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Testing the efficacy of an online interactive decision aid and educational tool to help parents of children with intellectually disabilities choose whether or not to have genome-wide sequencing

Submission date
06/02/2015

Recruitment status
No longer recruiting

Registration date
19/02/2015

Overall study status
Completed

Last Edited
23/10/2018

Condition category
Mental and Behavioural Disorders

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

Plain English summary of protocol
Background and study aims
DECIDE is an online multimedia tool to help parents choose whether or not to have a complex new test, GWS, to find the cause of their child's intellectual disability. Parents need to understand the meaning of GWS results because of their potential impact on the family. Tools like DECIDE are effective at promoting informed choices that are consistent with users' values. Our research also shows that parents want information in a variety of formats that are suited to their needs. DECIDE aims to fill this need. This study will measure the effectiveness of DECIDE compared with conventional in-person pretest genetic counselling and will determine what type of information participants want to know.

Who can participate?
Children with intellectual disabilities who have been seen by Dr Anna Lehman at the University of British Columbia (Canada)

What does the study involve?
Children will be randomly assigned to one of two groups: those in arm 1 will receive DECIDE first followed by genetic counselling and those in arm 2 will receive genetic counselling followed by DECIDE.

What are the possible benefits and risks of participating?
The results of the study will improve DECIDE for future users; however, this may not be a direct benefit to participants. In terms of GWS results, a diagnosis is expected for one in four participants. Some parents find it beneficial to finally know the cause of their child's problems. It means no more diagnostic testing. Sometimes parents say that it can help to obtain school resources or help in explaining a child's difficulties to others. They also say that it helps give them a direction for the future. Parents may also be able to connect with families with similar

conditions and that can be helpful. It is possible that participants will learn about a condition for which screening or early treatment could be medically important to reduce the impact of a condition. People who learn about conditions for which there is no medical treatment may choose to make lifestyle changes to adjust to living at a higher risk of this condition occurring. Although we have tested DECIDE in small groups of families, we have not used it in people who are actually making a decision about GWS. We do not know how well it will work. It is possible that it could be confusing or distressing. The research genetic counsellor and all other team members are available to help clarify any confusion that may result from using DECIDE. Any type of testing can be emotional and can create anxiety and GWS is no exception. If families do not receive a result that gives a genetic reason for their child's problems, this could be very disappointing to them. Sometimes sequencing reveals variants whose importance cannot be determined at the time. Sometimes these are discussed with parents, depending on whether we think they could be harmful. Receiving information about a variant of unknown significance can be stressful because it can introduce more uncertainty. If families choose to know about the incidental findings, they will not know ahead of time if there will be any such findings or what they might be. This may make some people quite anxious. In addition to the risks of physical harms, there are also possible non-physical risks associated with taking part in this study. For example, disclosure of genetic research data could result in discrimination by employers or insurance providers. Incidental findings that are not confirmed in a clinical laboratory do not have to become part of a patient's medical record but it is possible that these research results may still be demanded by insurance companies and could result in the patient being refused life or extended health insurance. These issues will be discussed in detail in the genetic counselling session and by DECIDE.

Where is the study run from?
Child and Family Research Institute, University of British Columbia (Canada)

When is the study starting and how long is it expected to run for?
January 2014 to March 2019

Who is funding the study?
Canadian Institutes of Health Research (Canada)

Who is the main contact?
Dr Patricia Birch

Contact information

Type(s)
Scientific

Contact name
Dr Patricia Birch

Contact details
Box 150
4500 Oak Street
Vancouver
Canada
V6H 3N1

Additional identifiers

Protocol serial number
N/A

Study information

Scientific Title
Assessing the clinical utility of a novel e-counselling aid for clinical genome-wide sequencing (DECIDE): a randomised cross over trial

