

Journal of Mental Health for Children and Adolescents with Intellectual and Developmental Disabilities: *An Educational Resource*

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Contents

Article	Author	Page
A pilot multidisciplinary clinic at Aspect Autism School by a specialist mental health team	Singhal, D., Dossetor, D., Wong, M. & Butterworth, T.	4
Resources	Editors	12
YAM: Youth Aware of Mental Health	Erin Pilon and Tanya Lancaster	13
Interview with Leanne Dowse from the University of New South Wales	Leanne Dowse	14
Case Report - The Successful Use of ECT in a Youth with Autism, Major Depression, Severe Intellectual Disability and Self-injury.	Wurth, P., McIntyre, S., Ratnayake, P. and Walker, J.	18
Readings	Editors	23
The Medicine Cabinet: Placebo or Nocebo	Judy Longworth and Introduction by David Dossetor	24
Society for the Study of Behaviour Phenotypes (SSBP) 21 st International Research Symposium 2018.	Peter Wurth	30
<i>I have promises to keep</i>	Jackson Karl Schomacker	36

The aim of this Journal is to improve the mental health of children and adolescents with intellectual and developmental disability through enabling academic debate, research and commentary on the field.

Description and purpose

This journal is a vehicle of expertise about mental health information of children and adolescents with intellectual and developmental disability. As a product of CHW School-Link, this journal is supported by School-Link and a collaborative effort with a multi-agency editorial group from the Benevolent Society and NSW Department of Education. We are extremely proud to present these ideas and invite you as authors to help develop this field and the knowledge base to help support children and adolescents.

On our Website:

www.schoollink.chw.edu.au

The website will be playing a crucial role in the information that CHW School-Link can provide to you.

- *The collection of previous and current editions is located there with the ability to download articles separately.*
- *An invitation for contributions can be found on the website with instructions for authors.*
- *Upcoming training at conferences, workshops and other professional development opportunities will be continuously updated.*

Editorial

Jodie Caruana

CHW School-Link Coordinator
The Children's Hospital at Westmead

www.schoollink.chw.edu.au

Dear readers,

School holidays are often a tricky time for some students. Please use this resource from South West Sydney Local Health District School-Link to help those students in need. <https://www.swslhd.health.nsw.gov.au/services/ICAMHS/resources.html>

We are pleased to announce that our webinar series for teachers of students with an intellectual disability has begun. This has been an amazing partnership between CHW School-Link, Network Centre Facilitator - Fairfield and the Benevolent Society. Thankyou Shiraz Patel for initiating this. More information is outlined below.

An important document was released on 6th December 2018 concerning the NDIS in NSW. The Portfolio Committee No.2 Health and Community Services of the Legislative Council of NSW submitted a report from their inquiry into the *Implementation of the National Disability Insurance Scheme and the provision*

of disability services in New South Wales <https://www.parliament.nsw.gov.au/lcdocs/inquiries/2496/Final%20report.pdf> .

The report outlines several issues and outlines 23 recommendations which the government has until 28 Feb 2019 to table a response.

We hope you enjoy this summer edition.

Jodie Caruana

School-Link Coordinator
The Children's Hospital at Westmead.



Webinar Series

Supporting the mental health of students with an intellectual disability

This **FREE** webinar series is brought to you by a partnership between the Networked Specialist Facilitator - Strathfield, The Benevolent Society, and CHW School-Link.

The webinars aim to assist teachers and other school professionals to support the mental health of students with an intellectual disability in the classroom. Topics are being added throughout 2019.

Current Topics include:

- Understanding & responding to behaviour (50 mins)
- Self-regulation (25 mins)

Webinars are available anytime, on demand using this link:

www.schoollink.chw.edu.au/webinar-series/



A pilot multidisciplinary clinic at an ASPECT autism school by a specialist mental health team

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Background

Around 50-70% of children with Autism Spectrum Disorder (ASD) and Intellectual Disability (ID) have at least one mental health diagnosis (AAASD, 2012, and Brereton *et al*, 2006). Many children with ASD and ID have mental health related difficult behaviours, which cannot be explained by ASD or ID. Appropriate psychiatric input is thus needed for diagnosis and management. While there has been a steady increase in the prevalence of autism worldwide (Brereton *et al*, 2006), there is a scarcity of child psychiatrists or paediatricians trained in neurodevelopmental and mental health disorders. This report describes the experience of two interns who were training in the only Neurodevelopmental Child Psychiatry Team in the public health system in NSW, at the Department of Psychological Medicine, at **the Children's Hospital at Westmead, Australia**. This report explores the experience and possible benefit of a multidisciplinary special school-based mental health clinic for children with ASD.

Autism Spectrum Australia (Aspect) is Australia's largest service provider for people on the autism spectrum in Australia and caters to students with co-morbid ID. Their curriculum includes mental wellbeing and most of the children have access to school /community-based services including a paediatrician, speech therapist, psychologist and behaviour management specialists. However, the specialist psychiatrist input is low, reportedly around 6% in children attending an Aspect school in Sydney.

This report follows a year-long monthly multidisciplinary Mental Health clinic pilot at an Aspect school. The clinic provided a comprehensive mental health assessment for the child/young person and the family by a team made up of a dual trained paediatric/child psychiatry fellow and clinical psychologist from the **Developmental Psychiatry Team at the Children's Hospital at Westmead**.

Aim

The clinic aimed to increase collaboration and mental

health awareness between families, educational staff and community-based professionals working with this population whilst providing care for children and young people.

In addition to establishing a collaborative relationship and a multidisciplinary service, this study assessed the following four questions:

1. Do these students have unidentified mental health diagnoses? We describe the new mental health diagnoses made and medications prescribed for these conditions.
2. Can available mental health screening instruments identify co-morbid psychiatric disorder in this population compared with clinical assessment (gold standard)? We trialled 3 screening questionnaires for identifying mental health disorders.
3. Do these cases have associated family problems as assessed by mental health status and parenting style of the parents by clinical assessment and using screening instruments?
4. Is a school-based multidisciplinary clinic felt to be a useful service by the parents and teachers? The usefulness of the clinic was evaluated with the Clinic Satisfaction Scale.

Method

This pilot clinic was set up and managed by a steering committee of senior representatives of Aspect and the Neurodevelopmental Psychiatry Team and ethics approval was obtained by **Aspect and the Children's Hospital at Westmead**.

Cases of concern were selected by the school; approval and involvement was sought from the parents, and a referral from the involved paediatrician (See Figure 1).



Figure 1. Organisational process of a school clinic.

Initially, some information was collected about the young person’s presenting symptoms. Pre-clinic perception of teachers and parents regarding the child or young person’s presenting symptoms table was used (see Appendix 1)

A single comprehensive diagnostic assessment generally took half a day and included: reviewing previous medical and educational health records, interviewing the parents, class teacher, child (if verbal), school counsellor, occupational therapist, speech therapist and observing the child in the classroom and playground. Other information was collected by questionnaires (as listed below). The MH (mental health) team provided verbal and written feedback to both the parents and school staff. The usefulness of the clinic was assessed by the Clinic Satisfaction Scale, informal telephone feedback and feedback from steering committee feedback.

Three MH screening tools were completed by parents and teachers:

- an online DAWBA (Development and Wellbeing Assessment) (Goodman *et al*, 2000),
- DBC (Developmental Behavioural Checklist) (Dekker *et al*, 2002) and
- MHID Screener with 9 Likert Scale questions (developed by The Neurodevelopmental team at the Children’s Hospital at Westmead).

DAWBA was chosen as it is easy to administer, accessible online, low cost and does not require expert input. The package is designed to generate ICD-10 and DSM-IV psychiatric diagnoses on 5-16 year olds. The DBC is widely used in Australia and assesses a broad range of behavioural and emotional disturbance in young people with intellectual or developmental disability between the ages of 4-18 years. The MHID was developed as a quick screener which can be completed in less than a minute. These screening tools were quick, easy to administer and did not require special training. Administering these tools was a way to ex-



“The clinic provided a comprehensive mental health assessment for the child/ young person and the family”

plore whether these questionnaires could provide a ready source of information for the school to identify the presence of co-morbid psychiatric disorder themselves and then decide whether there was a need to refer to specialist child psychiatry services.

The MH team assessment included an assessment of parental relationships and wellbeing and this was supplemented with the DASS-21 (The Depression Anxiety Stress Scale) (Lovibond and Lovibond, 1995) and the 30 questions Parenting Scale from Stepping Stones Triple P (SSTP, 2012).

A Clinic Satisfaction scale (See appendix 2) was used to provide data on what the parents and teachers thought of the clinic, which was modified from the Service Satisfaction Scale (Plapp and Rey, 1994). The process, outcomes and teacher and parent satisfaction were evaluated.

Results

Assessments

8 clinic assessments were completed in the course of a school year, (7 boys, 1 girl, aged 8-12 years). All had new or additional psychiatric diagnoses: 6/8 for sleep disorder, 4/8 with Anxiety Disorder and 2/8 with Atten-

tion Deficit Hyperactivity Disorder (ADHD). All had pharmacological intervention for these disorders (details in Table 1) and were given behaviour management advice.

Medication Recommendations	Number of Patients (out of 8)
Antidepressant/Anxiolytics (Recommended)	6 (SSRI and 4 Amytriptylline)
Stimulant started	2 (Ritalin)
Clonidine	4
Risperidone	1
Increase in medication	2 (Fluoxetine, Risperidone)
Stopped resperidone	1
Melatonin	2

Table 1 Describes the changes in medication management recommended as a result of the clinic.

All medication changes received positive feedback from teaching staff or parents or both when contacted either by phone or verbal feedback. ASPECT staff said there was a decrease in behavioural concerns at school and improved school participation.

Parental Mental Health

Clinical assessments identified parental mental health issues in seven of the eight parents (87.5%). Maternal depression was identified in 5/8 (62.5%), and paternal depression in 1/8 (12.5%). Only one mother was already on antidepressants and was seeing a psychologist.



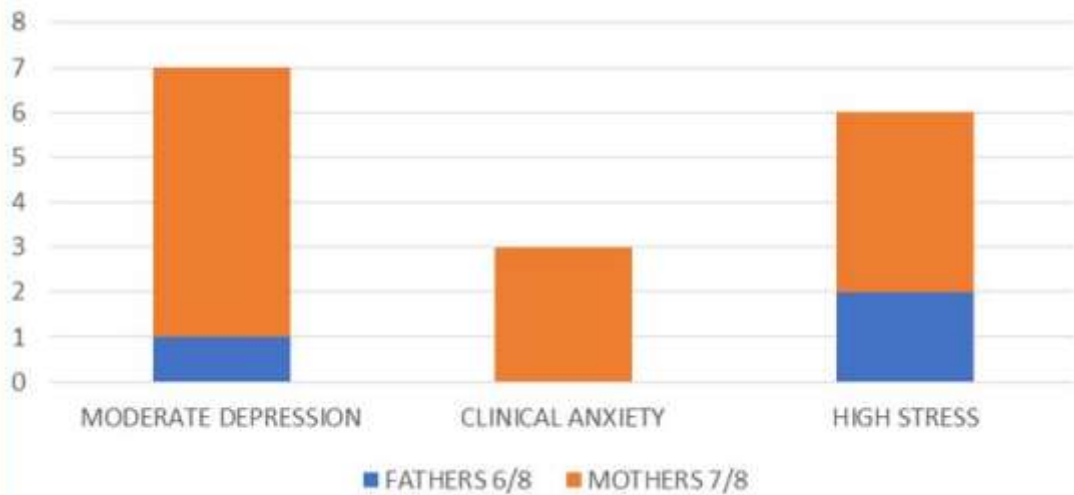
DASS results. The DASS was used to assess parent ill health. 7/8 (87.5 %) mothers and 6/8 (75%) fathers filled the DASS- 21 form. 6/7 (85.7%) mothers scored high for depression and 1/7 (16.6%) scored in the normal range. 1/7 (16.6%) had moderate depression and 5/6 (83.3%) fathers scored in a normal range for depression. 3/7 (42.8 %) mothers scored a clinical level of anxiety. 4/7 (57.1%) mothers and 6/6 (100%) fathers scored normally for anxiety. 4/7 (57.1%) mothers and 2/6 (33.3%) fathers had clinical level of stress. 3/7 (42.8%) mothers had a normal stress level and 4/6 (66.6%) of fathers. Figure 2 displays the results in the clinical range.

The results indicate that the DASS-21 performed well against clinical assessment and was therefore clinically useful.

100% of the completed DAWBA, MHID Screener and **DBC showed ‘high probability’ of mental health or behavioural disorder** showing high sensitivity, which indicates that the school were selecting cases appropriate for a mental health clinic. However, all the tools had their limitations in diagnostic accuracy. DAWBA did not identify ID vs 7/8 clinical assessment (CA), anxiety (vs 4/8 CA) and mood disorder (vs 1/8 CA) and underestimated ASD (37.5% vs 100% in CA).

MHID Screener was good at recognising ASD (100% as CA) and ID (83.3% vs 85% in CA). Both MHID Screener (100%) and DAWBA (37.5%) over-estimated the ODD (vs 25% CA). Both MHID Screener (100%) and DBC (100%) overestimated anxiety vs 50% CA.

