, technical, administrative) | | | |
| Total Staff In-Kind Contribution | 1.4 | | 1.4 |
| Non-Staff In-Kind | | | |
| Non-staff in-kind contribution | \$2,080 | - | \$2,080 |

| Staff In-Kind – Participating Institution 1 | 2018 | 2019 | Total |
|--------------------------------------------------|----------|------|----------|
| <i>Position</i> | | | |
| Program Leader/Senior Manager | | | |
| Key Researcher/Manager/Project Leader | | | |
| Researcher/Professional | 0.67 | | 0.67 |
| Other (support staff, technical, administrative) | | | |
| Total Staff In-Kind Contribution | 0.67 | | 0.67 |
| Non-Staff In-Kind | | | |
| Non-staff in-kind contribution | \$46,360 | - | \$46,360 |

Schedule 4 – Payment of Project Funds

Item 1 – Payment Schedule

| No. | Payment Description | Amount (GST excl.) | Conditions |
|-----|-----------------------|--------------------|-----------------------------------------------------------------------------------------------------------------------------------------------------|
| 1 | Project payment 1 | \$49,538 | Initial payment in Feb 2018 |
| 2 | Project payment 2 | \$29,722 | Submission of all reports and other deliverables due by 30 Sept 2018, including all in-kind contributions for the reporting period met or exceeded. |
| 3 | Final project payment | \$19,816 | Submission of all reports and other deliverables of the project and all in-kind contributions for the reporting period met or exceeded. |
| | Total Payments | \$99,076 | |

Appendix R Evidence of Lowitja Institute Funding Support - Cont

EXECUTED as an AGREEMENT

EXECUTED by the National Institute for
Aboriginal and Torres Strait Islander Health
Research Limited,)

(ABN 70 138 780 695)


Signature of Chief Executive Officer

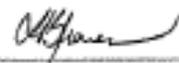
Mr Romlie Mokak
Name of Chief Executive Officer

23/11/17
Date of Execution


Signature of Witness

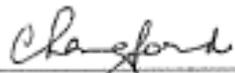
SHAYNE BELLINGHAM
Name of Witness

EXECUTED by James Cook University)
(ABN 46 253 211 955))


Signature of Witness

Lenore Brown
Name of Witness (print)

22/11/17
Date of Execution


Signature of Authorised Signatory

Tina Langford
Name of Authorised Signatory (print)
Manager
Research Ethics & Grants

James Cook University