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[Original article: 3 tables, 2 figures, 14 online tables, 8 online figures, 1 appendix]

## Self-reported impact of developmental stuttering across the lifespan

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## PUBLICATION DATA

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[abstract]

**AIM** To examine the phenomenology of stuttering across the lifespan in the largest prospective cohort to date.

**METHOD** Participants aged 7 years and older with a history of developmental stuttering were recruited. Self-reported phenotypic data were collected online including stuttering symptomatology, co-occurring phenotypes, genetic predisposition, factors associated with stuttering severity, and impact on anxiety, education, and employment.

**RESULTS** A total of 987 participants (852 adults: 590 males, 262 females, mean age 49 years [SD=17 years 10 months; range=18–93 years] and 135 children: 97 males, 38 females, mean age 11 years 4 months [SD=3 years; range=7–17 years]) were recruited. Stuttering onset occurred at age 3 to 6 years in 64.0%. Blocking (73.2%) was the most frequent phenotype; 75.9% had sought stuttering therapy and 15.5% identified as having recovered. Half (49.9%) reported a family history. There was a significant negative correlation with age for both stuttering frequency and severity in adults. Most were anxious due to stuttering (90.4%) and perceived stuttering as a barrier to education and employment outcomes (80.7%).

**INTERPRETATION** The frequent persistence of stuttering and the high proportion with a family history suggest that stuttering is a complex trait that does not often resolve, even with therapy. These data provide new insights into the phenotype and prognosis of stuttering, information that is critically needed to encourage the development of more effective speech therapies.

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### **What this paper adds**

- Half of the study cohort had a family history of stuttering.
- While 75.9% of participants had sought stuttering therapy, only 15.5% identified as having recovered.
- There was a significant negative correlation between age and stuttering frequency and severity in adults.

[main text]

Stuttering, or stammering, is a complex trait that imposes significant personal and economic burden on the individual and wider society.<sup>1,2</sup> Stuttering can be characterized by dysfluent speech, including word, sound, or phrase repetition, sound prolongation, and blocks in sound production.<sup>3</sup> People who stutter may also experience non-verbal secondary behaviours, such as tics and grimacing, or exhibit covert behaviours, such as avoiding specific words or situations.<sup>3</sup>

Stuttering is common, with up to 11% of children experiencing stuttering by the age of 4 years, and a lifetime population prevalence of approximately 1%.<sup>4,5</sup> Most prevalence studies found stuttering to be more common in males, with observed male-to-female ratios of up to 4.6.<sup>5,6</sup> The lifetime recovery rate has been estimated at almost 70% from retrospective interview data,<sup>7</sup> although no prospective data have been examined across the lifespan.

Interventions targeting fluency typically centre on cognitive behavioural approaches and operant conditioning for younger children (e.g. Lidcombe Program) and behavioural speech restructuring for school-age children and adults (e.g. Smooth speech, Camperdown program).<sup>8,9</sup> These interventions are effective for many, but not all, individuals.<sup>8,10,11</sup>

Reports on a family history of stuttering vary, with clinically ascertained cohorts consistently reporting a positive family history in more than 50% of individuals.<sup>12–14</sup> Linkage

studies in large families have implicated four putative causal variants in genes involved in lysosomal metabolism (*GNPTAB*, *GNPTG*, *NAGPA*)<sup>15</sup> and intracellular trafficking (*AP4E1*).<sup>16</sup> More recently, genome-wide association studies have identified common genetic variants associated with developmental stuttering and related traits in population-based cohorts,<sup>17,18</sup> providing further evidence that stuttering may be a genetically complex trait.

When stuttering persists, the negative impacts are far-reaching and can be lifelong. People who stutter are more likely to experience poorer quality of life, which is characterized by bullying and social anxiety disorder.<sup>1,19,20</sup> Emotional reactivity and neuropsychiatric disorders, such as anxiety and depression, are also reported at increased levels.<sup>21</sup> Studies exploring the lived experience of stuttering found unfavourable educational outcomes, employment experiences, and reduced earnings,<sup>1,22,23</sup> although in relatively small cohorts ( $n < 262$ ).

In this study, we examined stuttering phenomenology in the largest cohort of people who stutter recruited prospectively. We present extensive phenotypic data, including self- or parent-reported information on treatment for stuttering, prognosis, associated quality of life, and psychosocial outcomes. We document (1) stuttering, broader communication and neuropsychiatric phenotypes, and access to treatment; (2) a family history of stuttering; (3) factors associated with stuttering severity; and (4) the impact of stuttering on anxiety, education, and employment.

## METHOD

### Study population and recruitment

Participants were recruited between April 2018 and May 2020 from the Australian arm of our Genetics of Stuttering Consortium ([www.geneticsofstutteringstudy.org.au/consortium](http://www.geneticsofstutteringstudy.org.au/consortium)). Inclusion criteria were (1) participants aged 7 years or older who (2) currently stuttered or stuttered in the past. People who stuttered were asked: Have you ever stuttered? The following description was provided for the specificity of our phenotype.

People who stutter have trouble getting their words out. Stuttering is when people repeat sounds over and over (e.g. C-c-can I go?), repeat words or syllables over and over (e.g. Can-can-can-I go?); make long prolonged sounds (e.g. Caaaaaaaaaaaaan I go?); have speech ‘stoppages’ or ‘blocks’ where no sounds come out.

The age range was specified to limit cases who may demonstrate brief periods of dysfluency that resolve by the age of 7 years.<sup>24</sup> Participants were excluded if they experienced any acquired neurological disorders, such as traumatic brain injury, before the onset of their stuttering.

Our recruitment strategy included national media campaigns and promotion through support organizations, university departments, and stuttering clinics via mail-outs, e-mail, and social media as successfully used by our team in several studies.<sup>25,26</sup> The human research ethics committee at the Royal Children’s Hospital, Melbourne approved the study (no. 37353).

### **Data collection**

Participants consented to study participation via the website ([www.geneticsofstutteringstudy.org.au](http://www.geneticsofstutteringstudy.org.au)). Two surveys were used based on the stuttering study nested within the longitudinal Early Language in Victoria Study, which examines communication development in children:<sup>2,4,24</sup> 7 to 17 years and 18 years and older. The former was completed by the child’s parent or guardian. Surveys took approximately 15 minutes to complete. Both surveys were intended to capture the individual’s or parent’s perceived experiences of the nature and impact of stuttering.