Number of Parents in clinical range for Mental Illhealth on DASS-21



“Results indicate that the DASS-21 performed well against clinical assessment”

Figure 2, Number of parents in clinical range for mental ill health on the DASS-21

Parenting Questionnaire

7/8 (87.5%) mothers and 4/8 (50%) fathers answered the Parenting Questionnaire and results showed at least one of the parents who answered the questions had suboptimal style of parenting including laxness, over-reactivity and verbosity. This suggests that greater attention to parenting skills and style may be helpful to the identified clinic children.

Mental Health Screening tools

7/8 (87.5%) teachers and 6/8 (75%) parents completed the online DAWBA, 7/8 (87.5%) parents and 5/8 (62.5%) teachers filled the DBC. 8/8 (100%) parents and 6/8 (75%) teachers filled the MHID Screener.

DAWBA (37.5%), MHID Screener (66.7%) underestimated ADHD vs 75% CA. None of the above screening scales looked specifically for sleep difficulty which needed attention and management in six of the eight children (75%). Accordingly, these screening instruments had high level of under identification and over identification and therefore appear to lack clinical validity in this population. Some of these failings are because the instrument is not designed to identify some diagnoses, such as sleep disorders in all of them and ID in the DAWBA. It is not possible to comment more broadly on the clinical significance of these diagnostic differences due to low numbers, but in general they lacked clinical utility. However, the DAWBA may have value in a mainstream population, but may not be suitable for a population with Autism. The DBC is primarily a behavioural measure and does not serve well as a measure of psychiatric diagnosis. The MHID was of

Comparison of Different Assessment Tools used in a School Clinic at an ASPECT school for kids with Autism

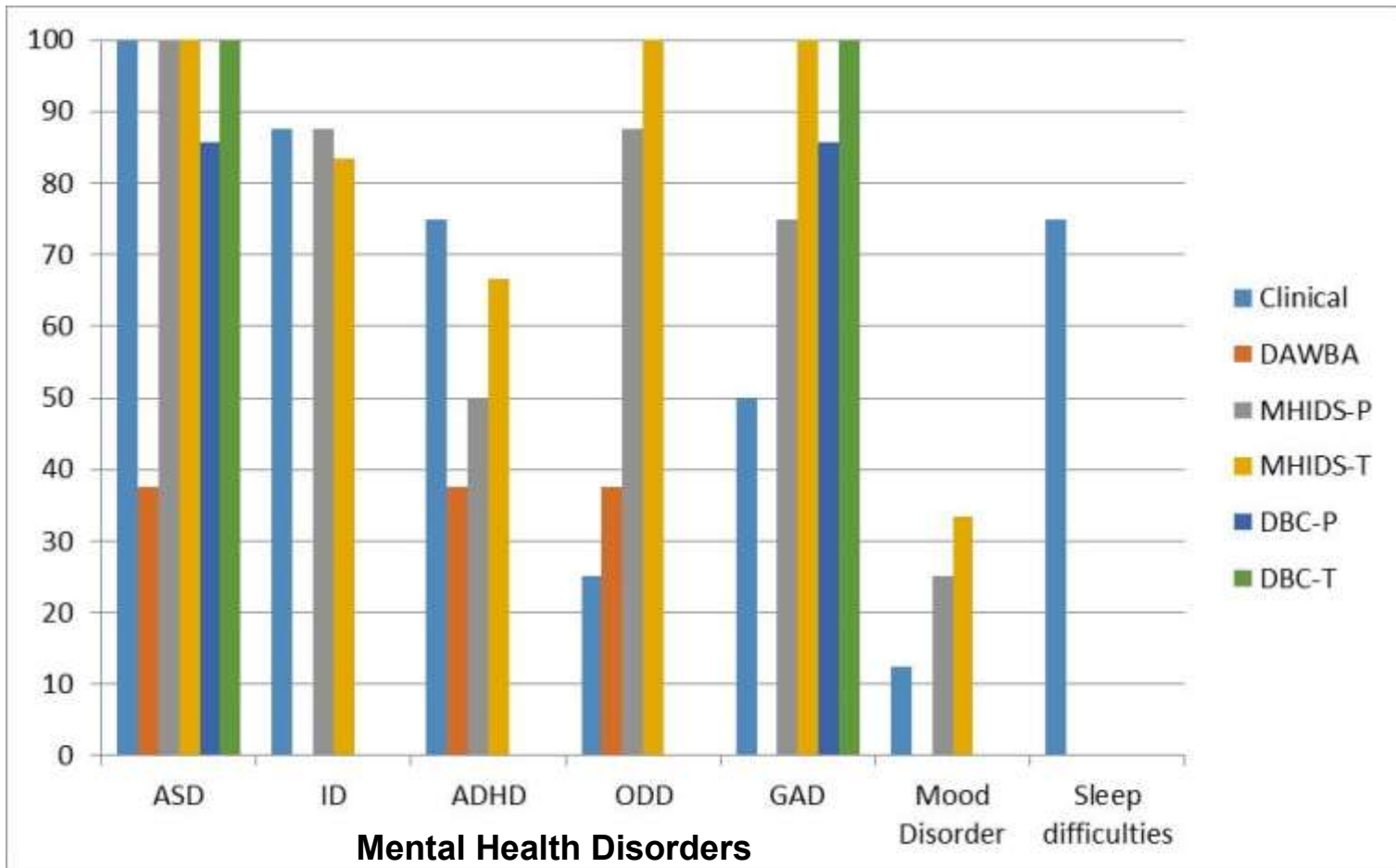


Figure 3: Overview of all assessment tools and the disorders they measured in an ASPECT school

greater convenience in terms of time taken, but needs further development. See figure 3 for an overview of all assessment tools and the disorders they measured.

Clinic Evaluations

5/8 (62.5%) clinic evaluations forms were returned from the parents and the teachers. Of these, 4/5 (80%) of teachers and parents strongly or very strongly agreed that the clinic was useful. 100% of both parents and teachers strongly/very strongly agreed that participation of school in the clinic was useful. 4/5 (80%) parents and 5/5 (100%) teachers agreed that multidisciplinary approach is essential. The Clinic Service Evaluation Questionnaire confirms that parents and teachers valued the Multidisciplinary MH School-based Clinic.

Conclusion

This school-based specialised psychiatry clinic had high levels of satisfaction from parents and teachers, and brought new understanding about troubled children with ASD. These mental disorders were not considered particularly severe but illustrated the importance of co-morbid sleep problems, anxiety and

ADHD not identified by 'treatment as usual' and intervention led to improved outcomes for the child, based on feedback from parents and teaching staff. It highlights the importance of specialised mental health assessment in addition to an educational and behavioural perspective in this population. Hence a multidisciplinary mental health approach is essential for children with ASD/ID and mental health comorbidity.

How can we enable a school-based multidisciplinary mental health approach, when the future of services based on the National Disability Insurance Scheme and Health Activity-Based Funding targets individual disability and mental health practitioners and does not fund collaborative multidisciplinary team work? Currently, mental health services expect clients with Autism or ID to be managed by mainstream mental health services. With the increasing incidence of Autism and the high level of documented mental health co-morbidity, the specialised mental health knowledge of this population is essential for professionals working with them. There is a need for upskilling and education of the professionals including the paediatricians who are the primary medical professionals looking af-

“simple and quick instruments can enable parents and teachers to identify mental health issues in this population ”

ter these children.

There is also a high level of mental health issues (mainly Depression) in parents and suboptimal parenting style was identified in at least one of the parents, most probably due to a lack of adequate community-based support for these children and families. However, the interactions between autism, intellectual disability, parental depression, child's and parents temperament, social support, cultural background, circumstances and parenting style is complex. Currently the **'My Say' project is rolling out universal availability of Stepping Stones Parent Training** which is shown to improve parenting skills, parental wellbeing and childhood behaviour (Gray *et al*, 2017). The School-Link study of Stepping Stones Parent Training in Schools for Special Purposes catering for ID also demonstrated powerful effects. The need to maintain the funding of such parent training is clear and would no doubt greatly benefit special schools. Parental (Carer) mental health, support and wellbeing in this population needs a broad-based public health approach to address identified need of carer burn out.

We found simple and quick (like MHID) instruments can enable parents and teachers to identify mental health issues in this population and facilitate the referral to the neurodevelopmental specialist. From this experience, we have made some modifications: we have omitted psychosis as one of the possible diagnoses due to its rarity. We are interested in adding a question on sleep issues. Further understanding of the relationship between level of disturbance and developmental age, may be important diagnostically, but was difficult for parents and teachers to understand.

We conclude that although screening questionnaires can sensitise parents and staff to mental health problems, there are problems of over and under-identification of problems. There are also diagnostic overshadowing issues. DAWBA showed poor sensitivity to ID, Anxiety, mood disorder, ADHD and ASD and over-identified ODD. DBC tended to over-identify anxiety. Both take significant time to complete and score and neither consider sleep problems. Due to small numbers the results are inconclusive. However, there is a need for an easily available, and simple screening tool



to enable parents and teachers to identify the mental health disorder. This will help people with autism (with or without ID) to get appropriate and quicker specialist services. We have not been able to demonstrate that MH screening instruments for students with ASD have reached acceptable standards, although those for **parents' wellbeing appear to.**

This pilot clinic demonstrates the importance of identifying co-morbid psychiatric disorder in students with ASD. Does this require such multidisciplinary clinics to be established as a means of helping the educational progress of the students and the wellbeing of the families? This is a large funding challenge. Are there other models of multidisciplinary mental health practice that can be supported to give these students and families to flourish? However, currently neither screening instruments, nor community treatment-as-usual are a substitute for a multidisciplinary school-based clinic.

The research is funded by the Department of Psychological Medicine at The Children's Hospital at Westmead. No one received any payment for participating in this research.

References

AAASD. (2012). *The Interface between Autism Spectrum Disorders and Mental Health: The Ways Forward*. Discussion Paper December 2012, Australian Advisory board on Autism Spectrum disorder.

ASPECT. 2018. www.autismspectrum.org.au. Last accessed October 2018.

Brereton, A.V, Tonge, B.J., Einfeld, S.L. (2006). Psychopathology in children and adolescents with autism compared to young people with intellectual disability. *Journal of Autism and Developmental Disorders*. Volume 36, Issue 7, Pages 863-70.

Dekker, M.C., Nunn, R.J., Einfeld, S.E., Tonge, B.J. and Koot, H.M. (2002). Assessing Emotional and Behavioural Problems in Children with Intellectual Disability: Revisiting the Factor Structure of the Developmental Behaviour Checklist. *Journal of Autism and Developmental Disorders*. Volume 32, Issue 6.

Goodman, R., Ford, T., Richards, H., Gatward, R. and Meltzer, H. (2000). The Development and Well-Being Assessment: description and initial validation of an integrated assessment of child and adolescent psychopathology *Journal of Child Psychology and Psychiatry*. Volume 41, Issue 5, pages 645-656.

Gray, K., Tonge, B., Taffe, J., Sanders, M., Sofronoff, K., Einfeld, S. (2017). A community-based parenting intervention for parents of children with a disability: outcomes in the Australian Stepping Stones Triple P Project. *Mental Health Research in Intellectual Disabilities*. Volume 10, Supplement 1, Page 140.

Lovibond, S.H. & Lovibond, P.F. (1995). *Manual for the Depression Anxiety & Stress Scales*. (2nd Ed.) Sydney: Psychology Foundation.

SSTP. (2012). **Practitioner's Kit for Standard Stepping Stones Triple P (includes Practitioner's Manual, and Stepping Stones Triple P Family Workbook)**

Plapp, J. and Rey, J. (1994). Child and adolescent psychiatric services: case audit and patient satisfaction. *Journal of Quality in Clinical Practice* Volume 14, Pages 51-56.

“This pilot clinic demonstrates the importance of identifying co-morbid psychiatric disorder in students with ASD.”



Appendix 1

Pre-clinic perception of teachers and parents regarding the child's/young person's presenting symptoms

(0 =not at all, 1=may be, 2 =likely, 3=agree, 4=strongly agree, 5=definitely)

What do you think is contributing to the child's current presentation?

1	Behavioural problem	0	1	2	3	4	5
2	ADHD	0	1	2	3	4	5
3	Autistic Obsession/ stereotypy	0	1	2	3	4	5
4	Learning difficulty	0	1	2	3	4	5
5	Psychosis	0	1	2	3	4	5
6	Anxiety including social anxiety	0	1	2	3	4	5
7	Mood Difficulty including depression	0	1	2	3	4	5
8	Is his/her behaviour appropriate for the developmental age	0	1	2	3	4	5
9	Is his/her behaviour inappropriate for developmental age	0	1	2	3	4	5

Appendix 2

Clinic Satisfaction scale

Questionnaire asked to the parents and the teachers

We used a 1-5 Likert scale (strongly disagree, disagree, not sure, agree and strongly agree).

The School clinic was useful in helping to meet the needs of my child					
The School Clinic was useful in helping address my family's needs					
The participation of the school staff is useful in the School Clinic					
The school is the best place to hold this clinic					
It is helpful to have school staff, health staff, disability support staff in the same room and working together.					
It is stressful to have a large number of people in the same room during the clinic					
Please indicate whether you feel the following people are essential to attend the clinic, useful but not essential, or you are unsure.					
This clinic provided a service that I couldn't have got elsewhere					
Did you feel that there was sufficient time allocated to discuss all your concerns?					
How can we improve on this service?					

