CoIN Study
Christmas Newsletter 2020

Hello everyone!

The CoIN Study has now been running for 7 months. As we approach the Christmas holidays, we would like to update you with our progress so far as well as tell you about our plans for the future. The CoIN Study aims to identify specific challenges facing families of children with rare neurodevelopmental and genetic (hereon 'neurogenetic') disorders, track changes in child behaviour, and examine associations between these and parental mental health during the Covid-19 pandemic. Parents/carers (hereon 'parent') of children with rare neurogenetic disorders are invited to complete a baseline and monthly follow-up survey, asking about family life and relationships, access to healthcare and education, overall health and wellbeing, child behaviour and coping during the Covid-19 pandemic. Information collected in the CoIN Study will be used to identify and provide better support to families of children with rare neurogenetic disorders now and in the future.

What have we been doing?

Since the launch of the CoIN Study in May, 219 families have completed the baseline survey. We really appreciate all your responses and continued support of the CoIN Study. We have started to take a look at these responses, which you can read more about below. We have also developed Community Feedback and Parent/carer Interview sub-studies.

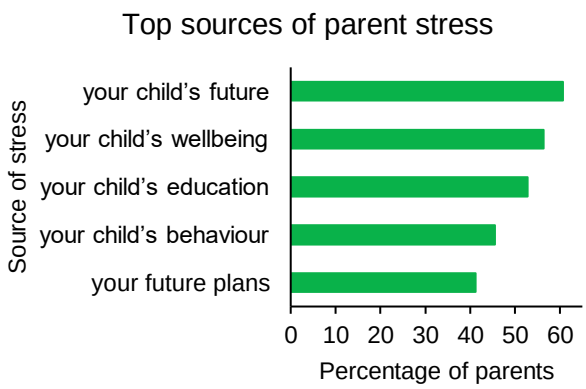
Community Feedback invites members of the CoIN Study community to review the study survey and answer a short one-off questionnaire about the content of the survey and their experience completing the survey. We have now finished collecting responses from 14 parents and our initial review reveals the majority of parents had a positive experience completing the CoIN Study survey. For example, 100% of parents reported the survey was easy to use, follow and understand (i.e., responded 'Fairly easy' or 'Very easy') and 85% of parents found the survey length agreeable. Further, some parents reported the survey helped them 'understand what [they] were feeling' and 'reflect on how [their] family was coping'. These responses will be further analysed in the new year and used to improve the final study survey to capture the priorities of families.

Parent/carer Interviews invite members of the CoIN Study community to complete a one-off 40-minute interview over Zoom about their experiences of the Covid-19 crisis. Interviews facilitate collection of rich in-depth information which will complement survey responses. They also offer the opportunity for parents to discuss experiences, positive or negative, that might not be captured by the study surveys. Alike responses from Community Feedback, responses from Parent/carer Interviews will also be used to improve the final study survey to capture the priorities of families. If you are interested in taking part in an interview, please see our website for more information and how to take part (https://www.coinstudy.co.uk/take-part).

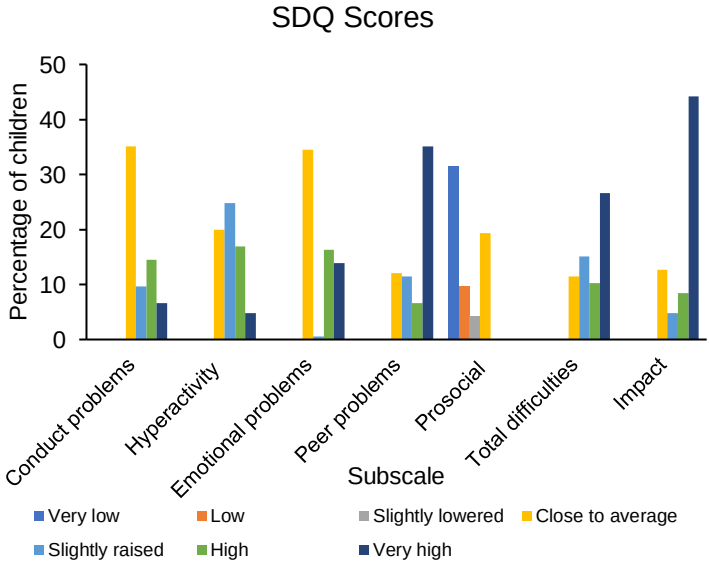
What have we found so far?