The survey examined demographic data, stuttering phenotypes (age at onset, resolution or persistence, blocks in speech, sound, syllable, part-word, word, or phrase repetition, and frequency of stuttering), stuttering management (advice, assessment, and therapy), the number and relationship of family members who stuttered and other health-related information, such as communication (speech and language disorder subtypes), neuropsychiatric, and neurodevelopmental phenotypes (Figure S1). Participants were asked: In the last 12 months, have you had a stutter? Stuttering was defined as ‘recovered’ if individuals had not stuttered in the 12 months before completing the survey. Self-reported stuttering severity ratings were also

collected; these previously utilized and published measures were made using a rating scale from 1 (no stuttering in the past week) to 10 (extremely severe stuttering in the past week).<sup>27,28</sup> Participants were asked to report stuttering frequency on an ordinal scale from 1 (sometimes, not every day) to 5 (several times per sentence).

Anxiety ratings included: (1) Are/were you anxious while stuttering? (yes/no); and if yes (2) How anxious are/were you while stuttering? (a little, fairly, very, extremely). Yes/no questions about the impact of stuttering on educational attainment and employment were answered.<sup>4,24,29</sup> Participants' educational attainment levels were classified into four categories: (1) high school certificate/trade apprenticeship; (2) certificate/diploma; (3) undergraduate degree; (4) postgraduate degree.

Survey questions were revised throughout the early recruitment process in response to feedback; hence, the number of survey responses varied according to the question.

### **Stuttering validation**

Stuttering was validated via videoconference with a speech pathologist in 54 participants (5.5%), chosen using stratified random sampling for sex, socioeconomic status, and stuttering status. Speech pathologists assessed the presence or absence of stuttering and rated stuttering severity on the same scale used for self-report.<sup>27,28</sup> Participants were also asked to rate the severity of their stuttering on the day and ratings were compared.

### **Statistical analyses**

#### ***Stuttering phenotype and factors associated with stuttering severity***

The correlation between stuttering severity and frequency was estimated using Kendall's tau ( $\tau$ ). For binary factors (sex, family history of stuttering, presence of additional communication difficulties), we tested for significant differences in the distribution of severity scores using the Mann–Whitney  $U$  test. Associations between age and both severity and stuttering frequency were examined using Kendall's  $\tau$ .

#### ***Stuttering and anxiety, education, and employment***

Differences in the distribution of anxiety ratings between participants with persistent stuttering, compared to people with recovered stuttering, were examined using the Mann–Whitney  $U$  test. Univariable associations with both anxiety and educational attainment level in adults were examined with the Mann–Whitney  $U$  test for binary factors, the Kruskal–Wallis test for age, and Kendall’s  $\tau$  for stuttering severity.

The effect of stuttering severity on both anxiety and educational attainment level was then examined using ordinal logistic regression models. In the first instance, models were fitted with any factors (sex, age, family history of stuttering, presence of additional communication difficulties) showing association in the univariable analyses ( $p < 0.05$ ) considered as covariates and assuming proportional odds. Covariates were selected for inclusion in the final model using a backward elimination approach (removing covariates if  $p \geq 0.05$ ). After covariate selection, the proportional odds assumption was tested for each variable in the model using a Brant test. A final generalized ordinal logistic regression model was fitted, allowing the effect estimate to differ across outcome levels for any variable violating the proportional odds assumption ( $p < 0.05$ ).

In children, the association between stuttering severity and parental education level (four-level ordinal variable) was tested using Kendall’s  $\tau$ .

## RESULTS

### Participants

The cohort comprised 1001 people who stutter and who registered for the study. We restricted analyses to those with complete data for sex, age, and stuttering status, leaving 987 participants (852 adults: 590 males, 262 females, mean age 49 years [SD=17 years 10 months; range=18–93 years] and 135 children: 97 males, 38 females, mean age 11 years 4 months [SD=3 years; range=7–17 years]); Table 1); 2.3 times as many males as females were recruited.

### Stuttering phenotype

Eight hundred and thirty-four participants reported stuttering in the past 12 months (individuals with persistent stuttering), while 153 participants (89 males, 64 females) had recovered (people with recovered stuttering). Of those who had recovered, 24 were children (16 males, eight females) whose parents or guardians reported that they stopped stuttering when they were 8 years old or younger; they had all received therapy from a speech pathologist. Stuttering onset occurred at age 3 to 6 years in 64.0% of participants (Figure S2). Of the 129 adults with recovered stuttering, 28 (10 males, 18 females) reported that stuttering resolved at age 8 years or younger, and 101 (63 males, 38 females) at 9 years or older. Ninety-three adults with recovered stuttering had sought advice, assessment, or treatment for stuttering and 36 had stopped stuttering without accessing treatment (Figure S3).

Self-reported stuttering severity ratings ( $n = 831$ ) spanned from 1 to 10, while stuttering frequency ratings ( $n = 308$ ) ranged from 1 (sometimes, not every day) to 5 (several times per sentence) (Table S1). Severity was positively correlated with the frequency of stuttering ( $\tau = 0.499$ ,  $p = 6.25 \times 10^{-24}$ ). Severity scores in adults (median = 4.0, interquartile range [IQR] = 2–5) were lower than in children (median = 6, IQR = 3–7,  $p_{\text{diff}} = 2.61 \times 10^{-8}$ ).

The most commonly reported stuttering phenotype was blocking (722 out of 987, 73.2%), followed by single sound ( $n = 573$  out of 987, 58.0%) and syllable ( $n = 380$  out of 987, 38.5%) repetition (Table S2 and Figure S4). Twenty-four individuals did not specify their stuttering phenotype.

Most reported additional responses to stuttering, such as avoiding saying certain words (787 out of 966, 81.5%) and beginning to speak and then giving up (584 out of 966, 60.5%). A subset of 308 adults were asked about physical responses to stuttering: 206 reported physical behaviours such as grimacing and blushing before, during, or after stuttering (Table S3).

### **Stuttering validation**

In our validation subset, consisting of 54 participants (5.5% of the study sample), there was 100% agreement between participant self-report and clinical rating of the presence or absence of stuttering. All participants with persistent stuttering ( $n = 51$ ) were confirmed to stutter via

videoconference. The three participants who did not stutter during the videoconference were those with recovered stuttering. Speech pathologist ratings of stuttering severity were strongly correlated with the participants' own ratings ( $r_s = 0.77$ ,  $p = 9.74 \times 10^{-12}$ ; Figure S5).