Headspace: Allied Health Professional Skills Module: Developmental Disorders in Young People: 6 hours

<https://www.thinkgp.com.au/education/allied-health-professional-skills-module-developmental-disorders-young-people?start>

This 6 hour online activity is for allied health professionals.

Welcome to the headspace Allied Health Professionals (AHP) Skills Module: Developmental Disorders in Young People. This activity seeks to educate health professionals on issues surrounding the identification and support of young people with developmental disabilities, along with their mental health care. Utilising a number of case studies, ways of offering the best possible evidence-based care for young people in an effective and inclusive manner are explored.

This ALM consists of four sequential parts. Each part will take approximately one and a half hours to complete.



Be You <https://beyou.edu.au/>

Beyond Blue was appointed to lead Be You by the Australian Government in June 2017

The National Mental Health Commission Review of Mental Health Programmes and Services Report of 2014 found that there were multiple initiatives promoting social and emotional health and wellbeing for children and young people across education settings. These Australian Government funded programs included:

Response Ability , KidsMatter Early Childhood, KidsMatter Primary, MindMatters and headspace School Support

All of these initiatives were designed and delivered with the best possible intent, but had the potential to be so much more if they were integrated into one single, national end-to-end education-based program.

This led to the Australian Government's National Support for Child and Youth Mental Health Program, which has the vision to support infants, children and young people to achieve their possible mental health.

This program has two components: the Mental Health in Education Program, focused on the education and training of early learning, primary, secondary and pre-service educators; and the National Workforce Support Program, focused on providing education and training to clinicians and non-clinicians working with children between the ages of 0-12 outside of the education space.

Supporting People with Intellectual Disability at Risk of Self-Harm and Suicide

The 3DN e-learning team are pleased to announce a new module "Supporting People with Intellectual Disability at Risk of Self-Harm and Suicide" is now available. This module will cover:

- Understanding what is meant by self-harm and how it may differ from self-injurious behaviour
- Why self-harm may occur and its impact on people with ID
- Supporting people with ID who self-harm
- Risk factors for suicide in people with ID
- Recognising and responding to signs and symptoms of suicide in people with ID
- Supporting people with ID who feel suicidal
- Supporting people with ID who are affected by suicide
- Self-care when supporting people with ID who are at risk of self-harm and suicide

You can access this module at www.idhealtheducation.edu.au

3DN are continuing to develop new modules, and more modules are scheduled to be released over the next few months. For queries please contact: ldh-ealtheducation@unsw.edu.au

Emerging Minds

The Emerging Minds: National Workforce Centre for Child Mental Health supports professionals and organisations working in the health, community or social sector to better support children and parents/carers and improve the mental health outcomes of children aged 0-12 years.

Through the National Workforce Centre for Child Mental Health, professionals have free access to a national web hub providing online training, implementation and practice support tools and information, webinars, events and the latest evidence and news about infant and child mental health.

These resources are centred around the following principles:

- **Keeping children's mental health in mind.**
- **Strengthening children's resilience.**
- **Supporting children at risk of, or experiencing, mental health issues.**
- **Responding to infants and children who may experience trauma, grief and loss.**

Whether you work with children aged 0-12 years, with adults who are parents/carers, and/or with families, you have a crucial role to play in strengthening infant and child emotional and social wellbeing, and recognising when a child may be at risk of developing mental health issues. To find out more, visit [Emerging Minds](#).

YAM: Youth Aware of Mental Health

Erin Pilon

Psychology Service Workforce Advisor, NSW Department of Education

Tanya Lancaster

Wellbeing and Counselling Services Advisor, NSW Department of Education

The Department of Education is working closely with the Black Dog Institute to support the implementation of the Youth Aware of Mental Health (YAM) program. YAM is being progressively delivered in sites across NSW as part of the LifeSpan whole of community, evidence-based approach to suicide prevention.

YAM is a mental health program for young people aged 14 – 16 years and is a universal program delivered by accredited instructors that are external to the school. YAM uses role play and lectures to promote increased discussion and knowledge about mental health as well as developing emotional intelligence and problem-solving skills. The department is coordinating the delivery of YAM in NSW public schools and has established 16 Head Teacher positions to lead the implementation of YAM.

The Black Dog Institute reviewed Australian and international school programs with the best evidence of reducing suicidal behaviour. Analysis of YAM shows significant improvements in youth mental health by effectively reducing depression, conduct problems, hyperactivity, suicide attempts, severe suicidal ideation and suicide plans. It has also been shown to facilitate healthy lifestyle choices by young people. YAM supports the cognitive, emotional, social and physical wellbeing of students and can contribute to the **school's planned approach to wellbeing**. All Year 9 students are the identified cohort to receive the YAM program. The program is designed to be delivered to groups of up to 30 students.

Student voice is a key component of the program and our young people have actively engaged in the content and have reported they have really valued the opportunity to speak about topics that are important to

them. Having an opportunity to open up, share their points of view in a non-judgemental way allows deep conversations in order to explore their options when faced with a problem. The YAM program highlights the importance of seeking support when it is needed and not having to deal with a problem alone, connecting young people with local and online mental and general health services, as well as specific youth resources in the community.

Instructor feedback indicates that students with mild to moderate levels of intellectual disability have engaged well with the program. YAM is inclusive in its nature because the students' ideas and thoughts form the basis for the role plays and discussions and therefore are relevant to the individual contexts which allows for differentiation and adjustments. The YAM instructor acts as a facilitator, valuing each student's contribution while encouraging discussion among peers which means they learn from each other. As the program is based on dialogue and role play, it minimises the requirement for reading and writing. Furthermore, where required, School Learning Support Officers (SLSOs) familiar to the students are encouraged to participate in the groups to support the engagement of students with additional learning needs. Further information about Youth Aware of Mental Health (YAM) can be accessed at <http://www.y-a-m.org/>

Further information about LifeSpan can be accessed at <https://www.blackdoginstitute.org.au/research/lifespan>.

If you have any queries about [YAM](#) in NSW Department of Education schools, please contact Student Engagement and Interagency Partnerships on 02 9244 5610.



Interview with Leanne Dowse

Leanne Dowse

Professor, Chair in Intellectual Disability Behaviour Support

University of New South Wales



Please tell us about your career to date including your current role.

As a speech therapist working with people with Intellectual Disability during the time of deinstitutionalisation I learned about the need for understanding the clinical as well as the social context of disability. Many changes happened during that time that we were not really prepared for, particularly what it meant for people with intellectual disability to live in the community and what a community needed to be like to be welcoming of them. I moved to a research position at the University of NSW where I worked with the first academics in Australia working around and understanding the social model of disability. Even though we think that is a well-established idea now; it really only **emerged in Australia in the 1990's. Projects that I worked on really took seriously the idea that disability is primarily the social consequence of living in a society that values normalcy above all else. From those beginnings of disability studies in Australia I now hold the Chair in Intellectual Disability Behaviour Support at UNSW, and this year was appointed UNSW's first Professor of Disability Studies.**

What or who were your key influences?

As I have moved through an understanding of disability from a clinical to a more social perspective my biggest influences have been people with disability themselves and their organisations and the experiences they have shared with me through my research. My PHD was about self-advocacy so I worked with self-advocacy groups both in the UK and Australia. I have also had many academic colleagues who have taught me the skills needed to be a good scholar and researcher.

What is the IDBS and its strategic roadmap?

The Intellectual Disability Behaviour Support Program (IDBS) was established under the Chair of Intellectual

Disability Behaviour Support which I hold. It was a NSW Government funded initiative aimed at addressing issues for people who have intellectual disability or cognitive disability and what we think of as complex support needs. People might have complex behavioural issues or they might have dual diagnoses, they might have contact with the criminal justice system, they might have drug and alcohol issues, so the work tries to broaden out what we understand by the intersection of the social and the clinical and intellectual disability. The program of work focuses on **bringing research to policy and practice. I think that's probably the most important work.** People often have an expectation of researchers that we have all the answers to the difficult questions at our finger tips. What we have really understood over the time of designing and moving the program forward is that for research to have an impact on those who need it or **want it, it's got to be a very collaborative process from the word go.**

The work of the IDBS program is focused on **translating and exchanging knowledge that's co-produced.** We work with organisations and through them with people with disabilities and practitioners. Almost all

“The IDBS program is focused on translating and exchanging knowledge that is CO-produced”



of our work has a translational component so we have a lot of guides for practice that we have developed in conjunction with people who will use them. We work with various designers and we have a process of making sure that our final products are usable by its end users. You can see them here <https://www.arts.unsw.edu.au/research/intellectual-disability-behaviour-support-program/resources/>

The strategic roadmap sets out where we see the future direction of research for this group. It shows what needs to change in our systems, in our policies and in our practices so that people with intellectual disability who have complex support needs have a system that properly supports them and a workforce that will be able to provide for those support needs. We see it as an important map in developing knowledge that is based in the lives of people and their experiences rather than what we think is a good idea for research.

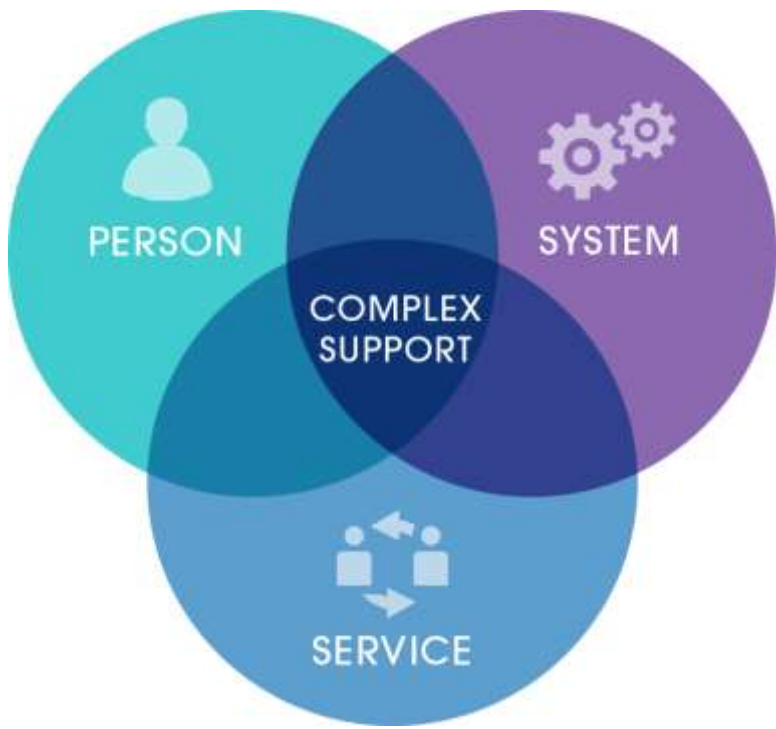
What are some of the challenges you face in your industry?

That's an interesting question - I work in the education industry within the university, and specifically in disability studies and research. I suppose the biggest challenge in the university context is that academics are under a lot of pressure to do all sorts of things and there is a big expectation from the community that academics should produce knowledge that is us-

able and that also it doesn't just use people as subjects of research but actually develops research that is needed by the community. As an academic there is a lot of pressure to publish, to get research money, to teach courses, to do a whole range of things which may not be in line with what the community thinks academics should do. So there is always a real challenge there between what the end users of research expect from researchers and what universities expect from researchers.

How can training help professionals support their clients or students?

Training is an incredibly important aspect of anybody's career development but I would say probably for me, it's not just about training but education. Education is bit of a broader idea where people come to understand not just how to do something or what to do, but *why* and how to evaluate, to challenge and to innovate. I think that's particularly important in disability where we are seeing all sorts of changes in the way that people with disability think about their supports and services. This requires us to think about what education really is. There is so much information around now about everything; you can Google just about anything. Understanding though, why you do something and being able to argue for why something is a good or a bad idea I think is absolutely key to professional practice.



Consultation and partnership is highlighted in your work, why is this important to you, your team and frontline professionals?

We take as our starting point ‘nothing about us, without us’, which is what people with disability demand from research. Our approach is never do any sort of research that doesn’t partner with people whose lived experience is the thing that we are exploring. Morally, we wouldn’t do research unless it was in partnership or very much in consultation with people. I think this has been a political shift; there has been such a long history of exploitation where research often caused damage or distress to people with disability. One of the things that we have to do first for our own professional ethics is to ensure that our work is co-produced and that it is addressing an issue that is important to people with disability as subjects of research.

What is your approach to working with complex patients who have complex support needs and an example of that approach?

