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Keywords Intellectual disability, challenging behaviour, cumulative risk, relative risk, population sample.

Objectives. Considerable variation has been reported in the prevalence and correlates of challenging behaviour in adults with intellectual disabilities. To provide a robust estimate of prevalence, we identified the entire administrative population of adults with ID in a defined geographical area and used a behaviour assessment tool with good psychometric properties.

Methods. Data from 265 adults who were known to services were collected using a demographic survey tool and the Behavior Problems Inventory – Short Form. The prevalence of self-injurious, aggressive/destructive, stereotyped, and overall challenging behaviour was evaluated. We explored the potential of developing Cumulative Risk Indices (CRI) to inform longitudinal research and clinical practice.

Results. The prevalence of overall challenging behaviour was 18.1% (95% CI: 13.94%-23.19%). The prevalence of self-injurious behaviour was 7.5% (95% CI: 4.94%-11.37%), aggressive and destructive behaviour 8.3% (95% CI: 5.54%-12.25%), and stereotyped behaviour 10.9% (95% CI: 7.73%-15.27%).

Communication problems and severity of ID were consistently associated with higher risk of challenging behaviours. CRIs were significantly associated with challenging behaviours and the five methods of CRI development produced similar results.

Conclusions. Findings suggest a multi-element response to challenging behaviour is likely to be required that includes interventions for communication and daytime activity. Exploratory analyses of CRIs suggested these show promise as simple ways to capture cumulative risk in this population. Subject to longitudinal replication, such a tool may be especially useful in clinical practice to identify adults who are priority for interventions and predict future demand on services.

Practitioner Points:

- The prevalence of challenging behaviour was 18.1% in this total population study. Stereotypy was the most frequent type of challenging behaviour.
- Communication difficulties and severe profound ID were most systematically related to the presence of challenging behaviour.
- Establishing the effect of multiple risk factors is likely to identify people who are priority for interventions. Addressing multiple, rather than singular risks, is likely to be more efficacious.
- We tested five different methods of putting together a Multiple Risk Index.
- All methods provided a reasonable association with challenging behaviour. The most user-friendly method was the additive CRI.

Limitations:

- This is a cross sectional design which enabled factors currently associated with challenging behaviour to be identified for the whole cohort, but these variables may not be those conferring risk for the development or maintenance of challenging behaviour over time.
- Future longitudinal research is required to replicate these CRI analyses before concluding about the CRI method with the highest predictive validity.

Introduction

The most robust methodology for estimating prevalence of challenging behaviour in adults with intellectual disabilities (ID) is to sample at a population level. However, the prevalence of challenging behaviour reported in administratively defined population studies shows considerable variation: from 4% in Holden & Gitlesen's (2006) study in Norway to 22% in Jones *et al's* (2008) study in Scotland, for example. Inconsistencies are also apparent in the correlates of challenging behaviour. The presence of autism, for example, has been identified in some studies to be associated with higher levels of challenging behaviour (Lundqvist, 2013) but not in others (Jones *et al*. 2008). Variability in prevalence and correlates may be a function of differences in sampling methodology and in the definitions of challenging behaviour (Emerson *et al.* 2001a). This variability is confusing and may impede accurate service planning and the development of effective interventions (Lowe *et al.* 2007; Jones *et al*. 2008; Lundqvist, 2013)

Variability is likely to emerge in the estimation of prevalence when definitions of challenging behaviour differ (Cooper *et al.* 2009a). Kiernan and Qureshi (1993) attempted to overcome this problem by differentiating between challenging

behaviour that is ‘more’ or ‘less’ demanding. They defined challenging behaviour as ‘more demanding’ when it occurred daily, restricted engagement, required physical intervention, or resulted in a major injury. Using this definition, with mixed age groups, has yielded estimates of ‘more demanding’ challenging behaviour of 3.8% (Holden & Gitlesen, 2006), 5.7% (Kiernan & Qureshi, 1993), 7.8% (Emerson & Bromley, 1995), 5-10% (Emerson *et al*. 2001a), and 10% (Lowe *et al*. 2007). Including ‘less demanding’ challenging behaviour (aggression, destruction, self-injury or other problem behaviour, which did not meet the above criteria) increased prevalence estimates to 10-15% (Emerson *et al*. 2001a) and 11.1% (Holden & Gitlesen, 2006). It is important to note that there remains no agreed consensual, conceptual or operational definition of challenging behaviour.

Two more recent population-based studies report challenging behaviour prevalence in adults rather than mixed age population samples using classification tools designed for adults with ID and challenging behaviour. Jones and colleagues (2008) included adults aged 16-years and older and defined challenging behaviour using psychiatrists’ assessment and diagnostic criteria for psychiatric disorders for use with adults with learning disabilities/mental retardation (DC-LD) (Royal College of Psychiatrists, 2001). These adults were assessed by psychiatrists using a purpose-designed measure based on DC-LD criteria and other assessment tools including the Psychiatric Present State for Adults with Learning Disabilities (PPS-LD) (Cooper, 1997), an autism assessment derived from DCR-ICD 10 (WHO, 1993), and the Vineland Adaptive Behavior Scales (Sparrow, Balla & Cicchetti, 1984). Psychiatric assessment produced an overall estimate for challenging behaviour of 22.5%, compared with 18.7% using the DC-LD criteria. In the other population-based study, Lundqvist (2013) used the Behavior Problems Inventory (Rojahn *et al*. 2001)

to identify challenging behaviour in adults aged over 18-years in Sweden. Only behaviour rated on the BPI as having a severe impact (on a 4 point scale ranging from 0=never/no problem, to 3=severe) was defined as challenging. The overall prevalence of challenging behaviour was estimated at 18.7%. These two studies indicate that prevalence rates may be higher than the earlier population studies above that defined challenging behaviour in terms of 'more' or 'less demanding'.