### **Communication and neuropsychiatric phenotypes**

Communication difficulties other than stuttering, diagnosed by a speech pathologist, were present in 74 adults and 36 children (Figure 1). Language difficulties were most common in four (0.5%) adults and 16 (11.9%) children and speech sound disorder in nine (0.9%) adults and 12 (8.9%) children (Table S4). Behavioural or attention difficulties (e.g. attention-deficit/hyperactivity disorder, behaviour or conduct problems) and neurodevelopmental difficulties (e.g. autism spectrum disorder, developmental delay, learning difficulties, and intellectual disability) were also reported at a higher frequency in children than adults: 34 (25.2%) children were reported to have one or more behavioural or attention difficulty, compared to 67 (7.9%) adults, while 42 (31.1%) children had one or more neurodevelopmental difficulty compared to 47 (5.5%) adults (Table S5). Adults provided information on neuropsychiatric diagnoses: 187 (22.0%) reported one or more neuropsychiatric condition, most commonly depression in 146 (17.1%) and anxiety in 139 (16.3%) participants (Table S5).

### **Access to treatment for stuttering**

Help-seeking for stuttering was noted by 834 people who stuttered (84.5%) in the form of advice, assessment, or therapy, with 740 having been assessed by a speech pathologist. A majority (749 out of 987, 75.9%) reported receiving therapy from a speech pathologist. Of those who did not seek help, 36 (23.5%) reported that they had recovered.

Smooth speech was the most common treatment accessed by adults and the Lidcombe Program by children (Figure S6 and Table S6). A small proportion of participants ( $n = 18$  [1.8%]) had not received treatment from a speech pathologist but had tried hypnotherapy or other complementary treatments.

### **Family history of stuttering**

Nine hundred and seventy-seven participants provided family history information (Figure 1c). Almost half (487, 49.9%; 168 females and 319 males) reported that at least one other family member stuttered. A third (333, 33.7%) reported at least one first-degree relative (parent, sibling, and/or child) who stuttered (119 females and 214 males). Twenty-six families reported five or more family members who stuttered. Of note, 413 (41.8%) reported at least one male relative who stuttered compared with 164 (16.6%) who reported at least one female relative who stuttered.

### **Factors associated with stuttering severity**

In people with persistent stuttering, stratified by adult or paediatric status, no significant associations were found between severity score and sex, reported family history of stuttering, nor presence of additional communication difficulties (Table S7). In adults with persistent stuttering, a significant negative correlation was observed between age and severity ( $\tau = -0.213$ ,  $p = 2.44 \times 10^{-15}$ ; Figure 2) and between age and stuttering frequency ( $\tau = -0.191$ ,  $p = 4.04 \times 10^{-5}$ ). This relationship between age and stuttering severity was not seen in children ( $r_s = 0.084$ ,  $p = 0.242$ ; Figure 2).

### **Stuttering and anxiety**

Anxiety responses during stuttering were stratified by persistent and recovered status (Table 2). People with recovered stuttering reported that they experienced higher levels of anxiety during stuttering compared to people with persistent stuttering ( $p_{\text{diff}} = 4.0 \times 10^{-3}$ ).

For people with persistent stuttering who responded to the anxiety questions ( $n = 261$ ), no significant associations were found between anxiety level and sex, age, or presence of an additional communication difficulty (Table S8). Reported family history was nominally associated with anxiety, with higher levels reported in participants with a family history of stuttering ( $p = 0.033$ ; Table S8). There was a non-linear relationship between stuttering severity and anxiety, with lower severity scores in the lowest and highest anxiety categories ( $n = 235$ ; Table S9 and Figure S7).

The regression model showed that for each 1-point increase in severity, participants had 1.21 times greater odds of reporting being ‘fairly’, ‘very’, or ‘extremely’ anxious, compared to ‘not anxious’ or ‘a little anxious’ ( $p = 6.86 \times 10^{-3}$ ; Table S10).

Participants with a persistent stutter who reported they were anxious whilst stuttering were asked which of a series of items made them anxious. ‘Situations’ were most commonly reported to cause anxiety (69.7%), followed by ‘people’ (52.1%), ‘letters or sounds’ (50.6%), and ‘phone calls’ (44.4%; Table S11).

### **Stuttering, education, and employment**

For adults, the perceived negative impact of stuttering on educational attainment, career progression, and employment is summarized in Table 3. Stuttering severity scores were significantly higher ( $p = 4.10 \times 10^{-9}$ ) in the 575 participants who reported that their stuttering had impacted their education and/or employment (median = 4, IQR = 3–6), compared to those who felt it had not had an impact (median = 3, IQR = 2–4; Figure S8).

In 722 adults, educational attainment level and severity scores had a significant negative correlation ( $\tau = -0.116, p = 1.0 \times 10^{-4}$ ; Table S12). Sex, a family history of stuttering, and other communication difficulties were not associated with the highest educational attainment level (Table S13). The distribution of age differed in the four education categories ( $p = 0.014$ ; Table S13), with participants tending to be older in the lowest and highest educational attainment level categories.

With adjustment for age, each point increase in severity score was associated with 0.86 reduced odds of attaining a qualification at a higher educational level ( $p = 3.23 \times 10^{-5}$ ; Table S14). This relationship between stuttering severity and educational attainment may be explained by a range of factors, including confounding by socioeconomic outcomes. To assess this, we examined the correlation between parental educational attainment level (often used as a proxy for socioeconomic status) and parent-reported stuttering severity in children who stuttered. We found a significant negative correlation between parental educational attainment level and stuttering severity ( $\tau = -0.268, p = 4.9 \times 10^{-4}$ ), suggesting that the relationship

between stuttering severity and educational level in adults may, at least in part, be due to confounding.

## **DISCUSSION**

This study examined self-reported stuttering phenotype and family history in 987 individuals. This is the largest prospective sample of people who stutter reported to date and concurrently with this depth of phenotypic and demographic details.

### **Stuttering phenotype**

The age at stuttering onset in both adults and children reported in this study was consistent with smaller published clinical studies, where most individuals start stuttering between 2 and 4 years of age.<sup>5,24,30</sup> However, adults reported that their stuttering began at older ages than children overall; recovery (where applicable) happened later compared to what is usually reported in the literature. These seemingly atypical trajectories of stuttering reported by adults in this study may reflect a recall bias towards an event that occurred many years ago. Furthermore, unexpected results may also reflect varying times at which an individual became aware of their stuttering. Blocks were the most common stuttering phenotype followed by single sound and syllable repetition in both adults and children. Existing studies show contrasting findings across paediatric and adult studies whereby word, part-word, and syllable repetition are most common in paediatric populations<sup>5,29,31</sup> and blocks most common in adult populations.<sup>32</sup> Our data are in line with findings in adult populations. The different stuttering types reported in children recruited to our cohort may be attributed to differences in methodology, that is, parent report versus clinical assessment, thereby making direct comparisons difficult.