We don’t talk about patients as complex; we talk about people who have complex support needs. The one thing I would say that is really important in terms of what informs our work is that we don’t think of the person as being complex. Instead we talk about complex support needs; the reason we do that is because complexity doesn’t reside in the person, a person may have multiple things happening, they might have multiple diagnoses, they may also have circumstantial

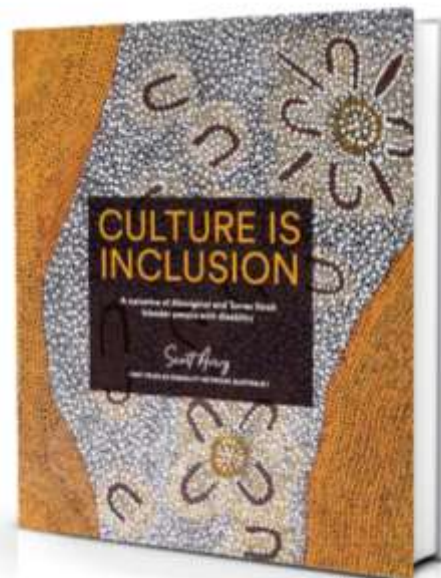
disadvantage, they may have a whole range of issues in their family, they may be socially disconnected and they may have experiences and needs for support which our service system itself is not able to support. When we talk about complex support needs we are really talking about the idea where a person with a set of needs, require support that a service system isn’t equipped to meet. The responsibility is not just on the person to be different, but the responsibility is on the service system to be responsive to the needs that the person has.

Those needs can never just be about impairment or diagnoses; they are always about the person in their social context, in their family context, with all of their strengths as well as the issues they need support with. We use this diagram to show how support needs arise at the intersection of the person; their service environment and the system that shapes what both do.

Image to the left is courtesy of Intellectual Disability Behaviour Support Program UNSW

Favourite book

I read quite broadly, both fiction and non-fiction, and my favourite book is usually whatever I am reading at the moment. Right now that is a most beautiful and important book called ‘Culture is Inclusion: A narrative of Aboriginal and Torres Strait Islander people with disability’ by Scott Avery recently released by First People’s Disability Network. It is a culmination of several of years of intensive work and presents a narra-





tive of Aboriginal and Torres Strait Islander people with a disability. It's a ground breaking book with world first research and I recommend it to anyone with an interest in the experiences of our Aboriginal brothers and sisters with disability. The book is available on the FPDN website. <https://fpdn.org.au/product-category/publications/>

A web link you would like readers to see
IDBS website (image above and link below)
<https://www.arts.unsw.edu.au/idbs>

What do you take on holidays?

Probably not what but who - I always take my partner, I like to holiday in Italy and he can speak Italian.

Intellectual Disability Behaviour Support Program

Program Objectives

The Intellectual Disability Behaviour Support (IDBS) program works to address the research-to-policy-and-practice nexus to improve support for people with cognitive disability and complex support needs. The term 'cognitive disability' includes many labels - including intellectual disability, borderline intellectual disability, acquired brain injury and autism. Generally, having a cognitive disability

means that a person will have difficulty with things such as self-management, decision making and communication and experience some level of social exclusion.

The program achieves this through:

- Consulting with key stakeholders and developing collaborative relationships with academic, government and sector agencies both nationally and internationally;
- Leading the development and delivery of educational programs for frontline and managerial staff to support people with cognitive disability who have complex support needs and behaviours of concern;
- Leading a research program to inform support practices for those with cognitive disability and complex support needs across the disability and community sectors;
- Contributing to policy and practice approaches to cognitive disability support in alignment with international best practice;
- Focusing on areas where there is a specific need to address knowledge deficits within the Australian and international context; and
- Translating knowledge emerging from the IDBS program to ensure the work is informed by, and communicated to, a broad range of stakeholders.

Case Report - The Successful Use of ECT in a Youth with Autism, Major Depression, Severe Intellectual Disability and Self-injury.

Wurth, P., McIntyre, S., Ratnayake, P. and Walker, J.



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ABSTRACT

Objectives:

To report the successful use of electro-convulsive therapy (ECT) in a youth with Autism Spectrum Disorder (ASD), intellectual disability (ID), Major Depression, self-injurious behaviour and epilepsy.

Method:

Clinical case report

Results:

A fifteen year old youth with severe ID, severe ASD and epilepsy presented with an eight year history of treat-

ment for varying levels of withdrawal, anhedonia, sleep disturbance and severe self-injurious behaviour that had worsened considerably over the previous six months. A diagnosis of Major Depression with melancholic features was made. Positive behavioural support interventions and trials of a variety of anti-depressant and antipsychotic medications over an 18 month period produced minimal change. Medication options were limited by the youth's inability to swallow tablets. Major Depression and self-injurious behaviour improved dramatically after nine bilateral ECTs and remained in remission for the next three years.

Conclusion:

This case report adds to the literature supporting the effectiveness and safety of ECT for people with ID. In addition, this case demonstrates that the diagnosis of mental illness can be made in the presence of ID and that ECT should be a readily available treatment option for this population.

KEYWORDS autism, intellectual disability, epilepsy, depression, ECT.

INTRODUCTION

ECT remains an effective treatment for severe depression but there are numerous obstacles to its use, rendering it typically the treatment of last resort (Khalid *et al*, 2008), especially for minors (Walter *et al*, 1997 and Little *et al*, 2002). Despite profound changes over the decades in the manner in which it is administered, with safety and efficacy superior to many medications used in depression, it retains a high degree of stigma (Smith, 2001 and McDonald & Walter, 2013). Its use is very uncommon in children and adolescents, and even more so in those with ID (Collins *et al*, 2012). Literature on the use of ECT in patients with ID is scarce, despite a higher prevalence of psychiatric disorders than the general population (Collins *et al*, 2012). Due to the nature of problems conducting randomised control trials within this population, case reports are the best source of information. The limitations of decision-



making capacity requires a much higher standard of substitute consent, typically by a Mental Health Tribunal, than other treatments (Dare & Rasmussen, 2015).

The diagnosis of mental illness in those with ID, especially in those with limited language, is challenging (Rush *et al*, 2004). Nevertheless, with a suitably modified assessment and appropriate diagnostic criteria, reliable diagnoses can be made (Moss *et al*, 1998). Deliberate self-injury is common in those with ASD, with the prevalence inversely proportional to IQ (Holden & Gitlesen, 2006) and communication skills (Chiang, 2008). The aetiology is poorly understood but multiple factors are usually involved. Non-specialist mental health clinicians will often regard self-injury as a consequence of ID rather than as evidence of a mental health disorder requiring treatment (Reiss *et al*, 1982). In Australia it is commonplace for patients with ID to experience major obstacles to adequate mental health care (Bennett, 2014 and Wurth & Brandon, 2014). Inpatient care in a general psychiatric ward can be problematic. Individuals with ID can be at risk of harm in an acute admission unit, will typically find the environment very stressful, and can challenge the management skills of staff unfamiliar with their needs (Donner *et al*, 2010). In contrast to some other countries, there are no specialised admission units for this population in Australia. For example Scotland, with a population of 5.295 million, had 226 acute inpatients; patients with Learning Disability under the care of psychiatrists specialising in ID at a census in 2014 (TSG,

2015). The area of Lothian, with a population of 800,000 which includes Edinburgh, has a 24 bed specialist admission unit within a general psychiatric hospital (Lyll & Kelly, 2007).

CASE HISTORY

The patient was a fifteen year old male who presented to the ACT Mental Health Service for People with Intellectual Disability (MHS-ID). Autism and significant developmental delay were diagnosed at the age of 2 years and 9 months. He was repeatedly assessed throughout childhood to be functioning within the severe range of ID (IQ 20-35). He was non-verbal and had uniformly low skills on a Vineland Adaptive Behaviour Score at the age of 11. Previously he had appeared happy, and enjoyed activities such as swimming and eating out. A modest degree of self-injury would occur in protest at changes to routine or frustration of expectations.

“His mood and behaviour had deteriorated over six months, with increased withdrawal from preferred activities”

His mood and behaviour had deteriorated over six months, with increased withdrawal from preferred activities, excessive amounts of time in bed hiding under

bed clothes, and severe self-injury by punching himself on the chin or ears. His parents reported that he looked miserable. His sleep and appetite had worsened substantially. He would not even eat his previously favoured fast food. He appeared to his parents to have lost weight, but would not cooperate with being weighed. He would not go to school. He had been treated with risperidone in varying doses for ten years, which had been helpful for chronic insomnia, but with minimal benefit on autistic behaviours, including long term low level self-injury. He was also taking fluoxetine, prescribed for an episode of probable depression when he was eight, with useful benefit at the time. There was a strong family history of depression and anxiety in the families of both parents.

His physical health was good and there was no reported or documented history of epilepsy. His medication was fluoxetine 30 mg daily and risperidone 2.75 mg daily. Physical examination by his general practitioner was limited by severe tactile defensiveness but no abnormalities were detected. Comprehensive blood tests revealed no significant abnormalities. A CGH microarray revealed no significant copy number variations. He had had dental treatment under general anaesthetic the year prior to presentation.

It was initially unclear whether he was suffering from an exacerbation of anxiety secondary to ASD, or a relapse of Major Depression. He did not respond to treatment of anxiety with clonidine up to 50µg tds or propranolol up to 30mgs tds. Further exploration of the history and presentation focussing on behavioural change led to a diagnosis of Major Depression with melancholic features. There was no response to an increase of fluoxetine to 40mg *mane*; or to the combination with mirtazapine up to 90mg daily. There was transient improvement with the replacement of fluoxetine with escitalopram 20mg *mane*, with mirtazapine

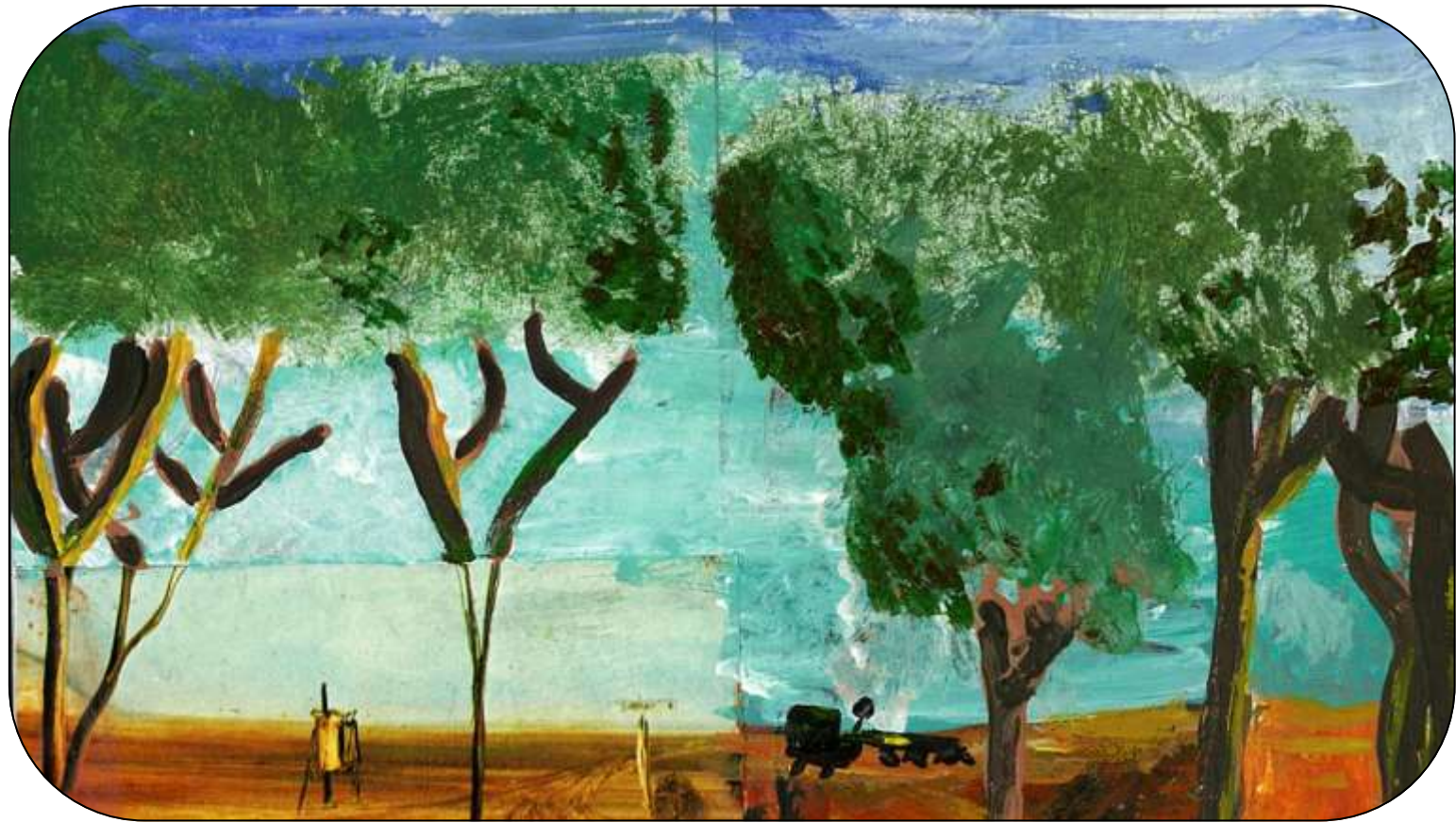
90mg daily and risperidone 2.75mg daily unchanged. Escitalopram was tapered off and venlafaxine 37.5mg *mane* introduced, with the capsule contents sprinkled on food. He then had an epileptic seizure and the family revealed that he had suffered two previously, three and fourteen months beforehand. Given the overwhelming demands of his care, his parents had not sought medical attention for these isolated events. Mirtazapine was reduced to 60mg, but he became progressively more unsettled. After two months treatment venlafaxine was withdrawn and mirtazapine increased to 75mg *nocte*.