In total population studies, increased severity of ID and the presence of communication difficulties have typically been found to be associated with more frequent or severe challenging behaviour (Emerson & Bromley, 1995; Emerson *et al*. 2001a; Holden & Gitlesen, 2006; Jones *et al*. 2008). There is more conflicting evidence regarding other potential correlates. Some studies found men were at greater risk of presenting challenging behaviour than women (Kiernan & Qureshi, 1993; Emerson *et al*. 2001a). Other studies, however, found women were at greater risk than men (Jones *et al*. 2008), or that there is no association between gender and challenging behaviour (Holden & Gitlesen, 2006). Associations have been reported between challenging behaviour and psychiatric diagnoses (Lowe *et al.* 2007; Lundqvist, 2013), type of residence (Holden & Gitlesen, 2006; Lowe *et al.* 2007; Jones *et al*. 2008) and age (Kiernan & Qureshi, 1993; Holden & Gitlesen, 2006), but not consistently across population studies.

The focus on correlates is important for its potential to provide a first level identification of likely risk factors for challenging behaviours. To date, correlates of challenging behaviour have been examined individually. We could locate no research, in population-based samples, on the cumulative impact of correlates. Following early work by Rutter (1979), evidence from non-ID populations is suggesting that behaviour problems are associated with *cumulative* risk, as opposed

to individual risk factors acting independently (Appleyard, Egeland, van Dulmen & Sroufe, 2005). Being able to measure cumulative risk, and understand how variations in levels of cumulative risk may relate to challenging behaviour in adults with ID is important for the development of preventative intervention approaches.

The purpose of the present study was to estimate the prevalence of challenging behaviour in the total administrative population of adults with ID residing in Jersey. Jersey is an island measuring 118 square kilometres, 14 miles off the coast of Normandy, France. We aimed to build on available evidence from recent population studies (Jones *et al*. 2008; Lundqvist, 2013) by using a psychometrically sound behaviour assessment tool to define challenging behaviour. A second aim of the study was to examine socio-demographic correlates of challenging behaviours and explore methods of combining information about correlates into cumulative risk indices.

Method

Participants

Participants were 265 persons ≥ 18 years of age administratively defined as having ID (i.e., who were receiving, or had received, support from services in Jersey). Table 1 summarises participant characteristics.

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Procedure

Figure 1 presents an overview of the population ascertainment process. The Health and Social Services (H&SS) administrative database, in Jersey, FACE (Functional Analysis of Care Environments, 2012) was reviewed and 330 potential participants were identified. FACE records were then cross-referenced with current Education Department Record of Needs, and records of individuals maintained by local service providers from the voluntary sector and employment support services. This identified 52 additional individuals. A further 8 people were identified by researchers during the data collection process (new referrals to H&SS), providing 390 potential participants. Researchers screened all potential participants to ensure they met eligibility criteria. Persons were removed from the sample if they were deceased (N=13), had no ID diagnosis (from FACE records, N=45), were duplicate name entries on FACE (N= 9), had moved away permanently from Jersey (N= 8) or were below the age criterion (N= 4). This left 311 potential participants. Nine declined consent and a further 11 were traced but contact was not reciprocated. Some individuals were difficult to trace mainly due to minimal historical contact with services and their details were no longer accurate. Health and social care colleagues attempted to locate these individuals at the request of researchers and local death records were checked. Twenty six people were not traced. Surveys were completed with 265 participants which is equivalent to 97% of eligible and traceable participants (N=274) or 85% of eligible people (N=311).

Based on the most recent population census (States of Jersey, 2011), there are 78,342 adults over the age of 18 resident in Jersey. This suggests the administrative prevalence of ID is 0.40% in Jersey. The administrative ID prevalence

estimate is similar to adult population ID estimates in other studies (0.33-0.48%: Jones *et al*. 2008; Lundqvist, 2013).

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Ethical approval

The study was approved by the School of Psychology research ethics and governance committee at Bangor University and by the States of Jersey, Health and Social Services ethics committee. The consent process and accompanying documentation was designed using guidance from the Mental Capacity Act (2005) and the National Research Ethics Service (NRES) (http://www.nres.nhs.uk/). A lead professional was initially identified from the relevant databases who had previously supported the participant. Researchers met with the lead professional to complete a capacity assessment to determine whether the participant had the capacity to give consent to take part in this research. If the participant was assessed by the lead professional as not having capacity a personal consultee was approached. If the participant was assessed as having capacity this was further assessed face to face by researchers. If the participant was then assessed as not having capacity a personal consultee was identified and approached and asked to consider consent using detailed study information provided. If the participant was assessed as having capacity, then a researcher completed the consent process with them alongside a witness. If participants, or their personal consultees, did not give consent then they were not involved in the research project. Informed consent was obtained from 162

adults. For 103 adults who did not have capacity to provide independent consent consultees gave consent.

Measures

A proxy informant was identified to complete the surveys for each participant. Informants were mainly either family members or key workers within a supporting organisation. All Informants who provided information about the participant had a minimum of regular weekly contact and had known the participant for at least one year. Researchers met face-to-face with proxy informants to complete the demographic survey and the Behavior Problems Inventory - Short Form (BPI-S).

1. Individual and Demographic Survey.

Individual and demographic data were gathered using a 22-item structured questionnaire adapted from the Individual Schedule of the Challenging Behaviour Survey (Alborz, Bromley, Emerson, Kiernan & Qureshi, 1994) and the Wessex Scale (Kushlick, Blunden & Cox, 1973). Questions gathered information about the presence of Down syndrome, autism, dementia and any other syndromes / conditions; information about the degree of ID, formally recorded psychiatric disorders, epilepsy, sensory functioning, mobility, health, daytime engagement, and communication skills. Researchers selected most variables on an empirical basis due to their associations with challenging behaviour in previous studies. The Individual Schedule from the Challenging Behaviour Survey has been used in other population studies involving adults with ID (Holden & Gitlesen, 2006; Lowe *et al*.