We have taken an initial look at some of the information collected in our baseline survey, to give you an idea of the experiences of families of children with rare neurogenetic disorders. We will be working on further analyses in the new year and preparing findings for circulation to the community and submission to academic journals.

Parent worries: We assessed parent worries by asking parents how stressed they had felt over the previous week in response to several potential stressors. We found the most significant worry for parents was their child's future (60.61%), followed by their child's wellbeing (56.36%), education (52.73%) and behaviour (45.45%) and their own future plans (41.21%). These stressors vary from those reported by parents in the Covid-19: Supporting Parents, Adolescents and Children during Epidemics (Co-SPACE) Study, a national study of how families in the general population are coping in response to the Covid-19 pandemic and how parents can support their child's mental health. They found parent/carers top five sources of stress (between 30th March and 4th April 2020) related to work, their child's wellbeing, education and screen-time, and family/friends outside of the household. In contrast to parents in the Co-SPACE Study, the top stressors reported by parents of children with rare neurogenetic disorders focused on their child and their future.



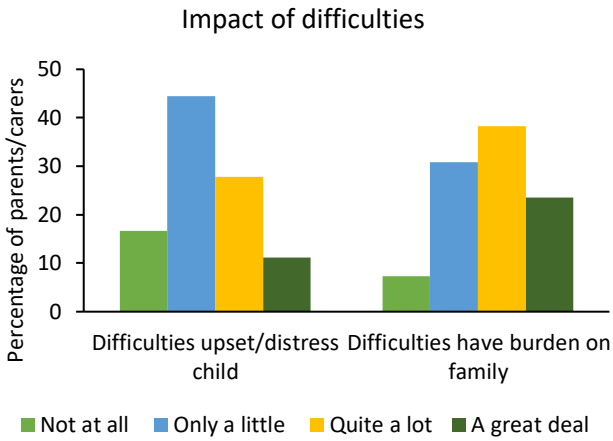
Child behaviour: We assessed child behaviour using a standardised assessment called the Strengths and Difficulties Questionnaire (SDQ). We found the majority of children did not experience significant difficulties (i.e., scored 'Close to average' and 'Slightly raised') in comparison to the percentage of children who experienced significant difficulties (i.e., scored 'High', and 'Very high') on the Conduct problems (44.85% vs. 21.21%) , Hyperactivity (44.85% vs. 21.28%) and Emotional problems (35.15% vs. 30.30%) subscales. In contrast, the majority of children experienced significant difficulties (i.e., scored 'High' and 'Very high'/'Low' and



Note: The Prosocial subscale is reverse scored, such that it is scored from 'Close to average', 'Slightly lowered', 'Low', to 'Very low', whereas the other subscales are scored from 'Close to average', 'Slightly raised', 'High', and 'Very high'. 'Very low' reflects more difficulties on the Prosocial subscale, whereas 'Very high' reflects more difficulties on the other subscales.

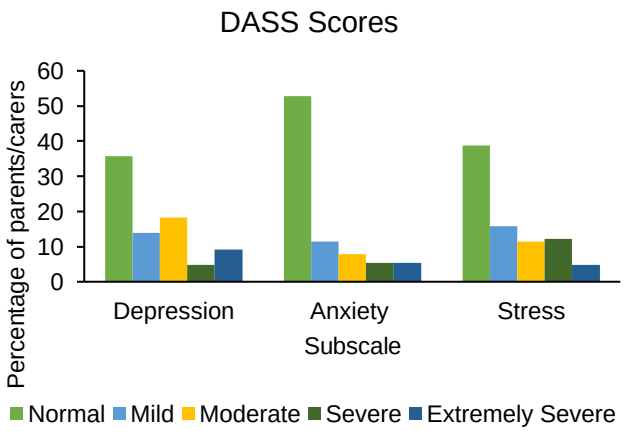
'Very low') versus children who did not experience significant difficulties (i.e., scored Close to average' and 'Slightly raised'/ Slightly lowered') on the Peer problems (41.82% vs. 23.64%) and Prosocial (42.21% vs. 23.64%)) subscales. In total, a greater percentage of children (36.97%) experienced significant difficulties (i.e., scored 'High' and 'Very high' on the Total difficulties score) compared to those that did not (26.67%; i.e., scored 'Low' and 'Very low' on the Total difficulties score).The impact of these difficulties on a child's life was 'Very high' in 44.24% of children, such that it would cause significant stress or impairment.

The SDQ also specifically distinguishes the extent the impact of a child's difficulties causes them to be upset and distressed and their burden on the family as a whole. We found the extent child difficulties impacted the child (i.e., whether they were significantly upset and/or distressed by them) was less significant than the burden they had on the family (38.98% responded 'Quite a lot' and 'A great deal' vs. 61.76%).



