Institution: University of Birmingham		
Unit of Assessment: 4 – Psychology, Psychiatry and Neuroscience		
Title of case study: Improving the mental health and challenging behaviour of people with neurodevelopmental disorders and severe intellectual disability		
Period when the underpinning research was undertaken: 2003–2020		
Details of staff conducting the underpinning research from the submitting unit:		
Name(s):	**Role(s) (e.g. job title):**	**Period(s) employed by submitting HEI:**
Prof. Chris Oliver	Professor of Neuro-developmental Disorders	1993–December 2020
Dr Kate Woodcock	Senior Lecturer	2008–present
Dr Caroline Richards	Senior Lecturer	2015–present
Period when the claimed impact occurred: 2013–2020		
Is this case study continued from a case study submitted in 2014? No		

1. Summary of the impact

The CEREBRA Centre for Neurodevelopmental Disorders at the University of Birmingham translates latest research findings into effective, practical assessments and interventions for people with severe intellectual disability. Their approaches **have been added to national clinical guidelines**, resulting in the widespread application of **new clinical assessment tools** in 83 NHS trusts and in 24 other countries. CEREBRA has improved awareness of rare disorders and has demonstrably **changed management practices** across the world, providing information for families and healthcare professionals in over 200 countries. In the UK alone, over 2,000 families have received advice and support through assessments, support groups, workshops and nationally commended lay guides.

2. Underpinning research

The goal of Oliver's programme of research is to improve the understanding, clinical assessment and effective treatment of psychological disorders in children and adults with severe intellectual disability associated with a diverse set of neurodevelopmental disorders and genetic syndromes. These collectively cause severe psychological and behavioural problems in about 21,000 children and up to 47,000 adults in the UK.

Professor Oliver's **research team has delineated many unique behavioural, cognitive and neuropsychological profiles in children and adults with severe intellectual disability** [R1, R2, R3, R4]. They have identified relationships between cognitive impairments, physical health conditions, environmental influences and specific genetic disorders. This has led to improved understanding and assessment of the causes of clinically significant behaviours, as well as recommendations for treatment. These behaviours include severe self-injurious behaviour and temper outbursts, as well as mental health conditions such as anxiety [R5]. Additionally, the research has established the risk of development of psychological disorder, based on an individual's characteristics such as genetic disorder or specific behavioural markers [R1, R3, R4 R5].

Many standard clinical assessments of behaviour and emotional states cannot be used in practice with people who have severe intellectual disability: the tests are imprecise with regard to assessing their behaviour, may require self-reporting that such participants cannot provide or may refer to situations that are unlikely to be experienced by people with severe intellectual disability. To fill this gap, measures of behaviour disorders (self-injury, aggression), mood, dimensions of Attention Deficit and Hyperactivity Disorder, and repetitive and social behaviours with robust psychometric properties have been developed for use in clinical practice [R6]. The team then develop and apply behavioural and cognitive assessments for these groups of individuals [R6].

The study of genetic disorders has enhanced understanding of the atypical presentation of clinical disorders in people with severe intellectual disability. For example, autism is common in individuals with rare genetic syndromes but the variable profile of their symptoms can impede efficient diagnosis. Through their application of standardised autism assessments in samples with homogenous genetic cause of intellectual disability, **Oliver's team has contributed to both causal theoretical models of idiopathic autism and contributed knowledge about the differing profiles associated with specific genetic disorders** [R4]. Similarly, longitudinal studies of the natural course of clinically significant behaviours like self-injury have identified behavioural markers, such as repetitive behaviour and impulsivity, that predict both the onset and chronicity of self-injury [R3, R5].

In summary, the key research findings [KF] from Oliver's team are:

KF1: Provision of precise descriptions of the psychological characteristics and disorders experienced by people with genetic disorders and severe intellectual disability [R2, R5];

KF2: Identification of the person characteristics that elevate the risk of developing rare disorders [R1, R3, R4, R5];

KF3: The development of assessments of affect and behaviour that can be used in clinical settings [R2, R6];

KF4: The development of causal models of the disorders to inform targeted intervention [R4].

3. References to the research

[R1] McClintock, K., Hall, S., & Oliver, C. (2003) Risk markers associated with challenging behaviours in people with intellectual disabilities: A meta-analytic study. *Journal of Intellectual Disability Research*, 47(6): 405–416. DOI: 10.1046/j.1365-2788.2003.00517.x

This output has been cited 397 times (Scopus, 26th February 2021), the journal is in the 2nd quartile (CiteScore Rank 2019).This output is referenced in sources **S2** and **S3**.

[R2] Adams, D., & Oliver, C. (2010) The relationship between acquired impairments of executive function and behaviour change in adults with Down syndrome. *Journal of Intellectual Disability Research*, 54(5): 393–405. DOI: 10.1111/j.1365-2788.2010.01271.x

[R3] Richards, C., Oliver, C., Nelson, L., & Moss, J. (2012) Self-injurious behaviour in individuals with autism spectrum disorder and intellectual disability. *Journal of Intellectual Disability Research*, 56: 476–89. DOI: 10.1111/j.1365-2788.2012.01537.x

This output has been cited 92 times (Scopus, 26th February 2021) and is referenced in source **S3**.