2007) and has adequate inter-rater reliability (Emerson *et al*. 2001b; Lowe *et al*. 2007). The Wessex Scale has similarly been used in previous studies (Moss, Oliver, Arron, Burbridge & Berg, 2009) and there is evidence for good inter-rater reliability at subscale and item level (Kushlick *et al*. 1973; Palmer & Jenkins, 1982). Definitions for degree of ID were taken from the UK Department for Work and Pensions Guidance (2012). This definition categorises degree of ID based on IQ score (mild: 50-69; Moderate: 35-49; severe: 20-34; profound: less than 20) and describes typical daily living skills and support needs associated with each category.

2. The Behavior Problems Inventory - short form for use with individuals with intellectual disabilities (BPI-S).

The BPI-S was developed to measure challenging behaviours in adults with an ID presented during the previous six months (Rojahn *et al*. 2012a). The BPI-S is a shorter version of the Behavior Problems Inventory-01 (BPI-01) which has been used in a number of studies and shown to have good psychometric properties (Rojahn *et al*. 2001). The BPI-S has 30 questions in three categories. The self-injurious behaviour (SIB) subscale contains eight items, the aggressive-destructive behaviour (ADB) sub-scale ten items, and the stereotyped behaviour (SB) sub-scale twelve items. The BPI-S has two Likert-type rating scales per item – a five point frequency scale (never = 0; monthly = 1; weekly = 2; Daily = 3; Hourly = 4) and a three point severity scale (mild = 1; Moderate = 2; Severe = 3). A mild rated behaviour is defined as behaviour that does not cause significant damage to the individual or others, such as reddening of the skin, and any property damage does not require repair or replacement. A moderate severity behaviour is one that causes

damage to the individual or others such as bruising or causes damage to the environment requiring repair. A severe impact behaviour is one that inflicts moderate to severe damage to the individual or others requiring medical intervention or causes damage to items which are beyond repair. Frequency and severity of behaviour are measured for the SIB and ADB subscales, and frequency alone for the SB subscale. A total frequency score can also be obtained across all items on the BPI-S, and a total severity score for ADB and SIB only.

The BPI-S has good psychometric properties (Rojahn *et al.* 2012b; Mascitelli *et al*. 2015). In the present study, Cronbach's alpha for the total BPI-S frequency and severity scales was .891 and .773 respectively. The α-coefficient for the SIB subscale was .681 and .627 for the frequency and severity scale respectively. The α-coefficients for the ADB subscale were .792 and .788 for the frequency and severity scale respectively. The SB subscale, which only includes frequency ratings, had an α-coefficient of .867. Overall, internal consistency of the BPI-S was considered adequate to good depending on the domain measured. The comparatively lower α-coefficients for the SIB subscale have also been reported in previous studies, and may be related to either lower frequencies of behavioural topographies or ambiguity of the SIB construct (Rojahn *et al*. 2012b; Mascitelli *et al*. 2015).

There was a significant relationship between all subscales including SIB and ADB, rho=.253, p<.001; SIB and SB, rho=.445, p<.001; SB and ADB, rho=.442, p<.001; SIB and BPI-S Total, rho=.596, p<.001; ADB and BPI-S Total, rho=.731, p<.001; SB and BPI-S Total, rho=.815, p<.001.

Definition of challenging behaviour

To develop a definition of challenging behaviour a working group of clinicians and researchers initially considered the BPI-S structure. Clinical experience highlighted the importance of considering both frequency and severity variables as considered in previous definitions of challenging behaviour - including Diagnostic Criteria with Learning Disabilities (DC-LD) (Royal College of Psychiatrists 2001). The definition was piloted initially to check that it could be applied to the BPI-S data before being finalised and used in the study to establish prevalence. The process of developing a definition was also informed by a comprehensive review of previous research from early studies that defined ‘more’ or ‘less’ challenging behaviour (e.g. Kiernan et al. 1997; Emerson et al. 2001ab) and recent population studies such as Lundqvist (2013) who applied a clinical definition to the rating tool (Behavior Problem Inventory -01) utilised to collect data. Whereas Lundqvist (2013) only used severe rated behaviour listed on the BPI-01 we felt this too restrictive and included a high frequency element in all categories, especially as the SB scale on the BPI-S has no severity score. This led to a working definition of challenging behaviour, used to code behaviour problems as measured by the BPI-S:

a) SIB: Self-injurious behaviour is “challenging” if either it is rated as severe and occurs at least weekly, or is rated as moderate but occurs at least daily. Any other occurrence of self-injurious behaviour is not rated as challenging.
b) ADB: Aggressive destructive behaviour is “challenging” if either it is rated as severe and occurs at least weekly, or is rated as moderate but occurs at least daily. Any other occurrence of behaviour is not rated as challenging.
c) SB: Stereotyped behaviour is “challenging” if it occurs at the highest rated frequency (hourly). Any other occurrence of behaviour is not rated as challenging.

d) CB: Overall challenging behaviour is defined by the presence of a least one behaviour defined as “challenging” in the above categories.

Data analysis

Data were analysed using Statistical Package for the Social Sciences Version 21 (SPSS, Inc., Chicago, Il, US). The point prevalence of types of challenging behaviour (as defined above) was calculated with 95% confidence intervals.