The significant positive correlation between stuttering severity and frequency suggested that those who stuttered more often reported a higher severity rating score.<sup>27,33,34</sup> Interestingly, for adults, there was a negative correlation between age and both self-reported severity and frequency of stuttering. Without objective longitudinal measurements of stuttering severity in the same individuals, we cannot definitively conclude that stuttering severity changes with age.

An alternative explanation may be that the perception of severity changes with age, an area that is less explored in the literature.

### **Communication and neuropsychiatric phenotypes**

Communication, behavioural and attention, and neurodevelopmental difficulties were reported at a higher rate in children compared to adults. A possible reason for this difference may be that many of these conditions occur and are diagnosed in childhood. Rates of diagnoses for many of these conditions have also increased over time.<sup>35</sup>

Neuropsychiatric diagnoses were reported in 22% of adults with stuttering. The most commonly reported conditions were depression (17.1%) and anxiety (16.3%). The 2007 Australian National Survey of Mental Health and Wellbeing estimated a lifetime population prevalence of experiencing any mental disorder of 45.5%, with 14.4% of Australian adults experiencing anxiety disorders in the past 12 months,<sup>36</sup> suggesting that our cohort of adults with stuttering did not experience neuropsychiatric illnesses at a higher rate than the general population. Yet, given our self-selecting recruitment strategy, these findings are not necessarily representative of all people who stutter.

### **Access to treatment for stuttering**

A high proportion (83.6%) reported seeking advice, assessment, or therapy for their stuttering, most of which was from a speech pathologist. This proportion is notably higher than that reported in community-based studies of stuttering (13%–40%)<sup>7,24</sup> and may again reflect the self-nominated recruitment methodology. Most treatments accessed were evidence-based therapies, such as cognitive behavioural approaches or speech restructuring.<sup>8,9,37</sup> Other less researched treatments, such as hypnotherapy, were also reported; however, in most cases, these complementary therapies were received alongside conventional therapy with a speech pathologist.

Most people with recovered stuttering reported receiving treatment, although 36 (3.6%) recovered without treatment. This proportion is lower than previously reported in retrospective<sup>7</sup> or longitudinal<sup>24,38</sup> studies, where natural recovery rates trend upwards of 50%. This

discrepancy likely reflects potential recruitment bias, in that individuals who recovered from stuttering at a young age were less likely to have participated.

### **Family history of stuttering**

Our reports on family history are the most comprehensive to date, detailing relatives of first, second, third degree, and greater. Notably, a larger proportion of participants reported at least one male relative who stuttered (41.8%) compared to at least one female relative who stuttered (16.6%). In line with existing studies,<sup>12–14,39</sup> 49.9% of participants reported at least one family member who stuttered. However, these reports have not been validated further, so we cannot rule out erroneous reporting.

### **Factors associated with stuttering severity**

Factors previously found to be associated with stuttering persistence include being male, having a positive family history of stuttering, and poorer speech and language skills.<sup>14</sup> We were unable to confirm these associations with persistence nor with stuttering severity in our cohort.

### **Impact of stuttering on anxiety, education, and employment**

An increase in self-reported stuttering severity was associated with a greater likelihood of being anxious while stuttering, which is consistent with the existing literature.<sup>40</sup> The non-linear relationship reported in this study reflects inconsistencies in past work examining the relationships between anxiety and stuttering.<sup>2,19,41</sup> Of note, people with persistent stuttering reported lower anxiety levels than people with recovered stuttering. This may be due to stronger negative associations with stuttering in people with recovered stuttering or to people with persistent stuttering having developed better coping mechanisms to manage their anxiety; however, further exploration is required to draw firm conclusions. The complex interrelationships between stuttering, anxiety, and depression warrant deeper investigation, as highlighted by a recent study of young adults that found depressive symptoms to be elevated in people who stutter compared to their fluent peers.<sup>42</sup>

A majority (80.7%) reported perceived impacts on educational attainment and/or employment prospects. However, our participants were more qualified than the general

population; 69% of Australians aged 20 to 64 years have a post-high school qualification<sup>43</sup> compared to 78.3% of adults in this study. There was also a higher proportion of adults with an undergraduate degree or higher compared to the general population (49.7% vs 35%).<sup>43</sup> These results are likely due to selection bias.

### **Methodological considerations**

The large sample size and detail in stuttering and demographic data is a strength of this study. Existing studies with comparable sample sizes have extracted data from epidemiological surveys, limiting the depth of information on stuttering phenotype.<sup>1,32</sup> Alternatively, others have recruited participants retrospectively on the basis of medical records, which also brings about a different sampling bias.<sup>44</sup> Our sample recruited a cohort of people who stuttered beyond the clinical setting, arguably capturing a broader sample. Nonetheless, participants were self-selecting and the impact of this ascertainment bias should be considered when interpreting results, particularly the high proportion of adult participants, stuttering persistence, and family history. Furthermore, while our data provide valuable insight into the lived experience of stuttering, the non-epidemiological recruitment method limited robust comparisons to previously published epidemiological or true population-based data, making it difficult to make broader conclusions about this population.

This study relied on patient self-report gathered from a detailed online survey, with many items from the published Early Language in Victoria Study survey.<sup>2,4,24</sup> Therefore, we were unable to collect data on clinician-rated stuttering phenotype, including stuttering recovery, for our large sample. While we obtained a comprehensive representation of the lived experience of stuttering, our data could have been augmented with additional information from a standardized tool, such as the Overall Assessment of the Speaker's Experience of Stuttering.<sup>45</sup> Administration of standardized tools comes with a cost that was not possible in the present study and this would have also doubled the time taken for current respondents. Furthermore, the validation study, and a complex system of data cleaning and cross-checks, confirmed consistency across participants' responses, thus maintaining data fidelity. Pleasingly, and of

relevance for other studies seeking to recruit case cohorts via online recruitment, our validation study demonstrated a 100% concordance between self-reported stuttering and stuttering assessed by videoconference with a speech pathologist, as outlined earlier. Furthermore, we repeated several analyses, restricting them to only those individuals who reported seeking help or therapy for their stutter (704 out of 852 adults), due to the greater confidence that this subset did not simply experience typical dysfluencies. Our findings were overall similar where analyses were restricted to this subset, compared to the analyses including all participants (Appendix S1).

### **Conclusions**

This is the largest prospective sample of people who stutter reported to date. Our summaries of family history are the most comprehensive to date, detailing each family member who stuttered and showing that close to 50% of participants reported a family history of stuttering. While 75.9% reported that they had sought stuttering therapy, only 15.5% identified as having recovered. Additionally, a negative correlation with age for both stuttering frequency and severity was reported in adults. The frequent persistence of stuttering and the high proportion with a family history suggest that stuttering is a complex trait that does not often resolve, even with therapy. These rich data provide additional evidence of the complex nature of stuttering.