Acronym
DECIDE

Study objectives
1. DECIDE, a multi-media, online information source and decision aid for genome-wide sequencing (GWS), is an effective tool that will:
1.1. Empower participants about GWS decision-making (measured with the Genetic Counselling Outcome Scale [GCOS-24]); empowerment is a clinical construct that describes individuals' belief that they have decisional, behavioural, emotional and cognitive control, and hope over their situation
1.2. Help parents to acquire accurate and balanced GWS information (measured with a knowledge questionnaire)
1.3. Inform and support parents' decision-making about GWS (measured with a decisional comfort scale)
1.4. Be acceptable to parents (measured with structured interviews)
2. By comparison with conventional in-person pretest genetic counselling, DECIDE will:
2.1. Result in equivalent levels of decisional conflict, which is a short-term measure
2.2. Result in equivalent levels of knowledge of GWS and its consequences
3. Half the participants will see a genetic counsellor first and then use DECIDE; the other half will use DECIDE first and then see a genetic counsellor; we hypothesise that participants who use DECIDE before seeing a genetic counsellor will:
3.1. Ask qualitatively more sophisticated questions
3.2. Have a better understanding of the testing implications of GWS
3.3. Need less in-person genetic counselling time
3.4. Have less decisional regret, which is a longer term outcome measure than is decisional conflict
4. When parents are offered choices relating to incidental findings, they are able to articulate which incidental findings they would choose for:
4.1. Themselves
4.2. Their children
This is a pilot project to provide preliminary evaluation of these hypotheses

Ethics approval required
Old ethics approval format

Ethics approval(s)
University of British Columbia Research Ethics Board (Canada), 21/10/2014, ref: H14-01139

Study design

Single-centre interventional randomised cross over study

Primary study design
Interventional

Study type(s)
Treatment

Health condition(s) or problem(s) studied
Intellectual disabilty

Interventions
Two different methods of providing genetic counselling-type information for parents to help them to choose whether or not to have genome-wide sequencing:
1. DECIDE
2. Conventional genetic counselling
Arm 1 will have DECIDE first followed by genetic counselling; arm 2 will have genetic counselling followed by DECIDE.

Intervention Type
Other

Primary outcome(s)
Participants' empowerment: will be measured with GCOS-24 at time 0, 1 month and study exit (6 months)

Key secondary outcome(s))
1. Participants' knowledge
2. Participants' decisional conflict (validated scale by O'Connor, Ottawa Decision Support Framework group)
3. Participants' decisional regret scale (validated scale by O'Connor, Ottawa Decision Support Framework group)
These will be measured at time 0, 1 month and study exit (6 months).

Completion date
30/03/2019

Eligibility

Key inclusion criteria
1. Two-parent family with child who has a significant developmental delay or intellectual disability with an unidentified cause after standard testing, including microarray, and who has been seen by Dr Anna Lehman (University of British Columbia, Department of Medical Genetics, Canada)
2. Fluent in English
3. Access to the internet
4. All ages, but mostly age 0–18 years

Participant type(s)
Patient

Healthy volunteers allowed
No

Age group
All

Sex
All

Key exclusion criteria
Not meeting inclusion criteria

Date of first enrolment
01/11/2017

Date of final enrolment
01/04/2018

Locations

Countries of recruitment
Canada

Study participating centre
Child and Family Research Institute, University of British Columbia
Vancouver
Canada
V6H 3N1

Sponsor information

Organisation
Canadian Institute of Health Research (Canada)

ROR
https://ror.org/01gavpb45

Funder(s)

Funder type
Government

Funder Name
Canadian Institutes of Health Research

Alternative Name(s)
Instituts de Recherche en Santé du Canada, The Canadian Institutes of Health Research (CIHR), Canadian Institutes of Health Research (CIHR), Canadian Institutes of Health Research | Ottawa ON, CIHR - Welcome to the Canadian Institutes of Health Research, CIHR, IRSC

Funding Body Type
Government organisation

Funding Body Subtype
National government

Location
Canada

Results and Publications

Individual participant data (IPD) sharing plan
The data sharing plans for the current study are unknown and will be made available at a later date.

IPD sharing plan summary
Data sharing statement to be made available at a later date

Study outputs

Output type	Details	Date created	Date added	Peer reviewed?	Patient-facing?
Results article	results	01/07/2018		Yes	No