To examine the association between demographic characteristics and challenging behaviour we followed two steps. In Step 1, we estimated a Relative Risk (RR) between each demographic indicator and challenging behaviour outcomes, as an indicator of their association. Variables from the demographic questionnaire were first cleaned and response categories collapsed, where appropriate. For example, response categories for incontinence (doubly incontinent, incontinent once a week or more, sometimes incontinent but less than once a week) were collapsed to create a binary variable ‘incontinent’ and ‘fully continent’. This process produced 19 dichotomous variables and 2 other continuous variables – age and time in the current setting (family, individual home, or service setting). RRs were calculated to explore the strength of association between each demographic factor and challenging behaviour outcomes. RR was calculated using the formula RR=(a/(a+b))/(c/(c+d)) for the four possible subgroups in a 2x2 table. This formula is appropriate for binary outcomes. The 95% confidence intervals of the RR were computed using the formula RR=logRR±1.96×SE where SE(RR)=sqrt[1/a+1/c]-[1/(a+b)]-[1/(c+d)] (Altman, 1991). Where there were potential associations with continuous covariates, these variables were centred at their median value (Kraemer

& Blasey, 2004). We then estimated RR using a Generalised Linear Model (GLM) where the exponentiated coefficient ‘b’ is equivalent to RR (Knoll, Le Cessie, Algra, Vandenbroucke & Groenwold, 2012).

In Step 2, we explored the strength of association between a cumulative number of demographic characteristics and challenging behaviour outcomes. For this, we used five different methods to create a cumulative risk index (CRI). In this step, we used 20 of the 21 potential correlates: living with a partner was removed from further consideration due to the very low numbers of people living with a partner.

CRIs are typically used to predict behavioural outcome following exposure to risk (Small & Luster, 1994; Evans, Dongping & Sepanski-Whipple, 2013). In this study, CRIs were not developed to predict any outcomes, as this is a cross-sectional study. Our aim was to explore the type of CRI that would demonstrate the strongest concurrent association with challenging behaviour. Such findings can then inform future longitudinal research, and clinical practice that aims to prioritise adults for intervention and predict future demand on services.

We first recoded all demographic indicators to binary variables, as CRIs are typically constructed by summing dichotomised risk factors (Evans *et al*. 2013). The two non-binary variables (Step 1 above) were thus recoded. Chronological age was dichotomised at 25 years. Under current UK service configuration, at 25 years of age individuals with ID transition from child to adult services. Time in setting indicated the length of time individuals had lived in their current residence and was dichotomised at 12 months, following an examination of the variable’s distribution (Table 1) and on the basis that 12 months may be an adequate period for settling in following a move.

Apart from its potential clinical utility, this approach presents several methodological advantages over other approaches most commonly used, such as multiple regression. Firstly, collinearity between risk factors does not distort estimates of multiple risk in the CRI model (Evans *et al*. 2013). Where regression models may reject certain risk factors in the final model, the CRI can accommodate independent and interdependent risk factors (Evans *et al.* 2013). Having multiple, correlated predictors in a regression model can affect estimates and reduce statistical power (Myers & Wells, 2003). Additionally, CRIs are parsimonious and statistically sensitive even with small samples (Cohen, Cohen, West & Aiken, 2003; Evans *et al*. 2013).

Five CRIs were constructed as follows:

CRI Method 1. All 20 available demographic variables were considered. Participants received a score of '1' for each 'risk' indicator present and '0' if absent (e.g. non-verbal=1, verbal=0). The number of factors present for each participant was estimated with 20 as the potential maximum value of the CRI.

CRI Method 2. A weighted CRI was calculated by multiplying each participant's CRI (Method 1) with the relevant RR scores produced for each challenging behaviour outcome. The summation of these scores produced a weighted CRI for each challenging behaviour. Where the initial CRI summed the number of 'risk' factors present, this weighted CRI also accounts for the intensity of risk exposures.

CRI Method 3. An outcome-specific CRI was created by summing the number of factors identified in step 1 as having a RR equal to or larger than 1.5 for each challenging behaviour category. Therefore, each challenging behaviour outcome had

a different CRI, depending on which of the 20 factors was univariately associated with a strength of (RR) 1.5 or higher.

CRI Method 4. A cumulative domain risk index (CDRI) was constructed not by considering individual 'risk' factors as in the previous methods, but by grouping individual factors in conceptually coherent domains of 'risk': (a) personal characteristics: age, gender; (b) diagnostic status characteristics: degree of ID, autism present, Down syndrome present, other syndrome present, psychiatric disorder, dementia; (c) placement characteristics: type of residence, time in setting, daytime engagement; (d) communication skills: presence of speech, speech clarity, understanding of communication; and (e) physical and sensory health: continence, mobility, epilepsy, seizures, vision and hearing. We examined whether any risk factor was present (1 vs 0) within each domain, and then created a CDRI by summing the number of domains where at least one 'risk' was present (range of CDRI scores 0-5). This method aimed to explore the suggestion that adverse outcomes come about when individuals are exposed to a higher number of risk domains (Campbell, Shaw & Gilliom, 2000).

CRI Method 5. A weighted CDRI was calculated by multiplying the number of 'risk' variables present within each risk domain (Method 4) by the total number of 'risk' variables within the domain, and dividing by the overall number of 'risk' variables available (i.e. 20). For example, the communication domain included three variables. If the participant scored yes to two of these the weighting would involve (2*3)/20. The summation of these results from each domain produced the weighted CDRI. This method is conceptually similar to method 4, but aims to account for any high concentration of 'risk' within a domain.

To explore the strength of the association between each different CRI type and challenging behaviour outcomes, we fitted Receiver Operating Characteristic (ROC) curves and compared the resulting Area Under the Curve (AUC) scores between methods. AUCs are equivalent to the Wilcoxon test of ranks (Hanley & McNeil, 1982) and an informative effect size denoting clinical significance (Kraemer *et al*. 2003).