After eighteen months, his sleep had improved but self-injury and depression were no better. Treatment options were severely limited by his requirement for liquid medication. He would not accept crushed tablets. He was intermittently constipated, and his mood and behaviour would improve transiently after bowel movements. He had lost 20kg over two years. His medication at this point was mirtazapine 75mg and risperidone 2.75mg daily. Over the years his parents had used a helmet and arm splints to limit the damage he could inflict. When arm splints were removed he would vigorously punch himself in the head and open old wounds. He looked miserable and was irritable and distressed. When he did attend school he stayed under a doona in a corner of the classroom. Otherwise he was at home in bed. He was therefore referred for the second psychiatric opinion required by ACT legislation on ECT to the second author, who concurred with the decision to seek Tribunal consent for ECT.

Obstacles to the delivery of ECT were his age, inability to give informed consent, seizure disorder, potentially disruptive behaviour within the hospital ward and ECT suite, and the challenge of persuading colleagues unfamiliar with ID of the validity of the diagnosis of Major Depression and melancholia and the need for this treatment.

Extensive discussions were held between the MHS-ID team and the hospital mental health and ECT teams about these issues, and about the logistics of administering ECT. Staff from MHS-ID made numerous visits to the inpatient and ECT unit to address the anxiety of medical and nursing staff about the possible negative impact of such a patient on the milieu of a voluntary psychiatric ward. Hospital staff were concerned about

“Treatment options were severely limited by his requirement for liquid medication”



the history of seizures and the risk of status epilepticus. A neurologist recommended ensuring the availability of midazolam in the ECT suite to terminate prolonged seizures. MHS-ID inspected the ward and the ECT suite and identified potential environmental risks specific to this patient. Detailed planning took place, including allocation of roles to specific staff throughout the process of administration of ECT. He was assigned a single room to enable a parent to stay with him at all times.

He was admitted the day prior to ECT, and promptly had a generalised seizure. The neurologist remained supportive of ECT. Physical examination, pathology results and brain CT were normal. Premedication with olanzapine 10mg one hour prior to ECT was administered.

Bilateral ECT was administered, with a successful, slightly prolonged seizure. Following this he was agitated and self-injurious for several hours. Seven bilateral ECT treatments were administered as an inpatient over a three week period. MHS-ID nursing and psychology clinicians attended the first two administrations to assist unit staff. Premedication with olanzapine was unnecessary after the second ECT session. No agitation was seen after the third and subsequent procedures, and he rapidly became much more relaxed, in-

teractive and happy. He ate well and his bowels opened more regularly. MHS-ID staff visited periodically to assist ward staff and his family. In addition to working with the MHS-ID team, the second author was also a senior consultant psychiatrist in the inpatient unit, enabling her to provide continuity of care and stability in addressing any daily concerns on the ward for both the family and staff.

Arm splints and helmet were no longer required. On weekend leave after seven ECT treatments he appeared much happier, roamed around the house rather than retreating to bed, and ate and slept well. Lamotrigine was introduced as an anticonvulsant and adjuvant antidepressant, building up to a dose of 150mg bd. He continued mirtazapine 45mg nocte and risperidone 1mg bd. He returned to school where his behaviour and engagement were noted to be greatly improved. There was then a partial relapse of depressed mood and self-injury and eighteen days after discharge he was readmitted for a further two ECTs. Depression has now been in remission for three years with his medication unchanged. He has regained weight to a healthy weight range. Self-injury continues at low levels consistent with his premorbid behaviour, he is participating well in activities and his epilepsy is under good control.

Confirmation

** It is confirmed that this case study has been de-identified and that written consent has been received from the legal guardians for publication of this article.

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Glossary of Terms

Anhedonia: Inability to feel pleasure in normally pleasurable activities

Mane: Morning

Nocte: Night

Bd: Twice a day

References

1. Khalid, N., Atkins, M., Tredget, J., Giles, M., Champney-Smith, K., Kirov, G. The effectiveness of electroconvulsive therapy in treatment resistant depression: a naturalistic study. *Journal of ECT* 2008; **24**: 141-145.
2. Walter, G., Rey, J.M. and Starling, J. Experience, knowledge and attitudes of child psychiatrists regarding electroconvulsive therapy in the young. *Australian and New Zealand Journal of Psychiatry* 1997; **31**: 676-81
3. Little, J.D., McFarlane, J., Ducharme, H.M., Phil, D. ECT Use Delayed in the Presence of Comorbid Mental Retardation: A Review of Clinical and Ethical Issues. *The Journal of ECT* 2002; **18** (4): 218-222
4. Smith, D. Shock and Disbelief. *The Atlantic Monthly* February 2001.
5. McDonald, A., Walter, G. *ECT and Stigma*. In: Ghaziuddin, N., Walter, G., eds. *Electroconvulsive Therapy in Children and Adolescents*. USA: Oxford University Press, 2013; 29-55.
6. Collins, J., Halder, N., Chaudhry, N. Use of ECT in patients with an intellectual disability: review. *The Psychiatrist Online* 2012; **36**: 55-60.
7. Dare, F.Y. and Rasmussen, K.G. Court-Approved Electroconvulsive Therapy in Patients Unable to Provide Their Own Consent A Case Series. *The Journal of ECT*. Sept 2015 **31** (3): 147-9
8. Rush, K., Bowman, L., Eidman, S., and Mortenson, B.. Assessing Psychopathology in Individuals with Developmental Disabilities. *Behavior Modification* 2004; **28**: 621-637.
9. Moss, S., Prosser, H., Costello, H.N., Patel, P., Rowe, S., Turner, S., Hatton, C. Reliability and validity of the PAS-ADD checklist for detecting psychiatric disorders in adults with intellectual disability. *Journal of Intellectual Disability Research* 1998; **42**: 173-83.
10. Holden, B., Gitlesen, J.P. A total population study of challenging behaviour in the county of Hedmark, Norway: Prevalence and risk markers. *Research in Developmental Disabilities* 2006; **27**: 456-465.
11. Chiang, H-M. Expressive communication of children with autism: the use of challenging behaviour. *Journal of Intellectual Disability Research* 2008; **52**: 966-972.
12. Reiss, S., Levitan, G.W, and Szyszko, J. Emotional disturbance and mental retardation: Diagnostic overshadowing. *American Journal of Mental Deficiency* 1982; **86**: 567-574.
13. Bennett, C. Understanding systemic problems in providing mental health services to people with an intellectual disability and co-morbid mental disorders in Victoria. *Australasian Psychiatry* 2014; **22**: 48-51.
14. Wurth, P. and Brandon, S-A. The ACT Mental Health Service for People with Intellectual Disability, 10 years on. *Australasian Psychiatry* 2014; **22**: 52-55.
15. Donner, B., Mutter, Scior, K. Mainstream In-patient Mental Health Care for people with Intellectual Disabilities: Service User, Carer and Provider Experiences. *Journal of Applied Research in Intellectual Disabilities* 2010; **23**: 214 - 225
16. The Scottish Government. *Mental Health and Learning Disability Inpatient Bed Census*. <http://www.gov.scot/Publications/2015/06/7555/0>
17. Lyall, R., Kelly, M. Specialist psychiatric beds for people with learning disability. *BJPsych Bulletin* 2007; **31**: 297-300.



Reading List

Corsano, P., Musetti, A., Guidotti, L. and Capelli, F. (2017). Typically developing adolescents' experience of growing up with a brother with an autism spectrum disorder. *Journal of Intellectual and Developmental Disability*. Vol 42, Iss 2, Pp. 151-161.

Karrie A. Shogren, Michael L. Wehmeyer, Nirbhay N. Singh (Eds.), *Handbook of Positive Psychology in Intellectual and Developmental Disabilities: Translating Research into Practice*, (pp. 339-355). Cham: Springer

Stancliffe, R., Wiese, M., Read, S. (2017). Special Issue: End of life and people with intellectual disability. *Journal of Applied Research in Intellectual Disabilities*, 30(6), 977-1164.

Weise, J. and Trollor, J. (2018). Preparedness and training needs of an Australian public mental health workforce in intellectual disability mental health. *Journal of Intellectual and Developmental Disability*. Vol 43, Iss 4, Pp. 431-440.

Research News

1. School non-attendance in students with intellectual disability

By Monash University

<https://research.monash.edu/en/projects/school-non-attendance-in-students-with-intellectual-disability>

Melvin, G.A., Gray, K.M., Tonge, B.J. Hastings, R., Heyne, D. and Totsika, V. Psychiatry Monash Health

School non-attendance is a major societal problem and a current national and international priority. Children and adolescents with intellectual disability face significant inequalities and challenges, including participation in education. This Monash University project therefore aims to address a major gap in knowledge - understanding of types and rates of school non-attendance problems in school students with intellectual disability and the factors that influence non-attendance. Improved understanding of the pathways to non-attendance will facilitate the development of specific interventions to provide much needed improvements in attendance in this disadvantaged group.

2. Staff and Parent Wellbeing in Disability Services

By the University of Melbourne

[https://mispgh.unimelb.edu.au/centres-institutes/centre-for-health-equity/research-group/jack-](https://mispgh.unimelb.edu.au/centres-institutes/centre-for-health-equity/research-group/jack-brockhoff-child-health-wellbeing-program/research/physical-health-and-wellbeing/ooralla-partnership)

[brockhoff-child-health-wellbeing-program/research/physical-health-and-wellbeing/ooralla-partnership](https://mispgh.unimelb.edu.au/centres-institutes/centre-for-health-equity/research-group/jack-brockhoff-child-health-wellbeing-program/research/physical-health-and-wellbeing/ooralla-partnership)

This study aims to test the effectiveness, cost and acceptability of a program to increase key workers self-efficacy to support parents mental health.

This research will provide a strong evidence base upon which disability organisations can base organisational redesign to optimize health and wellbeing of children and carers within the context of the new National Disability Insurance Scheme (NDIS). The 4-year research project is in partnership with Yooralla, a leading Victorian disability organisation, and has worked in collaboration with families of children with a disability.

Strategies to improve the mental health of parents of children with a disability are urgently needed, and are timely given the current rollout of the National Disability Insurance Scheme (NDIS) in Australia. This program is needed to break the vicious cycle of poor mental health in parents of children with a disability that leads to poorer short and long term outcomes for themselves, their child and their family.

For more information please contact Dana Young. E: dana.young@unimelb.edu.au P: 03 9035 9870

The Medicine Cabinet: Placebo or Nocebo

Judy Longworth, Senior Clinical Pharmacist

Introduction by Dr David Dossetor, Child Psychiatrist

Department of Psychological Medicine

The Children's Hospital at Westmead

Introduction by Dr David Dossetor

These Latin terms for 'I please' or 'I harm' underlie common significant difficulties and complexities in treating children with medications. For example, in depression a third of young people respond to 'pink medicine' (a traditional form of placebo medication) whereas two thirds respond to an antidepressant. In effect, **we don't know whether the medication has worked, or whether they would have got better anyway.** And yet there is still one third that has failed to respond. Fortunately, it is found that on a second antidepressant another two thirds will respond, leaving a yet smaller group who still need further treatment. Overall, the placebo effect of any treatment tends to get less, the more severe and persistent a condition is.

This situation gets more complex if, as commonly occurs, the child and family report a medication works for 3 weeks or 3 months and then no longer works. Was this a placebo effect for the first period, or just part of a natural variation of the symptom over time or is it due to some sort of accommodation to the effect of the medication, through a pharmacological, neurotransmitter mechanism, metabolic or pharmacokinetic mechanism? All these are scientifically shown to be possible but very difficult to distinguish as it is not possible to do tests on these options in an individual patient.

In contrast, in a recent randomised control study of

fluoxetine in children and adolescents with Autism, 45% of the fluoxetine group experienced adverse events or side effects but this was not statistically significantly different to the 42% of the placebo group who also experienced adverse events or nocebo effects or negative placebo. Common adverse reports for fatigue, anxiety, nausea, headaches, sleeplessness, skin rash occur in both groups, meaning that side effects occur as much on no active treatment as on active treatment. Indeed, these are common everyday experiences even if you are not unwell or having treatment.

Thus, how do you know a negative effect is a result of a misinterpretation or some sort of persuasive process **in examining one's own wellbeing rather than due to an effect of the medication?** Particularly if a patient or parent comes to the consultation with a suspicion or pre-conviction that psychotropic medications are harmful and people should get better from illness without medication, then such anxiety or thinking becomes a causal factor in getting such nocebo symptoms. I recall a patient with severe anxiety and severe tics or **Tourette's syndrome, whose mother was suspicious and sceptical about medication.** Every side effect that the parent was warned of, the patient dutifully complained about. This lack of trust in the potential of a medication to help, led to premature breakdown in treatment in the context of other complex family social adversities that were also difficult to influence. This illustrates the level of trust a child and family need to



have for their clinician's judgement and the possibility that a medication can be helpful. If side effects occur, the child, family and clinician need to examine the seriousness and significance openly together, being aware of both placebo and nocebo effects. People often show greater scepticism or anxiety about a psychotropic medication than for a medication for a somatic disorder albeit they may be subject to the same level of scientific evidence. The article below describes the complexity to some of the mechanisms of placebo and nocebo, which clinician and patient need to be aware of to get the optimal benefit from treatment and to minimise nocebo effects.