Recently, the SDQ was also used by the Office for National Statistics (ONS) to generate the Mental Health of Children and Young People (MHCYP) in England 2020 report. When examining average scores of children aged 5-16 years old (surveyed in July 2020), they found the average score on all of the SDQ subscales fell in the 'Close to average' range, as well as the Total difficulties and Impact scores. This differs to children in the CoIN community, who experience elevated difficulties in peer interactions and executing prosocial behaviours as well as total difficulties. Difficulties also have a significant impact on children's abilities to function and stress in the CoIN community, as well as having an increased burden on the family. This implies children, with rare neurogenetic disorders and their families require extra support alleviating the burden of child difficulties on the family during an unprecedented period such as the Covid-19 pandemic.

Parent mental health: We assessed parent mental health using a standardised assessment of depression, anxiety and stress called the Depression, Anxiety and Stress Scales (DASS-21). We found most parents scored in the 'Normal' range on all three subscales (Depression, 35.67%; Anxiety, 52.73%; Stress, 46.72%). In addition, we found a greater proportion of parents scored in the 'Severe' and 'Extremely Severe' range for Stress (20.44%) than Depression (17.04%) or Anxiety (13.14%). In contrast, general population studies exploring the rate of these mental health conditions during Covid-19 find a greater proportion of adults report severe or



extremely severe levels in depression, followed by anxiety then stress. Additionally,, the rates of all three are slightly elevated in our sample versus the general population. Specific changes during the pandemic might have cause elevated stress in parents of children with rare neurogenetic disorders versus adults in the general population. We will explore these specific factors to identify areas that can be targeted now and in the future to reduce experiences of extreme stress in parents .

What happens next?

We will be sending out our penultimate follow-up survey in February 2021 and our final survey in April 2021. Our final survey will include questions informed by the Community Feedback and Parent/carer Interview sub-studies to ensure we are capturing the priorities of families. Once we have finished collecting responses, we will hold focus groups and/or workshops with parents to discuss our research findings and the best way to share these findings with families.

News from the core CoIN Study team

Since launching in May, two new members have joined the core CoIN Study team. They will also be joined by an additional member (a post-doctoral researcher) in spring 2021. To keep you up to date, here is some information about the core study team and what everyone is up to now.

Abby is now in her final year as a medical student at St. George's, University of London and hopes to graduate and begin her career as a doctor in August 2021. She also works part-time as a research assistant at King's College London and was instrumental in setting up the CoIN Study and the recruitment of families in the first national lockdown.

Katie is an undergraduate placement student from the University of Plymouth where she is studying for a BSc in Psychology. Recently, Katie has been running the CoIN Study social media accounts and our fantastic website. Check out our website here (www.coinstudy.co.uk) for more information about the study and our extensive list of resources. Katie is hoping to continue working with members of the CoIN Study by exploring data collected in the Early Development in Tuberous Sclerosis study (EDiTS), supervised by Dr Tye, for her final year research project at Plymouth. Looking forward to seeing your findings, Katie!

Jess is a research assistant on the CoIN Study and is currently coordinating the Community Feedback and Interview sub-studies. She has also generated short digests of descriptive data from the baseline surveys, which will be published on our website in the New Year. Before joining KCL, she obtained a BA in Experimental Psychology from the University of Oxford. She wishes to continue to pursue a career in academia and has applied for PhDs in developmental psychology. Good luck Jess!

Finally, **Charlotte** is a research fellow and the principal investigator on the CoIN Study. Charlotte's research involves exploring predictors and correlates of neurodevelopmental conditions, like autism and ADHD, in children with rare neurogenetic disorders and epilepsy. This includes the EDiTS and Brain development in Early Epilepsy (BEE) studies. EDiTS aims to chart development in infants with tuberous sclerosis complex (TSC) in comparison to infants without TSC, in order to identify features of development that predict behavioural outcomes later on in life. BEE is a new study, which aims to track the development of autism in infants with epilepsy. These conditions commonly co-occur with

great impact on quality of life of the individual. To find out more about either of these studies contact Charlotte at charlotte.tye@kcl.ac.uk.

CoIN Study Team Members

Abigail Runicles | Katie Robertson | Jessica Martin | Charlotte Tye

Thank you!

With best wishes,

Dr Charlotte Tye & the CoIN Study team

Contact us: coinstudy@kcl.ac.uk