[**R4]** Richards, C., Jones, C., Moss, J., Groves, L., & Oliver, C. (2015) The prevalence of autism spectrum disorder phenomenology in genetic disorders: A systematic review and meta-analysis. *Lancet Psychiatry*, 2: 909–916. DOI: 10.1016/S2215-0366(15)00376-4

This output has been cited 149 times (Scopus, 26th February 2021) and the journal is ranked number 3 for Psychiatry and Mental Health (CiteScore Rank 2019). It has been selected as one of our UoA outputs for REF2021.

[R5] Arron, K., Oliver, C., Moss, J., Berg, K., & Burbidge, C. (2011) The prevalence and phenomenology of self-injurious and aggressive behaviour in genetic syndromes. *Journal of Intellectual Disability Research*, 55(2): 109–120. DOI: 10.1111/j.1365-2788.2010.01337.x

This output has been cited 144 times (Scopus, 26th February 2021) and is referenced in source **S2**.

[R6]. Oliver, C., Royston, R., Crawford, H., Moss, J., Waite, J., Arron, K., Burbidge, C., Ellis, K., Nelson, L., Ross, E., Russell, H., & Welham, A. (2019) Informant assessments of behaviour and affect for people with intellectual disability.

https://www.researchgate.net/publication/337899095_Informant_assessments_of_behaviour_and_affect_for_people_with_intellectual_disability

This is an open access technical report describing the freely available clinical assessments that can be downloaded after registration on http://findresources.co.uk/professionals/login

Key research grants:

The Cerebra Centre for Neurodevelopmental Disorders (continued funding; Oliver, Centre Director). Cerebra. £1,621,000, January 2008–December 2019.

Understanding and changing sleep disorders in children with developmental disability (Oliver, PI). Cerebra. £350,000, January 2014–December 2019.

Development and preliminary evaluation of a clinical assessment protocol for self-injurious behavior in children with severe intellectual disability and autism spectrum disorder (Oliver, PI). Baily Thomas Charitable Foundation. £111,000, January 2013–December 2014.

Social cognition and its relation to social development in neurodevelopmental disorders (Oliver, PI). The Leverhulme Trust. £206,184, 2012–2015.

4. Details of the impact

CEREBRA is a charity that helps children with brain conditions and their families discover a better life together. The charity co-funds *The CEREBRA Centre for Neurodevelopmental Disorders* at the University of Birmingham to generate impact by translating the latest research findings into effective and practical assessments and interventions. This then enables the Centre to provide information, advice and direct support to parents, carers and practitioners for the ongoing treatment of rare neurodevelopmental disorders. Oliver and his team have impacted on **clinical guidelines** as well as **care and educational practice**. They **have improved outcomes for those who suffer from neurodevelopmental disorders and severe intellectual disability**.

Informing clinical guidelines and practice for rare neurodevelopmental disorders

National clinical guidelines have been informed by research with the 2015 NICE Guidance for Challenging Behaviour [S1] citing 14 research papers published by Oliver and his team between 2001–2013, including R1, R3 and R5. The guidelines are further underpinned by a briefing paper co-authored by Oliver and Richards for the Challenging Behaviour Foundation on risk markers [S2; KF1, KF2]. Specifically, the recommendations on assessment of challenging behaviour provide guidance on the co-occurrence and persistence of characteristics associated with these behaviours. **New clinical assessment tools have also been developed** based on 11 measures developed from KF3 to assess behaviour and effect in children and adults with severe intellectual disability. The tools are freely available online [R6], along with psychometric and normative data for comparison. The measures have now been downloaded by practitioners in 83 NHS Trusts and by approximately 100 intellectual disability services, such as schools and care providers [S3].

Further **national guidelines were also changed** on the assessment and diagnosis of dementia in people with intellectual disabilities, co-produced by the British Psychological Society and Royal College of Psychiatrists. Oliver's evidence [KF2, KF3] of the behavioural indicators of dementia and of the utility of modified neuropsychological assessment of dementia in individuals with Down syndrome is cited [S4].

Guideline changes and the provision of bespoke assessment tools have led **to changes in management practices** for patients with rare neurological disorders across the world. Following the success in the UK, the tools developed by Oliver's team [KF3] have been translated into 11 languages and are used in 23 countries outside the UK. To provide an additional, more detailed,

example, a consensus statement on the diagnosis and management of Cornelia de Lange syndrome published in *Nature Reviews Genetics* (2018) contained 68 detailed recommendations heavily influenced by Oliver's work [KF2, KF4]. The President of the World Federation of Cornelia de Lange Syndrome Support Groups writes:

> It is a testament to the work of Professor Oliver that so much of his research is included and referenced in this prestigious paper. This has become a defining document for management of the syndrome for the 4,242 [Cornelia de Lange Syndrome] people registered with us globally. [S5]