Placebo or Nocebo by Judy Longworth

Definitions:

Placebo: Google describes it as a medicine or procedure prescribed for the psychological benefit to the patient rather than for any physiological effect; a substance that has no therapeutic effect, used as a control in testing new drugs (Google, 2018)

Nocebo: a detrimental effect on health produced by psychological or psychosomatic factors such as negative expectations of treatment or prognosis (Google 2, 2018).

The term placebo is often used in relation to something that has an effect, even a therapeutic effect, **however that "something" should be inert. Placebo effects** are a recent area of study and with more understanding there could be better clinical trial research. Clinical trials are the backbone of evidence based medicine and thus with the recognition of best practice and evidence based practice the understanding of placebos and nocebos has increased.

Traditionally, placebo is seen as an inert or "inactive" substance or procedure and the placebo effect (or response) is something that follows on from the administration of the placebo. There is a paradox in that an inert substance should not elicit a response or effect **on patient's mind, brain or body. The association of placebo effects with randomised controlled trials (RCTs)** has caused confusion because the response in the placebo group is not necessarily a genuine psycho-

“Thus, how do you know a negative effect is a result of a misinterpretation or some sort of persuasive process in examining one's own wellbeing rather than due to an effect of the medication?”

social response to the simulation of treatment. The response to a placebo in RCTs might reflect the natural course of disease, fluctuations in symptoms, regression to the mean, response bias with respect to a patient reporting subjective symptoms and other concurrent medications (Finnis et al, 2010). To better understand placebo effects in clinical trials and practice, a shift in focus from the inert substance to what is actually happening for the patient is a good place to start. Does the literature suggest that the placebo effect is a genuine psychobiological event attributable to the overall therapeutic context? Understanding this leads to a better clinical trial and stronger basis for evidence based medicine. These will be discussed later in this article.

Before a medication can be marketed or licensed it needs to fulfil certain regulatory requirements. The FDA (USA Federal Drug Administration) wants the sponsor, usually a pharmaceutical company, to show through adequate and well-controlled clinical studies the superiority of one substance or procedure over another. This also applies to the TGA (Therapeutic Goods Administration) in Australia. A well controlled study involves a comparison of subjects treated with the new medication and a suitable control population so that the effect of the new medication can be seen without influences such as spontaneous change, placebo effects, concomitant medications or observer expectations. Placebo control, no-treatment control (suitable where objective measurements are felt to make blinding unnecessary), and dose-comparison control studies are all study designs in which a difference is intended to be shown between the test article and some control (FDA, accessed 2018).

The FDA wants a new medication to show superiority over the control substance. If the control or inert substance is unable to show superiority then there would be no licensing and hence marketing of the product. This would be a problem for trials that are comparator trials i.e. trials against a product that was already licensed/marketed as the difference between comparator and new substance might not be statistically different to lead to approval by the governing authority.

Why placebo controlled trials?

One way as discussed to show superiority is by placebo controlled trials. There is an ethical question regarding whether clinical subjects should be **'washed out'** from their current active medication and then randomly assigned to a treatment arm that might consist of a placebo (or inert substance) exposing the subject to significant risks, especially when there is available efficacious approved medications. There are advocates for the approach that new medications should be approved for clinical trials that compare the investi-

“Neuroimaging studies have shown nocebo affects brain activation in different sites to placebos ”

gational medication with established approved medications. These trials are often done post marketing by research institutes instead of the traditional pharmaceutical company trials. This research approach is called equivalence or non-inferiority and is in contrast with current criteria of the FDA for superiority.

One limitation of placebo trials, especially when these are only being used for the licensing of medication, is that there is no comparison with conventional medication; i.e. equivalence studies when applying the results to the conventional or normal population (Krauss, 2018). These trials are used for bodies such as the Pharmaceutical Benefits Advisory Committee (PBAC) when deciding about pricing of medications to go onto the Pharmaceutical Benefits Scheme (PBS). Superiority studies are necessary for the investigational medication to show prominence over the comparator, thus for practical reasons the comparator is always placebo.

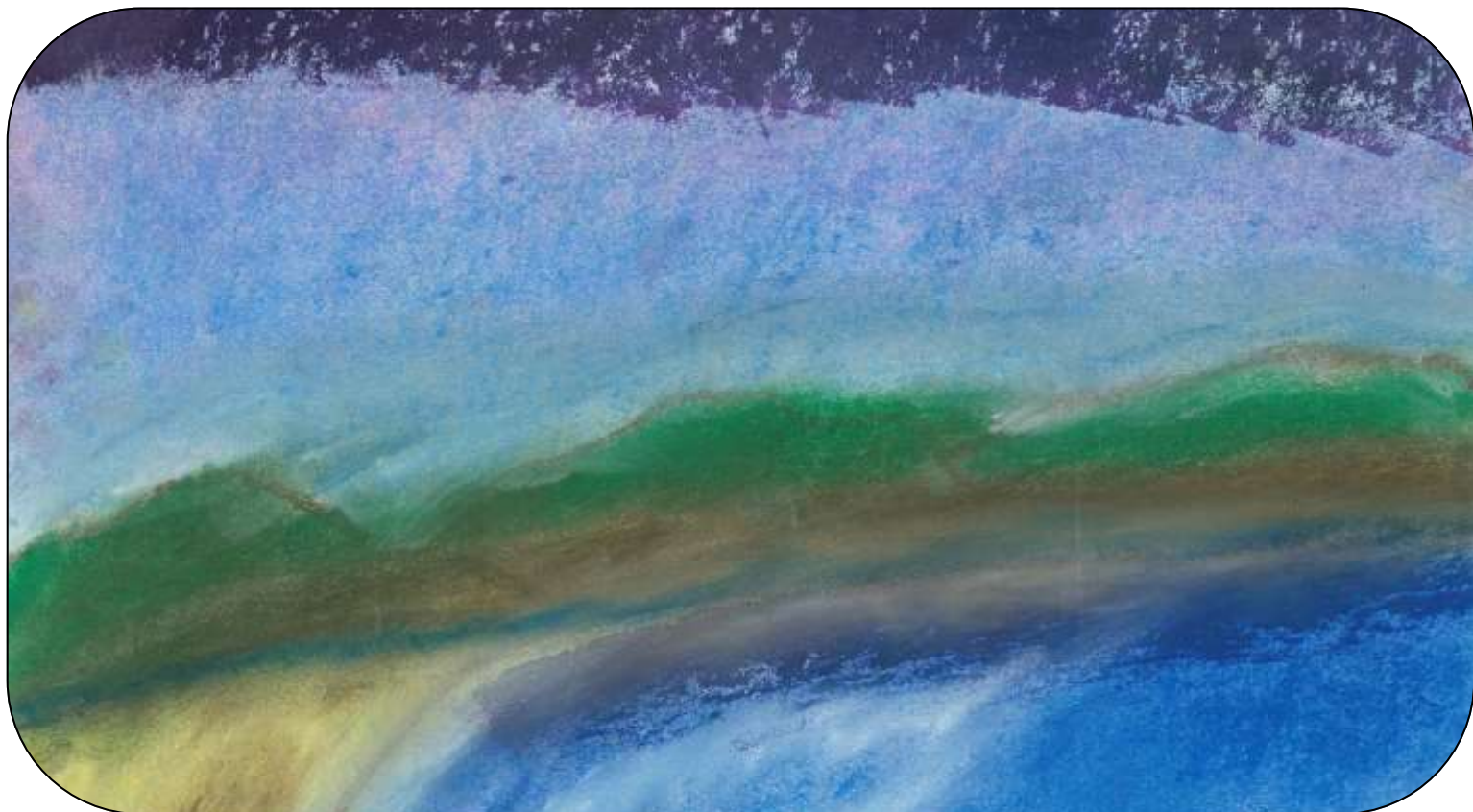
Placebo trials

The neurobiology of placebo responsiveness has ad-

ressed placebo analgesia, and is considered in terms of opioid and non-opioid mechanisms. The use of naloxone (opioid antagonist) in studies has shown the involvement of endogenous opioids in some placebo effects. Placebo analgesic effects are also likely to be inhibited by cholecystokinin as trials with cholecystokinin antagonist have shown a potentiated effect (to increase the power of the drug). Cholecystokinin has a key role in nocebo hyperanalgesia which occurs through anxiety mechanisms including the hippocampal regions. Neuroimaging studies have shown nocebo **affects brain activation in different sites to placebo's** (Finnis *et al*, 2010).

Understanding how a placebo works clinically in different patient groups over time has not kept pace with research into the mechanisms of placebo effects which have mainly been laboratory experiments rather than those in a clinical setting (Finnis *et al*, 2010). There have been several clinical trials in adults showing the clinical relevance and aetiology of placebo-induced somatic sensations in irritable bowel syndrome and allergic rhinitis. The allergic rhinitis study (Schaefer *et al*, 2018) showed that placebos without deception can improve symptoms of allergic rhinitis and especially the quality of life but no effects on the improvement of symptoms.

Clinically focused research is needed to explore non-deceptive techniques for prescribing treatment aimed at promoting placebo effects; there has been some progress as there is clinically relevant evidence show-



Pain	Activation of endogenous opioids and dopamine (placebo; activation of cholecystokinin and deactivation of dopamine (nocebo))
Parkinson's disease	Activation of dopamine in the striatum and changes in activity of neurons in basal ganglia and thalamus
Depression	Changes in electrical and metabolic activity in different brain regions (eg ventral striatum)
Anxiety	Changes in activity of the anterior cingulate and orbitofrontal cortices; genetic variants of serotonin transporter and tryptophan hydroxylase
Addiction	Changes of metabolic activity in different brain regions
Autonomic responses to brain stimulation	Change of neuronal excitability in limbic regions
Cardiovascular system	Reduction of β adrenergic activity of heart
Respiratory system	Conditioning of opioid receptors in the respiratory centres
Immune response	Conditioning of some immune mediators (eg, interleukin 2, interferon γ , lymphocytes)
Endocrine system	Conditioning of some hormones (eg growth hormone, cortisol)
Physical performance	Activation endogenous opioids and increased muscle work
Alzheimer's disease	Prefrontal executive control and functional connectivity of prefrontal areas

Table of Mechanisms for placebo effects in medical conditions and physiological by Finnis et al, 2010.

ing placebo effects can have therapeutic effects in different populations (Finnis *et al*, 2010).

Hall and colleagues consider the possible interaction between placebo and drug molecular pathways, especially the genomic effects, and the implications for randomised control trial studies (Hall *et al*, 2015). This leads to the probability of identifying potential responders and non-responders through their genetic profile. The first evidence that there is a biological process giving rise to the placebo response which is more than just pleasing the experimenter was published in 1978 involved a series of molar teeth extraction and pain control experiments.

Placebo effects

Placebo effects are often considered as innocuous but **this can be misleading as improvement in patient's symptoms** that can be attributable to their participation in the therapeutic encounter with all its interactions (Kaptchuk and Miller, 2015). Placebo effects have been shown to rely on complex neurobiological

mechanisms involving neurotransmitters such as endorphins, cannabinoids as well as dopamine (Kaptchuk and Miller, 2015). There has even been fMRI studies which show the areas in the brain affected and these include the prefrontal cortex, anterior insula, rostral anterior cingulate cortex, and amygdala (Tetreault *et al*, 2016). There has also been genetic studies showing different alleles of genes displaying different responses such as different allele polymorphisms in the COMT (enzyme important in dopamine synthesis) studies in irritable bowel syndrome (IBS).

Finniss and colleagues have produced a table of mechanisms for placebo effects in medical conditions and physiological systems as replicated above (Finnis *et al*, 2010).

Nocebo effects

Less is known about the mechanism for nocebo response which can also be quite anxiogenic and stressful and thus limits ethical research into this mind-brain interaction. Study of nocebo effect relates to the nega-

“The placebo-nocebo effects are just being recognised ”

tive psychosocial context surrounding the treatment. As for placebo, it is the administration of an inert substance together with the suggestion that the substance will do harm. Nocebo-related effect also refers to the negative expectation of symptoms worsening with the administration of an inert substance. Most of this research has been done in pain where the negative expectations can lead to an amplification of pain response.

More recent studies have found nocebo effects were also associated with a decrease in dopamine and opioid activity in the nucleus accumbens, thus reducing the role of the reward system in nocebo effects as well. This suggests there is a complex relationship between different neurotransmitters such as dopamine and opioids when either placebo or nocebo are administered (Enck *et al*, 2008). Further to this, one suggestion of negative information can induce long-lasting negative effects (Colloca and Finniss, 2012).

The way information is delivered with regards to disclosure of potential adverse events in the clinical setting has been highlighted in several clinical trials involving significant nocebo responses (Colloca and Finniss, 2012). Nocebo effects can modulate the outcome of a given therapy in a negative way as do placebo effects in a positive way (Colloca and Finniss, 2012). Thus, the way information is delivered can have major implications on the results of an intervention.

The placebo-nocebo effects are just being recognised and as further studies are done more information will come to light as well as confirming studies that have already been done. The nature of how we test new medications in clinical trials will also change with new studies in pharmacogenomics and the impact of our genome on our response to medication. When looking at these issues in respect to children and adolescents there are further ethical considerations to be made. Studies in RCTs on the same active medication has shown adults and children have varied placebo response rates (Weimer *et al*, 2013). It has been noted in a review that placebo responses in children with psychiatric conditions (major depression, obsessive compulsive disorder and other anxiety disorders) when pooled are higher than those known in adults

(Weimer *et al*, 2013).