### **ACKNOWLEDGEMENTS**

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### **Supporting information**

The following additional material may be found online:

**Table S1:** Summary of stuttering frequency

**Table S2:** Stuttering phenotypes

**Table S3:** Physical responses and additional behaviours accompanying stuttering

**Table S4:** Communication problems diagnosed by a speech pathologist

**Table S5:** Summary of other neuropsychiatric conditions and health problems reported by participants

**Table S6:** Access to speech therapy services and specific treatments in adults and children

**Table S7:** Associations with severity scores

**Table S8:** Associations with anxiety due to stuttering in 261 adult participants with persistent stuttering

**Table S9:** Stuttering severity score distributions by reported anxiety level

**Table S10:** Results from a generalized ordinal logistic regression model, examining the association of speech severity score on anxiety level

**Table S11:** Items causing anxiousness in participants with persistent stuttering, who reported that they were anxious while stuttering

**Table S12:** Severity score distributions, by education level, in participants with persistent stuttering

**Table S13:** Associations with highest educational level in participants with persistent stuttering

**Table S14:** Results from a generalized ordinal logistic regression model, examining the association of speech severity score on educational level, in participants with persistent stuttering

**Figure S1:** Overview of stuttering phenotyping survey contents.

**Figure S2:** Age of stuttering onset for adult and child participants.

**Figure S3:** Summary of participants with persistent and recovered stuttering.

**Figure S4:** Combinations of stuttering phenotypes showing the proportions of males and females.

**Figure S5:** Comparison of self-rated stuttering severity scores versus speech pathologist ratings of severity in a subset of 54 individuals included in the severity study.

**Figure S6:** Combinations of specific treatment programmes and therapies received.

**Figure S7:** Anxiety level while stuttering against stuttering severity in adult participants with persistent stuttering.

**Figure S8:** Severity scores in individuals who did, or did not feel, their stutter had affected their education and/or employment prospects.

**Appendix S1:** Supplementary results

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**Table 1:** Basic demographics<sup>a</sup>

	Males	Females	Total
Total participants, <i>n</i> (%)	687 (69.6)	300 (30.4)	987
Age, years:months, median (range)	47:0 (7–93)	38:6 (7–86)	45:0 (7–93)
Age group, years, <i>n</i> (%)			
<18	97 (14.1)	38 (12.7)	135 (13.7)
18–24	55 (8.1)	37 (12.3)	92 (9.3)
25–39	133 (19.4)	76 (25.3)	209 (21.2)
40–54	139 (20.2)	50 (16.7)	189 (19.2)
55–69	176 (25.6)	65 (21.7)	241 (24.4)
≥70	87 (12.7)	34 (11.3)	121 (12.3)
Self-reported ancestry, <i>n</i> (%)			
European	596 (86.8)	263 (87.7)	859 (87.0)
South Asian	35 (5.1)	4 (1.3)	39 (4.0)
Aboriginal/Torres Strait Islander	5 (0.7)	3 (1.0)	8 (0.8)
African	4 (0.6)	5 (1.7)	9 (0.9)
East Asian	12 (1.8)	5 (1.7)	17 (1.7)
Mixed/other	32 (4.7)	20 (6.7)	52 (5.3)
Not available	3 (0.4)	0 (0)	3 (0.3)
Highest level of education (participants ≥18 years), <i>n</i> (%)			
High school	76 (12.9)	48 (18.3)	124 (14.6)
Trade/apprenticeship	73 (12.4)	3 (1.2)	76 (8.9)
Diploma/certificate	160 (27.1)	88 (33.6)	248 (29.1)
Undergraduate degree	148 (25.1)	65 (24.8)	213 (25.0)
Postgraduate degree	133 (22.6)	58 (22.1)	191 (22.4)
Highest level of education (primary carer of participants aged <18 years), <i>n</i> (%) <sup>b</sup>			
High school	7 (7.2)	3 (7.9)	10 (7.4)
Trade/apprenticeship	4 (4.1)	3 (7.9)	7 (5.2)
Diploma/certificate	31 (32.0)	12 (31.6)	43 (31.9)
Undergraduate degree	27 (27.9)	12 (31.6)	39 (28.9)

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Postgraduate degree	28 (28.9)	8 (21.1)	36 (26.7)
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<sup>a</sup>Where figures do not add up to the corresponding totals, data were missing.

<sup>b</sup>For participants aged <18 years, stratified by sex.

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**Table 2:** Anxiety due to stuttering, stratified according to recovered and persistent stuttering

<b>Anxiety level due to stuttering</b>	<b>Recovered stuttering</b> <b>(<i>n</i> = 46)</b>	<b>Persistent stuttering</b> <b>(<i>n</i> = 261)</b>
Not anxious	4 (8.7)	25 (9.6)
A little anxious	6 (13.0)	65 (24.9)
Fairly anxious	7 (15.2)	71 (27.2)
Very anxious	17 (37.0)	70 (26.8)
Extremely anxious	12 (26.1)	30 (11.5)

Data are *n* (%).

**Table 3:** Perceived impact of stuttering on education and employment outcomes

<b>Perceived impact</b>	<b>Persistent stuttering (<i>n</i> = 713)</b>
Participants felt their stuttering had affected their educational attainment	459 (64.4)
Participants felt their stuttering had been a barrier to finding employment	395 (55.4)
Participants felt their stuttering had been a barrier to career progression	467 (65.5)
Participants felt their stuttering had affected at least one of the above	575 (80.7)

Data are *n* (%).

## Figure legends

**Figure 1:** Summary of co-occurring communication, developmental, and neuropsychiatric phenotypes. (a) Summary of communication difficulties (other than stuttering), behavioural and attention difficulties, neurodevelopmental difficulties, and neuropsychiatric conditions reported by adult participants. (b) Summary of co-occurring communication difficulties (other than stuttering), behavioural and attention difficulties, and neurodevelopmental difficulties, reported in paediatric participants. See Table S5 for a full breakdown of individual conditions. (c) Family history of stuttering by degree of relatedness. First-degree relatives include parents, children, and siblings; second-degree relatives include aunts, uncles, nieces, nephews, grandparents, and grandchildren; other relatives include great-grandparents, great-grandchildren, great aunts, great uncles, cousins, and other relatives.