Results

Prevalence of challenging behaviour

Using the definitions derived from BPI-S responses (see above) 18.1% of participants presented challenging behaviour (CB) (n=48) (95% CI: 13.94%-23.19%), 7.5% (n=20) presented SIB (95% CI: 4.94%-11.37%), 8.3% (n=22) presented ADB (95% CI: 5.54%-12.25%), and 10.9% (n=29) presented SB (95% CI: 7.73%-15.27%). Table 2 lists the most prevalent topographies within each subtype.

+++ Insert Table 2 +++

Correlates of challenging behaviour - Relative Risks

Figure 2 shows the results from the initial relative risk (RR) analysis on the association between each demographic characteristic with challenging behaviour outcomes. RRs above 1 indicate an elevated risk of presenting challenging behaviour, whereas RRs below 1 indicate a reduced risk of challenging behaviour.

Where 95% confidence intervals for the RR included 1, the RR was deemed not statistically significant.

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Three characteristics were systematically and significantly associated with all four categories of challenging behaviour (SIB, ADB, SB and overall CB). These were being non-verbal, having limited understanding of communication and having a severe-profound ID (RRs ranged from 2.471 to 12.16; see Figure 2). Living in paid or congregate care, the presence of an autism diagnosis, having no daytime engagement, the presence of another syndrome, impaired vision, no clear speech, incontinence, the presence of seizures, epilepsy and mobility problems were significantly associated with some topographies of challenging behaviour but not consistently across all categories. Age, gender, living with partner, Down syndrome, time in current setting, impaired hearing, dementia or psychiatric disorder were not significantly associated with challenging behaviour.

Cumulative Risk Indices (CRIs)

Having identified pairwise associations between demographic variables and challenging behaviour, we compared five methods of constructing CRIs. Where demographic characteristics explored were continuous variables (as was the case for age and years living in setting), the first step was to obtain RRs on binary versions of these variables. The RRs for these binary versions were as follows: For age RR (95%CI) = SIB: 0.819 (0.284-2.358); ADB: 0.963 (0.37-2.504); SB: 1.042

(0.467-2.322); overall CB: 0.862 (0.456-1.627). For time in current setting RR (95%CI) = SIB: 0.809 (0.197-3.325); ADB 1.618 (0.584-4.48); SB: 1.165 (0.433-3.131); overall CB: 1.04 (0.481-2.25).

Table 3 presents the AUCs resulting from the ROCs. As indicated by the magnitude and confidence intervals of the AUCs there was little variation among the five methods. AUCs close to 1 indicate that the CRI method accurately distinguishes between participants with challenging behaviour and no challenging behaviour. AUC scores closer to 0.5 indicate the CRI provides a risk prediction that is no better than chance (Zhou, Obuchowski & Obuchowski, 2002). Each CRI method performed resulted in a statistically significant AUC that was reasonably strongly associated with SIB (AUCs ranged from .719 to .756), ADB (AUCs ranged from .687 to .730) and overall CB (AUCs ranged from .750 to .784), and very strongly with SB (AUCs ranged from .827 to .891). The weighted CRI (Method 2) produced the largest AUCs (Table 3), however these were not significantly better than AUCs produced by any of the other methods as indicated by overlap in confidence intervals.

+++ Insert Table 3 +++

Discussion

The overall prevalence of challenging behaviour in adults with ID in Jersey was 18.1% (95% CI: 13.94%-23.19%). This is similar to other adult population studies that have used behaviour assessment tools designed for adults with ID (Jones *et al.* 2008: 18.7%-22.5%; Lundqvist, 2013: 18.7%).

The prevalence of discrete subtypes of challenging behaviour is seldom reported in total population samples. In the present sample, stereotyped behaviour (SB) was the most common behavioural form (SB = 10.9%) followed by aggressive-destructive behaviour (ADB = 8.3%) and self-injurious behaviour (SIB = 7.5%). High rate stereotyped behaviour can have a significant negative impact on quality of life and requires further investigation. The prevalence of stereotypy has been considered in only one other population study (Lundqvist, 2013) and was estimated at 6.1%. Other studies have reported ADB as the most prevalent behavioural form, followed by SIB (e.g., Cooper *et al*. 2009ab: ADB 9.8%, SIB 4.9%; Lundqvist, 2013: ADB 11.9%, SIB 8.4%). The present study also suggests that ADB appears to be more prevalent than SIB.

Three characteristics were significantly associated with all subtypes of challenging behaviour. These were non-verbal speech, limited understanding of verbal communication, and severe-profound ID. Communication impairments have been previously associated with challenging behaviour (Kiernan *et al.* 1997; Emerson *et al*. 2001ab; McClintock, Hall & Oliver, 2003; Holden & Gitlesen, 2006; Lundqvist, 2013). In the present study, 119 (45%) participants had an impairment of expressive communication (75 were non-verbal and 44 had impaired clarity of speech). Seventy-three (27.5%) had limited receptive understanding of communication. However, only 32 (12%) had augmentative alternative communication (26 used Makaton, 3 the Picture Exchange Communication System (PECS), 2 British Sign Language (BSL) and 1 an iPad application). Even when a person knew Makaton, it was not always clear whether their carers / staff were competent in its interpretation or use. Improving the ability of adults with ID and others to communicate effectively will be vital given the association between

impaired communication and challenging behaviour. The association between degree of ID and challenging behaviour also supports previous research (Emerson & Bromley, 1995; Kiernan *et al*. 1997; Emerson *et al*. 2001ab; McClintock *et al*. 2003; Holden & Gitlesen, 2006; Jones *et al*. 2008; Lundqvist, 2013).

A strong association was seen between lack of daytime engagement and self-injury (RR=3.729) and stereotypy (RR=2.615). Although the cross-sectional design of the present study cannot determine whether lack of engagement is a cause or effect of SIB/SB, 38.5% of participants in this study had no daytime engagement. This is high but similar to previous estimates of one-third (Lowe *et al*. 2007), emphasising the need for to increase levels of meaningful engagement.