There is the assumption that placebo response is mainly generated by two distinct mechanisms: expectancies on one hand and Pavlovian conditioning on the other. In adolescents, these can be assumed as well as instrumental learning and learning by imitation as well as the genetic contribution (Weimer *et al*, 2013). Clinical trials are beginning to acknowledge that the higher placebo response especially in paediatric depression trials might not be attributable to most known factors such as amount of contact with staff or chances of receiving active treatment as it is in adult studies and further studies are needed into the cause of the placebo response.

In light of the paucity of placebo studies in children and adolescents there should be more studies as well as the need for gender studies as well as genetics to cover confounders.

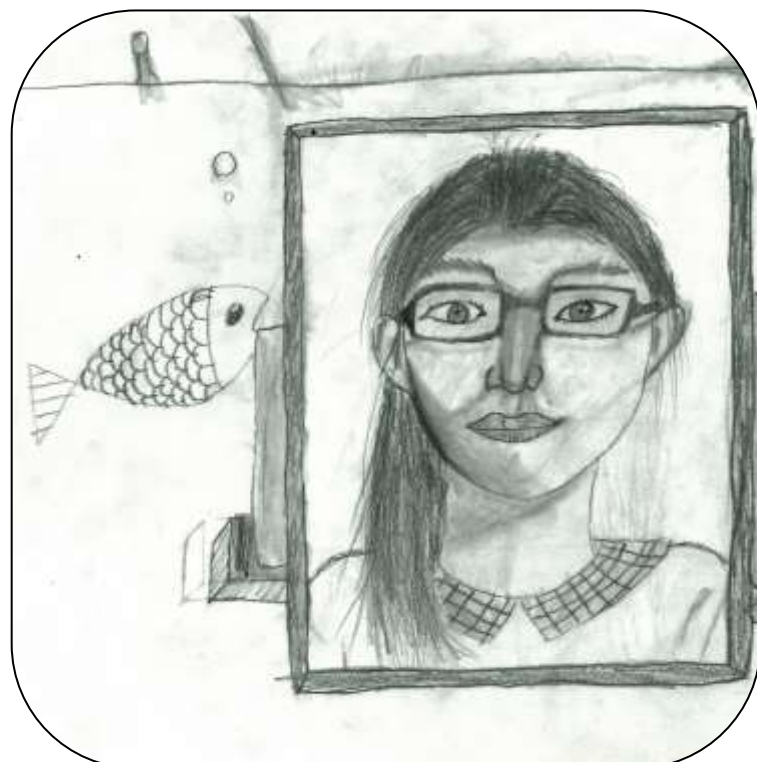
References:

Colloca, L., Finniss, D. (2012). Nocebo effects, patient-clinician communication, and therapeutic outcomes. *JAMA*. Vol 307, Iss 6, Pp. 567-8

Enck, P., Benedetti, F. and Schedlowski, M. (2008). New insights into placebo and nocebo responses. *Neuron*. Vol 59, Iss 31, Pp. 195- 206.

FDA (2018). <https://www.fda.gov/RegulatoryInformation/Guidances/ucm126501.htm> (Accessed 30/4/18).

Feifel, D. (2009). The use of placebo-controlled clinical





cal trials for the approval of psychiatric drugs: part I—**statistics and the case for the “greater good”**. *Psychiatry (Edmont)*. Vol 6, Iss 3, Pp. 41-43.

Finnis, D.G., Kaptchuk, T.J., Miller, F. and Benedetti, F. (2010). Biological, clinical, and ethical advances of placebo effects. *Lancet*. Vol 375, Iss 9715, Pp. 686-95.

Google (2018). https://www.google.com.au/search?source=hp&ei=8KPmWp_d08Wp8OX9ia7YCw&q=placebo+definition&oq=placebo+definiton&gs_l=psy-ab.1.0.0i10k1110.2905.6873.0.10384.18.17.0.0.0.0.206.2272.0j10j2.12.0...0...1c.1.64.psy-ab.6.12.2264.0.0j35i39k1j0i20i263k1j0i131k1.0.XElaWc_una4 (Accessed 30/4/18)

Google 2 (2018). https://www.google.com.au/search?source=hp&ei=8KPmWp_d08Wp8OX9ia7YCw&q=nocebo&oq=nocebo&gs_l=psy-ab.3.0i67k1i4j0i2j0i67k1j0i3.2576.4205.0.4672.8.6.0.1.1.0.208.998.0j2j3.5.0...0...1c.1.64.psy-ab.2.6.1011.0.46j35i39k1j0i131i67k1j0i131k1j0i46k1.0.mD4n-3wXFEw (Accessed 30/4/18)

Hall, K.T., Loscaizo, J. and Kaptchuk, T.J. (2015). Genetics and the placebo effect: the placebome. *Trends in Molecular Medicine*. Vol 21, Iss 5, Pp. 285-94

Hall, K.T., Lembo, A.J., Kirsch, I, et al. (2012). Catechol-O-methyltransferase val158met polymorphism predicts placebo effect in irritable bowel syndrome. *Public Library of Science*. October 23.

Jakovljevic, M. (2014). The placebo-nocebo response: controversies and challenges from clinical and research perspective. *Eur Neuropsychopharm*. Vol 24, Pp. 333-41.

Kaptchuk, T.J., Miller, F.G. (2015). Placebo effects in medicine. *New England Journal of Medicine*. Vol 373, Pp. 8-9.

Krauss, A. (2018). Why all randomised controlled trials produce biased results. *Annals of Medicine*. Vol 50, Iss 4, Pp. 312-22.

Schaefer, M., Sahin, T., Berstecher, B. (2018). Why do open-label placebos work? A randomized controlled trial of an open-label placebo induction with or without extended information about the placebo effect in allergic rhinitis. *PLoS ONE*. Vol 13, Iss 3: e0192758.

Tetreault, P., Mansour, A., Vachon-Presseau, E., Schnitzer, T.J., Apkarian, A.V. and Baliki, M.N. (2016). Brain connectivity predicts placebo response across chronic pain clinical trials. *Public Library of Science Biology*. Vol 14, Iss 10: e1002570

Weimer, K., Gulewitsch, M.D., Schlarb, A.A., Schwille-Kiuntke, J., Klosterhalfen, S., Enck, P. (2013). Placebo effects in children: a review. *Pediatric Research*. Vol 74, Iss 1, Pp. 96-102.

Society for the Study of Behaviour Phenotypes (SSBP) 21st International Research Symposium 2018.

A report by Dr Peter Wurth

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Abbreviations: NBPSA (Neurodevelopmental and Behavioural Paediatric Society of Australia), FMRP (Fragile X Mental Retardation Protein), FXS (Fragile X Syndrome), ID (Intellectual Disability), PWS (Prader Willi Syndrome), ASD (Autism Spectrum Disorder), AS (Angelman Syndrome), DS (Deletion syndrome), WS (Williams Syndrome), TSC (Tuberose Sclerosis Complex), PKU (Phenylketonuria).

The meeting was in Melbourne, 28-30th August hosted by Honey Huessler. The first two days were devoted to research papers with the educational day shared with the Neurodevelopmental and Behavioural Paediatric Society of Australia (NBPSA) on the final day.

The first keynote address was by David Godler, Group Leader, Cyto-Molecular Diagnostics Research Group of **the Murdoch Children's Research Institute (MCRI)** in Melbourne on the molecular model and clinical aspects of FMRP in FXS, highlighting the significance of mosaicism. Mosaicism is a complex phenomenon which can have a major impact on diagnosis and research into rare genetic syndromes. It is a common cause for delayed or absent diagnosis, and the degree of mosaicism can vary over time in one individual. Mosaicism may well have diluted the efficacy of the Novartis trial of AFO056 in FXS. It remains unclear how low is the threshold above which mosaicism becomes meaningful. He presented an overview of the molecular pathology in this syndrome. Although targeted treatment in mice with FXS correct the phenotype, this is unsuccessful in humans. One possible reason is that the mice are clones, whereas human pathology varies immensely, including as a result of mosaicism. Current studies are examining the prevalence of FXS and chromosome 15 imprinting disorders (PWS & AS) in 100,000 and 75, 000 newborns respectively.

Claudine Kraan from the same MCRI research group presented on FMRP allele size distribution in 35,000 males and females. Firstly a new group has been identified with an abnormally low number of CGG (cysteine, guanine, guanine) repeats, less than 27, and this may be associated with some disorders. This low count is found in 1:5 males and 1:3 females. The grey zone of 45-54 repeats does not in itself appear to be associat-

ed with ID, and some studies that show such an association may well have included undetected full mutations given sensitivity problems with current diagnostic tests, and the issue of mosaicism. CGG repeats greater than 80 are associated with FXTAS (Fragile X-associated tremor/ataxia syndrome) and FXPOI (Fragile X-associated primary ovarian insufficiency), but not with ID. Some individuals in the pre-mutation range are shy and awkward, but ID in this range may **be associated with a second 'genetic hit'**. She reported on a study of 19,000 referrals to the Victorian Genetics Service, which were compared with two independent cohorts from the general population. There was no association between expansions in the grey zone and pre-mutation zones, and developmental delay.

<https://www.youtube.com/watch?v=iFNUOZXcv-M>

Emma Baker also from the same MCRI research group presented on FMRP mRNA in blood as a predictor of intellectual functioning and autism severity in FXS. She noted in females 22-50% have an ID, and 16-20% have ASD. In males 100% have ID and 30-55% ASD. Lower IQ is the driver for the higher rates of ASD in males more than females. 60% of males and 100% of females still have some FMRP depending on IQ, i.e. mRNA is not completely silenced. She presented a study of 125 individuals (28% female) with FXS, which showed that FMRP mRNA levels in blood were strongly associated with IQ in males but not females, and with symptoms of ASD in females but not males. This discrepancy remains unexplained.

Randi Hagerman, Director of the MIND Institute, UC Davis, presented the results of a controlled trial of sertraline in children between 2 and 6 years with ASD but without FXS. A previous trial showed efficacy in FXS. Sertraline stimulates BDNF (brain-derived neurotrophic factor) which improves neuronal connectivity and can lead to improvements in expressive language and a reduction in reactivity. 58 children were recruited, 4 with identifiable genetic syndromes. 6 patients discontinued, and the remainder were treated with 2.5-5mg sertraline daily depending on age. A number of individuals did well but the overall results failed to reach significance, possibly a consequence of the het-

“Emotional dysregulation was significantly predictive of behavioural problems including aggression”

erogeneity of ASD.

Stephan Huijbregts a neurodevelopmental psychologist from Leiden presented a study of the impact on metabolic control of early treatment of PKU with tetrahydrobiopterin (BH4). BH4 is a cofactor of phenylalanine hydroxylase (PAH) which is deficient in PKU. PAH is required for the conversion of phenylalanine into tyrosine. Subjects had been treated early with a PKU diet. He noted that even in early treated PKU IQ was in the low normal range and there were higher rates of executive dysfunction, depression and anxiety, depending on phenylalanine levels. The addition of BH4 produced small but significant differences to Health-Related Quality of Life measures in adults but not children. (This medication is only on the Australian PBS for BH4 deficiency and costs >\$5000/month otherwise.)

Flora Tassone from UC Davis presented on Global Methylomic Profiling in children with ASD. She noted that methylation is a common epigenetic modification of DNA. The study involved 44 age-match participants with and without ASD, and found significant numbers of altered methylation in 47 genes in the ASD group, mainly hypermethylation with some hypomethylation. She noted the potential to contribute to diagnostic classification and possible therapies.

The second keynote address was by Tony Simon from UC Davis on the impact of cognitive-affective interaction on risk and protection for psychosis in 22q11.2 DS (VCFS). These individuals have IQs varying between 55 and 100. ADHD is found in 20-50%, typically the inattentive type. ASD is frequently diagnosed on the ADI given to parents but not on the ADOS. Anxiety is seen in 50-60% and schizophrenia in perhaps 20%, with typical onset between the ages of 18 and 35. He noted that while IQ is predictive of adaptive function in neurotypicals, anxiety is far more predictive in those with this syndrome. He noted that anxiety may be due to **‘misattribution of salience of environmental stimuli’, leading to high levels of ‘allostatic cognitive load compared to capacity’**. **Affectively laden distractions impair attention excessively in this syndrome.** The apparent ASD often diagnosed appears to resemble a combination of anxiety and avoidance, and most of these individuals are very socially motivated. High anxiety in ear-

ly adolescence is probably a predictor of the later development of psychosis. This hypothesis is the focus of a current longitudinal study. (<https://www.youtube.com/watch?v=VVcvPkUSoVA>)

Tony Simon again presented on 22q11.2 DS, comparing various measures of ASD in 17 children with this syndrome compared to a matched group of children with idiopathic ASD. There were divergent scores on language, some overlap on scores on ADOS items, and qualitative differences in social impairment. He speculated that ASD in this population could represent a psychotic prodrome.