**Figure 2:** Relationship between stuttering severity and age. (a) Scatter plot of participant age versus severity score, with locally estimated scatter plot smoothing curve, fitted in adult and paediatric participants separately. The vertical dotted line at 18 years is the age cut-off for adult participants. Above the plot is a histogram of ages. The points and histogram are coloured according to age categories. (b) Distribution of severity scores in the different age categories.

# Reporting checklist for cross sectional study.

Based on the STROBE cross sectional guidelines.

## Instructions to authors

Complete this checklist by entering the page numbers from your manuscript where readers will find each of the items listed below.

Your article may not currently address all the items on the checklist. Please modify your text to include the missing information. If you are certain that an item does not apply, please write "n/a" and provide a short explanation.

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	Reporting Item	Page Number
<b>Title and abstract</b>		
Title	<a href="#">#1a</a> Indicate the study's design with a commonly used term in the title or the abstract	1
Abstract	<a href="#">#1b</a> Provide in the abstract an informative and balanced summary of what was done and what was found	4
<b>Introduction</b>		
Background / rationale	<a href="#">#2</a> Explain the scientific background and rationale for the investigation being reported	5-6
Objectives	<a href="#">#3</a> State specific objectives, including any prespecified hypotheses	6
<b>Methods</b>		

Study design	<a href="#">#4</a>	Present key elements of study design early in the paper	7-9
Setting	<a href="#">#5</a>	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	7-8
Eligibility criteria	<a href="#">#6a</a>	Give the eligibility criteria, and the sources and methods of selection of participants.	7
	<a href="#">#7</a>	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	7-9
Data sources / measurement	<a href="#">#8</a>	For each variable of interest give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group. Give information separately for for exposed and unexposed groups if applicable.	7-9
Bias	<a href="#">#9</a>	Describe any efforts to address potential sources of bias	7-9
Study size	<a href="#">#10</a>	Explain how the study size was arrived at	10
Quantitative variables	<a href="#">#11</a>	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen, and why	NA
Statistical methods	<a href="#">#12a</a>	Describe all statistical methods, including those used to control for confounding	7-9
Statistical methods	<a href="#">#12b</a>	Describe any methods used to examine subgroups and interactions	7-9
Statistical methods	<a href="#">#12c</a>	Explain how missing data were addressed	7-9
Statistical methods	<a href="#">#12d</a>	If applicable, describe analytical methods taking account of sampling strategy	7-9
Statistical methods	<a href="#">#12e</a>	Describe any sensitivity analyses	7-9
<b>Results</b>			

Participants	<a href="#">#13a</a>	Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed. Give information separately for exposed and unexposed groups if applicable.	10
Participants	<a href="#">#13b</a>	Give reasons for non-participation at each stage	NA
Participants	<a href="#">#13c</a>	Consider use of a flow diagram	NA
Descriptive data	<a href="#">#14a</a>	Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders. Give information separately for exposed and unexposed groups if applicable.	10-11
Descriptive data	<a href="#">#14b</a>	Indicate number of participants with missing data for each variable of interest	10-14
Outcome data	<a href="#">#15</a>	Report numbers of outcome events or summary measures. Give information separately for exposed and unexposed groups if applicable.	10-19
Main results	<a href="#">#16a</a>	Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included	10-19
Main results	<a href="#">#16b</a>	Report category boundaries when continuous variables were categorized	10-19
Main results	<a href="#">#16c</a>	If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period	10-19
Other analyses	<a href="#">#17</a>	Report other analyses done—e.g., analyses of subgroups and interactions, and sensitivity analyses	10-19
<b>Discussion</b>			
Key results	<a href="#">#18</a>	Summarise key results with reference to study objectives	15-19
Limitations	<a href="#">#19</a>	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias.	18

Interpretation	<a href="#">#20</a>	Give a cautious overall interpretation considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence.	15-19
Generalisability	<a href="#">#21</a>	Discuss the generalisability (external validity) of the study results	15-19
<b>Other Information</b>			
Funding	<a href="#">#22</a>	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	20

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## *Developmental Medicine & Child Neurology*

### DISCLOSURE OF INTERESTS

**Manuscript Number (if known):** DMCN-OA-21-08-0519.R2

**Manuscript title:** The nature and impact of developmental stuttering across the lifespan, as reported by study participants

All authors must sign the Disclosure of Interests form.

#### Author Disclosures

Any competing interest of any the authors must be declared. The aim of the statement is not to eradicate competing interests, as they are almost inevitable. Papers will not be rejected because there is a competing interest.

Name	Signature	Date	Competing Interest to Declare	
			Yes	No
1. Nathan Gillespie	1.	1/3/22		<i>no</i>
2.	2.			
3.	3.			
4.	4.			
5.	5.			
6.	6.			
7.	7.			
8.	8.			

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## *Developmental Medicine & Child Neurology*

### DISCLOSURE OF INTERESTS



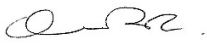
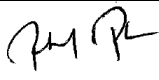


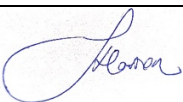
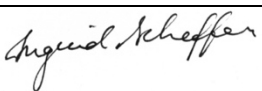


Manuscript Number (if known):

Manuscript title: *The nature and impact of developmental stuttering across the lifespan*

All authors must sign the Disclosure of Interests form.

#### Author Disclosures

Any competing interest of any the authors must be declared. The aim of the statement is not to eradicate competing interests, as they are almost inevitable. Papers will not be rejected because there is a competing interest.

Name	Signature	Date	Competing Interest to Declare	
			Yes	No
1. Jessica O. Boyce	1. 	03/08/2021		no
2. Victoria E. Jackson	2. 	03/08/2021		no
3. Olivia van Reyk	3. 	03/08/2021		no
4. Richard Parker	4. 	03/08/2021		no
5. Adam P. Vogel	5. 	03/08/2021	yes	
6. Else Eising	6. 	03/08/2021		no
7. Sarah E. Horton	7. 	04/08/2021		no
8. Ingrid E. Scheffer	8. 	04/08/2021	yes	
9. David J. Amor	10. 	03/08/2021		no
10. Michael S. Hildebrand	11. 	03/08/2021		no



11. Simon E. Fisher	12. <i>Simon E R</i>	03/08/2021		no
12. Nicholas G. Martin	13. <i>Nick Martin</i>	03/08/2021		no
13. Sheena Reilly	14. <i>Sheena</i>	03/08/2021		no
14. Melanie Bahlo	15. <i>Melanie Bahlo</i>	03/08/2021	yes	
15. Angela T. Morgan	16. <i>Angela</i>	03/08/2021		no

