

Centre for Educational Development, Appraisal and Research (CEDAR)

University of Warwick

**Cerebra 1000 Families Study**  
**End of Grant Report - December 2022**



Centre for Educational Development  
Appraisal and Research

## ***Introduction to the Cerebra 1000 Families Study***

In 2013, Cerebra made an initial investment over six years (2014-2019) in the 1000 Families Study in addition to other family focused research at the University of Warwick. The case for this study was the lack of a large scale (several hundred families or larger) research sample of families of children with intellectual disability in the UK established explicitly with longitudinal research opportunities in mind. Both of these features were important: a large sample would mean that unique research questions could be asked adopting some of the latest statistical modelling techniques; and the ability to follow families over time would mean that scientific issues of causality could be addressed. Establishing causality is significant since the focus of our family research programme is to understand how children with intellectual disability may affect their family members' well-being, and also how family members affect the mental health and well-being of children with intellectual disability. To examine such questions requires data collected over time.

The Warwick team, with the assistance especially of Cerebra, successfully recruited 1,013 families of children aged 4-15 years by December 2017 (1000 Families Study Wave 1). Wave 2 of the study involved the follow-up approximately 2.5 years later of the families whose child with intellectual disability was still under 16 years of age. In total, 650 families were retained in the study at Wave 2 (59.7% of the total sample; 70.1% of the families eligible for follow-up at Wave 2 – that is, those whose child was under 16 and had consented to be invited at Wave 2).

Additional funding was made available by Cerebra to support a second follow-up of the study (Wave 3) during 2020-2022. We proposed to follow up families approximately 1.5-2 years after their participation in Wave 2 of the study, and to continue to use the data gathered from the first two waves of the study to produce high quality scientific outputs and other dissemination outputs in a variety of formats for families.

### ***Why the 1000 Families Study is important***

One of the biggest influences on the development of *all children* is their family. Children with brain conditions are no different to all other children in this respect – their development is also affected by what happens within their family. The context of the family affects all children's development in at least three ways:

1. Parents and carers approach parenting in different ways, and how parents raise their children can affect how children develop
2. The economic context in which families live has a big impact on children's development. This includes poverty, parents being out of work, and factors such as the quality of the housing in which families live
3. Parents and other family members who are under significant stress may affect the well-being of children within the family

Within this context, the research carried out by the team at Warwick focuses on three big issues for families of children with brain conditions, specifically children with a learning disability and/or autism:

- **Exploring whether the same family factors that influence the development of all children affect the development of children with learning disability and/or autism.** The *Cerebra 1,000 Families Study* is designed to contribute to this area of research as we can examine these families over time to generate new research findings that can be used to improve family life.
- **How can parents and other family members can be supported to maximize positive developmental outcomes for their children with learning disability and/or autism?** We address this issue by developing and/or testing interventions that can give parents key skills to be more effective in their task of raising children with learning disability/autism. We also develop and/or test interventions designed to directly improve parents' well-being and mental health. Findings from the *Cerebra 1,000 Families Study* are used to inform the development of new supports and interventions for families.
- **Attempting to redress the balance for families of children with learning disability and/or autism.** Parents and other family members of children with learning disability/autism are typically portrayed as suffering significant stress when raising their children. This is perhaps true, but misses the fact that raising a child with learning disability/autism is also a positive experience and results in many rewards for families. In the *Cerebra 1,000 Families Study*, we explicitly study family members' (especially parents') positive perceptions and experiences when raising children with learning disability/autism; in addition to recognizing the challenges.

### **Progress summary - January 2020 to December 2022**

In the funding period for the third wave of the 1000 Families Study, we:

- Revised the 1000 Families Survey to include questions for the first time relating to grandparent support and grandparent relationships – creating a unique opportunity to explore extended family research questions
- Obtained full research ethics approval (from the Humanities and Social Sciences Research Ethics Committee, University of Warwick) to carry out the Wave 3 follow-up. This included versions of the survey for families of children below and above the age of 16 years at the time of the Wave 3 follow-up (for Wave 2 we only had ethical approval to follow-up children who were still <16 years of age at Wave 2)
- Carried out the Wave 3 follow-up, receiving a total of responses from 538 families (79.6% of the eligible sample – those who had given us permission to contact them about a third wave of the study)
- Continued to produce research based on Wave 1 of the study
- Began to carry out research drawing on Waves 1 and 2 of the study (see Appendix 2)
- **Published, or had accepted for publication, 11 peer review scientific journal papers** drawing on 1000 Families Study data (see Appendix 1)
- Used £33k funding from the Cerebra grant (as originally proposed) to leverage an additional £35k+ from the University of Warwick for a collaborative full time three-year PhD scholarship (to which Caitlin Williams was appointed, and started in October 2020)
- Gave more than 10 talks/presentations drawing on 1000 Families Study data, mainly to audiences including family members and/or professionals, and including to audiences in Canada and India as well as the UK

- Produced a further other outputs about our family research, typically referencing the 1000 Families Study, including regular newsletters, and articles and videos produced with Cerebra and available through the Cerebra website
- Obtained research ethics approval to carry out a **1000 Families Study – Fathers** project, approaching Wave 3 respondents to engage fathers in the families in a version of the survey (this started following completion of Wave 3 in 2022 and will carry on into 2023; managed by Caitlin Williams via her continuing PhD scholarship)

### ***Continuing use of the 1000 Families Study data***

A key rationale for the 1000 Families Study is that the data will continue to have uses over time, and might enable collaborations with other researchers to lead to new scientific findings. This use of the study data is of three types.

First, research teams in the UK, Canada, and Australia have been adding some of the same measures as those we have in 1000 Families to their own research studies with the idea that this may create some additional opportunities for collaborative research (especially comparing UK data and data from other countries).

Second, we are creating more active links with other studies. We have been working with a researcher in Jordan to create a replication of the 1000 Families Study there. The survey has been designed and some measures have been translated into Arabic, and the study is being submitted for ethical approval. We have also been approached by a team funded by NIHR in the UK to study childhood to adolescent trajectories in behavioural and emotional problems in autistic children. This research is looking to combine data across multiple international longitudinal studies, and we are hopeful that 1000 Families Study data will be included.

Third, the data from all three waves of the 1000 Families Study have been or are being used by PhD students and to support additional scientific outputs. Our research team are focusing on positively-orientated research questions (i.e., about positive outcomes for family members) and on family systems questions (how family members influence each other's well-being). Eight current projects, and their status, are summarised in Appendix 2. We have also begun to embrace Open Science principles (e.g., to increase transparency and the replicability of findings) by pre-specifying and registering planned analyses of the 1000 Families Study data; typically using the Open Science Framework. We will continue to develop new analyses/projects using data from the study.

## Appendix 1. 1000 Families Study Publications

1. Bailey, T., Hastings, R. P., & Totsika, V. (2021). COVID-19 impact on psychological outcomes of parents, siblings and children with intellectual disability: longitudinal before and during lockdown design. *Journal of Intellectual Disability Research*, **65**, 397-404.
2. Hastings, R. P., Totsika, V., Hayden, N. K., Murray, C. A., Jess, M., Langley, E., & Margetson, J. (2020). 1,000 Families Study, a UK multi-wave cohort investigating the wellbeing of families of children with intellectual disabilities: Cohort Profile. *BMJ Open*, **10**:e032919.
3. Hayden, N. K., Hastings, R. P., & Bailey, T. (in press). Behavioural adjustment of children with intellectual disability and their sibling is associated with their sibling relationship quality. *Journal of Intellectual Disability Research*.
4. Jess, M., Bailey, T., Pit-ten Cate, I., Totsika, V., & Hastings, R. P. (2020). Measurement invariance of the Positive Gains Scale in families of children with and without disabilities. *Research in Developmental Disabilities*, **103**, 103662.
5. Jess, M., Flynn, S., Bailey, T., Hastings, R. P. & Totsika, V. (2021). Failure to replicate a robust Down syndrome advantage for maternal well-being. *Journal of Intellectual Disability Research*, **65**, 262-271.
6. Langley, E., Totsika, V., Hastings, R. P., & Bailey, T. (2021). Family relationships and their associations with perceptions of family functioning in mothers of children with intellectual disability. *American Journal on Intellectual and Developmental Disabilities*, **126**, 187-202.
7. Murray, C. A., Hastings, R. P., & Totsika, V. (2021). Clinical utility of the parent-report Strengths and Difficulties Questionnaire as a screen for emotional and behavioural difficulties in children and young people with intellectual disability. *British Journal of Psychiatry*, **218**, 323-325.
8. Rixon, L., Hastings, R. P., Kovshoff, H., & Bailey, T. (2022). Outcomes for siblings associated with sub-groups of autistic children with intellectual disability identified by latent profile analysis. *Research in Developmental Disabilities*, **130**, 104337.
9. Stanford, C. E., Hastings, R. P., Riby, D. M., Archer, H. J., Page, S. E. & Cebula, K. (2022). Psychological distress and positive gain in mothers of children with autism, with or without other children with neurodevelopmental disorders. *International Journal of Developmental Disabilities*, **68**, 479-484.
10. Williams, C., Bailey, T., & Hastings, R. P. (2022). Modelling triadic relationships in families of children with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities*, **35**, 843-855.
11. Zabidi, A. S., Hastings, R. P., & Totsika, V. (2023). Spending leisure time together: Parent-child relationship quality in families of children with an intellectual disability. *Research in Developmental Disabilities*, **133**, 104398.