Linda Campbell from Newcastle presented on emotional dysregulation in 22q11.2 DS. She noted that these individuals have higher scores than typicals on anger and aggression, and 7-14% are diagnosed with disruptive disorders. A cross-sectional sample of 129 subjects compared with 116 typical children aged 4-22 was assessed with a range of measures, showing 50% had significant problems with emotional control compared to 8% of typicals. Emotional dysregulation was significantly predictive of behavioural problems including aggression.

Donna McDonald-McGinn from Philadelphia Children’s Hospital presented on language decline in 22q11.2



DS. Schizophrenia is reported to follow a decline in cognitive and language scores in this syndrome. A retrospective chart review of 730 children up to the age of 21 was performed. Significant declines in cognitive functioning occurred in many children before the age of 10, including a striking decline in language scores. FSIQ declines as VIQ falls to match PIQ. Further work to determine any association between the degree of decline and the subsequent onset of schizophrenia is planned. She also noted that some individuals with **this syndrome develop early onset Parkinson's disease.**

Pat Howlin from Kings College London described efforts to identify what interventions work with which children with ASD. ASD is clearly heterogeneous yet most research focuses on group outcomes. She explored factors predictive of treatment response as opposed to variables related to prognosis on autism more generally, in two large groups of children. Interventions tended to produce significant group effects, with variability of response within groups. None of the variables identified reliably distinguished responders and non-responders, illustrating the complexity of the field.

Lauren Lawson from La Trobe University Melbourne described gender differences in internalising psychopathology in young adults with ASD. The limited studies in this area typically find little difference in the prevalence of anxiety and depression between males and females, but her study of 111 subjects found the same overrepresentation of anxiety and depression in females with autism as is found in neurotypicals. This correlated with a significantly higher use of emotional

regulation involving suppression in females than males.

Melanie Porter Director of the Centre for Research in Atypical Neurodevelopment at Macquarie University described the complex gene and environmental contributions to the phenotype of WS. She noted that phenotype can be defined as the observable expression of the interaction of genotype and environment. WS is due to a 7q11.23 deletion which typically involves the loss of 26, occasionally 28 genes. While there is significant heterogeneity of expression, individuals are typically hypersocial, empathic, musical, with hyperacusis, poor emotional regulation and a high frequency of mental illness, especially ADHD, Generalised Anxiety Disorder, depression and specific phobias. They have a mild to moderate level of ID, significant executive dysfunction, and a higher frequency of cardiovascular and connective tissue disease. They exhibit a positive cognitive bias, and tend to focus on eyes more than the whole face compared to neurotypicals. There are widespread changes in brain structure with loss of volume and connectivity. 34-40% of children with WS take stimulants, and 26% of adults take psychotropic medication.

Liz Pellicano from Department of Educational Studies, Macquarie University, Sydney described the need for autism researchers to involve patients and families to a much greater extent. There has been a surge in publications on autism since 2000 from 500 to 3500 papers per year, but it is not clear that this work has made any difference to the lives of people with autism and their families. Most of them feel that higher rates





of anxiety and depression are due to their experience of others as a result of stigma, exclusion, the pressure to be normal and a failure of empathy by neurotypicals. People with autism do not adapt to stimuli as much, and tend to see the world more accurately and less biased by previous experience. The need to involve subjects and their families in research was a theme that recurred throughout the conference.

Anna Jansen paediatric neurologist from University Hospital Brussels described the top fifteen research priorities in TSC derived from focus groups and interviews with patients and caregivers. The major problems identified were refractory epilepsy, disfiguring skin lesions and TAND (TSC-associated neuropsychiatric disorders). This was the second presentation highlighting the need to involve individuals with genetic disorders themselves in research.

Continuing this theme, Dawn Adams from the Autism Centre of Excellence Griffith University Queensland presented on parent thoughts on clinical trials for children with a number of rare genetic disorders. Many were surprisingly keen to participate in trials of approaches that have only previously been researched in mice. Most were keen to see the personality of their children not affected by any treatment, and attitudes to 'cure' varied. **Parents of individuals with 22q11.2** wanted research on mental illness, and parents of those with AS wanted research on speech and communication.

Rachel Cvejic from 3DN, University of NSW Sydney presented preliminary data on the use of 'big data' in behavioural phenotype research. Linkages are now possi-

ble between data on presentations to EDs, hospital admissions, ambulance trips, and births and deaths. These linkages can create cohorts by diagnosis. A retrospective study of 492 people with AS and 3570 with Down Syndrome who were admitted to New South Wales hospitals over a 14 year period was conducted. Individuals with AS were five times more likely to be admitted for accidental injury than those with DS or neurotypicals. Rates for interpersonal violence and falls were much higher for AS. There are still big gaps in the data, which relies on the accuracy of coding. ICD10 lumps multiple rare copy number variations together.

Rachel Royston from Birmingham presented on risk factors for psychopathology in WS, FXS and PWS. Affective and anxiety disorders are much more common in FXS, specific phobias in WS and FXS and psychosis in PWS. Sensory hypersensitivities predict anxiety. There was no association with age, and none of the variables studied predicted psychopathology in FXS.

Laura Groves from School of Psychology University of Birmingham presented on anxiety disorders in Cornelia de Lange and FXS. Both conditions have high rates of anxiety disorders. This study found a disconnection between subjective experience and diagnosis reflective of the lack of sensitivity of the diagnosis of anxiety disorders in this population. DSM 5 is particularly insensitive, given the excessive reliance on subjective phenomena in diagnostic criteria.

Kylie Gray from the Centre for Developmental Psychiatry & Psychology, Monash University presented a comparison of the efficacy of community-based parent intervention for children with and without ASD. 365 families who took part in the Stepping Stones Triple P program were studied. The diagnosis of ASD was based on parent report only. The children with ASD had significantly higher rates of behavioural and emotional problems at all time points compared to those without, but both groups demonstrated significant decreases in these problems after treatment. This program relies on parent education about behavioural management. Benefit was maintained at 3 and 12 month follow ups. Financial hardship was controlled for and the average IQ of the children was 63.

“Parents of individuals with 22q11.2 wanted research on mental illness, and parents of those with AS wanted research on speech and communication.”

“School non-attendance is 50% more common in intellectual and developmental disorders”

Effie Pearson from the School of Psychology, University of Birmingham presented on communication in AS, a disorder caused by a deletion or alteration of the gene UBE3A (Ubiquitin-protein ligase E3A), which occurs in 1:12000-20000 births. Non-deletion forms of AS (ie. uniparental disomy or imprinting problems) had better communication skills than those with a deletion. Speech is absent or severely impaired across this spectrum however regardless of cognitive ability. Spoken language skills dissociated from other communicative abilities, suggesting specific involvement of this gene in speech production. Alternative communication aids are therefore likely to be more effective in this population.

Megan Tones from the Mater Medical Research Institute, Brisbane presented on developmental milestones in AS using data derived from the Global AS Registry, a Queensland initiative. There are 700 individuals on the register, and 75% have the deletion form. Only 20% are older than 18 years. There are more mutations of UBE3A than cases of uniparental disomy in this group. There was a wide range of ages in meeting standard developmental milestones, which were typically more delayed in those with the deletion form than other subtypes.

Bruce Tonge, Emeritus Professor of Psychiatry, Monash University gave the Tom Oppe lecture on school non-attendance in young people with developmental disabilities. He described the evolution of his understanding and the shift in name from school refusal to school non-attendance. This can be authorised, ie. agreed upon at all necessary levels including the Education Department, and unauthorised, a local initiative of either school or families. He identified four categories – refusal, associated with anxiety and depression; truancy, associated with disruptive behaviour disorder; withdrawal by parents as a result of parental need; and exclusion by the school, either local, inappropriate and unlawful, or formal and sanctioned by the Education Department. There is no good information on long-term outcomes which are likely to be poor. Problematic non-attendance is 50% more common in intellectual and developmental disorders than in neurotypicals, and 300% more common when per-

sistent. About 10% of students with ID experience exclusion from schooling. A surprising figure was the number of days missed as a result of attendance at medical appointments. Children with ASD, WS and PWS had an increased prevalence of non-attendance whereas those with Down Syndrome and FXS had a reduced prevalence. Authorised non-attendance was particularly prevalent in ASD. 45% of parents struggled to overcome their children's resistance to going to school. A high prevalence of this problem was also found in a study from Warwick, UK.

Petrus de Vries, Professor of Psychiatry, University of Cape Town presented on TSC-associated neuropsychiatric disorders (TAND). TSC affects 1:6000, and 30% are inherited in an autosomal dominant pattern, with males equalling females, and the rest new mutations. Mortality under 5 years has plummeted in the last 25 years but the level of ID has not changed. The TSC1 or hamartin gene on chromosome 9 and the TSC2 gene tuberin on chromosome 16 combined to form a complex which inhibits mTOR (mammalian or mechanistic target of rapamycin) which regulates cell growth. The failure of this complex to form in TSC results in much higher levels of mTOR, which cause the overgrowth and tumours seen throughout the body. Sirolimus and analogues inhibit mTOR and shrink tumours, as well as improving seizures, and are now on the Pharmaceutical Benefits Scheme for selected indications in this



condition. There are numerous neuropsychiatric complications including ASD, ADHD, learning disorders, executive function disorders, depression and anxiety, as well as ID. There is no increase in psychotic illnesses. There is a freely available fifteen minute TAND checklist in fifteen languages, and the aim is to have all individuals with TSC screened annually, in order to correct the pervasive underdiagnosis of TAND around the world.

Honey Heussler from Child Medical Research Centre, University of Queensland presented on medical cannabinoids. She described **Scott's parabola**, developed to describe the phases through which new surgical treatments pass, from non-use, to excessive hype, to peak use, to the recognition of problems and eventual disuse. Cannabinoid usage is now in the upwards slope of this parabola. The most common compounds are cannabidiol and THC. Different brands of medicinal cannabis have various ratios of these two compounds. It is **used in MS, oncology and possibly in Parkinson's disease**. Cannabidiol has multiple sites of action. 30-50% of cases of refractory epilepsy respond although some show worsening. It may be useful in lessening anxiety in FXS. There is currently insufficient available information to recommend its use, although many parents of individuals with ID are already obtaining various compounds.

ACT paediatrician Felicity Williams presented on her experience of having a four year old child with AS. Gross motor delay was evident at six months leading to diagnosis. She described the inevitable need to socialise with other parents who understood the issues involved, and with whom there was frequent contact during the different therapies required. She described the multiple appointments and associated costs, and the need to celebrate small achievements. Her personal experience has helped her understand the perspective of parents with children with severe ID, and helped her explain to these parents the current deficit model of funding, which requires parents to emphasise their **child's weaknesses rather than their strengths and attributes**. It has helped her recognise both the value and burden of having a child with ID in a family. She noted that five gene-based therapies for AS are entering development.

Stewart Einfeld from Sydney University presented on behavioural phenotypes in PWS, WS and 22q11.2. He focused especially on individuals with PWS. Emotional ability is much lower than IQ, which is typically in the mild to average range of ID. If they are upset they regress quickly, and are very egocentric. Skin picking is common and NAC (N-Acetyl Cysteine) and topiramate are occasionally helpful. They perseverate, check and



hoard, but there are none of the classic cognitive associations seen in OCD, the behaviour is not ego dystonic, and these behaviours do not respond well to OCD medication. 10-30% develop psychosis, at seven times the rate in UPD (Uniparental Disomy) compared to deletions, with typical onset before 40 and as young as 9. The peak incidence occurs in late adolescence, and is typically relapsing-remitting but occasionally chronic. Depression is more common than psychosis and responds to standard treatment. Temper outbursts including rage attacks are a major problem, which can last until exhaustion sets in. No medication has been shown to be helpful. There is a lack of functional oxytocin but trials of oxytocin have been ineffective or counterproductive. There are trials of the longer acting carbococin and mindfulness underway. Tantrums diminish over the age of 30. There are deletions of the COMT gene in 22q11.2. The psychotic illness looks identical to schizophrenia and is often fairly treatment resistant. Anxiety is common and treatment follows the usual lines. WS individuals exhibit high levels of anxiety, including specific phobias which respond to SSRIs. Hyperacusis responds to avoidance of stimuli and ear-plugs. They have low anxiety about strangers and are indiscriminately positive towards them. There is a very high incidence of childhood sexual abuse.

There were numerous poster presentations of a high standard, covering a range of syndromes. The next conference will be in Birmingham, UK from the 4th to the 6th of September 2019.

I have promises to keep
They're stuck in my head, real deep

I've got Autism and ADHD
People treat me like I'm a disease
I have a body that just can't stop moving
Which would be ok, if the world was always
grooving
My senses are always alive
Leaves me in constant overdrive
I try so hard to be good
But people just think I'm a hood
I struggled so much in school
Some kids can be so cruel
I want to be just like the others
Not feel like I'm always a bother
My mind keeps going non-stop
Until I finally drop
When I hit over-load, my brain shuts down
Leaving me feeling, like I'm going to drown
I have only one or two friends
Who I'll look after, until the end
If you could just see past my behaviour
It would feel like I have another saviour
I wish you could see the real me
I just want to fly and be free

I have promises to keep
Which I think about each night, before I sleep

Jackson Karl Schomacker
11 Yrs Old

The beautiful artworks in this journal are taken from the participants of the **Operation Art project** at the Children's Hospital at Westmead. You can find out more at <https://www.artsunit.nsw.edu.au/visual-arts/operation-art-2014>

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