Living in paid / congregate care (associated with SIB and SB here) has been reported as an associate of challenging behaviour in other studies (Holden & Gitlesen, 2006; Lowe *et al.* 2007; Jones *et al*. 2008). Incontinence (associated with SIB and SB) has been related to aggression in previous studies (Cooper *et al*. 2009ab), suggesting future research attention is warranted. Epilepsy (associated with SIB and SB) has been related to challenging behaviour in some studies (Deb, Thomas & Bright, 2001), but not in recent population-based samples (Jones *et al*. 2008; Lundqvist, 2013). Autism (associated with ADB and SB) has also been associated with challenging behaviour in previous research (McClintock *et al*. 2003; Lundqvist, 2013).

There was an equitable gender distribution in this sample (50.6% male, 49.4% female). This may be explained by the fact there was a greater proportion of men not included in the study either because researchers were unable to trace them, they did not reciprocate contact, or they declined consent (33 males vs. 13 females). It should be noted, however, that the precision of gender estimates in population studies is

unknown because of lack of confidence intervals (e.g., 562 (54.9%) men, 461 (45.1%) women: Jones *et al*. 2008). Of interest was also the lack of gender differences in the prevalence of challenging behaviour, contrary to findings in other studies (e.g. Emerson *et al*. 2001a; Jones *et al*. 2008). However, population studies with a similarly wide age range and larger sample (18-87 years, n=915; Lundqvist, 2013) also did not identify gender differences for most challenging behaviour categories. It is likely that the reported variability regarding gender differences in the literature is related to studies' sampling designs, participant age, and gender ratio.

The findings on correlates may highlight populations at risk of presenting higher levels of challenging behaviour. We extended this work by building exploratory CRIs and by testing different methods of constructing these. The aim of our approach was to identify a method of developing a CRI that clinical services will be able to use in the future (following longitudinal replication to establish its predictive validity) to predict service need and design interventions targeting multiple risk factors. Identifying adults with multiple risk factors is likely to identify vulnerable adults who are priority for interventions and addressing multiple rather than singular risks is likely to prove more efficacious (Evans *et al*. 2013). Our results indicated that any type of CRI was reasonably associated with challenging behaviour including the potentially service-friendly, uniform, additive CRI (Method 1). The advantage of this CRI method is that it could be developed into a user-friendly, easily interpreted tool for professionals to use in practice thus avoiding the need for more complex analysis. Weighting the CRIs as in methods 2 and 5 was associated with slightly better estimates, but the differences were not statistically significant from the simpler additive methods.

Some of the observed variation in AUCs is related to differences in the range of scores of each CRI method. The largest AUC was obtained for the weighted CRI which has the longest scale range. Using the weighted method, SB had the highest AUC score (.891) and the longest scale range (0-71) and ADB the lowest AUC score (.730) and the narrowest range (0-24). Similarly, the lowest AUC scores are seen in the CDRI (Method 4) which has the narrowest range (0-5). The CDRI and weighted CDRI, which were calculated using subgroups of risk factors, might be useful for future risk management as opposed to risk prediction. It should be noted that AUCs in this study were used to explore the potential association, not as a risk prediction test. The latter requires testing in a prospective study. Future longitudinal research is required to replicate these analyses before concluding about the CRI method with the highest predictive validity.

There are four main limitations of the present study. First, study findings apply only to the administratively defined population with ID in Jersey, as other adults with IQ < 70 in the community and not known to services were not included. Those with complex presentations, physical health problems, mental health problems or challenging behaviour may have greater support needs, be more likely known to services, and thus over-represented in the sample. However, findings from this study are likely useful in practice since specialised support (such as for challenging behaviour) might be best planned on the basis of a population of people with ID already known to services. A previous meta-analysis indicated an adult ID prevalence rate of 4.94/1000 (95% CI: 3.66-6.22) (Maulik *et al*. 2011) which would indicate that 80% of adults with ID in Jersey are listed on the FACE database. Given that surveys were completed on 85% of eligible participants results are a robust representation of the Jersey population in receipt of ID services. Secondly, the FACE

database was checked to explore whether participants had a record of a cognitive assessment or psychiatric diagnosis. Where they did not there was a reliance on proxy informants to identify level of ID and psychiatric diagnosis. There was a descriptive key listing typical presentations associated with level of ID (ability to complete tasks of daily living and support needs) taken from the Department of Work and Pensions (2012) definition to support informants with this. The third limitation is that the cross sectional design enabled factors currently associated with challenging behaviour to be identified for the whole cohort, but these variables may not have been those conferring risk for the development or maintenance of challenging behaviour over time. Finally, the list of potential risk factors investigated may not be exhaustive. Factors such as socio-economic position, traumatic life events, quality of environments and other psychosocial factors may contribute. Whilst there are methodological difficulties in assessing these a potential impact should be acknowledged.

The number of population based studies is small and they have multiple differences in terms of population, sample size, age, definitions, behaviours included, and research tools utilised. There is currently no clear way to systematically evaluate the impact of these methodological differences on prevalence data (e.g., via meta-analysis) because of the very small number of population based studies. With more population based studies published this will be a valuable exercise in future.

In summary, the present study identified the total administrative population of adults with ID in a defined geographical area and measured challenging behaviour with a well-validated tool that is appropriate for people with ID. The estimated prevalence of behaviour problems was similar to estimates provided in other recent population studies (Jones *et al.* 2008; Lundqvist, 2013). Communication difficulties

and severity of ID were consistently associated with a higher risk of presenting any type of challenging behaviour. These findings highlight the hypothesised function of challenging behaviour as communicative act, and point to specific subgroups that are more susceptible to presenting high levels of behaviour problems. Last, the study compared different methods of measuring cumulative risk, a method that has implications for clinical practice. Our findings indicated that CRIs are significantly associated with challenging behaviours and, subject to longitudinal replication, any method of CRI can measure risk with a similar potency.