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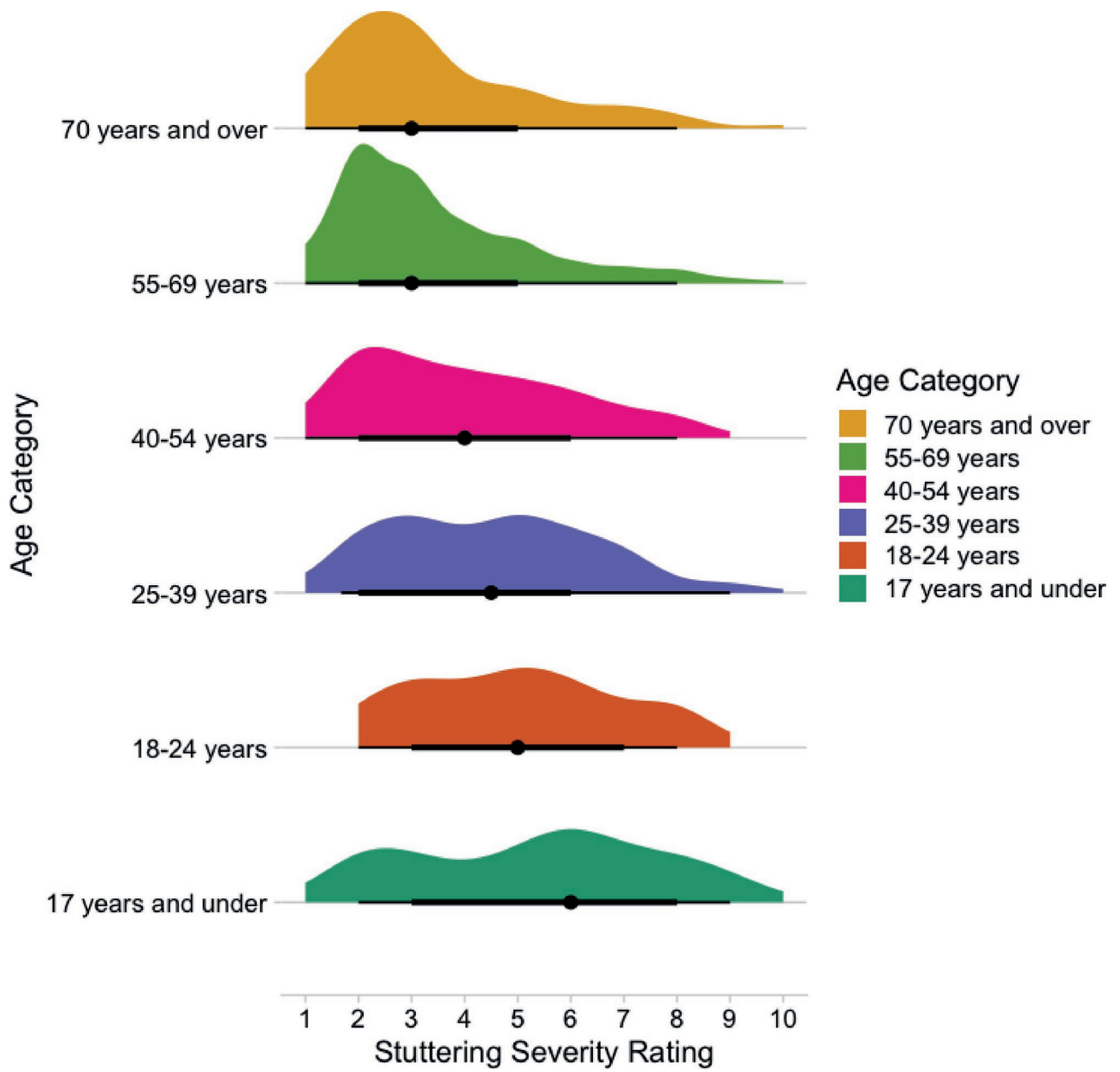


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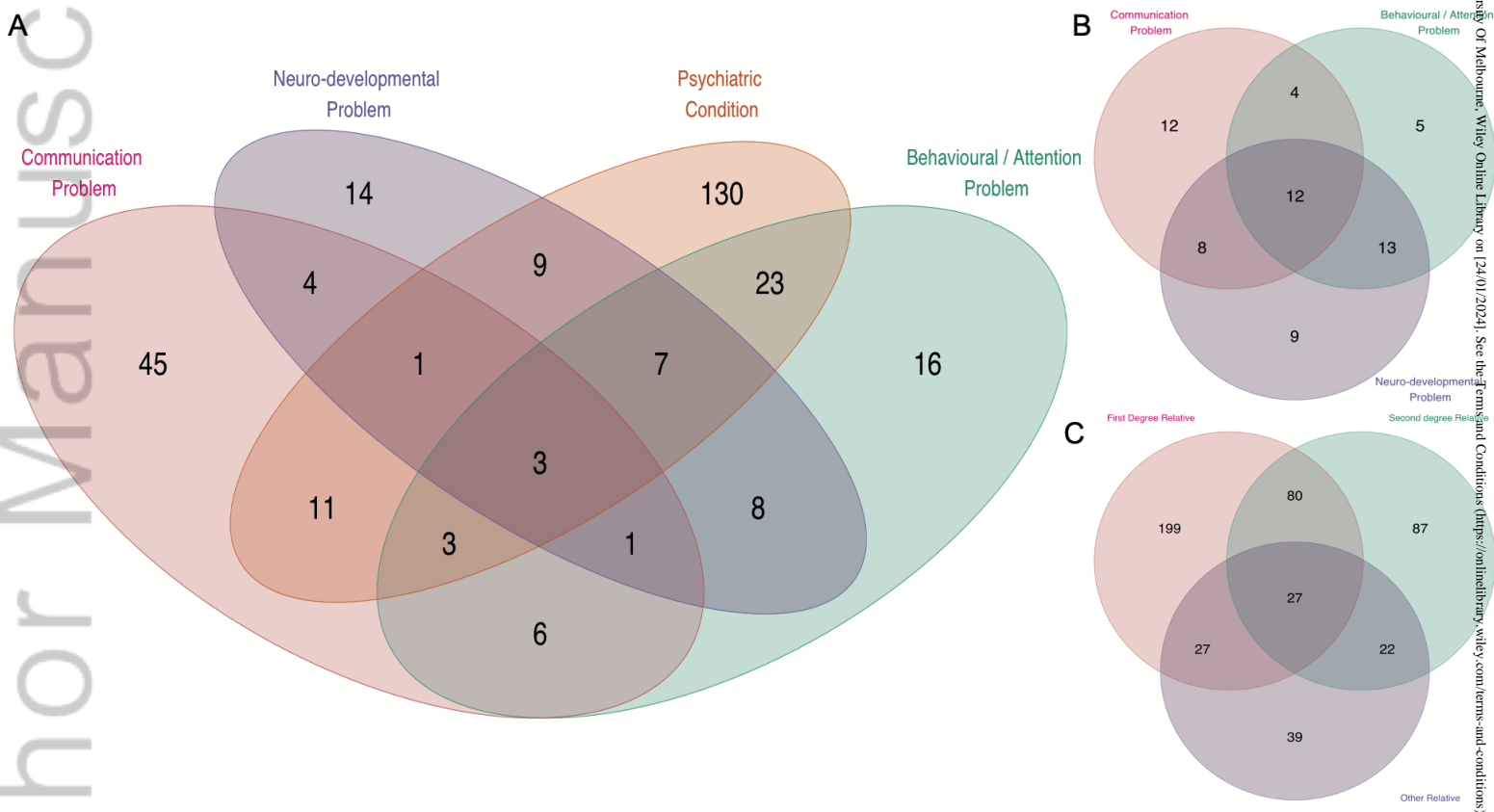
*APV is Chief Science Officer of Redenlab Inc, a speech neuroscience company.*