The CEREBRA Centre has also attracted global engagement via their Further Inform Neurogenetic Disorders (FIND) website (www.findresources.co.uk) whose focus is to better disseminate information on rare neurodevelopmental disorders. Google analytics [S3] show it receives an average of 966 visits and 3,666 page-views per month, from 200 visiting countries: 728,000 so far in total and growing rapidly. In house evaluation has also indicated that the website is visited by both clinicians and families. In its first year, 79% of respondents felt that the website had improved their knowledge whereas 95% found the information easy to understand. An NHS Consultant Paediatric Neuropsychiatrist writes:

> The FIND website […] is remarkable in that it includes evidence-based information that is accessible to parents/carers, whilst also including rich information and resources that clinicians (like paediatricians, psychiatrists, psychologists, and occupational therapists) can consult and use in their clinical practice. [S6]

Improved outcomes for patients and their families

The CEREBRA Centre works directly with families and has **provided over 2,000 UK families with personalised feedback reports** using the research-led clinical assessment tools, to detail and aid interpretation of the assessment of their child's performance. These reports, which are based on the findings of KF1–KF4, are highly valued by families to **improve their understanding and to help them educate and advocate with local services and professionals**. The impact of these reports is best evidenced by testimony from the many charities and organisations supported by Oliver and his team [S4, S6, S7, S8]. These include UNIQUE (a charity which focuses on very rare syndromes), the Fragile X Society, the Down's, Cornelia de Lange, Prader-Willi, Angelman, Williams, Lowe, Tuberous Sclerosis, Cri du Chat, Rubinstein-Taybi, Smith-Magenis, Kleefstra, Child Growth Foundations. For these charities, Oliver's team provide assessments and online advice, serve as chairs and members of scientific and clinical advisory committees, run support groups, present national and international workshops, and produce nationally commended lay guides and online materials. Through these charities **CEREBRA often reach a significant proportion of the UK families affected by a particular syndrome** (e.g.~15% for the most common, Fragile X, up to 93% for the very rare Kleefstra syndrome [S10]). The Smith-Magenis Syndrome charity CEO and Chair testifies:

> 10 years ago most families affected by Smith-Magenis syndrome were struggling with unmanageable challenging behaviours, sleep exhaustion and the prospect of their child likely to be sectioned […] The research provided by Chris's team has enabled our SMS population to manage the challenges more aptly with identified practical solutions, bringing about greater optimism and hope for the future. [S6]

As well as using FIND as an information source, families of patients also use co-authored guides with the Cerebra charity for self-injury, pain, autism, sleep and anxiety [S9]. These guides are primarily designed to translate research findings to **improve wellbeing** for parents and carers, but are also being used by clinicians. The anxiety guide was "highly commended" [S8] and the Sleep tips [S9] was "commended" in the BMA Patient Information Awards [S10] which recognise excellence in accessible, well-designed and clinically balanced patient information. Overall, the Head of Research and Information at the Cerebra Charity reports:

> Through his research, [Oliver] and his team have had a significant impact on the lives of many […] children, and their families. In particular, the University of Birmingham researcher team have written a number of guides that we have produced for parents covering topics that are important to these families. [S10]

5. Sources to corroborate the impact

[S1] NICE guidelines: Challenging behaviour and learning disabilities: prevention and interventions for people with learning disabilities whose behaviour challenges (2015). *National Institute for Health and Care Excellence Guidance: Methods, evidence and recommendations*

These guidelines cite 14 papers from the Oliver team, published between 2001–2013, including R1.

[S2] Briefing Paper Early intervention for children with learning disabilities whose behaviour challenges (2014). *Challenging Behaviour Foundation*

The briefing cites 3 papers from the Oliver team, published 2003–2011, including R1 and R5; Oliver and Richards are co-authors.

[S3] Google analytics usage statistics for FIND website.

[S4] BPS-RCP guidelines: Dementia and People with Intellectual Disabilities: Guidance on the assessment, diagnosis, interventions and support (2015). *The British Psychological Society and Royal College of Psychiatrists*

These guidelines cite 3 papers from the Oliver team, published 2010–2012, including R2.

[S5] Testimonial from the World Federation of Cornelia de Lange syndrome (CdLS) support groups, signed by the President, Vice President, Treasurer and their advisory Council President.

The testimony highlights Oliver's contribution to the 2018 consensus report and the impact of the FIND website for families.

[S6] Testimonial signed by the CEO and the Chairperson of the Board of Trustees, Smith-Magenis Syndrome Foundation UK.

This testimony includes several quotations from clinicians and lay members associated with the Foundation.

[S7] Testimonial from CEO for The Fragile X Society.

[S8] Testimony from the Chair and Trustee of the Kleefstra Syndrome Organization.

[S9] Cerebra guides on Self injurious behaviour in children with intellectual disability (2013); Pain (2015); Anxiety (2015); Autism in genetic disorders (2012); Sleep (2017).

[S10] Testimonial from Head of Research and Information, Cerebra.

This includes details of six lay guides, the commendation by the BMA for three of these, and numbers of downloads of the guides from the Cerebra site (totalling 29,433 in June 2020).