## Appendix 2. 1000 Families Study scientific papers drafted or in active development

<i>Research team</i>	<i>Data use</i>	<i>Status</i>
Daniel Sutherland with Richard Hastings, Samantha Flynn, and Jo Griffin.	Investigating whether positive family functioning protects against the negative effect of child behavioral and emotional difficulties on parental mental health - using three waves of 1000 Families data.	Study currently being designed and prepared for ethics review
Caitlin Williams, with Paul Thompson and Richard Hastings	Evaluating the Family Stress Model in a sample of families of children with intellectual disabilities: A replication study using data from all three waves of the 1000 Families Study ( $n=360$ )	Pre-registration on the Open Science Framework (OSF) in progress
Caitlin Williams, with Paul Thompson, Nikita Hayden, and Richard Hastings	Associations between sibling relationship quality and prosocial behaviors in families of children with developmental disabilities: Cross-lagged analysis using data from Wave 1 and Wave 2 of the 1000 Families Study ( $n=297$ )	Analysis completed and paper drafted, being revised for submission to a peer review journal. Pre-registered ( <a href="https://osf.io/u2qnm/">https://osf.io/u2qnm/</a> )
Caitlin Williams, with Paul Thompson and Richard Hastings	Cross-lagged associations between parent relationship satisfaction, and sibling relationship quality in families of children with developmental disabilities- using data from Wave 1 and Wave 2 of the 1000 Families Study ( $n=223$ )	Analysis completed and paper drafted, being revised for submission to a peer review journal. Pre-registered ( <a href="https://osf.io/xz7bg/">https://osf.io/xz7bg/</a> )
Emma Langley, with Richard Hastings, Vaso Totsika and Tom Bailey	A rare study focusing on the single parent families in the 1000 Families Study dataset – examining child, demographic and family predictors of the well-being of single mothers of children with intellectual disability ( $n=213$ )	Included in Emma's PhD thesis (PhD viva passed in December 2018). Paper drafted for publication
Jane Margetson (Cerebra), with Richard Hastings, Vaso Totsika and Tom Bailey	Associations between parenting behaviours and parent-child relationship quality and the mental health and well-being of children with intellectual disability – using all of the 1000 Families Study sample ( $n=606$ )	Analysis completed and paper drafted, being revised for submission to a peer review journal
Caitlin Murray, with Richard Hastings and Vaso Totsika	Associations between parental wellbeing and child internalizing and externalizing difficulties: Cross-lagged analysis using data from Wave 1 and Wave 2 of the 1000 Families Study ( $n=650$ ).	Analysis completed and paper drafted, being revised for submission to a peer review journal
Caitlin Murray, with Vaso Totsika, Richard Hastings, Kylie Gray, and Tom Bailey	Anxiety in children with intellectual disabilities with and without autism using Wave 1 data, looking at associations with family poverty, parental distress, and child anxiety ( $n=648$ )	Analysis completed and paper drafted, being revised for submission to a peer review journal