*IES has served on scientific advisory boards for UCB, Eisai, GlaxoSmithKline, BioMarin, Nutricia, Rogcon, Chiesi, Encoded Therapeutics, Xenon Pharmaceuticals, Zynerba and Knopp Biosciences; has received speaker honoraria from Liva Nova, GlaxoSmithKline, UCB, BioMarin, Biocodex, Chiesi and Eisai; has received funding for travel from UCB, Biocodex, GlaxoSmithKline, Biomarin and Eisai; has served as an investigator Zogenix, Zynerba, Ultragenyx, GW Pharma, UCB, Eisai, Xenon Pharmaceuticals, Anavex Life Sciences, Ovid Therapeutics, Epigenyx, Encoded Therapeutics and Marinus; and has consulted for Zynerba Pharmaceuticals, Atheneum Partners, Ovid Therapeutics, Care Beyond Diagnosis, Epilepsy Consortium and UCB. She may accrue future revenue on pending patent WO2009/086591; has a patent for SCN1A testing held by Bionomics Inc and licensed to various diagnostic companies, and has a patent for a molecular diagnostic/therapeutic target for benign familial infantile epilepsy (BFIE) ) [PRRT2] WO/2013/059884. She receives/has received research support from the National Health and Medical Research Council of Australia, Health Research Council of New Zealand, CURE, Australian Epilepsy Research Fund, Medical Research Future Fund and NIH/NINDS.*

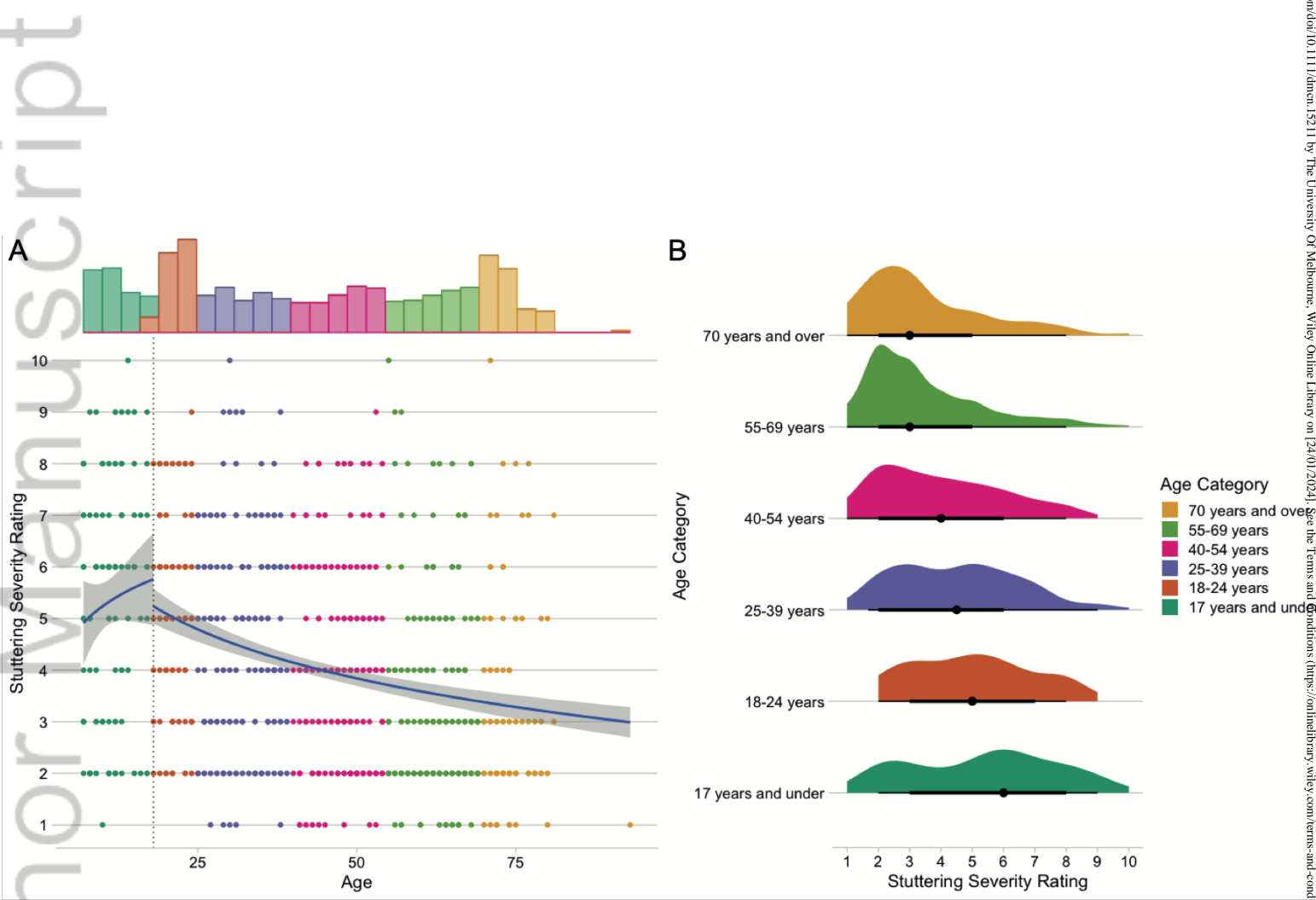
*Melanie Bahlo has received funding for research carried out in this work.*



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DMCN\_15211\_Morgan OA Fig 1.tif



DMCN\_15211\_Morgan OA Fig 2.tif