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Table 1. *Characteristics of participants*

Characteristic	**Summary statistic**
Mean age in years (SD)	41.44 (16.278)
Gender	Male: 134 (50.6%)
	Female: 131 (49.4%)
Type of accommodation	Congregate care: 108 (40.8%)
	Paid carer: 24 (9.1%)
	Family carer: 91 (34.3%)
	Independent living: 42 (15.8%)
Time in setting	Less than 1 year: 32 (12.1%)
	1-5 years: 60 (22.6%)
	6-10 years: 62 (23.4%)
	11-20 years: 53 (20%)
	21 years plus: 58 (21.9%)
Degree of intellectual disability	Profound: 26 (9.8%)
	Severe: 32 (12.1%)
	Moderate: 83 (31.3%)
	Mild: 124 (46.8%)
Other diagnoses (include)	Autism: 31
	Down Syndrome: 36
	Cerebral Palsy: 15
	ADHD: 2
	Fragile X: 2
	Soto syndrome: 2
	Other: 11
Daytime engagement	Paid work: 37 (14%)
	Voluntary work: 39 (14.7%)
	Vocational training: 22 (8.3%)
	Education: 5 (1.9%)
	Day service: 60 (22.6%)
	No daytime engagement: 102 (38.5%)
Epilepsy	57 (21.5%)
Psychiatric condition	70 (26.4%) including:
	Depression: 31 (11.7%)
	Schizophrenia: 18 (6.8%)

Figure 1. *Flow diagram illustrating the population ascertainment process*

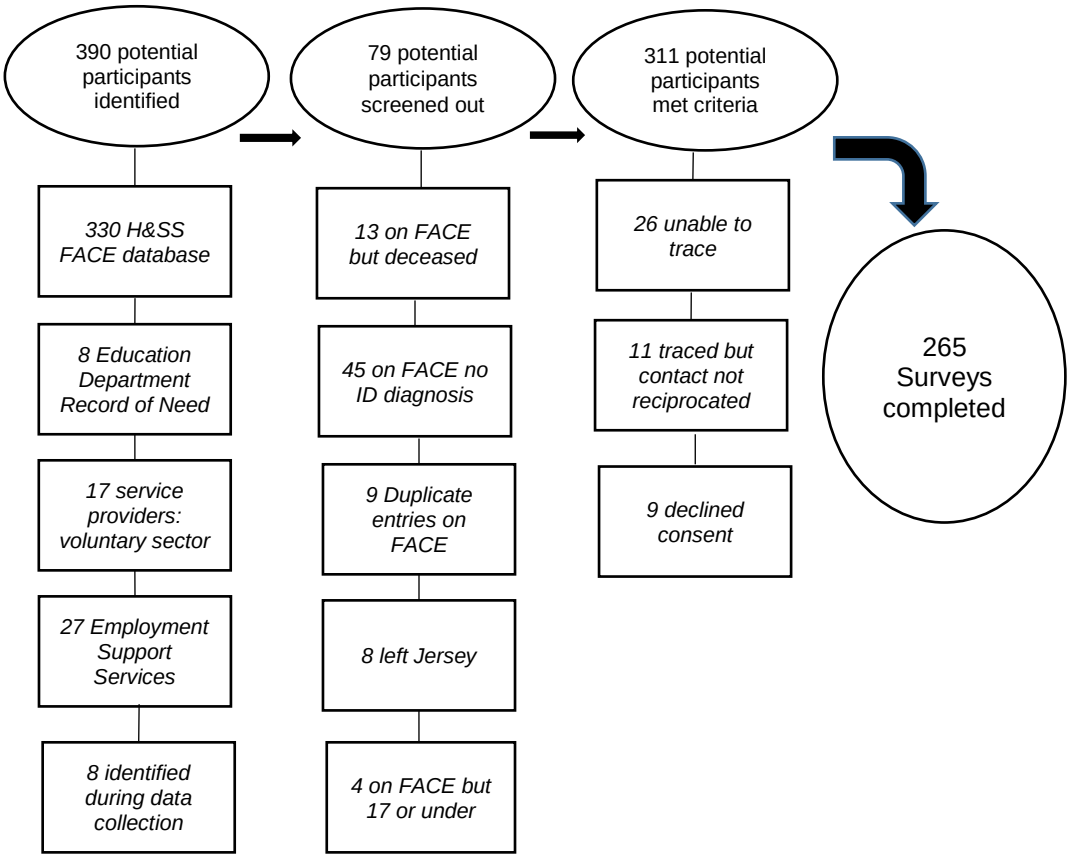

Table 2. *Prevalence of specific behaviours*

Category	Specific behaviour	Prevalence (%)	95% CI
Self-injurious behaviour (SIB)	SIB Total	7.5	4.94-11.37
	Self-scratching	3.0	1.54-5.84
	Head hitting	2.3	1.04-4.85
	Hair pulling	1.9	0.81-4.34
	Teeth grinding	1.9	0.81-4.34
Aggressive destructive behaviour (ADB)	ADB Total	8.3	5.54-12.25
	Verbally abusive	4.2	2.33-7.28
	Biting others	2.3	1.04-4.85
	Bullying	2.3	1.04-4.85
Stereotyped behaviour (SB)	SB Total	10.9	7.73-15.27
	Repetitive hand or finger movements	5.7	3.46-9.13
	Rocking / repetitive body movements	3.4	1.8-6.33
	Pacing, jumping, bouncing, running,	2.6	1.28-5.35
	Yelling and screaming	2.6	1.28-5.35
	Waving or shaking arms	2.6	1.28-5.35

Figure 2. *Forest plots illustrating Relative Risk (95% CI) between demographic factors and self-injurious behaviour, aggressive destructive behaviour, stereotyped behaviour and overall challenging behaviour*

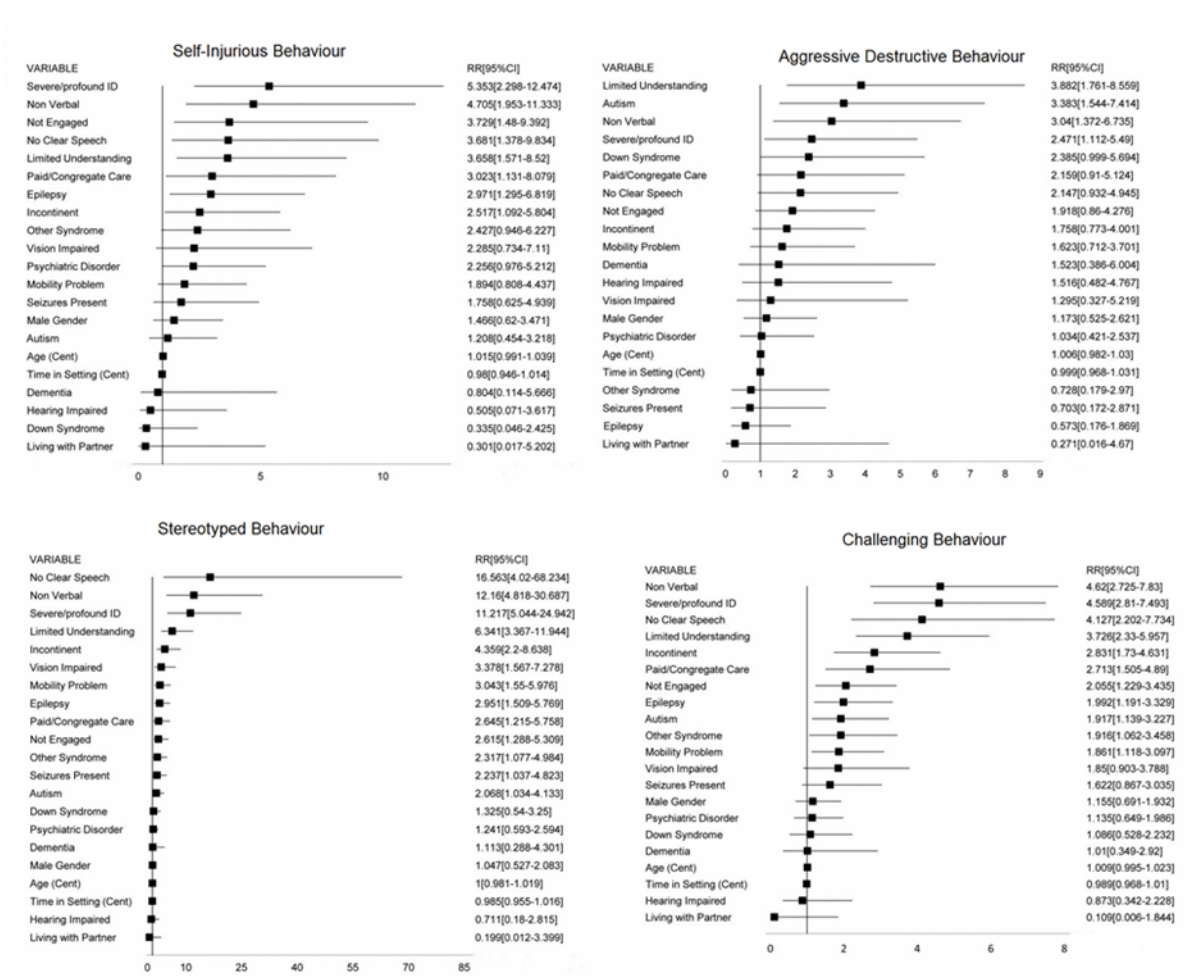

Table 3. *Area Under the Curve (AUC) results for the five cumulative risk indices (CRIs)*

Area Under the Curve (AUC)										
Method		**Range**	**Mean**	**SD**	**Behaviour**	**AUC**	**Asymp sig.**	**SE**	**Asymptotic 95% CI**	
									Lower Bound	**Upper Bound**
1.	**CRI**	0-13	4.65	2.96	SIB	.742	<.001	.064	.616	.867
					ADB	.703	.002	.056	.594	.812
					SB	.869	<.001	.034	.802	.935
					CB	.767	<.001	.041	.687	.846
2.	**Weighted CRI**	0-39	12.04	9.53	SIB	.756	<.001	.064	.630	.882
		0-24	8.54	6.19	ADB	.730	<.001	.054	.624	.837
		0-71	21.8	20.7	SB	.891	<.001	.026	.840	.941
		0-35	10.9	8.86	CB	.784	<.001	.039	.708	.859
3.	**Custom CRI**	0-13	3.77	2.83	SIB	.751	<.001	.063	.628	.874
		0-10	3.11	2.55	ADB	.724	.001	.056	.614	.833
		0-12	3.23	2.75	SB	.874	<.001	.030	.816	.932
		0-12	3.23	2.75	CB	.778	<.001	.038	.704	.852
4.	**CDRI**	0-5	2.88	1.27	SIB	.719	.001	.061	.599	.838
					ADB	.687	.004	.055	.579	.796
					SB	.827	<.001	.035	.759	.895
					CB	.750	<.001	.039	.674	.826
5.	**Weighted CDRI**	0-2.95	0.96	0.69	SIB	.731	.001	.064	.605	.858
					ADB	.696	.002	.055	.588	.804
					SB	.861	<.001	.033	.795	.927
					CB	.759	<.001	.040	.680	.838